

The Impact of Development Defects of Enamel on Young People

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Abstract

Background: in order to ascertain whether Developmental Defects of Enamel (DDE) constitute a public health problem it is necessary to consider whether the condition impacts on the lives of affected individuals.

Aim: to describe the impact of DDE on individual young people.

Objectives:

- 1) Describe the extent to which contemporary dental research on DDE has included the perspective of children and young people.
- 2) Describe the impact of DDE on the oral health-related quality of life (OHRQoL) of young people.
- 3) Explore, in detail, the impact of DDE on young people.

Methods: a systematic review of the literature, a cross sectional study and a qualitative interview study were conducted.

Results: Little research about DDE has previously attempted to capture the subjective experience of those with the condition. The impact of DDE, as measured with a child specific OHRQoL measure, was generally of low frequency and was equivalent to that of young people with relative oral health. In qualitative interviews the impact of DDE varied markedly between young people. DDE impacted on individuals' whose sense of self was defined by appearance and who depended on perceived approval from others about their appearance. No links between gender, age, severity of DDE and impact were apparent.

Discussion: The impact of DDE was generally of low frequency with marked variation in impact between individuals. This research is the first to discover that some of this variation can be accounted for by young people's sense of self. This concept has not been identified in relation to the impact of visibly different conditions before. More research is needed to further investigate the impact of DDE.

Contents

1. Chapter One Introduction.....	7
1.1 Background	7
1.2 Aim and objectives	8
2. Chapter Two Literature Review	10
2.1 Introduction	10
2.2 Developmental Defects of Enamel (DDE).....	10
2.3 Aetiology of DDE.....	11
2.3.1 Localised causes	12
2.3.2 Generalised causes	13
2.3.3 Aetiology of fluorosis	15
2.4 Are DDE a public health problem?.....	16
2.4.1 Prevalence.....	18
2.4.2 Impact of the condition on the individual	31
2.4.3 Impact of the condition on wider society	42
2.4.4 Condition is preventable and effective treatments are available	43
2.4.5 Conclusion.....	45
2.5 Approaches available to investigate the impact of dental conditions	47
2.5.1 Models of health	47
2.5.2 Investigating impact.....	50
2.5.3 Oral health-related quality of life (OHRQoL).....	56
2.5.4 Qualitative approaches to impact.....	63
2.5.5 Research with children.....	70
2.5.6 Methodological considerations of research with children.....	72
2.5.7 Contemporary approaches to research with children.....	74
2.5.8 Measurement of HRQoL of children	75
2.5.9 Measurement of OHRQoL in children.....	77
2.5.10 OHRQoL measures for children	79
2.5.11 Qualitative research of the impact of oral health on children.....	85
2.5.12 Conclusion.....	87
2.6 Rationale.....	87
2.7 Aim and objectives	88
2.8 Publications arising from the work in this chapter	89
3. Chapter Three.....	90
The extent to which contemporary dental research on DDE has included the perspectives of children & young people	90
3.1 Introduction	90
3.1.1 Objectives of the study.....	91
3.2 Methods.....	91
3.2.1 Objective a) Developing the classification.....	91
3.2.2 Objective b) Identify research papers	93

3.2.3	Objective c) Application of categories.....	96
3.3	Results	96
3.3.1	DDE literature.....	96
3.3.2	General child dental literature	100
3.3.3	Comparison of DDE and child dental literature	102
3.4	Discussion	102
3.5	Publications arising from the work in this chapter	105
4.	Chapter Four	106
	The impact of DDE on the OHRQoL of young people.....	106
4.1	Introduction	106
4.1.1	Objectives of the study.....	107
4.1.2	CPQ ₁₁₋₁₄	107
4.1.3	P-CPQ.....	109
4.2	Method	110
4.2.1	General clinic-based sample.....	110
4.2.2	DDE Sample	112
4.3	Results.....	113
4.3.1	Results of the evaluation of CPQ ₁₁₋₁₄ and P-CPQ from the general clinic-based sample	114
4.3.2	Results of the impact of DDE on young people	123
4.3.3	Results of the impact of DDE using P-CPQ.....	128
4.4	Discussion	128
4.5	Publications arising from the work in this chapter	137
5.	Chapter Five	138
	Exploring the impact of DDE on young people.....	138
5.1	Introduction	138
5.1.1	Objectives of the study.....	139
5.1.2	Design of the study	139
5.2	Methods.....	140
5.2.1	Sample.....	140
5.2.2	Procedure.....	140
5.2.3	Interviews	141
5.2.4	Data analysis.....	143
5.3	Results.....	144
5.3.1	Sample.....	144
5.3.2	DDE status of the young people	144
5.3.3	Qualitative data.....	147
5.4	Discussion	160
5.4.1	Impacts of marks.....	161
5.4.2	Influence of sense of self on the impacts of marks.....	162
5.4.3	Social interactions.....	165
5.4.4	Methodological aspects of the study	167
5.5	Publications arising from the work in this chapter	172

6. Chapter Six	173
Discussion	173
6.1 Introduction	173
6.2 Summary of findings.....	174
6.3 Implications of the research for policies about DDE.....	175
6.4 Implications of the research for clinical care of patients with DDE.....	177
6.5 Recommendations for research about DDE	177
6.6 General implications	179
6.6.1 Implications for research with children.....	179
6.6.2 Implications for research on OHRQoL with children.....	180
7. Chapter Seven.....	182
Conclusions and Recommendations	182
7.1 Summary of findings.....	182
7.2 Recommendations for policies about DDE.....	183
7.3 Recommendations for clinical care of patients with DDE.....	183
7.4 Recommendations for research about DDE	183
7.5 Recommendations for research with children	184
7.6 Recommendations for research on OHRQoL with children	184
8. Chapter Eight	188
References.....	188
9. Chapter Nine.....	209
Appendices.....	209
9.1 Appendix A List of dental journals included in systematic review	209
9.2 Appendix B Child Perceptions Questionnaire for 11-14 year olds	214
9.3 Appendix C Clinical Data Collection Sheet.....	229
9.4 Appendix D Evaluation of P-CPQ using different approaches to the analysis of ‘don’t know’ responses.....	231
9.5 Appendix E Information sheets and consent forms for Chapter Four	240
9.6 Appendix F Topic Guide.....	244
9.7 Appendix F Information sheets and consent forms for Chapter Five.....	246
9.8 Appendix G Publications arising from this thesis	252

1. Chapter One Introduction

1.1 Background

Developmental defects of enamel (DDE) have been defined as:

‘disturbances in hard tissue matrices and in their mineralisation arising during odontogenesis’ (Commission on Oral Health 1982).

Over ninety different aetiological factors have been associated with DDE of the permanent dentition, including both localised and generalised causes. Much research, usually conducted in children and young people, has been aimed at the aetiology of DDE, particularly of fluoride.

Several authors have discussed whether DDE are a public health problem (Burt and Eklund 1999; Martinez-Mier *et al.* 2004; Whelton *et al.* 2004; Cutress *et al.* 2006) and have applied a variety of criteria in different countries to answer this question. Sheiham and Watt (2003) suggested the following criteria for determining the public health significance of a health condition:

- Prevalence
- Impact on individuals
- Impact on wider society
- Condition is preventable with effective treatments available

To consider whether DDE are a public health problem in the UK, this thesis reviews the literature for each of the above criteria and identifies gaps in the current evidence. One significant gap is the impact DDE have on individual children and young people. Little research appears to have been conducted to gain their perspectives on DDE generally, with no research on the impact of DDE in the UK. There have been repeated recommendations for further research in this area (Lalumandier and Rozier 1998; Milsom *et al.* 2000; Medical Research Council Working Group Report 2002; Sigurjons *et al.* 2004; Sujak *et al.* 2004). In addition, the small number of existing studies have highlighted important methodological considerations that should be taken into account in future investigations.

Research into the impact of any condition requires an approach that recognises both the biological and psychosocial components of health. Current approaches to investigating the impact of conditions include the use of complementary quantitative and qualitative methods. Quantitative assessments, using oral health related quality of life (OHRQoL) measures, provide an overall description of the impact of a condition, whereas qualitative methods enable a more in-depth detailed exploration, from the individual's perspective on the meaning of the condition to their everyday lives. However, contemporary thinking on research with children and the apparent lack of children's perspectives on DDE demands further analysis of the extent to which research has considered their perspectives, to ensure that future research is child-centred, rather than purely describing 'what adults think children think' (Alderson 1995).

In conclusion, there has been little attention paid to the impact of DDE on children and young people and much of this work may have been restricted methodologically. Accurate information about the impact of DDE is required to determine whether they constitute a public health problem.

1.2 Aim and objectives

The aim of this thesis is to describe the impact of DDE on young people.

The objectives of the research are to:

- 1) Describe the extent to which contemporary dental research on DDE has included the perspective of individual children and young people.
- 2) Describe the impact of DDE on the OHRQoL of young people.
- 3) Explore, in detail, the impact of DDE on young people.

This thesis is structured as follows:

Chapter Two is a narrative review of the literature on DDE, including the various factors associated with them. It then considers their significance as a public health problem in terms of prevalence, impact on the individual and on society and also the prevention and availability of treatments for DDE. Gaps in the literature are highlighted, particularly concerning the impact of DDE on the individual. A detailed description of current approaches to researching the impact of dental conditions and conducting research with children is presented. The chapter concludes by presenting the rationale, aims and objectives of the research described in this thesis.

Chapter Three details a systematic review of the extent to which the perspectives of children and young people have been taken into account in DDE research. This chapter also makes suggestions on the benefits of research *with*, rather than *on*, children.

Chapter Four reports on a quantitative study, using a child-specific OHRQoL measure, to investigate the impact of DDE on young people. As well as describing levels of impact associated with DDE, an evaluation of the properties of this measure is included.

Chapter Five provides a complementary, detailed, qualitative exploration of the impact of DDE on young people, from their own perspectives. Symbolic interactionism is used as the theoretical framework to guide this study.

Chapter Six brings together the findings of the studies contained within the thesis and discusses their implications for policies about DDE, for the clinical care of patients with DDE and more general implications for research with children. Recommendations for research are also outlined.

Chapter Seven highlights the major conclusions and summarises recommendations for clinical care, policy and research.

2. Chapter Two Literature Review

2.1 Introduction

This chapter first describes the background and aetiology of developmental defects of enamel (DDE). It then considers their significance as a public health problem, against four criteria (Sheiham and Watt 2003):

- Prevalence
- Impact of the condition on the individual
- Impact of the condition on society
- Prevention and availability of effective treatments

Gaps in the current literature regarding these criteria are highlighted, particularly those concerning the impact of DDE on the individual. The second section details the approaches available to investigating the impact of dental conditions and concludes by describing the rationale, aims and objectives of the research undertaken for this thesis. Box 1 provides an outline of the chapter.

Box 1. Outline of Chapter

- Background to developmental defects of enamel (DDE)
- Significance of DDE as a public health problem against four criteria:
 - Prevalence
 - Impact of the condition on the individual
 - Impact of the condition on society
 - Prevention and availability of effective treatments
- Approaches available to investigate the impact of dental conditions
- Rationale
- Aims and objectives

2.2 Developmental Defects of Enamel (DDE)

Abnormalities of enamel are commonly referred to as developmental defects of enamel, enamel opacities, enamel hypoplasia, discoloured enamel and dental fluorosis (Clarkson and O'Mullane 1989). These terms are often used interchangeably (Cutress *et al.* 2006), even though they have specific definitions:

Enamel opacities:

‘a qualitative defect of enamel identified visually as an abnormality in the translucency’

Enamel hypoplasia:

‘a quantitative defect of enamel visually and morphologically identified as involving the surface of the enamel and associated with a reduced thickness’

Discoloured enamel:

‘an obvious abnormal appearance of the enamel excluding coloured opacities’

Developmental defects of enamel:

‘disturbances in hard tissue matrices and in their mineralisation arising during odontogenesis’ (Commission on Oral Health 1982)

Dental fluorosis:

‘specific disturbance of tooth formation caused by excessive intake of fluoride during the formative period of the dentition’ (Moller 1982)

The term ‘DDE’ will be used in this thesis to refer to enamel abnormalities generally, as the definition covers both qualitative and quantitative defects. The term ‘fluorosis’ will be used when the cause of the defect can be ascribed to fluoride.

2.3 Aetiology of DDE

Over ninety different aetiological factors have been associated with DDE of the permanent dentition, mostly reported in review articles or case studies. Localised causes include trauma or infection (Winter and Brook 1975) and generalised causes include genetic or systemic influences (Winter 1997) (Table 1).

Table 1. Aetiology of abnormal enamel in permanent teeth

Examples	
Localised causes	
Trauma	Intrusion or avulsion of deciduous tooth
Infection	Periapical infection of deciduous tooth
Generalised causes	
Genetic:	
Confined to dental structures	Amelogenesis imperfecta
Associated with systemic disease	Epidermolysis bullosa, Pseudohypoparathyroidism
Systemic:	
Nutritional deficiencies	Malnutrition
Infections	Gastric, respiratory
Metabolic disorders	Vitamin-D dependent rickets, Hypoparathyroidism
Idiopathic	Molar-incisor hypomineralisation
Substances	Tetracycline, fluoride

2.3.1 Localised causes

Localised DDE of the permanent dentition can be due to trauma and infection of deciduous teeth. Injuries to the deciduous teeth commonly occur between the ages of 18 months to 4 years, affecting 19% of children (Cutress *et al.* 2006). The frequency of abnormalities of permanent teeth, secondary to such injuries, ranges from 12% to 69% (Ravn 1968; Selliseth 1970; Sennhenn-Kirchner and Jacobs 2006). Depending on the age of the child and the type of injury to the deciduous tooth (for example, intrusion, subluxation or avulsion) different abnormalities of the permanent tooth are observed. Injuries sustained in children aged 18 months or younger tend to disturb tooth morphology as well as enamel formation (Sennhenn-Kirchner and Jacobs 2006). Periapical infections of the deciduous tooth, consequent to trauma or caries, can, if long-standing, cause defects of the developing permanent tooth germs (Winter 1997; Cordeiro and Rocha 2005).

DDE due to localised causes usually affect small numbers of teeth and appear as well-demarcated white/yellow opacities or hypoplastic areas.

2.3.2 Generalised causes

Generalised causes affect the structure and appearance of teeth, due to genetic or systemic influences.

2.3.2.1 Genetic influences

Abnormal enamel may result from genetic disorders, either as a feature of a syndrome or from a single gene abnormality confined to enamel.

Various DDE have been linked to syndromes such as epidermolysis bullosa, pseudohypoparathyroidism and oculodentodigital dysplasia. These syndromes are relatively uncommon; epidermolysis bullosa and pseudohypoparathyroidism occur in approximately 49 (Horn *et al.* 1997) and 3.4 people per million population respectively (Nakamura *et al.* 2000). Only about 100 cases of oculodentodigital dysplasia have been reported since the syndrome was characterised in 1957 (Gorlin 1994). Pitted hypoplastic enamel is characteristic of all these syndromes (Winter and Brook 1975).

Abnormalities of enamel without generalised genetic conditions are collectively known as amelogenesis imperfecta (Winter 1997). Many different classification systems have been described, based on the mode of inheritance and type of defect. Generally, these conditions are a result of a single gene mutation, due either to autosomal dominant, autosomal recessive or X-linked patterns of inheritance. The abnormal enamel may be hypoplastic, hypomineralised or both (Crawford *et al.* 2007). Occurrence of amelogenesis imperfecta ranges from 1 in 700 to 1 in 14,000 depending on the populations studied (Winter 1997). The appearance of the enamel varies by type of defect, ranging from thin, shiny and discoloured enamel in Hypoplasia Type IA, to Hypomaturation Type IIIC, where the appearance of the incisor teeth is described as 'snow-capped' due to the opaque areas of affected enamel limited to the incisal edge (Winter and Brook 1975).

2.3.2.2 Systemic factors

Systemic or chronological DDE of permanent teeth have been associated with a range of conditions occurring during the neo-natal period and in the early years of life (Winter 1997). Malnutrition has been linked to DDE in children from developing countries

(Rugg-Gunn *et al.* 1997) but with complex relationships postulated between nutrition, gastric and respiratory infections and socio-economic status (Correia Sampaio *et al.* 1999). Other infections implicated are childhood viruses such as chickenpox and mumps (Cutress *et al.* 2006). Metabolic disorders, particularly those affecting calcium homeostasis, such as Vitamin-D dependent rickets and hypoparathyroidism, have also been associated with DDE (Nikiforuk and Fraser 1981; Walls and Soames 1993; Zambrano *et al.* 2003).

A specific type of systemic DDE that features hypomineralised molars and incisors was named Molar-Incisor Hypomineralisation in 2001 (Weerheijm *et al.* 2001). The aetiology of this condition remains unknown, although infections, dioxins in breast milk and possibly genetic influences have been considered (Weerheijm and Mejare 2003).

DDE due to these systemic factors range from hypomineralised enamel to more severely hypoplastic pitted or grooved presentations. The timing of the upset can sometimes be estimated by the chronology of the enamel changes.

Finally, ingestion of substances, including tetracyclines and fluoride, can cause DDE. Prolonged or recurrent use of tetracyclines has been linked to discolouration of enamel and dentine since the 1960s. The severity of the defects depend on the drug type, dose, number of courses and age of the child (Tredwin *et al.* 2005). The appearance of the affected teeth vary from yellow discolouration with tetracycline to grey-brown discoloration with oxytetracycline (Driscoll *et al.* 1993).

The link between DDE and fluoride was first suggested in the 1930s and stemmed from work by Dr Frederick McKay (Murray *et al.* 1991). It was confirmed by Dean, who observed an increase in the proportion of children with DDE as the concentration of fluoride in the drinking water increased (Dean *et al.* 1938; Dean and McKay 1939). Dean then introduced fluoridation to Grand Rapids, Michigan, by adding 1 part fluoride per million parts of water (1ppm F) to the water supply and showed a reduction in caries levels (Arnold *et al.* 1956).

The main effect of fluoride on teeth occurs during the maturation phase of enamel development (Fejerskov *et al.* 1990). The appearance of fluorosis is associated with cumulative fluoride ingestion during amelogenesis, but the severity depends on the

dose, timing and duration of the intake. The central incisors are considered to be most at risk of fluorosis between the ages of 21 and 30 months for females and 15 to 24 months for males (Evans and Stamm 1991). Any source of fluoride available systemically during amelogenesis can cause fluorosis (Ophaug and Singer 1988).

2.3.3 Aetiology of fluorosis

The association between the use of fluorides advocated by dental professionals and dental fluorosis has received significant attention.

2.3.3.1 Sources of fluoride

2.3.3.1.1 Water fluoridation

Several studies have investigated water fluoridation as a risk factor for fluorosis (Ismail *et al.* 1990; Riordan and Banks 1991; Riordan 1993; Heller *et al.* 1997) with odds ratios (OR) for fluorosis ranging from 2 to 8.5. The York Review, a systematic review of the safety and effectiveness of water fluoridation, found fluoride levels to have a significant relationship with fluorosis (OR = 2.1) (McDonagh *et al.* 2000). The use of fluoridated water to mix infant formula milk has also been suggested as a risk factor for fluorosis, although this only appears to be a risk for prolonged use in children over 12 months of age (Osuji *et al.* 1988; Clark 1994; Browne *et al.* 2005).

It has been estimated that 60% of the total prevalence of fluorosis is attributable to fluoride sources other than from drinking fluoridated water (Whelton *et al.* 2004).

2.3.3.1.2 Fluoride toothpaste

Toothpaste is a very widespread source of fluoride (Murray *et al.* 1991). The fluoride compounds and concentrations found in toothpastes vary both between brands and between countries. The usual concentration is 1000-1500 ppm F, with higher (over 2000 ppm F) and lower (less than 600 ppm F) formulations available (Levine *et al.* 2004). Many studies have investigated fluoridated toothpaste use as a risk factor for fluorosis (Osuji *et al.* 1988; Milsom and Mitropoulos 1990; Holt *et al.* 1994; Rock and Sabieha 1997; Mascarenhas and Burt 1998; Tabari *et al.* 2000; Conway *et al.* 2005; Franzman *et al.* 2006), including the age at which brushing commenced, the frequency of brushing, the concentration of the toothpaste, the amount used and swallowed. No conclusions as to the relative importance of these factors can be made, as the findings of these studies are inconsistent. In addition, it has been suggested that socio-economic

status is associated with fluorosis and use of fluoridated toothpaste but, again, the evidence is inconsistent (Milsom and Mitropoulos 1990; Hamdan and Rock 1991; Nunn *et al.* 1994). Tabari and colleagues found children from more deprived areas to be at lower risk of developing fluorosis and suggested this may be due to children from less deprived areas starting the use of toothpaste earlier or with more frequency (Tabari *et al.* 2000).

2.3.3.1.3 Fluoride supplements

The use of fluoride supplements (tablets and drops) began in the 1950s, with different countries having different dosing regimens based on child's age, weight, caries-risk and fluoride levels in the water. Again, many studies have investigated fluoride supplements as a risk factor for fluorosis. A meta-analysis of 14 such studies reported that ORs of regular supplement users, compared to non-users, ranged from 2.4 to 2.6 (Ismail and Bandekar 1999). The authors estimated the odds of developing fluorosis when combining the use of supplements and toothpaste during the first year of life compared to the use of toothpaste alone and found the odds increased from 1.5 (for toothpaste alone) to 6.2 (for toothpaste with supplements).

2.3.3.1.4 Other sources of fluoride

Other possible sources of fluoride include gels and varnishes, which, although applied topically, can be swallowed. Little is known about the risk of fluorosis in humans from these sources.

In summary, fluoride ingestion as a result of preventive strategies is a cause of DDE. However, the research on risk factors for fluorosis has tended to be retrospective and univariate, without accounting for potential confounding. In addition, studies requiring parents to supply information on infant-rearing and dental health practices are subject to recall bias.

2.4 Are DDE a public health problem?

Several authors have attempted to answer this question for DDE generally, but particularly for fluorosis (Burt and Eklund 1999; Martinez-Mier *et al.* 2004; Whelton *et al.* 2004; Cutress *et al.* 2006). Establishing whether fluorosis is a public health problem is important to contribute to risk-benefit assessments of fluoridation, namely comparison of the effectiveness of fluoride at reducing caries to the increased risk of fluorosis.

Cutress and colleagues considered whether DDE were a public health problem in terms of aesthetics and need for treatment in children. They were not able to reach a conclusion because of the complexity of considering the type of defect, the size, the number of teeth involved, individual's interest in appearance and children's age. They recommended further research on ways of measuring the impact of the aesthetics of DDE.

Burt and colleagues (1999) felt that, while fluorosis was a public health problem in some countries of East Africa and in areas of India, it could not be classified as a public health problem in the US because, although prevalent, most fluorosis was of the mildest forms. They recognised that changes in the public's concern about aesthetics could increase the potential for it to become a public health problem in the future. In Mexico, fluorosis has been deemed a possible public health problem due to the impact of the condition and, again, the importance of physical appearance in society was felt to be an important determining factor (Martinez-Mier *et al.* 2004). In Europe, fluorosis, at current levels of prevalence and severity, is not considered a public health problem, although more detailed consideration of the impact of the aesthetics of fluorosis is recommended (Whelton *et al.* 2004).

The deliberations of these authors used varying criteria, particularly prevalence and severity, to determine whether DDE are a public health problem, with most alluding to the aesthetics of the condition and its consequent impact. However, no data were given about the impact of DDE. Several authors recommended future research in this area (Lalumandier and Rozier 1998; Sigurjons *et al.* 2004).

Sheiham and Watt (2003) suggested the following criteria for determining the public health significance of a health condition:

- Prevalence of the condition
- Impact of the condition on individual
- Impact on wider society
- Condition is preventable and effective treatments are available

For a condition to be considered a public health problem, the prevalence should be high, or if uncommon, the condition should be serious. The condition should have an impact on an individual in terms of symptoms, functioning, psychological and social considerations. The condition should have a societal impact due to the costs of treatment or time missed from school/work for that population. Finally, the condition should be preventable or treatable (Sheiham and Watt 2003).

The next section of this review attempts to assess the extent to which DDE can be considered a public health problem against these criteria. While the review of the literature focuses on current UK data, data from elsewhere are used if helpful.

2.4.1 Prevalence

The first criterion for a public health problem concerns the prevalence of the condition, including trends in prevalence over time.

2.4.1.1 Prevalence of DDE in the UK

Most studies describe the prevalence of DDE in children and young people. From the UK Children's Dental Health Surveys, the proportion of 12-year-olds with DDE on their anterior teeth was 34% in 2003 and 36% in 1993 (Table 2). The most common types were demarcated (17%) and diffuse defects (16%) (O'Brien 1994; Chadwick and Pendry 2004). The vast majority (92%) of the demarcated defects were confined to less than one third of the tooth surface, compared to 59% for diffuse defects. Of the diffuse defects, 65% were symmetrical, which the authors suggested could be attributed to fluoride. The central incisors were the teeth most commonly affected by any type of defect (Chadwick and Pendry 2004). The findings from the national surveys are compatible with those of local studies in the UK (Table 2).

Table 2. Prevalence of DDE in UK from national and local surveys

Area	Author	Year	Prevalence (%)
National Children's Dental Health Survey	(Chadwick and Pendry 2004)	2003	34
North West	(Milsom <i>et al.</i> 2000)	1996/7	34
North West	(Ellwood and O'Mullane 1996)	1993	36
National Children's Dental Health Survey	(O'Brien 1994)	1993	36
Midlands	(Elley and Charlton 1992)	1991	36
North West	(Milsom and Mitropoulos 1990)	1988	44
North West	(Clarkson and O'Mullane 1989)	1987	30
South Yorkshire	(Clerehugh 1979)	1976	36

2.4.1.1.1 Prevalence of fluorosis

The prevalence of fluorosis was estimated at different levels of fluoride in water in the York Review. At 0.1 ppm F prevalence was estimated to be 15%. At 1ppm F, it was 48% and for fluorosis of 'aesthetic concern', 12.5% (McDonagh *et al.* 2000). The estimated numbers of additional people who have to be exposed to water fluoridation at 1ppm F for one additional person to develop fluorosis of any level was 6 and of 'aesthetic concern' was 22.

The Review's estimates came from 88 studies (83 of which were cross-sectional) from 30 different countries. Forty studies came from the UK. Study areas above 5ppm F were excluded. One study was judged to be of moderate quality, with the others being of the lowest level of evidence. The two main faults were failure to control for confounding factors (such as temperature and altitude) and reduction of observer bias. The level of fluorosis chosen to be of 'aesthetic concern' was taken from a study by Hawley and colleagues (Hawley *et al.* 1996) based on young people's ratings of the

appearance of photographed teeth, rather than the experiences of those with the condition.

The term 'fluorosis' was used in the Review, but it stated that DDE other than those caused by fluoride could have been measured in the included studies, leading to the likelihood of an overestimation of the prevalence of fluorosis.

2.4.1.1.2 Prevalence of fluorosis in the UK

Since the York Review, several UK studies have further investigated the prevalence of fluorosis specifically. In the North East, the prevalence in children aged 8 or 9 years in a fluoridated area was 54% compared to 23% in an area without fluoride in the water (Tabari *et al.* 2000). In Ireland the prevalence in 12-year-olds in the fluoridated Republic of Ireland was 30% compared to 21% in non-fluoridated Northern Ireland (Whelton *et al.* 2006).

The Medical Research Council's Report on water fluoridation and health recommended further studies to investigate the prevalence of fluorosis in fluoridated and non-fluoridated areas, controlling for confounding factors (Medical Research Council Working Group Report 2002).

2.4.1.2 Trends in the prevalence of DDE

No clear trend can be observed for changes in the prevalence of DDE in the UK (Holloway and Ellwood 1997). Elsewhere there is some weak evidence of a slight increase in the prevalence of fluorosis in North America and the Republic of Ireland (O'Mullane *et al.* 1986; Rozier 1999; Whelton *et al.* 2001). The York Review investigated trends in prevalence from 32 water fluoridation studies conducted in nine countries and based on the percentage prevalence found no increase in fluorosis over time (McDonagh *et al.* 2000). The studies included were of the lowest quality of evidence.

2.4.1.3 Measurement of the prevalence of DDE

The estimates of the prevalence of DDE described above are based on epidemiological surveys, involving clinical examinations, using various indices. Many indices have been developed to record the prevalence and severity of DDE. Two types of indices, 'specific' and 'descriptive', have emerged, distinguished by the ascription of aetiology from clinical appearance. Specific indices consider only DDE associated with fluoride.

They are favoured by those who believe it is possible to differentiate between fluoride and non-fluoride induced defects. Clearly, descriptive indices do not attempt to ascribe aetiology but rather record the full range of DDE (Ellwood *et al.* 1994). They are used by those who believe that the clinical diagnosis of fluorosis is problematic, due to the similarities with other DDE not induced by fluoride.

2.4.1.3.1 Fluorosis-specific indices

Dean's Index of Fluorosis and Thystrup and Fejerskov Index (TFI) have been the most widely used indices designed specifically to measure fluoride-induced defects, and to distinguish them from DDE due to other causes.

2.4.1.3.1.1 Dean's Index of Fluorosis

Dean developed his original classification so he could map, in a given area, the fluoride content of the drinking water and the severity of 'mottling' (Dean 1934). His original index involved examining individuals in 'good natural light', to establish if defects were present. Children with defects other than mottling were classified as 'normal', thereby attempting to restrict the index to measure only fluorosis. If fluorosis was found, the individual was classified according to the clinical appearance of the second most severely affected tooth in the mouth. Each child was given a 'mouth classification', using one of the seven categories described in Table 3. Later modifications reduced the number of categories to six, by removing 'moderately severe' (Dean *et al.* 1942).

Table 3. Dean's Index (Dean 1934)

Category	Score	Criteria
Normal	0	Normal calcification and hypoplasia other than mottling of enamel
Questionable	0.5	Slight aberrations in the translucency of normal enamel on teeth of those continuously resident in areas with fluoride in the water supply
Very mild	1	Small opaque paper white areas scattered irregularly over the surface
Mild	2	White, opaque areas on the surfaces of the teeth involving at least half of the tooth surface
Moderate	3	White, opaque areas on the surfaces of the teeth involving all the tooth surfaces
Moderately severe	4	Smoky white appearance or brown stain and pitting
Severe	5	Pits are deeper and often confluent, stains are widespread.

There are several limitations of Dean's Index. First, Dean acknowledged the vague description of the 'questionable' category and suggested that distinguishing between 'very mild' and 'mild' required experience (Dean 1934). Second, it is based on the two most severely affected teeth so does not allow for the measurement of the extent of the fluorosis on the remaining teeth, although the simplicity of this approach aids use in the field (Clarkson 1989). Third, it tells us nothing of what people think about fluorosis.

Several studies have reported the examiner reliability of Dean's Index. Inter-examiner agreement was good ($k = 0.75-0.94$) (Kumar *et al.* 2000). Kappa values of 0.89-0.98 were obtained for intra-examiner reliability (Mabelya *et al.* 1994) indicating substantial reliability (Landis and Koch 1977). Dean's Index remains one of the most widely used fluorosis-specific indices (Rozier 1994). A search of electronic databases (Medline via Ovid and Embase) revealed over fifty studies worldwide that have used Dean's Index to measure prevalence of fluorosis, often comparing prevalence in fluoridated and non-fluoridated communities. This search identified only two published studies in the UK (Forrest 1956; Whelton *et al.* 2006).

2.4.1.3.1.2 Thystrup and Fejerskov Index (TFI)

Thystrup and Fejerskov believed that distinctions could be made about the aetiology of defects in the vast majority of instances and that a specific fluorosis classification was justified (Thylstrup and Fejerskov 1978). They identified the need for an index that was based on the histopathological appearance of enamel.

Thystrup and Fejerskov examined 120 children, with varying degrees of fluorosis, from an area of Tanzania with 3.5 ppm F in the water supplies. Ten classes of macroscopic enamel changes were identified. Extracted teeth were then sampled and histological sections representing each of the 10 categories were prepared. The degree of microscopic changes corresponded to the visible macroscopic changes so validating the histological findings. Drying extracted teeth was found to make the perichymata more pronounced. Therefore, the authors recommended drying teeth with cotton-wool and examining in daylight, using a plane mirror and probe. Occlusal and buccal/lingual surfaces were classified. Enamel opacities not due to fluoride were ignored. Table 4 describes the 10-point system of TFI. Data are expressed as the proportion of a community with each TFI score.

Table 4. Thystrup and Fejerskov Index (Thylstrup and Fejerskov 1978)

Score	Clinical Appearance
0	Normal translucency of enamel remains after prolonged air drying
1	Narrow white lines located corresponding to the perikymata
2	Smooth surfaces - more pronounced lines of opacity which follow the perichymata. Occasionally confluence of adjacent lines. Occlusal surfaces - scattered areas of opacity <2mm in diameter and pronounced opacity of cuspal ridges
3	Smooth surfaces - merging and irregular areas of opacity. Accentuated drawing of perichymata often visible between opacities. Occlusal surfaces - confluent areas of marked opacity. Worn areas usually circumscribed by a rim of opaque enamel
4	Entire surface exhibits marked opacity
5	Entire surface exhibits marked opacity with focal loss of outermost enamel (pits) <2mm in diameter
6	Smooth surfaces - pits are regularly arranged in horizontal bands < 2mm in vertical extension Occlusal surfaces - confluent areas <3mm in diameter exhibits loss of enamel. Marked attrition
7	Smooth surfaces - loss of outermost enamel in irregular areas involving less than one-half of entire surface Occlusal surfaces - changes in the morphology caused by merging pits and marked attrition
8	Smooth and occlusal surfaces - loss of outermost enamel involving > half of surface
9	Smooth and occlusal surfaces - loss of main part of enamel with change in anatomic appearance of surface. Cervical rim of almost unaffected enamel is often noted

The index was later modified to be based solely on examination of the buccal tooth surfaces and with a shorter scale (Fejerskov *et al.* 1990), although the original index remains the most widely used.

Assessments of the reliability of the index revealed a kappa value of 0.70 (Mabelya *et al.* 1994) and 0.58 (Ellwood *et al.* 1994) for inter-examiner agreement and 0.94-0.99

(Mabelya *et al.* 1994) and 0.73 (Tabari *et al.* 2000) for intra-examiner agreement indicating moderate agreement.

TFI is more precise than Dean's Index, because of the increased number of categories. For example, in areas of 6 ppm F, the most frequently observed category using the original index was score 7 and in areas of 21 ppm F, it was score 8. Using Dean's Index, both these degrees of fluorosis would be classified as 'severe' (Thylstrup and Fejerskov 1978). However, difficulties interpreting these categories have led to the value of this greater precision being questioned (Horowitz 1986). Another significant limitation is that TFI is based purely on a normative assessment of teeth and their structure.

TFI has been widely used. Many studies have also compared Dean's Index with TFI (Burger *et al.* 1987; Cleaton-Jones and Hargreaves 1990; Zietsman 1991; Mabelya *et al.* 1994). A comparison in Tanzania found TFI revealed more fluorosis than Dean's Index in communities with minor and moderate fluorosis but the two were comparable in communities with severe fluorosis (Mabelya *et al.* 1994). TFI has been widely used in the UK (Hamdan and Rock 1991; Holt *et al.* 1994; Ellwood and O'Mullane 1995; Ellwood and O'Mullane 1996; Hawley *et al.* 1996; Stephen *et al.* 2002; Tavener *et al.* 2004; Tavener *et al.* 2006) either alone or in combination with another index.

2.4.1.3.1.3 Visual analogue scale for fluorosis

More recently, a visual analogue scale (VAS) for measuring fluorosis severity has been developed, to overcome perceived disadvantages of the existing ordinal scales. Calibrated examiners score the worst affected tooth on a 100mm scale from 'best you can imagine' to 'worst you can imagine'. Inter- and intra-examiner agreement was good (Intra-class correlation coefficient (ICC) = 0.79 and ICC = 0.88-0.97) (Vieira *et al.* 2005). However, the VAS was applied by examiners based on their perceptions of the severity of the fluorosis, without considering the perceptions of those affected.

2.4.1.3.2 Descriptive indices

While some researchers favour using specific measures, others argue that making a diagnosis of fluorosis is problematic and favour an approach that relies purely on the description of the appearance of the teeth.

2.4.1.3.2.1 Federation Dentaire Internationale Index: Developmental Defects of Enamel

A lack of a well-defined and internationally accepted classification of enamel defects was identified by a Working Group of the Federation Dentaire Internationale Commission on Oral Health, Research and Epidemiology. This group deemed as inappropriate those classifications that presumed the aetiology of defects and thought indices should be based on descriptive criteria.

Developmental Defects of Enamel Index (DDEI) was designed to record the type, number, demarcation and location of defects with flexibility for recording data on a person, tooth or tooth surface basis as required (Table 5).

The index can be used, with or without a probe, to explore the tooth and lighting can be natural or artificial, depending on the 'field conditions'. The Working Group recommended that, ideally, teeth should be professionally cleaned and dried before examination. However, recognising that this may not be practicable, they suggest that the inspection conditions should be noted.

Table 5. Federation Dentaire Internationale Index: Developmental Defects of Enamel Index

	Codes for scoring
Types of defect: Normal	0
Opacity: White/cream	1
Yellow/brown	2
Hypoplasia: Pits	3
Grooves - horizontal	4
Grooves - vertical	5
Missing enamel	6
Discoloured enamel not associated with opacity	7
Other defects	8
Number and demarcation of defects: Single	1
Multiple	2
Diffuse, fine white lines	3
Diffuse, patchy	4
Location of defects: Gingival one-half	1
Incisal one-half	2
Gingival and incisal halves	3
Occlusal	4
Cuspal	5
Whole surface	6
Other combinations	7

No standard method of measuring the reliability of DDEI has been published. However, agreement between the examiners proved 'generally satisfactory' when the extent of the disagreements was examined (Clarkson *et al.* 1988).

Criticisms of DDEI include:

- Generation of large amounts of data that are difficult to both analyse and present in a meaningful way (Clarkson and O'Mullane 1989). For example, results from studies using DDEI have been presented as a percentage of:
 - white opacities: demarcated single, demarcated multiple, diffuse horizontal parallel lines, diffuse patchy distribution.
 - yellow opacities: demarcated single, demarcated multiple.
 - hypoplasia: pits, groove horizontal, grooves vertical etc.

Some have chosen to record and analyse results from all surfaces but have presented data for the labial surfaces of incisors separately (Suckling and Pearce 1984; Clarkson and O'Mullane 1992).

- It does not identify the most common types of defect (Clarkson and O'Mullane 1989).
- The inability of the index to record the extent of the defects, that is the area of the tooth surface affected (Clarkson and O'Mullane 1989).
- Comparison of results obtained with DDEI and other indices and between results obtained with DDEI by several investigators can be difficult. The use of different light sources and examining teeth either wet or dry contributes to these comparability problems (Clarkson and O'Mullane 1992).

In response to these criticisms, a modified DDEI was proposed. It had a revised scoring system, placed less emphasis on the colour of the defect and included a method of recording severity (Table 6)(Clarkson and O'Mullane 1989).

Table 6. The Modified DDE Index-general purpose (Clarkson and O'Mullane 1989)

Type of defect	Codes for scoring
Normal	0
Demarcated opacities:	
White/cream	1
Yellow/brown	2
Diffuse opacities:	
Lines	3
Patchy	4
Confluent	5
Confluent/patchy + staining + loss of enamel	6
Hypoplasia:	
Pits	7
Missing enamel	8
Any other defects	9
Extent of Defect:	
Normal	0
<1/3	1
At least 1/3 <2/3	2
At least 2/3	3

The recommended conditions for use of the modified DDEI were for teeth to be examined wet and under artificial light. The authors emphasised that the index should be used flexibly, depending on the objectives of the study, and that investigators could interchange components of the index to suit their requirements. The authors acknowledged that, like the original version of the index, the general purpose version of the modified DDEI also results in the generation of large amounts of data that prove difficult to analyse. The modified DDEI was thus simplified further, for use in surveys (Table 7)(Clarkson and O'Mullane 1989). However, still there is no consideration of the affect of the condition on the individual.

Table 7. Modified DDE Index-surveys (Clarkson and O'Mullane 1989)

	Codes for scoring
Normal	0
Demarcated opacities	1
Diffuse opacities	2
Hypoplasia	3
Other defects	4

The survey version of the modified index categorises three broad types of defect: diffuse opacities, demarcated opacities and hypoplasias. Data are usually presented as the percentage of diffuse and demarcated defects. Diffuse opacities are broadly considered to encompass those defects thought to be due to fluoride (Holloway and Ellwood 1997) and discriminate between fluoridated and non-fluoridated areas (Clarkson and O'Mullane 1989). However, as diffuse opacities may have other, less common origins, their prevalence can only be considered an approximation to that of fluorosis.

Reliability assessment of the survey version of the modified DDEI has been attempted; kappa values at the level of the tooth were 0.78. At the level of the mouth for demarcated defects the kappa value was 0.80 and for diffuse defects it was 0.94 (Ellwood *et al.* 1994). This indicates substantial agreement.

The survey version of the modified DDEI is the most widely used index in the UK to study enamel defects (Holloway and Ellwood 1997). It has mainly been used to assess their prevalence (Elley and Charlton 1992; Nunn *et al.* 1992; Nunn *et al.* 1993; Downer *et al.* 1994; Holt *et al.* 1994; Milsom *et al.* 1996; Tavener *et al.* 2004) as it does not lend

itself to recording severity. It was used in the national Children's Dental Health Surveys (O'Brien 1994; Office for National Statistics 2004).

Other descriptive measures (Al-Alousi *et al.* 1975; Murray *et al.* 1984) have not been widely reported.

2.4.1.3.2.2 Use of photographic techniques

In addition to direct scoring of participant's teeth, specific and descriptive indices have also been applied to photographs of teeth. The use of photographs reduces the observer bias when comparing the prevalence of defects in fluoridated and non-fluoridated areas. Despite some early concerns about the modification of contrast between normal and defective enamel when using photographic techniques (Ellwood *et al.* 1994) more recent studies have found clinical and photographic data to agree closely (Tabari *et al.* 2000).

A standardised photographic method was developed to reduce the variation in technique of equipment (camera, lens, film), lighting and conditions (Cochran *et al.* 2004). When reliability was assessed from seven European study sites, kappa values for intra-examiner reliability of the transparencies when scored using TFI were 0.45-0.6 and 0.43-0.7 when using the simplified modified DDEI. For inter-examiner reliability, the values were 0.32-0.55 (TFI) and 0.34-0.69 (modified DDEI). This technique has been advocated to allow comparison of the prevalence of enamel opacities from different study sites and for longitudinal studies and has since employed a digital camera using both TFI and the modified DDEI (Tavener *et al.* 2004). The Medical Research Council Report recommended that future research on the prevalence of fluorosis should involve photographic techniques (Medical Research Council Working Group Report 2002).

2.4.1.3.3 Summary and conclusion

DDE are a common condition, affecting approximately one-third of children in the UK. The prevalence of fluorosis varies between fluoridated and non-fluoridated areas. No clear trends in prevalence have been established. Many studies have investigated the prevalence of DDE in the UK and worldwide, mainly in children and young people, rather than adults.

The measurement of DDE has been complicated by controversy about the appropriateness of ascribing aetiology to fluoride-induced defects. Fluorosis-specific indices have been widely used in studies of the effect of water fluoridation or other sources of fluoride on the prevalence of defects but, as fluorosis is only one of many causes of DDE, this approach has been criticised. In response, some researchers have taken a pragmatic approach and developed descriptive indices based on characteristics of the appearance of teeth. While these indices avoid the difficulties of presuming aetiology, they have limitations, particularly in amenability to analysis. More recently, rather than entering into such debates, DDE has been measured by both TFI and the modified DDEI, using a standardised photographic technique (Cochran *et al.* 2004; O'Mullane *et al.* 2004).

Prevalence is, however, just one of the criteria for determining the public health significance of DDE. While the current literature suggests DDE are a common condition in the UK, to establish the importance of this, the literature on the impact of the condition on the individual also needs to be considered. In terms of fluorosis, the Medical Research Council highlighted the need to supplement customary clinical outcomes with measures of impact on individuals, to assist in risk-benefit assessment of fluoridation (Medical Research Council Working Group Report 2002).

2.4.2 Impact of the condition on the individual

The second criterion to establish whether a condition is a public health problem is its impact on individuals. The most common method used for assessing the impact of conditions are health-related quality of life measures (Bowling 1997; Brondani and MacEntee 2007). Important considerations of such measures are that: they are based on an explicit model of health, have been evaluated for use in that particular population (in terms of age, culture and language) and fully address aspects important to those completing the measures (Guyatt *et al.* 1993; Gill and Feinstein 1994). More recently, qualitative research methods have been used to supplement data, with complementary insights that are both more comprehensive and detailed.

Within the field of oral health, socio-dental indicators were the first measures used to assess impact, largely focusing on the impact of oral health on social functioning (Cohen and Jago 1976; Cushing *et al.* 1986). These narrowly-focused indicators were later replaced by oral health-related quality of life (OHRQoL) measures, which include

symptoms, functional limitations, psychological and social impacts (Allen 2003). Many such measures have been developed for completion by adults, two of which are based on a conceptual model of oral health, namely: Oral Health Impact Profile, with has a full-length 49-item version (OHIP-49) and a shorter, 14-item version (OHIP-14) (Slade and Spencer 1994; Slade 1997b) and Oral Impacts on Daily Performance (OIDP) (Adulyanon and Sheiham 1997). Both these measures have been evaluated and widely used in the UK.

Two measures have been developed for use with children. Both are based on models of health, Child Oral Health Quality of Life (COHQoL) (Jokovic *et al.* 2002) and CHILD-OIDP (Gherunpong *et al.* 2004). Before the commencement of this thesis, neither had been used or evaluated for use in the UK.

Qualitative research has been used to explore the impact of oral health, including, for example the psychosocial consequences of dental pain in adults (Pau *et al.* 2000; Cohen *et al.* 2007) and oral health generally on older people (MacEntee 1996). Little qualitative research has been published specifically on the impact of oral health on children and young people. More detail on OHRQoL and qualitative explorations of impact will be provided later in this chapter.

The main psychosocial concern in discussions about whether DDE are a public health problem, is the appearance of affected teeth (Burt and Eklund 1999; Martinez-Mier *et al.* 2004; Whelton *et al.* 2004; Cutress *et al.* 2006). This section describes the literature on the appearance of DDE, before discussing studies investigating the impact of the condition.

2.4.2.1 Appearance of DDE

The appearance of DDE range from barely detectable white marks, through to severely affected enamel, which is brown-stained, pitted and grooved. Some research has been conducted on the perceptions of lay people or dentists of the appearance of DDE. These studies have typically asked observers to assess the appearance of images of teeth with differing severities of defects.

2.4.2.1.1 Lay people's perceptions of DDE in the UK

Several studies have investigated lay people's perceptions of fluorosis specifically. In the UK, adult perceptions of the appearance of fluorosis were increasingly negative as

severity increased (Alkhatib *et al.* 2004) and social judgements made about those with severe fluorosis were negative compared to those without the condition (Williams *et al.* 2006a; Williams *et al.* 2006b). Elsewhere, lay people in Australia differentiated between different severities of fluorosis with general agreement that TFI 3 was not pleasing (Riordan 1993). The Medical Research Council Report recommended improving knowledge of the public's perceptions of the aesthetic impairment associated with different severities of fluorosis (Medical Research Council Working Group Report 2002).

A survey of adults in the UK investigated perceptions of the appearance of fluorosis (Alkhatib *et al.* 2004). Participants were shown seven photographs representing different severities of fluorosis and were asked to respond to questions about the attractiveness of the teeth, satisfaction with colour and need for treatment. The proportion of participants perceiving teeth to be unattractive, unsatisfactory and in need of treatment increased with greater severity. Most (60.3%) perceived mild (TFI 1 and 2) fluorosis as satisfactory, decreasing to 40.3% for moderate levels (TFI 3 and 4) and 0.3% for severe (TFI 5 and above).

Several studies have investigated lay people's perceptions of the characteristics of individuals with fluorosis. Ninety participants were shown four photographs of an individual with increasing severity of fluorosis (normal, mild, moderate and severe as per Dean's Index of Fluorosis 1934) and asked to attribute characteristics to that individual. Initially participants were asked to write down descriptions of the individual. None of these spontaneously-generated characteristics showed significant variation with the severity of fluorosis. However, when participants were asked to choose from a list of 22 characteristics, the percentage who endorsed 'attractive' and 'clean' decreased with increasing severity. The number of characteristics that varied significantly increased still further when participants were told to look closely at the mouth (attractive and clean plus careful, intelligent, kind, reliable, sociable, unattractive) and also when intra-oral photographs were used. The main limitation of this study was the Hawthorne effect, as participants were aware their judgements were being investigated and there may have been social acceptability bias (Williams *et al.* 2006a).

A follow-up study included a response latency method, where the time taken to make a judgement was used as an indicator of the strength of the attitude. Forty volunteers viewed 12 life-sized extra-oral images of smiling faces, eight of which had been digitally modified to simulate mild fluorosis, severe fluorosis or untreated caries. Forty-five percent of participants attributed negative characteristics to an individual with severe fluorosis compared to 8% for normal enamel, 10% for mild fluorosis and 30% for untreated caries. For images of severe fluorosis and untreated caries these negative attitudes were also more strongly held as they elicited the fastest responses. For images of normal or mildly fluorotic enamel, the fastest responses were those endorsing positive characteristics. Lay people held more negative attitudes to severe fluorosis and untreated caries and also held them more strongly (Williams *et al.* 2006b).

In Australia, Riordan asked 110 lay people (including students, parents and dentists) to complete six statements about their perceptions of the appearance of fluorosis when viewing, in person, 28 children with fluorosis ranging from TFI score of 0 to 3 from a 'conversational distance'. Participants could distinguish between different fluorosis levels, with general agreement that the appearance of TFI 3 was not pleasing. A quarter of lay people felt that, for children with a TFI score of 3, the appearance of their teeth would be embarrassing. Some lay people reported that higher TFI scores indicated neglect on the part of the child. This study purported to investigate the impact of fluorosis on individual children but, rather than asking the children themselves whether their teeth had any impact, they relied on the perceptions of others (Riordan 1993).

2.4.2.1.2 Perceptions of parents

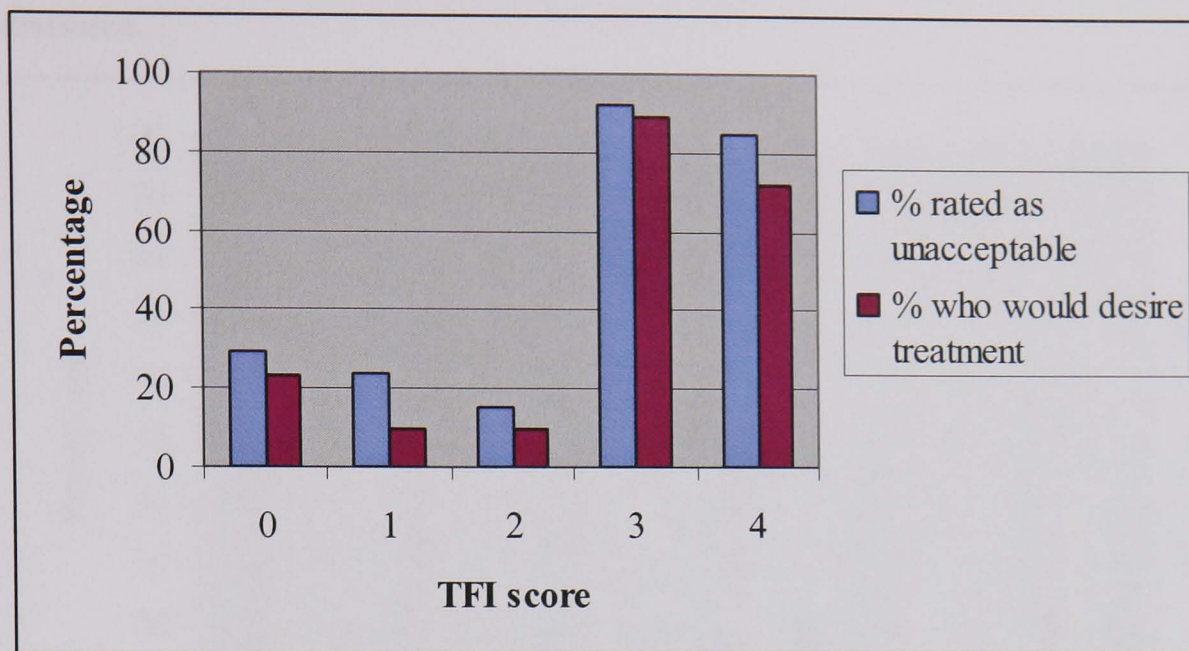
There is a consistent significant inverse relationship between fluorosis and parent's satisfaction with their children's teeth (Clark *et al.* 1993; Lalumandier and Rozier 1998; Sigurjons *et al.* 2004; Levy *et al.* 2005). A study of parents from Iceland, Ireland and England found an increased parental dissatisfaction with the appearance of their child's teeth ranging from 38% dissatisfied by TFI score of 0 compared to 53% for TFI 3. However, alignment, and not tooth colour, was the main reason cited by parents for dissatisfaction (Sigurjons *et al.* 2004). Further research was recommended into the impact of fluorosis on affected children (Lalumandier and Rozier 1998; Sigurjons *et al.* 2004).

2.4.2.1.3 Perceptions of young people

Ellwood and O'Mullane investigated perceptions of different types of DDE in three areas of North Wales and North West England (< 0.1 ppm F, 0.7 ppm F, 0.9 ppm F). Participants were asked about the appearance of their own teeth. Different areas were used in case perceptions differed between communities where defects were prevalent or not. The size of demarcated opacities and the degree of enamel hypomineralisation was weakly related to satisfaction with appearance, with small demarcated opacities and mild enamel hypomineralisations not compromising the appearance of the anterior teeth, but larger demarcated opacities and more severe hypomineralisation identified as being less acceptable. Interestingly, 41% of children with large demarcated opacities or severe hypomineralisation of their anterior teeth reported they had not noticed any marks that spoiled the appearance of their teeth. There was no significant difference in satisfaction with the appearance of the teeth among the three areas in the study (Ellwood and O'Mullane 1995).

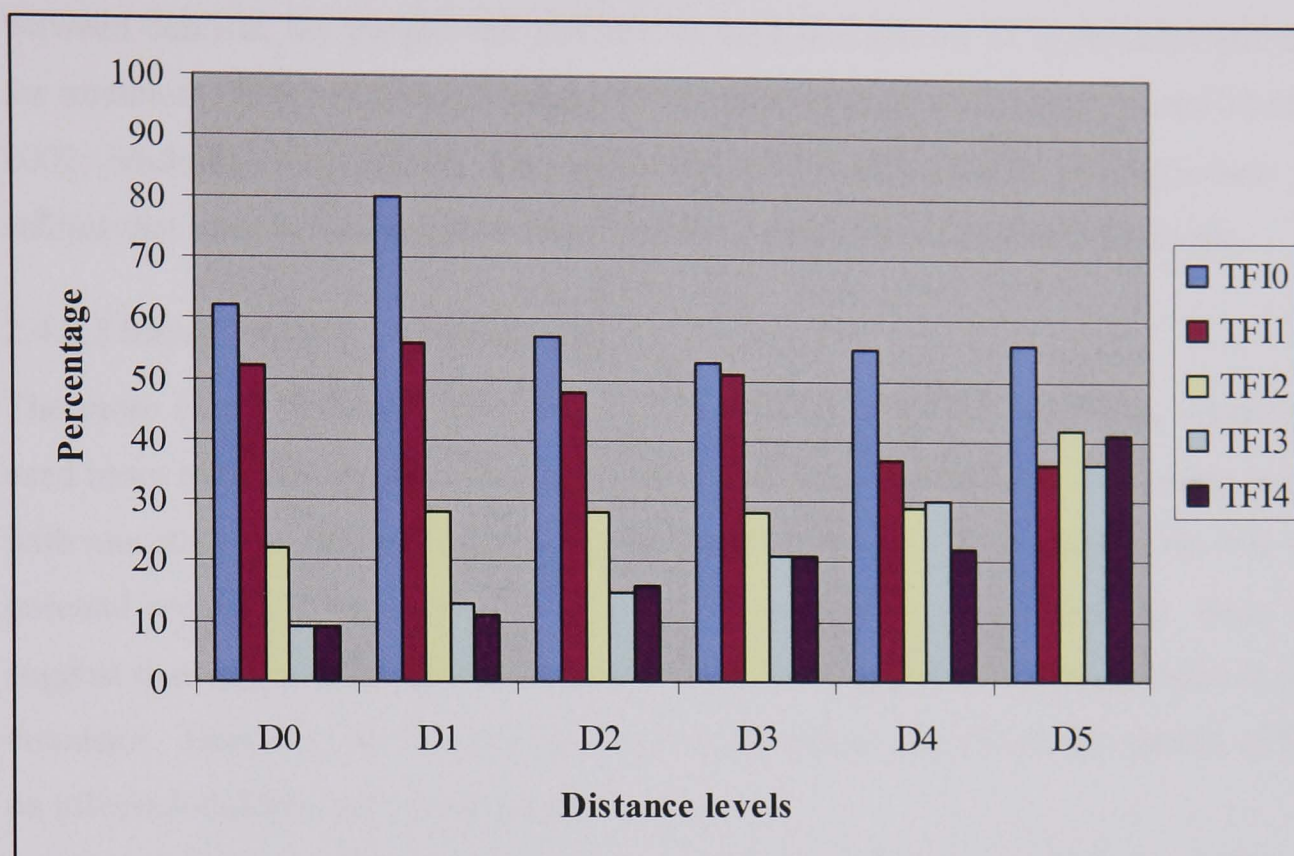
Hawley and colleagues 1996 asked a random sample of 14-year-olds from a non-fluoridated area of the North West of England to examine six photographs of upper anterior teeth with TFI scores 0-4. Photographs were of life-sized teeth with lips cropped off. Participants were asked to rate the appearance of each photograph as either very poor, poor, acceptable, good or very good and to indicate whether they would request treatment if their teeth were so affected. The proportion who rated the photographs as poor/very poor fell from 29% for TF scores of 0 to 15% for TF 2 and then increased to 92% for TF score 3 (Figure 1). While the trend for appearance to be unacceptable with increasing TFI score was significant, there was also evidence of variation in acceptability that was not related to severity. The responses regarding the desire for treatment matched closely with their opinions on appearance. No difference was found between males and females, but the authors felt that unacceptability would probably be affected by a large number of factors including age, social class and how the teeth were viewed (teeth in this study were viewed with the lips omitted). The authors concluded that teeth with some milder forms of fluorosis may be more aesthetically pleasing than those without (Hawley *et al.* 1996). The findings from this study were used in the York Review to define the level of fluorosis of 'aesthetic concern', as equivalent to TFI of 3 or more. However, this study involved young people rating clinical photographs of teeth only, rather than teeth in the context of a person's mouth or face and didn't include the perspective of those affected by DDE.

Figure 1. Acceptability and desire for treatment according to severity of fluorosis



A web-based UK study, used digitally altered images to investigate young peoples' perceptions of the appearance of fluorosis (Edwards *et al.* 2005). Fluorotic markings, reflecting TFI scores of 0 to 4, were added to images of the teeth of a young female. Participants were asked to rate different views (teeth only (D0), mouth at different simulated distances (D1-D4) and full face (D5)) with different severities of fluorosis in terms of their acceptability and whether they would request treatment if their teeth had that appearance. Acceptability reduced as severity of fluorosis increased (particularly for TFI scores of 2 or over). Acceptability increased when teeth were viewed surrounded by lips, compared to teeth only, and also with increasing distance (Figure 2). The authors commented that the young people found views of teeth with retracted lips 'distasteful', whether fluorosis was present or not. This finding has implications for previous studies of young people's perceptions of fluorosis (Edwards *et al.* 2005).

Figure 2. Percentage of responses rated as acceptable for each TFI level at every distance.



2.4.2.1.4 Agreement between children, parents and dentists

Several studies have found poor agreement between dentists, lay people and children on their perceptions of the appearance or perceived need for treatment of DDE including fluorosis (Ellwood and O'Mullane 1995; Milsom *et al.* 2000; Astrom and Mashoto 2002; Shulman *et al.* 2004). These data suggest normative assessments do not predict those children with DDE who are dissatisfied. In a study of 12-year-olds in North West England, children reported they had far fewer cases of DDE than dental professionals. Further research into the impact of DDE was recommended (Milsom *et al.* 2000).

2.4.2.1.5 Summary

In summary, the studies described above investigated the perceptions of the appearance of DDE. Many studies have been carried out on this subject. Lay people were found to have negative perceptions of the appearance of teeth with fluorosis (Alkhatib *et al.* 2004) and to make negative social judgements about those with severe fluorosis (Williams *et al.* 2006a; Williams *et al.* 2006b). Among parents, dissatisfaction with appearance increased with increasing severity (Clark *et al.* 1993; Lalumandier and Rozier 1998; Sigurjons *et al.* 2004; Levy *et al.* 2005). The findings from studies of young people's perceptions have varied, with suggestions that viewing mouth or full-

face images and viewing distance are important factors in determining acceptability of appearance (Edwards *et al.* 2005). Finally, several studies have found poor agreement between dentists, lay people and children in their perceptions of appearance and need for treatment (Ellwood and O'Mullane 1995; Milsom *et al.* 2000; Astrom and Mashoto 2002; Shulman *et al.* 2004). The following section will describe the apparently few studies that have investigated the impact of the condition on affected individuals.

2.4.2.2 Impact of DDE on individuals

The more recent studies of the impact of DDE used OHRQoL measures, others have used more rudimentary questionnaires. Most have involved children and young people, with one study specifically examining the impact on adults. One study in the UK used parental reports of the impact of DDE on their children. Cumulatively, these data suggest that severe fluorosis has an impact on some children and young people in other countries. However, no research has been conducted in the UK on the impact of DDE on affected children and young people.

2.4.2.2.1 Impact of DDE on adults

A small study (n=30) on US adults with and without severe amelogenesis imperfecta compared impact on OHRQoL, social avoidance and self-esteem. The study was conducted to encourage dental insurers to cover the treatment of conditions such as amelogenesis imperfecta. The authors were concerned both with the impact of the appearance of the enamel of patients with severe amelogenesis imperfecta and also the corresponding malocclusions and sensitivity. There were significantly higher levels of social avoidance in adults with severe amelogenesis imperfecta, but no difference in self-esteem. Affected participants reported higher levels of self-consciousness and embarrassment on OHIP-14 (Coffield *et al.* 2005). The limitations of this study include non-response bias due to the poor response rate (44.1% among those with severe amelogenesis imperfecta and 40.3% for those without) and the use of a self-esteem measure designed for use with adolescents, used with adults (mean age of 36.9 years).

2.4.2.2.2 Parental reports of the impact of DDE

The 2003 UK Children's Dental Health Survey used parents as proxies to report the impact of DDE on their children. More parents of children with DDE reported their child's oral health had an impact on their self-confidence and had experienced pain in the past 12 months than parents of children without DDE (Nuttall and Harker 2004).

2.4.2.2.3 Impact of DDE on children and young people

The first study to investigate the impact of DDE on young people was conducted in Tanzania with 13-15-year-olds with severe fluorosis (TFI score of greater than 4). Participants were asked three questions: first about the distress the appearance of their teeth caused, secondly how much they worried about the appearance of their teeth and finally how much their smiling was affected. Most of the young people reported no distress or worry, but 70% reported that the way their teeth looked hindered them from smiling freely. This was a surprising finding, in a community with endemic fluorosis, where severe fluorosis is very common. Usually, concern about appearance is a result of being visibly different (Thompson and Kent 2001) but it would be anticipated in this case that, as fluorosis was endemic, the differences would be minimal. However, this finding may be because participants compared their appearance to peers who originated from another area (van Palenstein Helderma and Mkasabuni 1993). The questions used in this study were chosen by the authors to cover the physical, psychological and social aspects of health, rather than being based on a theoretical model. The questions were administered as an interview by a researcher who knew the participants, which may have resulted in acceptability bias.

An 8-item questionnaire was designed to measure concerns among children (7-11 years) and their parents of the appearance of fluorosis and to compare concerns held in Mexico City and Indianapolis. This questionnaire included questions used in earlier studies (van Palenstein Helderma and Mkasabuni 1993; Clark and Berkowitz 1997), with additional items to distinguish satisfaction with fluorosis from satisfaction with other tooth factors, such as alignment and overall colour. Generally, perceptions of discolouration were associated with the concerns of the participants, with higher levels of concern in Indianapolis compared to Mexico City. The questionnaire was found to have acceptable reliability and validity (Martinez-Mier *et al.* 2004). It was not made explicit whether this measure was based on a model of health and it has not been used or evaluated for use elsewhere.

In a fluoridated area of Malaysia, 16-year-old young people, with and without DDE, were asked several questions about the way their front teeth had an impact on their social activities (Sujak *et al.* 2004). No differences were found between groups in terms of covering their mouth when smiling, avoiding going out with friends or lack of confidence when socialising. Results of a logistic regression model indicated that

having DDE alone did not predict dissatisfaction with the condition. Further research on the psychosocial impact of enamel opacities was recommended. Again, the choice of the questions was derived from a review of the literature and not based on a model of health or the views of Malaysian young people themselves.

The three studies described above attempted to investigate the impact of DDE on young people with the condition. Relationships between normative assessment of DDE and data on psychosocial impact were apparently weak. These studies used several questions, chosen apparently arbitrarily by the authors, without description of the theoretical model used. Additionally, it is not clear to what extent this research includes the views of children. Several other studies have also investigated the impact of DDE, but have used OHRQoL measures.

A more detailed investigation of the impact of fluorosis on young people was conducted in Arusha, Tanzania. The prevalence of fluorosis of TFI scores of 2 or higher was 74.9% in an area of water containing 3.6 ppm F. A sample of 478 young people, with a mean age of 15.7 years, completed a modified version of OIDP, asking participants, during the past six months, how often problems with their mouth or teeth had caused them difficulties with eating, speaking and pronouncing clearly, cleaning teeth, sleeping and relaxing, smiling without embarrassment, maintaining emotional state and enjoying contact with people. Participants were also asked to assess the discolouration of their maxillary anterior teeth and to report the condition of their mouth and teeth. Significantly more females than males reported themselves to be affected on at least one daily performance. Overall, young people who reported oral impacts on their daily performance and discolouration of upper anterior teeth were more likely to be dissatisfied with both their oral condition and their dental appearance. Again, despite being members of a community where dental fluorosis is endemic, unexpectedly high proportions of participants confirmed their oral condition impacted on their daily lives (Astrom and Mashoto 2002). One of the limitations of this study was the choice of measure as they used a version of OIDP that had been modified by removing the item 'carrying out major work or social role'. The authors did not evaluate the use of the modified version or use of this 'adult' measure for this age group in Tanzania.

A similar study in Brazil also investigated the impact of fluorosis using a modified version of OIDP with 513 schoolchildren aged 6-15 years. No differences in impact were found between those normatively assessed as having fluorosis or not (Michel-

Crosato *et al.* 2005). The version of ODP used in this study assessed eight performances and replaced the 'carrying out major work or social role' performance with 'studying'. No details were given of how this modified measure was evaluated or even how it was administered (as an interview or self-completed questionnaire), this is particularly relevant as children as young as six years participated.

Only one study has been published that used an OHRQoL measure designed for children (Robinson *et al.* 2005). The aim was to describe the OHRQoL of children and compare the impact of caries and fluorosis. The participants were 211 12-year-olds in rural Uganda, completing the Child Perceptions Questionnaire for 11-14 years (CPQ₁₁₋₁₄) in the school setting. CPQ₁₁₋₁₄ is one questionnaire of the COHRQoL battery of measures (section 2.4.2). This study also included an evaluation of the reliability and validity of CPQ, which were found to be acceptable for use in this age group, setting and language. More young people with fluorosis of TFI score greater than 2 had impacts on their OHRQoL than those without. However, the number of children with fluorosis was small (n=12), so it is difficult to make conclusions regarding the nature or extent of the impact of fluorosis.

2.4.2.3 Summary

There is some evidence to suggest that severe fluorosis has an impact on some children and young people in other countries, particularly in areas of Africa. These countries differ from the UK in the levels of fluoride in the water, the prevalence and severity of fluorosis and social and cultural views of health. The relationships between normative assessments of DDE and young people's reports of impact were weak. No research has been conducted on the impact of DDE on children and young people in the UK. The methods used in this body of research apparently included rudimentary questionnaires and modified versions of 'adult' OHRQoL measures that may not have been evaluated for use in these countries, with participants of this age, in their modified forms or in relation to DDE specifically. Evaluation for use in a particular population (in terms of age, culture and language) is an important methodological consideration when using such measures (Guyatt *et al.* 1993; Gill and Feinstein 1994). There is also no evidence that the specific aspects of DDE have been included in the design of these studies.

Only one study used an OHRQoL measure designed for children, which was evaluated for use in the target population. Further detailed research on the impact of DDE.

particularly fluorosis, has been recommended repeatedly (Lalumandier and Rozier 1998; Milsom *et al.* 2000; Medical Research Council Working Group Report 2002; Sigurjons *et al.* 2004; Sujak *et al.* 2004).

The review has identified a gap in knowledge about whether DDE impact on individuals and, therefore, whether they constitute a public health problem in the UK. It has also identified an apparent dearth of data, as well as potential methodological problems in relation to assessing impact, particularly among children and young people.

2.4.3 Impact of the condition on wider society

The third criterion for a public health problem regards the impact on society. For DDE, this impact would be the cost of treatment and time missed from school or work. For example, the cost of treating dento-facial trauma has been estimated by Scandinavian researchers to be US\$ 3.2-3.5 million per million population (Andreasen and Andreasen 1997). In the UK the average total cost of treating one patient with a dental traumatic injury has been estimated at £856 (Wong and Kolokotsa 2004). Oral disease and its treatment in the US results in the loss of over 20 million work days and 51 million school hours per year (Department of Health and Human Services 2000). However, no research of this kind has been conducted in relation to DDE.

In order to be able to assess the impact of DDE on society, data would be required on both the number of individuals needing treatment and the corresponding direct and indirect costs. In dentistry, estimates of the need for treatment or oral health promotion interventions have, traditionally, relied on surveys, including only normative assessments of the prevalence of oral diseases and conditions. However, normative methods have several shortcomings. First, professional assessments often differ from those of lay people, as was noted for DDE in section 2.4.2.1.4. Poor agreement was found between the perceptions of the need for treatment among lay people, dentists and children (Ellwood and O'Mullane 1995; Milsom *et al.* 2000; Astrom and Mashoto 2002; Shulman *et al.* 2004). Secondly, agreement between professionals often differs, for DDE this was evident by the less than perfect inter- and intra-examiner reliability for DDE and fluorosis indices. Thirdly, for complex conditions, such as DDE, which have many different causes and a range of clinical appearances, the normative assessments are particularly prone to inaccuracies. Finally, they neglect the psychosocial aspects of health such as the impact of conditions on individuals (Sheiham *et al.* 1982).

More recently, oral health needs assessments have also included measures of the impact of oral health on individuals, acknowledging that the presence of a condition does not always mean the individual will perceive the need for treatment (Sheiham and Spencer 1997). To this end, OHRQoL measures have been incorporated into needs assessment, mainly in older adult populations (Sheiham and Tsakos 2007). Supplementation of clinical data with assessments of the impact of conditions have been recommended in oral health needs assessments for children (Weintraub 1998; McGrath *et al.* 2004; Tsakos *et al.* 2006).

Future research on the impact of DDE on wider society would need to take account of needs for treatment from both clinical and individuals' perspectives. Therefore, research on the impact of DDE on the individual is needed before further research can be conducted on the impact on wider society.

2.4.4 Condition is preventable and effective treatments are available

The final criterion of a public health problem considers the potential for prevention and effective treatment of DDE.

2.4.4.1 Prevention of DDE

The literature deals mainly with minimising the risk of DDE by reducing the ingestion of substances including tetracyclines and particularly fluoride.

Tetracycline-induced DDE have largely been eliminated by the implementation of guidelines restricting the use of the antibiotic (Cutress *et al.* 2006). The British National Formulary advises caution prescribing tetracyclines during pregnancy, breastfeeding and to children under 12 years of age (British National Formulary 2007).

2.4.4.2 Prevention of fluorosis

Various measures have been recommended to reduce the risk of fluorosis from different sources of fluoride.

In 1978 the concentration of fluoride in the water in Hong Kong was reduced from 1.0 ppm F to 0.7 ppm F because of concerns about fluorosis. The prevalence of fluorosis in children 7 to 12-years-old reduced from 64% in 1979 to 47% in 1985 (Evans and Stamm 1991). To reduce the risk of fluorosis in Ireland, the optimal range for water

fluoridation has been changed from 0.8-1.00 ppm F to 0.6-0.8 ppm F (Clarkson *et al.* 2003).

Recommendations have been made in several countries, including Australia, the United States, Canada and Ireland, for fluoride intake from toothpaste to be reduced. Measures taken include restricting use of fluoride toothpaste to those over 2 years of age, use of low fluoride toothpaste formulations, guidance on the amount of toothpaste used and need for parental supervision (Horowitz 1992; Canadian Dental Association. 2000; Department of Health and Children 2002; Riordan 2002; Do and Spencer 2007). However, lower fluoride dosage in toothpaste reduces its effectiveness in caries prevention (Mitropoulos *et al.* 1984; Holt *et al.* 1994; Ammari *et al.* 2003). In the UK, toothpastes containing 1000 ppm F are currently recommended for use in pre-school children, to balance the risks of fluorosis and caries (Scottish Intercollegiate Guideline Network 2005; Department of Health & the British Association for the Study of Community Dentistry 2007).

Similarly, concerns about the risk of fluorosis, particularly due to compliance with fluoride tablets/drops regimens have led to a reduction in their use as a public health measure including in the UK (Burt and Eklund 1999; Riordan 1999; Fomon *et al.* 2000; Riordan 2002; Scottish Intercollegiate Guideline Network 2005).

2.4.4.3 Effective treatment of DDE

The techniques recommended in the UK for the management of DDE include various methods to improve the appearance of teeth. Options include: microabrasion (either hydrochloric acid/pumice or phosphoric acid/pumice); bleaching either at the chairside or using a nightguard technique; composite resin applied to discrete areas of defective enamel or as a veneer over the tooth surface. Porcelain veneers are only recommended for patients of 16 years or over and then only when other techniques have failed. It is recommended that the shade of the tooth is recorded pre- and post-operatively, to assess the effectiveness of the treatment for an individual patient (Wray and Welbury 2001).

Several studies have investigated the effectiveness of these techniques in improving the appearance of DDE. A clinical trial of nightguard bleaching in Mexico found it to be effective for teeth with mild fluorosis as judged by clinicians. The effectiveness from the patient's perspective was not assessed; patients were asked only about side-effects

(Loyola-Rodriguez *et al.* 2003). A study conducted in the UK of 32 patients with DDE attending paediatric dentistry clinics judged the effectiveness of microabrasion as assessed by patient satisfaction. Patients were asked their opinion of their teeth at the end of treatment and six months later. Opinions were not sought before treatment as the authors postulated that the patients would not attend for treatment unless they were dissatisfied with their teeth. Twenty-one patients were satisfied with the appearance of their teeth immediately after microabrasion, with 17 patients still satisfied six months later (Wong and Winter 2002). The assessment of satisfaction was purely based on a 'yes' or 'no' answer to one question: are you satisfied with your teeth? (Wong, personal communication).

OHRQoL measures have been used in trials to assess the effectiveness of treatments including implant-retained or conventional prosthesis (Heydecke *et al.* 2005; Allen *et al.* 2006) and reservoir biteguards in patients with xerostomia (Robinson *et al.* 2005). Such measures have not been used to assess the effectiveness of treatments in children. Future studies of the effectiveness of treatments for DDE could include such measures of the effectiveness from the patient's perspective.

2.4.5 Conclusion

This section has reviewed the literature on DDE against the criteria for a public health problem. Most research has focussed on DDE in children and young people. The first criterion regarding the prevalence of DDE in the UK, has received considerable attention. DDE occur in about one-third of UK children, with the most common types being demarcated and diffuse defects (O'Brien 1994; Chadwick and Pendry 2004). Fluorosis is more common in the presence of fluoride in the water supply (McDonagh *et al.* 2000).

It is difficult to find data that answer the second criterion regarding the impact of DDE on children and young people, with no research previously conducted in the UK. Other studies of the impact of DDE have been conducted on children and young people in low or middle-income countries, often in areas with endemic fluorosis. These countries differ from the UK in the levels of fluoride in the water and the prevalence and severity of fluorosis. The relationship between DDE and impact in those studies is neither strong nor consistent. The methods used in these studies have limitations, particularly regarding the choice of measures. To this end, measures developed for adults have been

applied unquestioningly to children. In addition, further research on the impact of DDE in the UK has been recommended by the Medical Research Council and other authors (Lalumandier and Rozier 1998; Medical Research Council Working Group Report 2002; Sigurjons *et al.* 2004).

Similarly, no research has been conducted on the impact of DDE on wider society. Previously, such assessments of need for treatment at a population level would have been based on the prevalence of the condition, but more recent needs assessments have included the impact of a condition on the individual. In this way, the impact on the individual and wider society are closely linked, as only when the impact at the individual level has been established can oral health needs assessments of wider society be investigated.

The final criterion for a public health problem considers effective interventions. Measures to reduce DDE induced by tetracyclines and fluoride have been adopted in the UK, with various treatments recommended to improve their appearance. Some research has been conducted into the effectiveness of such treatments, with only one study attempting to gain children's perspective.

Overall, DDE have been the subject of much research, particularly in terms of their aetiology and prevalence. However, it is difficult to come to any conclusions about whether they are a public health problem in the UK, due to the gaps in the existing evidence. Authors who have raised this question in other countries have frequently identified the lack of information on the impact of the aesthetics of DDE on those with the condition (Burt and Eklund 1999; Martinez-Mier *et al.* 2004; Whelton *et al.* 2004; Cutress *et al.* 2006).

The remaining section of this chapter will consider in more detail current approaches to the study of subjective impact of dental conditions (Box 1).

Box 1. Outline of Chapter

- Background to developmental defects of enamel (DDE)
- Significance of DDE as a public health problem against four criteria:
 - Prevalence
 - Impact of the condition on the individual
 - Impact of the condition on society
 - Prevention and availability of effective treatments
- **Approaches available to investigate the impact of dental conditions**
- **Rationale**
- **Aims and objectives of the thesis**

2.5 Approaches available to investigate the impact of dental conditions

The complex challenges associated with research of the impact of DDE require careful consideration. This section begins with a methodological review of current approaches to the study of the impact of dental conditions on individuals. This involves understanding:

- the difference between a biomedical model of disease and the biopsychosocial model of health and
- the current state of the art approaches to the study of the impact of disease

The problem is further complicated as the population of interest are children and young people. As a result, there is a need to review contemporary approaches to research *with* children. The degree to which the measurement of impact in children has adopted current best practice regarding research *with* children will be discussed.

2.5.1 Models of health

The literature reviewed earlier in this chapter emphasised the aetiology and prevalence of DDE, rather than its impact. This emphasis is characteristic of most public health research in that it has concentrated on the epidemiology and biostatistics of the condition (Lupton 1995). Public health and biomedicine have, traditionally, been

disease-orientated, with little consideration of the subjective experiences of those with various health states. This has largely been as a consequence of the dominance of the biomedical model of disease in medicine and dentistry, which is orientated to answer certain research questions, but neglects others.

2.5.1.1 The Biomedical Model of Disease

The biomedical model has been the dominant model of disease since the late 19th century. The emphasis is on pathological processes and the way in which they compromise the body (Annandale 1998). Research within this model has led to the discovery of the causes of many infectious diseases including measles, polio and elimination of diseases, such as smallpox, with the development of vaccines (Lupton 1995). The role of the clinician within the biomedical tradition is to repair these deviations from the normal biological processes (Imrie 2004).

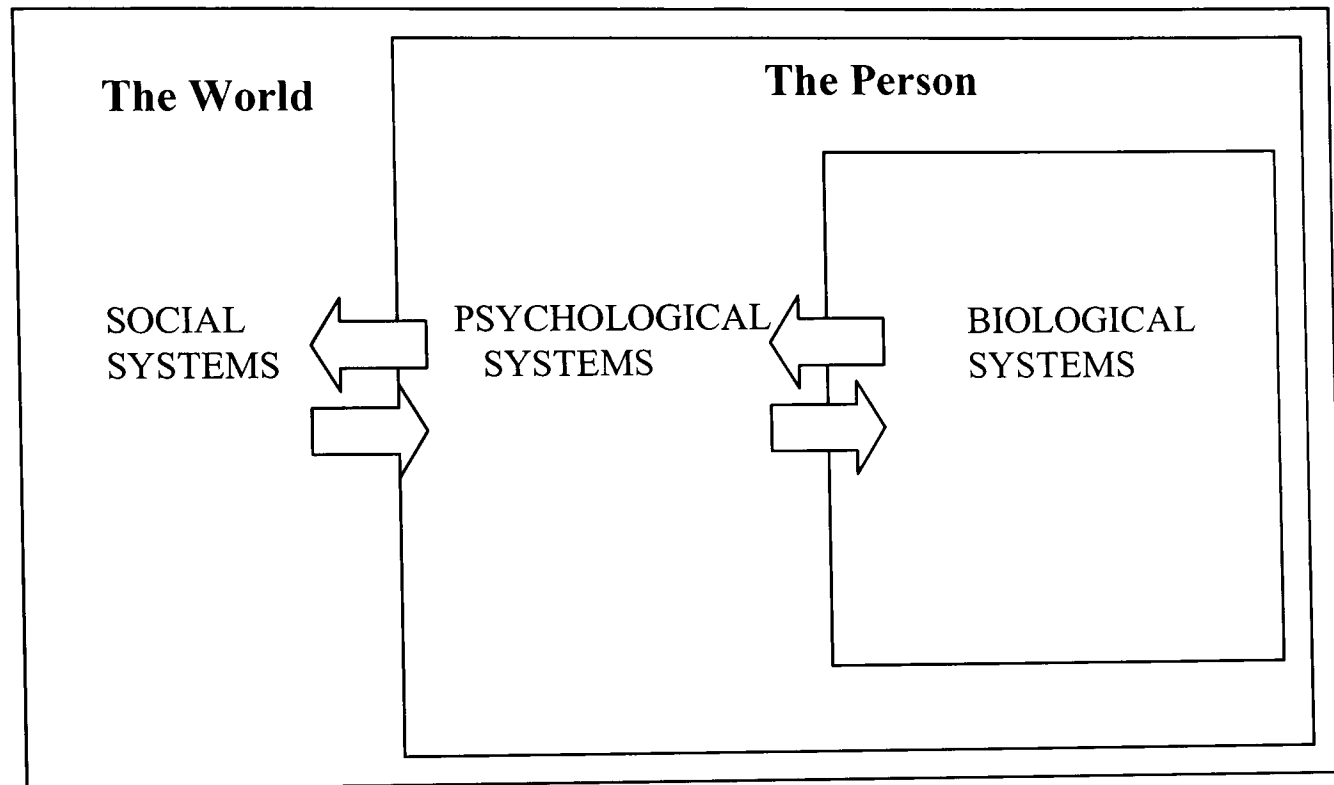
More recently, the biomedical model has been criticised for its narrow disease-focus (Annandale 1998; Nettleton and Gustafsson 2002). Within public health, assessment of the health needs of the population that rely purely on disease surveillance fail to account for the impact of the disease, as was discussed in section 2.4.3 (Sheiham *et al.* 1982; Bowling 1997; Sheiham and Spencer 1997). In the clinical setting, the focus of the biomedical model has been criticised for neglecting the patient's perspective: for example, a person may have a disrupted biological process, as evidenced by a laboratory finding, but may feel well, while another person may feel ill, but no disease processes are identifiable. Similarly, while clinicians may be able to use a patient's history of symptoms and clinical investigations to diagnose disease, interventions aimed purely at correcting physical abnormalities have limited ability to ensure the full recovery of the patient (Engel 1977).

2.5.1.2 The Biopsychosocial Model of Health

The focus of the biopsychosocial model is on health and illness, rather than disease. This focus recognises the dynamic interaction of physiological processes with personal and social factors. Rather than reducing disease down to its constituent variables, the model sees biological and psychological systems as the two interrelated systems of the person, with the person interrelating with the social systems of their world (Sarafino 2002) (Figure 3). The biopsychosocial model provides a framework for health care, public health, research and teaching (Engel 1977) and has been applied to chronic pain (Talo *et al.* 1995), psychiatric (Engel 1980) and gastro-intestinal conditions (Drossman

1996). Over the past two decades, research has been shifting from the biomedical to the biopsychosocial model of health (Alonso 2004).

Figure 3. Systems of the biopsychosocial model (Sarafino 2002)



The World Health Organization (WHO) devised the International Classification of Functioning, Disability and Health (ICF) based on the biopsychosocial model (World Health Organisation 2002). Although the ICF is not described in terms of 'systems', it is similarly arranged to cover functioning and structures of the body, activities of the individual and participation in society. These domains are set against contextual factors, from external environmental sources and internal personal factors, including gender, age, social status and ethnicity. The context influences how a health condition is experienced by an individual. ICF was designed as a planning and policy tool to shift focus from a biomedical emphasis on the causes of disease, to the biopsychosocial approach of the impact of health conditions. The ICF and the biopsychosocial framework have received little attention in relation to oral health (Reisine *et al.* 1994; MacEntee 2006).

A biopsychosocial approach would include not just the prevalence and determinants of defects, but also the psychological and social systems, such as people's thoughts, feelings and beliefs about the condition and how they feature in interactions between people, families and society. The framework provided by the biopsychosocial model,

therefore, allows an emphasis on the impact of DDE on children and young people. This thesis attempts to remain within this framework, wherever possible.

2.5.2 Investigating impact

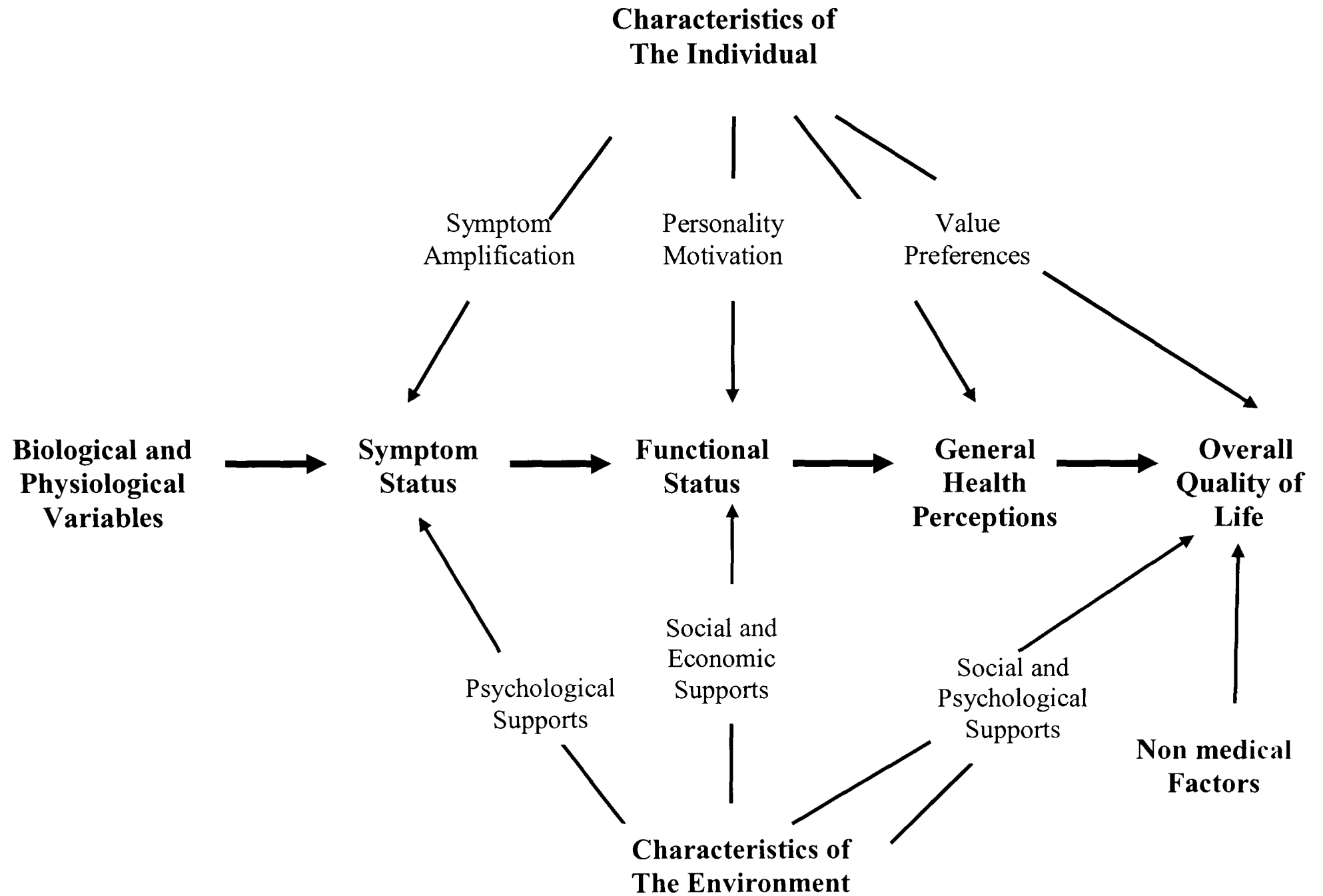
Quantitative and qualitative research may be used to investigate the impact of conditions on individuals. As stated earlier, quantitative HRQoL questionnaires have dominated research on the psychosocial impact of DDE and, indeed, health in general. Less commonly, qualitative approaches seek to explore the impact of health through the ‘eye of the experiencer’ (Ziller 1974) rather than using predefined domains that may not be important to the individual. This section will describe quality of life and qualitative approaches to investigating the impact of conditions.

2.5.2.1 Health-related quality of life (HRQoL)

HRQoL has been the predominant approach in research of individuals’ perspectives on their health and healthcare treatment. The terms ‘quality of life’, ‘health status’ and ‘HRQoL’ are often used interchangeably (Fitzpatrick *et al.* 1998). From a patient’s perspective, ‘quality of life’ and ‘health status’ are distinct constructs (Smith *et al.* 1999). The term ‘HRQoL’ is used to acknowledge that widely valued aspects of life exist that are not generally considered as ‘health’, including income, freedom and quality of the environment (Guyatt *et al.* 1993). ‘HRQoL’ is different from ‘quality of life’ in that it focuses on the impact of a disease/condition on the ability of a person to live a fulfilling life (Bullinger *et al.* 1993). In the literature, it includes dimensions of physical, social and role functioning as well as mental health and general health perceptions.

Wilson and Cleary suggest measuring HRQoL in levels, ranging from biological and physiological domains at one end of a continuum, through symptom status, functioning and general health perceptions, to overall quality of life at the other end. Each of the levels are related and influenced by characteristics of the individual and of the environment. This model was intended to facilitate understanding of the relationship between traditional variables and measures of HRQoL (Wilson and Cleary 1995) (Figure 4). The model, effectively operationalises the biopsychosocial model of health, and has been used in oral health research as a framework for investigating the relationship between clinical variables and their impact in patients with xerostomia or edentulousness (Baker *et al.* 2007; Baker *et al.* 2007).

Figure 4. The Wilson and Cleary model



2.5.2.2 HRQoL measures

HRQoL measures may be generic or specific. Generic measures assess HRQoL generally whereas specific measures focus on problems relevant to that condition, site or disease. Each has strengths and weaknesses for different circumstances.

2.5.2.2.1 Generic HRQoL measures

The main advantages of generic measures are their broad applicability, allowing comparisons between groups or people with different conditions. They have been used more frequently than disease-specific instruments in the general population and can be used when no disease-specific instruments exist in a particular area. Due to the broad range of aspects of health status and consequences of illness, they may detect unexpected problems associated with a condition or its treatment (Guyatt *et al.* 1993).

The main shortcoming of generic instruments relate to their broad nature, which render them less responsive to change, and less relevant and acceptable to participants with different specific conditions.

2.5.2.2.2 Specific HRQoL measures

Specific instruments may be disease-, site- or dimension-specific and so overcome some disadvantages of generic instruments. The relevant content can make them more sensitive, more acceptable to participants and, therefore, higher completion rates are more readily achievable. Their specific nature makes them more likely to respond to change (Robinson *et al.* 2002). The disadvantages are that they are not suitable in samples who do not have the relevant condition and they are too specific to detect effects not anticipated (Fitzpatrick *et al.* 1998). Some specific instruments are not based on a concept of quality of life, being simply composed of 'narrow mental, physical, and social functioning subscales alongside symptom checklists' (Carr *et al.* 2002).

The two categories of measure, generic and specific are not mutually exclusive and may be used in combination.

2.5.2.2.3 Discriminative and evaluative measures

Discriminative HRQoL measures are intended to distinguish between participants at a point in time, such as those with better or worse HRQoL (Guyatt *et al.* 1992). Discriminative measures can be used to quantify the burden of illness or compare the burdens of different illnesses or conditions. Evaluative instruments detect changes within individuals over time, such as in clinical trials, when changes may be due to the intervention under investigation.

2.5.2.3 Properties of HRQoL measures

The important properties of HRQoL measures are: appropriateness, reliability, validity, responsiveness, precision, interpretability, acceptability and feasibility (Fitzpatrick *et al.* 1998). Definitions for these criteria exist, but are not universally accepted.

2.5.2.3.1 Appropriateness

As stated earlier, measures must be appropriate to the aims of the study, based on an explicit definition of health and should fully capture factors important to those completing them (Gill and Feinstein 1994; Jokovic *et al.* 2002). Without consideration of these factors, research could underestimate some impacts and give undue weight to others (Brondani and MacEntee 2007).

2.5.2.3.2 Reliability

Reliability, in this context, is the consistency with which a questionnaire measures what it is designed to measure (Carr *et al.* 2002). It has two different aspects: internal consistency and reproducibility.

Internal consistency refers to whether a questionnaire enquires about a unified concept. Individual items should correlate strongly, both with each other and with the summed score of the total of items in the same scale. However, if items of a scale correlate perfectly, it is likely that there is some redundancy among items. A balance is needed between minimal redundancy and a measure that is too homogenous (Fitzpatrick *et al.* 1998).

Reproducibility evaluates whether an instrument yields the same results on repeated application. There is little agreement about the length of time that should elapse between repeated applications. However, the interval should be sufficient to ensure that participants

are unlikely to recall their previous answers, but not so long as to allow actual changes in the underlying dimension.

2.5.2.3.3 Validity

Validity is the degree to which a questionnaire measures what it is designed to measure. There are several different types of validity: face, content, criterion and construct. In order to assess the different types, a range of evidence, including how the content of the questionnaire was determined, inspection of the content and relationships to other variables need to be considered (Carr *et al.* 2002).

2.5.2.3.3.1 Face validity and content validity

Face validity examines whether instruments appear to make sense to participants, whereas content validity examines the extent to which the domain of interest is comprehensively covered in the instrument. Face validity and content validity are related, but discrete. Together, they assess whether measures clearly address the intended subject matter and whether all aspects of HRQoL are adequately covered. Assessment of both face and content validity requires qualitative techniques, to examine the questionnaire and to look at how the questionnaire was developed. The testing of face and content validity have been criticised for insufficient rigour and often involves panels of ‘experts’, rather than those to whom the measure is directed (Brondani and MacEntee 2007).

2.5.2.3.3.2 Construct validity

The first step in construct validation is to establish or select a model or theoretical framework that represents an understanding of what investigators are trying to measure. The theoretical framework provides a basis for understanding the behaviour of the system being studied and allows hypotheses or predictions about how the instruments being tested should relate to other measures (Jokovic *et al.* 2002). Typically, construct validity is examined by the correlation of a new measure against a range of other evidence, such as clinical or laboratory data and use of health services (Fitzpatrick *et al.* 1998).

2.5.2.3.3.3 Criterion validity

Criterion validity is the extent to which a questionnaire measures what it claims to measure, assessed by comparison with a ‘gold standard’ of the same attribute (Guyatt *et al.* 1993). As no gold standard of HRQoL exists, global single-item questions (such as the global oral

health question, ‘how do you rate the health of your teeth, lips and mouth’ (Atchison and Gift 1997)) are often used instead (Robinson *et al.* 2003).

The validity of a measure needs to be assessed in relation to a specific purpose and particular population or setting. As was evident in the literature described earlier in this chapter, many validated measures are adopted for use in circumstances which bear little relation to the original sample (Brondani and MacEntee 2007). The language and cultural context in which a measure is used can have a bearing on the validity, as can the intended purpose of the measure (Weintraub 1998). To overcome this, it is necessary to carry out validation work in the specific sample, to ensure that the measure retains its psychometric properties (Bowling 1997; Robinson *et al.* 2005).

2.5.2.3.4 Responsiveness

Responsiveness is the ability to detect meaningful change (Carr *et al.* 2002). It is particularly important when measures are used in trials, when it is necessary to detect changes over time, within individuals, that might reflect therapeutic effects.

2.5.2.3.5 Precision

Precision refers to the size of the gradations within a scale. A more precise measure will distinguish between finer gradations of the underlying concept. The main influences on precision is the format of the response categories (for example a Likert scale or a visual analogue scale), the method of numerical scoring and the method of weighting (Fitzpatrick *et al.* 1998). Whilst useful to distinguish between groups and detect small changes, greater precision increases the complexity of the scoring and is redundant if poor reproducibility renders fine gradations irrelevant.

2.5.2.3.6 Interpretability

Interpretability is concerned with the meaning of the scores. HRQoL measures have often been criticised by clinicians and others for their lack of interpretability, when compared to other measures such as clinical indices. This may be due to lack of familiarity (Carr *et al.* 2002).

2.5.2.3.7 Acceptability

Acceptability relates to whether the instrument is acceptable to the participant. Influences on acceptability include: characteristics of the measure, such as the ease of completion or complexity of a measure, length and appearance; format factors, such as whether it is interviewer-administered or self-completed; cultural factors, such as language, method of translation and applicability (Fitzpatrick *et al.* 1998; Carr *et al.* 2002; Jokovic *et al.* 2002). Several generic HRQoL measures for children have been criticised for being unacceptably long, including the 87-item Child Health Questionnaire (Landgraf *et al.* 1998) and the 188-item Child Health and Illness Profile (Starfield *et al.* 1993; Ravens-Sieberer *et al.* 2006).

The instrument must be acceptable to participants to minimise stress and obtain high response rates. A questionnaire that is difficult to complete may result in participants omitting to answer some questions or the whole questionnaire. The method of administration can also affect the response rates (Fitzpatrick *et al.* 1998).

2.5.2.3.8 Feasibility

In addition to participant acceptability, it is important to evaluate the ease of administration and processing from the researchers' perspective. Consideration needs to be given to costs, training needs and time demands (Fitzpatrick *et al.* 1998; Carr *et al.* 2002).

In summary, assessment of a HRQoL measure should include whether it is conceptually clear, valid, reliable and if the scoring system is interpretable and sufficiently precise. It needs to be acceptable to both the participants and the researchers.

2.5.3 Oral health-related quality of life (OHRQoL)

2.5.3.1 Oral Health

The most frequently used definition of oral health in the UK is:

Oral health is a standard of health of oral and related tissues which enables an individual to eat, speak and socialise without active disease, discomfort, embarrassment and which contributes to general well-being (Department of Health 1994).

This definition makes reference to functional, psychological and social concerns and reflects the characteristics of the biopsychosocial model of health. Its focus is not on the oral cavity itself, but on the individual and the way in which oral diseases and conditions

impact on health, well-being and the quality of life; it is 'person centred'. rather than 'mouth-centred'(Locker 1997).

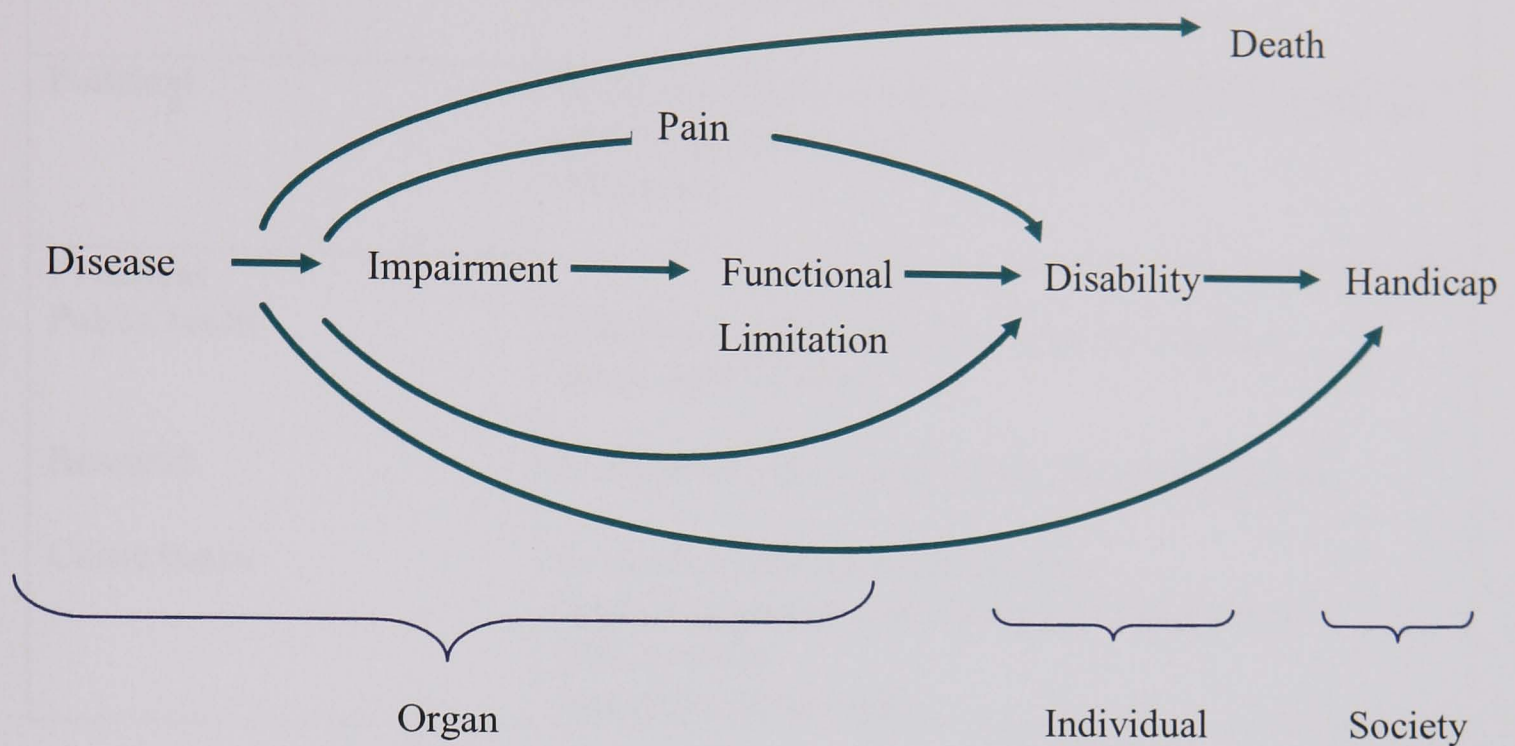
2.5.3.2 Measuring oral health

Assessments of oral health by epidemiologists, dental public health professionals and health services researchers have, traditionally, relied on the biomedical approach of using clinical indicators (Gift 1996). Measures most often employed to record oral status are measures of existing or past dental disease, rather than health (Corson *et al.* 1999). This is also the case for the indices used for DDE outlined earlier. However, clinical indicators rely on the dental professional's judgements, give no indication about the functioning of the oral cavity or the person as a whole and say nothing about subjectively-perceived symptoms, such as pain and discomfort (Locker 1997).

Cushing and colleagues suggested that dental diseases are ideally suited to the use of subjective measures, alongside objective measures, because they are largely social and behavioural in origin and are experienced by people in social and psychological ways (Cushing *et al.* 1986). Socio-dental indicators, defined as 'measures of the extent to which dental and oral disorders disrupt normal role functioning' (Locker 1988) were developed to capture the full impact of oral disorders within populations (Slade 1996).

However, Locker (1988) suggested that the term 'socio-dental indicators' and the concept of health on which they are based are too narrowly-defined to do full justice to the range of events involved in the measurement of oral health. The term 'indicator' is generally used when documenting the health of populations, whereas 'individual measures' are needed to reflect the level of health of individuals or groups. Recognising that new measures should be based on a contemporary model of health, Locker adapted the WHO International Classification of Impairments, Disabilities and Handicaps (ICIDH) (World Health Organisation 1980). His model of oral health depicts disease, impairment, functional limitation, disability and handicap as a related sequence of events (Figure 5) (Locker 1988). This model is broadly compatible with the Wilson and Cleary model (Figure 4), with disease and disability/handicap broadly equivalent to biological/physiological variables and functional status respectively. Although the ICIDH has been superseded by the ICF, Locker's model remains in use.

Figure 5. Locker's model of health (Locker 1988)



2.5.3.2.1 Application of OHRQoL

The appropriateness of linking HRQoL to the oral cavity has been widely endorsed. In keeping with the approaches throughout this thesis, the quality of life perspective allows assessment of the impact of different oral diseases and exploration of the individual factors that influence the extent of the impact.

The potential applications of OHRQoL measures can be categorised into theoretical, political, and practical (Locker 1996)(Table 8).

Table 8. Summary of the applications of OHRQoL measures

Theoretical	Exploring models of oral health Describing factors influential to health
Political	Demonstrating involvement of the public in healthcare Identifying the public's priorities Advocacy
Practical Public health Research Clinic based	Planning, monitoring and evaluating services Needs assessments Evaluating outcomes of healthcare interventions Evaluating individual patient care Improving patient-practitioner communication Clinical audit Marketing of services

2.5.3.2.1.1 Theoretical applications

Potential theoretical applications include: exploring models of oral health and disease, describing income inequalities and determining influential psychosocial factors (Locker 1996). In addition, other theoretical applications include elucidating the relationship between different aspects of oral health (Robinson *et al.* 2002). This approach, therefore, potentially enables research on DDE to extend beyond simple presence or absence. It allows research to explore the broader psychosocial consequences of these conditions.

2.5.3.2.1.2 Political applications

Advocating for resource is a political application of these measures. This is particularly relevant for oral health services and research, which is often isolated from mainstream health care systems. Such measures could place oral health in context, by showing whether oral conditions such as DDE have an impact, thereby giving oral health legitimacy with policy makers and government (Reisine 1985). Other applications described include harnessing public opinion and encouraging lay involvement in policy making (Robinson *et al.* 2002). With the increasing political emphasis on patient and public involvement in healthcare (Department of Health 2001), the findings of studies of OHRQoL have the potential for demonstrating how policy makers are consulting with, and listening to, the

public. The results could be used to identify the public's priorities and to advocate for changes in policy, for example in the provision of oral health promotion programmes (Gift 1997). Political demands exist, particularly as healthcare costs rise, for evidence of the benefits of treatments, to ensure best use of resources (Department of Health 1997). Taking into account the political imperatives of evidence-based healthcare, evidence is required of cost-effectiveness, clinical effectiveness and benefits, as perceived by patients, carers, health care professionals and society as a whole (Gift 1997). OHRQoL measures may therefore be ideally suited to fulfilling some of these requirements.

This literature review has shown that, in relation to DDE, there is a need to include the views of the population. This is especially the case for fluoridation, as part of risk-benefit assessments for caries reduction strategies. The increased risk and impact of fluorosis should be evaluated against the reduced risk and impact of caries. The comparison of the impact of these conditions would inform debate and decision-making, about fluoridation schemes.

2.5.3.2.1.3 Practical applications

The potential practical uses of OHRQoL measures are for research, public health and in clinical practice. In research terms, OHRQoL can be used as an outcome measure in clinical trials (Fitzpatrick *et al.* 1998), with oral health measures developed specifically for this purpose (Locker *et al.* 2005).

Public health applications include: describing and monitoring the health status of populations, the results of which can be used to assess population needs; identifying target populations; priority setting. Such measures could also be used to screen population groups, to identify the need for referral for dental treatment (Weintraub 1998).

In clinical practice, measures can be used to monitor and evaluate individual patient care (Corson *et al.* 1999). Other clinical applications include facilitating communication and identifying patient preferences and as a means of marketing dental services to patients (McGrath and Bedi 1999).

Some of these potential applications may never be realised due to such barriers as ideology, lack of interpretability, difficulties choosing a measure and concerns over validity. However, many measures of OHRQoL have been developed and used over the past decade and their use is increasing (Allen 2003). As we have seen, there does not seem to have been widespread use of such measures for assessment of the impact of DDE.

2.5.3.3 Measures of OHRQoL

Measures of OHRQoL differ in content, dimensions assessed, weightings, psychometric properties, assessment of frequency and formats. The formats differ in length, response choices and methods of administration (Weintraub 1998). Most have been developed for use with adult populations (Table 9).

Table 9. Measures of oral health status and OHRQoL (Allen 2003)

Name of measure
Social Impacts of Dental Disease (Cushing <i>et al.</i> 1986)
Geriatric Oral Health Assessment Index (Atchison and Dolan 1990)
Dental Impact Profile (Strauss and Hunt 1993)
Oral Health Impact Profile (Slade and Spencer 1994)
Subjective Oral Health Status Indictors (Locker and Miller 1994)
Dental Impact of Daily Living (Leao and Sheiham 1996)
Oral Impacts on Daily Performances (Adulyanon and Sheiham 1997)
OH-QoL UK (McGrath and Bedi 2001)

Different approaches have been taken in their development, but only two are explicitly based on a theoretical model of oral health, namely: OHIP and OIDP. These two measures are described briefly below.

2.5.3.3.1 Oral Health Impact Profile (Slade and Spencer 1994)

OHIP has been the most widely applied instrument in the UK (Corson *et al.* 1999). It was intended to provide a comprehensive measure of self-reported dysfunction, discomfort and disability, arising from oral conditions with the purpose of providing information about the burden of illness and the effectiveness of health services in reducing that burden (Slade 1997a). The development of OHIP involved three steps. First, Locker's model was used to

define seven dimensions of impact: functional limitation, physical pain, psychological discomfort, physical disability, psychological disability, social disability and handicap. Interviews of Australian adult dental patients were then conducted to yield statements about the adverse impacts of oral conditions, which resulted in 46 statements, categorised into the seven dimensions. Three additional statements were included in the handicap dimension. OHIP consists of 49 items, with a reference period of twelve months.

OHIP has acceptable levels of reliability, including internal consistency (Cronbach's alpha = 0.70-0.83 for six subscales, handicap subscale = 0.37), and reproducibility (Intra-class Correlation Coefficient = 0.42-0.77 for six subscales, social disability subscale = 0.08). Construct validity was assessed against perceived need for a dental visit, with statistically significant findings for five of the subscales (pain/discomfort, functional limitation, physical pain, psychological disability, psychological discomfort) and was described as moderate (Slade and Spencer 1994).

A short form, OHIP-14, has since been developed, because it was felt that, for some research settings and types of participants (including children), the use of the full questionnaire was unacceptable. OHIP-14 has been found to have good internal consistency, validity and precision (Slade 1997b). Robinson and colleagues examined the properties of OHIP-14. Internal consistency was in the range of Cronbach's alpha = 0.88-0.92. Face and content validity were reasonable, correlations between OHIP-14 and pain and global oral health ratings indicated acceptable criterion validity and construct validity was adequate for use in the UK (Robinson *et al.* 2003).

OHIP-14 was used to assess the impact of dental disease in the UK Adult Dental Health Survey 1998 (Kelly *et al.* 2000). Half (51%) of dentate adults experienced one or more of the problems (mostly in the categories of physical pain, psychological discomfort and psychological disability) occasionally or more often in the previous 12 months.

2.5.3.3.2 Oral Impacts of Daily Performance (Adulyanon 1996)

OIDP aims to measure serious oral impacts on a person's ability to perform daily activities. It was derived from a modified version of Locker's conceptual model focussing predominantly on handicap and ultimate impacts. It assesses impact of oral disorders on

seven daily tasks and attempts to quantify frequency and severity of impacts. OIDP was designed for use in adults with an interview format and uses a six month reference period.

Internal consistency was good (Cronbach's alpha = 0.88-0.92). In UK evaluations, face validity was poor (because it contained contingency questions). It had reasonable content validity. OIDP correlated with both the global oral health rating and the pain rating, indicating acceptable criterion validity. The precision and construct validity of OIDP was questioned, due to difficulties transforming the data (Robinson *et al.* 2003).

As stated in section 2.4.2.2.3, OIDP was used to investigate the impact of DDE on children in Tanzania and Brazil (Astrom and Mashoto 2002; Michel-Crosato *et al.* 2005). In these studies, a version of OIDP was used without evaluation of the modified measure for use in these countries or for use in children. As OIDP was developed for use with adults, the use of this measure may be inappropriate.

2.5.4 Qualitative approaches to impact

An additional approach to investigating the impact of health and illness is qualitative research. This approach emerged about fifteen years ago to improve knowledge of the way specific conditions impact on people's lives (Skevington 1994). It was acknowledged that quantitative measures often cannot reveal the range or depth of individuals' subjective experiences of health and illness (Fontana and Frey 1994). Since then qualitative research has been widely used, both alone and in combination with quantitative research, to answer questions of the impact of chronic illness (Gerhardt 1990), including mental illness (Cutting and Dunne 1989), diabetes (Dzurec 1990), chronic pain (Kortesluoma *et al.* 2003), epilepsy (Bishop and Allen 2003), endometriosis (Jones *et al.* 2004) and sickle cell disease (Thomas and Taylor 2002).

In oral health qualitative methods have been used alone to explore the impact of dental pain in adults in the UK (Pau *et al.* 2000) and in combination with quantitative measures to investigate the impact of oral health on older people in Canada (MacEntee 1996). Seemingly, little has been published specifically on the impact of oral health on children and young people. One study explored the experiences of young people with Treacher Collins syndrome (Beaune *et al.* 2003) and several studies have explored children's general

perceptions of oral health (Ostberg *et al.* 2002; Fitzgerald *et al.* 2004). This review of the literature has not been able to identify studies that have explored the impact of DDE on children and young people using qualitative methods.

The following section will describe qualitative research, the methods used and how it is evaluated. It will then describe how qualitative studies have been used to explore the impact of oral health in adults.

2.5.4.1 Defining qualitative research

There is no consensus definition of qualitative research, but it is generally agreed that it is an interpretive, naturalistic approach, that attempts to make sense of, or to interpret, phenomena in terms of the meanings people attach to aspects of their social world (Denzin and Lincoln 2000). The perspective generally taken is to explore the ideas and perceptions of the participants, with the researchers taking an ‘insiders’ view’ to examine the experiences, feelings and perceptions of the people they study (Holloway and Ellwood 1997). Thus, qualitative research takes the ‘emic’ perspective, by eliciting meaning from the participant’s point of view, rather than the researcher’s view or ‘etic’ perspective (Morse 1992).

Qualitative research is often defined in terms of its differences from quantitative research (Brannen 2004). For example, qualitative research uses words, while quantitative research uses numbers. Qualitative research is largely concerned with meanings, as opposed to behaviours. However, as the two methods are increasingly used together (Hammersley 1992), it is more helpful to consider them as complementary (Bower and Scambler 2007).

2.5.4.2 Data collection methods

Qualitative research aims to provide an in-depth understanding, so methods such as observation, interviews and focus groups are used, with a small number of people selected for the purpose. Interviews and focus groups accommodate emerging topics, with data generated from the interaction between the researcher and the participants (Legard *et al.* 2003).

Qualitative interviews can be semi-structured or unstructured (also known as in-depth), depending on the nature of the information being sought, with semi-structured interviews having a more specific focus (Crabtree and Miller 1999). Features of qualitative interviews include:

- combining structure with flexibility, to allow responsiveness to relevant topics raised by the participant.
- the use of a range of probes and other techniques to achieve depth of answer in terms of penetration and explanation.
- the generation of new thoughts occurring to the interviewee that they had not explored before (Kvale 1996).

A focus group is a systematic discussion planned on a defined topic of interest. The group should be relatively homogenous, with a supportive and permissive atmosphere (Kruger 1998). The size of groups varies from 3 to 12 participants, with an optimum of 6 or 7 (Holloway 1997).

The choice of method of interviews, focus groups, or a combination, is determined by the nature of the topics, with greater breadth of information from groups, but more depth from interviews (Crabtree and Miller 1999).

2.5.4.3 Analysis of qualitative data

Analysis of qualitative data should result in a detailed description of the data, which identifies patterns and develops explanations, while remaining faithful to the data in its original form (Sandelowski 1995). Methods differ, for example, some approaches, such as symbolic interactionism, focus on language. Others, such as ethnography, focus on understanding or the building of theory, such as grounded theory (Tesch 1990).

Methods of analysis include:

- Content analysis

Content analysis involves systematic analysis of the content of verbal or written communications by categorisation and classification (Weber 1990).

- Conversation analysis

Conversation analysis was developed from ethnography and involves studying the structure and patterns of interactions, rather than just the content in terms of words (Silverman 2000).

- Narrative analysis

Narrative analysis is concerned with identifying a basic story and focuses on the way the story is constructed and the meaning of the 'plot' (Reissman 1993).

- Constant comparative analysis

Constant comparative analysis involves characterisation of the variation within data by looking for comparisons and differences (Glaser and Strauss 1967). Although it was developed for use in grounded theory, the method is often used in other approaches (Thorne 2000).

- Framework

Framework analysis is concerned with classifying data by organisation according to themes and emerging categories. It has developed from social policy research to expedite handling large volumes of data (Ritchie and Spencer 1994).

2.5.4.4 Quality in qualitative research

In quantitative research, reliability and validity serve as evidence of rigour. Indeed, the properties of (oral) HRQoL measures are often evaluated in this way (section 2.5.2.3). These properties can also be applied, in a broad sense, to qualitative research (Emden and Sandelowski 1998; Seale *et al.* 2004).

2.5.4.4.1 Reliability

Reliability concerns the reproducibility and consistency of the findings (Hammersley 1992). Others have described this as 'two levels of replicability: external and internal' (Lewis and Ritchie 2003). External replication concerns whether similar findings would occur if another study was undertaken (Seale *et al.* 2004). Methods to ensure external replication involve recording, in detail, procedures such as: sample selection, data collection, method of analysis and providing interpretations that are well supported by evidence. Internal replication relates to the extent to which factors or judgements are replicated between researchers, for example the consistency with which instances are assigned to the same category by different observers (Hammersley 1992).

2.5.4.4.2 Validity

Validity refers to how far the findings reflect the subject under study, in terms of representation, understanding and interpretation. External validity, also known as plausibility, (Glaser and Strauss 1967) refers to verifying findings externally, for example triangulation and participant validation. Forms of triangulation include: methods triangulation (comparing data generated by different methods, such as qualitative and quantitative), triangulation of sources (comparing data from different qualitative methods), triangulation through analysis by different researchers and theory triangulation from different theoretical perspectives. Participant validation involves asking the same participants to confirm the interpretation of their data (Lewis and Ritchie 2003) or to ask other participants to comment on this interpretation. The latter technique is known as continuous feedback.

Internal validity, sometimes referred to as credibility (Glaser and Strauss 1967), involves the degree to which the findings accurately reflect the study population. Methods of internal validation include checking the accuracy of fit of the explanations by comparison between different parts of the data (Silverman 2000).

Validity is closely linked to generalisability. A common criticism of qualitative research is the perception of a lack of generalisability of the findings (Ritchie and Spencer 1994).

2.5.4.4.3 Generalising from qualitative research

Generalisation is concerned with whether findings from a study sample can be relevant beyond the sample and the context of the research. Two kinds of generalisation have been described: empirical and theoretical (Hammersley 1992).

2.5.4.4.3.1 Empirical generalisation

Empirical generalisation concerns two aspects of the applicability of findings from qualitative research, namely: representational and inferential generalisation (Lewis and Ritchie 2003).

Representational generalisation refers to whether the phenomena found in the sample would similarly be found in the parent population. The degree to which this is the case depends on the quality of sampling, data collection, analysis and interpretation in the study.

Inferential generalisation, also termed ‘transferability’, is concerned with generalisation to other settings and contexts. The application of findings to other situations depends on sufficient detail being given to allow an assessment to be made of the similarity of two settings and whether transfer of findings might be appropriate.

2.5.4.4.3.2 Theoretical generalisation

Theoretical generalisation draws theoretical principles from the findings of a study for wider application (Gobo 2004). This type of generalisation includes using findings from a study to support or develop existing theories or to generate theories for further testing.

2.5.4.4.4 Reflexivity

In addition to reliability and validity, reflexivity is another important aspect of quality in qualitative research. Reflexivity is the term used to express the ways the researcher and the research process have shaped the data. Assumptions and personal and intellectual biases should be considered during the formulation of the research question, data collection and interpretation, including how the researcher responded to events during the study (Mays and Pope 2000). Systematic and self-conscious research design, data collection, interpretation and communication enhance quality in qualitative research (Mays and Pope 1995).

2.5.4.4.5 Quality assessments

Checklists and guidelines, specifically designed for qualitative research can be applied to assure the quality of reported research findings (Seale 1999; Public Health Resource Unit. 2002). Checklists can play a role in answering criticisms about rigour, but the nature of qualitative research means over-zealous use of checklists can be counterproductive (Barbour 2001). The quality of dental qualitative research was found to be mediocre, with deficiencies in the areas of research design, methodological rigour, reflexivity and the presentation of findings. The authors concluded that, while qualitative methods were under-utilised in oral health research, to improve the quality in future work, guidelines should be employed (Thaliath *et al.* 2006).

2.5.4.5 Qualitative research of the impact of oral conditions

Qualitative methods has been used alone and in combination with quantitative measures to explore the impact of oral health in adults (MacEntee 1996; Pau *et al.* 2000). Medline searches for this narrative review have identified few reports of research on the impact of either oral conditions in general or DDE on children and young people using qualitative methods.

Pau and colleagues (2000) identified a gap in the literature regarding adults' subjective experience of toothache and so explored the impact of dental pain on those attending an emergency clinic. Thirty-five adults were interviewed by a dentist in a clinical setting. Framework analysis was used, with two researchers categorising the data. The key themes to emerge were the perceived inability of people to cope with toothache and the care-seeking patterns of the participants. Quotes were used in the article to illustrate these themes (Pau *et al.* 2000). The main limitation of this study was the lack of reflexivity, particularly regarding the setting for the research (the participants were interviewed in a room in the clinic while waiting for treatment) and the influence of the dentist conducting the interviews. The study did not include a discussion of the generalisability of the findings, either beyond the sample or in other settings.

A Canadian study used a combination of quantitative and qualitative methods to investigate the impact of oral health of older adults. A large quantitative survey was complemented by a smaller study involving in-depth interviews. Participants were interviewed until the data were saturated and no further information emerged (n = 24). The data were analysed using constant comparative method, with attention paid to reliability and validity, although discussions about reflexivity were absent. The findings of the qualitative study complemented the quantitative data and revealed additional insight into the participants' perspectives and experiences, particularly how older people find ways to adapt positively to the changes in their oral health (MacEntee *et al.* 1997).

In summary, qualitative methods have been used alone and in combination with quantitative methods to explore the impact of oral conditions on individuals. The findings

from such studies allow the participants' perspectives to be gained in their own words, without imposing the rigid format of questionnaires.

2.5.4.6 Conclusion

In order to investigate the impact of DDE on individuals, current concepts suggest research needs to be based on an appropriate conceptual model of health and to consider the important properties of HRQoL measures. To date, there has been minimal use of OHRQoL measures in relation to DDE and most studies have used measures designed for use with adults. Qualitative methods, offer complementary opportunities to explore, in detail, the perspective of the participant, particularly in terms of what having a condition means to them. No qualitative research has been conducted on the impact of DDE.

A substantial body of research now critiques the use of 'adult' measures, and methods more generally in child populations. Research with children involves a number of important theoretical and methodological concerns that will now be reviewed.

2.5.5 Research with children

2.5.5.1 Changing position of children in society

Over the past 30 years children have moved to the forefront of personal, political and academic agendas (James *et al.* 1998).

These changes are partly due to demographic shifts in the population. In the UK, the proportion of the population aged under 16 has declined since the 1970s (Office for National Statistics 2006). In addition, there has also been a change in family structure, with an increase in one-parent families (Office for National Statistics 2004). Collectively, such changes have led to the idea of the child as a scarcity and thus more 'precious' (Zelitzer 1985).

Political legislation has reflected this trend, with changes to children's rights, both globally and nationally. The Convention on the Rights of the Child is the most universally accepted human rights instrument in history, being ratified by every country in the world, except for the USA and Somalia (United Nations 1989). The Convention confirms that children have

a right to have their views taken seriously and given due weight. In English law, the Children Act 2004, requires welfare agencies to take account, not only of children's best interests, but also the wishes and desires of individual children (Office of Public Sector Information 2004). The English government has also shown its commitment to ensuring health and other services are child-centred (Department for Education and Skills 2003; Department of Health 2003). The National Service Framework for Children, Young People and Maternity Services (Children's NSF) requires services to give children and their parents increased information, power and choice over the treatment they receive and involve them in planning their care.

Academically, this changed position of children in society has had implications for the involvement of children in research, particularly within the social sciences. This has been reflected in evolving approaches to the sociological study of childhood through three phases: pre-sociological, transitional and sociological (James *et al.* 1998).

2.5.5.2 Pre-sociological model

Up until the 1970s much of the research was based on a pre-sociological model of childhood. There were two main features of this model: childhood was viewed without regard for the social structure in which children live and children were seen as developmentally-incomplete adults. Piaget and others attempted to define the stages of child development, from infant through early childhood, middle childhood, adolescence to adulthood (Piaget 1972; Bee and Boyd 2004). Piaget's idea, that development takes place in stages, has been questioned, in view of its consequences for children who do not achieve standardised developmental targets. Within the Piaget model, research often took the form of age-based studies of children, without explaining why a particular age group was chosen.

2.5.5.3 Transitional model

The transitional model was also based on a developmental scheme but, as ideas of how society shapes the individual emerged in the 1970s, the emphasis shifted to the 'socially developing child'. Children were seen as 'waiting to be processed through the particular rite of passage that socialisation within society demands'. Researchers adopting this approach gave little time to children themselves, tending to conduct research *on* children.

2.5.5.4 Sociological model

As more weight was given to the rights and views of the child, the ‘voices’ of children became increasingly recognised in research. The sociological model brought with it the concept of child-centred research that:

- Regards children as competent and reflexive in reporting their own experiences.
- Gives children a voice and taking seriously what they say.
- Works for and *with* children, rather than *on* them (Mayall 1996).

These models demonstrate the evolution of studies of childhood, from research *on* children to research *with* children and the adoption of the concept of child-centred research. Such child-centred methods are appropriate for research of the impact of DDE on individual children and young people, with several important considerations for research of this kind.

2.5.6 Methodological considerations of research with children

Several methodological considerations require attention when the researcher is an adult and the participant a child: power imbalance, language use, setting for the research, analysis and quality of the data.

2.5.6.1 Power imbalance

Children have, in the past, been excluded from research. Researchers have preferred to ask adults, such as parents or teachers, to report on children’s lives, rather than ask the children themselves. Research conducted *on* children assumes the superiority of adult knowledge. Social studies of childhood adopt a stance that doesn’t assume adult superiority. Instead, children are asked to help the adults, understand childhood so that they can gain access to children’s own unique knowledge (Mayall 2000). Research conducted *with* children minimises differences, in an attempt to reach understandings of children’s own perspectives.

A number of authors have discussed the role an adult should employ when researching young people, to alter the power relations between adults and children. Roles such as non-authoritarian adults, ‘friends’ and ‘least adult’ are recommended.

Other recommendations to reduce the power imbalance include research methods that allow children to shape the agenda, methods that focus on real life events and participatory techniques (Morrow 1999; Backett-Milburn *et al.* 2003). Other strategies include: providing child-centred information, giving children the choice to participate (as well as the choice of when, where, how and with whom the research takes place), maintaining confidentiality, the use of humour, seeking children's views on how to improve the research and valuing the time children give (O'Kane 2000).

2.5.6.2 Language use

Children's use of language differs from that of adults. Children may experience difficulties in comprehension leading to discrepancies between the children's understanding and the researcher's. However, the difficulties can be two-way, with researchers also misunderstanding children's language (Punch 2002).

2.5.6.3 Setting

The setting for the research is especially important with children, as it may influence their responses. The expression of the child's personality, in terms of their attitudes and behaviour, is often more context-dependent than that of adults (Scott 2000).

Children are most often studied in schools. School-based research is typically more cost-effective than at home, but children's responses may be influenced by the presence of teachers or class mates. Research conducted at home may help some children view the researcher as an interested adult, rather than as a figure of authority (Faux 1988). However, the home may not always be an ideal space: the regulation and discipline, like those of the school can be problematic for some children. In either environment, complete privacy is often elusive. Moreover, these settings, created by adults and designated as 'places for children', may not be settings they themselves relate to (Rasmussen 2004). Oral health research often studies children in clinical settings which, due to its inherent hierarchy and patient's anxiety, may influence children's responses.

2.5.6.4 Analysis

The analysis and interpretation of data from children can reflect the researcher's, rather than children's, beliefs and concerns (Qvortrup 1994; Woodhead and Faulkner 2000). It is important, therefore, not to impose an 'adultist' view on the data gathering and analysis, but to be receptive to children's own interests and concerns. The influences of both academic and personal preconceptions on the processes of interpretation of data from children must be considered (Davis *et al.* 2000).

2.5.6.5 Quality of data

Researchers working with children are often asked if they can 'really believe' children's accounts of their experience (Morrow 1999). Regardless of the age of the participant, there is no evidence that bias (including acquiescence and social desirability bias) is greater in data from children than adults (Scott 2000). It has been suggested that if children are not providing valid and reliable data, it is not the fault of the child, but of the researcher.

Methods suggested to assure the quality of data include assuring confidentiality, developing a rapport between researcher and child, giving the child unambiguous and comprehensive instructions at the start, asking questions relevant to children's own experience, avoiding leading questions and permitting 'don't know' responses to avoid guesses (Punch 2002).

2.5.7 Contemporary approaches to research with children

The shift towards child-centred research has resulted in improved methods that are sensitive to the competence of children and young people. The emphasis is on capturing their perspectives and putting them at ease with adult researchers. Methods may be quantitative or qualitative, such as questionnaires, interviews (individually or in groups) and participatory techniques such as drawing, photographs, diaries and worksheets (Harden *et al.* 2000; Backett-Milburn *et al.* 2003). These methods represent a hierarchy of involvement of children.

Questionnaires, though completed by children, are often designed by adults. Therefore, while they are useful to answer certain research questions they may not capture all aspects relevant to the young people. Focus groups and interviews seek information directly from them in their own words, rather than 'what adults think children think' (Alderson 1995;

Borland *et al.* 1998; Morrow 1999). They have been used successfully with children from six years of age (Docherty and Sandelowski 1999; Heary and Hennessy 2002). However, some difficulties may arise with interviews, individually or in groups if spontaneous conversation is not forthcoming, particularly with young children. Participatory techniques, that involve 'handling things' rather than 'just talking' have also been used (Morrow 1999; Backett-Milburn *et al.* 2003). For example, drawing is something children often do on their own or with friends. They often chat as they draw and this technique can provide the opportunity to further engage children to participate in research. However, some difficulties may arise generating useful data for analysis (Pain and Francis 2003).

In a review of methods for conducting research *with* children, Punch concluded that both traditional and innovative methods can be used to overcome some of the issues of research *with* children, but the choice of method for a study should depend on the aim of the study, the experience and preference of the participants and the competencies of the researcher (Punch 2002).

In summary, changes in the position of children in society and legislation to promote the rights of children have resulted in methodological shifts in the way research is conducted with children. The review of the existing literature on the impact of DDE on children and young people identified several potential concerns, namely: the choice of questions used to assess impact and the use of modified OHRQoL measures, designed for adults. Bearing in mind the emphasis on child-centred research and the methods currently available to ensure research is conducted *with* children, this thesis attempts to embrace this approach as far as possible.

2.5.8 Measurement of HRQoL of children

Work on HRQoL in children is less well-developed than in adults and has mainly been in the fields of oncology, rheumatology, asthma and epilepsy (Vincent and Higginson 2002). A recent literature search identified more than 50 HRQoL instruments in children, of which nine were generic (Ravens-Sieberer *et al.* 2006).

Methodological challenges to the measurement of health-quality of life arise in three main areas: the concept of HRQoL in children; problems caused by the changes children undergo both physically and cognitively; the use of a proxy.

2.5.8.1 Difficulties with the concept of HRQoL in children

There is neither a consensual definition nor a conceptual model of health or HRQoL in children (Ravens-Sieberer *et al.* 2006). Existing measures tend to be based on adult models or use quality of life questionnaires that are adapted from adult measures (Titman *et al.* 1997). These approaches reduce the content validity, as items in adult questionnaires may not be relevant to children and may not address aspects of daily life that children value. This is a central requirement of HRQoL measures (Guyatt *et al.* 1986). For example, some HRQoL measures have been adapted through the substitution of adult items, such as ‘work’ with ‘school’ (Eiser and Morse 2001). In a review of HRQoL measures for children, Vincent and Higginson found most existing scales focus on physical and symptom-related aspects of HRQoL and do not incorporate school-related items. They concluded that if the content of questionnaires was to reflect the main areas of children’s lives, factors such as family and social relationships, activities and schooling should be included (Vincent and Higginson 2002).

2.5.8.2 Cognitive and physical changes in children

The measurement of HRQoL is complicated by the rapid changes seen as children develop (Allison *et al.* 1997; Jokovic *et al.* 2002; Ravens-Sieberer *et al.* 2006). These changes have implications for reading levels, type of scales, time frames and general comprehension of questionnaires (Vincent and Higginson 2002). To some extent, simple questionnaires, pictorial representations and the assistance of an interviewer may overcome some of the problems in children’s levels of literacy and understanding, but different scales for different age-groups have also been suggested. The Child Asthma Questionnaire (CAQ) has forms for three different age groups (4 to 7 years, 8 to 11 years and 12 to 16 years) tailored to the level and life context (Christie *et al.* 1993). The CAQ has demonstrated it is possible to obtain valid and reliable reports of HRQoL from children (French *et al.* 1994).

2.5.8.3 Use of proxies

Information from parents or carers can be used to supplement children's assessments of HRQoL or can be used as a proxy for the child assessment. Such information can be useful, as it may influence decisions about clinical management. Furthermore, involving parents in the assessment of their child's quality of life provides an opportunity to raise awareness in parents/carers (Inglehart *et al.* 2002).

The use of proxies may be the only solution for very young or sick children (Pantell and Lewis 1987). Most HRQoL measures for children under six years of age are proxy measures (Eiser *et al.* 2000). However, only modest agreement is found between parents and children's reports of HRQoL (Achenbach *et al.* 1987). Eiser and Morse's systematic review of the relationship between the ratings of children's quality of life made by parents and children (Eiser and Morse 2001) indicated that the accuracy of proxy ratings depends on the specific domains of quality of life considered. There is greater agreement for observable functioning (e.g. physical quality of life) than for non-observable functioning (e.g. emotional or social quality of life). Agreement is better between parents and chronically sick children compared with parents and their healthy children (possibly due to greater communication about illness and treatment). However, the importance of any agreement and the common assumption that information from proxies should 'match' that provided by children is questioned. The standard practice, in developing new measures, of reporting validity by determining correlations between child and proxy ratings, is also highlighted. Although strong correlations between child and proxy data demonstrate some validity, it should not be assumed that they are interchangeable. The conclusion reached by the review is that differences between ratings made by children and adults had to be anticipated and treated as important.

Where possible and in keeping with conducting research *with* children, information from parents should only be used to complement the views of children.

2.5.9 Measurement of OHRQoL in children

Seemingly few studies have assessed OHRQoL in children or young people generally, this is not just a deficiency in the DDE literature. As was the case in existing research on DDE,

some studies have used parents as proxies for children or children have completed questionnaires designed for adults without evaluation of the measures for this new purpose.

2.5.9.1 Parents as proxies

As stated in section 2.5.8.3, the use of proxy assessments of OHRQoL is appropriate for young (under six years of age) or sick children (Pantell and Lewis 1987). Two studies have used parent's assessments of the OHRQoL of their children before and after dental treatment under general anaesthesia (Anderson *et al.* 2004; Baens-Ferrer *et al.* 2005). In one study, the children involved had a mean age of 5.1 years, these children were too young to complete a quantitative measure themselves (Anderson *et al.* 2004). In the second study, the mean age was 9.6 years, no comments were made by the authors why they had relied on the parents as proxies instead of asking the children themselves (Baens-Ferrer *et al.* 2005).

As stated in section 2.4.2.2.2, the UK Children's Dental Health Survey 2003 questionnaire included, for the first time, a section on the impact of oral health. Parents alone completed 91% of the questionnaires with the remainder completed by parents with the assistance of their child. Proxies were used as the authors stated that a measure designed for children themselves had not been evaluated for use in the UK and that the sample included children of a wide age range (5 to 15 years) and therefore differing competencies to complete a questionnaire (Nuttall and Harker 2004). Clearly such research falls some way short of being sufficiently inclusive of children's views.

2.5.9.2 Use of 'adult' OHRQoL measures with children

In addition to the studies already described of the use of adult OHRQoL measures to investigate the impact of DDE on children, several other studies have also used such measures for an age group which they were not developed for. The relationship of two measures, a generic HRQoL measure RAND SF-36 (Ware 1993) and OHIP (Slade and Spencer 1994) to clinical indicators among young people was investigated in the USA (Broder *et al.* 2000). The young people were between 12 and 17 years of age. The authors stated that Rand SF-36 could be completed by individuals of 14 years of age although some participants in this sample were younger. The authors commented that this was the first study to use OHIP in young people. However, OHIP may not be appropriate for use in this

group, as it contains 49 items and much of the work evaluating OHIP has been in older adults (Slade 1996).

Translated versions of OHIP-14 have also been used without modification with young people in Uganda (12-20 years) (Astrom and Okullo 2003) and Myanmar (14-year-olds) (Soe *et al.* 2004). Aspects of OHIP-14 such as ‘satisfaction with diet’, ‘difficulty doing usual jobs’ and ‘finding life less satisfying’ may not be as appropriate for young people as for adults. The use of a measure developed for older adults was acknowledged as a limitation by Soe and colleagues (2004).

OIDP was used in Brazil with young people (12-14 years) without acknowledgement of the use of an adult questionnaire; no attempt was made by the authors to assess the validity for use in this age group (de Oliveira and Sheiham 2003). OIDP includes eight physical, psychological and social activities, one being ‘carrying out a major work or social role’. In a subsequent study, OIDP was modified, to include the item ‘school activities’, possibly as a substitute for ‘work activities’, but this was not made clear (de Oliveira and Sheiham 2004).

The inappropriateness of using an adult OHRQoL measure with children led to the development of several child-specific questionnaires. The following section will describe the current state of knowledge about these measures.

2.5.10 OHRQoL measures for children

Although the need for measures that are relevant to children, young people and their families has long been identified (Weintraub 1998) none had been developed until recently (Tapsoba *et al.* 2000; Jokovic *et al.* 2002). The first, the Oral Health Related Quality of Life questionnaires (ICSII-OHRQOL) (Chen *et al.* 1997) were developed to assess OHRQoL across nations, although it was not developed from a recognised conceptual framework of child health or oral health. It has been evaluated for use in New Zealand, Germany and Poland (Tapsoba *et al.* 2000).

Two further measures have been developed based on models of child oral health: Child Oral Health Quality of Life (COHQoL) questionnaires (Jokovic *et al.* 2002) and CHILD-OIDP (Gherunpong *et al.* 2004).

2.5.10.1 COHQoL questionnaires

The Child Oral Health Quality of Life (COHQoL) questionnaires were developed for children and young people with a wide range of oral and oro-facial conditions including caries, malocclusions and clefts. They were designed to have discriminative and evaluative properties. COHQoL currently consists of two Child Perceptions Questionnaires (CPQ): one for children aged 8 to 10 years (CPQ₈₋₁₀), and one for those aged 11 to 14 years (CPQ₁₁₋₁₄) as well as a Parental-Caregiver Perceptions Questionnaire (P-CPQ) (Jokovic *et al.* 2002; Jokovic *et al.* 2003; Jokovic *et al.* 2004). The measures were designed by a process recommended by Juniper (Juniper *et al.* 1996) and Guyatt (Guyatt *et al.* 1986) to ensure the final questionnaires contained items of the most relevance to children with oral and oro-facial conditions.

2.5.10.1.1 Child Perceptions Questionnaire (CPQ₁₁₋₁₄)

The first of the COHQoL measures to be developed was CPQ₁₁₋₁₄. First, a preliminary pool of 46 items was developed from a review of the literature. These items were modified, following comments from an expert panel of health professionals and parents. In-depth interviews were then conducted with 11 young patients to modify this pool further. Items for the final questionnaire were then selected, following an item impact study. Young people with oral disorders were asked if they had experienced the problem described in each item and were then asked to rate how important this problem was on a 5-point scale ('does not bother me at all' = 0 to 'bothers me very much' = 4). For each item, an impact score was calculated by multiplying the percentage of children who had experienced the problem by the item's mean importance rating. Items were assembled into 4 health domains and were then selected based on their ranking within these domains.

CPQ₁₁₋₁₄ contains 37 items, encompassing domains of oral symptoms, functional limitations, emotional well-being and social well-being. Domains suggested by the World Health Organisation guidelines were covered, including schooling, relationships and activities (World Health Organisation 1994). CPQ₁₁₋₁₄ asks about the frequency of events

in the previous three months using a 5-point Lickert scale (for children this time frame may reduce recall bias and be more appropriate than the six month period used by some adult measures). The questionnaire is self-completed.

The validity and reliability of CPQ₁₁₋₁₄ in Canada were assessed in a clinic-based sample of young people (Jokovic *et al.* 2002). The internal consistency of the scale was good (Cronbach's alpha = 0.91) and for the domains was acceptable to good. The intra-class correlation coefficient (ICC = 0.90) for CPQ₁₁₋₁₄ indicates excellent agreement. Construct validity was assessed by means of association between scale scores and two global indicators. There were significant correlations between CPQ₁₁₋₁₄ scale scores and global ratings of oral health ($p < 0.05$) and overall well-being ($p < 0.01$) in the expected direction. CPQ₁₁₋₁₄ also has acceptable reliability and validity in settings in New Zealand, Uganda and Saudi Arabia (Foster Page *et al.* 2005; Robinson *et al.* 2005; Brown and Al-Khayal 2006) (Table 10). However, no studies have reported on the acceptability, face or content validity of this measure. Before the studies described in this thesis, CPQ₁₁₋₁₄ had not been evaluated for use in the UK.

Table 10. Evaluation studies of COHQoL questionnaires

Measure	Country	Type of population
CPQ ₁₁₋₁₄ (Jokovic <i>et al.</i> 2002)	Canada	Clinic-based
CPQ ₁₁₋₁₄ (Foster Page <i>et al.</i> 2005)	New Zealand	School-based
CPQ ₁₁₋₁₄ (Robinson <i>et al.</i> 2005)	Uganda	School-based
CPQ ₁₁₋₁₄ (Brown and Al-Khayal 2006)	Saudi Arabia	Clinic-based
CPQ ₁₁₋₁₄ Short Forms (Jokovic <i>et al.</i> 2006)	Canada	Clinic-based
CPQ ₈₋₁₀ (Jokovic <i>et al.</i> 2004)	Canada	Clinic-based
CPQ ₈₋₁₀ (Humphris <i>et al.</i> 2005)	N Ireland	School-based
P-CPQ (Jokovic <i>et al.</i> 2003)	Canada	Clinic-based

In addition to studies to evaluate the measure, CPQ₁₁₋₁₄ has been used to compare the impact on OHRQoL of orofacial and craniofacial conditions with the impact of dental caries (Locker *et al.* 2005). The differences between the two groups were small, with significantly higher scores in the orofacial/craniofacial group compared to the caries group for the functional limitations and social well-being domains. The authors suggest the emotional and social well-being domains were the most important with respect to overall quality of life. Section 2.4.2.2.3 detailed the only identifiable study including the impact of DDE that used an OHRQoL measure designed for children. This study used CPQ₁₁₋₁₄ in a sample of 12-year-old school children in Uganda (Robinson *et al.* 2005).

Subsequently, 16- and 8-item short forms were developed, to increase acceptability and appropriateness for use in clinical and population-based surveys. Good reliability and validity of these short forms, for clinic-based Canadian children, has been confirmed (Jokovic *et al.* 2006).

A CPQ for 8-10 year olds, again, has exhibited substantial agreement, internal consistency and good construct validity in children from clinics in Canada (Jokovic *et al.* 2004) and in a school-based sample in Northern Ireland (Humphris *et al.* 2005).

2.5.10.1.2 Child Oral Health Impact Profile (COHIP)

More recently, the Child Oral Health Impact Profile (COHIP) has been developed for children aged 8-15 years in the US, based on the pool of items used to develop CPQ₁₁₋₁₄. COHIP has 34 items across six domains: oral health, functional well-being, social-emotional well-being, school-environment, self-image and treatment expectations. COHIP uses the same scoring system as CPQ₁₁₋₁₄. The total COHIP scale was found to have acceptable internal consistency, test-retest reliability and validity in children attending clinics in the US, although further modifications of the subscales were recommended before the measure is used more widely (Broder and Wilson-Genderson 2007; Slade and Reisine 2007). It has not been evaluated for use elsewhere.

The main difference between CPQ₁₁₋₁₄ and COHIP is the inclusion of items that reflect positive aspects of OHRQoL (Broder *et al.* 2007). However, the performance of positively worded items on OHRQoL questionnaires for children has recently been deemed to be unsatisfactory (Locker *et al.* 2007).

2.5.10.1.3 Parental-Caregiver Perceptions Questionnaire (P-CPQ)

P-CPQ is a measure of parental/carer perceptions of the OHRQoL of children. P-CPQ was not intended to be a 'proxy' measure, but as a supplement to the information obtained from the children. The main rationale for developing P-CPQ was that parents/carers are intimately involved in the health of their children, and children's treatment is as likely to be influenced by parental perceptions of need as it is by the needs of the children themselves. P-CPQ has 31 items and includes a 'don't know' response option.

P-CPQ validity and reliability were assessed in a similar way to CPQ₁₁₋₁₄ (Jokovic *et al.* 2003). P-CPQ showed good construct validity with significant associations between the total scores and global ratings of oral health ($p < 0.05$) and overall well-being ($p < 0.0001$). It had good internal consistency for the total score (Cronbach's alpha = 0.94) and for the subscales. The test-retest reliability indicated excellent agreement (ICC = 0.85). Different methods for managing the 'don't know' response did not affect the properties of P-CPQ (Jokovic *et al.* 2004). Again, before the studies described in this thesis, P-CPQ had not been evaluated for use in the UK.

The level of agreement between children's OHRQoL and that perceived by their mothers using CPQ₁₁₋₁₄ has been investigated (Jokovic *et al.* 2003). Agreement between mothers and children was good (ICC = 0.70) for overall scores, indicating substantial agreement. However, the results from the subscales show how the level of agreement varied according to the characteristics of interest, with agreement for the emotional and well-being subscales indicating only moderate agreement. This echoed the findings of the systematic review of HRQoL alluded to earlier (Eiser and Morse 2001). The authors concluded that mothers may be used as proxies in some circumstances, but advised the views of children were preferable.

2.5.10.2 CHILD-OIDP (Gherunpong *et al.* 2004)

A version of OIDP that suited children's capabilities and interests was developed for use with Thai school children aged 11-12 years (Gherunpong *et al.* 2004). Unlike the COHQoL questionnaires, CHILD-OIDP was designed to measure impact in order to estimate dental treatment need and for service planning purposes. The process began with the adult Thai version of the index, which was adapted following interviews with children and paediatric dentists. Changes were made to the language, sequence of questions, response options and recall period (reduced from 6 to 3 months). Nine daily performances were originally included and covered the main domains recommended in the WHO guidelines (World Health Organisation 1994). The scoring system quantifies impacts using both frequency and severity scores. A set of 18 pictures were developed, to improve the acceptability to children, and reduced the time taken from 20 minutes to 10 minutes. However, when internal consistency was assessed, the consistency of the performance 'doing light physical activity' was questionable, so this performance was removed. After

excluding this item, internal consistency was improved (Cronbach's alpha = 0.65). Reliability was almost perfect (kappa = 0.91). Validity was confirmed as children who perceived oral treatment need had much higher CHILD-OIDP scores than those who did not perceive a need ($p < 0.001$) and children who thought their overall oral health problems were severe had much higher CHILD-OIDP scores than those who perceived their problems as moderate and low ($p < 0.001$).

CHILD-OIDP has been used in Thailand where 89.8% of children had one or more oral impacts, with eating being the most common performance affected. The severity of impacts varied by performance, being high for eating and smiling and low for study and social contact. Sensitivity, toothache, oral ulceration and tooth exfoliation accounted for most of the impacts (Gherunpong *et al.* 2004).

CHILD-OIDP has also demonstrated satisfactory properties for use among 10-year-old school children in France (Tubert-Jeannin *et al.* 2005) and 10-11-year-olds in the UK (Yusuf *et al.* 2006). In the UK study, face and content validity of Child-OIDP were assessed qualitatively in a pilot study by administering the measure to a sample of 20 children. As a result amendments were made to the measure used in the main validation study (Yusuf *et al.* 2006).

2.5.11 Qualitative research of the impact of oral health on children

A small number of studies have investigated the impact of oral health on children and young people. One study has explored the experiences of young people with Treacher Collins syndrome (Beaune *et al.* 2003) and several studies have explored children's general perceptions of oral health (Ostberg *et al.* 2002; Fitzgerald *et al.* 2004).

The impact of Treacher Collins syndrome on young people was investigated in Canada. Six participants aged from 12 to 18 years took part in semi-structured interviews at a hospital. Content analysis revealed three main themes of 'forming friendships and fitting in', 'handling staring and teasing' and 'balancing sameness and difference', with quotes used to illustrate these themes. While this study confirmed findings from other quantitative studies, several new findings emerged that can be generalised to the clinical management of these patients and interventions aimed at helping patients with cranio-facial differences to

improve their social interactions. The quality of the study was discussed in terms of its reliability, validity and reflexivity (Beaune *et al.* 2003).

Two studies have explored young people's experiences of oral health. One, conducted in Sweden, was stimulated by the need to improve the understanding of young people's perceptions of oral health after a large school survey. Seventeen participants aged 15 to 18 years had semi-structured interviews before no new data emerged. The interviews were conducted by a dentist, but not in a clinical setting. Data were analysed using constant comparative method. The two main themes were general and personal perceptions of oral health and the perceived influences on oral health. The impact of oral health was principally related to the affect of the appearance of teeth on social interactions. The authors discussed validity and reflected upon the influence on the data of a dentist conducting the interviews (Ostberg *et al.* 2002).

A second study involving young people was conducted to inform the re-design of dental services for children and young people in the South Island, New Zealand. This study used focus groups of two to 16 young people, held in a variety of settings by non-dentist researchers. Content analysis found that, while young people could recite the oral health messages they had been taught their own perceptions held oral health and dental services to be largely irrelevant to their everyday lives. The only exceptions given were the importance of having attractive teeth when socialising. The authors acknowledged that, while using a focus group enabled a wide range of perspectives to be accessed, the results could be affected by peer group pressure (Fitzgerald *et al.* 2004).

While qualitative methods involve greater participation of children in research by giving them a voice to explain their perspectives in their own words, the studies need to be designed to take account of factors such as power imbalance, language use, setting, analysis and ensuring quality of data. Some studies involved dentists acting as interviewers, interviews conducted in clinical settings, analysis without feedback from participants and failed to reflect on the influences of these factors on the quality of the data.

2.5.12 Conclusion

Changes to the position of children in society over the past 30 years have seen the emergence of a new approach to researching *with* children. Several important methodological recommendations have been made:

- Minimising the power imbalance that exists between children and adults
- Considering the language and comprehension of children participating in research
- Ensuring the settings for research are appropriate
- Using appropriate models of data analysis
- Assuring the quality of the data

Existing approaches to investigating children's experience of oral conditions, both quantitative and qualitative, do not appear to have heeded these recommendations and as a result may not reflect children's perspectives on the impact of DDE and dental conditions more generally. A more systematic approach to establish the degree to which research on DDE in children has involved them in research would be needed to confirm this hypothesis.

2.6 Rationale

Box 1. Outline of Chapter

- Background to developmental defects of enamel (DDE)
- Significance of DDE as a public health problem against four criteria:
 - Prevalence
 - Impact of the condition on the individual
 - Impact of the condition on society
 - Prevention and availability of effective treatments
- Approaches available to investigate the impact of dental conditions
- **Rationale**
- **Aims and objectives of the thesis**

This chapter has described the background and aetiology of DDE. It was difficult to come to any conclusions about whether DDE are a public health problem in the UK due to

several gaps in the evidence. One of the main deficiencies was the lack of information on the impact of DDE on individuals, despite repeated recommendations for research of this kind (Lalumandier and Rozier 1998; Milsom *et al.* 2000; Medical Research Council Working Group Report 2002; Sigurjons *et al.* 2004; Sujak *et al.* 2004). The chapter went on to describe how investigating the impact of dental conditions would necessitate a shift from the biomedical model of disease approach to research about DDE to embrace a biopsychosocial approach. In keeping with a biopsychosocial model of health the review has considered state of the art approaches to investigating impact, including complementing OHRQoL measures with qualitative methods. However, as much of the existing research has studied children and young people it was also necessary to review the current position of this group of the population in research. When the principles of research *with* children were discussed, in relation to the existing attempts to investigate children's perspectives on DDE and oral health generally, it appeared that many of the main factors were apparently neglected.

In summary, in order to ascertain whether DDE constitute a public health problem it is necessary to consider whether the condition impacts on the lives of affected individuals. The little research that has attempted to answer this question may have been restricted methodologically.

2.7 Aim and objectives

The aim of this research is to describe the impact of DDE on individual young people.

The objectives are to:

- 1) Describe the extent to which contemporary dental research on DDE has included the perspective of individual children and young people.
- 2) Describe the impact of DDE on the OHRQoL of young people.
- 3) Explore, in detail, the impact of DDE on young people.

To investigate the impact of DDE on young people, several important considerations need to be taken into account. First, the methods chosen should embrace contemporary views on involving children and young people with research, thereby taking a child-centred approach. Different methods of investigating impact are available that vary in the degree to which they capture the individual's own perspective, particularly when young people are

involved. Consequently, this thesis will adopt a multifaceted approach, investigating the impact of DDE on young people at a range of different levels. The thesis therefore includes a series of complementary studies on different aspects of the research question.

This traditional review has identified potential ethical and methodological shortcomings in the existing research in that it does not appear to be child-centered. The first research chapter of the thesis will therefore review systematically the contemporary literature to establish the degree to which it has included children's view of DDE. This study will be conducted to meet Objective One.

The second stage of the research involves drawing on existing biopsychosocial approaches through the use of a child-specific OHRQoL measure to provide an overall description of the impact of DDE and to evaluate the psychometric and child-centred properties of this measure for use in the UK. This study will fulfil Objective Two.

Finally, while quantitative research can measure the extent of the impact it is insufficient alone to capture the rich detail of young people's experiences of a condition and what it means to their everyday lives. The third stage will therefore involve an in-depth qualitative exploration of the perspective of young people in their own words. This study will satisfy Objective Three.

The next three chapters report these studies as discrete but related investigations. Chapter Three reports a systematic review of contemporary DDE and child dental literature.

2.8 Publications arising from the work in this chapter

Z. Marshman, P.G. Robinson. Child and Adolescent Oral Health-Related Quality of Life. *Seminars in Orthodontics* 2007; 13: 75-126

3. Chapter Three

The extent to which contemporary dental research on DDE has included the perspectives of children & young people

3.1 Introduction

Chapter Two involved a traditional narrative review of the literature about DDE. It appeared that there was a paucity of research about their impact on children and young people and a hypothesis that the existing DDE research fails to obtain their perspectives, more generally.

‘Children seen, but not heard’ is a saying that originated in early Victorian times. At that time, children did not have a childhood as it is known today, but were seen as ‘imperfect’ adults, wearing smaller versions of adult’s clothes and going to work rather than school (Heywood 2001). However, their position in society has changed. Children are no longer ‘seen, but not heard’ and there is growing interest in gaining greater understanding of children and young people’s experiences (Scott 2000). The evolution of child-centred research places increasing importance on obtaining children’s own perspectives, changing the position of children from an ‘object of concern’ to an ‘active participant’ (O’Kane 2000). As previously stated, active participation of children requires consideration of factors such as power imbalance, language use, setting, appropriate analysis and quality assurance.

Therefore, the aim of the study described in this chapter was to establish the extent to which contemporary dental research on DDE has included the perspective of individual children and young people. This study allowed the current literature, outlined narratively in Chapter Two, to be systemically and critically reviewed, with the focus on involvement of child participants. General child dental literature was also reviewed to allow comparison. Including the general child dental literature also ensured that reports that had used novel or unexpected terms for DDE would be more likely to be found.

3.1.1 Objectives of the study

- a) To develop a classification of how children were involved in research.
- b) To identify recently published DDE and general child dental literature.
- c) To quantify the identified literature within the categories from objective a).
- d) To compare DDE research with child dental research more generally.

3.2 Methods

Systematic reviews involve identifying, appraising and synthesising all relevant studies, in order to answer a research question, often about the effectiveness of an intervention. They can, however, provide an accurate picture of previous research in any particular field (Petticrew and Roberts 2006).

3.2.1 Objective a) Developing the classification

The categorisation framework was developed using framework analysis, which classifies qualitative data by organisation according to key themes and emerging categories (Ritchie and Spencer 1994). This matrix-based method, which has been widely used in applied policy research, allows data to be synthesised quickly when specific information is needed. It is more appropriate than other qualitative approaches for an analysis on a narrow topic.

A sample of child dental literature was chosen purposively to include a wide range of studies from both different specialties (including paediatric dentistry, orthodontics, oral pathology, oral surgery, dental public health) and from biological and psychosocial perspectives. The two themes of *on* children and *with* children were explored and characterised, based on an initial sample of 20 papers. Four main categories were identified and frameworks devised so the properties of these categories could be developed. A further fifteen papers were then examined to enable the properties to be fully described. Table 11 summarises the categories and arranges them in decreasing order of involvement of children.

Table 11. Categories of child dental literature developed from framework analysis

Category	Properties
1. <i>With</i> children: children seen as active participants	a) children involved in research process b) children's own accounts
2. <i>With</i> children: children seen as subjects	a) children completing measures designed by adults b) case report/series with child's input throughout case
3. Proxies for children used	a) parent/carer used appropriately as proxy b) clinician used appropriately as proxy
4. <i>On</i> children	Children seen as the objects of research

The first category of research, where children were active participants (i.e. being seen, listened to and heard), attached a priority to fully involving children. Within this category, two sub-categories were derived, based on the degree to which the children were involved. The first sub-category (category 1a) was research conducted with children as participants actively engaged throughout the research process (e.g. involving them in research design, in piloting, using participatory data collection techniques and getting their feedback on results during the analysis). The second sub-category (category 1b) comprised research where children participated by giving accounts of their experiences in their own words, such as through semi-structured or in-depth interviews or focus groups.

The second category, also classed as research *with* children, included studies where children were 'seen' and listened to as the subjects of research, although their own words were 'not heard'. This category was sub-divided into studies where children completed measures designed wholly by adults (category 2a) including self-complete questionnaires, structured interviews and visual analogue scales. The second sub-category included clinical case studies showing evidence of the child's views and feelings being taken into account throughout the reporting of the case, such as reporting the presenting complaint in

the child's own words and evidence of discussions with the child about the treatment (category 2b).

The third category included research that used either parents/carers (category 3a) or clinicians (category 3b) as appropriate proxies for children. It was felt appropriate to use proxies for young children (under six years) or those who lacked verbal articulacy, as research with the children themselves would not be a feasible option (Pantell and Lewis 1987).

The final category (category 4) included research where children were simply the objects to be studied i.e. children were seen, but not heard. Included within this category was research that saw children as:

- a set of teeth or a mouth to be treated
- a source of samples of plaque, saliva or hard/soft tissue
- an age group of patients to be managed
- patients with medical conditions
- a population group to be surveyed clinically
- patients on whom 'special investigations' were conducted
- recipients of an oral health promotion intervention

3.2.2 Objective b) Identify research papers

The search strategy was based on published studies using child-related keywords (child* or young person or young), DDE-related keywords (enamel and defects or enamel and opacities or fluorosis) and general dental-related keywords (caries or erosion and dent* or trauma and dent* or malocclusion or orofacial or oral or periodont* or orthodont*).

The search strategy was performed on the databases Medline (via Ovid) and Embase and limited to the English language. The resulting references were exported to an Endnote library (Thomson ResearchSoft 2005) and all duplicates removed. A list of dental journals was compiled (Appendix A) and articles from non-dental journals were excluded from the electronic library.

The time frame of 2000-2005 was chosen as the new social studies of children and childhood were consolidated in 1998 (James *et al.* 1998), with methods described for conducting such research published in 2000 (Christensen and James 2000).

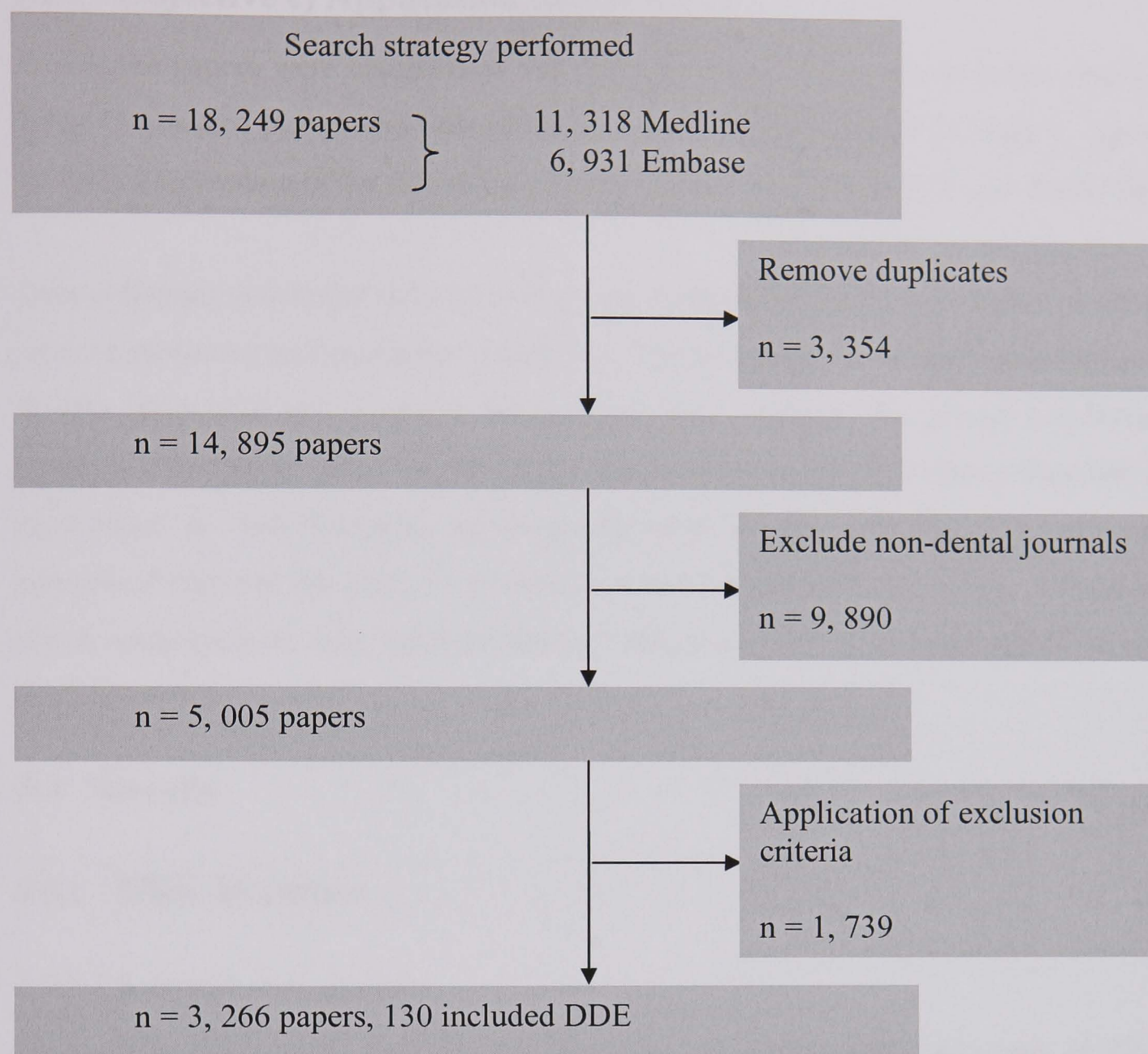
At the first pass through the electronic library of references the following exclusion criteria were applied:

- reports before 2000
- studies with participants over 16 years of age
- studies with no primary data
- articles reporting in vitro studies
- conference proceedings
- articles that did not have children and aspects of their oral health as their main topic.

A team of researchers from different disciplines (paediatric dentistry, sociology, health psychology, dental public health and orthodontics) was recruited to conduct the review. Two trained reviewers from the team independently applied the exclusion criteria based on the abstracts and where necessary the full length papers. Agreements between the reviewers about application of exclusion criteria occurred for 77% of the papers and disagreements were resolved through discussion.

The initial search resulted in 18,249 papers representing 14,895 individual papers after duplicates were removed. After excluding articles from non-dental journals the number of papers reduced to 5,005. Application of the exclusion criteria resulted in 3,266 papers, of which 752 were case reports/series. Of these 130 articles (4.0%) considered DDE (Figure 6).

Figure 6. Flow diagram for results of literature search



3.2.3 Objective c) Application of categories

Before the papers were categorised, the eight reviewers were trained in the classification using 15 selected papers and then calibrated on two further sets of 15 papers. Agreement on the categorisation of the individual papers ranged from 63% to 100% on the second set.

Data collection proceeded with two reviewers categorising each paper independently: four pairs of reviewers assessed approximately 817 papers per pair. Where a paper appeared to fit into more than one category, the category that presumed the greater involvement of children was chosen. The two reviewers then compared which category they had placed each paper in. Inter-examiner agreement between the two reviewers was assessed. The agreement between the pairs of reviewers was excellent (88% to 92%). Disagreements about categorisation were resolved through discussion and if necessary involvement of a third reviewer.

3.3 Results

3.3.1 DDE literature

3.3.1.1 Research with children

Of the 130 DDE papers that resulted from the literature search, only nine (6.9%) were categorised as research *with* children. These papers were all in category 2a (Table 12). No papers were found that involved children or young people as active participants (category 1).

Table 12. Frequency distribution of categories of papers

Category	Properties	No. of papers: DDE (%)	No. of papers: Overall (%)
1. <i>With</i> children – children seen as active participants	a) children involved in research process	0	2 (0.1)
	b) children’s own accounts	0	6 (0.2)
2. <i>With</i> children – children seen as subjects	a) children completing measures wholly designed by adults	9 (6.9)	220 (6.7)
	b) case report/series with child’s input throughout case	0	10 (0.3)
3. Proxies for children used	a) parent/carer used appropriately as proxy	0	173 (5.3)
	b) clinician used appropriately as proxy	0	12 (0.4)
4. On children	Children seen as the objects of the research	121 (93.1)	2843 (87.1)

The nine papers including DDE-related keywords in category 2a involved children and young people being asked questions or completing questionnaires. Although these studies involved children through the completion of these measures, all of the measures were designed by adults, most without input from children on the content, wording or format (Table 13). Four of these papers referred to children as ‘subjects’, the others used child-related terms, such as adolescents or children. Eight reported studies involving children and young people commenting on their own oral health, although none of these studies involved just children with DDE. One study referred to young people assessing images of another young person’s teeth that were digitally altered to appear fluorosed (Edwards *et al.* 2005).

Table 13. List of DDE papers of research *with* children

Authors	Age (yrs)	Country	Main constructs	Term used to refer to children	Method with children
(Milsom <i>et al.</i> 2000)	12	UK	Perceptions of appearance of own teeth	Subjects	2 questions asked by clinical examiners
(Astrom and Mashoto 2002)	12-20	Tanzania	Perceptions of own teeth & OHRQoL	Students	8 questions from OIDP asked by interviewers
(Wondwossen <i>et al.</i> 2003)	12-15	Ethiopia	Perceptions of appearance of own teeth	Adolescents	2 questions asked by interviewers
(Shulman <i>et al.</i> 2004)	6-15	USA	Perceptions of appearance of own teeth	Subjects	1 question asked by clinical examiners
(Sujak <i>et al.</i> 2004)	16	Malaysia	Perceptions & social impact of own teeth	Subjects	Self-completed questionnaire devised from the literature
(Martinez-Mier <i>et al.</i> 2004)	7-11	Mexico	Perceptions & impact on OHRQoL of own teeth	Children	Development of 8-item questionnaire then administration by interviewers
(Michel-Crosato <i>et al.</i> 2005)	6-15	Brazil	Impact on OHRQoL of own teeth	Children	8 questions from OIDP asked by interviewers
(Robinson <i>et al.</i> 2005)	12	Uganda	Impact on OHRQoL of own teeth	Children	CPQ self-completed with supervision
(Edwards <i>et al.</i> 2005)	14-15	UK	Acceptability of image of fluorosed teeth	Subjects	Web-based testing of lay adolescents

As described in Chapter Two, two papers used a modified version of OIDP developed for adults, with questions that may not be appropriate for younger people (Astrom and Mashoto 2002; Michel-Crosato *et al.* 2005). Neither study gave details of which of the aspects of daily performance fluorosis impacted upon. For example, in the Brazilian study, children as young as 6-years-old were asked questions from OIDP about: difficulty with oral hygiene, difficulty sleeping, difficulty smiling or laughing, difficulty maintaining emotional stability, difficulty studying and difficulty playing or having fun with people (Michel-Crosato *et al.* 2005). The exact wording of these questions was not described and, as previously mentioned, no details were given of the method of administration of this measure. In addition, no mention was made of any evaluation of this measure particularly in terms of acceptability and face and content validity for the children participating. Of the properties of OHRQoL described earlier, these three properties are the most pertinent for assessing the level of involvement of children and young people in the research.

Another paper reported a questionnaire study, designed to measure the perceptions and concerns of children with fluorosis in Mexico. This questionnaire was developed based on questions used in previous studies, rather than asking children what items were important to them. The final stage of the development of this measure involved piloting with children, but the comprehensiveness, relevance and clarity of the questions was assessed by a panel of dental experts, not by children (Martinez-Mier *et al.* 2004). Only one study used a measure specifically designed for use with children and young people. While the reliability and construct validity of this measure was assessed in the target population (12-year-old children in Uganda) neither the acceptability, face validity nor content validity were investigated (Robinson *et al.* 2005).

Five case reports were written about techniques for improving the appearance of the teeth of patients with DDE. Three of these reported the patient's presenting complaints, although none of them did so in the patient's own words (Quinonez *et al.* 2000; Wakefield and Woods 2002; Bussadori *et al.* 2004). Only one of them reported on patient satisfaction after treatment (Quinonez *et al.* 2000). No case reports involved the child throughout the reporting of the case, therefore they were included in category 4 rather than category 2b.

3.3.1.2 Use of proxies

No articles reported using clinicians or parents as proxies for children's views (category 3, Table 12). One study, about parental perceptions of fluorosis, involved a telephone interview with parents, about their satisfaction with the appearance of the teeth of their 8-year-old children (Sigurjons *et al.* 2004). This article was not included in category 3a, rather it was categorised as research *on* children, as the children were old enough to report their own satisfaction, without the need to use a proxy. However, the choice of parents, instead of children, may have been dictated by the use of the telephone as the method of approach, which may not have been felt to be appropriate for 8-year-old children.

3.3.1.3 Research on children

Finally, the vast majority (n = 119, 93%) of DDE papers were categorised as research *on* children (category 4, Table 12). Typically, the DDE literature viewed children as 'a population group to be surveyed clinically', often to investigate the aetiology or prevalence of the condition. Implicit in these papers was the idea of children as the objects of research, with no involvement of children or their parents to any extent.

3.3.2 General child dental literature

3.3.2.1 Research with children

Whilst no studies from the DDE literature involved children as active participants, eight studies (0.3%) from the general child dental literature had done so. The reports of two of these studies (0.1%) had evidence of children being included throughout the research process (category 1a). Both studies involved children and young people in the development and evaluation of child OHRQoL questionnaires (CPQ₁₁₋₁₄ and CHILD-OIDP) (Jokovic *et al.* 2002; Gherunpong *et al.* 2004).

A further six (0.2%) papers reported studies using qualitative methods (category 1b). These studies explored young people's perspectives in their own words on: oral health generally, dental services, dental health education, habits (drinking soft drinks) and compliance with orthodontic treatment. In terms of investigating the impact of conditions, one study investigated the impact of Treacher Collins Syndrome and two studies investigated the impact of oral health generally. Most of these studies involved young

people, although, in one study, children from eight years of age were interviewed (Table 14) (May and Waterhouse 2003). Only one used interviews, most were focus groups. A variety of methods of analyses were used, with only one study using an explicit theoretical framework to inform the study (Beaune *et al.* 2003).

Table 14. List of studies using qualitative methods

Authors	Age (yrs)	Method	Analysis	Focus of enquiry
(Bennett <i>et al.</i> 2001)	< 18	Focus groups	Content	Compliance with orthodontic treatment
(Ostberg <i>et al.</i> 2002)	15-18	Interviews	Constant Comparison	Impact of oral health
(May and Waterhouse 2003)	8-14	Focus groups	Framework	Drinking soft drinks
(Beaune <i>et al.</i> 2003)	12-18	Focus groups	Content	Impact of Treacher Collins syndrome
(Fitzgerald <i>et al.</i> 2004)	13-18	Focus groups	Content	Impact of oral health
(Ostberg 2005)	14-19	Focus groups	Constant Comparison	Views of oral health education

A total of 220 (6.7%) papers were in the second category, with children completing measures designed by adults (category 2a). Unlike papers in category one, these papers used measures developed without children's' input into the topics they felt were relevant or in the format or wording of the measures. Only 10 (0.3%) case reports/series had evidence of the child's involvement.

3.3.2.2 Use of proxies

A total of 185 (5.7%) papers used proxies to gain the child's perspective, of which 173 (5.3%) used parents/carers and 12 (0.4%) used clinicians. The children of interest in studies involving parents/carers (category 3a) were either less than six years of age or older children with communication difficulties. The papers reporting the use of clinicians as proxies (category 3b) investigated the impact of dental treatment or treatment services on young children or those unable to communicate themselves.

3.3.2.3 Research on children

Finally, the vast majority (n = 2,843, 87.1%) of papers were categorised as research *on* children. Within the child dental literature were extreme examples that referred to the children studied as ‘the material’.

3.3.3 Comparison of DDE and child dental literature

As shown in Table 12, similar proportions of DDE papers and papers from the general child dental literature involved research *with* children. The notable differences were the lack of case reports and use of proxies in the DDE.

3.4 Discussion

This systematic review of the extent to which the experiences of individual children and young people have been researched provides a critique of recent literature about DDE. The categories developed represented a hierarchy of involvement of children in research, ranging from full involvement in the research process, to no involvement. It found that although there have been some attempts to involve children in research about DDE the vast majority of research has been *on* them. This approach broadly reflects the dominant research approach in paediatric dentistry.

The findings confirm the suggestions made in Chapter Two. Most of the research on DDE has used methods that involve children, merely as objects of research, to investigate the aetiology or prevalence of the condition. While this information is important it satisfies only one criterion when deciding whether DDE are a public health problem. Specifically, research of this kind neglects the patient’s perspective and provides no information on the impact of conditions on those affected. We can conclude that the existing research does not adequately assess the impact of DDE on affected individuals and does not tell us whether they constitute a public health problem.

The results from the DDE literature were mirrored by the child dental literature. Only a small number of studies in this general literature had involved children as active participants or had been concerned with the impact of oral health. Involving children in this way has the potential to ensure that aspects pertinent to them are properly considered in dental research. This is important from both clinical and policy perspectives.

In paediatric dentistry it is the child who undergoes the treatment and who lives with the consequences. It is therefore important to consider their perspectives, desires, and expectations in decision-making about their care (Mouradian 1999). While communication with patients is stressed as a key feature of dentistry for children (Blinkhorn 2005), this emphasis does not appear to be present in research. Further research *with* children, particularly about the effectiveness of clinical interventions from their perspective could improve the quality of individual patient care.

In terms of policy implications, existing policies, such as the Children's NSF suggest professionals should listen to children, value their views and take these into account in decisions about their care and the planning, delivery and evaluation of services (Department of Health 2003). In Scotland, the action plan for improving dental services suggests services should be 'child-friendly' (Scottish Executive 2005). Further information from children could be used to advocate for resources to improve the child-friendliness of the way dental services are delivered. Equally, research *with* children to contribute to policies regarding DDE, such as guidelines on the use of various sources of fluoride, could result in policy changes that reflect children's perspectives more accurately. The increasing recognition of research *with* children (James *et al.* 1998; Barron 2000), rather than research defined by adult interests, biases and agendas (Balen *et al.* 2006) has already resulted in changes to social policy for children (Grover 2004).

The findings of this systematic review verify suggestions made in Chapter Two that existing research has methodological problems. No studies about DDE were categorised as actively involving children throughout the research process (Category 1) and the nine studies involving children completing measures designed by adults (Category 2a) often failed to evaluate the acceptability, face and content validity of the measures used. Without such evaluations, these studies may not include items relevant to children, may not be worded and formatted in a way appropriate and may miss aspects of their lives on which DDE impact. No studies investigated young peoples' perspectives or impact in their own words (Category 2b). Such an approach adds the advantage of giving children a voice to describe their own experiences, rather than relying on 'what adults think children think' (Alderson 1995). The subsequent studies described in this thesis will involve research *with*

children about the impact of DDE. The study described in Chapter Four will describe the impact of DDE on children and young people, using an OHRQoL measure designed for use with children. This study will include an evaluation of the properties of the measure pertinent to ensuring the research is child-centred. The study described in Chapter Five will use qualitative interviews to capture and explore, in detail the participants' perspectives.

Like all research, this systematic review has limitations. Firstly, for practical reasons, the search was restricted to electronic databases, the English language and dental journals. The adoption of this strategy meant that some relevant studies may have been omitted. However, as the general child dental literature was also included this ensured that, whatever terms were used for DDE in the reports, they were more likely to be found. Secondly, as child-centred research is at a relatively early stage in dentistry, such studies may not yet have reached the stage of publication. Inclusion of conference proceedings might have included some such studies, but would have resulted in an unmanageable number of articles. Thirdly, studies reporting research with children may have been published elsewhere and therefore may have been overlooked. Finally, the representation of the research provided in the published reports may not include comprehensive details of the actual study. Children may have been more fully involved in some studies without this being described in the text.

In summary, this systematic review found there to be few data available on the impact of DDE and children's experience of this condition is under-represented in clinical and public health policy. In most child dental research, including research about DDE, children are seen, but not listened to or heard. What research has been carried out has methodological limitations. Research on the impact of DDE on individual children and young people is required. Such research should embrace contemporary methodological principles.

3.5 Publications arising from the work in this chapter

i) Presentation:

Z. Marshman, B.J. Gibson, J. Owens, H.D. Rodd, H. Mazey, S.R. Baker, P. E. Benson, P. G. Robinson. A systematic review of child oral health research 2000–2005. International Association of Paediatric Dentistry Congress. International Journal of Paediatric Dentistry 2007; 17: s1, Abstract No. OS093.

ii) Publication:

Z. Marshman, B.J. Gibson, J. Owens, H.D. Rodd, H. Mazey, S.R. Baker, P.E. Benson, P.G. Robinson. 'Seen but not heard'. A systematic review of the place of the child in 21st century dental research. International Journal of Paediatric Dentistry 2007; 17: 320-327.

4. Chapter Four

The impact of DDE on the OHRQoL of young people

4.1 Introduction

The aim of the body of research in this thesis is to describe the impact of DDE on individual young people in order to contribute to discussions about whether DDE are a public health problem. To date, little research has been carried out to investigate the impact of DDE, particularly in the UK, despite repeated recommendations for research in this area (Lalumandier and Rozier 1998; Milsom *et al.* 2000; Medical Research Council Working Group Report 2002; Sigurjons *et al.* 2004; Sujak *et al.* 2004). The paucity of research was confirmed by the findings of the study described in Chapter Three. Investigating the impact on individuals is an important criterion for determining the public health significance of a condition. Previous research has mainly concentrated on the prevalence and determinants of DDE based on normative assessments, rather than considering young people's own perspectives.

Chapter Two reviewed the various approaches to investigating the impact of dental conditions and the important considerations when the population group of interest are children and young people. One such approach was the use of a child-specific OHRQoL measure. However, before a measure can be used, properties such as reliability and validity should be evaluated in the country, setting and age-group under question, including an assessment of how well the measure addresses aspects important to those completing it (Guyatt *et al.* 1993; Gill and Feinstein 1994).

Two measures of OHRQoL have been developed for children and young people (COHRQoL and CHILD-OIDP). Both measures are based on a conceptual framework of oral health, but neither measure has been used with children and young people with DDE in the UK. CPQ (one of the COHRQoL measures) is more suitable to assess the impact of DDE, as CHILD-OIDP only detects the ultimate impacts of dental conditions and, so, may be relatively insensitive to the whole range of impacts that result from enamel defects.

CPQ₁₁₋₁₄ has previously been used in Ugandan young people including those with fluorosis (Robinson *et al.* 2005). The properties of CPQ₁₁₋₁₄ have been evaluated for use in Canada, New Zealand, Uganda and Saudi Arabia (Jokovic *et al.* 2002; Foster Page *et al.* 2005; Robinson *et al.* 2005; Brown and Al-Khayal 2006), but neither CPQ₁₁₋₁₄, nor its analogous parental questionnaire P-CPQ have been evaluated for use in the UK.

The aim of this study was to describe the impacts of DDE on the OHRQoL of young people, using a measure that was rigorously tested.

4.1.1 Objectives of the study

- a) To assess the reliability and validity of CPQ₁₁₋₁₄ and P-CPQ for use in a clinic-based sample in the UK
- b) To detail the impact of DDE on the OHRQoL of young people attending for treatment of the condition
- c) To compare the OHRQoL of young people with DDE to that of orally healthy young people

The objectives of this study included evaluation of both CPQ₁₁₋₁₄ and P-CPQ. It was not the intention to use data from P-CPQ as a proxy for CPQ₁₁₋₁₄. Rather, its use provides additional information on what parents/carers know about the impact of their child's oral condition on their lives.

4.1.2 CPQ₁₁₋₁₄

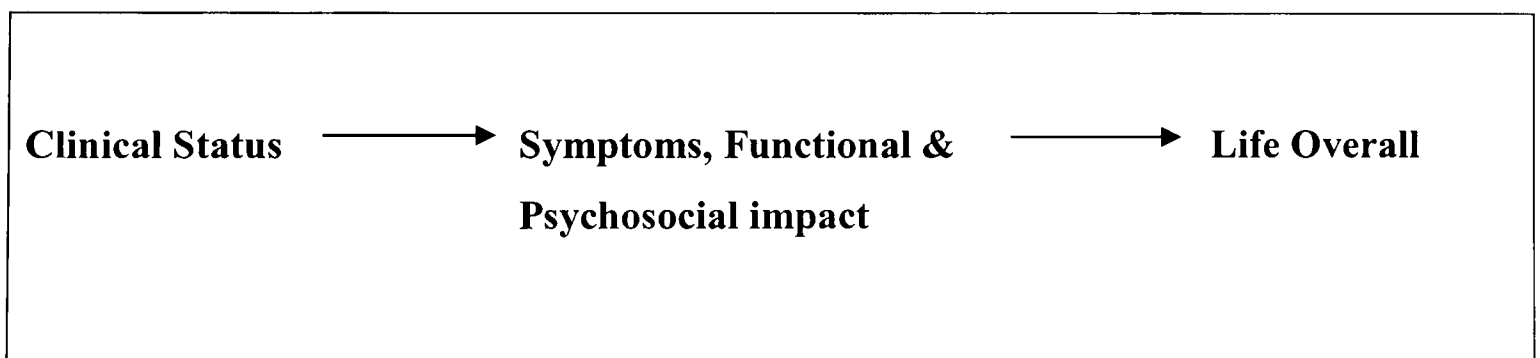
CPQ₁₁₋₁₄ was developed by a process recommended to ensure it contained items of the most relevance to young people (Jokovic *et al.* 2002) (see section 2.5.10.1). The measure contains 37 items, asking about the frequency of events in the previous three months. The measure encompasses four health domains: oral symptoms, functional limitations, emotional well-being and social well-being. Response options are: never = 0; once or twice = 1; sometimes = 2; often = 3; everyday or almost everyday = 4 (Appendix B). Summing the response codes for all items generates an overall CPQ₁₁₋₁₄ score. Scores for each domain can also be computed.

Reliability and validity of CPQ₁₁₋₁₄ were initially assessed in a sample of 123 Canadian young people attending oro-facial, paediatric dentistry and orthodontic clinics in a hospital. Global indicators of oral health and the extent to which the oral and oro-facial condition affects life overall were also obtained. These items were worded as follows: “Would you say that the health of your teeth, lips, jaws and mouth is ...?” (global oral health rating) with a 5-point response format ranging from ‘Excellent’ to ‘Poor’ and “How much does the condition of your teeth, lips, jaws or mouth affect your life overall?” (life overall rating) with a response range from ‘Not at all’ to ‘Very much’.

The internal consistency reliability of the scale was good (Cronbach’s alpha = 0.91) and for the subscales was acceptable to good. The test-retest reliability found good agreement (ICC = 0.90).

Discriminant validity was confirmed by significant differences in scale scores among the three clinical groups (oro-facial patients had the highest scores; paediatric dentistry patients had the lowest). Construct validity was confirmed by significant correlations between scale scores and the two global indicators (Jokovic *et al.* 2002). This method of assessment of validity was based on a simplified form of Wilson and Cleary’s (1995) model of disease and its consequences (see section 2.5.2.1) (Figure 7).

Figure 7. Simplified Wilson and Cleary model applied to oral diseases and disorders (Locker *et al.* 2001)



During the derivation of the measure in Canada, neither the face, nor content validity of the measure were reported. Other studies have assessed these properties of OHRQoL measures qualitatively, by administering the measure to a small sample of children (Yusuf *et al.* 2006).

Clinical data were extracted from patient's notes, but were not available for all participants. Caries data were available for the paediatric group and showed a significant correlation between total CPQ₁₁₋₁₄ score and the number of decayed tooth surfaces ($r=0.64$). No other relationships between clinical data and scale scores were reported. Data on DDE were not included.

Other studies using CPQ₁₁₋₁₄ have used clinical examinations, rather than extraction of clinical data from patient records (Foster Page *et al.* 2005; Robinson *et al.* 2005). In New Zealand, clinical data on caries (number of decayed, missing and filled surfaces (DMFS) and malocclusion (Dental Aesthetic Index) (Cons *et al.* 1986) were collected. There was a significant difference between CPQ₁₁₋₁₄ scores for young people with DMFS greater than four compared with those of DMFS=0. CPQ₁₁₋₁₄ scores increased with increasing severity of malocclusion (Foster Page *et al.* 2005). In Uganda, data on caries (DMFT) and fluorosis (TFI) were reported. Two summary measures of CPQ₁₁₋₁₄ were computed: total score and the mean number of impacts 'often' or 'everyday'. The total score was associated with the presence of caries experience (DMFT>0), but not with fluorosis. The number of impacts 'often' or 'everyday' correlated with caries experience and marked fluorosis (TFI > 2) (Robinson *et al.* 2005).

4.1.3 P-CPQ

P-CPQ is a measure of parental-carer perceptions of the OHRQoL of children. P-CPQ was designed to supplement the information obtained by CPQ from children. It includes 31 questions covering the same four domains, with 14 additional questions on the impact on the family, the Family Impact Scale (FIS) (Jokovic *et al.* 2003). The participants are asked to indicate, using a six-point Likert scale ('never' = 0, 'once or twice' = 1, 'sometimes' = 2, 'often' = 3, 'everyday or almost everyday' = 4, and 'don't know'), the frequency at which the events have affected their child in the past three months. The P-CPQ contains a 'don't know' response because the authors were aware of the limited knowledge a parent may have of their child's activities and feelings. 'Don't know' response categories have been used in other questionnaires to reassure respondents that it is acceptable not to know the answer, as well as to minimise guessing (Bowling 1997). P-CPQ also includes the global

oral health and life overall ratings, although these ratings do not include a ‘don’t know’ response.

P-CPQ was evaluated in Canada in a similar way to CPQ₁₁₋₁₄ (Jokovic *et al.* 2003). It had good internal consistency for the total score (Cronbach’s alpha = 0.94) and for the subscales. The test-retest reliability indicated excellent agreement (ICC = 0.85). P-CPQ showed good construct validity, with significant associations between the total scores and global ratings of oral health and life overall. Only questionnaires with zero ‘don’t know’ responses were included in the analyses. However, when different methods for handling ‘don’t knows’ were investigated, the properties were not affected (Jokovic *et al.* 2004).

4.2 Method

The aim of this study was to describe the impacts of DDE on the OHRQoL of young people, using a measure that was rigorously tested. To meet the three objectives of this study, different samples of young people were recruited:

- A general clinic-based sample, which included young people with a range of oral conditions including caries, malocclusion, gingivitis and DDE.
 - From this general sample, a sub-sample of young people with ‘relative oral health’ was composed. The relative oral health sub-sample was defined as no unrestored decay, an IOTN of three or less and no gingivitis or DDE present.
- A group of young people with DDE attending for treatment of the condition.

In each case, individual young people were invited to participate by the clinicians; assent was gained from the young people and consent from their parents/carers.

4.2.1 General clinic-based sample

To evaluate the measure, a consecutive sample of young people between 11 and 14 years of age and their parents/carers were invited to take part. These young people were attending for an examination at the orthodontic or paediatric dentistry clinics of Charles Clifford Dental Hospital, Sheffield or a Sheffield General Dental Practice.

Data were collected by asking young people and parents/carers to complete, at the time of their visits to the clinics, the CPQ₁₁₋₁₄ and P-CPQ respectively. The initial questionnaires also invited participants to complete a follow-up questionnaire, posted two weeks later, to assess test-retest reliability. The second questionnaire asked if either the oral/oro-facial condition or its impact on the young person's well-being, had changed since recruitment. Data on ethnicity, age, gender and socio-economic status were also obtained.

The sample size calculation was based on caries data from the Canadian study as there were no data on either DDE or caries from the UK. From the original Canadian study a Spearman's rank correlation coefficient of 0.64 was found between the number of decayed teeth and CPQ₁₁₋₁₄ scores (Jokovic *et al.* 2002), requiring a total sample of 22 to be significant at an alpha of 0.01. A total sample of 90 young people was chosen to allow for possible cultural differences between the UK and Canada.

Clinical data were collected by clinicians who had been calibrated in the use of the clinical variables (Appendix C). The existence of DDE on anterior teeth was recorded as present or absent. Caries status was assessed, based on the British Association for the Study of Community Dentistry criteria, by enumerating the number of decayed, missing or filled teeth (DMFT) due to caries (Pine *et al.* 1997) (see Appendix C). Malocclusion was categorised using the dental health component of the Index of Orthodontic Treatment Need (IOTN) (Brook and Shaw 1989). Gingivitis was recorded as present or absent. The clinical data were used to identify a sub-sample of young people with relative oral health, defined as no unrestored decay, an IOTN of three or less and no gingivitis or defects present. The results for this 'relative oral health sub-sample' were compared to the DDE group.

4.2.1.1 Data analysis

Where participants failed to indicate a score for an item, missing values were dealt with in two ways: those participants who failed to complete more than one-seventh of the questions were excluded from the analysis. A similar threshold for excluding missing values has been adopted in other OHRQoL research (Slade 1997a). Missing values from the remaining participants were replaced with the sample mean score for that item. No analytical strategy to deal with missing values was described in the Canadian study.

For P-CPQ, the different approaches to the analysis of ‘don’t know’ responses used in the Canadian study were employed (Jokovic *et al.* 2004) to assess whether the properties of the measure were affected (Appendix D). Of the four methods, one had markedly inferior properties. Of the other three, the approach selected was replacing the ‘don’t knows’ with zero, as the reliability was slightly superior to the other methods.

After taking account of missing or ‘don’t know’ responses, the total score for each participant was calculated by summing the item codes. A second summary measure for each participant, recorded the number of impacts reported ‘often’ or ‘everyday or almost everyday’. Subscale scores were calculated by summing the codes for questions within the four health domains. For P-CPQ, the FIS scores were calculated by summing the responses to these 14 questions.

Internal consistency was assessed by means of Cronbach’s alpha; test-retest reliability was assessed by ICC. The latter was based on data from those who participated in the follow-up study, and who did not report that their oral health and/or its impact had changed between the two administrations of the questionnaire.

Construct validity was assessed by testing associations of the scale and the subscale scores with the life overall scores and the clinical data. Criterion validity was examined by comparing the global rating of oral health to scale scores. Face validity was assessed for CPQ₁₁₋₁₄ only, by discussions with five participants, about whether the questionnaire made sense to them, and by examining the number of missing responses to items. The content validity of CPQ₁₁₋₁₄ was assessed for the DDE group only, based on the extent to which participants felt the measure captured the range of impacts of DDE. This was assessed by discussions with five participants, after they had completed the questionnaire.

4.2.2 DDE Sample

To obtain the sample of young people with DDE, young people between 11 and 14 years of age awaiting treatment of the condition at the paediatric dentistry clinics of Charles Clifford Dental Hospital, Sheffield and their parents were approached.

Clinical data were collected by calibrated clinicians, using the survey version of the modified DDEI to score the upper incisors. This simple index was used to minimise disruption on the clinics.

4.2.2.1 Analysis of DDE data

After taking account of missing responses (and ‘don’t know’ responses for P-CPQ), the two summary measures (total score and the number of impacts reported ‘often’ or ‘everyday or almost everyday’), mean subscale scores and the percentage of young people experiencing one or more impacts in each subscale were calculated. An extreme group approach was adopted in which OHRQoL data from the DDE group were compared to data from the relative oral health sub-sample. The difference between the groups was assessed using the effect size (mean difference in scores between the groups divided by the pooled SD of the scores)(Locker *et al.* 2005). Cohen described an effect size of 0.2 as small, 0.5 to be moderate and 0.8 to be large (Cohen 1988). For content validity, all comments about the extent to which the measure captured the impact of DDE on young people, were recorded.

An item impact assessment was carried out to give an indication of which aspects of young people’s lives DDE impacted upon. Items were assessed on the basis of their frequency and importance to the young people. The impact score for each question was the product of the prevalence (percentage of children giving a positive response) and the mean score for each item. Item impact scores were calculated for the DDE sample and compared to the item impact scores for the relative oral health sub-sample.

This study was approved by the South Sheffield Research Ethics Committee (see Appendix E for information sheets and consent forms).

4.3 Results

The results of the evaluation of the measures (from the general clinic-based sample), the comparison of impact between the DDE sample and the relative oral health sub-sample and the detailed investigation of the impact of DDE on OHRQoL will be described separately.

4.3.1 Results of the evaluation of CPQ₁₁₋₁₄ and P-CPQ from the general clinic-based sample

Ninety-one young people, with their parents/carers, were invited to participate in the study. These participants will be described as the general clinic-based sample. No young people refused or were unable to complete the questionnaire due to literacy problems. Data from two of the young people and four parents/carers were excluded due to missing data. Of the parents/carers, mothers completed 61 (71.3%) questionnaires, fathers completed 25 and one was completed by a carer.

The resultant general clinic-based sample of 89 comprised 29 young people attending a paediatric dentistry clinic, 30 attending an orthodontic clinic and 30 attending a general dental practice. The mean age of participants was 12.4 years; there were 47 females; 83 children were White British. Postcodes of the participants were used to gain an Index of Multiple Deprivation 2000 score and rank, as a measure of socio-economic status; postcode data were available for 74 participants. The sample was made up of young people from areas with varying levels of deprivation.

4.3.1.1 Clinical data of the general clinic-based sample

Twenty-percent of the young people in the general clinic-based sample had DDE. The DMFT was 1.25 (SD 2.75) with a mean total number of missing teeth, for any reason, of 0.41 (SD 0.97) (Table 15). Seventy-nine percent had no unrestored carious teeth.

Table 15. Caries experience of the general clinic-based sample (n = 89)

	Mean	SD
Missing teeth due to caries	0.08	0.41
Filled teeth	0.78	2.12
Decayed teeth	0.39	1.20
Decayed, missing and filled teeth	1.25	2.75

Sixty-nine percent of young people had an IOTN score of less than four and 64% had good gingival health.

Thirty-eight young people had no unrestored caries, an IOTN of less than four and no defects or gingivitis. This group was used as the relative oral health sub-sample.

4.3.1.2 CPQ₁₁₋₁₄ scores of the general clinic-based sample

The mean total CPQ₁₁₋₁₄ score was 18.07 (SD 11.59). Of the full range of CPQ₁₁₋₁₄ scores from 0 to 148, scores ranged from 3 to 53 therefore no floor or ceiling effects were seen. The mean number of impacts experienced ‘often’ or ‘everyday or almost everyday’ was 1.56 (SD 1.92). Table 16 shows the mean score for the individual subscales.

Table 16. Mean scores for individual subscales of CPQ₁₁₋₁₄ of the general clinic-based sample (n = 89)

Subscale scores	Mean	SD
Symptoms	5.35	3.16
Function	5.42	3.75
Emotion	4.22	4.45
Social	3.08	3.09

One-fifth of the young people said the health of their teeth, lips and mouth (global oral health rating) was ‘fair’ or ‘poor’ in the prior three months. The condition of their teeth, lips, jaws or mouth affected 11% of participants lives ‘a lot’ or ‘very much’ (life overall rating).

4.3.1.3 Reliability of CPQ₁₁₋₁₄ from the general clinic-based sample

The Cronbach’s alpha for the total CPQ₁₁₋₁₄ scale was 0.87 and for the subscales ranged from 0.59 to 0.83, indicating substantial to excellent internal consistency (Table 17).

Table 17. Reliability of CPQ₁₁₋₁₄ and subscales of the general clinic-based sample (n = 89)

	No. of items	Cronbach's alpha
CPQ ₁₁₋₁₄	37	0.87
Symptoms	6	0.63
Function	9	0.59
Emotion	9	0.83
Social	13	0.65

Seventy-nine percent of participants (n=70) indicated a willingness to complete a second questionnaire, two weeks later, 84% of whom did so (n=59). From the second questionnaires, 73% (n=43) reported that the condition of their mouth was unchanged and thus were analysed for test-retest reliability. The ICC, on repeated application of the measure, was 0.83 (95% CI= 0.76-0.90), suggesting almost perfect agreement (Landis and Koch 1977).

4.3.1.4 Validity of CPQ₁₁₋₁₄ from the general clinic-based sample

4.3.1.4.1 Face validity

Discussions with participants revealed several important points about the questionnaire. Participants found it difficult to remember, despite regular reminders, that their responses should only concern aspects related to their teeth, lips, jaws or mouth and that this caveat applied to each item. A specific cause of confusion arose with the item 'because of your teeth, lips, jaws or mouth, in the past 3 months how often have you: breathed through your mouth? Participants were unsure how to answer this question, as they did not know whether their mouth breathing was related to their teeth, lips, jaws or mouth or some other cause.

Participants were also unsure how to respond to items that included several activities, for example 'taking part in activities like sports, clubs, drama, music, school trips'. The participants' oral condition may have limited their participation in some of these activities, but not others. Neither did participants know how to answer questions that included

activities of which they had no experience such as ‘chewing corn-on-the-cob or steak’ or ‘playing musical instruments’.

Despite these comments, the number of missing values was low, with a mean of 0.21 per young person. The final item regarding the frequency with which participants had been asked questions about their teeth, lips, jaws or mouth by other children, met with a considerable number of missing responses (n=9). None of the participants were able to offer an explanation, other than its position at the bottom of the page, just before the instruction ‘nearly finished’.

4.3.1.4.2 Construct validity

To assess construct validity, appropriate bivariate analyses were used to test relationships between summary measures of CPQ₁₁₋₁₄ and clinical data and ratings of life overall (Table 18). Summary and subscale scores of CPQ₁₁₋₁₄ were consistently associated with life overall, but only occasionally so with clinical data. For example, only the functional subscale scores of CPQ₁₁₋₁₄ were related to the presence of DDE. The number of children with impacts ‘often’ or ‘everyday’ correlated with the total number of missing teeth and missing teeth due to caries. No relationships were apparent between IOTN scores or the presence of gingivitis and CPQ₁₁₋₁₄.

Table 18. Relationship between life overall ratings, clinical data and CPQ₁₁₋₁₄ scores of the general clinic-based sample (n = 89)

	Total CPQ₁₁₋₁₄	No. often or everyday	Symptom	Function	Emotion	Social
Spearman's rank correlation coefficient						
Life overall	0.40*	0.29*	0.28*	0.28*	0.29*	0.35*
No. missing teeth	0.07	0.23*	-0.03	0.12	0.13	0.10
Missing due to caries	0.05	0.21*	0.08	0.11	0.11	0.01
Filled	0.09	0.10	-0.01	0.16	0.11	0.06
Decayed	0.10	0.17	0.09	0.11	0.08	0.11
IOTN	0.14	0.12	0.13	0.13	0.11	0.11
p-values (Mann Whitney U test)						
DDE present	0.11	0.05	0.19	0.02*	0.53	0.14
Gingivitis	0.54	0.47	0.72	0.31	0.82	0.99

*=statistically significant, $p < 0.05$

4.3.1.4.3 Criterion validity

Summary measures of CPQ₁₁₋₁₄ correlated with the global oral health rating, indicating acceptable criterion validity (Table 19).

Table 19. Rank correlations between CPQ₁₁₋₁₄ scores and global measure of oral health of the general clinic-based sample (n = 89)

	Spearman's rank correlation coefficient
CPQ ₁₁₋₁₄	0.28*
No. often or everyday	0.34*
Symptoms	0.37*
Function	0.22*
Emotion	0.18
Social	0.19

*=statistically significant, $p < 0.05$

4.3.1.4.4 P-CPQ scores

The total score and subscales of P-CPQ are described in Table 20. Of the subscales, the highest mean score was in the symptoms domain.

Table 20. Mean scores for the subscales of P-CPQ and FIS of the general clinic-based sample (n = 87)

	Mean	SD
P-CPQ	14.2	14.5
Symptoms	4.1	3.3
Function	2.8	3.4
Emotion	2.9	4.1
Social	1.7	3.0
FIS	2.7	4.1

The distribution of the parents/carers global ratings was similar to the young people's in that 20% of parents rated their child's oral health as 'fair' or 'poor' and 11% reported levels of impact on life overall of 'a lot' or 'very much'.

4.3.1.4.5 'Don't know' responses of parents

The distribution of ‘don’t know’ responses was evaluated as an indicator of what parents/carers know about the impact of their child’s oral health. Twenty-seven parents/carers used ‘don’t know’ responses: 20 were mothers, six were fathers and one was a carer. In total, 49 ‘don’t know’ responses were used. Distribution of these responses was not even, with most occurring in the oral symptoms and social well-being subscales (Table 21). Fifteen parents/carers gave ‘don’t know’ responses to questions about the frequency their child got food stuck in their teeth or the roof of their mouth, ten didn’t know the frequency their child breathed through their mouth and seven parents/carers didn’t know the frequency their child was asked questions by other children.

Table 21. Distribution of ‘don’t know’ responses of the general clinic-based sample

	No. of ‘don’t knows’
Oral Symptoms:	
Bad breath	1
Food stuck in or between teeth	9
Food stuck in roof of mouth	6
Functional Limitations:	
Breathing through mouth	10
Difficulty with hard foods	1
Emotional well-being:	
Anxious or fearful	1
Shy/embarrassed	1
Concerned what others think	2
Upset	1
Social well-being:	
Hard time playing attention	2
Not wanting to speak in class	2
Not wanting to talk to other children	1
Arguing with other children/family	1
Being teased/called names	3
Worried fewer friends	1
Asked questions by other children	7

Table 22. Reliability of P-CPQ, subscales and FIS of the general clinic-based sample (n = 87)

	No. of items	Cronbach's alphas
P-CPQ	45	0.93
Symptoms	6	0.69
Function	8	0.68
Emotion	7	0.85
Social	10	0.79
FIS	14	0.82

The internal consistency (Table 22) and test-retest reliability of P-CPQ were acceptable (the test-retest analysis included 52% of parents, with an ICC of 0.95).

4.3.1.5 P-CPQ validity from the general clinic-based sample

4.3.1.5.1 Construct validity

Ratings of life overall were related to all P-CPQ measures. No relationship was apparent between P-CPQ and the presence of DDE. Significant correlations were found between the number of children with impacts 'often' or 'everyday' and the DMFT.

Table 23. Relationship between life overall ratings, clinical data and P-CPQ scores of the general clinic-based sample (n = 87)

	Total P-CPQ	Often or everyday	Symptom	Function	Emotion	Social	FIS
Spearman's rank correlation coefficient							
Life overall	0.40**	0.27*	0.34**	0.22*	0.41**	0.35**	0.41**
IOTN	0.11	0.01	0.09	0.02	0.15	0.07	0.06
Decayed	0.09	0.17	0.01	0.13	0.18	0.11	0.15
DMFT	0.15	0.25*	0.08	0.01	0.20	0.14	0.19
p-values (Mann Whitney U test)							
DDE present	0.25	0.11	0.22	0.31	0.86	0.51	0.40
Gingivitis present	0.45	0.74	0.49	0.37	0.51	0.85	0.55

*=statistically significant, $p < 0.05$

**statistically significant, $p < 0.01$

4.3.1.5.2 Criterion validity

Criterion validity was examined by comparing the P-CPQ scores and the global oral health rating (Table 24). Correlations between both summary measures and two of the subscales and the global oral health rating were found.

Table 24. Rank correlations between P-CPQ scores and global measure of oral health of the general clinic-based sample (n = 87)

	Spearman's rank correlation coefficient
P-CPQ	0.23*
No. often or everyday	0.25*
Symptoms	0.30**
Function	0.18
Emotion	0.09
Social	0.10
FIS	0.21*

*=statistically significant, $p < 0.05$

**statistically significant, $p < 0.01$

4.3.2 Results of the impact of DDE on young people

A consecutive sample of thirty young people with DDE attending paediatric dentistry clinics for treatment of this condition were invited to participate, with data from 29 young people analysed (one participant was excluded due to excessive missing data). This group of young people represented the DDE sample. The mean age of participants was 12.4 years. There were 16 females.

4.3.2.1 Clinical data of the DDE sample

The majority of the DDE were categorised as diffuse, with 10.3% scored as 'other defects' (a combination of types of opacities) (Table 25).

Table 25. Distribution of modified DDE scores of the DDE sample (n = 29)

	Number	Percentage
Demarcated opacities	7	24.1
Diffuse opacities	14	48.3
Hypoplasia	5	17.2
Other defects	3	10.3

The mean DMFT of the DDE sample was 1.60 (SD 2.06) (D = 0.28, M = 0.6, F = 0.72).

4.3.2.2 CPQ₁₁₋₁₄ scores of the DDE sample

After excluding one participant who missed one page of 6 questions, there were only two missing values.

The mean CPQ₁₁₋₁₄ score was 22.55 (SD 13.92) and ranged from 1 to 69 (of the full range of 0-148). Mean CPQ₁₁₋₁₄ scores, by type of opacities, ranged from 19.51 (SD 17.5) for participants with diffuse opacities, to 36.72 (SD 6.03) for participants with a combination of defects (Table 26).

Table 26. Mean CPQ₁₁₋₁₄ scores for different types of opacities of the DDE sample (n = 29)

	Mean	SD
Demarcated opacities	22.00	6.98
Diffuse opacities	19.51	17.50
Hypoplasia	23.40	11.59
Other defects	36.72	6.03

There was no significant difference between the mean CPQ₁₁₋₁₄ score for DDE and for the oral health sub-sample (mean CPQ₁₁₋₁₄ score = 19.78) ($p = 0.41$, t-test), and the effect size of 0.20 was 'small'. The mean number of impacts experienced 'often' or 'everyday or almost everyday' was 1.93 (SD 2.96), compared to 1.97 for the oral health sub-sample ($p = 0.95$, t-test). Therefore, neither the total CPQ₁₁₋₁₄ score nor the number of impacts 'often' or 'everyday or almost everyday' were associated with DDE in this group seeking treatment for the condition.

Table 27 shows the mean score and percentage with one or more impacts for the individual subscales. The highest mean subscale score was for the functional limitation subscale, although this had the lowest percentage of participants reporting one or more impacts. No significant differences were found between the mean subscale scores for the DDE group, compared to the oral health sub-sample.

Table 27. Mean scores and percentage impact for individual subscales of CPQ₁₁₋₁₄ of the DDE sample (n = 29)

	Mean	SD	% with ≥ 1 impact
Symptoms	5.41	2.85	96.5
Function	6.89	4.49	79.3
Emotion	5.55	5.88	96.5
Social	4.52	3.62	93.1

The percentage of young people who said the health of their teeth, lips and mouth was ‘fair’ or ‘poor’ in the prior three months was 59% compared to 18% in the oral health sub-sample ($p = 0.009$, t-test). The condition of their teeth, lips, jaws or mouth affected 14% of participants lives ‘a lot’ or ‘very much’; for the oral health sub-sample it was 13% ($p = 0.54$, t-test).

4.3.2.3 Content validity of CPQ₁₁₋₁₄ in relation to DDE

After completing the questionnaire, five young people were asked about the appropriateness of the content of CPQ₁₁₋₁₄ to the impact of their DDE. Most of their comments related to the relevance of the items or the wording of the questions.

The relevance of several items was raised, particularly ‘difficulty saying words’ and ‘drinking through a straw’, (both from the functional limitation subscale). Again, comments were made about the item ‘breathing through the mouth’; several participants were aware they did this frequently but despite thinking it was not relevant to the effects of their oral condition, answered ‘everyday or almost everyday’. One relevant topic noted as absent, was whether other people had said they had not brushed their teeth properly.

The wording of several questions resulted in surprising responses from some participants. For example, two young people who were repeatedly called ‘yellow tooth’ and ‘black tooth’ by others at school indicated they had never been ‘teased or called names’. On questioning, they did not perceive these comments as either teasing or name-calling, but just something that happened between class mates. Another young person said she had

responded to the question about ‘avoiding smiling or laughing around other children’ with ‘never’ because, rather than avoiding smiling, she always looked down, to hide her teeth.

4.3.2.4 Item impact assessment

An item impact assessment was used in the development of CPQ₁₁₋₁₄ to identify aspects important to young people (Jokovic *et al.* 2002). In this study it was used among children with DDE and the healthy subsample. The impact score for each question was calculated to assess which items were the most relevant to DDE and compared with scores of the oral health sub-sample (Table 28). The mean impact score for the DDE sample was 32.5 compared to 29.3 for the oral health sub-sample ($p=0.74$). The highest impact score, for both groups, was ‘breathing through the mouth’. The items where the DDE group scored higher were ‘difficult to eat or drink hot or cold foods’, ‘concerned what other people think of your teeth, lips, mouth or jaws’, ‘missed school because of pain, appointments and surgery’ and ‘arguing with other children/family’.

Table 28. Item impact scores among children with DDE and relative oral health sub-sample

	Relative oral health sub-sample (n = 38)	DDE group (n = 29)
Oral Symptoms:		
Pain in teeth/mouth	48.1	69.0
Bleeding gums	46.8	33.1
Sores in mouth	36.8	38.6
Bad breath	78.1	52.7
Food stuck between teeth	126.3	143.5
Food stuck in roof mouth	33.9	20.1
Functional Limitations:		
Breathing through mouth	251.7	126.0
Taking longer to eat	90.8	70.3
Trouble sleeping	29.4	28.9
Difficulty with hard foods	52.1	26.9
Difficulty opening wide	2.5	12.4
Difficulty saying words	3.0	3.6
Difficulty with foods you like	4.0	17.3
Difficulty using a straw	0.2	0.69
Difficulty to drink/eat hot/cold foods	18.2	91.0
Difficulty playing an instrument	0.2	1.03
Other children asked questions	21.2	41.0
Emotional well-being:		
Irritable/frustrated	18.7	17.3
Unsure of yourself	12.4	22.7
Shy/embarrassed	44.1	44.2
Concerned what others think	31.3	95.16
Worried less attractive	29.3	22.7
Upset	7.9	19.0
Nervous/afraid	12.9	24.8
Worried that are less healthy	7.1	7.2
Worried different from others	5.7	6.2
Social well-being:		
Missed school	27.0	87.2
Hard time playing attention	12.9	1.03
Difficulty with homework	4.0	3.6
Not wanting to speak in class	10.8	18.6
Not taking part in sports/clubs	0.6	1.03
Not wanting to talk to other children	0.8	1.03
Avoiding smiling around other children	6.4	15.2
Not spending time with others	0.2	1.8
Arguing with other children/family	1.9	22.7
Being teased/called names	6.4	15.2
Made to feel left out	0.1	0.0
Other children asked questions	21.0	41.0

4.3.3 Results of the impact of DDE using P-CPQ

Data from 29 parents/carers of young people attending for treatment of DDE were analysed. Of these, 24 were mothers and five were fathers.

The mean P-CPQ score was 19.4 (SD = 13.6) and ranged from 4 to 47 (of a total of 124). There was no significant difference between the mean P-CPQ score for DDE and for the oral health sub-sample (mean P-CPQ = 14.0) ($p = 0.10$); the effect size of 0.39 was between 'small' and 'moderate'.

The mean number of impacts experienced 'often' and 'everyday or almost everyday' was 1.40 (SD = 1.83) compared to 1.06 for the oral health sub-sample ($p = 0.50$, t-test). No significant differences were found between the subscale or Family Impact Scale scores for the DDE group compared to the oral health sub-sample. These results echo those for CPQ₁₁₋₁₄, in terms of the lack of difference between the DDE group and the oral health sub-sample.

4.4 Discussion

The aim of this study was to investigate the impact of DDE on the OHRQoL of young people. This was identified as a gap in the current literature and important information to contribute to debates about the public health significance of DDE in the UK. In Chapter Two, the existing evidence about whether DDE are a public health problem was considered against four criteria. Much research has been conducted on the first criterion, regarding the prevalence of DDE in the UK. Some research has considered the effectiveness of interventions at preventing or treating DDE, but it was difficult to find UK data on the impact of DDE on individual children and young people or on wider society. CPQ₁₁₋₁₄ was identified as a suitable measure of the impact of DDE, but had not previously been evaluated for use in the UK. In this section the investigation of the impact of DDE will be discussed followed by the results of the evaluation of the measure, although some overlap is inevitable.

Based on this study, DDE appeared to have a low frequency of impact on the OHRQoL of young people attending for treatment of the condition. The frequency of impact of DDE

was similar to that experienced by those with no oral conditions, using two summary measures of CPQ₁₁₋₁₄ and P-CPQ, subscale scores and ratings of the affect on life overall. One explanation for the low frequency of impacts of DDE is that they had little impact on young people with the condition. Several studies, from other countries, have compared the impact of those with and without DDE, with inconsistent findings (Astrom and Mashoto 2002; Sujak *et al.* 2004; Michel-Crosato *et al.* 2005; Robinson *et al.* 2005). However, the current study was the first to take an extreme group approach and compare the impact on OHRQoL of those attending for treatment of DDE and those with relative oral health, thus an effect would have been expected. Moreover, that DDE has little impact on those attending for treatment appears counter-intuitive and further explanations were sought.

The other explanations include:

- The study was under-powered
- Problems due to the method of assessment of impact including:
 - responses based on the frequency of impacts
 - discriminative properties of CPQ₁₁₋₁₄
 - content validity
- Impact was mediated by other characteristics of the environment or the individual

The lack of difference between the DDE and relative oral health groups may be because the study was under-powered. Other studies have used similar sample sizes. For example, a study in Canada also using CPQ₁₁₋₁₄ had a sample of 71 children comparing the impact of caries (mean number of decayed teeth = 4.5) and orofacial conditions (including clefts and other craniofacial anomalies) on OHRQoL. Impact was not markedly different, although scores for several items differed significantly (Locker *et al.* 2005). Due to the absence of data for DDE in the UK, the sample size for the study was calculated using caries data from Canada. In Canada, a significant correlation between CPQ₁₁₋₁₄ score and the number of decayed tooth surfaces was found (Spearman's rank correlation coefficient = 0.64) (Jokovic *et al.* 2002). The sample size calculation suggested a total sample of 22 was needed, although a sample size of 30 per group was chosen to allow for cultural differences between the UK and Canada.

In addition, no relationships were found between the summary measures of CPQ₁₁₋₁₄ and DDE, and the only significant correlation for caries was between the number of impacts ‘often’ and ‘everyday’ and missing teeth due to caries (Spearman’s rank correlation coefficient = 0.21). The findings from this study suggest a sample size of 800 would be needed to have an 80% chance of detecting a difference between the impact of DDE on those attending for treatment and oral health (at a 5% level of significance with an effect size of 0.20). A sample of this size would be very large for an extreme group approach. Similarly, using the CPQ₁₁₋₁₄ data from the children with caries in this study, a comparison of the impact of DDE and caries would require a sample size of approximately 500. Future studies using CPQ₁₁₋₁₄ to detect differences between clinical groups will require larger sample sizes than used in this study.

A second explanation may be due to the properties of CPQ₁₁₋₁₄, particularly the response system, ability to discriminate between groups and content validity for DDE.

CPQ₁₁₋₁₄ scores the frequency with which oral conditions impact on young people. Assessing impact in terms of frequency may not be as appropriate for DDE as it is for other conditions. While the impacts of DDE on young people may be infrequent, when such impacts do occur they may be sufficient for young people to seek treatment. The item impact study, used by Jokovic 2002 to develop the measure originally, was based on the degree to which young people were ‘bothered’ by their oral health, with the frequency scoring system only introduced for the final measure. Many other HRQoL measures for children assess the degree of impact (Christie *et al.* 1993; Juniper *et al.* 1998; Landgraf *et al.* 1998; Edwards *et al.* 2005), although several adult OHRQoL measures record frequency (Slade and Spencer 1994; Locker and Jokovic 1996). More qualitative research is required to understand the nature (frequency and degree) of the impacts of DDE. Such a study is reported in Chapter Five.

Even taking an extreme group approach, CPQ₁₁₋₁₄ did not discriminate between those attending for treatment of the condition and a sub-sample that were orally healthy, this approach was taken to maximise the difference between the clinical status of those in the two groups. Interestingly, in a Ugandan study, CPQ₁₁₋₁₄ was able to detect differences in OHRQoL of young people with fluorosis of TFI score > 2 compared to those without

(Robinson *et al.* 2005), although numbers of young people with fluorosis were small. In Canada, CPQ₁₁₋₁₄ was able to discriminate between groups of young people attending different clinics, although the levels of disease present were much higher than in this study (Jokovic *et al.* 2002). Correspondence with the author revealed 53% of the sample in Canada had untreated caries, compared to only 21% of children in this sample (A. Jokovic personal communication). Such high caries levels in the sample originally used to evaluate the properties of the measure, may have over emphasised the sensitivity and discriminative powers of CPQ₁₁₋₁₄.

The Medical Research Council recommended further research on the impact of fluoridation on quality of life (Medical Research Council Working Group Report 2002). A study to meet this aim would require an OHRQoL measure able to discriminate between the impact of dental caries and fluorosis. If CPQ₁₁₋₁₄ was to be used for such an investigation, further testing of the discriminative properties of the measure would be needed, at the levels of the conditions in the population, and the substantive study would require a very large sample indeed.

Content validity is another relevant property of the measure that may account for the lack of difference between the groups. Discussions with young people revealed several important considerations about the items of CPQ₁₁₋₁₄ for young people with enamel defects, particularly relating to the relevance and wording of questions. The relevance of two items was questioned as young people were unable to see how ‘difficulty saying words’ and ‘drinking through a straw’ were important aspects of their oral health. These items had low scores in the item impact assessment. In addition, being accused of not brushing their teeth, an impact young people felt was relevant, was not included in the questionnaire. It may be that CPQ₁₁₋₁₄ did not capture all impacts relevant to young people with or without DDE, which is one of the disadvantages of using measures largely designed by adults with young people. CPQ₁₁₋₁₄ represents a generic OHRQoL measure for children with broad applicability to different dental and oro-facial conditions, rather than a DDE-specific measure. As discussed in section 2.5.2.2, two broad types of HRQoL measure exist: generic and specific. The main disadvantage of generic measures is their lack of relevance to all participants (Guyatt *et al.* 1993). The lack of difference between the groups may be

due to weak content validity of the measure for young people with DDE. Further testing of the content validity of CPQ₁₁₋₁₄ in the UK is required.

A third explanation for the lack of difference may be that the impact of DDE (and other diseases/conditions) was mediated by other factors. In quality of life research, the impact of severe chronic illness on individuals has also been reported to be similar to that of healthy people (Albrecht and Devlieger 1999; Rapkin and Schwartz 2004). In oral health research, Gregory and colleagues explored how people with very striking dental disease did not feel an impact (Gregory *et al.* 2005). An explanation offered for this paradox concerns characteristics of the environment or individual (Sprangers and Schwartz 1999). Wilson and Cleary's original model (section 2.5.2.1, Figure 4) includes the influence of environmental and individual factors on symptoms, functional and psychosocial impacts (Wilson and Cleary 1995). Possible environmental factors include cultural, social and material deprivation (Locker 1992). Variables such as general health status, household income and life stress have been shown to explain as much variance in the impact of oral conditions on adults, as clinical indicators such as missing teeth (Locker and Slade, 1994). Individual resources, such as personal control, optimism, social support and coping strategies, have also been suggested as possible determinants of the impact of health conditions (Sprangers and Schwartz 1999; Taylor and Seeman 1999; Locker *et al.* 2005). Indeed, in a study to evaluate the CPQ for 8 to 10-year-olds, relationships were found between OHRQoL and self-esteem (Humphris *et al.* 2005). The relationship of these environmental and individual factors, to OHRQoL in young people, needs further investigation. These factors will also be explored using qualitative methods in Chapter Five. In addition, more complex quantitative analysis, possibly using structural equation modelling (Baker *et al.* 2007), should be conducted in future.

Although there was no significant difference between the frequency of impact, as measured by CPQ₁₁₋₁₄, on OHRQoL of those in the DDE and oral health groups, there was a significant difference between the global ratings of oral health between these groups. This may be because the global rating does not include frequency as the basis of the responses, but an overall assessment of oral health generally. Single item global ratings have been used to summarise several health concepts, including biological, physiological, symptoms and functional states, when the use of HRQoL measures is not feasible (Robinson *et al.*

2003). Although some research has been conducted about older adult's use of global oral health ratings (Locker and Gibson 2005), little is known about the meaning of global rating for young people and further research is needed.

Although the frequency of the impact of DDE was found to be low, this study revealed certain aspects of young people's lives with which DDE was apparently associated, namely discomfort with hot and cold foods or drinks, breathing through the mouth, concern about what others think of their teeth, arguing with friends or family and missing school. Possible explanations may be that young people, with hypoplastic or hypomineralised enamel, experience more sensitivity to hot or cold. The item regarding breathing through the mouth had a high impact score for both samples and young people raised concerns about this item in the evaluation of both face and content validity. Concern about what others think was also an item of importance to young people with DDE. This may be due to the perceived visibility of defects to others, particularly at an age when approval from peers is important (Harter 1990). The higher score, for arguing with friends or family, may be due to negative interactions with others about DDE. Finally, as the young people were all attending the Dental Hospital for treatment, they may have experienced multiple visits to their own dental practitioners, as well as, visits to the hospital clinic, which resulted in time away from school.

Alone, quantitative data are not sufficient to fully investigate the impact of DDE and qualitative research is required. A more detailed exploration of these impacts is undertaken in the qualitative study described in Chapter Five.

Also of relevance to the aim of the study was the finding of weak relationships between the presence of DDE and summary measures of CPQ₁₁₋₁₄ (except for the functional subscale score). No relationships were apparent between the summary measures of P-CPQ and the presence of DDE. Indeed, the relationships between the clinical and OHRQoL data were also inconsistent. The relationship between the presence of DDE and the functional subscale score may be because young people with DDE have difficulty drinking or eating hot or cold foods, due to dentine sensitivity (this item had a high impact score), or due to type 1 error. Studies of OHRQoL in adults have shown tenuous links with clinical indicators (Cushing *et al.* 1986; Locker 1992; Locker and Slade 1994; Soe *et al.* 2004) and

various explanations have been offered, including the suggestion that overall differences between the concepts of health and disease may be responsible (Gregory *et al.* 2005). OHRQoL measures such as CPQ₁₁₋₁₄ are based on a conceptual model of oral health that embraces biological and psychosocial dimensions of health, in contrast to the focus of clinical data on normative assessments of biological process. These differences were described in Chapter Two, when the emphases of the biomedical model of disease and the biopsychosocial model of health were outlined. The relationship between the impact and clinical assessment of DDE will be explored qualitatively in Chapter Five.

The first objective of this study was to assess the properties of CPQ₁₁₋₁₄ (and its analogous parental/carer version) in a general clinic-based sample, including the young people's perspective on the measure. CPQ₁₁₋₁₄ had satisfactory internal consistency and test-retest reliability. Some concerns were raised about the face validity, although the number of missing responses was low. The criterion validity of the summary measures of CPQ₁₁₋₁₄ was acceptable (Table 19), although the correlations between the emotional and the social subscale were not statistically significant. Construct validity was acceptable, in relation to personal assessment of life overall, but sporadic and inconsistent for measures of clinical status (Table 18).

The face validity of CPQ₁₁₋₁₄ was investigated through discussions with five participants and revealed several aspects of the questionnaire that caused confusion. Double questions, where several activities are included in one question, are commonly used in HRQoL measures and leave participants with a dilemma to either ignore some of the activities or to average out their problems by selecting a middle response option (Mallinson 2002). The number of missing responses in this study was low. However, participants do respond to items, even if they don't understand the question (Clarke and Schober 1992). The young people also interpreted certain questions differently than might be expected, particularly relating to teasing or being called names. The meaning of these words to young people may differ from the meaning intended in the measure. Other research has also shown that people interpret survey questions in unexpected ways (Tanur 1992).

Face validity of health-related and OHRQoL questionnaires is often neglected and, when it is assessed, it is often investigated by a panel of 'experts', scrutinising the items during the

pilot stage of development (Guyatt *et al.* 1993). An expert panel of health professionals and parents was involved in the development of CPQ₁₁₋₁₄. However, they failed to identify several problems raised by young people in this study. In future, assessment of face validity should include the perspective of the participants. The potential problems discovered with the face and content validity of CPQ₁₁₋₁₄ suggests that attempts to involve young people in its development may not have been sufficient and further qualitative testing is required, before this measure is used more widely. Mallinson suggested a method, which has been applied to a generic HRQoL measure, and resulted in recommendations to improve the clarity for participants (Mallinson 2002). These findings support the importance of language use in child-centred research, as was discussed in Chapter Two (section 2.5.6.2).

The paper about the development of CPQ₁₁₋₁₄ (Jokovic 2003) was classed as category 1 in the systematic review (Chapter Three, section 3.3.2.1). The measure was designed using a recommended method to include items of importance to young people with oral and orofacial conditions. In-depth interviews were carried out with 11 young people, although it wasn't clear whether advice was sought from them on the exact wording of the items. For the selection of the final items, the authors relied on the item impact study. However, the method of selection was not based purely on items with the highest impact scores. Instead, the items were organised into health domains and the highest ranking items in these domains were included. For example, 'difficulty drinking through a straw' had an impact score of 2.5 and was included in the functional limitations subscale, whereas 'being jealous of family members', with an impact score of 16.9, was not included in the emotional well-being subscale. The domains of the measure were chosen by the adult researchers to organise the items, but may not have been domains that were important to young people with DDE or young people more generally.

The reliability and validity of P-CPQ were acceptable for use in the UK if 'don't know' responses were adjusted. The distribution of 'don't know' responses was investigated as an indicator of what parents/carers know about the impact of their child's oral condition on their lives. An examination of the items that resulted in the highest number of 'don't know' demonstrated that parents/carers were not always able to detect some unobservable impacts of oral conditions on their children, such as 'breathing through the mouth' or

'getting food stuck between teeth or in the roof of the mouth'. The greater ability of parents to rate their child's HRQoL for observable functioning is consistent with the findings of a systematic review (Eiser and Morse 2001). The initial evaluation of P-CPQ had similar findings regarding the questions that elicited the highest number of 'don't know' responses. The authors considered removing these items, but decided against it, for fear of compromising the content validity (Jokovic *et al.* 2004).

As is the case with any research, aspects of the design and conduct of this research may have affected the findings. Firstly, from a child-centred perspective, the choice of the clinic as a setting for completion of the measure may have affected the responses and the young people may have felt under pressure to participate, due to the power imbalance between themselves as patients and their dentist recruiting them to the study. This method was chosen, as opposed to asking children to complete the questionnaire at home, to improve the response rate. An information sheet, designed for children of between 11 and 14 years was given to participants informing them that it was their choice to participate and their treatment would not be affected if they declined. Assent from children was gained, rather than consent, in line with the research ethics committee's recommendation at the time; such committees now encourage consent from children themselves.

In conclusion, DDE had a low frequency of impacts on young people attending for treatment of the condition, when assessed using a rigorously tested OHRQoL measure. Possible explanations for this finding include under-powered sample, low levels of impact associated with DDE, the properties of the measure or mediating individual or environmental effects. A more exploratory consideration of the impacts of DDE on young people is required.

4.5 Publications arising from the work in this chapter

i) Poster:

Z. Marshman, H.D. Rodd, M. Stern, C. Mitchell, D. Locker, A. Jokovic, P.G. Robinson
An evaluation of the Child Perception Questionnaire in the UK. International Association
of Dental Research Conference, Honolulu, March 2004.

ii) Presentation:

Z. Marshman, H.D. Rodd, M. Stern, C. Mitchell, P.G. Robinson. Treatment of 'Don't
know' responses in a parental Oral Health Related Quality of Life questionnaire. British
Society of Dental Research Conference, Birmingham, April 2004.

iii) Publications:

Z. Marshman, H.D. Rodd, M. Stern, C. Mitchell, P.G. Robinson. Evaluation of the Parental
Perceptions Questionnaire, a component of the COHQoL, for use in the UK. Community
Dental Health-in press, to be published December 2007.

iv) Z. Marshman, H.D. Rodd, M. Stern, C. Mitchell, D. Locker, A. Jokovic, P.G. Robinson.
An evaluation of the Child Perceptions Questionnaire in the UK. Community Dental Health
2005; 22: 151-155

5. Chapter Five

Exploring the impact of DDE on young people

5.1 Introduction

Chapter Four reported a quantitative study of the impact of enamel defects on young people. The frequency of the impact of DDE on young people attending for treatment was low and was broadly similar to that experienced by those with no oral conditions. While the study allowed the description of the impact of DDE on OHRQoL and enabled comparison, it may not have captured all aspects of impact relevant to children and young people themselves. Several areas of impact of DDE were identified as requiring further exploration. For example, ‘concern what other people think of your teeth’ had a high item impact score and would benefit from further expansion on the role of peer approval about the appearance of teeth for young people. Also, the study found missing school and arguing with other children or family important for those with DDE, but did not provide information on how young people perceived this impacted on their lives. Another possible determinant of the impact of dental conditions was identified in the discussion of the study, namely individual factors, such as self-esteem and coping (Sprangers and Schwartz 1999; Taylor and Seeman 1999; Humphris *et al.* 2005). As the aim of the thesis is to investigate the impact of DDE on individuals, these factors require further attention.

Qualitative research provides a complementary method to quantitative research. It explores experiences, feelings and perceptions (Morse 1992) and allows the emergence of topics important to participants, rather than restricting data to the preconceived ideas of researchers. Such methods have not previously been applied to investigating the impact of DDE. Qualitative research has been used widely, in relation to general health, and has already been used with young people with Treacher Collins syndrome (Beaune *et al.* 2003) and to gain children’s general perceptions of oral health (Ostberg *et al.* 2002; Fitzgerald *et al.* 2004). The impact of Treacher Collins syndrome was mainly related to social interactions and the importance of making friends (Beaune *et al.* 2003). Young people in New Zealand perceived oral health as largely irrelevant to their everyday lives, with the exception of having attractive teeth for social reasons (Fitzgerald *et al.* 2004).

Qualitative methods allow the impact of DDE to be explored at a higher level of child-centeredness, achieving greater participation of children by giving them a voice to explain their perspectives in their own words. This approach also allows exploration of variation between individuals, so children are not viewed as a homogenous group, just because they belong to a certain age-group (Christensen and James 2000).

This study aimed to explore, in detail, the impact of DDE on children and young people, through their experiences of the condition and its meaning to their everyday lives.

5.1.1 Objectives of the study

- a) To explore the meaning of DDE for young people
- b) To explore variation between individuals on their experiences of DDE

5.1.2 Design of the study

Methodological considerations of research with children and young people were discussed in section 2.5.6. To maximize the involvement of children, the study was designed to consider factors such as power imbalance, language use, choice of setting, an appropriate framework for analysis and assuring the quality of data.

5.1.2.1 Theoretical framework for analysis

Theoretical frameworks are used in qualitative research to help interpretation of the data. To ensure the involvement of children, the selected framework reflected the lives of young people. Previous research on the impact of oral conditions and the findings from the quantitative study identified social interactions as an important factor. Therefore, after a review of possible frameworks, symbolic interactionism was chosen.

The main principle of symbolic interactionism is that the self is established and developed through interactions. The self is the interactionist term for the continually changing individual (Strauss 1962). Development of the self begins during childhood from the process of interaction and interpretation of how others react (Mead 1934) and continues throughout adulthood. Symbolic interactionism has three premises. First, that human beings act towards things e.g. physical objects, other people, activities or situations on the basis of the meanings they have for them. Secondly, the origin of the meaning of a thing is

formed through interactions, particularly language used between people. The third premise then explains that subsequent use of meaning by a person involves interpretation, transforming the meaning in the light of the situation (Blumer 1969). Thus this framework emphasises the self and interactions, particularly from the perspective of meaning, with attention paid to use of language.

Symbolic interactionism is well established in medical sociology and has already been applied to research of young people's experiences of chronic illness (Woodgate 1998). This framework allowed a focus on the interplay between social interactions and the meaning of DDE for young people in the situations in which they live.

5.2 Methods

5.2.1 Sample

The study involved young people with varying severities of DDE on their incisor teeth. Recruitment occurred in two areas: one with no fluoride added to the drinking water (Sheffield) and an area with 1 part per million added (West/North Lincolnshire). This was in case of differences in the experiences of DDE in communities where defects are relatively more prevalent, in comparison to those where they are less common (Ellwood and O'Mullane 1995; Wondwossen *et al.* 2003). Young people, aged 10 to 15 years, were chosen because this is the age when the impact of physical attributes and changes associated with puberty affect the individual (Roth and Brooks-Gunn 2000).

Young people with medical or dental conditions that might influence their experiences of their teeth, including those with fixed appliances or a definite need for orthodontic treatment were excluded as were young people with cognitive or language difficulties.

5.2.2 Procedure

Participants were recruited by a Consultant in Paediatric Dentistry in Sheffield and by primary care dentists in Lincolnshire. Potential participants were first asked by the dentists, as part of their routine care, if they were aware they had the condition. Only those who were aware were invited to participate for ethical reasons. Young people who met the inclusion criteria were introduced to the study by their dentist and an age-appropriate

information sheet was given to them and their parent/carer. They were given a response slip and stamped addressed envelope to express their desire to be involved. Respondents were then contacted and a convenient date for the interview was arranged. Using dentists as ‘gate-keepers’ allowed participants the opportunity for further discussions or treatment after the research, if necessary. Consent was gained from both the young person and the parent/carer.

The dentist also indicated the approximate severity (mild/moderate/severe) of each potential participant’s incisors to assist sampling by severity. In addition, age and gender were included in the sampling framework as these factors had been suggested as being responsible for variation in the impact of DDE (van Palenstein Helderma and Mkasabuni 1993; Astrom and Mashoto 2002). This sampling framework suggested that between 18-36 participants would be required (Table 29). Qualitative research involves relatively small samples that do not attempt to be statistically representative, thus sample size calculations are not applicable (Sandelowski 1995). Recruitment continued until the point where no new information emerged during the interviews and data became repetitious, indicating ‘saturation’ (Glaser and Strauss 1967).

Table 29. Sampling framework showing estimated number of participants required

	10/11 year old		12/13 year old		14/15 year old	
	Male	Female	Male	Female	Male	Female
Mild	1-2	1-2	1-2	1-2	1-2	1-2
Moderate	1-2	1-2	1-2	1-2	1-2	1-2
Severe	1-2	1-2	1-2	1-2	1-2	1-2
Estimated sub-totals	3-6	3-6	3-6	3-6	3-6	3-6

5.2.3 Interviews

Semi-structured interviews were used, providing a loose structure for open-ended questions to permit young people to describe their own experiences in their own words (Bailey 1978). Interviews, rather than focus groups were chosen to ensure a depth of coverage (Crabtree and Miller 1999). The interviews started by introducing the study and establishing that participants could stop the interview at any point, to reassure them of the confidentiality of

their responses, to inform them of the purpose of the voice recorder and to establish that they were the experts on the subjects covered. All interviews were tape recorded.

The areas to be covered were outlined in an initial topic guide that was informed by the theoretical framework, the findings from the quantitative study, the literature review and informal conversations with young people with DDE. The topic guide began with a general enquiry about the young person (such as hobbies and likes/dislikes) before moving onto more specific and personal areas (Horowitz *et al.* 2003) (Appendix F). It was piloted with several young people before data collection began. As the interviews proceeded and initial analyses were carried out, the topic guide was modified to further explore emerging topics. Probes and prompts were employed such as the use of comparisons, hypothetical questions and paraphrasing (Stone and Lemanek 1990). Participants were also asked to comment on ideas from other participants (Woodgate 1998); a technique known as continuous feedback. Field notes were made after the interviews, including observations about non-verbal cues and points for the analysis.

The interviews were conducted in participants' own homes, most without the presence of parents or siblings, but for several interviews a request was made for a parent to be in attendance (noted in the field diary). At the end of each interview the participants were debriefed about the study. A summary of the findings was sent to each young person at the end of the study with participants given the opportunity to return feedback in a stamped addressed envelope.

Following training in a standardised photographic technique, a digital camera was used to take two photographs of each participant's upper and lower incisors after the interview. A Finepix S2 Pro camera (Fujifilm, Tokyo, Japan), an AF Micro Nikkor 105mm lens (Nikon, Tokyo, Japan) and a Macro Speedlight SB-29 (Nikon, Tokyo, Japan) were used. One photograph was taken of the teeth wet with saliva and a second photograph was taken 60 seconds later when teeth had dried out, as drying can effect the grading of teeth (Cochran *et al.* 2004). The photographs were scored using the TFI and the modified DDEI for general purposes by a distant assessor who was calibrated in the use of these indices. Intra-examiner reproducibility was measured using 15% of the photographs. The assessor was masked to the fluoride levels in the water supplies of the participants.

5.2.4 Data analysis

Interview recordings were transcribed verbatim by the interviewer as soon as possible after the interview to allow data to be analysed as soon as collected.

Consistent with symbolic interactionism and the objectives of the study, each transcript was studied to try to establish the meaning of DDE for that participant, the language used to describe them, whether DDE had featured in social interactions and whether or not the defects had an impact on the young person as a result. The second stage of the analysis looked for variation within the data. This stage began after the first six interviews. Constant comparative analysis was used to allow full characterisation of variation by looking for comparisons and differences (Glaser and Strauss 1967). Although developed for use in grounded theory, this approach has been widely used to expand understanding of experiences (Thorne 2000).

As well as comparing the meaning and impact of DDE, variation between individuals in terms of age, gender and severity were investigated within the data. As the analysis proceeded and themes emerged the relevant literature was consulted to provide a fuller interpretation of the data. This approach is characteristic of the iterative nature of qualitative research.

The results are presented as the concepts that emerged from the data rather than preconceived ideas. Quotes are used to support points and illustrate ideas; pseudonyms are used to ensure confidentiality. Participant's own words are used where appropriate in keeping with traditions in qualitative research, this also allows the results to remain as close as possible to representing young people's own perspectives.

This study was approved by the North Sheffield Research Ethics Committee (Appendix G contains the information sheets and consent forms).

5.3 Results

5.3.1 Sample

Twenty-one participants were interviewed before data saturation occurred. To recruit this number, 25 young people were approached, but 4 declined. Thirteen were female. The participants were aged from 10 to 15 years and came from both rural and urban areas across South Yorkshire, Lincolnshire and North Nottinghamshire. Interviews lasted between 21 and 48 minutes.

5.3.2 DDE status of the young people

Two photographs of each of the 21 participant's teeth were scored, but one participant's photographs were excluded due to the presence of restorations. Table 30 shows the characteristics and DDE scores of participants, with differences in grading between wet and dry photographs for some participants. Intra-examiner reproducibility (involving scoring 15% of photographs twice) found 85% agreement. The TFI scores ranged from 0 to 5. A TFI score of 5 indicates 'the entire surface exhibits marked opacity with focal loss of outermost enamel less than 2mm in diameter'. Eleven participants had an incisor that scored TFI 3 or above, categorised as being of aesthetic concern (McDonagh *et al.* 2000). Five participants scored zero on the TFI suggesting the defect did not have the appearance of fluorosis. Using the general purpose version of the modified DDEI, 11 participants had diffuse opacities, most of which were 'patchy' and 5 had demarcated opacities. Extent of defects on participants' incisors ranged from normal to covering at least two-thirds of the tooth surface.

Name	Age	Gender	State	DDE-type				DDE-extent				TFI score			
				UL1	UL1	LL1	LR1	UL 1	UL1	LL1	LR1	UR1	UL1	LL1	LR1
Phillipa	14	F	Wet	4	4	0	0	1	1	0	0	3	3	1	1
			Dry	3	3	0	0	1	1	0	0	2	2	0	0
Richard	11	M	Wet	2	0	1	0	1	0	1	0	0	0	0	0
			Dry	2	0	1	0	1	0	1	0	0	0	0	0
Joseph	12	M	Wet	0	1+2	0	0	0	1	0	0	0	0	0	0
			Dry	0	1+2	0	0	0	1	0	0	0	0	0	0
Nick	13	M	Wet	5	6	3	3	3	3	1	1	4	5	2	2
			Dry	5	6	3	3	3	3	1	1	4	5	2	2
Michelle	12	F	Wet	6	6	4	4	2	2	3	3	4	4	3	3
			Dry	6	6	4	4	2	2	3	3	4	4	3	3
Martin	12	M	Wet	5	4	0	0	1	1	0	0	4	3	0	0
			Dry	5	4	0	0	1	1	0	0	4	3	0	0
Jane	10	F	Wet	1	1	0	0	1	1	0	0	0	0	0	0
			Dry	1	1	0	0	1	1	0	0	0	0	0	0
Natasha	14	F	Wet	5	5	5	5	3	3	3	3	4	4	4	4
			Dry	5	5	5	5	3	3	3	3	4	4	4	4
James	15	M	Wet	1	1	0	2	1	1	0	1	0	0	0	0
			Dry	1	1	0	2	1	1	0	1	0	0	0	2
Peter	14	M	Wet	4	4	-	-	2	2	-	-	3	3	-	-
			Dry	4	4	-	-	2	2	-	-	3	3	-	-
Julie	13	F	Wet	6	6	5	5	2	2	3	3	5	5	4	4
			Dry	6	6	5	5	2	2	3	3	5	5	4	4
Helen	14	F	Wet	5	5	4	4	2	2	1	1	4	4	3	3
			Dry	5	5	4	4	2	2	1	1	4	4	3	3
Emma	12	F	Wet	4	4	0	0	1	1	0	0	0	0	0	0
			Dry	4	4	0	0	1	1	0	0	0	0	0	0

				UL1	UL1	LL1	LR1	UL 1	UL1	LL1	LR1	UR1	UL1	LL1	LR1
Clare	13	F	Wet	1	1	0	1	1	1	0	1	0	0	0	0
			Dry	1	1	0	1	1	1	0	1	0	0	0	0
Lucy	12	F	Wet	4	4	0	0	1	1	0	0	1	1	0	0
			Dry	4	4	0	0	1	1	0	0	1	1	0	0
Gemma	12	F	Wet	3	3	3	3	2	2	1	1	2	2	1	1
			Dry	3	3	3	3	2	2	1	1	2	2	1	1
Louise	13	F	Wet	4	4	3	3	3	3	1	1	3	3	2	2
			Dry	4	4	0	0	2	2	0	0	3	3	0	0
Andrea	14	F	Wet	4	4	0	0	1	1	0	0	3	3	0	0
			Dry	4	4	0	0	1	1	0	0	3	3	0	0
Jessica	11	F	Wet	5	6	4	4	2	3	1	1	4	5	3	3
			Dry	5	6	3	2	2	3	2	2	4	5	3	3
Andrew	14	M	Wet	3	3	3	3	2	2	1	1	2	2	1	1
			Dry	3	3	3	3	1	1	1	1	1	1	1	1
Sam	12	M	Wet	Excluded from scoring as had anterior restorations											
			Dry												

DDE = Modified Developmental Defects of Enamel score

TFI = Thylstrup and Fejerskov Index score

'-' = unable to see teeth sufficiently on photographs

5.3.3 Qualitative data

Participants were asked initially about the language they themselves used to describe people of their age, words used were *'young people'*, *'teenagers'*, *'pre-teens'* and *'young adults'*. The term *'children'* was not used by participants. Participants are therefore referred to as young people.

The data provided an insight into the psychological and social systems of young people in terms of their sense of self and the nature of their social interactions. DDE were a feature of such interactions and for some young people had an impact on their sense of self. The variation in the impact was characteristically described in terms of the degree to which DDE *'bothered'* them. Other possible explanations for variation in impact such as age, gender and severity were also considered.

5.3.3.1 Young people's sense of self

Participants often described themselves in terms of interpersonal traits. For example, while descriptors such as *'happy'*, *'honest'*, *'stupid'*, *'forgetful'* and *'clumsy'* were used by some, the main emphasis of most descriptions was on interpersonal characteristics such as *'being friendly'*, *'a good listener'*, *'caring'*, *'popular'* or *'sensitive to others'*. Gemma's description of herself wholly in terms of interpersonal traits was characteristic:

'I'm hopefully kind to my friends. I'm friendly, friendly to talk to at school, polite hopefully. I'm not very confident in myself like, I don't get up and speak to people I don't know' Gemma, 12 years old.

5.3.3.2 Young people's social interactions

Not surprisingly, given the emphasis of the descriptors, relationships, particularly *'having friends'*, were very important to participants. This was evident as the subject of friends was talked about expansively and without prompting. Some participants talked of layers of friends beginning with their best friend/friends then groups of close friends, then peers in their class and, finally, a wider circle in their school or neighbourhood. Others talked of friends they knew through school, who lived nearby or those known through hobbies. The social relations described were principally with people their own

age. Other significant relations were with siblings, with little mention of parents or teachers.

5.3.3.3 Types of social bonds

The nature of social relations varied by the strength of the bonds between individuals. Three types of bond were evident; the secure bonds of friends, threatened bonds that resulted from conflicts between individuals and newly-formed bonds. Interactions about DDE featured in each type of bond.

5.3.3.4 Secure bonds

Secure bonds existed between best friends and between groups of close friends. Having and naming a best friend was a consistent finding along with counting numbers of friends in *'their group'*. The language used to describe interactions with friends ranged from *'playing with friends'* by younger participants to *'hanging out'* or *'going out with friends'* by older young people. Younger participants had an emphasis on activities undertaken with friends, for older participants, trust and having a confidante featured more highly. As young people grew older the concept of friendship appeared as a more 'adult' model:

'we'll sit in the house playing with my toys, making games up and everything or we'll go back outside and build dens' Jane 10 years

'trustworthy, got to be trustworthy, nice, I wouldn't want friends who were horrible to other people' Peter 14 years

The use of nick names was common among close friends:

'... my friend Nadia, we call each other nick names, she's called fish face and I'm rabbit teeth, we don't really care because we call each other silly names'
Michelle 12 years

References to DDE within secure bonds included asking a friend's opinion and receiving compliments. Jessica described the discussions with her best friends about teeth:

'They say "what's wrong with them?", cos they don't see what's so bad about them, Jennifer hates hers, they are bigger than she wants them to be and a bit crooked here and there, but it doesn't matter to me really because I know that deep down she's a nice person' Jessica 11 years

5.3.3.4.1 Threatened bonds

A recurrent feature was episodes that appeared to threaten social bonds such as teasing, particularly in the form of name-calling or 'falling out with friends'. These threatened bonds resulted from disagreements between friends within groups or comments made by people from rival groups.

The topics for teasing frequently featured physical aspects such as facial features (hair colour and style, nose and ear size, skin in terms of spots/freckles and teeth), sensory impairments (hearing aids, speech and glasses) and body weight. Apart from physical aspects, other topics for teasing were mental illness, personality and bereavement:

'There was a girl, her mother died after Boxing Day and her younger brother was getting teased, saying "we have a mother and you don't" and Eve got teased because erm everybody was saying she was bragging she was the best athlete when she wasn't.' Lucy 12 years

These topics were consistently related to 'being different', particularly visible physical difference.

'Some people get teased because they are fat and some people just get teased because they are not right' Emma 12 years

Teasing about teeth related to the size and colour of teeth and orthodontic appliances. Consistent with other aspects of physical appearance that made individuals different, DDE were a topic for teasing. Some young people had experienced teasing and name-calling about their DDE, usually as a result of conflict between individuals:

'I don't like the thing on my tooth and I don't like my big ears. Whenever I get into an argument with someone that's the first things they go on about' Richard 11 years

'Well at school there is this boy who calls me the plaque blob. It's a bit stupid to call me the plaque blob when he is actually very fat...at first I thought he were jealous because I was more of a friend to this other friend than him....' Joseph 12 years

5.3.3.4.2 New social bonds

Young people talked about the social bonds that resulted from meeting people for the first time. This was particularly relevant to the major transition from primary to secondary school:

'Cos our primary school was very small, there was only 200 odd people and when we went to St Johns, it's massive and meet more people, become more

wary of your surroundings...there is a lot more people to compare yourself to'
Phillipa 14 years

Jessica described her recent experiences of moving to secondary school and how this made her feel about her DDE:

'I started to become more self-conscious of my teeth, it's probably as I met more and more new people and saw more new teeth, it got a bit more daunting'
Jessica aged 11 years

Natasha recalled that at primary school she was unconcerned by her DDE and described when she first became conscious of them:

'Secondary school I started to think, I don't know, I just looked in the mirror and thought I was different' Natasha 13 years

Another occasion for new social bonds was holidays. Holidays were mentioned as an occasion when DDE became the subject of inquisitive interactions:

'Erm whenever I go on holiday or something when I meet some friends I don't know they say, "I don't mean to be bad, but I think you've got something on your teeth there"they thought I wasn't brushing them and stuff...I just went "I don't know why it's there, I brush my teeth everyday"' Richard 11 years

In summary, social interactions had an important place in young people's lives, particularly the relations they had with people of their own age. The nature of the social bonds between individuals varied where interactions were with close friends, new acquaintances and when conflict threatened the bonds. Close friends with secure bonds provided someone to talk to about concerns and provided support about DDE. When bonds were threatened DDE became a topic of teasing and were used as ammunition in relationship breakdowns. When new social bonds formed, during holidays and changing schools, DDE were also a feature of social interactions.

5.3.3.5 The impact of DDE on young people: 'not bothered' to 'really bothered'

The phrases used by participants to refer to their DDE included 'marks', 'markings', 'white spots', 'speckles', 'things', 'lines', 'stains' and 'bits'. DDE, from the perspective of participants, will therefore be referred to as marks for the remainder of this chapter. Marks caused a range of impacts, 'bothering' young people to varying degrees. No participants reported symptoms such as sensitivity to hot and cold or breathing through their mouths. For some young people, marks appeared to have no impact; this was characteristically described as being 'not bothered'. For example:

'The dentist told me, er not the last time I went, but the time before, but I'm not really bothered' Martin 12 years

'They are not white, but not brown, sort of in-between, it wouldn't bother me if I had it for the rest of my life' Andrea 14 years

In contrast, for others having marks had a negative impact.

'I don't like the colour, I'm conscious about it, when I'm talking I don't like showing them...I'm actually quite bothered' James 15 years

Richard described how his teeth made him feel:

'I don't like the things on my teeth, it has never really made me cry, but I've been upset a few times' Richard 11 years

However, an advantage of having marks was missing time from school to go to the dentist:

'I think its quite fun going to the dentist cos I get to miss time off school' Nick 13 years

Marks also made some young people feel good about themselves as illustrated by Peter:

'I'm quite proud probably, I've got white teeth' Peter 14 years

5.3.3.6 Variation in the impact of marks

It emerged that the impact of marks on an individual was dependent on aspects of the individual; their self. With the emergence of this theme of sense of self, the literature was reviewed.

Two early symbolic interactionists, William James and Charles Cooley, were amongst the first scholars to study the self. James suggested that people differ in what domains of their lives define their sense of self:

'self-feelings depend entirely on what we back ourselves to be and do' (James 1890)

He went on to describe how the defining characteristics of sense of self develop:

'so the seeker of his truest, strongest, deepest self must review the list carefully, and pick out the one on which to stake his salvation' (James 1892).

More recent empirical research has confirmed James' hypotheses in young people in that their sense of self is contingent on success in the domains of importance to them, with the most common domains being appearance and popularity, rather than academic competence (Rosenberg 1979; Harter 1985).

Cooley, however, focused on the self as a social construction and considered the importance of others in the formation of sense of self. According to Cooley, 'self-feelings' arise from monitoring of one's own appearance and actions by viewing one's self from the perceived standpoint of others. He described the development of self during interactions as using other people as a looking glass, a notion described as the 'looking-glass self' (Cooley 1902). The 'looking-glass self' has three components:

- Self - appraisals
- Actual appraisal of significant others e.g. close family and friends
- Perceptions of the appraisals of others (reflected appraisals)

Thus a 'sense of self' develops as people gaze into a social mirror to determine whether they have other people's approval. The notion of the looking-glass self has also been confirmed in young people. Moreover, there is variation between individuals on the need for approval from others to validate their own sense of self (Harter *et al.* 1996; Cash and Fleming 2002).

James' and Cooley's approaches to self have different emphases, but neither is mutually exclusive. Both processes have been identified as important in the development of sense of self in young people (Harter 1986; Harter 1990; Harter *et al.* 1996). Both were observed in the present study and appeared to explain some of the variation in impact of marks between young people.

5.3.3.7 Marks and defining aspects of sense of self

Variation was observed between participants whose sense of self was contingent on appearance and those who attached more importance to other domains such as personality. For example, for James, appearance was an important part of his sense of self. He disliked the colour of his teeth and overall, having defects '*bothered*' him. In contrast, Nick's sense of self was contingent on personality, he was unconcerned about the appearance of his teeth generally, or about having marks specifically.

There was also variation in the importance of the perceived approval of others as a contributor to sense of self. For some young people perceptions of how others appraised their teeth was particularly important. It was not what others had actually said about their teeth that they internalised, but the reflected appraisals of others and how they might be judged on the basis of this. For example, no one had ever said anything negative about Jessica's marks, friends had reassured her about them, but she described her perceptions of what other people at school thought about them:

'They say "they are fine, nothing is wrong with them", but even though they are not saying anything, I bet people are thinking it and I don't want them too'
Jessica 11 years

For these young people varying significance was attached to gaining approval depending on the appraiser. The perceived approval of peers (class mates and young people in the neighbourhood) was more important than actual approval of significant others such as close friends.

Other young people were unconcerned about peer approval, even when they had received negative comments:

'This boy called David, his twin called Jade and Jamie and Darren would go around saying I've got stickers on my teeth and trying to wind me up, they started teasing me... I wasn't really bothered about it' Jane 10 years

When both James' defining characteristics and Cooley's looking-glass self approaches to sense of self were considered together they helped explain the variation in the effect of DDE. Examples to illustrate the application of these components to the data both to those 'bothered' and those 'not bothered' are given in Table 31 and 32. Those for whom appearance was a defining characteristic of their sense of self and who relied on the reflected appraisals from others were bothered by their marks (Table 31). Their consciousness about their appearance led them to perceive the appearance of their teeth to be poor (self-appraisal). They also interpreted reflected appraisals of their teeth to be negative. This perception was independent of what other close friends had actually said to them (actual appraisal).

Table 31. Example of analysis- young people 'bothered' by marks

	Richard	Natasha	James	Jessica
Age	11	14	15	11
DDEI type	2	5	1	5/6
DDEI extent	1/0	3	1	2/3
TFI score	0	4	0	4/5
Appearance and sense of self	<i>'I'm confident about stuff, but I'm not confident about my appearance'</i>	<i>'Self-conscious' about appearance</i>	<i>'I'm conscious of the way I look, it's a personal thing'</i>	Particularly as changing schools. <i>'I don't want to look at myself in the mirror 'cos I know I'll look horrible, I hate photos, I always look awful'</i>
Actual appraisals of others about teeth	Negative: <i>'Whenever I get into an argument with someone that's the first thing they go on about'</i>	Positive comments from friends & sister: <i>'Nat goes "they are a nice shape and a nice colour"'</i>	Neutral appraisal from Mum: <i>'She said nowt, its just a natural thing'</i>	Positive appraisals from friends: <i>'Jennifer doesn't see what's wrong with mine'</i>
Self-appraisal of teeth	<i>'I don't like that thing on my tooth'</i>	<i>'I always wanted to change my teeth'</i>	<i>'The colour, I don't really like it'</i>	<i>'I was about 7 or 8 when my teeth got bad'</i>
Reflected appraisals about teeth	Appraisals from others negative, this reinforced self-appraisals	Perceived others appraisals to be negative, although actually positive	Perceived others appraisals to be negative, although neutral	Perceived others appraisals to be negative, although actually positive
Impact	<i>'I've been upset a few times'</i>	<i>'I'm loads bothered'</i>	<i>'I'm actually quite bothered'</i>	<i>'become more self-conscious'</i>

Photographs of teeth of young people described in Table 31.

Richard



Natasha



James



Jessica



For example, Natasha was conscious of her appearance and *'loads bothered'* by the colour of her teeth. Despite reassuring actual appraisals from those close to her that her teeth were attractive she still perceived the way other people interacted with her was different because of her teeth. Without the marks she said:

'People would smile more' Natasha 14 years

Some young people for whom appearance was important had received actual negative appraisals from others about their teeth and this reinforced their own negative feelings about them and challenged their sense of self further:

'They used to say "your teeth are going yellow" ...I didn't want to show my teeth when I smiled. Teeth have to be white, I used to worry and now I can't wait to get them things off' Richard 11 years

In contrast, some of those *'not bothered'* by their marks (Table 32) had also received negative interactions about them. However, these young people differed in that their sense of self was not contingent on appearance. For example, Michelle who said:

'it doesn't really matter what you look like, its personality' Michelle 12 years

had received negative appraisals from others about her teeth, but, overall, perceived others felt her teeth were acceptable. Her *'things'* had little impact on her:

'I don't moan about the things on my teeth because it doesn't matter, they are stuck there and there's no point bothering about it' Michelle 12 years

Unlike those bothered by their teeth, these young people were able to dismiss negative interactions as a consequence of conflict or a topic for teasing. In symbolic interactionism the way people interpret interactions differently is said to depend on their *'definition of the situation'*. For those for whom appearance was not an important characteristic of their sense of self, the definition of the situation was different and consequently reactions were processed differently.

Table 32. Example of analysis- young people ‘not bothered’ by marks

	Michelle	Jane	Nick	Lucy
Age	12	10	13	12
DDEI type	6	1	4/5	4
DDEI extent	2	1	3	1
TFI scores	4	0	5/6	1
Appearance and sense of self	<i>‘I’m happy with the way I am, it doesn’t really matter what you look like its personality’</i>	<i>‘I notice personality, how people act’</i>	<i>‘I think it’s the personality that counts’</i>	<i>‘I really think personality is more important’</i>
Actual appraisals of others about teeth	<i>Negative: ‘When people are being nasty they always say things about my teeth, I don’t really care’</i>	<i>Negative: ‘they would say “you’ve got stickers on your teeth”</i>	<i>Negative: ‘they say I’ve got bad teeth’</i>	None
Self-appraisal of teeth	<i>Negative: ‘Its annoying ‘cos I can’t brush them off’</i>	<i>OK: ‘Sort of whitey, creamy colour, around the marks a tanny, whitey colour’</i>	<i>Good: ‘My teeth are perfect’</i>	<i>Good: ‘I’ve got a few yellow bits but the rest are white, I think I have quite good teeth’</i>
Reflected appraisals about teeth	<i>Perceived others to think their teeth were ‘alright’</i>	<i>Perceived others appraisals to be part of teasing ‘they would say it to tease me, trying to wind me up’</i>	<i>Perceived others appraisals to be because: ‘they are jealous of me that I’ve got good teeth, I just ignore them’</i>	No data
Impact	<i>‘They don’t really bother me’</i>	<i>‘I’m not really that bothered if I haven’t got perfect teeth’</i>	<i>‘I hardly ever notice them’</i>	<i>‘I’ve noticed it but it doesn’t bother me ‘cos they are there & they are healthy’</i>

Photographs of teeth of young people described in Table 32

Michelle



Jane



Nick



Lucy



5.3.3.8 Age, gender and severity of marks

Age (van Palenstein Helderma and Mkasabuni 1993), gender (Astrom and Mashoto 2002) and severity of defect (Ellwood and O'Mullane 1995; Hawley *et al.* 1996) were also considered as possible explanations of the variation in impact. In this study, young people of all ages experienced the range of impacts. However, the transition to secondary school, an age-related life event appeared, for some, to be a period when marks became a concern. This might have been a function of the demands of establishing new social bonds and one's sense of self with a new social group, rather than because of age.

No links between either gender or the severity of defects and the degree of impact were evident (Table 31 and Table 32). For example, Julie's marks had little effect (for her upper incisors: DDEI = confluent/patchy covering between one-third and two-thirds of the surface, TFI = 5):

'I've got some marks on the side of these two teeth, but they don't bother me'
Julie 13 years

Photograph of Julie's anterior teeth



In contrast, Natasha (for her upper incisors: DDEI = confluent covering at least two-thirds of the surface, TFI = 4) was markedly affected by her teeth:

'I always wanted to change my teeth, they are like multi-coloured...I'm loads bothered' Natasha 14 years

Photograph of Natasha's anterior teeth



While it is not possible with qualitative data to test the associations between these variables and the degree of impact, the apparent lack of any relationships deserved to be highlighted. Overall, the important factor in whether marks had an impact on an individual was the process by which they derived their sense of self.

5.4 Discussion

This study used qualitative interviews to explore, in detail, the impact of marks on young people and variation in impact. This study elucidated the findings from the quantitative study, providing in-depth insights into the impact of marks.

Marks were found to feature in young people's interactions. Close friends provided advice and support about marks. When conflict arose between young people marks became a topic for teasing and when new young people were encountered, questions arose about marks. Marks had a range of impacts; young people were *'bothered'* to varying degrees. Neither age, gender, nor severity provided strong explanations for this variation, rather individuals for whom appearance was a defining characteristic of their sense of self and who relied on the reflected appraisals from others tended to be *'bothered'* by their marks. Each of these aspects of the results will be discussed, before more general discussions about the methodological considerations of the research are considered.

5.4.1 Impacts of marks

Marks had a range of impacts on a spectrum from *'pride'* in the colour of the teeth to *'not bothered'* to *'really quite bothered'*. Missing school to attend dental appointments was seen as a positive impact of having marks.

Those *'bothered'* by the marks expressed feelings of self-consciousness and not wanting to *'show'* their teeth when speaking or smiling. Self-consciousness has not previously been mentioned in relation to DDE in children, although a controlled study of adults with severe amelogenesis imperfecta found higher levels of self-consciousness in those with the condition (Coffield *et al.* 2005). Self-consciousness was included in this study based on previous studies of young people with craniofacial anomalies (Bjornsson and Agustsdottir 1987). A study exploring the relationship between dental aesthetics (using the aesthetic component of the Index of Orthodontic Treatment Need) and OHRQoL found tooth appearance to have a greater impact in individuals with high levels of self-consciousness (Klages *et al.* 2004). One implication of these observations is that dental professionals should be aware of the potential for their patients to be self-conscious and should approach discussions with them accordingly. There are also implications for item selection in quantitative OHRQoL research.

Other studies have had mixed results about the impact of DDE on smiling. In a study in Tanzania, most young people with fluorosis reported the way their teeth looked hindered them from smiling freely (van Palenstein Helderma and Mkasabuni 1993). Although in Malaysia, no differences were found between those with and without DDE in terms of covering their mouth when smiling (Sujak *et al.* 2004). These differences may be cultural, due to the wording of the questions and young people's interpretation of them or may reflect local norms. In this study, the use of the term *'showing your teeth'* was a recurrent finding, rather than young people describing an impact on smiling specifically, highlighting the importance of the subtleties of language use.

The impact of marks was described in terms of the degree to which young people were *'bothered'*. Participants did not express impact in terms of frequency. As stated in Chapter Four, in the item impact assessment to develop CPQ₁₁₋₁₄, the response format rated how much young people's oral condition bothered them. However, in the final

measure, the frequency of impact was assessed. Some quality of life measures for children use response options that include the degree to which children are bothered by their medical conditions (Christie *et al.* 1993; Juniper *et al.* 1998). In future, measures of the impact of oral conditions on children and young people could consider rating the degree to which participants are 'bothered', rather than expressing impact in terms of frequency.

Marks also had a positive impact via increased whiteness of teeth and time off school. Hawley and colleagues (1996) concluded that teeth with some milder forms of fluorosis may be more aesthetically pleasing to 14-year-olds than those without (Figure 1). Time off school was reported as an advantage of having marks. This finding compares with the results from the quantitative study described in Chapter Four, where the item on CPQ₁₁₋₁₄ about missing school was found to be of high importance to children attending for treatment of their marks. However, in CPQ₁₁₋₁₄ missing time off school is regarded as a negative impact, rather than the positive impact described here. This highlights one of the advantages of gaining young people's own perspectives directly, rather than relying on adults interpretations.

Another item from the quantitative study that required further exploration was sensitivity to hot and cold. This impact did not feature in the participants discourses even when enquired about directly. While this impact is biologically plausible, it was apparently not an impact of importance to these young people. Rather, the important impact of marks on young people was on social and psychological aspects of their lives. This finding has implications for the interpretation of quantitative data.

5.4.2 Influence of sense of self on the impacts of marks

The important factor in whether marks impacted on an individual was their sense of self. The data reflected the components of James' hypotheses on the self (James 1892) and Cooley's looking-glass self (Cooley 1902). These two processes have both been observed as important in the development of sense of self in young people (Harter 1986; Harter 1990; Harter *et al.* 1996). Neither James nor Cooley provided a definition of sense of self although they described how it develops from childhood as a function of social interactions. The interactionist view is that sense of self develops as young people observe the way others react to them and gradually become aware that others have different perspectives and become able to apply these perspectives to themselves

(Carver 2003). Little research about sense of self, or James' or Cooley's approaches to it, has been carried out in relation to oro-facial visible differences or aesthetic anomalies more generally. While other studies have identified self-image (Edwards *et al.* 2005) or social interactions (Beaune *et al.* 2003) as important for young people with craniofacial differences, without the appropriate theoretical framework, both the social and psychological aspects of the self were not discovered.

James hypothesised that an individual's sense of self is based on how adequately they perform in the domains of life that are important to them. Performance in unimportant areas has little impact on the self (James 1892). Marks appeared to impact on young people who desired a good appearance, whereas those for whom appearance was unimportant were apparently unaffected. Clinicians have anecdotally noted this individual variation in demand for treatment with this study confirming this phenomenon. The most important domains relevant to marks for these young people were appearance and personality. This corresponds with other research that has found appearance and popularity to be the most common domains of importance for young people (Rosenberg 1979; Harter 1985; Harter 1990).

Research of the applicability of the looking glass self in young people has also found differences in the extent to which an individual's sense of self is dependent on approval from others. Some individuals base their sense of self on approval from peers and experience fluctuations as peer approval fluctuates. These individuals are described as having a 'looking glass self orientation' (Harter *et al.* 1996). For others, sense of self does not rely on approval from others. The notion of the looking-glass self has also been applied to reflected appraisals of body appearance and image (Cash and Fleming 2002). Other research has shown that most young people experience short-term fluctuations of sense of self associated with major life events such as changes of schools and become temporarily more reliant on social feedback, until their sense of self in this new situation develops (Simmons 1979; Harter and Whitesell 2003).

In this study, marks had an impact on individuals' whose sense of self was defined by appearance and who depended on approval from others about their appearance. These young people saw the appearance of their teeth as a threat to their sense of self. Even though, in some cases, the defects on the teeth were normatively assessed as being of mild severity (Table 30). Perceived approval from peers was particularly important,

more so than approval of significant others such as close friends. This finding echoes other research with young people, where peers had more influence on sense of self than close friends (Harter 1989).

Sense of self includes both social and psychological components, yet research in this field is often restricted to narrower self-related concepts such as self-esteem, self-consciousness and self-confidence (Leary and Tangney 2005). The psychological aspects of the self have recently been suggested in relation to the impact of oral health conditions on children (Humphris *et al.* 2005; Locker *et al.* 2005; Gussy and Kilpatrick 2006). However, in addition to these psychological components of the self, young people of this age must also establish their own identity in terms of their roles within the groups of people around them and society more generally (Erikson 1959). Several attempts have been made to describe the progress of identity development throughout the teenage years to adulthood (Erikson 1959; Marcia 1966; Marcia 1980), although it is now acknowledged that the process of identity development varies between individuals and cultures (Bee and Boyd 2004). Young people's perceptions of the appearance of their own bodies and those of others are an important source of identity for some young people (James 1993). Further research, particularly longitudinal research into the contribution of the social and psychological components of the self to young people's experiences of marks, and oral health more generally, is required.

No links between gender, age, severity of marks and impact were apparent among these young people. In the past, gender differences between men and women on the need for approval about appearance have been cited, although this observation may have been due to the assumption that women are more concerned about their appearance because they discuss it more often than men (Rumsey 1997). Findings from the literature on the differences in impact of DDE on male or female young people are inconsistent (Astrom and Mashoto 2002; Sujak *et al.* 2004). Transition to secondary education, was the only age-related event that had any apparent relationship to impact. In terms of severity, a previous study in the UK with young people with DDE, found smaller defects to generally be more acceptable to young people than larger defects, but noted individual variation with some young people being unaware of what the researchers considered large or severe defects (Ellwood and O'Mullane 1995). Other studies have also found poor agreement between normative and children's assessment of the appearance and need for treatment of DDE (Milsom *et al.* 2000; Astrom and Mashoto 2002; Shulman *et*

al. 2004). Normative assessments of facial disfigurement are also poorly related to their impact on individuals (Thompson and Kent 2001).

The biopsychosocial model of health adopted in this thesis has provided a framework to ensure consideration of, not just, the impact of a condition on biological systems, but also on the social and psychological systems of the person and the world around them. Establishing the impact of DDE on the individual is an important criterion in contributing to discussions about whether DDE are a public health problem. The variation between individuals and lack of relationship with severity found in this study, have implications for discussions on the impact of fluorosis. In the York Review, fluorosis was considered an adverse effect of fluoridation and fluorosis of TFI ≥ 3 was classified as being of 'aesthetic concern'. This study provides some evidence that for some young people with TFI ≥ 3 , fluorosis is of no concern, but may be for others with lower TFI scores. Multifactorial research is needed to further investigate the relationship of biological variables and characteristics of the individual (such as sense of self) and their environment. As the Wilson and Cleary model, (1995) described in Chapter Two (section 2.5.2.1, Figure 4), operationalises the biopsychosocial approach to health, it could provide an appropriate theoretical model to guide this research. Seen in this model, sense of self is an individual factor that mediates the relationship between clinical factors and impact.

5.4.3 Social interactions

Young peoples' interactions involving marks included teasing and questions from people they met for the first time. Teasing was reported by the participants usually as a reaction to conflict between young people. Other researchers have reported teasing due to DDE (Welbury and Shaw 1990). The main topics reported in this study for teasing were features that singled an individual out as being different, usually their physical appearance. As DDE are an aspect of physical appearance that can make an individual different it is not surprising that participants reported being teased and called names just as they were about other aspects of their physical appearance.

The qualitative data echoed findings from the informal discussions in the quantitative study (section 4.3.2.3) which revealed subtleties in the meaning of name-calling for young people, including the use of (teeth-related) nicknames between close friends. The quantitative study also identified 'arguing with friends or family' as being of

greater impact in those with marks compared to those without. These observations suggest differences in meaning between the anti-social use of names during name-calling and arguing, compared to the pro-social use of names as nick-names. There are many definitions of teasing in the literature (Shapiro *et al.* 1991; Bollmer *et al.* 2003). One definition that recognises both the anti-social and pro-social aspects defines teasing as:

‘an intentional provocation accompanied by playful off-record markers that together comment on something relevant to the target’ (Keltner *et al.* 2001)

Children from the age of 8 years are able to discern the intended meaning of teasing. There is further consolidation of the concept of teasing by 11 or 12 years (Harter and Whitesell 1990). The most commonly reported forms of teasing in children and young people are name-calling and making fun of an attribute or behaviour; the most common topics being physical appearance and intellectual performance (Scambler *et al.* 1998). When examining the motives stated by children who teased, the most common causes were getting someone back for teasing them, to play or joke around, to indicate they disliked the target or to make themselves feel better when they were in a bad mood (Shapiro *et al.* 1991). Features of teeth were the fourth most common topic for teasing with 7% of children reporting being teased about their teeth once a week or more (Shaw *et al.* 1980). Other dental reasons for teasing reported in the literature are denture wearing (Rodd and Atkin 2000) and malocclusion (Helm *et al.* 1985; Onyeaso and Sanu 2005).

How interactions such as teasing and name-calling about marks were interpreted by individuals, again, appeared to be related to aspects of their sense of self, with variation in how these situations were defined. For those for whom appearance was important and contingent on approval from others, interactions about marks were defined as negative, in contrast to those for whom appearance was unimportant who were able to dismiss these interactions.

Young people reported being asked questions by others they were meeting for the first time. One such question was whether the appearance of the teeth was due to the individual neglecting to brush them. Again, this impact was observed in the informal discussions in section 4.3.2.3. Young people felt this topic was relevant, but it was absent from CPQ₁₁₋₁₄. Other studies have also reported children with fluorosis being accused of neglecting their teeth (Riordan 1993; Fantaye *et al.* 2003; Edwards *et al.*

2005), but have not specifically related this to the formation of new social bonds. The feature of interactions about marks during the transition to secondary education was another important finding and is relevant as young people may seek treatment around this time. Other authors have found a change in school to be a situation which threatens a child's sense of self. This may be explained by the disruptions in social networks and increased social comparisons that occur (Simmons 1979).

Social interactions have also featured heavily in other qualitative studies of the impact of cranio-facial conditions and oral health more generally on young people (Ostberg *et al.* 2002; Beaune *et al.* 2003; Fitzgerald *et al.* 2004). The impact of Treacher Collins syndrome on young people in Canada revealed three main themes of 'forming friendships and fitting in', 'handling staring and teasing' and 'balancing sameness and difference', which were all related to social interactions (Beaune *et al.* 2003). Two studies have explored young people's experiences of oral health. One, conducted in Sweden, found the impact of oral health was principally related to the affect of the appearance of teeth on social interactions. A second study from New Zealand found that while oral health was perceived to be largely irrelevant to the everyday lives of young people, the only exception was the importance of having attractive teeth when socialising (Fitzgerald *et al.* 2004).

In summary, DDE featured in young people's social interactions with differing interpretations of these interactions depending on the individual's sense of self. Other studies have also found the impact of oral and craniofacial conditions to be related to social interactions.

5.4.4 Methodological aspects of the study

Dimensions of quality in qualitative research include reflexivity and reliability and validity.

5.4.4.1 Reflexivity

A reflexive stance was taken both in terms of the involvement of young people in the research and the influence the researchers had on the research process.

5.4.4.1.1 Involvement of young people

Young people were actively involved in this study. It was the first study to explore young people's experiences of DDE in their own words, allowing their perspectives to

be listened to and heard directly. While previous researchers have anticipated some of the ways DDE would impact on young people's daily lives, other aspects, particularly the degree of variation between individuals, have not previously been described. It also allowed clarification of some of the items from the quantitative study that had been identified as being of importance.

Throughout this thesis the methodological considerations of power imbalance, language use, setting and analysis have been referred to as important aspects of child-centred research. The degree to which this study has adequately considered these factors will now be discussed.

5.4.4.1.1.1 Power imbalance

Research with children and young people often discusses the potential power imbalance between the participant and the adult researcher (Chapter Two, section 2.4.6.1) (Punch 2002). A number of authors have discussed the role an adult researcher should employ to alter the power relations with young people. Roles such as non-authoritarian adults, 'friends', 'least adults' and detached observers are recommended to provide the researcher with the opportunity to interact with children and young people (James *et al.* 1998; Davis *et al.* 2000; Woodhead and Faulkner 2000). In this study the role adopted was best described as 'least adult'. To prevent further complications arising from the researcher being a dentist and alteration of the power relation, the young people were not told of this during the interviews, unless they asked directly.

Power imbalance was also reduced by involving young people in piloting the topic guide and gaining their consent. They were also given a choice of when and where they wanted to be interviewed. Before each interview the process was explained fully with assurance of confidentiality. The interviews were kept informal through the use of humour. After the interviews the young people were de-briefed and thanked for the donation of their time and effort. Copies of the results were sent to them for feedback after the analysis. These methods have been recommended to reduce the power imbalance (O'Kane 2000). No participants took the opportunity to provide feedback; on reflection more active methods could have been employed to gain their perspectives on the analysis.

5.4.4.1.1.2 Language

As young peoples' language differs from that of adults, there was an awareness of the potential for misunderstandings in both directions; the participant misunderstanding the researcher and vice versa (Punch 2002). Several potentially problematic occasions arose. For example, one participant referred to herself as *'crafty'*. Her intended meaning of this expression was probed as the interviewer was unsure whether she meant she was good at crafts or devious.

Expressions such as being *'bothered'* and *'showing their teeth'* were used frequently and were obviously turns of phrase to which young people could relate to describe their experiences of such conditions. The terms used by young people to describe their DDE, *'marks'*, *'speckles'* and *'thingies'* were unlike those used by dental professionals, this also has relevance for clinicians in discussions with young people about the appearance of their teeth.

5.4.4.1.1.3 Setting

The home was chosen as the setting for the study despite schools or clinics being the places where children are studied most often. It was hoped that the more 'natural' environment would provide richer data and put the participants at ease without the inhibitive effect of the educational or clinical environment (Scott 2000). Privacy can be difficult to obtain in each of these settings. In this study, on several occasions, parents requested to be present during the interviews or siblings were within ear shot. Their proximity may have influenced the responses of some participants. This was recorded in the field notes with one particular instance when an interruption by a parent caused a participant to suddenly change the subject. After the parent had left the interview was steered around to this topic again to complete the in-depth probing.

5.4.4.1.1.4 Analysis

This study aimed to provide new insights into the impact of DDE on young people. Rather than claiming that the results are a representation of young people's views it is acknowledged that they are an adult researcher's presentation of the views of others from a different generation. The results attempted to remain as faithful to the participants experiences as possible, but inevitably they reflect the researcher's interpretation and chosen theoretical framework. The use of a theory was necessary to guide the study, but while this helped to make sense of the data it will also have shaped its construction (Mauthner and Doucet 2003).

Symbolic interactionism was selected as the theoretical framework for this study. The premise of this perspective is that the meaning individuals attribute to things are the products of social interactions. This framework was chosen as it was appropriate to young people and findings from previous studies. The results support its assumptions as interactions about marks were frequently reported by the young people and interpreted according to each individual's definition of the situation. Symbolic interactionism has already been applied to research of young people's experiences of chronic illness (Woodgate 1998), but not in relation to oral health.

In summary, young people were involved throughout the research process and this study would be classified as Category 1, research *with* children using the categories derived in the systematic review described in Chapter Three. In future, the child-centred research concept could be taken a stage further by involving children as researchers themselves and allowing them to shape research agendas (Alderson 2000).

5.4.4.1.2 Influences on the research process

Secondly, the influence of the sample and recruitment needs consideration. Ethical concerns inevitably dominate choices about the methods of research involving children and young people (Punch 2002) as was the case in this study. The participants' dentists acted as gate-keepers, first, to make sure that the research didn't obviously draw participants' attention to their marks for the first time and, secondly, so that if, as a result of the study, any participants wanted further discussions or treatment they would be available. This method did limit the sample to participants who visited a dentist, who were aware of their marks and who were willing to take part. Pressure on young people to participate in this study was minimised as, although the potential participants were approached by their dentist, they were given several weeks to decide to participate, and then, through contact with the researcher, rather than their dentist.

These sampling and recruitment techniques could have affected the results. However, a range of experiences were expressed by the participants. This wide coverage of the topic suggests the data are valid. The purposive sample, informed by a framework involving age, gender and severity of defect achieved saturation at 21 participants. Whilst participants were from a range of geographical areas across South Yorkshire, Lincolnshire and North Nottinghamshire they did not include any from minority ethnic groups. Although ethnicity may be a factor related to the impact of young people with

marks, the literature on the self from the United States suggests a complex relationship between self, gender, ethnicity and social class (Thompson and Keith 2001). These factors would also need to be included in future studies on the impact of DDE.

5.4.4.1.3 Reliability and validity

The properties of reliability and validity can be applied in a broad sense to qualitative research (Chapter Two, section 2.5.4.4) (Emden and Sandelowski 1998; Seale *et al.* 2004). To ensure reliability, the details of the study such as: sample selection (including the sample framework), data collection, method of analysis and support for interpretations (Seale *et al.* 2004) are provided in the description of the study. External validity was ensured through triangulation with the data from other research including the study described in Chapter Four, analysis by different researchers and continuous feedback. Participants were asked to comment on the interpretation of data from other interviews and were also sent a summary of the findings of the study for verification. Achieving internal validity involved checking the accuracy of fit of the explanations by comparison between different parts of the data (Silverman 2000). The use of constant comparative analysis (Glaser and Strauss 1967) enabled these comparisons.

Previously the quality of qualitative research in dentistry has been reported to be mediocre, particularly in terms of design, rigour, reflexivity and presentation of findings (Thaliath *et al.* 2006). Application of checklists such as the Critical Appraisal Skills Programme appraisal tool have a role in assessing quality, with an appraisal tool specifically for qualitative research (Public Health Resource Unit. 2002). This study satisfies the important criteria of this tool. For example full details are given of recruitment, data collection, the relationship between the researcher and the participants and the process of analysis.

5.4.4.1.3.1 Generalisability

Qualitative research findings are not statistically representative, but conceptually so. The findings of this study can be applied beyond this sample to other young people with DDE, but also to young people with other minor dental aesthetic conditions such as malocclusion or dental trauma. For these conditions it is anticipated that there will also be variation in impact between individuals related to aspects of the self. In addition, these data serve to support existing theories of sense of self in young people, particularly the concept of the looking-glass self.

In conclusion, having DDE resulted in a range of impacts on young people. Rather than these variations being related to age, gender or severity, they were related to defining aspects of sense of self.

5.5 Publications arising from the work in this chapter

i) Prizes

Z. Marshman, B.J. Gibson, H.D. Rodd, P.G. Robinson. Young people's "tips" to dentists. British Society of Paediatric Dentistry Conference, Cardiff, International Journal of Paediatric Dentistry 2005; 15: 395.

ii) Posters

Z. Marshman, B.J. Gibson, P.G. Robinson. Perceptions of young people with enamel defects. IADR Brisbane June 2006 Abstract No. 1203.

Z. Marshman, B.J. Gibson, P.G. Robinson. The impact of developmental defects of enamel on young people. World Congress on Preventive Dentistry, September 2005 Abstract No. 21.

6. Chapter Six

Discussion

6.1 Introduction

The public health significance of DDE has not been established, although several authors have considered this question (Burt and Eklund 1999; Martinez-Mier *et al.* 2004; Whelton *et al.* 2004; Cutress *et al.* 2006). These authors have all alluded to the aesthetics of the condition and its consequent impact. Sheiham and Watt (2003) suggested the following criteria for determining the public health significance of a health condition:

- Prevalence of the condition
- Impact of the condition on the individual
- Impact on wider society
- Condition is preventable and effective treatments are available

While there are abundant data on the prevalence of DDE, there is a paucity of research on the impact of DDE, with several authors recommending future research in this area (Martinez-Mier *et al.* 2004; Whelton *et al.* 2004). Indeed, the narrative review conducted in Chapter Two was unable to find any data from the UK on children and young people's perspectives on the impact of DDE.

This thesis forms an original addition to existing knowledge. First, the existing literature was reviewed systematically to ascertain the degree to which young people's views on DDE had been considered. On identifying that the impact of DDE had not been studied from young people's perspectives, the thesis reports the first study in the UK to describe the impact of DDE in comparison to relatively healthy young people. With the discovery that DDE did not cause a significant frequency of impacts, the final study was the first worldwide to explore the experiences of DDE in young people using qualitative methods. No other qualitative research in dentistry has explicitly adopted a symbolic interactionist approach. Furthermore, the identification of the importance of sense of self in mediating the relationship between clinical status and the impact of conditions is an addition to knowledge that may have implications beyond dentistry. This research was also novel in so far as it was the first in dentistry to use an array of child-centred techniques that reflect the state of the art from social sciences.

6.2 Summary of findings

The gap in the literature on the impact of DDE on individual children and young people identified in the narrative review was confirmed by the systematic review described in Chapter Three. This review established that, while some attempts had previously been made to gain children's perspectives about DDE, the vast majority of studies had not included their input. This systematic review found little evidence of consideration of the impact of DDE on affected individuals. This emphasis was characteristic of the general child oral health literature.

Both quantitative and qualitative methods were then chosen to investigate the impact of DDE on children and young people. The frequency of the impact of DDE on the OHRQoL of young people attending for treatment was found to be low and equivalent to that of young people with relative oral health. Several explanations were offered for this lack of difference, including the low impact of DDE, the power of the study, properties of the measure used and the possibility of mediating factors. Aspects of impact identified as being important to children with DDE were difficulty with hot or cold foods or drinks, concern what others thought of their teeth, arguing with friends and family and missing school.

The mixed-method approach allowed these findings to be explored qualitatively. Indeed this was the first study to take this approach to the study of DDE. The impact of DDE was found to vary markedly between young people. This variation was expressed as the degree to which DDE '*bothered*' them, rather than the frequency of impacts. Specific positive aspects of impact were pride in the appearance of teeth and missing time off school for appointments. Negative impacts were self-consciousness and not wanting to '*show*' their teeth during speaking or smiling. No mention was made in this study about difficulties with hot or cold foods/drinks, rather the emphasis was on psychological and social components.

The impact of DDE was apparently related to the defining aspects of young people's sense of self. DDE had an impact on individuals' whose sense of self was defined by appearance and who depended on approval from others about their appearance. This finding reflects James' hypotheses on the self (James 1892) and Cooley's concept of the

looking-glass self (Cooley 1902). Both processes have been identified as important in the development of sense of self in young people (Harter 1986; Harter 1990; Harter *et al.* 1996). Both were observed in the qualitative study and explained some of the variation in experience of DDE between young people. Sense of self has not previously been identified as an important factor in the impact of conditions that affect the appearance of young people.

Individual differences in sense of self also resulted in differing interpretations of social interactions, including teasing/name-calling and the use of nicknames. These different perceptions of interactions also emerged from the assessment of CPQ₁₁₋₁₄ (Chapter Four). Events such as changing schools and holidays were times when social interactions about DDE were stimulated. Neither gender, age nor severity of defects were apparently related to their impact.

While no research had previously been conducted in the UK on the impact of DDE on affected children and young people, several other studies have been conducted elsewhere. Those data suggest that severe fluorosis has a weak and inconsistent impact on some children and young people in other countries, particularly in areas of Africa. Overall, the studies contained within this thesis add to knowledge by explaining why that relationship is inconsistent. It is mediated by an individual factor (sense of self). Therefore, the presence of defects alone does not necessarily result in impact on children and young people, rather the relationship is more complex. These findings need to be supplemented with further quantitative research before the impact of DDE on individual children and young people in the UK can be fully described. Once the impact of DDE on the individual is understood, information on the impact of DDE on wider society is also needed before conclusions on the public health significance of DDE can be drawn.

6.3 Implications of the research for policies about DDE

The importance of establishing the impact of DDE has been flagged in the literature to inform debates about their public health significance. The public health importance of fluorosis, as one type of DDE, has received significant attention (Burt 1999; Martinez-Mier *et al.* 2004; Whelton *et al.* 2004). In 1999 Burt and colleagues asserted that fluorosis could not be classified as a public health problem in the US because most fluorosis was mild. However, they recognised that changes in the public's concern

about aesthetics could increase the potential for it to become a public health problem in the future (Burt and Eklund 1999). In Mexico, fluorosis has been deemed a possible public health problem due to investigations of the impact of the condition, using a rudimentary measure, and again, the importance of physical appearance was felt to be an important determining factor (Martinez-Mier *et al.* 2004). In Europe, fluorosis, at current levels of prevalence and severity, was not considered a public health problem, although more detailed consideration of the impact of fluorosis was recommended (Whelton *et al.* 2004).

The findings of the present study have implications for debates about fluorosis and the use of fluorides, particularly water fluoridation. Fluorosis is often cited as an adverse effect of fluoridation and, in the York Review, fluorosis of a TFI score of greater than or equal to three was classified as being of 'aesthetic concern' (McDonagh *et al.* 2000). First, these data add to knowledge in so far as the presence of fluoride-induced defects *per se* does not necessarily result in a negative impact. Secondly, for some young people with TFI greater than or equal to three, fluorosis is of no concern, but it may be a concern for others with lower TFI scores. The variation between individuals and the lack of relationship with severity found in this study, could contribute to risk-benefit assessments of new and existing fluoridation schemes. Indeed, the Medical Research Council Report on Water Fluoridation and Health recommended the need to communicate the degree of impact of fluorosis to the public to inform the public's perception of the risk of fluoridation (Medical Research Council Working Group Report 2002).

Chapter Two discussed policies such as those on the use of various sources of fluoride (section 2.4.4.2) and clinical guidelines on the treatment of DDE (section 2.4.4.3). The findings from this study have implications for policy development in future, with a need to incorporate the perspectives of children and young people with DDE. For example, the British Society of Paediatric Dentistry guidelines on the treatment of intrinsic discolouration of permanent teeth, which are currently under review, make little mention of asking patients for their own concerns and whether they are satisfied with the outcome of their treatment (Wray and Welbury 2001). Were such approaches to be included in future guidelines, treatment would be enhanced by making it more appropriate to the needs of affected children.

6.4 Implications of the research for clinical care of patients with DDE

The findings of the studies contained within this thesis have several implications for clinicians caring for individual children and young people with DDE. First, regarding communication, dental professionals need to be aware of the differences between the terms they use to describe DDE and those used by young people. Being aware of these differences in language and attempting to minimise them will improve clinicians' ability to access the true meaning of their patients' concerns. Also, when dealing with young people with DDE they need to be aware that young people may be self-conscious about their appearance. Furthermore, such young patients may be so self-conscious that they cannot express their concerns about their appearance. Consequently they may need to be offered the option of treatment sensitively. Anecdotally, clinicians have noted the variation between individuals in their demands for treatment. The findings of this research reveal that variations in individual characteristics such as sense of self provide some explanation for this.

Secondly, changes of school were apparently important life events for young people. They may therefore present around this time for treatment if they begin to become concerned about their appearance. Timely treatment of these cases before the start of their new school might ease the impact of DDE for some young people.

As reported earlier, qualitative approaches adopting a symbolic interactionism perspective have been little used in research of young people's experience of oral disease. Yet the approach yielded new knowledge about the impact of DDE and the meaning they have for the everyday lives of young people with the condition. It is possible that sense of self may mediate the impact of other clinical conditions that affect appearance. Future research should therefore use the methodological approaches employed in this study to investigate the impacts on young people of other dental and craniofacial anomalies and even conditions away from the head and neck.

6.5 Recommendations for research about DDE

While this thesis has advanced knowledge about the impact of DDE, further investigation is required. Once the impact on the individual has been established, research is also needed on the impact of DDE on wider society, another important criterion for establishing the public health significance of a condition on which little information is available.

One method to research the impact of DDE further would be to develop a condition-specific measure of the impact of DDE on the OHRQoL of children and young people. The qualitative study contained within this thesis could provide the basis for the development of such a measure with further involvement of children and young people throughout the process. Such methods were used to develop the Youth Quality of Life Instrument-Facial Difference module to ensure pertinence to young people with craniofacial differences (Edwards *et al.* 2005). The six domains perceived to be important and therefore included in this module were stigma and isolation, intimacy and trust, positive consequences, self-image, and negative emotions. Although this measure was developed for use with young people with craniofacial conditions, these domains concur with the main themes from the qualitative study in this thesis. The response format for the DDE-specific measure could include the degree to which participants were '*bothered*' by the defects, rather than the frequency of the impacts. Once developed and tested this measure could be used with children with DDE, including those children not seeking treatment. It could also be used to gain children's perspectives in clinical trials of treatment to improve the appearance of DDE.

However, the disadvantage of a condition-specific measure would be the inability to compare the impact of DDE with other diseases. The Medical Research Council Report recommended research on the impact of fluoridation on quality of life (Medical Research Council Working Group Report 2002), a study to satisfy this recommendation would require a comparison of the impact of fluorosis and caries, which would not be possible with a DDE-specific measure.

Findings from the quantitative and particularly the qualitative study provide evidence that the impact of DDE is related to social and psychological characteristics of the individual such as sense of self. While these two studies have generated a hypothesis about the importance of such individual factors on the impact of DDE, quantitative testing is recommended. The World Health Organisation's International Classification of Functioning, Disability and Health (Chapter Two, section 2.5.1.2) identifies personal factors as influences on the impact of health conditions on an individual, but fails to classify them further (World Health Organisation 2002). Wilson and Cleary (Chapter Two, section 2.5.2.1, Figure 4) include characteristics of the individual and the environment as potential predictors of the impact of clinical condition on HRQoL and

overall well-being. However, the influence of these factors and the nature of the relationships are poorly understood, although direct and indirect relationships within the model have been tested in xerostomia and pooled datasets of older adults (Baker *et al.* 2007; Baker *et al.* 2007). The Wilson and Cleary model appears to provide an appropriate theoretical framework to guide further research on the impact of DDE. Such research would involve selecting measures to represent the variables within the model and the use of statistical methods such as structural equation modelling to examine relationships between them. Impact could be measured using the DDE-specific OHRQoL measure; however, difficulties may arise in the choice of measures to represent individual characteristics which appear to involve both psychological and social components of the self. For example, there are as yet no measures of sense of self and while there are measures of self-esteem and self-concept, these concepts do not adequately include the social components of the self.

In addition to implications for policies, clinical care and research about the impact of DDE, the findings of these studies also have more general implications.

6.6 General implications

6.6.1 Implications for research with children

A theme running through this thesis was of research *with* children and the adoption of child-centred methods. Understanding of the impact of DDE was gained from the perspective of the young people themselves. This novel approach to oral health research yielded rich benefits. When young people were given a voice to express impact in their own words, unanticipated ideas emerged.

The feasibility of conducting dental research in young people's own homes was also established. The systematic review (Chapter Three) of child dental research identified little research on children and young people's perspectives on oral health generally. Child-centred research on the impact of other common conditions such as caries, malocclusion, trauma and treatment itself might also yield unexpected insights.

These conditions could also be studied quantitatively using the Wilson and Cleary model as a framework. Symbolic interactionism provided an appropriate theoretical

framework for guiding the qualitative study of the impact of DDE. It could also be used in similar studies of the impact of other dental or cranio-facial anomalies.

Other opportunities for research *with* children include evaluating the safety and effectiveness of treatments and also investigation of the impact of oral conditions over time as young people make the transition from primary to secondary school. To take the research *with* children approach further, children and young people could be more fully involved as researchers by allowing them to shape oral health research agendas (Alderson 2000).

6.6.2 Implications for research on OHRQoL with children

To ensure rigour, this thesis reported an evaluation of the properties of CPQ₁₁₋₁₄ and P-CPQ. CPQ₁₁₋₁₄ was evaluated for use in the UK and was found to have satisfactory reliability, criterion validity and construct validity in relation to ratings of affect on life overall. Construct validity in terms of clinical measures was inconsistent. Some concerns, particularly about the content and face validity of the measure were identified. The reliability and validity of P-CPQ was acceptable for use in the UK.

CPQ₁₁₋₁₄ is a generic OHRQoL measure for use with children, yet some items were perceived not to be relevant to children with DDE, a known disadvantage of generic measures (Guyatt *et al.* 1993). The measure was designed by a method recommended to ensure it contained the most relevant aspects for children (Guyatt *et al.* 1986). However, the response format for the item impact study was different to that used in the final questionnaire and the input from the children on the wording of the items is unknown. The content validity for the use of CPQ₁₁₋₁₄ with other specific conditions requires further testing. Evaluation of the face validity is also needed to investigate the meaning of the measure to participants. An area of particular interest would be the appropriateness of a scoring system based on frequency. Such a study could provide recommendations for amendments to enhance CPQ₁₁₋₁₄ and prevent the problems encountered in this study being perpetuated. A suitable qualitative method using framework analysis has already been used to test a generic HRQoL measure (Mallinson 2002). This qualitative method could be used to evaluate the content and face validity of CPQ₁₁₋₁₄ and also the global ratings of oral health in children and young people.

The discriminative properties of CPQ₁₁₋₁₄ were also questioned. Such properties may have been over-emphasised in the original study to evaluate the measure as the sample included children with much higher levels of caries than in the UK. Consequently, further testing of CPQ₁₁₋₁₄ would be needed and population-based studies would require very large sample sizes.

In terms of construct validity, weak relationships were found between clinical and OHRQoL measures. Other studies have also found tenuous links between clinical assessment and OHRQoL (Cushing *et al.* 1986; Locker and Slade 1994; Gregory *et al.* 2005). This finding has implications for oral health needs assessments that rely on normative assessments of need to plan services (Sheiham *et al.* 1982). A weak relationship was found between clinical assessment of malocclusion and impact, an orthodontic needs assessment found significant numbers of young people who had a normative need (assessed using the Index of Orthodontic Treatment Need), but registered no impacts on OHRQoL (de Oliveira and Sheiham 2003).

The use of oral health needs assessments to ascertain the likely impact of a condition on wider society was discussed in Chapter Two. The findings from this study support the need for supplementation of clinical data with assessment of the impact of conditions.

In conclusion, the studies contained within this thesis have added to knowledge about the public health importance of DDE. Little research about DDE had previously included young people's perspectives. The impact of DDE was generally of low frequency with marked variation in impact between individuals. This research is the first to discover that some of this variation can be accounted for by young people's sense of self. This concept has not been identified in relation to the impact of visibly different conditions before. More research is needed to further investigate the impact of DDE.

7. Chapter Seven

Conclusions and Recommendations

In order to ascertain whether DDE constitute a public health problem in the UK the studies within this thesis aimed to describe the impact of DDE on the lives of affected young people. The objectives were to:

- 1) Describe the extent to which contemporary dental research on DDE had included the perspective of individual children and young people.
- 2) Describe the impact of DDE on the OHRQoL of young people.
- 3) Explore, in detail, the impact of DDE on young people.

An innovative child-centred approach was taken to achieve these objectives and the research reported in this thesis forms an addition to knowledge as there were few data on the impact of DDE on individual children and young people. This chapter summarises the findings and recommendations arising from the research.

7.1 Summary of findings

- Very little research about DDE had attempted to capture the subjective experience of those with the condition. This finding was mirrored by the child dental literature more generally.
- The frequency of the impact on the OHRQoL of young people attending for treatment of DDE was generally low and was equivalent to that of young people with relative oral health. Several explanations for this finding were discussed.
- The impact of DDE varied markedly between young people with the main areas of impact important to young people being social and psychological aspects.
- The impact of DDE was apparently related to the defining aspects of young people's sense of self.
- DDE had an impact on individuals' whose sense of self was defined by appearance and who depended on perceived approval from others about their appearance.
- Young people whose sense of self was defined by personality did not experience an impact.
- The concept of sense of self has not previously been applied to the impact of conditions on young people.

- No links between gender, age, severity of DDE and impact were apparent.

7.2 Recommendations for policies about DDE

- Fluorosis, as a type of DDE, has received significant attention, particularly as a potential adverse effect of fluoridation. These findings call into question whether fluorosis is an adverse effect for all individuals with the condition.
- This information could contribute to risk-benefit assessments of new and existing fluoridation schemes, particularly the finding that the presence of fluoride-induced defects does not inevitably have a negative impact.
- Policies such as those on the use of various sources of fluoride and clinical guidelines on the treatment of DDE should be developed to include the perspectives of children and young people.

7.3 Recommendations for clinical care of patients with DDE

- Dental professionals need to be aware when treating young people with DDE of the different terms used and the potential for self-consciousness about appearance and approach this subject sensitively.
- Variations in sense of self provide some explanation for variation between individuals in their demands for treatment.
- Changing schools was a time when some young people began experiencing some concerns about their DDE and may lead them to seek treatment. This could also be the case for other dental or cranio-facial irregularities that affect appearance.

7.4 Recommendations for research about DDE

- Multifactorial, longitudinal research is required on the relationship between the impact of DDE and characteristics of the individual (including sense of self) and the environment.
- The Wilson and Cleary model could provide an appropriate theoretical framework to guide this study.
- Impact could be measured using a condition-specific OHRQoL measure for DDE, developed based on the findings from the quantitative and qualitative studies, with involvement of children and young people throughout the process.

- Future research on the effectiveness of treatments of DDE should include the patient's perspectives, if a DDE-specific OHRQoL measure was developed this could be used as an outcome measure in clinical trials.
- Once the impact of DDE on the individual has been established, further research is also needed on the impact of DDE on wider society, another important criterion for establishing the public health significance of a condition.

7.5 Recommendations for research with children

- Research *with* children would allow children and young people's perspectives on other oral and craniofacial conditions to be explored
- The impact of other common conditions such as caries, malocclusion and trauma could also be studied using the Wilson and Cleary model as a framework.
- The impact of oral conditions on young people making the educational transition from primary to secondary school also requires longitudinal research and should include an assessment of sense of self throughout this life event.
- Qualitative research, in particularly the symbolic interactionism approach could also be used in similar studies of the impact of other dental and cranio-facial anomalies

7.6 Recommendations for research on OHRQoL with children

- Before CPQ₁₁₋₁₄ is used more widely some of its properties should be re-evaluated.
- Qualitative testing of the face validity of CPQ₁₁₋₁₄ is required and, if necessary, modifications to the measure should be made.
- Studies using CPQ₁₁₋₁₄ for specific conditions need to assess the content validity of the measure for these conditions.
- Research involving a comparison of the impact on OHRQoL of fluorosis with the impact of caries would require further testing of the discriminative properties of CPQ₁₁₋₁₄ and the study would require a very large sample size.
- More generally, the response scoring system for measures of the impact of oral conditions on OHRQoL could include the degree to which young people are '*bothered*' rather than the frequency of impacts.
- Further testing of the meaning of global ratings of oral health in children and young people is required

In conclusion, the studies contained within this thesis have found the frequency of the impacts of DDE to be generally low, with marked variation between individuals. Variation was apparently related to the defining aspects of young people's sense of self. More research is needed to investigate the impact of DDE further.

Publications and presentation arising from the work in this thesis

This research has also been reported in the following peer-reviewed research papers (Appendix G):

Z. Marshman, P.G. Robinson. Child and Adolescent Oral Health-Related Quality of Life. *Seminars in Orthodontics* 2007; 13: 75-126

Z. Marshman, B.J. Gibson, J. Owens, H.D. Rodd, H. Mazey, S.R. Baker, P.E. Benson, P.G. Robinson. 'Seen but not heard'. A systematic review of the place of the child in 21st century dental research. *International Journal of Paediatric Dentistry*. 2007; 17: 320-327

Z. Marshman, H.D. Rodd, M. Stern, C. Mitchell, P.G. Robinson. Evaluation of the Parental Perceptions Questionnaire, a component of the COHQoL, for use in the UK. *Community Dental Health*-in press, accepted November 2005

Z. Marshman, H.D. Rodd, M. Stern, C. Mitchell, D. Locker, A. Jokovic, P.G. Robinson. An evaluation of the Child Perceptions Questionnaire in the UK. *Community Dental Health* 2005; 22: 151-155

The research was also reported at several scientific conferences:

Z. Marshman, B.J. Gibson, J. Owens, H.D. Rodd, H. Mazey, S.R. Baker, P. E. Benson, P. G. Robinson. A systematic review of child oral health research 2000–2005. *International Association of Paediatric Dentistry Congress*. *International Journal of Paediatric Dentistry* 2007; 17: 320-327

Z. Marshman, B.J. Gibson, P.G. Robinson. Perceptions of young people with developmental defects of enamel. *IADR Brisbane June 2006*; Abstract No. 1203

Z. Marshman, B.J. Gibson, P.G. Robinson. The impact of developmental defects of enamel on young people. *World Congress on Preventive Dentistry, September 2005*; Abstract No. 21

Z. Marshman, H. Rodd, M. Stern, C. Mitchell, P.G. Robinson. Treatment of 'Don't know' responses in a parental Oral Health Related Quality of Life questionnaire. British Society of Dental Research Conference, Birmingham, April 2004

Z. Marshman, H. Rodd, M. Stern, C. Mitchell, D. Locker, A. Jokovic, P.G. Robinson
An evaluation of the Child Perception Questionnaire in the UK. International Association of Dental Research Conference, Honolulu, March 2004

The British Society of Paediatric Dentistry Young Researcher Prize was won for:

Z. Marshman, B.J. Gibson, HD Rodd, PG Robinson. Young people's "tips" to dentists. British Society of Paediatric Dentistry Conference, Cardiff, International Journal of Paediatric Dentistry 2005; 15: 395

8. Chapter Eight

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9. Chapter Nine

Appendices

9.1 Appendix A List of dental journals included in systematic review

Acta Odontologica Scandinavica

Acta Odontologica Latinoamericana

Advances in Dental Research

American Dental Association. Journal

American Journal of Dentistry

American Journal of Orthodontics and Dentofacial Orthopedics

Angle Orthodontist

Annals of Periodontology

Annals of Australasian College of Dental Surgeon

Archives of Oral Biology

Asian Journal of Oral and Maxillofacial Surgery

Atlas of Oral and Maxillofacial Clinics of North America

Australian Dental Journal

Australian Endodontic Journal

Australian Orthodontic Journal

Biological Therapies in Dentistry

Brazilian Dental Journal

British Dental Journal

British Journal of Oral & Maxillofacial Surgery

Bulletin of Dental Education

Bulletin of Toyko Dental College

California Dental Association. Journal

Caries Research

Cleft Palate-Craniofacial Journal

Clinical Journal of Dental Research

Clinical Oral Implants Research

Clinical Oral Investigations

Community Dental Health

Community Dentistry and Oral Epidemiology

Compendium of Continuing Education in Dentistry
Cranio: the Journal of Craniomandibular Practice
Critical Reviews in Oral Biology and Medicine
Dental Clinics of North America
Dental Economics
Dental Health
Dental Materials
Dental Materials Journal
Dental Traumatology
Dentistry Today
Dentomaxillofacial Radiology
Endodontic and Dental Traumatology
European Journal of Dental Education
European Journal of Oral Sciences
European Journal of Orthodontics
European Journal of Paediatric Dentistry
European Journal of Prosthodontics and Restorative Dentistry
Evidence-Based Dentistry
Functional Orthodontics
General Dentistry
Hawaii Dental Journal
Head and Neck
Implant Dentistry
Indian Journal of Dental Research
International Dental Journal
International Endodontic Journal
International Journal of Dental Hygiene
International Journal of Oral and Maxillofacial Implants
International Journal of Oral and Maxillofacial Surgery
International Journal of Oral Biology
International Journal of Orofacial Mycology
International Journal of Orthodontics
International Journal of Paediatric Dentistry
International Journal of Periodontics and Restorative Dentistry
International Journal of Prosthodontics

Iowa Dental Journal
Irish Dental Association. Journal
Japanese Journal of Pediatric Dentistry
Journal of Applied Oral Science
Journal of American College of Dentists
Journal Canadian Dental Association
Journal of Clinical Dentistry
Journal of Clinical Orthodontics
Journal of Clinical Pediatric Dentistry
Journal of Clinical Periodontology
Journal of Contemporary Dental Practice
Journal of Craniofacial Surgery
Journal of Cranio-Maxillofacial Surgery
Journal of Dental Education
Journal of Dental Hygiene
Journal of Dental Research
Journal of Dentistry
Journal of Dentistry for Children
Journal of Endodontics
Journal of Esthetic and Restorative Dentistry
Journal of Forensic Odonto-Stomatology
Journal of General Orthodontics
Journal of Indian Society of Pedodontics and Preventive Dentistry
Journal of Indiana Dental Association
Journal of International Academy of Periodontology
Journal of Oklahoma Dental Association
Journal of Oral and Maxillofacial Surgery
Journal of Oral Implantology
Journal of Oral Pathology & Medicine
Journal of Oral Rehabilitation
Journal of Orofacial Orthopedics
Journal of Orofacial Pain
Journal of Oral Science
Journal of Orthodontics
Journal of Periodontal Research

Journal of Periodontology
Journal of Prosthetic Dentistry
Journal of Prosthodontics: Implant, Esthetic, and Reconstructive Dentistry
Journal of Public Health Dentistry
Journal of the History of Dentistry
Journal of the South African Dental Association
Massachusetts Dental Society. Journal
Medicina Oral
Michigan Dental Association. Journal
New Jersey Dental Association. Journal
New York State Dental Journal
New Zealand Dental Journal
Nihon University Journal of Oral science
Northwest Dentistry
Odontology
Odonto-Stomatologie Tropicale
Ontario Dentist
Operative Dentistry
Oral and Maxillofacial Surgery Clinics of North America
Oral Diseases
Oral Microbiology and Immunology
Oral Oncology
Oral Radiology
Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology and Endodontics
Orthodontics and Craniofacial Research
Pediatric Dental Journal: International Journal of Japanese Society of Pediatric
Dentistry
Pediatric Dentistry
Pennsylvania Dental Journal
Periodontal Abstracts
Periodontal Clinical Investigations
Periodontology
Periodontology 2000
Practical Periodontics and Aesthetic Dentistry
Primary Dental Care

Quintessence International
South Africa Dental Journal
Seminars in Orthodontics
Singapore Dental Journal
Special Care in Dentistry
Swedish Dental Journal
Tennessee Dental Association. Journal
Texas Dental Journal
TMJ Update
Tsurumi University Dental Journal
World Journal of Orthodontics

9.2 Appendix B Child Perceptions Questionnaire for 11-14 year olds

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, lips, mouth or jaws?”**
- Put an in the box for the answer that is best for you

Today's date: / /
DAY MONTH YEAR

<p style="text-align: center;">FIRST, A FEW QUESTIONS ABOUT YOU</p>
--

1. Are you a boy or a girl?

Boy

Girl

2. When were you born? / /
DAY MONTH YEAR

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

Excellent

Very good

Good

Fair

Poor

4. 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

QUESTIONS ABOUT ORAL PROBLEMS

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

5. Pain in your teeth, lips, jaws or mouth?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

6. Bleeding gums?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

7. Sores in your mouth?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

8. Bad breath?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

9. Food stuck in or between your teeth?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

10. Food stuck in the top of your mouth?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

For the next questions...

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

In the past 3 months, how often have you:

11. Breathed through your mouth?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

12. Taken longer than others to eat a meal?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

13. Had trouble sleeping?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

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

14. 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

15. Difficult to open your mouth wide?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

16. Difficult to say any words?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

17. Difficult to eat foods you would like to eat?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

18. Difficult to drink with a straw?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

19. 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, lips, jaws or mouth?
If you felt this way for another reason, answer 'Never'.*

In the past 3 months, how often have you:

20. Felt irritable or frustrated?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

21. Felt unsure of yourself?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

22. Felt shy or embarrassed?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

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

23. Been concerned what other people think about your teeth, lips, mouth or jaws?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

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

Never

Once or twice

Sometimes

Often

Everyday or almost every day

25. Been upset?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

26. Felt nervous or afraid?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

27. Worried that you are not as healthy as others?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

28. Worried that you are different than other people?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

QUESTIONS ABOUT SCHOOL

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

In the past 3 months, how often have you:

29. Missed school because of pain, appointments, or surgery?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

30. Had a hard time paying attention in school?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

31. Had difficulty doing your homework?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

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

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, lips, jaws or mouth? If it was for another reason, answer 'Never'.

In the past 3 months, how often have you:

33. Avoided taking part in activities like sports, clubs, drama, music, school trips?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

34. Not wanted to talk to other children?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

35. Avoided smiling or laughing when around other children?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

36. Had difficulty playing a musical instrument such as a recorder, flute, clarinet, trumpet?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

37. Not wanted to spend time with other children?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

38. 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, lips, mouth or jaws,
how often have:**

39. Other children teased you or called you names?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

40. Other children made you feel left out?

Never

Once or twice

Sometimes

Often

Everyday or almost every day

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

Never

Once or twice

Sometimes

Often

Everyday or almost every day

NEARLY FINISHED!

THE LAST PAGE

- 42 We would like to know whether the way young people think about their mouths is affected by their background and culture.**

Please tick the box that best describes your ethnic group

Bangladeshi
Black African
Black British
Black Caribbean
Black other
Chinese
Indian
Pakistani
Vietnamese
White English, Scottish, Welsh, Northern Ireland
White Irish
Other (Please state)

THE LAST QUESTION!

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

Would you be willing to help us by completing another copy of the questionnaire soon? We would post it to you in the next 2 weeks.

YES

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

THANK YOU FOR HELPING US

9.3 Appendix C Clinical Data Collection Sheet

COHQoL DATA FORM

1. Date of examination (DDMMYY)
2. Hospital Number
3. Examiner HR 1 MS 2 CM 3
4. Surname
5. First Name
6. Date of birth
7. Age (numerical)
8. Gender Male 0 Female 1
9. Post code
10. IOTN – dental health component
11. Gingival health 0 = Good 1 = Gingivitis
12. Enamel defects

8	7	6	5	4	3	2	1	1	2	3	4	5	6	7	8
8	7	6	5	4	3	2	1	1	2	3	4	5	6	7	8

Modified DDE codes for scoring

	Codes for scoring
Normal	0
Demarcated opacities	1
Diffuse opacities	2
Hypoplasia	3
Other defects	4

13. Dental caries

8	7	6	5	4	3	2	1	1	2	3	4	5	6	7	8
8	7	6	5	4	3	2	1	1	2	3	4	5	6	7	8

BASCD criteria codes for scoring

	Codes for scoring
Present and sounds	0
Arrested dentinal decay	1
Decay	2
Decay with pulpal involvement	3
Filled and decayed	4
Filled with no decay	5
Extracted due to caries	6

14. **Other dental findings** of note (Details overleaf)

9.4 Appendix D Evaluation of P-CPQ using different approaches to the analysis of ‘don’t know’ responses

Introduction

P-CPQ is a measure of parental-caregiver perceptions of the OHRQoL of children. P-CPQ was designed to supplement the information obtained from children. P-CPQ includes 31 questions covering the same four domains as CPQ₁₁₋₁₄ with 14 additional questions on the impact on the family, the Family Impact Scale (FIS) (Jokovic et al. 2003). The participants are asked to indicate, using a six-point Likert scale (‘never’=0, ‘once or twice’=1, ‘sometimes’=2, ‘often’=3, ‘everyday or almost everyday’=4, and ‘don’t know’), the frequency at which the events have affected their child in the past three months. The P-CPQ contains a ‘don’t know’ response because the authors were aware of the limited knowledge a parent may have of their child’s activities and feelings. ‘Don’t know’ response categories have been used in other questionnaires to reassure respondents that it is acceptable not to know the answer, as well as to minimise guessing (Bowling 1997). P-CPQ also includes the global oral health and life overall ratings, although these ratings do not include a ‘don’t know’ response.

P-CPQ was evaluated in Canada in a similar way to CPQ₁₁₋₁₄ (Jokovic et al. 2003).

It had good internal consistency for the total score (Cronbach’s alpha = 0.94) and for the sub-scales. The test-retest reliability indicated excellent agreement (ICC = 0.85). P-CPQ showed good construct validity, with significant associations between the total scores and global ratings of oral health and life overall. Only questionnaires with zero ‘don’t know’ responses were included in the analyses. However, when different methods for handling ‘don’t knows’ were investigated, the properties were not affected (Jokovic et al. 2004).

The aim of this study was to investigate whether different approaches to handling ‘don’t know’ responses affected the properties of P-CPQ for use in the UK.

Method

The evaluations of P-CPQ and CPQ₁₁₋₁₄ were carried out at the same time with a clinic-based sample of children and their parents (section 4.2.1.1). This sample included parents

of children attending for an examination at the orthodontic or paediatric dentistry clinics of Charles Clifford Dental Hospital, Sheffield or a Sheffield General Dental Practice. Data were collected by asking parents to complete the questionnaire at the time of their visits and a retest questionnaire two weeks later.

Clinical data were collected by examination of the young people for the presence of DDE, caries status, malocclusion and gingival health.

Data analysis

Where participants failed to indicate a score for an item, missing values were dealt with in two ways: those participants who failed to complete more than one-seventh of the questions were excluded from the analysis. A similar threshold for excluding missing values has been adopted in other OHRQoL research (Slade 1997a). Missing values from the remaining participants were replaced with the sample mean score for that item.

Four approaches to analysis of 'don't know' responses were assessed to see whether they affected the properties of the measure:

- Approach 1 Exclusion - only the data from the parents who had not used the 'don't know' response were analysed.
- Approach 2 Item mean - replacing 'don't know' responses with the item mean for the entire sample.
- Approach 3 Mean items answered - imputation of the mean score for the items answered.
- Approach 4 Replacement - replacing 'don't know' responses with zero value.

These four approaches were used in the original evaluation study of P-CPQ for use in Canada (Jokovic *et al.* 2004).

After taking account of missing or 'don't know' responses, the total score for each participant was calculated by summing the item codes. A second summary measure for each participant, recorded the number of impacts reported 'often' or 'everyday or almost everyday'. Subscale scores were calculated by summing the codes for questions within the

four domains and the FIS scores were calculated by summing the responses to these 14 questions.

Internal consistency was assessed by means of Cronbach's alpha and test-retest reliability by means of intraclass correlation coefficient (ICC). The latter was based on data from those who participated in the follow-up study and who did not report that the oral health of their child and/or its impact on them had changed between the two administrations of the questionnaire.

Construct validity was assessed by testing associations of the scale, the subscale scores and the FIS with the life overall scores and the clinical data. Criterion validity was examined by comparing the global rating of oral health to scale scores and FIS scores.

Results

P-CPQ scores

The total score, subscales of P-CPQ and FIS, using the four different approaches to analysis of 'don't know' responses, are summarised in Table 1. Of the subscales, the highest mean scores were in the symptoms domain.

Table 1. Mean scores for the subscales of P-CPQ and FIS using different approaches to ‘don’t know’ responses

	Exclusion Mean (SD)	Item mean Mean (SD)	Mean items answered Mean (SD)	Replacement Mean (SD)
P-CPQ	13.5 (12.5)	14.2 (14.5)	14.3 (14.5)	14.2 (14.5)
Symptoms	3.9 (3.1)	4.1 (3.2)	4.1 (3.3)	4.1 (3.3)
Function	2.8 (3.7)	2.8 (3.4)	2.8 (3.4)	2.8 (3.4)
Emotion	2.4 (3.3)	2.5 (3.6)	2.5 (3.6)	2.9 (4.1)
Social	1.3 (2.0)	1.5 (2.7)	1.8 (3.0)	1.7 (3.0)
FIS	2.9 (3.8)	3.3 (4.6)	2.7 (4.1)	2.7 (4.1)

Reliability using different approaches to ‘don’t know’ responses

The internal consistencies of the total scale derived by each method were acceptable, the exclusion and item mean approach had reliability of less than 0.60 for one subscale each (Table 2).

Table 2. Reliability of P-CPQ, subscales and FIS using different approaches to ‘don’t know’ responses

	No. of items	Exclusion α	Item mean α	Mean items answered α	Replacement α
P-CPQ	45	0.86	0.89	0.92	0.93
Symptoms	6	0.68	0.65	0.71	0.69
Function	8	0.72	0.52	0.67	0.68
Emotion	7	0.85	0.83	0.85	0.85
Social	10	0.41	0.77	0.79	0.79
FIS	14	0.77	0.87	0.82	0.82

For the exclusion approach to ‘don’t know’ responses, 56% parents reported their child’s oral health to be unchanged and the ICC was 0.69. In the adjusted approaches, 52% of parents could be included in the test-retest analysis, with an ICC of 0.92-0.95.

Validity

Construct validity using different approaches to ‘don’t know’ responses

Construct validity varied between the exclusion and adjusted approaches (Tables 3-6). For the ‘don’t know’ exclusion approach, ratings of life overall were related to both summary measures of P-CPQ, the emotional and social subscales and the FIS. The number of decayed teeth and the functional subscale of the P-CPQ were associated, but no other relationships were apparent between the P-CPQ and clinical variables.

With the item mean approach, global ratings of life overall were related to all P-CPQ measures except the functional subscale. Significant correlations were found between the parent/carer reports of children with impacts ‘often’ or ‘everyday’ and the number of decayed teeth and the DMFT.

Similarly, the mean items answered approach showed global ratings of life overall to be related to all P-CPQ measures, except the functional subscale. With this third approach, significant correlations were found between the number of children with impacts ‘often’ or ‘everyday’ and the DMFT and also between the emotional subscale and the number of decayed teeth and the DMFT.

When ‘don’t know’ responses were replaced by zero, global ratings of life overall were related to all P-CPQ measures. A significant correlation was found between the number of children with impacts ‘often’ or ‘everyday’ and the number of DMFT.

Table 3. Relationship between life overall ratings, clinical data and P-CPQ scores using exclusion approach

	Total P-CPQ	Often or everyday	Symptom	Function	Emotion	Social	FIS
	Spearman's rank correlation coefficient						
Life overall	0.33*	0.26*	0.06	0.13	0.30*	0.30*	0.41*
IOTN	0.21	0.08	0.18	0.08	0.21	0.18	0.23
Decayed	0.11	0.01	0.23	0.32*	0.07	0.00	0.10
DMFT	0.06	0.14	0.11	0.20	0.02	0.02	0.03
	p-values						
DDE present	0.9	0.51	0.42	0.98	0.83	0.89	0.90
Gingivitis present	0.73	0.97	0.82	0.96	0.76	0.56	0.57

Table 4. Relationship between life overall ratings, clinical data and P-CPQ scores using item mean approach

	Total P-CPQ	Often or everyday	Symptom	Function	Emotion	Social	FIS
	Spearman's rank correlation coefficient						
Life overall	0.40**	0.30**	0.33**	0.20	0.41**	0.31*	0.42**
IOTN	0.12	0.02	0.09	0.03	0.15	0.07	0.08
Decayed	0.08	0.21*	0.01	0.03	0.20	0.14	0.09
DMFT	0.15	0.28**	0.07	0.20	0.22*	0.14	0.16
	p-values						
DDE present	0.33	0.20	0.30	0.53	0.94	0.59	0.57
Gingivitis present	0.53	0.57	0.53	0.46	0.40	0.99	0.61

Table 5. Relationship between life overall ratings, clinical data and P-CPQ scores using mean items answered approach

	Total P-CPQ	Often or everyday	Symptom	Function	Emotion	Social	FIS
	Spearman's rank correlation coefficient						
Life overall	0.40**	0.27**	0.34**	0.20	0.41**	0.36**	0.41**
IOTN	0.12	0.01	0.09	0.03	0.15	0.09	0.70
Decayed	0.08	0.17	0.02	0.15	0.22*	0.08	0.15
DMFT	0.14	0.25*	0.08	0.03	0.22*	0.09	0.19
	p-values						
DDE present	0.25	0.11	0.22	0.31	0.86	0.51	0.40
Gingivitis present	0.45	0.74	0.49	0.37	0.51	0.85	0.55

Table 6. Relationship between life overall ratings, clinical data and P-CPQ scores using replacement approach

	Total P-CPQ	Often or everyday	Symptom	Function	Emotion	Social	FIS
	Spearman's rank correlation coefficient						
Life overall	0.40**	0.27*	0.34**	0.22*	0.41**	0.35**	0.41**
IOTN	0.11	0.01	0.09	0.02	0.15	0.07	0.06
Decayed	0.09	0.17	0.01	0.13	0.18	0.11	0.15
DMFT	0.15	0.25*	0.08	0.01	0.20	0.14	0.19
	p-values						
DDE present	0.25	0.11	0.22	0.31	0.86	0.51	0.40
Gingivitis present	0.45	0.74	0.49	0.37	0.51	0.85	0.55

*=statistically significant, $p < 0.05$

**statistically significant, $p < 0.01$
 p-values obtained from Mann Whitney U test

Criterion validity using different approaches to ‘don’t know’ responses

Criterion validity was examined by comparing P-CPQ scores and the global oral health rating (Table 7). In the ‘don’t know’ exclusion analysis, the number of children with impacts ‘often’ or ‘everyday’ and the FIS score were associated with the global rating. In the three ‘don’t know’ adjusted approaches, correlation between the summary measures and subscale scores and the global oral health rating were found, but varied between approaches.

Table 7. Rank correlations between P-CPQ scores & global measure of oral health

	Exclusion r_s	Item mean r_s	Mean items answered r_s	Replacement r_s
P-CPQ	0.23	0.25*	0.24*	0.23*
No. often or everyday	0.31*	0.20	0.25*	0.25*
Symptoms	0.20	0.24*	0.30**	0.30**
Function	0.21	0.26*	0.18	0.18
Emotion	0.02	0.06	0.07	0.09
Social	0.07	0.09	0.11	0.10
FIS	0.27*	0.27*	0.27*	0.21*

*=, $p < 0.05$

**= $p < 0.01$

Conclusion

Unlike in the Canadian study (Jokovic et al. 2004), the properties of P-CPQ were affected when different approaches to handling ‘don’t know’ responses were taken. The results indicate that their inclusion, handled by adjustment, render the reliability and validity of this measure acceptable for use in the UK. As there were only minor differences between the adjusted approaches with respect to validity, from the perspective of reliability and validity, the replacement approach may be marginally the most appropriate method. This method involves replacing ‘don’t know’ responses with a zero value and had the highest internal consistency.

9.5 Appendix E Information sheets and consent forms for Chapter Four

Information on the research project:

IMPACTS OF ORAL DISEASE ON YOUNG PEOPLE

We would like you to take part in a research project. We are trying to find out how much mouth problems affect young people and their families. Then we can find out which problems need treatment and whether our treatment helps.

Before you decide it is important for you to understand why the research is being done and what it will involve. Please take time to read the information on the both sides of this sheet carefully and discuss it with others if you wish. Ask us if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part.

If you have any questions about the research please ask your dentist or contact:

Professor Peter G Robinson
Dept of Oral Health and Development
School of Clinical Dentistry
Claremont Crescent
Sheffield, S10.
Telephone: 0114 271 7892

or
Dr. David Winstanley
Research and Consultancy
Unit
2-4, Palmerston Road,
Sheffield, S10
Telephone: 0114 271 1435

What will I have to do?

Taking part involves two stages:

In stage 1:

- Both the young person and their guardian complete a questionnaire about mouth problems experienced by the young person
- The dentist will make an extra record about the condition of your child's mouth
- We will ask your permission to send you another questionnaire in 2 weeks time

In stage 2:

- Both the young person and their guardian fill in the questionnaire again

The questionnaire takes about 5 minutes to fill in. No special treatments or tests are necessary. The only difference you will notice between taking part and your normal appointment will be completing the questionnaires.

Do I have to take part?

No, you are free to make your own choice. The research is not part of your child's treatment and whether or not you choose to take part, the treatment will not be affected. If you take part but then change your mind you can drop out without affecting your child's treatment in any way.

Is it safe?

Yes. No special treatments or tests will be done to your child. The only difference either of you will notice between taking part and a normal appointment will be completing the questionnaires.

Who will see my information?

All information that is collected about you during the course of the research will be kept strictly confidential. Any information about you that leaves the hospital/surgery will have your name and address removed so that you cannot be recognised from it. The only people who will see the information will be the researchers. Nothing that identifies you will be kept on a computer. All the forms from the research will be kept securely in the Dental School. The reports from this research will not mention any of the people who took part.

How will it help me?

There is no payment for taking part. The benefit from taking part will be that you are helping us in our research to improve dental care for young people.

What if I am not happy about the way the study has been conducted?

If you have any reason to complain about any aspect of the way you have been approached or treated during this research, the normal NHS complaints mechanisms are available to you and are not affected because you have taken part in the research. If you have any complaints or concerns, please contact Professor Peter Robinson on the number overleaf. If that is not satisfactory, please contact Dr. David Winstanley.

What do I do now?

The dentist will tell you about the research and ask you to sign a form to show you agree to take part. If you don't want to take part, just tell the dentist.

Keep this form for reference. We will also give you a copy of a signed consent form to keep.

Thank you for your help!

Study Number: 02/218

Patient Identification Number for this trial:

CONSENT FORM

Title of Project:

IMPACTS OF ORAL DISEASE ON YOUNG PEOPLE

We would like you to take part in a research project. We are trying to find out how much mouth problems affect young people and their families. Then we can find out which problems need treatment and whether our treatment works.

Taking part involves two stages:

In stage 1:

- Both the young person and their guardian complete a questionnaire about mouth problems experienced by the young person
- The dentist will make an extra record about the condition of your child's mouth
- We will ask your permission to send you another questionnaire in 2 weeks time

In stage 2:

- Both the young person and their guardian fill in the questionnaire again

The questionnaire takes about 5 minutes to fill in. No special treatments or tests are necessary. The only difference you will notice between taking part and your normal appointment will be completing the questionnaires.

Taking part in this project is entirely voluntary. If you or your child decide to take part you may change your mind at any time and this will not affect your future treatment or care in any way. All the information gathered in the study will be confidential. No one will have access to it except the researchers. Neither your name nor anything that identifies you will be used in any reports of the study.

If you have any problems or feel you would like to know more, please ask your dentist or contact:

Professor Peter G Robinson, Dept of Oral Health and Development, School of Clinical Dentistry, Claremont Crescent, Sheffield, S10. Telephone: 0114 271 7892 or

Dr. David Winstanley, Research and Consultancy Unit, 2-4, Palmerston Road, Sheffield, S10

Telephone: 0114 271 1435

The *parent or carer* should complete the whole of this sheet.

Please write the name of the person for whom you are consenting.

.....

Please initial box

1. I confirm that I have read and understand the information sheet dated
(version) for the above study and have had the opportunity to ask questions.

2. I understand that our participation is voluntary and that we are free to withdraw at any time, without giving any reason, without our medical care or legal rights being affected.

3. I understand that sections of any of medical notes may be looked at by responsible individuals from this clinic. I give permission for these individuals to have access to those records.

4. We agree to take part in the above study.

_____ Name of Parent or carer	_____ Date	_____ Signature
_____ Name of Person taking consent (if different from researcher)	_____ Date	_____ Signature
_____ Researcher	_____ Date	_____ Signature

9.6 Appendix F Topic Guide

Welcome

Establishment of ground rules

No right or wrong answers

Use of a tape recorder by the researcher to remember what the young people say without having to take notes

Confidentiality-responses will not be shared with parents, teachers etc.

Young person to choose which name they would like to be referred to

Can terminate the interview at any time if required

Ask about term they prefer for people their age? Teenagers? Young people? Children?

Warm-up questions

Ask about a favourite activity

Ask about school- favorite subjects, teachers, friends, -can you tell me how many close friends you have and their first names?

Ask about good qualities-what are you good at?

Ask about ability to do things compared to friends

Appearance

Can you tell me of anyone at school who is good looking? Can you describe them?

What does being 'good-looking' mean to you?

What do you first notice about how people look?

How do you think your friends see you?

Is there anything about your appearance that you like or do not like?

Probe with: do you like what you see in the mirror? Do you like what you look like in photographs?

What is important to you about the way you look?

Oral Health in general terms

Do you and your friends talk about teeth at all?

What does having a healthy mouth mean to you?

How important do you think are teeth as a feature of the face?

What colours are teeth?

What does having good teeth mean to you? Can you describe features of good teeth/bad teeth?

Oral Health-own teeth

How would you compare your own teeth with your friend's teeth?

What have your parents/friends said to you about your teeth?

How would you describe your teeth?

Do you think you have good or bad teeth?

Is having good-looking teeth important to you?

Have you ever had trouble with your mouth, teeth or gums?

How have your feelings about your teeth changed over time?

Any comments made about your teeth by others who are not friends? Dentist?

Other young people?

Closing

If you could give tips to dentists about how to go about treating young people what would they be?

Thank the participant for the discussion

De-brief the participant on the aim of the project and the research process

Take photographs

9.7 Appendix F Information sheets and consent forms for Chapter Five

Information on the research project:

Young people's views of teeth

We would like you to take part in a research project. We are trying to find out what young people think about permanent marks on front teeth. The purpose of the study is to help us find out whether these problems need treatment and whether we should do more to prevent them.

Before you decide it is important for you to understand why the research is being done and what it will involve. Please take time to read the information on the both sides of this sheet carefully and discuss it with others if you wish. Ask us if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part.

If you have any questions about the research please ask your dentist or contact:

Zoe Marshman

Dept of Oral Health and Development

School of Clinical Dentistry

Claremont Crescent

Sheffield, S10 2TA

Telephone: 0114 271 7893 (office)

07703179151 (mobile)

Prof Peter G Robinson

Dept of Oral Health and Development

School of Clinical Dentistry

Claremont Crescent

Sheffield, S10 2TA

Telephone: 0114 271 7892

What will I have to do?

If you agree, an interviewer will come to your house and interview you. Your parent can be in the room during the interview if you want.

The interview takes 60 minutes at the most and will include the interviewer taking a photograph of your front teeth only. No one will be able to identify you from the photograph. The interview will be tape-recorded. No other special treatments or tests are necessary.

If you agree, we may wish to contact you again after the interview to follow-up points raised. We will destroy these contact details after three years.

Do I have to take part?

No, you are free to make your own choice. The research is not part of your dental treatment and whether or not you choose to take part, your treatment at the dentists will not be affected. If you take part but then change your mind you can drop out without affecting your treatment in any way.

Is it safe?

No special treatments or tests will be done to you.

Who will see my information?

All information that is collected about you during the course of the research will be kept strictly confidential. Any information about you will have your name and address removed so that you cannot be recognised from it. The only people who will see the information will be the researchers. Nothing that identifies you will be kept on a computer. All the forms from the research will be kept securely in the University of Sheffield. The reports from this research will not mention any of the young people who took part. The audio-tapes will also be stored securely and kept for 3 years before being destroyed.

What are the benefits of taking part?

There is no payment for taking part. The benefit from taking part will be that you are helping us in our research to improve dental care for young people.

Who is organising and funding the study?

The study is being organised and funded by the Department of Oral Health and Development of the University of Sheffield.

What if I am not happy about the way the study has been conducted?

If you are harmed by taking part in this research, there are no special compensation arrangements. If you are harmed due to someone's negligence, then you may have grounds for a legal action but you may have to pay for it. Regardless of this, if you wish to complain, or have any concerns about any aspects of the way you have been approached or treated during the course of this study, the normal National Health Service complaints mechanism should be available to you.

If you have any complaints or concerns, please contact Zoe Marshman on the number overleaf. If that is not satisfactory, please contact Prof Peter G Robinson.

What do I do now?

The dentist will tell you about the research. After you have had some time to think about it, return the reply slip in the envelope provided letting us know if you would be willing to take part. If you are willing, the researcher will contact you and ask you and your parent to sign a consent form and arrange a time for the interview when it is convenient for you.

Keep this form for reference. We will also give you a copy of a signed consent form to keep.

Thank you for your help!

CONSENT FORM

Title of Project:

Young people's views of teeth

We would like your child to take part in a research project. We are trying to find what young people think about permanent marks on front teeth. Then we can find out whether treatment is needed or more work to prevent these marks. This will involve your child being interviewed, the interview will be tape-recorded.

The interview will last 60 minutes at the most and will take the form of a chat rather than questions and answers. We would like to take photographs of your child's front teeth. Your child will not be identifiable by this photograph. No special treatments or tests are necessary.

Taking part in this project is entirely voluntary. If you or your child decide to take part you may change your mind at any time and this will not affect your future treatment or care in any way. All the information gathered in the study will be confidential. No one will have access to it except the researchers. Neither your child's name nor anything that identifies them will be used in any reports of the study.

We also ask your permission at this stage, to keep your child's contact details for a period of three years as it may be desirable to contact them again in the future to clarify information or to request they participate in further research in this area.

If you have any problems or feel you would like to know more, please ask your dentist or contact:

Zoe Marshman, Dept of Oral Health and Development, School of Clinical Dentistry, Claremont Crescent, Sheffield, S10. Telephone: 0114 271 7892

Parent or carer please complete this sheet.

Name of the young person to be involved in the research.

Please initial box

1. I confirm that I have read and understand the information sheet for the above study and have had the opportunity to ask questions.

2. I understand that photographs will be taken and that the purpose for which the material will be used has been explained to me in terms which I have understood.

3. I understand that sound recordings will be made and that the purpose for which the material will be used has been explained to me in terms which I have understood.

4. I understand that our participation is voluntary and that we are free to withdraw at any time, without giving any reason, without our medical care or legal rights being affected.

5. I understand that sections of my child's clinical dental notes may be looked at by responsible individuals from this dental clinic. I give permission for these individuals to have access to those records.

5. I give my permission for my child to be contacted again within a three-year period

6. I agree for my child to take part in the above study.



Name of Parent or carer

Date

Signature

Researcher

Date

Signature

1 for patient; 1 for researcher

Young person please complete this sheet.

Please initial box

1. I confirm that I have read and understand the information sheet for the above study and have had the opportunity to ask questions.

2. I understand that photographs will be taken and that the purpose for which the material will be used has been explained to me in terms which I have understood.

3. I understand that sound recordings will be made and that the purpose for which the material will be used has been explained to me in terms which I have understood.

4. I understand that our participation is voluntary and that we are free to withdraw at any time, without giving any reason, without our medical care or legal rights being affected.

5. I understand that sections of my clinical dental notes may be looked at by responsible individuals from this dental clinic. I give permission for these individuals to have access to those records.

5. I give my permission to be contacted again within a three-year period

6. I agree to take part in the above study.

Name of young person

Date

Signature

Researcher

Date

Signature

9.8 Appendix G Publications arising from this thesis

This research has also been reported in the following peer-reviewed research papers:

Z. Marshman, P.G. Robinson. Child and Adolescent Oral Health-Related Quality of Life. *Seminars in Orthodontics* 2007; 13: 75-126

Z. Marshman, B.J. Gibson, J. Owens, H.D. Rodd, H. Mazey, S.R. Baker, P.E. Benson, P.G. Robinson. 'Seen but not heard'. A systematic review of the place of the child in 21st century dental research. *International Journal of Paediatric Dentistry*. 2007; 17: 320-327

Z. Marshman, H.D. Rodd, M. Stern, C. Mitchell, P.G. Robinson. Evaluation of the Parental Perceptions Questionnaire, a component of the COHQoL, for use in the UK. *Community Dental Health*-in press, accepted November 2005

Z. Marshman, H.D. Rodd, M. Stern, C. Mitchell, D. Locker, A. Jokovic, P.G. Robinson. An evaluation of the Child Perceptions Questionnaire in the UK. *Community Dental Health* 2005; 22: 151-155

Child and Adolescent Oral Health-Related Quality of Life

Zoe Marshman and Peter G. Robinson

The quality-of-life data for children are less well developed than that for adults. Measurement difficulties arise because of a lack of a concept of quality of life in children and problems caused by developmental changes and the appropriateness of using a proxy. Over the past few years several measures designed specifically for assessing the impact of oral and orofacial conditions on the quality of life of children have been developed and evaluated. In future it is likely that the use of these measures will progress along with the use of qualitative approaches. (Semin Orthod 2007;13:88-95.) © 2007 Elsevier Inc. All rights reserved.

Despite the adoption of broad definitions of health, health has traditionally been measured through narrow, disease-focused clinical indicators relying on the judgment of professionals.¹ The use of clinical measures alone has been criticized for failing to capture the subjective experience of individuals.¹⁻³ Measures of quality of life are increasingly being used to supplement clinical indicators to explore the individual's perspectives on their health and health care.

There is an enormous array of definitions of quality of life. The World Health Organization Quality of Life Group defined quality of life as:

*An individual's perception of their position in life in the context of the culture and value system in which they live and in relation to their goals, expectations, standards, and concerns. It is a broad-ranging concept affected in a complex way by the person's physical health, psychological state, level of dependence, social relationships, and their relationships to salient features of their environment.*⁴

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The concept is multidimensional and measures used to assess quality of life have included aspects such as emotional, social, and psychological well-being and physical symptoms and functioning.⁵

This review will cover the main methodological challenges for assessing quality of life in children, detail how the measurement of oral health-related quality of life in this group has developed, describe the two main measures evaluated for this purpose, and end with a discussion of future directions for the assessment of oral health-related quality of life in children.

Assessing Quality of Life in Children

The assessment of quality of life in children provides unique insights into how disease, medical treatment and the prospect of disability affect them. Existing work on quality of life using quantitative measures in children is less well-developed than in adults and has mainly been in the fields of oncology, rheumatology, asthma, and epilepsy.⁶

Methodological challenges to the measurement of quality of life arise in three main areas: the concept of quality of life in children, problems caused by the changes children undergo both physically and cognitively, and the use of a proxy.

Difficulties with the Concept of Quality of Life

There is neither a consensual definition nor a conceptual model of quality of life in children. This lack leaves two key questions unanswered,

namely what is quality of life and how does it relate to other factors relevant to health care workers such as disease and well-being? These and similar questions should be resolved before the application of the measures becomes more widespread. To compound this, children's concepts of quality of life change as children develop. Existing measures used with children tend to be based on models of quality of life in adults or use quality-of-life questionnaires that are adapted from adult measures.⁷ These methods of development are inappropriate because much of the content of adult quality-of-life questionnaires is not relevant for children and may not address aspects of daily life that children value. This is a central requirement of health-related quality-of-life measures.⁸ For example, some quality-of-life measures have been adapted through the substitution of adult items, such as "work" with "school."⁹ In a review of quality-of-life measures for children, Vincent and Higginson found that most existing scales focus on physical and symptom-related aspects of quality of life and that they do not incorporate school-related items. The conclusion was that if the content of child quality-of-life questionnaires was to reflect the main areas of life in childhood, factors such as family and social relationships, activities, and schooling are all important.⁶

Problems Due to the Cognitive and Physical Changes in Children

The measurement of quality of life in children is complicated by the rapid changes seen as children develop.^{10,11} These changes have implications for the reading levels, type of scales, time frames used, and the general ability of children to comprehend child quality-of-life questionnaires.⁶ To some extent, simple questionnaires, pictorial representations, and the assistance of an interviewer may overcome some limitations in children's levels of literacy and comprehension, but different scales for different stages of development are required. A number of questionnaire biases, such as position and acquiescence bias, have been associated with child respondents and may complicate the collection of self-report data in this group.¹² These problems have led to the psychometric properties of ques-

tionnaires for children being questioned. The Child Asthma Questionnaire (CAQ) has forms for three different age groups (4 to 7 years, 8 to 11 years, and 12 to 16 years) tailored to the level and life context.¹³ The CAQ has demonstrated that it is possible to obtain valid and reliable reports of health-related quality of life from children.¹⁴

Use of Proxies

Information from parents or caregivers can be used to supplement child assessments of quality of life or can be used as a proxy for the child assessment.

Supplemental information from parents can be useful as it may influence decisions about clinical management, and involving parents in the assessment of their child's quality of life does provide an opportunity to raise awareness in parents or caregivers.¹⁵

The use of parents or caregivers as proxies has advantages in overcoming some of the concerns about the ability of children to provide assessments that meet psychometric standards and practical problems of reading level and comprehension. Indeed, the use of proxies may be the only feasible option for very young or sick children.¹² However, only modest agreement is found between parents as proxies and children's reports of quality of life.¹⁶ Eiser and Morse performed a systematic review to determine the relationship between the ratings of children's quality of life made by parents and children.¹⁷ The results of the review indicate that the accuracy of proxy ratings is dependent on the specific domains of quality of life considered. There is greater agreement for observable functioning (eg, physical quality of life) than for nonobservable functioning (eg, emotional or social quality of life). Agreement is better between parents and chronically sick children compared with parents and their healthy children (possibly because of greater communication about illness and treatment). However, the importance of any agreement and the common assumption that information from proxies should "match" that provided by children is questioned. The standard practice in developing new measures of quality of life, of reporting validity by determining correlations between child and proxy ratings, is also highlighted. Although strong cor-

relations between child and proxy data demonstrate some validity, it should not be assumed that they are interchangeable. The conclusion reached by the review is that differences between ratings made by children and adults must be anticipated and treated as important.

Where possible, then, information from parents should be used to complement the views of children although in some circumstances the use of a proxy is unavoidable.

Qualitative Approaches to Health-Related Quality of Life

While much of the research on health-related quality of life in children has involved quantitative assessments, qualitative research involving children and parents can achieve greater understanding of child quality of life.⁶ A review of the use of qualitative methods in children and adolescents concluded such methods to be appropriate for capturing information on the experiences and meanings of quality of life.¹⁸

Oral Health-Related Quality of Life

Over the past decade the assessment of oral health-related quality of life of adults has increased markedly. The potential applications of such measures have been categorized into theoretical, political, and practical¹⁹ (Table 1).

Locker described the potential theoretical applications as exploring models of health and

disease, describing income inequalities and determining influential psychosocial factors.¹⁹ In addition, other theoretical applications described include elucidating the relationship between different aspects of health.²⁰

One political application described by Locker was in advocating for resources. This is particularly relevant for oral health services and research as it is often isolated from mainstream health care systems. Such measures could place dental health in context by showing the impact dental conditions can have on ability to function, for example, giving oral health legitimacy with policy makers.²¹ Other applications described include harnessing public priorities,²⁰ which is particularly important given increasing political emphasis on patient and public involvement in health care.²²

The main practical applications of health-related quality-of-life measures are in evaluating services for research, public health, and clinical purposes. Other public health uses include describing and monitoring health status of populations, the results of which can be used to assess population needs, identify target populations, and priority setting. In research terms, they can be used as outcome measures in clinical trials.²³ In this application, health-related quality-of-life questionnaires can measure alleviation of symptoms, restoration of function, and any iatrogenic effects. Measures have been developed specifically for this purpose in dentistry.¹⁰

In clinical practice, with individual patients, measures can be used to facilitate communication and identify patient preferences.¹⁹ In dentistry, measures could be useful for nondental health care workers to identify needs for referral for dental treatment.²⁴ Other clinical uses include assessing likely compliance with treatment, and in the United States, for example, the use of such measures has been suggested as a way for dentists to market dental services to patients.²⁵

Oral Health-Related Quality of Life in Children

Initial attempts to assess oral health-related quality of life in children used questionnaires designed for use in adults.

Table 1. Summary of the Applications of Health-Related Quality of Life Measures

Theoretical
Exploring models of health
Describing factors influential to health
Political
Demonstrating involvement of the public in healthcare
Identifying the public's priorities
Advocacy
Practical
Public health:
Planning, monitoring and evaluating services; Needs assessments
Research:
Evaluating outcomes of healthcare interventions
Clinic based:
Evaluating individual patient care
Improving patient-practitioner communication
Clinical audit
Marketing of services

Use of Adult Oral Health-Related Quality-of-Life Measures in Children

The two most commonly used adult oral health-related quality-of-life measures, namely the Oral Health Impact Profile (OHIP)²⁶ and the Oral Impacts of Daily Performance (OIDP),²⁷ have been used in children. The appropriateness of these measures for children in terms of their length, content (neither include items assessing relationships or activities), and psychometric properties have been questioned.²⁸ The psychometric properties of instruments such as validity and reliability can be affected when the measure is used for a purpose other than that for which it was intended.^{29,30}

Child Oral Health-Related Quality-of-Life Measures

Accordingly, several groups identified a need for an oral health-related quality of life measure that was relevant to children and their families.^{10,24,31} Several measures have since been developed, namely:

- Second International Collaborative Study Oral Health-Related Quality of Life Questionnaire for Children (ICSII-OHRQOL)³¹
- Michigan Oral Health-Related Quality of Life Scale-Child Version³²
- Child Oral Health Quality of Life (COHQoL) Questionnaire¹⁰
- Child-Oral Impacts of Daily Performance (Child-OIDP)³³

Only the COHQoL and the Child-OIDP have been developed from a defined conceptual model of health-related quality of life; likewise only these two measures have been subjected to psychometric evaluation. The present review will therefore focus on them.

Child Oral Health Quality of Life (COHQoL) Questionnaire

A team in Toronto designed the COHQoL Questionnaire to assess the oral health-related quality of life of children 6 to 14 years old.¹⁰ The COHQoL questionnaire is intended for use as an outcome measure in clinical trials and evaluation studies. It consists of a Parental-Caregiver Perceptions Questionnaire (P-CPQ) and Child

Perceptions Questionnaires (CPQ) for children aged 6 to 7 years, 8 to 10 years, and 11 to 14 years. The measures are to be used for children with a wide range of oral and orofacial conditions including caries, malocclusions, clefts, and other orofacial anomalies.

The CPQ₁₁₋₁₄ and P-CPQ questionnaires are based on a conceptual framework developed from a review of child quality-of-life measures. A novel process was used to ensure that the final questionnaire contained items of the most relevance to children and parents of children with oral and orofacial conditions.⁸ The CPQ₁₁₋₁₄ contains 36 items encompassing 4 health domains: oral symptoms, functional limitations, emotional well-being, and social well-being (peer interaction, schooling, and leisure activities). It asks about the frequency of events in the previous 3 months on a 5-point Likert scale between never (scores 0) and everyday or almost everyday (scores 4). For children a 3-month reference period reduces recall bias compared with the 6-month period used by some adult measures. Summing the response codes for all items generates an overall CPQ₁₁₋₁₄ score and in addition scores for each domain can be computed. The questionnaire is self-administered. The CPQ₁₁₋₁₄ has acceptable reliability and validity in Canada,¹⁰ New Zealand,³⁴ Uganda,³⁵ and the United Kingdom,³⁶ although it may be insensitive to impacts at the levels seen in the general populations of some of these countries.

As part of the project to validate the CPQ₁₁₋₁₄, relationships between children's global ratings of oral health and their oral health-related quality of life were investigated.³⁷ Global ratings are single-question measures that ask about current health. The single-question measures used were: the global rating of oral health worded "Would you say that the health of your teeth, lips, jaws and mouth is . . .", and a rating of the extent to which oral/orofacial conditions affect overall well-being worded "How much does the condition of your teeth, lips, jaws and mouth affect your life overall?" Multiple linear regression analyses were used to identify items from the CPQ₁₁₋₁₄ that predicted these global ratings. The oral symptoms subscale score predicted the oral health rating with those reporting more symptoms rating their oral health less favorably. Predictors of overall well-being were the functional limitation subscale score and the emotional well-

being subscale score. The authors concluded that the findings from children concur with contemporary thinking on the consequences of disease, in that health and health-related quality of life are not merely determined by the nature of the disease but also by attributes of the personality and environment.

The CPQ₁₁₋₁₄ has subsequently been used to compare the impact orofacial conditions and dental caries have on the oral health-related quality of life of children with these conditions.³⁸ The orofacial conditions in question were cleft lip, cleft palate, and other craniofacial anomalies. The results found no marked differences between the orofacial group (n = 39) and the dental group (n = 31). The authors concluded that this may be based on the high levels of clinical, social, and emotional support available and on psychological attributes of the orofacial conditions group giving them the resources to face the challenges their conditions bring. The authors acknowledged, however, the relatively small sample size used.

Since the development of the CPQ₁₁₋₁₄, a questionnaire for 8- to 10-year-old children (CPQ₈₋₁₀) has been developed and evaluated. Twenty-five questions from the CPQ₁₁₋₁₄ were selected based on advice from parents, a child psychologist, a teacher, and information from the literature. Again, this measure showed good validity and reliability in Canada³⁹ and the United Kingdom.⁴⁰

The P-CPQ is a measure of parental-caregiver perceptions of the oral health-related quality of life of children. It is not intended to be a proxy measure but to supplement information obtained from the children. The P-CPQ has 31 items and includes a "don't know" response option. An evaluation of P-CPQ in Canada found acceptable psychometric properties.⁴¹ The level of agreement between children's oral health-related quality of life and that perceived by their mothers was good, with substantial agreement between scores.⁴² However, the level of agreement varied according to the characteristics, with only moderate agreement for the emotional and well-being subscales. The authors concluded that mothers may be used as proxies in some circumstances but advised that the views of children were also required.

Child-OIDP

The Oral Impact on Daily Performance (OIDP) index was developed from a theoretical model of oral health proposed by Locker⁴³ as adapted from the World Health Organization Classification of Impairments, Disabilities and Handicaps.⁴⁴ The OIDP measures major impacts, equivalent to disability and handicap only. An OIDP index appropriate for children to aid assessment of need in child populations was developed in Thailand.³³

The Child-OIDP was developed from the OIDP index using input from children and pediatric dentists and paying attention particularly to the language used, performances included, number of questions, and the response format.

The Child-OIDP is suitable for 11- to 12-year-olds and is administered as an interview. It includes 8 performances: eating, speaking, cleaning mouth, sleeping, emotion, smiling, study, and social contact. The response format adopts a 4-point Likert scale (scored 0-3), which has better reliability than a 0 to 5 scale. For each performance the child scores the frequency (0 to 3) and the severity (0 to 3) of the impact; these scores are subsequently multiplied to give a score ranging from 0 to 9 per performance. The overall impact score is the sum of all 8 performances (ranging from 0 to 72). The recall period of 6 months for the OIDP was shortened to 3 months for the child version. Pictures to illustrate the performances were developed, the use of which helped reduce the duration of the interview.

The validity and reliability of the Child-OIDP in Thai children was found to be acceptable.³³ Since development the Child-OIDP has been used in a cross-sectional survey in Thailand to assess the prevalence and severity of oral impacts.⁴⁵ This was the first population-based study using an oral health-related quality-of-life questionnaire in children. Clinical data on caries levels, Community Periodontal Index, oral hygiene, and orthodontic treatment needs of the children were collected. The results show a very high prevalence of impacts, but the impacts were not severe. Ninety percent of children reported having experienced some kind of oral impact, with impact on eating and cleaning teeth being the performances with the highest prevalence. In terms of severity, 19% of children had severe

impacts, with eating and smiling being the most severely affected. Impacts on studies and contact with people were the least common and severe. As this was a low-carries population (mean number of decayed, missing and filled teeth = 1.5), the authors discussed likely causes of impacts to be exfoliating deciduous teeth, mouth ulcers, bleeding gums, and dissatisfaction with the position of teeth.

Summary of Child Oral Health-Related Quality of Life Questionnaires

In summary, the COHQoL questionnaires were developed for use in children with oral and orofacial conditions as an outcome measure in clinical trials. Several of the component questionnaires of the COHQoL have been evaluated for use in different countries and found to have acceptable reliability and validity. To date, use of the questionnaires has been restricted to a comparison of the impacts of orofacial conditions and caries. Although the measure was intended for use in clinical trials, the ability of the questionnaire to detect change in longitudinal studies has not been assessed.

The Child-OIDP was developed for population-based surveys of 11- to 12-year-old children and has been found to have acceptable psycho-

metric properties for use in Thailand. It has subsequently been used in a large cross-sectional study to describe oral impacts of Thai school children. Despite the Child-OIDP being designed specifically to be used to assist dental service planning, no indication is given as to how the index is applicable to this function.

Discussion

A review of the literature reveals how the increase in interest in measuring oral health-related quality of life in children has developed over the past 5 years. Table 2 summarizes the published literature on oral health-related quality-of-life measures designed for children.

Several measures have been developed, but as yet little has been published on the use of such measures despite the potential theoretical, political, and practical applications. To improve the theoretical basis of oral health-related quality of life in children, an explicit model would help to inform the continuing development of this field. Politically, developments such as the UK National Service Framework for Children and Young People⁴⁶ require services to be child-centered, and to meet the needs of children and their families thus providing an opportunity to

Table 2. Published Literature on Oral Health-Related Quality of Life Measures in Children

<i>Author</i>	<i>Year</i>	<i>Age of Children (yrs)</i>	<i>Questionnaire Used</i>	<i>Purpose of Study</i>
Tapsoba ³¹	2000	12-13	ICSII-OHRQOL	Evaluation of properties of measure in Poland, Germany and New Zealand
Jokovic et al ¹⁰	2002	11-14	CPQ	Development and evaluation of measure in Canada
Filstrup et al ³²	2003	>4	Michigan Oral Health-related Quality of Life Scale-Child Version	Describes use of measure but no evaluation
Jokovic et al ³⁹	2004	8-10	CPQ	Development and evaluation of measure
Gherunpong et al ³³	2004	11-12	Child-OIDP	Development and evaluation of measure in Thailand
Gherunpong et al ⁴⁵	2004	11-12	Child-OIDP	Use of measure in cross-sectional survey in Thailand
Humphris et al ⁴⁰	2005	8-10	CPQ	Evaluation of measure in UK
Foster Page et al ³⁴	2005	11-14	CPQ	Evaluation of measure in New Zealand
Marshman et al ³⁶	2005	11-14	CPQ	Evaluation of measure in UK
Locker et al ³⁸	2005	11-14	CPQ	Use in children with caries and orofacial conditions
Jokovic et al ³⁷	2005	11-14	CPQ	Relationship with global ratings
Robinson et al ³⁵	2005	12	CPQ	Survey to describe impacts and compare impact of caries and fluorosis

put oral health-related quality-of-life measures to use in this priority area. Third, a practical application in pediatric dentistry and orthodontics may be an education of dental and other health professionals on the aspects of children's quality of life affected by oral health conditions and the treatments of it.

When deciding on the most appropriate measure of oral health-related quality of life in children to use for a study, the choice will be dependent on the purpose.²⁰ It is anticipated that further research will also be required to evaluate the measures for use in other countries.

To date, little research has been conducted to capture in-depth information from children on how their oral and orofacial conditions affect quality of life despite qualitative techniques being used successfully in other fields.⁶ Qualitative methods would offer the opportunity to hear from children themselves about the areas of their lives affected and allow further exploration of the interplay of psychosocial factors.

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Seen but not heard: a systematic review of the place of the child in 21st-century dental research

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Background. The position of children in society has changed with increasing emphasis on children's rights and child-centred services. This study aimed to describe the extent to which contemporary oral health research has been conducted *with* or *on* children.

Design. A systematic review of the child dental literature from 2000–2005 was conducted. A purposive sample was used to develop categories describing the level of involvement of children in research. Four main categories were developed: children as the objects of research, proxies used on behalf of children, children as the subjects of research with some involvement and children as active participants with their perspectives explored.

Electronic databases were searched and exclusion criteria applied. Each of the resulting papers was examined and categorised. The frequency distribution in each category and the distribution of these categories according to subject were calculated.

Results. The search revealed 3266 papers after application of the exclusion criteria. Of these, 87.1% were categorised as research where children were used as objects, 5.7% were found to involve proxies (parents or clinicians), 7.0% involved children to some extent and 0.3% involved children actively.

Conclusion. Most oral health research is conducted *on* children, in future research should strive to be conducted *with* children, involving them as fully as possible.

Introduction

'Children seen, but not heard' is a saying that originated from early Victorian times. During such times, children did not have a childhood as we know it today; they were seen as 'imperfect' adults, wearing smaller versions of adult's clothes and went to work rather than school¹. The late nineteenth century was significant in the construction of the modern childhood and more recently, the past 30 years have seen the position of children in society change further, moving them to the forefront of personal, political, and academic agendas². From a personal and family perspective, changes in developed countries have been due to a demographic shift; for example, in the UK, the proportion of the population aged under 16 has declined since the 1970s³. In addition, there has also been a

change in family structure, with an increase in one-parent families⁴. Collectively, such changes have led to the idea of the child as a scarcity and thus more 'precious'⁵.

Politically, legislation has changed children's rights both globally and locally. The United Nations Convention on the Rights of the Child was adopted in 1989 and is the most universally accepted human rights instrument in history, being ratified by nearly every country in the world⁶. This Convention confirms, amongst many other things, children's participation rights to express their views and to have them taken seriously and given due weight. In English law, the *Children Act 2004* requires welfare agencies to take account, not only of children's best interests, but also their wishes and desires⁷. The English government has also shown its commitment to ensuring health and other services are child centred^{8,9}. The National Service Framework for Children, Young People and Maternity Services (Children's NSF) requires services to give children and their parents increased information, power, and choice over

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the treatment they receive and involve them in planning their care. Dental services are included in the Children's NSF⁸.

There has also been a change in the field of childhood study and the level of involvement of children in research². James and colleagues have described how most research until the 1990s viewed children as developmentally incomplete adults and gave little time to children themselves. Thus, there was a culture where research was conducted *on* children. Since then, as more weight has been given to the rights and views of the child, the voices of children have increasingly been recognized¹⁰ with a change in emphasis to research *with* children. Social sciences have embraced this shift and moved away from research methods that view children as 'objects of concern', to methods that engage children as 'active participants'¹¹. This research goes beyond just considering what is in the child's best interest to actually involving children in research to gain their perspectives and own experiences.

During dental care, the importance of seeking children's views is widely acknowledged. Paediatric dentistry texts on the subject outline the importance of understanding how children relate to the adult world, with dental students taught the importance of talking to children, listening to their answers, and giving children control over their dental care¹². However, we have no knowledge of how far research in this field has been conducted *with* or *on* children.

This study therefore aimed to conduct a systematic review of the extent to which contemporary oral health research has been conducted *with* or *on* children. The review will enable any deficiencies in approaches to research in this field to be highlighted.

Materials and method

Developing the categories

The study was conducted in two stages, first to develop categories to classify research and then systematically reviewing the literature to place papers in these categories.

The categorization framework was developed using framework analysis, which classifies qualitative data by organization according to key themes and emerging categories¹³. This matrix-based method, which has been widely used in applied policy research, allows data to be synthesized quickly when specific information is needed. A purposive sample of child dental literature from the past 5 years was chosen to include a wide range of studies from both different subject areas (including paediatric dentistry, orthodontics, restorative dentistry, oral pathology/oral surgery/oral medicine, and dental public health) and epistemological stances. The two themes of *on* children and *with* children were explored and characterized, based on an initial sample of 20 papers. Four main categories were then identified and frameworks devised so the properties of these categories could be developed (Table 1).

The first category (category 1) included research where children were active participants being seen, listened to, and heard; this research attached a priority to fully involving children. Within this category two subcategories were derived based on the degree to which the children were involved. The first subcategory (category 1a) was research conducted with children as participants actively engaged throughout the research process (e.g. involving

Table 1. Frequency distribution of categories of papers.

Category	Properties	No. of papers (%)
1. <i>With</i> children – children seen as active participants	(a) Children involved in research process	2 (0.1)
	(b) Children's own accounts	6 (0.2)
2. <i>With</i> children – children seen as subjects	(a) Children completing measures designed by adults	220 (6.7)
	(b) Case report/series with child's input throughout case	10 (0.3)
3. Proxies for children used	(a) Parent/caregiver used appropriately as proxy	173 (5.3)
	(b) Clinician used appropriately as proxy	12 (0.4)
4. <i>On</i> children	Children seen as the objects of the research	2843 (87.1)

them in research design, in piloting, using participatory data collection methods, and getting their feedback on results). The second subcategory (category 1b) comprised research where children participated by giving accounts of their experiences in their own words using methods such as semistructured or in-depth interviews or focus groups.

The second category (category 2), also classed as research *with* children, included studies where children were seen and listened to as the subjects of research, although their own words were not heard. This category was subdivided into studies where children completed measures designed wholly by adults (category 2a) including self-complete questionnaires, structured interviews, and visual analogue scales. The second subcategory included clinical case studies showing evidence of the child's feelings being taken into account throughout the reporting of the case, such as reporting the presenting complaint in the child's own words and evidence of discussions with the child about the treatment (category 2b).

The third category included research that used either parents/caregivers (category 3a) or clinicians (category 3b) as appropriate proxies for children. It was felt appropriate to use proxies for young children (under 6 years) or those who lacked verbal articulacy, as research with the children themselves would not be a feasible option¹⁴.

The final category (category 4) included research where children were simply objects to be studied. In this research, children were not listened to or heard but only seen. Included within this category was research that saw children as:

- a set of teeth or a mouth to be treated;
- a source of a sample of plaque, saliva, or hard/soft tissue;
- an age group of patients to be managed;
- a child patient with a medical condition;
- a population group to be surveyed clinically;
- a patient on whom a 'special investigation' was conducted; and
- a recipient of a oral health promotion intervention.

These four main categories with their subcategories were developed to classify papers identified in a systematic review.

Search strategy

The search strategy was based on published studies using child-related keywords (child* or young person or young) and dental-related keywords (erosion and dent* or trauma and dent* or fluorosis and dent* or periodont* or malocclusion or orofacial or oral or periodont* or orthodont* or caries).

The search strategy was performed on the databases MEDLINE (via Ovid) and EMBASE and limited to the English language. The resulting references were exported to an Endnote library¹⁵ and all duplicates were removed. A list of dental journals was compiled based on the research team's knowledge and experience and any articles from nondental journals were excluded from the electronic library.

The so-called 'new social studies of children and childhood' were consolidated in 1998² with methods described for conducting such research published in 2000¹¹. Consequently, the most appropriate time frame to explore how far dental research was conducted *on* or *with* children was from 2000 to 2005.

At the first pass through the library of references the following exclusion criteria were applied:

- reports before 2000;
- studies with participants over 16 years of age;
- studies with no primary data;
- articles reporting *in vitro* studies;
- conference proceedings; and
- articles that did not have children and aspects of their oral health as their main topic.

A team of researchers from different disciplines (paediatric dentistry, medical sociology, health psychology, dental public health, and orthodontics) was recruited to conduct the review. Two trained reviewers from the team independently applied the exclusion criteria based on the abstracts and where necessary the full-length papers. Agreements between the reviewers about application of exclusion criteria occurred for 77% of the papers and disagreements were resolved through discussion.

The initial search resulted in 18 249 papers although this represented 14 895 individual papers after duplicates were removed. After excluding articles from nondental journals the

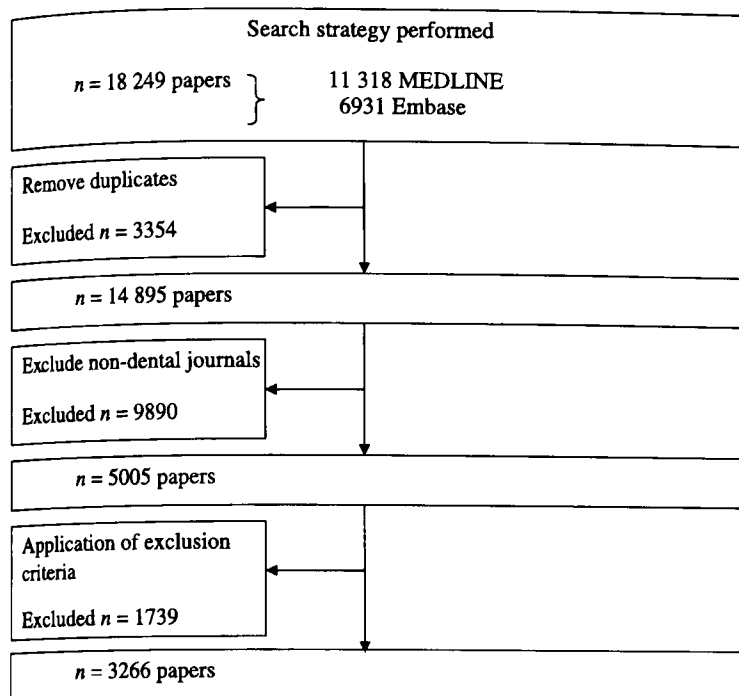


Fig. 1. Results of literature search.

number of papers reduced to 5005. Application of the exclusion criteria resulted in 3266 papers, of which 752 were case reports/series (Fig. 1).

Applying the categories

Before the papers were categorized the eight reviewers undertook training using 15 selected papers and then a calibration exercise on a further two sets of 15 papers. For the second set, agreement on the categorization of the individual papers ranged from 62.5% to 100%.

Two reviewers then categorized each paper independently, with four pairs of reviewers assessing approximately 817 papers per pair. When it was not possible to categorize the papers from the abstract, the full article was reviewed. Where a paper appeared to fit into more than one category, the category that presumed the greater involvement of children was chosen. Inter-examiner agreement between the two reviewers was assessed. The agreement between the pairs of reviewers ranged from 88% to 92%. Disagreements about categorization were resolved through discussion and, if necessary, involvement of a third reviewer.

After the papers had been categorized, they were grouped according to the subject area covered by the journal in which they were published. Journals fell into six broad areas: general dentistry, orthodontics, oral surgery/oral

medicine/oral pathology, restorative dentistry, dental public health, and paediatric dentistry.

Results

Of the 3266 papers that resulted from the literature search, only 238 (7.3%) were categorized as research *with* children (Table 1).

Research with children

Eight papers (0.3%) involved children as active participants, two (0.1%) with evidence of children being included throughout the research process (category 1a). These two papers both concerned developing questionnaires and involved children in the developing, compiling, and evaluating of the instruments^{16,17}. These papers were published in 2002 and 2004 in two different journals. A further six (0.2%) papers reported qualitative studies using interviews or focus groups (category 1b). The first of these studies was published in 2002. These studies explored children's perspectives on oral health generally, dental services, habits (drinking carbonated drinks and smokeless tobacco use), oral health education messages, their oral symptoms, and compliance with orthodontic treatment. Most studies involved adolescents, although in one study children from 6 years of age were interviewed.

In the second category, where children were seen as subjects, 220 (6.7%) papers involved children completing measures wholly designed by adults (category 2a). Unlike papers in category one, these papers used measures developed without children's input into the topics they felt were relevant or in the format or wording of the measures.

Only 10 of 752 case reports/series had evidence of the child's involvement (category 2b). These 10 cases included the patient's presenting complaint in his or her own words, a description of the patient's input into decisions about the treatment options, and the patient's perspectives on the outcome of treatment.

Use of proxies

A total of 185 (5.7%) papers used proxies to gain the child's perspective of which 173

(5.3%) used parents/caregivers and 12 (0.4%) used clinicians. The children of interest in studies involving parents/carers (category 3a) were either less than 6 years of age or older children with communication difficulties. The papers reporting the use of clinicians as proxies (category 3b) investigated the impact of dental treatment or treatment services on young children or those unable to communicate themselves.

Research on children

Finally, the vast majority ($n = 2843$, 87.1%) of papers were categorized as research *on* children. Implicit in these papers was the idea of children as the objects of research, with no involvement of children or their parents to any extent. Within these were extreme examples that referred to the children studied as 'the material'.

Subject area

Papers were also grouped according to the subject area covered by the journals in which they were published. When expressed as a percentage of the total number of papers per area, 9.2% of papers in general dental journals involved research with children; 5.9% in orthodontic journals; 7.5% in oral surgery, oral medicine, oral pathology journals; 5.8% in restorative dentistry journals; 11.4% in dental public health journals; and 5.4% in paediatric dentistry journals (Table 2).

Discussion

The categories developed for this systematic review represented a hierarchy of involvement of children in child dental research ranging from full involvement in the research process to no involvement. When these categories were applied to child-related dental research over the past 5 years most research used children as objects with no other involvement. This study highlights the need for future research to be conducted *with* children to capture their own experiences of oral health and treatment.

While we acknowledge that research *on* children may be appropriate to answer certain questions, when research opportunities arise, the potential to capture the perspectives of children should be considered. From this review, research *on* children typically views them as objects: as a set of teeth to be treated or a source of a sample of plaque or saliva. It treats children as a homogenous age group rather than as individuals and makes generalizations to this effect. A more holistic approach to studying oral health is required that recognizes the interaction of these biological perspectives with the social and psychological perspectives of the individual¹⁸. For example, a study that reports the survival of re-implanted avulsed teeth from the normative view of the clinician will ignore the children's subjective experience of the trauma-related treatment and outcomes. Similarly, traditional epidemiological surveys of children have also usually relied on normative assessments of the prevalence of oral

Table 2. Frequency distribution of papers by journal area.

Properties	General dentistry	Orthodontics	Restorative dentistry	Dental public health	Oral surgery, oral pathology, oral medicine	Paediatric dentistry	Total no. of papers
<i>With children:</i>							
(1a) Children involved in process	1	0	0	1	0	0	2
(1b) Children's own accounts	5	0	0	0	1	6	6
(2a) Children completing measures designed by adults	68	37	13	27	47	28	220
(2b) Case reports with child's input	0	3	0	1	1	6	10
<i>Proxies:</i>							
(3a) Parent/carer used appropriately	44	7	17	41	13	51	173
(3b) Clinician used appropriately	1	1	0	1	5	4	12
<i>On children:</i>							
(4) Children as objects	689	634	193	184	590	553	2843

disease. In the UK, the authors of the national Child Dental Health Survey 2003 were keen to supplement these data, for the first time, with an assessment of the impact of oral health on children. However, a validated measure of child oral health-related quality of life was not available in the UK at that time³. Due to developments in child-centred research these measures are now available^{19,20} and can be incorporated into future surveys of children's oral health. Only by conducting research *with* children will our understanding of children's oral health and their views of management be expanded.

Generally there has been an increasing emphasis placed on user involvement in research²¹. Research ethics committees and grant-awarding bodies require evidence of the level of involvement of participants throughout the research process, rather than merely assessing whether the research potentially puts participants at risk. Within health care the views of patients and the public on their experiences of health and health services have attained much higher significance in recent years²². Patient's rights to be listened to and have their needs acted upon have been clearly outlined²³. This emphasis has been particularly strong in services for children with an explicit requirement for services to be child-centred. Given that it is the child who undergoes the treatment and who lives with the consequences, it is important to consider, not just what clinicians think is in their best interests, but their own perspective, desires, and expectations²⁴. The Children's NSF suggests professionals should listen to children themselves, value their views, and take these into account in decisions about their care and the planning, delivery, and evaluation of services⁸. While communication with patients is stressed as a key feature of dentistry for children¹², this emphasis does not appear to be mirrored in research.

Within social science there has been an increasing recognition of the importance of listening to children to improve understanding of what is important to children, rather than research being defined by adult interests, biases, and agendas^{2,10}. The child-centred nature of this kind of research is not just evident in the research methods used, but also through

working with children to identify research questions that are meaningful to them and disseminating the findings back to them. This approach has led to social policy changes that reflect children's concerns more accurately²⁵. We acknowledge that there is an overlap between the categories developed in this study and the research methods reported in the articles, but conducting dental research *with* children requires more than just using participatory methods, it demands that we strive to involve children more fully throughout the research process.

This systematic review did identify several studies that have successfully involved children as active participants, exploring their perspectives on oral health or involving them in questionnaire development^{16,17}. Involving children in this way ensures that aspects pertinent to them are included, that language to which they can relate is used, and that questionnaire formats are appropriate²⁶. Interestingly, only 5.4% of articles published in specific paediatric dentistry journals were categorized as involving research *with* children. This was the lowest percentage of the six subject areas suggesting considerable opportunity for publishing further research of this kind in the paediatric dentistry literature.

Qualitative research with children has also been attempted²⁷, which adds the advantage of capturing children's perspectives in their own words rather than imposing adult-generated frameworks that might distort their ideas²⁸. Dentistry has been slow to adopt qualitative methodology with some initial resistance to its use²⁹. If more qualitative research is to be conducted with children, paediatric dentists will require training to develop these skills or may choose to work collaboratively with other disciplines with this expertise. This systematic review should be repeated in 5 years' time to investigate whether an increasing emphasis on research *with* children can be observed.

About 6% of papers involved parents/carers or a clinician as a proxy for the child. The use of proxies has advantages in overcoming some of the practical problems of reading level and comprehension in young children¹⁴. However, when comparing data obtained from parents

as proxies with children's self-reports of quality of life, agreement is modest³⁰. For this reason information from parents should be used to complement the views of children rather than as a substitute, although in some circumstances the use of a proxy is unavoidable.

It is acknowledged that the present study has some limitations. Notably, for practical reasons, the search was restricted to electronic databases, the English language and dental journals. The adoption of this strategy meant that some relevant studies may have been omitted. First, as child-centred research is at a relatively early stage in dentistry, such studies may not have reached the stage of publication yet. Inclusion of conference proceedings would have included some such studies, but would have resulted in an unmanageable number of articles. Second, studies reporting dental research *with* children may have been published out with the dental literature and therefore have been overlooked. Third, the reports of the research may not provide comprehensive details of the actual study. Children may have thus been more fully involved in some studies without this being documented as such in the text.

In summary, the findings of this systematic review demonstrate that in most child dental research, children are seen, but not listened to or heard. In future, researchers should attempt to involve children as far as possible in their studies to ensure that their perspectives are obtained.

What this paper adds

- An overview of the changing position of children in society
- An investigation of the extent to which child dental research is conducted *with* or *on* children
- Identifies opportunities for future research with children

Why this paper is important to paediatric dentists

- This paper challenges the way paediatric dentists involve children in research
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Evaluation of the Parental Perceptions Questionnaire, a component of the COHQoL, for use in the UK.

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Objective To assess the reliability and validity of the Parental Perceptions Questionnaire (PPQ) for use in the UK and to investigate whether different approaches to the treatment of ‘don’t know’ (DK) responses have any effect on the psychometric properties. **Methods** The parents of 89 children attending for an examination at a dental teaching hospital and a general dental practice completed the Parental-Caregiver Perceptions Questionnaire (PPQ), global oral health and global impact ratings. Clinical data were also collected. Four approaches were taken to the management of DK responses, one approach involved exclusion of DK responses and three approaches involved adjustment of DK responses (item mean, mean items answered and replacement of DK responses with zero). **Results** All four approaches demonstrated acceptable internal consistency and test-retest reliability of the total scale. The mean items answered and replacement approaches had optimal internal consistency of the subscales of the PPQ. Assessments of criterion validity in relation to global oral health rating were similar when the DK responses were adjusted, but the exclusion of DK responses had a detrimental effect. Construct validity of PPQ in relation to global impact rating and clinical data was acceptable only when responses were adjusted. **Conclusion** These data suggest that if DK responses are adjusted, the reliability and validity of this measure are acceptable for use in the UK

Key words: Oral health, psychometrics, quality of life

Introduction

Measures of quality of life are increasingly being used to supplement clinical indicators and to explore the impact of conditions on patients. In the case of the quality of life of children, information from parents or carers has been used to supplement child assessments of quality of life or as a proxy for the child assessment (Eiser and Morse, 2001).

The use of parents or carers as proxies has advantages in overcoming some of the concerns about the ability of children to provide assessments that meet psychometric standards, practical problems of reading level and comprehension (Rosenbaum and Saigal, 1996). Indeed, the use of proxies may be the only solution for very young or poorly children (Pantell and Lewis, 1987). However, only modest agreement is found between parents as proxies and children’s reports of aspects of quality of life (Achenbach *et al.*, 1987). Eiser and Morse, (2001) carried out a systematic review to determine the relationship between the ratings of children’s quality of life made by parents and children. The accuracy of proxy ratings varies with the specific domains of quality of life considered. There is greater agreement for observable physical functioning, and less for non-observable dimensions (e.g. emotional or social aspects). Agreement is also better between parents and chronically sick children compared with parents and their healthy children, possibly due to greater communication about illness and treatment. However, the importance of agreement and the common assumption that information from proxies should “match”

that provided by children is questioned as proxies and their children may not agree about many issues.

Nevertheless, parents do influence treatment choices, for example the motivation for orthodontic treatment often comes from parents, thus parental information can complement that from children (Stricker, 1970). Involving parents in the assessment of their child’s quality of life can also provide an opportunity to raise awareness in parents/caregivers (Inglehart *et al.*, 2002).

The Child Oral Health Quality of Life (COHQoL) questionnaire consists of a Parental-Caregiver Perceptions Questionnaire (PPQ) and Child Perceptions Questionnaires (CPQ) for children aged 8 to 10 years and 11 to 14 years. The measures were developed in response to a lack of a child-specific oral health related quality of life questionnaire (Jokovic *et al.*, 2002). The PPQ was designed to be analogous to the CPQ, to enable it to be used to complement the information gained from the CPQ, and also so that agreement between child and parent could be investigated. The measures are generic and were designed to be used for a wide range of conditions including caries, malocclusions, clefts and other oro-facial anomalies. The COHQoL questionnaires were derived using the item-impact method to ensure the final questionnaires contained items of the most relevance to children with these conditions and their parents (Guyatt *et al.*, 1986).

The PPQ includes 31 questions covering the four domains of oral symptoms (six questions), functional limitations (eight questions), emotion well-being (seven questions) and social well-being (ten questions) and an

additional fourteen questions on the impact on the family, the Family Impact Scale (FIS). The respondents are asked to indicate using a six-point Likert scale ('never'=0, 'once or twice'=1, 'sometimes'=2, 'often'=3, 'everyday or almost everyday'=4, and 'don't know') the frequency the events affected their children in the past three months. The PPQ contains a 'don't know' (DK) response specifically because the authors were aware of the limited knowledge a parent may have of their child's activities and feelings. DK response categories are used in other questionnaires to reassure respondents that it is acceptable not to know the answer as well as to minimise guessing (Bowling, 1997). The proportion of participants with at least one DK response was regarded as an essential characteristic of a parent's perception of their child oral health-related quality of life (Jokovic *et al.*, 2003).

The PPQ also includes two global ratings: parent's global ratings of the child's oral health (Atchison and Gift, 1997; Brook and Shaw, 1989) and the extent to which the oral/oro-facial condition affects his/her life overall. They are worded, respectively, as follows: "How would you rate the health of your child's teeth, lips, jaws and mouth?" with a 5-point response format ranging from 'excellent' to 'poor' and "How much is your child's overall well-being affected by the condition of his/her teeth, lips, jaws or mouth affect your life overall?" with a response range from 'not at all' to 'very much'. These ratings did not include a DK response.

The reliability and validity of the PPQ in Canada was assessed in 123 parents of children recruited at oro-facial, paediatric dentistry and orthodontic clinics (Jokovic *et al.*, 2002). The internal consistency and test-retest reliability of the scale and subscales were excellent. PPQ scale scores correlated with global ratings of oral health ($p < 0.05$) and overall well-being ($p < 0.01$) and a significant correlation between overall scale scores and the number of decayed tooth surfaces ($p < 0.01$) was demonstrated in the paediatric dentistry group. In the validation of the PPQ in Canada, only the questionnaires with no DK responses were included in the analyses. However, when DK responses were adjusted (by summing the response codes to all items and dividing this sum by the number of items for which a valid response was obtained), equally good validity was found. Several other methods for managing DK responses have subsequently been tested (Jokovic *et al.*, 2004). The different methods for handling DKs did not affect the properties of the PPQ.

The PPQ has not been validated for use in the UK, hence this study aimed to assess the reliability and validity in this setting, and to investigate whether different approaches to the treatment of DK responses had any effect on these psychometric properties.

Method

The evaluation of the PPQ was carried out at the same time as the validation of the CPQ. Parents of a consecutive sample of children attending for an examination at the orthodontic and paediatric dentistry clinics at a teaching dental hospital and one general dental practice were invited to participate. The parents were approached by the clinicians (MS, HR and CM) and consent gained.

A total sample size of 90 (30 parents from each of the three settings) was chosen based on the Canadian CPQ data but allowing for possible cultural differences. In the Canadian study, a Spearman's rank correlation coefficient of 0.54 between the number of decayed teeth and the CPQ scores was found (Jokovic *et al.*, 2002). A correlation coefficient of this magnitude would require a sample of 22 to be significant at an alpha of 0.01.

The PPQ is a self-administered questionnaire and parents were asked to complete it at the time of their child's visit to the clinics. The questionnaire contained 45 items with two global ratings as described previously, and invited parents to take part in the study to assess its test-retest reliability. The follow-up questionnaire included an additional question that asked parents if either the oral/oro-facial condition or its impact on the child's well-being had changed since recruitment. A reference period of three months was used for both administrations of the questionnaire.

The clinical status of the child with respect to dental caries and treatment experience, malocclusion, gingival health and the presence of enamel defects were collected by the calibrated clinicians (MS, HR, CM). Dental caries and treatment experience was assessed by enumerating the number of decayed, missing or filled teeth (DMFT) due to caries (Pine *et al.*, 1997). Malocclusion was categorised by using the dental health component of the Index of Orthodontic Treatment Need (IOTN) (Brook and Shaw, 1989).

The project was approved by the South Sheffield Research Ethical Committee.

Data analysis

Four approaches to analysis of DK responses were employed:

1. Exclusion, only the data from the parents who had not used the DK response were analysed.
- Data from all respondents were used in the other three methods for DK response adjustment.
2. Item mean, involved replacing DK responses with the item mean for the entire sample.
 3. Mean items answered, involved imputation of the mean score for the items answered.
 4. Replacement, involved replacing DK responses with zero value.

Missing values were replaced with the item mean for the entire sample but if participants failed to complete more than one-seventh of the questions they were excluded from the analysis. A similar threshold for excluding missing values has been adopted in other oral health related quality of life research (Slade, 1997).

After taking account of missing or DK responses, the total PPQ score for each participant was calculated by summing the item codes. A second summary measure 'number of impacts' for each participant recorded the number of impacts reported 'often' or 'everyday or almost everyday'. Subscale scores and the FIS score were calculated by summing the codes within these domains

Internal consistency was assessed by means of Cronbach's alpha, and test-retest reliability by means of intraclass correlation coefficient (ICC). The ICC was calculated based on data from parents who participated in the retest study and who did not report that their child's oral health and/or its impact on their life overall had changed between the two administrations of the questionnaire. This approach was taken to remove the effect of improvements or worsening of oral health in the intervening period between administration of the questionnaire as the test-retest reliability was the property of interest and not the responsiveness of the PPQ to change. This method is consistent with that taken during the development of the COHQoL (Jokovic *et al.*, 2002).

The feasibility of measuring a parent's perception of their child's oral health-related quality of life was assessed by examining the number and distribution of the DK responses as used in parental health related quality of life measures (Varni *et al.*, 1999). The face and content validity were assessed by examining the wording of the questionnaire and the number of missing responses to items. Construct validity was assessed by testing associations of the PPQ scale, subscale scores and the FIS with the life overall scores and the children's clinical data. Criterion validity was examined by comparing the parent's global rating of their child's oral health to summary measures of PPQ and the subscale/FIS score.

Finally, the reliability and validity of the four approaches to the treatment of the DK data were compared.

Results

Ninety-one children participated in the study. No parents refused to participate or to complete the questionnaire. Four participants were excluded due to excessive missing data. Mothers completed 62 (71.3%) of the 87 usable PPQs. The DK responses adjusted analyses therefore included data from 87 parents. Using the exclusion method, data from 61 parents were analysed after excluding all those respondents who provided one or more DK responses. Respondents providing DK responses were evenly distributed among the three settings and between mothers and fathers.

Overall, the mean DMFT was 1.23 with a mean number of decayed teeth of 0.41, a mean number of missing teeth due to caries of 0.06, a mean number of filled teeth of 0.77 and a mean number of teeth missing for any reason of 0.41. The mean DMFT for 12 year old children in the UK was 0.86 (DT=0.39, MT=0.06, FT=0.41) in 2000/1 (Pitts *et al.*, 2002).

Sixty eight percent of children had an IOTN score of less than 4.64% had good gingival health and 80% did not have enamel opacities. From the UK National Child Dental Health survey 2003, 65% had an IOTN score less than 4.35% had no gingival inflammation and 66% did not have opacities (Lader *et al.*, 2004).

The feasibility of using the PPQ was indicated by the frequency and distribution of the DK responses. In total, 1.7% (n=71) of responses were DK. Four questions accounted for over 50% of these responses, two of which belonged to the symptoms subscale and enquired about the child having food stuck in the roof of the mouth and

having food caught in or between the teeth. The third question was concerned with whether their child breathed through the mouth (functional limitation subscale). The final question that elicited high numbers of DK responses enquired about whether their child had been asked questions by other children about their teeth, lips, mouth or jaws (social well-being subscale).

The total score and subscales of the PPQ using the four different approaches to analysis of DK responses are summarised in Table 1. Of the subscales, the highest mean scores were in the symptoms domain.

In the global ratings 20% of parents rated their child's oral health as fair/poor and 11% reported levels of impact on life overall of a lot or very much.

Table 2 summarises the internal consistency derived from the four analytical approaches. The internal consistencies of the total scale derived by each method were acceptable, but the exclusion and item mean approach had sub-optimal reliability (<0.60) for one subscale each.

Most parents completed a follow up questionnaire after two weeks. In the DK exclusion approach, 56% of the parents reported their child's oral health to be unchanged and the ICC was 0.69. In the adjusted approaches, 52% parents could be included in the test-retest analysis with an ICC of 0.92-0.95.

The construct validity assessments are summarised in Table 3, which describes the relationships between life overall rating, clinical data and the measures of PPQ for all four approaches.

For the DK exclusion approach, ratings of life overall were related to both summary measures of PPQ, the emotional and social subscales and the FIS. The number of decayed teeth and the functional subscale of the PPQ were associated, but no other relationships were apparent between the PPQ and clinical variables.

With the item mean approach, global ratings of life overall were related to all PPQ measures except the functional subscale. Significant correlations were found between the 'number of impacts' and the number of decayed teeth and the number of DMFT.

Similarly, the mean items answered approach showed global ratings of life overall to be related to all PPQ measures except the functional subscale. With this third approach, significant correlations were found between the 'number of impacts' and the number of DMFT and also between the emotional subscale and the number of decayed teeth and number of DMFT.

When DK responses were replaced by zero, global ratings of life overall were related to all PPQ measures. A significant correlation was found only between the 'number of impacts' and the number of DMFT.

Table 4 summarises the significant relationships in construct validity assessments using the four approaches.

Criterion validity was examined by comparing the PPQ scores and the global oral health rating (Table 5). In the DK exclusion analysis, the 'number of impacts' and the FIS score were associated with the global rating. In the three DK-adjusted approaches to the handling of DKs, correlation between the summary measures and subscale scores and the global oral health rating were found, but varied between approaches.

Table 1. Mean scores for the subscales of Parental-Caregiver Perceptions Questionnaire and Family Impact Scale (FIS)

<i>Scores</i>	<i>Range</i>	<i>Exclusion mean (SD)</i>	<i>Item mean mean (SD)</i>	<i>Mean items answered mean (SD)</i>	<i>Replacement Mean (SD)</i>
Total score	0-180	13.5 (12.5)	14.2 (14.5)	14.3 (14.5)	14.2 (14.5)
Symptoms	0-24	3.9 (3.1)	4.1 (3.2)	4.1 (3.3)	4.1 (3.3)
Functional	0-32	2.8 (3.7)	2.8 (3.4)	2.8 (3.4)	2.8 (3.4)
Emotional	0-28	2.4 (3.3)	2.5 (3.6)	2.5 (3.6)	2.9 (4.1)
Social	0-40	1.3 (2.0)	1.5 (2.7)	1.8 (3.0)	1.7 (3.0)
FIS	0-56	2.9 (3.8)	3.3 (4.6)	2.7 (4.1)	2.7 (4.1)

Table 2. Reliability of the total scale, subscales and Family Impact Scale (FIS)

	<i>No. of items</i>	<i>Exclusion</i>	<i>Item mean</i>	<i>Mean items answered</i>	<i>Replacement</i>
		<i>Cronbach's alpha</i>	<i>Cronbach's alpha</i>	<i>Cronbach's alpha</i>	<i>Cronbach's alpha</i>
Total scale	45	0.86	0.89	0.92	0.93
Symptoms subscale	6	0.68	0.65	0.71	0.69
Functional subscale	8	0.72	0.52	0.67	0.68
Emotional subscale	7	0.85	0.83	0.85	0.85
Social subscale	10	0.41	0.77	0.79	0.79
FIS	14	0.77	0.87	0.82	0.82

Discussion

This study aimed to assess the reliability and validity of the PPQ for use in the UK. Four approaches tested whether the treatment of DK responses affected the properties of this measure.

Overall, the internal consistency of the total scale and test-retest reliability were acceptable, but the internal consistency (as measured by Cronbach's α) varied between the differing methods of handling the DK responses. There was also variation in the values for the subscales with the mean items answered and replacement method having superior internal consistency (Table 2). These findings contrast with those from Canada where the differing methods had no effect on internal consistency (Jokovic *et al.*, 2004).

Validity was assessed in several ways including looking for relationships between the PPQ scores and clinical data. Significant relationships were found with caries experience data but not with malocclusion, enamel opacities or gingivitis. Other studies have shown a tenuous link between oral health related quality of life measures and clinical data (Cushing *et al.*, 1986; Locker and Slade, 1994).

All adjusted approaches were associated with acceptable validity with only minor variations between approaches.

The exclusion of the DK responses had a detrimental affect on both construct and criterion validity of the measure. The exclusion approach yielded lower values for rank correlations than the other approaches. Again, other studies have not found differences between the different approaches (Jokovic *et al.*, 2002; Jokovic *et al.*, 2004). The difference found in our small study may

be due to the reduction in sample size or that the DK response plays an important contribution to the validity of this measure.

Another explanation for the poorer validity with the DK exclusion approach may be that respondents who use DK responses systematically differ from those respondents that don't choose them. In other research the use of DK responses in questionnaires was unrelated to gender, age or social status of the respondent (Ziller and Long, 1965). In this study, DK responses were evenly distributed across the three settings and between mothers and fathers, but we were unable to establish whether there was a relationship between the social status of the participants and the use of DK responses.

Generally, oral health-related quality of life measures do not include DK responses. DK responses can be offered for each question of the Oral Health Impact Profile, but are rarely used. For analysis purposes they are entered as missing values and if more than nine responses were missing or DK the questionnaire is excluded (total number of items=49) (Slade, 1997). When using the Oral Impacts on Daily Performance, missing or DK responses, are adjusted, but respondents with more than two missing items are excluded (Adulyanon and Sheiham, 1997).

The results of the evaluation of the PPQ suggest that if DK responses are included but handled by adjustment, the reliability and validity of this measure are acceptable for use in the UK. As there were only minor differences between the adjusted approaches with respect to validity, from the perspective of reliability and validity, the replacement approach may be marginally the most appropriate method. This method involves replacing DK responses with a zero value and had the highest internal consistency.

Table 3. Relationship between life overall ratings, clinical data and Parental-Caregiver Perceptions Questionnaire (PPQ) scores

A) Exclusion approach

	Total PPQ	No. of impacts	Symptom Subscale	Functional subscale	Emotional subscale	Social subscale	Family impact
	r_s	r_s	r_s	r_s	r_s	r_s	r_s
Life overall	0.33*	0.26*	0.06	0.13	0.30*	0.30*	0.41*
IOTN	0.21	0.08	0.18	0.08	0.21	0.18	0.23
Total missing teeth	0.05	0.14	0.10	0.10	0.04	0.06	0.10
Decayed	0.11	0.01	0.23	0.32*	0.07	0.00	0.10
DMFT	0.06	0.14	0.11	0.20	0.02	0.02	0.03
	Total PPQ (p-value)	No. of impacts (p-value)	Symptom Subscale (p-value)	Functional subscale (p-value)	Emotional subscale (p-value)	Social subscale (p-value)	Family impact (p-value)
Opacities present	0.9	0.51	0.42	0.98	0.83	0.89	0.90
Gingivitis present	0.73	0.97	0.82	0.96	0.76	0.56	0.57

B) Item mean approach

	Total PPQ	No. of impacts	Symptom Subscale	Functional subscale	Emotional subscale	Social subscale	Family impact
	r_s	r_s	r_s	r_s	r_s	r_s	r_s
Life overall	0.40**	0.30**	0.33**	0.20	0.41**	0.31**	0.42**
IOTN	0.12	0.02	0.09	0.03	0.15	0.07	0.08
Total missing teeth	0.02	0.06	0.05	0.09	0.05	0.04	0.09
Decayed	0.08	0.21*	0.01	0.03	0.20	0.14	0.09
DMFT	0.15	0.28**	0.07	0.20	0.22*	0.14	0.16
	Total PPQ (p-value)	No. of impacts (p-value)	Symptom Subscale (p-value)	Functional subscale (p-value)	Emotional subscale (p-value)	Social subscale (p-value)	Family impact (p-value)
Opacities present	0.33	0.2	0.30	0.53	0.94	0.59	0.57
Gingivitis present	0.53	0.57	0.53	0.46	0.40	0.99	0.61

C) Mean items answered approach

	Total PPQ	No. of impacts	Symptom Subscale	Functional subscale	Emotional subscale	Social subscale	Family impact
	r_s	r_s	r_s	r_s	r_s	r_s	r_s
Life overall	0.40**	0.27*	0.34**	0.20	0.41**	0.36**	0.41*
IOTN	0.12	0.01	0.09	0.03	0.15	0.09	0.70
Total missing teeth	0.01	0.07	0.02	0.06	0.05	0.02	0.07
Decayed	0.08	0.17	0.02	0.15	0.22*	0.08	0.15
DMFT	0.14	0.25*	0.08	0.03	0.22*	0.09	0.19
	Total PPQ (p-value)	No. of impacts (p-value)	Symptom Subscale (p-value)	Functional subscale (p-value)	Emotional subscale (p-value)	Social subscale (p-value)	Family impact (p-value)
Opacities present	0.25	0.11	0.22	0.31	0.86	0.51	0.40
Gingivitis present	0.45	0.74	0.49	0.37	0.51	0.85	0.55

Table 3. Continued overleaf...

Table 3. Continued...

	Total PPQ	No. of impacts	Symptom Subscale	Functional subscale	Emotional subscale	Social subscale	Family impact
	r_s	r_s	r_s	r_s	r_s	r_s	r_s
Life overall	0.40**	0.27*	0.34**	0.22*	0.41**	0.35**	0.41**
IOTN	0.11	0.01	0.09	0.02	0.15	0.07	0.06
Total missing teeth	0.02	0.07	0.02	0.05	0.04	0.02	0.07
Decayed	0.09	0.17	0.01	0.13	0.18	0.11	0.15
DMFT	0.15	0.25*	0.08	0.01	0.20	0.14	0.19
	Total PPQ (p-value)	No. of impacts (p-value)	Symptom Subscale (p-value)	Functional subscale (p-value)	Emotional subscale (p-value)	Social subscale (p-value)	Family impact (p-value)
Opacities present	0.25	0.11	0.22	0.31	0.86	0.51	0.40
Gingivitis present	0.45	0.74	0.49	0.37	0.51	0.85	0.55

r_s = Spearman's rank correlation coefficient, *statistically significant, $p < 0.05$, **statistically significant, $p < 0.01$, p-values obtained from Mann Whitney U test

Table 4. Relationships from construct validity analyses using four analytical approaches for Parental-Caregiver Perceptions Questionnaire (PPQ) data

	Exclusion	Item mean	Mean items answered	Replacement
Life overall with total PPQ	*	*	*	*
Life overall with 'number of impacts'	*	*	*	*
Life overall with symptoms subscale		*	*	*
Life overall with emotional subscale	*	*	*	*
Life overall with social subscale	*	*	*	*
Life overall with functional subscale		*		*
Life overall with Family Impact Scale	*		*	*
No. of decayed teeth with functional subscale	*			
No. of decayed teeth with emotional subscale		*	*	
No. of decayed teeth with often /everyday		*		
DMFT with number of impacts		*	*	*
DMFT with emotional subscale			*	

*statistically significant relationship, $p < 0.05$

Table 5. Rank correlations between Parental-Caregiver Perceptions Questionnaire (PPQ) scores and global measure

	Exclusion	Item mean	Mean items answered	Replacement
	r_s	r_s	r_s	r_s
Total PPQ	0.23	0.25*	0.24*	0.23*
No. of impacts	0.31*	0.20	0.25*	0.25*
Symptoms subscale	0.20	0.24*	0.30**	0.30**
Functional subscale	0.21	0.26*	0.18	0.18
Emotional subscale	0.02	0.06	0.07	0.09
Social subscale	0.07	0.09	0.11	0.10
FIS	0.27*	0.27*	0.27*	0.21*

*statistically significant, $p < 0.05$, **statistically significant, $p < 0.01$

The feasibility of using parents to supplement information given by children is confirmed by the low number of DK responses. However, an examination of the items that resulted in the highest number of DK responses demonstrates that parents are often not able to detect some unobservable impacts of oral conditions such as 'getting food stuck in the roof of the mouth'. The greater ability of parents to rate their child's health-related quality of life for observable functioning is consistent with the findings of a systematic review (Eiser and Morse, 2001). The Canadian validation of PPQ had similar findings regarding the questions that elicited the highest number of DK responses (Jokovic *et al.*, 2004). The authors of the PPQ considered removing these items but decided against it for fear of compromising the extent to which all aspects of oral health related quality of life are comprehensively covered by the PPQ.

The findings from this study may be of interest when considering the approach to the analysis of parental perceptions of their child's health related quality of life in the future.

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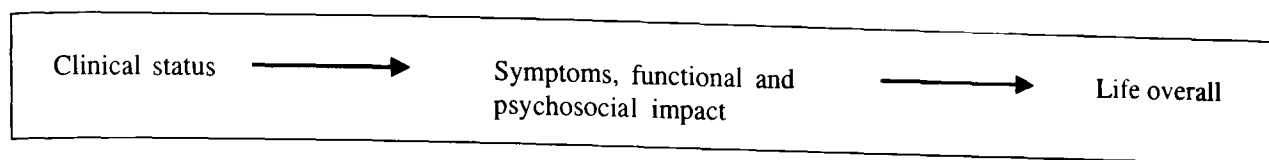


Figure 1. Simplified Wilson and Cleary Model applied to oral diseases and disorders (Locker, et al., 2001)

correlated with global ratings of oral health ($r_s = 0.23$; $p < 0.05$) and overall well-being ($r_s = 0.40$; $p < 0.001$) and the paedodontic group showed a significant correlation between overall scale scores and the number of decayed tooth surfaces ($r_s = 0.64$; $p < 0.01$). This method of assessment of validity was based on a model of disease and its consequences proposed by Wilson and Cleary (1995). This model is linear with biological, physiological and clinical variables at one end and overall quality of life at the other. A link between them is represented by symptoms of disease/disorder and functional, psychological and social experiences of the individual related to that disease/disorder. A simple form of this model as it applies to oral health is shown in Figure 1 (Locker *et al.*, 2001).

The need to test the psychometric properties of instruments such as those for measuring OHRQoL in a new environment have been stressed (Robinson *et al.*, 2003a; Bowling, 1997). The linguistic and cultural context in which a measure is used can have a bearing on the validity, as can the intended purpose of the measure. It was hoped that the questionnaire could be used to compare impacts perceived by children with clinical information on their oral conditions particularly with respect to enamel defects and dental caries. The CPQ₁₁₋₁₄ questionnaire has not been validated for use in the UK or for use in children with varying levels of disease. Therefore this study aimed to assess the reliability and validity of the CPQ₁₁₋₁₄ for use in the UK child population.

Method

As the CPQ₁₁₋₁₄ was designed for use with children with oral and oro-facial conditions, a consecutive sample of children between 11 and 14 years of age attending for an examination at the orthodontic and paediatric dentistry clinics at a teaching dental hospital and one general dental practice were invited to take part. These settings were similar to those used for the evaluation of the questionnaire in the Canadian study (Jokovic *et al.*, 2002).

The individuals were approached by the clinicians (MS, HR and CM) and consent gained. While the study sample were all children visiting a dentist they ranged from relative health to a variety of oral conditions including dental caries, malocclusion, gingivitis and enamel opacities.

Data were collected by asking children to complete the CPQ₁₁₋₁₄ at the time of their visits to the clinics. The questionnaire contained 37 items and two global ratings as described previously, and invited children to take part in the study to assess its test-retest reliability. The follow-up questionnaire two weeks later asked if either the oral/oro-facial condition or its impact on the child's well-being had changed since recruitment. Data on ethnicity, age, gender, socio-economic status were also

obtained, as these variables may confound or mediate relationships between clinical status and OHRQoL.

The sample size calculation was based on caries data from the Canadian study as caries was the most common clinical condition for which data were available. A Spearman's rank correlation coefficient of 0.64 was found between the number of decayed teeth and the CPQ₁₁₋₁₄ scores (Jokovic *et al.*, 2002) requiring a total sample of 22 to be significant at an alpha of 0.01. A sample of 30 children from each of the three settings (i.e. 90 in total) was chosen to allow for possible cultural differences between the UK and Canada.

Clinical variables were collected by the calibrated clinicians (MS, HR, CM). Caries status was assessed by enumerating the number of decayed teeth (from clinical examination at the D3 threshold) missing or filled teeth due to caries (Pine *et al.*, 1997). Malocclusion was categorised by using the dental health component of the Index of Orthodontic Treatment Need (Brook and Shaw, 1989). The presence of dental opacities on anterior teeth was recorded as present or absent.

The project was approved by the South Sheffield Research Ethical Committee. Written consent was obtained from the parents of all participants

Data analysis

Where children had failed to indicate a score for a item, such missing values were dealt with in two ways: those participants who failed to complete more than one-seventh of the questions were excluded from the analysis. A similar threshold for excluding missing values has been adopted in other oral health-related quality of life research (Slade, 1997a). Missing CPQ₁₁₋₁₄ values from the remaining participants were replaced with the sample mean score for that item. No analytical strategy to deal with missing values is described in the Canadian study.

The total CPQ₁₁₋₁₄ score for each participant was calculated by summing the item codes. A second summary measure for each participant, the number of impacts, recorded the number of impacts reported 'often' or 'everyday or almost everyday'. Subscale scores were calculated by summing the codes for questions within the four health domains.

Internal consistency was assessed by means of Cronbach's alpha and test-retest reliability by means of intraclass correlation coefficient (ICC). The latter was based on data from children who participated in the follow-up study and who did not report that their oral health and/or its impact on their life overall had changed between the two administrations of the questionnaire.

Construct validity was assessed by testing associations of the CPQ₁₁₋₁₄ scale and the subscale scores with the life overall scores and the clinical data.

Criterion validity was examined by comparing the global rating of oral health to CPQ₁₁₋₁₄.

Table 1. Caries experience of the sample (n=89)

	Mean	SD
Total number of missing teeth	0.41	0.97
Missing teeth due to caries	0.08	0.41
Filled teeth	0.78	2.12
Decayed teeth	0.39	1.20
Decayed, missing and filled teeth	1.25	2.75

Table 2. Mean scores for individual subscales of CPQ₁₁₋₁₄

Subscale scores	Mean	SD
Symptoms	5.35	3.16
Functional	5.42	3.75
Emotional	4.22	4.45
Social	3.08	3.09

Table 3. Reliability of the total scale and subscales

	No. of items	Cronbach's alpha
Total scale	37	0.87
Symptoms subscale	6	0.63
Functional subscale	9	0.59
Emotional subscale	9	0.83
Social subscale	13	0.65

Results

Ninety-one children aged 11 to 14 years were invited to participate in the study, no children refused or were unable to complete the questionnaire due to literacy problems. We report data from 89 children, as two were excluded due to excessive missing data.

The resultant sample comprised 29 children attending a paediatric dentistry clinic, 30 attending an orthodontic clinic and 30 attending a general dental practice. The mean age of participants was 12.4 years. There were 47 females. Eighty-three children were White British. Post-codes of the participants were used to gain an Index of Multiple Deprivation 2000 (IMD) score as a measure of socio-economic status; data were available for 74 participants. The sample was made up of children from areas in Sheffield with varying levels of deprivation.

The mean number of decayed, missing and filled teeth

was 1.25 (SD 2.75) with a mean total number of missing teeth for any reason of 0.41 (SD 0.97) (see Table 1). Fifteen percent of the sample had untreated decay present with 35% having a DMFT score of greater than zero.

Sixty-nine percent of children had an IOTN score of less than four. Sixty-four percent of children had good gingival health and 20% had enamel defects.

The questionnaire appeared to be of a suitable readability for children of this age to enquire about aspects of OHRQoL, and to cover a comprehensive range of oral impacts. The number of missing values were low with a mean of 0.21 per child. The final item about the frequency with which participants had been asked questions about their teeth, lips, jaws or mouth by other children had a considerable number of missing responses (n = 9). This high number of missing responses for the final item may be a symptom of the format of the questionnaire.

The mean total CPQ₁₁₋₁₄ score was 18.07 (SD 11.59). It ranged from three to 53. No floor or ceiling effects were seen for the total score. A mean number of impacts experienced 'often' or 'everyday or almost everyday' was 1.56 (SD 1.92). Table 2 shows the mean score for the individual subscales.

One-fifth of the children said the health of their teeth, lips and mouth (global oral health rating) was 'fair' or 'poor' in the prior three months. The condition of their teeth, lips, jaws or mouth reportedly affected 11.2% of the children's lives 'a lot' or 'very much' (life overall rating).

The Cronbach's alpha for the total scale was 0.87 and ranged from 0.59 to 0.83 for the subscales, indicating substantial to excellent internal consistency (Table 3). Seventy-nine percent of participants (n=70) indicated willingness to complete a second questionnaire two weeks later, 84% of whom complied (n=59). From the second questionnaires 73% (n= 43) reported the condition of their mouth was unchanged and thus were analysed for test-retest reliability. The intraclass correlation coefficient on repeated application of the measure was 0.83 (95% CI= 0.76-0.90) suggesting almost perfect agreement (Landis and Koch, 1977).

Table 4 summarises the relationships between the clinical data, summary measures of CPQ₁₁₋₁₄ and the life overall scores. Ratings of life overall were related to all summary measures of CPQ₁₁₋₁₄ (total CPQ₁₁₋₁₄ score, number of impacts and the subscale scores). The number of children with impacts 'often' or 'everyday' correlated

Table 4. Relationship between life overall ratings, clinical data and CPQ₁₁₋₁₄ scores

	Total CPQ ₁₁₋₁₄	No. often or everyday	Symptom subscale	Functional subscale	Emotional subscale	Social subscale
<i>Spearman's rank correlation coefficient</i>						
Life overall	0.40*	0.29*	0.28*	0.28*	0.29*	0.35*
IOTN	0.14	0.12	0.13	0.13	0.11	0.11
No. missing teeth	0.07	0.23*	-0.03	0.12	0.13	0.10
Missing due to caries	0.05	0.21*	0.08	0.11	0.11	0.01
Filled	0.09	0.10	-0.01	0.16	0.11	0.06
Decayed	0.10	0.17	0.09	0.11	0.08	0.11
DMFT	0.09	0.05	0.03	0.11	0.06	0.09
<i>p-values (Mann Whitney U test)</i>						
Opacity present	0.11	0.05	0.19	0.02*	0.53	0.14
Gingivitis	0.54	0.47	0.72	0.31	0.82	0.99

*p<0.05

Table 5. Rank correlations between CPQ₁₁₋₁₄ scores and global measure of oral health

	r_s
Total CPQ	0.28*
Total no. often or everyday	0.34*
Symptoms subscale	0.37*
Functional subscale	0.22*
Emotional subscale	0.18
Social subscale	0.19

(* $p < 0.05$)

with the total number of missing teeth and missing teeth due to caries. No relationships were apparent between IOTN scores or the presence of gingivitis and CPQ₁₁₋₁₄. Scores of the functional subscale of CPQ₁₁₋₁₄ were related to the presence of opacities (Table 4).

Summary measures of CPQ₁₁₋₁₄ correlated with the global oral health rating indicating acceptable criterion validity (Table 5).

Discussion

This study aimed to assess the reliability and validity of CPQ₁₁₋₁₄ for use in the UK. Overall, these data suggest the CPQ₁₁₋₁₄ has acceptable reliability with the internal consistency and test-retest reliability. Correlation coefficients, Cronbach's alphas and ICCs found in this study are equivalent to the results from Canada. Overall the criterion validity of the summary measures of CPQ₁₁₋₁₄ was acceptable although the correlations between the emotional and the social subscale were not statistically significant. Construct validity was acceptable in relation to personal assessment of life overall but sporadic and inconsistent for measures of clinical status.

Other studies have shown a tenuous link between patient-based measures and clinical indicators (Cushing *et al.*, 1986; Locker and Slade, 1994). Three principle explanations may account for the weak relationships between CPQ₁₁₋₁₄ and clinical data; there were low disease levels in the sample, the conditions under investigation may have caused immeasurably low levels of impact or that the impacts were mediated by a variety of factors such as culture and deprivation.

Cushing *et al.* (1986) found that DMFT data were not well correlated with measures of impact of dental disease as experienced by individuals. A review of validation studies of OHRQoL measures found only three studies using caries data (Jokovic *et al.*, 2002; Robinson *et al.*, 2003; Slade, 1997b). Despite the setting for the evaluation being similar to that in the Canadian study only fifteen percent of children in this sample had untreated caries compared to 53%. Mean DMFT levels were similarly lower therefore the degree of impact at these levels of disease on children may be too low for the CPQ₁₁₋₁₄ to detect.

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However, the mean DMFT of the sample, at 1.25, was higher than the mean DMFT of 0.79 found for children in this region (Pitts *et al.*, 2002). For this reason, the CPQ₁₁₋₁₄ may be unsuitable for discriminating between children at the low caries levels seen in UK population-based research.

The CPQ₁₁₋₁₄ was developed for use in evaluative research with children with oral disease and dental treatment needs. However, the findings from this study suggest that it may not be suitable for discriminating between groups in cross-sectional research in low disease populations. Further evaluation of CPQ₁₁₋₁₄ is warranted if it is to be used for this purpose in the UK.

Only the functional subscale score was associated with the presence of enamel opacities. This finding is surprising as opacities seem unlikely to affect functional abilities in comparison to social factors. It is possible that this result is due to type 1 error.

The findings from the caries and opacity data are surprising given that CPQ₁₁₋₁₄ has also been evaluated in a cross-sectional study of Ugandan children, many of whom had lower caries levels but evidence of fluorosis (Robinson *et al.*, 2003b). Despite using a back-translated version, the CPQ₁₁₋₁₄ successfully discriminated between children with and without the consequences of dental caries and/or dental fluorosis.

This study found no relationship between the dental health component of the IOTN and CPQ₁₁₋₁₄ scores. This relationship was not examined in the Canadian study. The use of OHRQoL measures in orthodontics has been considered but has been found to be problematic (Fox *et al.*, 1999). One difficulty may be that IOTN emphasises malocclusions that may not be important to quality of life (such as posterior cross bites). More research is required in this area.

Finally, the inconsistencies found in the relationship between the clinical data and the CPQ₁₁₋₁₄ scores may not be due to the psychometric properties of the measure but due to the fact that impacts are mediated by other factors. It has been suggested that culture, social and material deprivation can influence the extent of the impact of disease (Locker, 1992). Variables such as general health status, household income and life stress have been shown to explain as much variance in the impact of oral disorders on adults as clinical indicators such as missing teeth (Locker and Slade, 1994). We have not been able to account for these variables in analyzing the data obtained in the study.

In conclusion, the CPQ₁₁₋₁₄ shows good reliability, criterion validity and construct validity in relation to life overall but poor construct validity in relation to clinical variables in this relatively low disease population. If this measure is to be used in similar settings in the UK involving children with low levels of caries a large sample size will be required.

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