The impact of dental caries and its treatment under general anaesthetic on the everyday lives of children and their families

A thesis submitted in the fulfilment of the requirement for the Degree of Doctor of Philosophy

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Abstract

Objectives: To assess the impact of dental caries and treatment under general anaesthetic (GA) on the everyday lives of children and their families, using measures of quality of life (QoL) and oral health-related quality of life (OHRQoL).

Methods: Participants, aged 5-16 years old requiring treatment for caries under GA, were recruited. OHRQoL was measured before and three months after treatment using the Caries Impacts and Experiences Questionnaire for Children (CARIES-QC). Overall QoL was measured using the Child Health Utility 9D (CHU9D). Parents/caregivers completed the Family Impact Scale (FIS). Change in scores after treatment were analysed using Wilcoxon tests. Path analysis was conducted to investigate the relationships between clinical, individual and environmental factors and QoL outcomes, guided by a theoretical model.

Results: In total, 85 parent-child dyads completed the study. Three-quarters (76%) of children were living in the most deprived areas of England. There was a statistically significant improvement in OHRQoL (mean interval score difference in CARIES-QC=4.43, $p<0.001$) and QoL (mean score difference in CHU9D=2.48, $p<0.001$) following treatment, with moderate to large effect sizes. Path analyses revealed that 47% of the variance in OHRQoL scores was accounted for by the variables in the model. There were significant relationships between change in OHRQoL score and treatment type [extraction only vs. comprehensive care ($\beta=1.41$, $p=0.07$)] and number of extractions ($\beta=0.46$, $p<0.001$). There was statistically significant improvement in FIS scores following treatment (mean score difference= 5.48, $p=0.03$). Overall, 95% of parents felt their child's dental health had improved, and 74% reported improvement in their child's QoL.

Conclusion: Treatment under GA was associated with significant improvement in QoL and OHRQoL as reported by both children and their parents. Path analysis suggests that treatment type, via number of extractions, may impact on child OHRQoL and QoL following treatment under GA. Increased number of extractions was associated with worse OHRQoL and QoL. The results could have implications for treatment planning and the provision and commissioning of services.

Publications, presentations and awards

The following are a list to date of the research outputs arising from the work in this thesis.

Publications

Knapp R., Marshman Z., Rodd H., Gilchrist F., Zaitoun, H. (2018). Impact on Children of Dental Caries and Treatment Under GA [Conference abstract]. *Journal of Dental Research* 97, Special Issue B. Published online https://iadr.abstractarchives.com/abstract/18iags-2956425/impact-on-children-of-dental-caries-and-treatment-under-ga. Last accessed 08/09/2019.

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Frequently used abbreviations

BME	Black and Minority Ethnic group
BSPD	British Society of Paediatric Dentistry
CARIES-QC	Caries Impacts and Experiences Questionnaire for Children
CC	Combination Care for the treatment of dental caries. This included children having treatment on clinic followed by exodontia under general anaesthetic, and children receiving comprehensive care under general anaesthetic.
CCDH	Charles Clifford Dental Hospital
CHU9D	Child Health Utility 9D
CIS	CARIES-QC interval scale
dmft	Decayed, missing and filled primary teeth
DMFT	Decayed, missing and filled permanent teeth
EXO	Treatment for caries on clinic followed by exodontia under general anaesthesia
FG	Fiona Gilchrist, Senior Lecturer/Honorary Consultant in Paediatric Dentistry
GA	General anaesthetic
GTJ	Global Transition Judgement
HR	Helen Rodd, Professor of Paediatric Dentistry
HRQoL	Health-related quality of life
IMD	Index of Multiple Deprivation
IVS	Intravenous Sedation
MID	Minimally Important Difference
OHRQoL	Oral health-related quality of life
PICOS	Population, intervention, comparator, outcome and study design
PMC	Preformed Metal Crown
PROM	Patient Reported Outcome Measure
QATSDD	Quality Assessment Tool for Studies of Diverse Design
QoL	Quality of Life
RK	Rebecca Knapp, PhD student
UK	United Kingdom
WHO	World Health Organisation
ZM	Zoe Marshman, Professor in Dental Public Health

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Chapter 1: Introduction

1.1 Overview

Dental caries remains the most prevalent chronic disease worldwide and, despite being largely preventable, presents a significant global public health problem (Kassebaum et al., 2017). In the UK, the most recent child dental health survey of 5-year-old children reported 31% had 'obvious decay experience' in their primary teeth, which rose to nearly half of those surveyed by age 8-years (Steele et al., 2015). Dental caries impacts significantly on children and their families and, ultimately, many paediatric patients with dental caries require treatment under general anaesthetic (GA). In England alone, in 2017-18, approximately 42,000 children were admitted to hospital with a primary diagnosis of dental caries, many of whom will have then received extractions under GA (NHS Digital, 2018).

While the impacts of dental caries are well documented, what is less well understood is the subjective experience of children themselves. A number of instruments have been developed to investigate the subjective impact of oral diseases, seeking to measure oral health-related quality of life (OHRQoL). OHRQoL is defined as '*the impact of oral disease and disorders on aspects of everyday life that a patient or person values, that are of sufficient magnitude, in terms of frequency, severity or duration to affect their experience and perception of their life overall*' (Locker and Allen, 2007). Several instruments have been used in dental research to measure children's OHRQoL but, to date,

there has been little research carried out using child-completed questionnaires, with most studies relying on proxy-reported measures.

Furthermore, although several studies have looked at the impact of dental treatment under GA, none have previously assessed the impact of different treatment approaches under GA from the child's perspective. This research, therefore, contributes to the field as it is the first study to employ a disease-specific, child-reported measure to examine changes in OHRQoL following dental treatment for caries under GA, as well as examining which factors, especially treatment approach (extraction only or combination care involving restorations and extractions), have an impact on child reported quality of life outcomes.

1.2 Aims and objectives

The aim of this research is to examine the impact of dental caries and its treatment under GA on the everyday lives of children and their families. The specific objectives of the research are to:

1. Conduct a systematic review of the current literature on the effect of dental treatment for caries under GA on children's OHRQoL.
2. Investigate the impact of dental caries and its treatment under GA on children's everyday lives, using a child-reported measure of OHRQoL.
3. Investigate the impact of dental caries and its treatment under GA on the families of these children.
4. Examine the relationships between individual, clinical and environmental factors on children's OHRQoL and QoL, with respect to treatment for caries under GA, using path analysis.

1.3 Thesis structure

This report is structured as follows:

- **Chapter 2** is a narrative review of the current literature, which highlights the public health significance of dental caries, describes the treatment of dental caries under GA and considers methods of evaluating health outcomes from a patient perspective. Gaps in the current literature are identified, including the lack of information from a child's perspective on the impact of dental caries and interventions for its management. There is also a paucity of information on the impact of different treatment approaches under GA.
- **Chapter 3** is a systematic review that was undertaken of previous studies that have reported changes in child OHRQoL following treatment for dental caries under GA.
- **Chapter 4** describes the methods for the main study, which seeks to investigate the impact of dental caries and its treatment under GA on children's and their families.
- **Chapter 5** presents the results of this study, including descriptive and statistical analyses of the findings.
- **Chapter 6** discusses the research findings, reflects on the study design and highlights the implications of these findings for patient care, policy and further research.
- **Chapter 7** presents the main conclusions and recommendations arising from this research.

Chapter 2: Literature review

2.1 Introduction

Dental caries remains the most prevalent chronic disease worldwide, and the tenth most common condition in children, despite being largely preventable (Kassebaum et al., 2017). In the UK, approximately one third of five-year-old children have experience of caries, rising to nearly a half by the age of 8-years (Steele et al., 2015). Although the prevalence of dental caries in the UK is declining, there appear to be growing inequalities, with children from lower socioeconomic groups more likely to be affected than children from higher socioeconomic groups (Tsakos et al., 2015).

Dental caries is the most common reason for a child to be admitted to hospital for a general anaesthetic (GA) in the UK. However, such treatment is not without risk to the child, and it represents a significant financial cost to the National Health Service (NHS), estimated at £30 million annually (Faculty of Dental Surgery , 2015). The numbers of children being admitted for dental treatment under GA are growing, with approximately 42,000 children under 16-years being admitted in 2017-8 in England alone (NHS Digital, 2018). The treatment approach for the management of caries under GA is highly variable, depending on the available services and workforce in any geographical area. In some instances, exodontia (extraction) only is offered, while other providers may offer comprehensive care (a combination of extractions and restorative treatment) where appropriate.

Dental caries is associated with a number of negative impacts which have been shown to significantly affect the daily lives of children, including pain, impaired function and difficulty sleeping (Alsumait et al., 2015; Baghdadi, 2015; Gilchrist et al., 2015). Oral health related quality of life (OHRQoL) measures have been used to assess the impact of dental caries on individuals and their families, and take into account the functional, emotional and social impacts of various dental and oro-facial conditions (Locker and Allen, 2007).

This literature review considers the public health significance of dental caries, treatment under GA and the impact of dental caries and its treatment on the everyday life of children. Gaps in the current literature are highlighted, in particular the lack of information from children's perspectives on the impact of dental caries and its related treatment options.

2.2 Dental caries as a public health problem

2.2.1 Background to dental caries

Dental caries is the localised dissolution of dental hard tissues, caused by the by-products of fermentation of dietary carbohydrate by commensal bacteria which are present in the biofilm which covers the tooth surface. This fermentation process results in the production of acid, which causes dissolution of the tooth mineral surface. The process is dynamic, with dissolution and re-mineralisation occurring over months or years. When this dynamic process is in equilibrium there is no loss of tooth structure. Carious lesions, therefore, are a result of an imbalance in this process, whereby gradual loss of tissue occurs

(Fejerskov et al., 2009). The term dental caries, therefore, represents a disease continuum of increasing destruction and severity, from sub-clinical molecular changes to obviously observable cavitation into dentine (Selwitz et al., 2007).

This dynamic process means that, in most cases, dental caries is a chronic disease which progresses slowly. It is also a multifactorial disease which is affected by several different processes, for example, enamel quality, salivary flow, exposure to fluoride, presence of dietary sugars and tooth brushing. Any factor which enhances the process of re-mineralisation or disrupts the biofilm can prevent disease development, and thus dental caries is preventable. Even once the demineralisation process has begun, disease progression can potentially be halted or even reversed with appropriate change in diet and fluoride exposure (Selwitz et al., 2007).

2.2.2 Defining the problem

The concept of 'public health' is perhaps still best defined by Winslow (1920), who defined it as 'the science and art of preventing disease, prolonging life and promoting health through the organized efforts and informed choices of society, organizations, public and private, communities and individuals.'

In determining if a condition is a 'public health' issue, as opposed to simply a 'health' issue, Sheiham and Watt (2003) identified the following criteria:

- How prevalent the condition is in the population and whether the prevalence is increasing or decreasing (criteria 1)
- The impact of the condition on the individual (criteria 2)

- The impact of the condition on society as a whole (criteria 3)
- Whether the condition is preventable and there are effective treatments available (criteria 4)

Based on these criteria, a condition can be considered a public health problem if the prevalence is high or increasing, or where the prevalence is low, but the condition is serious. There should be demonstrable impact on the individual but also on the society, for example through direct treatment costs or indirect costs from loss of work and school days. The final criterion is that the condition is preventable or effective treatment available. In this section, each of these criteria will be considered in relation to dental caries in children.

2.2.2.1 The prevalence of dental caries in children (criteria 1)

Worldwide, dental caries affects 60-90% of children, but the distribution of the disease is not uniform across the globe (Petersen, 2003). The most commonly used method for assessing the burden of disease for caries is an assessment of the mean number of decayed, missing and filled teeth (DMFT, or dmft in the primary dentition). The World Health Organisation (WHO) Global Oral Health Report revealed that the global mean DMFT in 12-year-old children was 1.67, with geographic differences between and within countries. For example, in the Americas, Belize had a mean DMFT in 12-year-olds of 0.6, whereas the figure was 6.3 for Ecuador. In individual countries within Europe, the DMFT ranges from 0.7 to 7.8 (Petersen, 2003). A limitation of many epidemiological studies is that they only report total DMFT/dmft scores, so measure lifetime experience of the disease rather than current untreated disease levels. In addition, epidemiological studies may use

different thresholds and criteria for diagnosis of caries, so it is difficult to compare results between populations.

However, Kassebaum and colleagues (2017) conducted a systematic review to consolidate the available information on *untreated* caries globally. The study found that in 2015, untreated caries in the primary dentition affected 9% of children worldwide, or 621 million individuals, making it the tenth most prevalent condition amongst children. In the permanent dentition, caries became the most prevalent condition worldwide, affecting 35% of the population. In contrast to WHO data on DMFT/dmft, Petersen and co-workers (2005) found no significant difference in the prevalence and incidence of untreated caries worldwide between 1990 and 2015. However, their findings confirmed the WHO report that the burden of disease was not uniform globally.

In the UK, every ten years, the Child Dental Health survey is carried out to provide information on the dental health of children. This survey was last conducted in 2013 and found that in the five-year-old age group approximately one third of children had 'obvious decay experience' and 28% had untreated caries. By age eight, caries experience had risen to nearly a half of children and 39% had untreated disease. Although this represented an overall decrease in caries levels from the 2003 results, large numbers of children in the UK continue to be affected by the disease. The survey also found that children from lower socio-economic groups were disproportionately affected. In five-year-olds, 41% of those eligible for free school meals, which is linked to deprivation, had caries experience compared to 29% of other children (Steele et al., 2015).

This survey also found that 13% of children aged 5-years and 15% aged 15-years were classified as having 'extensive or severe caries', with caries affecting many teeth or some teeth where there was gross decay or sepsis. Severe caries places a disproportionate burden on the child and family, as well as health services, for example, severe caries may necessitate treatment to be carried out under GA. The number of children requiring treatment under GA in the UK is growing, and this management approach is covered in more detail in *Section 2.3*.

2.2.2.2 Impact of dental caries on the individual (criteria 2)

The effects of dental caries on the individual are well documented, and range from the physical, such as pain or difficulty eating to developmental effects, loss of school days and affected school performance. This section explores some of these impacts in more detail, before considering how these factors impact on oral health-related quality of life.

Pain

Pain is perhaps the most commonly reported symptom of dental caries. A review of the literature by Slade (2001) examined the results of epidemiological studies reporting on pain in children and adolescents with dental caries. The reported prevalence of 'pain' ranged from 5-33%, among 17 studies conducted in the USA, Canada, Australia, England, and Wales. A subsequent study in Brazil, which took a life course approach, found that 36-71% of children with caries had suffered dental pain by age six, increasing to 65-85% by age 12 (Bastos et al., 2008).

However, the studies included in the review by Slade and the study by Bastos and colleagues in Brazil, relied on parental reports of whether their child had experienced toothache, rather than asking children themselves. Although there are similarities between parent and child reporting of pain, reports are not identical. For example, another study by Ratnayake and Ekanayake (2005) of 567 children aged 8-years in Sri-Lanka found that that 31% of parents reported their child had experienced pain in the preceding two months, contrasting with the lower reported pain experience by children themselves (25%) . The reason for this difference may be due to difficulty of recall over this period for children. Interestingly, there was also a difference between the parent and child reports of whether this pain was impacting negatively on the child; with 74% of children reporting negative impacts and just 66% of parents recognising a negative impact on their child.

Furthermore, very few studies have included the views of younger children themselves. For example, in the UK, although the most recent Child Dental Health survey included self-reported impacts of caries in older children, with 18% of 12-year-olds and 15% of 15-year-olds experiencing toothache in the three months prior to the survey, the survey relied on parental reports of toothache in younger children. These parental reports revealed that 14% of 5-year olds had experienced toothache, rising to 18% of 8-year-olds, but as already mentioned this may not reflect the pain experience of children themselves. (Tsakos et al., 2015).

One study of 589 children in England by Shepherd and co-workers (1999), which sought the views of children, found that 47.5% of 8-year-olds reported previous toothache, although approximately one third of those reporting pain attributed it to a ‘wobbly tooth’, so the percentage of children impacted by pain from caries may be lower than this.

More recently, Gilchrist and colleagues (2015) found that pain was the symptom most commonly reported by children with dental caries. However, they found that children reported pain based on severity (how much it hurt) rather than frequency, which may reflect how children think about pain and has important implications for how this impact is recorded. In addition, this study found that children used a wide variety of words in indicate pain, which may not always be reflected in epidemiological studies, and therefore the true prevalence of caries-related pain may be higher than that reported in such studies.

Infection

When left untreated, caries can progress through the dentine to the pulp, which may become reversibly or irreversibly inflamed, and ultimately necrosis may occur, with potential for pathological changes such as periapical granuloma or cyst formation. These sequelae of dental caries may result in further pain, and potentially facial swelling and pyrexia. In severe cases, spreading infection and swelling can compromise the airway and even threaten life, particularly in children who are immunocompromised or have other medical conditions, such as cardiac defects. This has implications for the management of the disease, and in children with systemic symptoms and significant medical conditions, urgent treatment is required to prevent progression to a potentially life-threatening state.

Impact on anthropometric measures

Anthropometry is the ‘measurement of the size, weight, and proportions of the human body’ (Li et al., 2015). It has been hypothesised that, as growth and dental caries are influenced by common factors (for example aspects of nutrition, parenting and the environment), there could be a relationship between dental caries and anthropometric measures.

Some studies have shown a link between caries in children and a disruption to their growth and development. Although it has been defined in many ways, with no accepted definition, 'Failure to Thrive' (FTT) is the term generally used to describe a 'lack of expected normal physical growth' or 'failure to gain weight'. Acs and colleagues (1999) found that children with caries, where at least one tooth had pulpal involvement, weighed approximately 2kg less than children without caries. They noted that of all children with caries, 14% weighed less than 80% of their ideal weight, meeting that criterion for FTT. However, this study did not consider other environmental factors which may have impacted on weight, such as deprivation. More recent studies have shown higher levels of untreated dental caries were associated with significantly lower weight and height-for-age in children, even when adjusting for other demographic and social variables (Mishu et al., 2013; Alkarimi et al., 2014).

The exact mechanism and relationship between dental caries and FTT is unclear. However, there are a number of ways in which dental caries has been implicated in FTT. As already stated, the pain or discomfort from untreated caries is known to impact on children's eating habits. Difficulty eating, or selective eating, and associated reduced nutritional intake can affect growth (Alkarimi et al., 2014). In addition, the increased cytokine action from the chronic inflammation of pulpitis and dental infection can result in anaemia due to depressed bone marrow erythropoiesis. It has also been suggested that sleep disturbance, caused by pain from dental caries, could affect glucocorticosteroid production which in turn can affect growth (Acs et al., 1992; Sheiham, 2006; Alkarimi et al., 2014).

However, there is also evidence to suggest the opposite relationship between dental caries and development exists, whereby there is an association between dental caries and

above average weight. Body Mass Index (BMI) is commonly used as an indicator of obesity, and given that dental caries and obesity share diet-related influences, it is perhaps not surprising that an association has also been found between high rates of dental caries and higher BMI scores by some investigators (Hooley et al., 2012).

Two systematic reviews have been conducted which aimed to evaluate the evidence for the link between dental caries and certain anthropometric measurements. Hooley and co-workers (2012) conducted a review of 48 studies, which demonstrated considerable disagreement as to whether, and to what extent, there was an association BMI and dental caries. They found caries was associated with both low and high BMI, although half of the studies found no relationship between caries and BMI. Several factors were implicated in the difference in findings, including differences in early caries diagnosis, poor sampling technique and potentially incorrect assumption of a linear relationship. Severe dental caries was associated with low BMI, which was also shown to improve in studies where comprehensive dental rehabilitation treatment was carried out (Acs et al., 1999). Other studies showed a relationship between dental caries and obesity, which was potentially linked to diet, reduced salivary flow and protein-deficient malnutrition; all factors which underweight children may also be affected by. Therefore, the evidence from this reviewed seemed to suggest that there is a relationship between caries and BMI, but that this relationship is non-linear.

A subsequent systematic review by Li and colleagues (2015) included studies looking at a number of anthropometric measurements in children; namely BMI, height and weight. One third of the studies reviewed showed no significant relation between caries and anthropometric measures and, as in the review by Hooley and colleagues, many of the

studies had conflicting findings. While two studies, which used caries as the predictor for anthropometric measures, agreed that increased caries experience was associated with decreased children's BMI, the remaining 15 studies, which used anthropometric measures as a predictor for dental caries, had conflicting results.

The two systematic reviews mentioned suggest that the evidence for an association between dental caries and anthropometric measures is complex and apparently conflicting. While some studies reported a link between caries and FTT as described above, others found no link and other studies found the reverse trend: that dental caries was linked to obesity. A major limitation with the included studies in both these reviews, which may account for some of the variation in findings, is that there was considerable heterogeneity between the studies, particularly in relation to methodology. However, what appears to be emerging is a more complex picture of the relationship between dental caries and growth and development in children; and a need for future studies to adopt standardised caries measures and to better control for potential confounders in analysis.

Effect on school attendance and performance

Pain caused by dental caries has also been shown to impact on school attendance. In a study of a group of 8-year schoolchildren in the UK, 11.5% of those who had recent toothache reported missing school because of it (Shepherd et al., 1999). School attendance is also affected by children visiting the dentist for treatment of dental caries, with the same study reporting that of those children experiencing pain in the previous four weeks, 42% ended up visiting a dentist. A more recent study into the effect of treatment waiting times in the UK found that, of children waiting to have treatment under GA, approximately one quarter had missed school, and on average had missed three school days. When these

figures were combined with those school days lost for attendance of dental appointments, most children missed at least two days at school, with some children missing up to two weeks at a time (Goodwin et al., 2015).

A recent systematic review conducted by Rebelo and collegues (2019) examined the evidence for the impact of dental caries on school attendance and performance. Eighteen studies, from nine different countries, were included. The authors found that children with decayed teeth had 44% higher probably of poor school performance and were 57% more likely to have poor school attendance. A limitation with all the studies in the review, however, was that that they were observational in nature and did not account for potential confounding factors such as socioeconomic status.

Effect on quality of life

For a number of years now, there has been a move to assess the impact of oral health conditions on individual's overall quality of life (Marshman et al., 2005). As mentioned previously in this chapter, some consequences of dental caries, such as pain and impaired function, impact on the everyday lives of children and their families. Gilchrist and colleagues (2015) found that pain-related consequences had a significant impact on the daily lives of children with caries; the most common aspects to be affected were eating and sleeping. Dental pain has also been reported to impact on quality of life of children in other ways, for example, affecting social interaction, school attendance, and play and emotional aspects such as being upset or distressed about their mouth and worrying about being different to their peers (Pulache et al., 2016).

To assess the impact of caries and its treatment on the everyday life of individuals, several oral health-related quality of life (OHRQoL) instruments have been developed. The concept of OHRQoL and its measurement are explored in detail in *Section 2.4*, and the impact of treatment under GA on OHRQoL is discussed in *Section 2.3*.

2.2.2.3 Impact of dental caries on society (criteria 3)

There is compelling evidence that dental caries has significant and wide-ranging effects on the individual. However, the impacts from dental caries are not confined to individual experience, and dental caries has been shown to also have a significant impact on the wider family and society as a whole.

Effect on the family

Severe dental caries has been shown to have a negative effect on OHRQoL of families as well as children themselves (Abanto et al., 2012; Martins et al., 2015). Studies have found that parents of children with severe dental caries worry that the child will have fewer life opportunities, which has been linked to the parent's concerns about the poor aesthetics of their child's teeth (Abanto et al., 2012). The studies also reveal that children with high levels of dental caries required more attention from parents, and parents also reported greater disruption to family life and requiring more time off work (Locker et al., 2002; Abanto et al., 2012). Parents also reported feeling guilty, which was attributed to parents recognising the causes of dental caries as being preventable and therefore considered they were to blame for their child's poor oral health state (Abanto et al., 2012).

Economic Impact

According to the WHO, dental caries is the fourth most expensive chronic disease requiring treatment in most industrialised countries (Petersen, 2003). These costs include both direct costs of treatment to health services but also the indirect costs though, for example, loss of workdays. Listle and co-workers (2015) looked at the direct and indirect costs of oral diseases globally. The results suggested that the global economic burden of dental diseases was $442 billion in 2010; of this, $298 billion was the direct cost of treatment and a further $144 billion attributable to the indirect costs in terms of productivity losses. They found the total direct costs in Western Europe to be US$91 billion, with the indirect cost of a further $41 billion. The authors estimated that 17% of these costs were attributable to dental caries in permanent teeth.

Very little data exist regarding the total costs to society of treatment for dental caries in children. However, in the UK, the cost of treatment of dental caries under GA is well documented. In England alone in 2017/18, there were approximately 42,000 hospital admissions of children under 16-years with a diagnosis of dental caries. The number of children undergoing dental treatment is increasing, with figures for 2017/18 representing an 8% increase on those from the previous year (NHS Digital, 2018). The cost of this treatment to the NHS was estimated at £30 million. These growing numbers of children requiring treatment under GA present an increasing economic burden to the NHS each year, not to mention the wider costs to society and for individuals as previously mentioned.

2.2.2.4 The condition is preventable and effective treatments are available (criteria 4)

Prevention of dental caries

The multifactorial and dynamic nature of dental caries means that is possible to prevent and slow progression of this disease in several ways. This has made the prevention of dental caries a target for population oral health improvement strategies, the success of which has important implications for service commissioning. There is now a vast body of evidence which supports caries prevention at both an individual and population level, which has been used as the basis for oral health improvement programmes worldwide. A number such programmes have been introduced in the UK, and these are considered in more detail below.

In Scotland, the 'Childsmile' programme was introduced in 2006, which combines universal and targeted approaches to prevention, including free daily supervised tooth brushing in nurseries and priority primary schools, free dental packs and access to tailored programmes of care in primary care dental services (Macpherson et al., 2010). The programme has been linked to the significant improvement in the oral health of children in Scotland in recent years. The most recent Scottish National Dental Inspection Programme Report (NDIP) found that caries prevalence in primary school-aged children (aged 4-7 years) had fallen from 55% in 2003 to 31% in 2016. The average number of teeth affected in these children had also fallen, to less than half of the average number of teeth affected in 2003. Although this reduction cannot be proven to be due to the 'Childsmile' programme alone, the timing of implementation of the programme and little improvement in the oral health of children in Scotland prior to its implementation would suggest it has had a

significant role to play. However, significant inequalities still exist, with 45% of children with obvious decay experience in the most deprived areas, compared to just 18% in the least deprived areas (Macpherson et al., 2016).

Following on from the success of 'Childsmile', a similar programme was introduced in Wales. The programme, called 'Designed to Smile', was launched in 2009, and has seen improvements in terms of both frequency and severity of disease, with schools enrolled in the programme having fewer children with dental caries, but also fewer numbers of decayed teeth in children with the disease (Morgan, 2018).

In England, no similar national oral health promotion programme exists. Rather, an evidence-based prevention strategy, 'Delivering Better Oral Health', was produced in 2014 and was most recently updated in 2017. This document clearly outlines the guidelines for prevention to be employed by clinical dental teams (Public Health England, 2017a). The strategies can be broadly divided into three categories: increasing fluoride availability, reduction in sugar consumption and protection of susceptible tooth surfaces. The evidence surrounding these interventions, and guideline recommendations, are described below.

Increasing fluoride availability

The primary means of increasing fluoride availability is using fluoride toothpaste. A systematic review, which included studies in adults and children, over follow-up periods of at least a year, showed that toothpastes with a minimum of 1000ppm fluoride reduced the incidence of dental caries in the follow-up period. The studies also showed that higher concentrations of fluoride had greater effect, however, the decision to increase fluoride concentrations had to be balanced with the increased risk of fluorosis in younger children

(Walsh et al., 2010). As such, current recommendations given in the 'Delivering Better Oral Health' toolkit are for toothpaste containing 1000ppm fluoride to be used for low risk children aged under 6-years, and 1450ppm for those who are older, or considered to be at higher risk of dental caries.

Another method for increasing access to fluoride is through professional intervention, for example through regular topical fluoride varnish application. A systematic review of randomised and quasi-randomised trials found that topical fluoride application reduced incidence of caries in children. The review included 22 studies involving children aged between one and 15-years of age, who were followed up for between one and five years. Comparing fluoride varnish with no treatment or a placebo, the prevention fraction estimate was 43% in permanent teeth and 37% in primary teeth (Marinho et al., 2013). The current guidelines therefore recommend application of fluoride varnish twice a year for all children over three years of age, with consideration to applying the varnish in younger children and applying more frequently if children are deemed high-risk.

Reducing sugar consumption

There is a large body of evidence which links the role of dietary sugars to dental caries. A systematic review by Moynihan and Kelly (2014) found a positive association between frequency of sugar intake and caries. On average, across all the studies, those in 'high sugar' intake groups were more likely to experience dental caries than those in 'low sugar' groups (risk ratio= 7.15). This review also found that even when considering fluoride interventions, the relationship between sugar intake and caries remained. However, there was considerable heterogeneity in the studies and so not all of the data could be included in the final meta-analysis. Despite this limitation, however, the authors concluded the

available evidence was sufficient to support a clear association between caries prevalence and frequency of sugar intake.

Decreasing sugar consumption is, therefore, an important target area in prevention of dental caries, not only through diet advice to individuals but also through community-level interventions and national policies. Such interventions have the potential to not only help prevent dental caries, but also reduce the population burden of other health problems such as diabetes and obesity. Colchero and colleagues (2016) have investigated the early impacts of the introduction of a sugar tax in Mexico. They found a greater than expected decline in the purchase of taxed beverages by an average of 6% compared to pre-tax consumption. Purchase of untaxed beverages increased by 4% and was mostly for bottled water. However, these preliminary results do not yet reveal whether these changes are enough to confer health benefits to the population overall, and further longitudinal studies will be required to assess this.

In April 2018, in the UK, a soft drinks industry levy was introduced so that drinks are taxed more heavily if they have high sugar content. This implementation is too recent for the effects on dental caries in the UK to be known. The Faculty of Dental Surgery (FDS) have suggested that there is still much that needs to be done in this area and have argued that the levy needs extending to include sugary dairy drinks, which are currently exempt. They have also suggested that, to reduce children's sugar consumption further, all schools in England should become sugar free (Davies, 2019). A more radical approach has been suggested to introduce corporate policies to tackle the sugar industry's efforts to increase sugar consumption nationally and globally (Watt et al., 2019).

Protection of tooth surfaces

The final target for prevention is protection of susceptible tooth surfaces through the placement of fissure sealants. A systematic review of the available literature found that children with fissure sealants placed on the occlusal surface of their permanent molars were less likely to have dental caries (Ahovuo-Saloranta et al., 2013). For the main outcome, comparing resin fissure sealant with no treatment, the results from nine studies were pooled and significant reduction in dental caries in the sealant group compared to controls were found at 12, 24, 36, and 48-54 months. The odds ratios ranged from 0.12 (95% CI 0.07 to 0.19) at 24 months to 0.21 (95% CI 0.16 to 0.28) at 48-56 months. This means that, for example, at two years post treatment the odds of having decayed surfaces were 88% less if a fissure sealant was placed.

The robust evidence base for the effectiveness of measures to prevent dental caries has important implications for the commissioning of services. This has been highlighted in proposed reforms to the current dental contracts in the UK, which currently remunerate dentists on the basis of treatment rather than prevention. The proposed reforms will seek to create a system in which dentists are also rewarded for preventing future disease in their patients. A key aspect of this reform is also to evaluate quality of care, as well as clinical outcomes (Department of Health, 2015).

Treatment of dental caries

Typical treatment approaches for the management of dental caries in primary teeth in children fall into three broad categories. Firstly, is what might be termed the ‘traditional’ approach, whereby carious tooth tissue is completely removed, and a restoration placed. In some instances, pulp therapy may be also indicated, where the tooth pulp is felt to be

compromised or non-vital. Where restoration of the tooth has a poor prognosis, or it is felt the tooth is not restorable, a decision may be made to extract the tooth. The second approach is to 'seal' the carious tissue into the tooth using an adhesive restoration or preformed metal crown, which is often referred to as a biological approach Finally, others have advocated a 'best practice prevention' alone, which aims to use the reversible nature of the disease to slow the rate of decay (Innes et al., 2013).

However, there is currently much debate over the most appropriate and effective treatments for dental caries in primary teeth in the UK. Guidance by the British Society of Paediatric Dentistry (BSPD) advocates the removal of carious tissue and placement of a conventional restoration. However, this recommendation is largely based on evidence from studies conducted in specialist or secondary care; whereas most children are seen in general practice.

A retrospective case study of 677 children, who were seen by 50 different general dental practitioners (GDPs), found no significant difference in outcomes for carious teeth which had been restored and those which had never been restored. In both instances, approximately 12% of carious teeth were extracted due to pain or infection, with the remainder exfoliating naturally. In addition, no significant difference was found in prescribing rates of antibiotics between the two groups (Tickle et al., 2002). However, this retrospective study relied in reports in patient notes for data collection and this is a limitation with the data collection for the study. For example, 911 teeth which required extraction or prescription of an antibiotic were excluded from analysis as no information was available on whether the tooth had caries or a restoration. In addition, the findings are unable to give a picture of the impact of each treatment approach on children. While there

were no differences between extraction and prescribing rates, this information alone cannot give a full impression of the impact of caries and its treatment on the population, for example, whether different treatment approaches affect ongoing incidence and prevalence of pain or wider quality of life outcomes.

A randomised controlled trial, involving children aged three to 10-years-old, which compared the Hall Technique (a biological approach for management of caries where caries is 'sealed' in using preformed metal crowns) with the traditional approach of completely removing carious tissue and placing a restoration, and was conducted in general practice. The study found that the Hall Technique was significantly more effective over a 2-5 year period than conventional restorations, with significantly fewer failures radiographically or clinically (Innes et al., 2011).

More recently, a retrospective study by BaniHani and co-workers (2018) also sought to investigate the effect of traditional versus biological approaches for the management of caries in children. The study looked at the impact of treatment in children aged nine to 14-years-old, following them up over a period of up to 77 months, and was carried out in specialist hospital settings. No significant effect of treatment type on outcome was found, with most teeth remaining asymptomatic in both groups (95.3% in the conventional treatment group versus 95.8% in the biological approach group). This result may be different from that in the study by Innes and colleagues due to the shorter time frame of follow-up, or other factors such as setting, participant age or level of experience of the dentist.

In the UK, a longitudinal study was recently completed which compared traditional management of dental caries (i.e. traditional caries removal and placement of a restoration) with the biological management of caries (i.e. sealing in caries with crowns, or partial caries removal and fissure sealant placement). The 'Filling Children's Teeth: Indicated or Not?' (FiCTION) Trial aimed to answer the question 'What is the clinical and cost effectiveness of restoration caries in primary teeth, compared to no treatment?', and children were followed up over a 4-year period in the first instance (Innes et al., 2013). While full results cannot be published until funders have approved them, preliminary results were released at the BSPD conference in September 2018. Key findings from the trial were around the need for intensive prevention targeted to the child and parents to underpin any treatment approach to caries, and that early treatment, regardless of approach, is important to reduce clinical and patient-reported impacts from dental caries. The full trial results will have important implications for the management of dental caries in children.

However, irrespective of which approach is proven to be most effective, the delivery of dental care to children can be challenging, especially when the treatment needs are extensive and there are additional behavioural, social or medical considerations. Success of treatment has been shown to be compromised when full patient co-operation is not achieved, and therefore in some cases treatment under GA is required (Eidelman et al., 2000). A number of studies have suggested that GA allows treatment to be carried out under 'optimal conditions', which may be more successful than if treatment is carried out under different conditions (Acs et al., 2001; Tate et al., 2002; Amin et al., 2010). However, GA carries a risk, albeit small, of a serious adverse event, including death. In addition, GA is associated with a number of more commonly occurring morbidities and, as such, is only used as a last resort (Lee et al., 2013). Although traditionally there has been a focus on the

clinical outcomes of treatment under GA, such as success of restorations placed and rates of repeat GA, more recently a number of studies have sought to look at how this treatment impacts on the quality of life of patients (Jankauskiene and Narbutaite, 2010). The indications, risks and benefits of this management approach are considered in more detail in *Section 2.3*.

2.2.3 Summary of the problem

There is conclusive and abundant evidence that dental caries presents a significant public health problem, meeting all four criteria proposed by Sheiham and Watt (2003). The impact of untreated dental caries places a significant burden on both individuals and society. For individuals, untreated disease has implications biologically and socially. The societal costs of treating dental caries are also significant, both in terms of direct economic costs but also indirect costs in terms of loss of work and school days. Severe caries in children often requires treatment to be carried out under GA, which results in an additional burden to individuals, families and society. The numbers of children receiving treatment under GA in the UK are increasing each year, with an estimated cost last year of £30 million to the NHS. The effect of dental caries and its treatment have been explored using patient-reported outcome measures, to better understand the impacts on daily life for individuals. However, most studies which look at the impact of dental caries or its treatment on children have relied on parental reports, which may not fully represent the views of children themselves. There is, therefore, a need for further research which considers the impacts of dental caries and its management from children's perspectives.

2.3 General anaesthesia (GA) for the provision of dental treatment in children

2.3.1 Introduction

Delivery of dental treatment to children can be difficult, especially where there are multiple decayed teeth of poor prognosis or the child is young, anxious or has additional behavioural needs. In these instances, children may require a GA for treatment to be completed. In this section, the indications for treatment under GA are considered, alongside the relative risks and benefits. The provision of GA to children is given in context for the UK and, more specifically, Sheffield.

2.3.2 Indications for GA in children

Treatment under GA may be required where the child is unable to complete treatment in a general practice setting, for example due to anxiety or lack of cooperation. The indications for dental treatment under GA in children in the UK are described in more detail elsewhere (Davies et al., 2008), but some of the indications for treatment under GA include:

1. Children who do not have the psychological or emotional maturity to cooperate with treatment
2. Children with a mental, physical or medical disability
3. Children who are extremely uncooperative, fearful or anxious and where other management techniques (such as sedation) have failed or are unsuitable

4. Children with extensive or severe caries (e.g. caries affecting teeth in multiple segments)

It is recommended that treatment is carried out under GA only as a 'last resort', but ultimately it is up to the dentist to make a judgement based on factors such as those above. The most common reasons for children to be referred to a dental GA service are because of large numbers of teeth requiring treatment and poor co-operation of the child and subsequent inability to complete treatment under local anaesthesia (Sheller et al., 2003; Savanheimo et al., 2005). It has also been shown that referral for GA is also often influenced by non-clinical factors; including convenience, the attitude of the dentist, and parental attitude (Harrison and Nutting, 2000).

Dental caries is the most common reason for children in England to be admitted for a GA. There has been a rising trend in hospital admissions for dental caries in recent years. In England alone, in 2017-18, dental caries was the primary reason for 42,000 hospital admissions of children under 16-years with a diagnosis of dental caries which represented a rise of 8% on the previous year (NHS Digital, 2018). This number is significantly higher than for children undergoing a tonsillectomy, which is the second most common reason for a child to be admitted for a GA. Most admissions for dental caries are in the 5- to 9-year-old age group. In 2017/18, there were 33,871 cases of children under 10-years-old being admitted to hospital for a GA due to dental caries. There are also wide regional variations in GA rates, with numbers in the Yorkshire and Humber region being higher than any other region in England, with approximately 6,413 cases in 2017-18 alone (HSCIC, 2018).

The number of children receiving a GA for dental caries has been rising since 1997. Initially this rise was attributed to the requirement for all GA treatment to take place in hospital since 2002, following the publication of a Department of Health document, 'A Conscious Decision' (Goodwin, Sanders and Pretty, 2015). However, the continued increase in numbers since then suggests this is not the only reason for the rise. Some studies have noted that general dental practitioners may lack confidence and are reluctant to treat children (e.g. Seale and Casamassimo, 2003; Goodwin, Sanders and Pretty, 2015), which may be why large numbers of children are referred at late stages of caries progression.

2.3.3 Treatment approach under GA

There are two main approaches for the management of carious primary teeth under GA. The first is where only extractions are carried out (referred to as an exodontia service), and the second is where restorations (including pulp therapies) as well as extractions are carried out (referred to as a comprehensive care approach). A number of clinical factors may influence the treatment approach, such as how restorable the tooth is, how urgent treatment is, the caries risk of the child, whether comprehensive care services are available and whether the child has any co-existing medical conditions. The decision may also be influenced by parent or caregiver views.

In the UK, far fewer centres provide comprehensive care for children with caries compared to those providing exodontia services, and so GA is mostly used for dental extractions (Savanheimo et al., 2014). This is presumably because there are additional costs associated with comprehensive care, which requires more time and equipment than exodontia alone. While comparisons between GA and other management techniques (e.g.

sedation) are documented in the literature, there is a paucity of research on the relative costs of the different treatment approaches under GA. This is an important area which warrants further enquiry. As well as the financial implications, there is a lack of evidence as to which treatment approach under GA gives the best results, both from a clinical and patient perspective. This is therefore an area which requires further research.

Sheffield is one of the few centres in the UK where children (under the age of 16-years) may receive comprehensive care under GA. In 2017/18, the Charles Clifford Dental Hospital (CCDH) in Sheffield saw over 4500 children, referred for a new patient assessment. Around 45% of those who attended these new patient assessments were suitable for and received treatment under GA for dental caries. In total, 2039 children received treatment under GA for dental caries, of which there were 1205 cases of exodontia only and 468 of comprehensive care.

Optimal treatment planning aims to ensure that children do not require a repeat dental GA within a short period, because of untreated or inadequately managed dental caries at the time of the first GA. However, the number of children who have repeat GA for dental treatment remains an area of a concern. A retrospective study in the UK found that 8.9% of children returned for a repeat GA over a 6-year period, while a separate study found the rate to be higher, at 12%, with nearly half of these cases occurring within two years (Albadri et al., 2006). However, this figure may compare favourably with other treatment methods. A study comparing treatment outcomes under GA and sedation found re-treatment rates were significantly lower in the GA group, with 59% of children receiving treatment under GA requiring additional treatment within two years, compared to 74% of children treated under sedation (Eidelman et al., 2000).

In centres where exodontia only is the sole treatment approach available, dentists may choose to remove not only grossly decayed teeth but any tooth with signs of decay; aiming to return the child to a state where they are free of obvious caries (Goodwin, Pretty and Sanders, 2015). One study recommended this radical approach in order to prevent further GA; after they found 75% of tooth extractions required at repeat GA were for teeth where caries in had been left at the initial GA as teeth were considered restorable (Harrison and Nutting, 2000). However, what has not yet been explored is the impact of such a radical approach on the children involved.

Potentially, the facility to undertake comprehensive care under GA may be a way of reducing the overall number of extractions for a child with multiple decayed teeth (Harrison and Nutting, 2000). Furthermore, for children who are not able to cope with pre-operative radiographs the potential to take dental radiographs under GA, to aid treatment planning, also ensures optimum clinical outcomes, However, there are increased costs for comprehensive care, increased waiting times and potential greater morbidity due to the use of oral or nasal intubation, muscle relaxant drugs and longer operating and recovery times.

However, there has been little research carried out to date of the relative merits of each approach. There is lack of evidence as to whether exodontia only or comprehensive care under GA results in fewer repeat GAs for dental treatment, and a lack of information regarding the relative costs of each approach in the long term. There is also a lack of data as to which approach is best in terms of both clinical and patient-reported outcomes. One study, by de Souza and co-workers (2016), found no difference between treatment approaches under GA on child OHRQoL, as reported by their parents. However, the sample size in this study may have been too small to detect any between-group differences. Another

limitation with this study was that the measure of OHRQoL used was relying on parent reports and was a generic measure which may not have been sensitive enough to capture caries specific impacts. There is a need for further research which explores the impact of different caries management approaches from the child's perspective and using a caries-specific measure of OHRQoL.

2.3.4 Benefits, disadvantages and risks of dental treatment under GA

Many children who require a GA for dental treatment have high levels of treatment need, for example severe caries affecting teeth in multiple quadrants, and in these children a GA can allow all treatment to be carried out in a single visit. Studies reporting the views of parents found that they see treatment under GA as a way of addressing their child's oral health needs, allowing them to interact socially soon afterwards (Goodwin et al., 2015). As GA allows completion of treatment in a single session, parental satisfaction rates with the treatment their child has received are usually high (Anderson et al., 2004). Children themselves also respond positively to dental treatment under GA, including noting that they feel proud after having completed the operation, and are pleased that their dental problems have been treated (Rodd et al., 2014).

Reports of parental and dentist perceptions show that both groups believe that completing treatment under GA 'keeps the regular dentists separate from treatment', and therefore may prevent children becoming anxious about seeing the dentist in the future (Goodwin et al., 2015). However, where children are already anxious about dental treatment, studies have shown that treatment under GA does not reduce children's future levels of dental anxiety (Hosey et al., 2006; Goodwin, Sanders and Pretty, 2015). Klaassen

and colleagues (2008) found that there was no change in the Children's Fear Survey Schedule-Dental Subscale (CFSS-DS) score before and after treatment for dental caries under GA, indicating that the treatment process had no impact on dental anxiety in these children. However, a subsequent study by Cantekin and co-workers (2014) found that CFSS-DS scores actually increased following treatment under GA, indicating an increase in dental anxiety. While these studies represent a limited sample and are not generalisable to the population, there does appear to be a trend. If one of the reasons for children receiving treatment under GA is lack of compliance due to anxiety, it would seem that more needs to be done, in terms of a psychological intervention, to tackle these underlying fears in order to prevent the need for treatment under GA in the future (Cantekin et al., 2014).

Additionally, there can be long waiting times for children to receive treatment under GA. One study of six hospitals in North East England found that average waiting times for treatment was 8-months (Goodwin, Sanders and Pretty, 2015). A companion qualitative study by Goodwin, Pretty and Sanders (2015) found that some parents expressed concern over how long their child had to wait and the negative effect these waiting times had. Parents reported that children were affected by continuing or increased pain during this waiting time, which caused sleepless nights, and which may have affected their performance at school. This study also revealed that parents felt frustrated at having to wait for treatment, particularly when they felt it had resulted in further pain or infection.

The most serious risk associated with a GA is the risk of death, albeit low, at an approximate incidence of less than 1 in 100,000 (Association of Anaesthetists of Great Britain and Ireland, 2003). However, morbidity associated with a GA is significant, and considerably more common. On average, studies report that between 40 and 90% of

children experience post-operative morbidity, including pain, nausea, vomiting and bleeding (Atan et al., 2004; Hosey et al., 2006; Rodd et al., 2014). The most common causes of morbidity reported by parents following GA are post-operative pain and prolonged bleeding (Hosey et al., 2006). However, a qualitative study using video diaries, conducted by Rodd and team (2014) found that pain was not commonly reported by the children; instead the most negative impacts described by children themselves were disturbed eating and hunger. Other notable outcomes were nausea, bleeding and tiredness. Additional outcomes which had not been reported on by parents included discomfort from the cannula placed during the GA process and feelings of being scared or worried. A limitation of this study was the short follow-up period of just two weeks, and more research is needed to explore the long- term impacts of treatment under GA in children.

Impact of treatment under GA on OHRQoL

As previously discussed, dental caries has been shown to impact on the daily lives of children, and therefore change in OHRQoL is considered an important outcome measure when considering the effect of treatment for caries.

Jankauskiene and Narbutaite (2010) conducted a systematic review with the aim of reviewing the literature on child OHRQoL following dental treatment under GA. Eleven articles, from the period January 1978 to October 2009, were included in this review. Most of these studies were observational studies which employed a pretest-posttest design. There was significant heterogeneity between the studies, in particular relating to the instrument used to measure OHRQoL. The included studies had used a range of structured questionnaires, consisting of differing and un-validated questions. Of the included studies, only four had used validated instruments (Versloot et al., 2006; Klaassen et al., 2008, 2009;

Malden et al., 2008), and of these only two had used the same instrument (Klaassen et al., 2008; Malden et al., 2008). However, a limitation with these two studies is that the instrument they chose to use was not actually validated in their study population. Therefore, while treatment under GA was found to improve children's oral health and their quality of life overall, it was not possible to fully consolidate the results of the studies because of the differences in instruments used. The systematic review also revealed that in all the included studies the questionnaires were completed by parents or caregivers rather than the children themselves. There is therefore a need for further studies to use validated instruments and instruments designed to be completed by children. The limitations of this systematic review itself were that the search was limited to English language studies and no quality assessment was carried out of the included papers. A number of studies have been published since this review, and therefore there is a need for an update systematic review which also includes an appraisal of the quality of the included papers.

2.3.5 Dental GA summary

Treatment under GA for dental caries is sometimes necessary where other techniques to deliver dental care to children are not appropriate; particularly where there are extensive treatment needs. However, treatment under GA is not without risk of morbidity and mortality; therefore, there is a need to ensure it is only used when absolutely necessary. It is also important to consider the outcomes following treatment under GA and to justify the risks and costs associated with it. There is a need for future research to assess the impact of different treatment approaches under GA on the daily life of children, from their own perspective, in order to better understand children's experiences and improve their quality of care.

2.4 Health and quality of life

2.4.1 Introduction

When considering dental caries and its impact on individuals and society, the previous sections have focussed on a clinical viewpoint. The impact of caries on individuals and society has been discussed from a largely biomedical view of health; that is, a view which focusses on biological and physiological processes and is measured using clinical outcomes.

However, this approach has been regarded as too narrow to encompass what is understood by 'health'. As such, over recent years, different views of health have been proposed which take into consideration wider social and psychosocial factors. Alongside this change in how health is viewed, new methods to measure patient reported health outcomes have been developed which consider these wider aspects of health and take into account the subjective views of individuals.

In this section, the wider concept of health is discussed and the development of views of health traced historically to the present day. Methods to measure health are then reviewed, with a focus on patient reported measures of OHRQoL.

2.4.2 Defining health

The task of defining health is not merely an academic one; it is of practical concern because how health is defined affects where the efforts of healthcare are directed and the

goals that are being set. As a concept, however, it is notoriously difficult to define and views about health have changed considerably over time (Larson, 1999). This section provides an overview on how 'health' has been defined historically through to the present day and considers how the definition of health impacts on how health outcomes are measured.

The English word 'health' comes from the old English word '*hælþ'*, which means 'wholeness, being whole, sound or well' (Harper, 2016). The perception of health as wholeness also has roots in ancient Greece. Early documentation differentiates between this state, and that of 'illness'; a state which was considered abnormal and to be healed if possible (Huber, 2015).

Historically, health was seen as both desirable and achievable, albeit subject to fate and the will of the 'Gods'. However, there was also awareness of how health (or lack of it) was not just supernaturally imposed on the individual but was also influenced by lifestyle choices. For example, the Greek goddess Hygeia represented the 'attainment of health through rational living'; and this was linked to environment, food and exercise (Dubos, 1959; Tountas, 2009).

It is the 'father of medicine', Hippocrates, who is typically credited with liberating views of health from spiritual influences. Through observation, he began to develop a physical view of health which prompted the development of 'natural' treatments for diseases (Kleisiaris et al., 2014). His work was the foundation for the development of what is known as the *biomedical model* of health, which has formed the basis of healthcare interventions and healthcare services for many years (Wade and Halligan, 2004). However,

Hippocrates maintained a holistic view of health, describing it a dynamic process and a state of balance between different 'elements', which could be influenced by individual lifestyle choices and environmental factors (Huber, 2015).

It was during the period from the 16th to 19th centuries that some of these wider influences on health began to disappear, and the biomedical model dominated. As studies in basic science, anatomy, and subsequently cell biology and microbiology developed, definitions of health became condensed into a medical paradigm. Health began to be defined, in purely physical terms, as the 'absence of disease' (Huber, 2015). Diseases were no longer understood as revenge from the 'Gods' or imbalance of elements but were understood in terms of the physical body and causative microorganisms. Although a narrow view of health, it is this view of health which drove much of the development of effective treatments for diseases during the 1900s (Wade and Halligan, 2004).

Beyond the biomedical model

A key criticism of the biomedical model is that it does not take into consideration other factors which may influence health or encompass individual subjectivity (Huber, 2015). Therefore, in more recent history, thinking appears to have gone a full circle; back to the more holistic view of health held by physicians in earlier times. It has been recognised that 'health' cannot merely be defined in relation to physical aspects.

This shift in thinking towards a more multi-dimensional view of health, is recognised in the World Health Organization (WHO) definition which describes heath as 'a complete state of physical, mental and social wellbeing, and not merely the absence of disease or infirmity' (World Health Organisation, 1948). Early criticisms of this definition

were that it is utopian and that it makes 'invalids of us all' (Garner, 1979). Others criticised it for being too abstract, for not clarifying what is meant by 'wellbeing' (Saracci, 1997; Huber, 2015). In addition, it fails to recognise the subjective aspects of health. However, despite the criticisms, the WHO definition of health is still the most commonly used definition worldwide (Larson, 1999).

Some have argued for the WHO to change its definition of health to one that recognises that, even without 'complete' physical, social and mental wellbeing, a person may see themselves as being 'healthy' (Larson, 1999). It is argued that a holistic view of health needs to consider whether a person can function as they wish, despite physical, social or mental problems they may have. For example, Bircher (2005) defines health as 'a dynamic state of wellbeing characterized by a physical and mental potential, which satisfies the demands of life commensurate with age, culture, and personal responsibility'. This definition considers the changes over time in a person's life and recognises that 'health' is a subjective experience, affected by individual life experiences.

Models of health have been developed with a multidimensional definition of health underpinning them. One of the most commonly used multidimensional models in research is the biopsychosocial model (Alonso, 2004). The theory underpinning this model is most strongly linked to work by Engel (1977), although the concept of a holistic view of health can perhaps be traced back to ancient times, as previously mentioned. The biopsychosocial model incorporates biological, psychological, social and cultural aspects of health. It is now widely accepted by health care professionals and the public alike that health and illness are a result of an interplay between these different factors. However, despite popular acceptance of this model of health, research has often continued to use the biomedical

model as its framework, particularly in assessing the impact of healthcare interventions and making decisions about healthcare policy (Alonso, 2004).

Defining oral health

Considerations as to what constitutes ‘oral health’ have also followed this transition from a disease-centred and biomedical definition to a more patient-centred and bio-psychosocial ethos. Oral health has been defined as the ‘standard of health of the oral and related tissues which enables an individual to eat, speak and socialize without active disease, discomfort or embarrassment and which contributes to general wellbeing' (Department of Health, 2005).

This definition reflects a biopsychosocial model of health, recognising the importance of being able to carry out daily activities which impact on general wellbeing. There is also recognition in this definition of the inter-relationship of oral health and overall health. More recently, the World Dental Federation (FDI) have proposed a new definition of oral health, which although similar to the Department of Health definition above, has the advantage that it has a clear theoretical framework underpinning it, which should make the evaluation and assessment of oral health easier. According to their new definition, ‘oral health is multi-faceted and includes the ability to speak, smile, smell, taste, touch, chew, swallow and convey a range of emotions through facial expressions with confidence and without pain, discomfort and disease of the craniofacial complex’ (Glick and Williams, 2016). The framework which accompanies this definition describes the complex relationships between three main aspects of oral health: the disease and condition status, physiological and psychosocial function, as well as other determinants which affect oral

health and moderating factors that affect how individuals score their oral health. Finally, the framework recognises the impact of, and on, overall health and wellbeing (*Figure 1*).

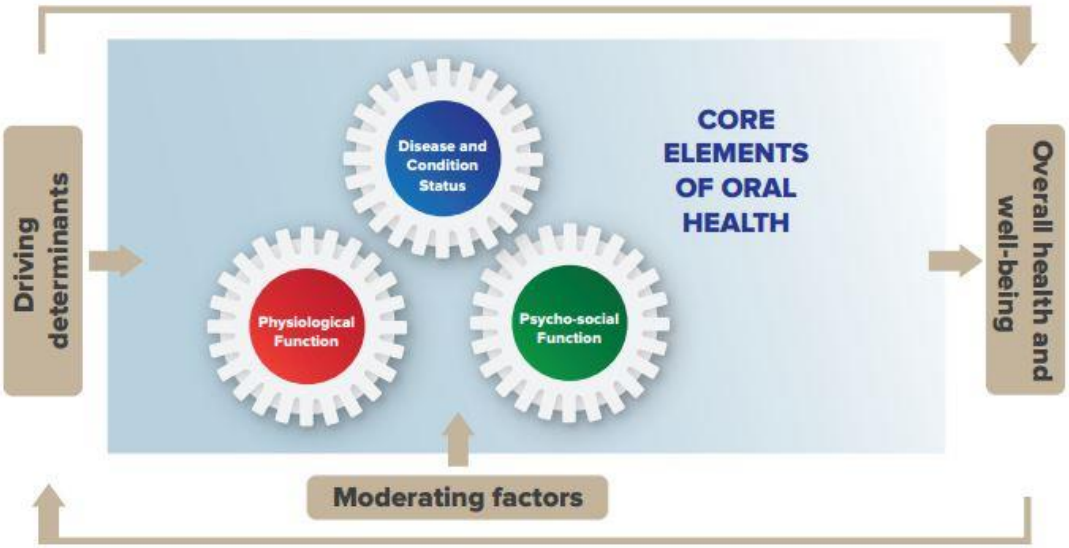

Figure 1: The World Dental Federation (FDI) framework, underpinning their new definition of oral health (Glick and Williams, 2016)

2.4.3 Measuring health

Just as the definition of health has changed over time, so too has the way in which health is measured had to change. With a purely medical view of health in mind, health outcomes can be measured on a purely objective basis using clinical indicators such as DMFT, but a limitation of studies relying on such indices is that they only reflect biological disease processes and are unable to record changes associated with the wider functional and psychosocial impacts on individuals (Barbosa and Gaviao, 2008).

Therefore, while easier to measure and quantify than multi-dimensional constructs, simplistic models such as the medical model may not lead to the best outcomes in terms of

health services and care. If one is to design interventions and plan services wisely, it is necessary to know how those who are to receive them perceive 'health'. It is also important to be aware of what influences a target population's perceptions of good health and which factors are important to them. This realisation has led to the development of measures to evaluate health in a way which incorporates the wider factors which impact on health.

Health-related quality of life

The concept of health-related quality of life (HRQoL) has emerged in this backdrop, with the focus on how diseases impact on individuals' daily life. Testa and Simonson (2009) defined HRQoL as the 'physical, psychological and social domains of health, seen as distinct areas that are influenced by person's experiences, beliefs, expectations and perceptions'.

The exact relationship between health and quality of life is still heavily debated. Some definitions would suggest that HRQoL equates to health, however, others would argue that HRQoL is broader, encompassing additional factors related to human experience (Locker and Allen, 2007). Wilson and Cleary (1995) developed a conceptual model of quality of life which attempts to explain the relationships between clinical variables and quality of life (*Figure 2*). It proposes five levels at which 'health' can be measured: biological and physiological, symptoms, functioning, general health perceptions and overall quality of life.

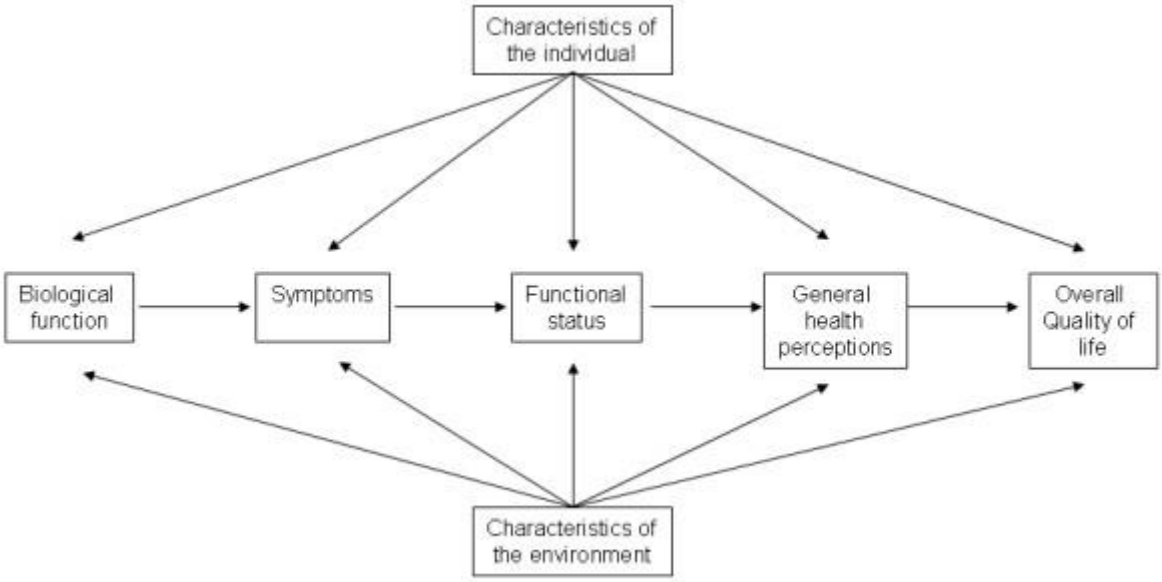

Figure 2: Wilson and Cleary model of health-related quality of life (Wilson and Cleary, 1995)

Each level is related to the others and influenced by individual and environmental factors. This model has provided an important framework for studies looking at health-related quality of life, as it attempts to relate traditional clinical variables to measures of HRQoL. This is important, as it is necessary to understand the underlying factors and pathways between them in order to develop effective interventions to improve HRQoL (Baker et al., 2007). To date, several studies have employed the Wilson and Cleary model in relation to various health conditions, and there is support for the direct pathways in the model. More recently a study by Baker and colleagues (2007) found evidence of indirect pathways and effects between non-adjacent levels, as well as direct relationships between non-adjacent levels, and highlighted the complexity of the relationships between clinical and non-clinical variables in participants with dry mouth. This highlights the need for

further research to build on current understanding of the pathways underpinning HRQoL in relation to specific conditions and their treatment.

Measures of health-related quality of life

Over the last thirty years there have been numerous attempts to develop methods to assess health-related quality of life, which have led to the use of patient reported outcome measures (PROMs). These measures fit into two broad categories, those which are generic and those which are disease specific, although the two are not mutually exclusive and can be used in combination (Guyatt et al., 1993).

Generic measures are designed to cover a range of health conditions. The main advantage of generic measures is that they are useful for comparing outcomes for populations or groups with different health conditions, due to their broad applicability. The range of impacts that are covered by generic instruments means they are sometimes able to detect unexpected problems associated with illnesses or conditions (Guyatt et al., 1993). They are more commonly used than disease-specific instruments and can be used where no disease-specific measure exists. However, a disadvantage of this broad applicability is that generic measures are less responsive to change and participants may find them less relevant and acceptable than disease-specific measures (Guyatt et al., 1993; Fitzpatrick et al., 1998).

Disease-specific measures have a number of inherent advantages when assessing change in individuals with a specific disease over time and/or following an intervention. They are considered more responsive to change and participants may find the content more acceptable and relevant and therefore potentially higher completion rates are achievable

(Robinson et al., 2002). The disadvantage is that they may be too specific to detect effects not anticipated (Fitzpatrick et al., 1998).

Several generic instruments have been developed for use with child populations, but very few have included children in their development. However, one measure which has fully engaged children in its development is the Child Health Utility questionnaire (CHU9D). Guyatt and co-workers (1993) suggested that for measures of HRQoL to be reliable, 'items on the questionnaire must reflect areas that are important to those suffering from the disease'. The CHU9D, a generic measure of HRQoL, was developed with children aged 7- to 17- years-old, to identify health dimensions that are important to them. The measure has also been used for children as young as 5-years-old, with adult, and has now been used in over 190 studies in a variety of situations, including clinical trials and observational studies, across a range of health conditions support (Furber and Segal, 2015).

However, as already discussed, it has been shown that generic measures may not be sensitive enough to measure the specific impact of some diseases. Foster Page and co-workers (2014) found that the CHU9D was not sensitive enough to detect the impact of dental caries when the level of dental caries, and subsequent impacts, in the individual were low. There is, therefore, sometimes benefit in using a disease-specific measure, and these are considered in relation to oral conditions in the next section, with a focus on dental caries.

2.4.4 Oral health-related quality of life

Introduction

While the concept of HRQoL emerged in the 1960s, the notion of oral health-related quality of life (OHRQoL) only emerged in the 1980s. This delay was perhaps because the impact of oral diseases on general health was not so well understood and there was a perception that oral disease had little impact on social issues (Bennadi and Reddy, 2013). However, as with overall health-related quality of life, researchers began to understand that objective clinical measures of health were insufficient to fully understand the impact of oral diseases on individuals. Studies which examined the association between objective measures of dental disease (such as presence of dental caries) and patient opinions on their oral health found only a weak relationship, and concluded that objective measures do not adequately reflect patient perceptions of their oral health (Allen, 2003). This understanding has led to the development of measures of OHRQoL. In this section, the definition of OHRQoL, its applications and current measures of OHRQoL for use in children will be reviewed.

Definition of OHRQoL

OHRQoL has been defined as 'the impact of oral diseases and disorders on aspects of everyday life that a patient or person values, that are of sufficient magnitude, in terms of frequency, severity or duration to affect their experience and perception of their life overall' (Locker and Allen, 2007). This definition explicitly links oral health to overall quality of life. While several conceptual frameworks for measuring health have been developed, which could be used in OHRQoL research, most studies of the impact of caries on OHRQoL do not make reference to which conceptual framework they are using (Gilchrist

et al., 2014). This has made it difficult to develop a knowledge base for OHRQoL research. In addition, while several factors have been associated with OHQoL, the research is often cross-sectional, studying only one or two factors at a time.

Some studies of the impact of oral conditions on quality of life have used the Wilson and Cleary model of HRQoL as the underlying framework to their research, and the findings have been largely compatible with this model (Baker et al., 2007; Gururatana et al., 2014). Baker and coworkers (2010) used the Wilson and Cleary model to inform their choice of outcomes in a longitudinal study investigating the OHRQoL of young people. They found that by including additional outcomes, such as income (an environmental factor) and sense of coherence (an individual factor), they could more fully explain the impact of oral health on overall wellbeing. However, there is still need for research which can clarify the relationships between variables in relation to other oral conditions and their management.

For this PhD, therefore, the Wilson and Cleary model (see previous section) has been chosen as the underlying framework. This model was chosen because it effectively operationalises the biopsychosocial model of health, incorporating factors which reflect the broader concepts of health, and has been used previously in oral health research as a framework for investigating the relationship between clinical factors and the impact they have on patients (Baker et al., 2007, 2008). This framework is helpful for identifying variables which may impact on OHRQoL outcomes and forms the basis for subsequent path analysis of direct and indirect relationships between clinical factors, individual factors, OHQoL and overall quality of life.

Applications of OHRQoL

OHRQoL measures have applications in three broad areas: theoretical, practical and political (*Table 1*). Theoretical applications could include exploring models of oral health or describing factors which influence health, which in turn has numerous practical applications. Foster Page and colleagues (2013) suggested that measures of OHRQoL offer valuable clinical applications, such as insight into treatment needs and assistance with clinical decision making. Other practical applications in public health and research could help evaluate interventions and services. Political applications may include such things as identifying the public's priorities in healthcare through to public involvement.

Table 1: Potential applications of OHRQoL measures, adapted from Robinson et al., (2003).

Application	Examples of usage
Theoretical	• Exploring models of oral health • Describing which factor influence health
Practical	• Planning, monitoring and evaluating services • Health needs assessments • Evaluating outcomes of healthcare interventions • Evaluating individual patient care • Improving patient-practitioner communication • Clinical audit • Marketing of services
Political	• Demonstrating involvement of the public in healthcare • Identifying priorities from the public's perspective • Advocacy

Measures of OHRQoL in children

Several measures of OHRQoL exist for use with children or using parents as proxies. However, only moderate agreement has been found between parent- and child-reported quality of life, and is highly dependent on the dimension of quality of life being considered (Eiser and Morse, 2001). There is generally better agreement on observable factors (such as functioning) than for non-observable factors (such as emotional or social ones). Therefore, it has been recommended that wherever possible, parental reports should be used to supplement child reports of OHRQoL, rather than acting as a proxy for them (Marshman and Robinson, 2007).

Of the few measures of OHRQoL designed to be completed by children themselves, the most commonly used are the Child Perceptions Questionnaire (CPQ) component of the Child-OHRQoL Questionnaire, the Child Oral Impacts on Daily Performances (C-OIDP) and the Child Oral Health Impact Profile (COHIP). Gilchrist and collegues (2014) carried out a systematic review to assess the methodological quality of the development and testing of these measures. The most commonly used measure in the included studies was the CPQ. An overview of these measures, and key findings from the systematic review, are detailed below.

Child Perceptions Questionnaire (CPQ)

The CPQ forms part of the Child-OHRQoL (C-OHRQoL) Questionnaire. This questionnaire was designed to incorporate both child and parental perceptions of OHRQoL. There are three separate components to the C-OHRQoL questionnaire, with each age-appropriate version of the CPQ reflecting differences in children's cognitive development. The following are the component questionnaires:

1. Parental- Caregiver Perceptions Questionnaire (P-CPQ), which was designed to assess impacts from the parent perspective, to supplement the views provided by children themselves.
2. Family Impact Scale (FIS) that assessed impact on the parents and rest of the family.
3. Child Perceptions Questionnaire (CPQ) for children 6 to 7 years of age (CPQ_{6-7}), 8 to 10 years of age, (CPQ_{8-10}), and 11 to 14 years of age (CPQ_{11-14}) that assess impact from the child's perspective. Short-form versions of the CPQ_{11-14} have also been developed.

Several studies have employed the CPQ to investigate the impacts of dental caries in children, but the studies have some conflicting results. While some studies show that children with caries have significantly higher scores (i.e. poorer OHRQoL) than those who do not have caries (Jokovic et al., 2004; Foster Page et al., 2008), others do not demonstrate an association between caries experience and CPQ (Barbosa et al., 2009; Gururatana et al., 2011). This may be because the instrument is not sensitive enough to measure change in caries specific impacts, especially in populations with low levels of disease (Marshman et al., 2005).

The CPQ was developed originally for use with a wide range of dental and oro-facial disorders and therefore may not be specific enough when exploring some impacts related to caries alone (Jokovic et al., 2002). The development of the CPQ included children in the process, however children were not involved in item generation (Gilchrist et al., 2014). This may impact on the validity of the measure. In particular, as previously mentioned, it has been shown that children tend to think about symptoms in relation to

severity rather than frequency (Gilchrist et al., 2015); but the response format in the CPQ relates to the latter.

Child-Oral Impacts of Daily Performance (C-OIDP)

The C-OIDP was derived from the adult version Oral Impacts on Daily Performance (OIDP) and is designed for use with children aged 11-12 years. It is based on an explicit conceptual framework: the WHO international classification of impairments, disabilities and handicaps (ICIDH), and assesses the ability to perform daily activities. It has been used in population surveys and has been validated for use in children in Thailand, France and UK. It has been suggested it could be used for planning services, and also in cross sectional surveys (Locker and Allen, 2007; Marshman and Robinson, 2007). However, to date, it's use has been restricted to validation studies and studies looking at the impact of various oral and medical conditions (Gilchrist et al., 2014).

A number of studies have employed the C-OIDP to investigate the effect of dental caries in children, and have shown children with dental caries report significantly more impacts than those without (Krisdapong et al., 2013). Notably, the 2013 Dental Health Survey in England employed the C-OIDP to assess impact of dental caries in 12- and 15-year olds and found that 58% of 12-year-olds and 46% of 15-year olds reported experiencing one impact from their oral health, although it is not clear what percentage of these reports were due to caries alone (Tsakos et al., 2015).

Child Oral Health Impact Profile (COHIP)

The COHIP was developed from the CPQ initial item pool, for use with children aged 8-15-years-old. As with the CPQ, it was designed for use across various oral

conditions. However, in contrast to the CPQ, it includes positive aspects of OHRQoL such as confidence and attractiveness, as well as the negative aspects of OHRQoL. The decision to include these aspects was derived from the theoretical perspective of health highlighted by the WHO definition; with health being 'more than the absence of disease' (Broder, 2007).

The COHIP has been employed in relatively few studies to date. However, it has been used to examine the impact of caries, and studies show a significant correlation between overall COHIP score and dental caries (Broder and Wilson-Genderson, 2007). A short form version, COHIP-SF 19, has also been developed (Broder et al., 2012). The questionnaire has also been used longitudinally to measure change following treatment for other oral conditions, such as molar-incisor hypomineralisation (Hasmun et al., 2018).

Limitations with these measures

None of the above measures were specifically designed to be evaluative. Evaluative measures must be responsive and demonstrate longitudinal construct validity (Guyatt et al., 1993). These aforementioned measures have only been used in a handful of longitudinal studies to date, and therefore their validity in certain populations has not been sufficiently tested (Terwee et al., 2007). In addition, all the aforementioned measures of child OHRQoL were designed for use with a range of oro-facial conditions, and may not be sensitive enough to measure disease-specific impacts on OHRQoL (Guyatt et al., 1993).

The Locker and Allen (2007) definition of OHRQoL affirms the subjective nature of OHRQoL and therefore the need for patient-centred measures that address aspects of daily life that are important to them. It is clear from the results of the systematic review by

Gilchrist and co-workers (2014) that the above measures may not have sufficiently incorporated these two aspects during development. The C-OIDP was developed from adult versions of the questionnaire, a process which may be inappropriate as the content of adult questionnaires may not address aspects of daily life which are relevant or valued by children (Marshman and Robinson, 2007). While the CPQ, and by extension the COHIP, was developed with children, children were not fully involved in item generation, which may also affect the validity of the questionnaire (Gilchrist et al., 2014). Therefore, to address these limitations, Gilchrist and co-workers (2018) developed the 'Caries specific measure of oral health related quality of life' (CARIES-QC), for use in children aged between 5- and 16-years-old.

Caries specific measure of oral health related quality of life - CARIES-QC

CARIES-QC was developed based on the Locker and Allen (2007) definition of OHRQoL, and was designed to address some of the acknowledged limitations of existing OHRQoL instruments. Unlike other measures of OHRQoL, CARIES-QC was developed with input from children at all stages. Importantly, the involvement of children during item generation helped to identify impacts related to caries which affected their daily lives, and which were important to them. Children also contributed to item reduction and the design of the measure. The development process revealed that children generally discussed the severity of the impacts they had experienced, rather than the frequency with which they occurred. This was an important finding as other measures of OHRQoL (e.g. CPQ and COHIP) use a response format that is frequency-based, which is not how children tended to think about the impacts. Therefore, the format of CARIES-QC is severity-based rather than frequency-based, in the language children themselves used in the development process (Gilchrist et al., 2018). CARIES-QC contains 12 items and one global question. All

questions are scored on a 3-point Likert scale, rather than a 5-point scale used in other measures, based on severity responses by children as to which terms they could differentiate. Children are asked whether each impact bothers them ‘Not at all’, ‘A bit’ or ‘A lot’. The responses are scored 0, 1 and 2, respectively, giving a total possible score of 24. Increasing score equates to increased impact on OHRQoL. In addition, the raw ordinal score can be converted to an interval score to allow calculation of change scores and effect sizes. The measure has been evaluated and has been shown to have acceptable validity, reliability and responsiveness (Gilchrist et al., 2018).

2.4.4 HRQoL and OHRQoL summary

Definitions of health and quality of life remain debated, and the reason for lack of consensus is the multidimensional, complex nature of the concepts. The confusion grows as terms such as health, happiness and wellbeing are often used interchangeably. In addition, as a concept, health is continually evolving. While the debate over definitions is likely to continue, affected by social and cultural contexts, it is necessary to choose a definition in order to know what one is aiming to achieve in health research. The model of health which will underpin this thesis is the biopsychosocial model, chosen as it is the most commonly used model in the literature, and because it represents a holistic view of health which has been linked to quality of life through the framework proposed by Wilson and Cleary (1995), which was described in more detail in *Section 2.4.3*.

There have been several instruments developed to measure OHRQoL in children, although the majority are generic measures of OHRQoL and as such, may be unable to detect the impacts which are specific to dental caries. There has only been one caries-

specific measure for children developed, which has been used in few studies to date. CARIES-QC is the only measure which was developed with input from children at all stages of its development. While there have been several studies published which have assessed the impacts of dental caries on OHRQoL in children, relatively few have investigated the effects of dental treatment for dental caries on OHRQoL.

2.5 Conclusions

Having reviewed the literature relating to the ubiquitous nature of caries in children, identified some key issues relating to the provision of dental care under GA and highlighted the need to consider OHRQoL in this population, the following conclusions have been made:

1. The significance of dental caries as a public health problem, as well as the wide-ranging impact of dental caries on individuals, establishes this as a disease of importance and worthy of investigation.
2. While several studies have explored the impacts of dental caries from a clinical perspective, or sought views of parents, there is little research to date which has sought the views of children themselves. There is a need for future research to use child-reported outcome measures to assess the impact of caries and its treatment, particularly under GA.
3. Most studies which examine the impact of caries on everyday life have used generic measures which may not be sensitive enough to detect caries specific impacts. Future studies should therefore include caries-specific instruments to measure OHRQoL.

4. There is limited research available on the relative merits of different treatment approaches; exodontia only or comprehensive care. There is a need to assess the impact of these different approaches from children's perspectives.

Therefore, based on these findings from the literature, the aims and objectives of this thesis are as follows:

Aim: To examine the impact of dental caries and its treatment under GA on the everyday lives of children and their families.

Objectives:

1. Conduct a systematic review of the current literature on the effect of dental treatment for caries under GA on children's OHRQoL.
2. Investigate the impact of dental caries and its treatment under GA on children's everyday lives, using a child-centred measure of OHRQoL.
3. Investigate the impact of dental caries and its treatment under GA on the families of these children.
4. Examine the relationships between individual, clinical and environmental factors on children's OHRQoL and QoL, with respect to treatment for caries under GA, using path analysis.

Chapter 3: Systematic review on changes in children's oral health-related quality of life following dental treatment under general anaesthesia

3.1 Introduction

As previously discussed in *Chapter 2*, oral health-related quality of life (OHRQoL) measures have been used to assess the impact of dental caries and it's treatment on children, which aim to take into account the wide-ranging impacts of dental caries on everyday life, including physical, emotional and social aspects (Locker and Allen, 2007).

Jankauskiene and Narbutaite (2010) were the first to conduct a systematic review to summarise the literature on child OHRQoL following dental treatment under general anaesthesia (GA). The review detailed studies reporting OHRQoL in children undergoing dental treatment under GA published from January 1978 to October 2009. This review highlighted the use of proxy-reported measures and the need for future studies to engage children themselves, using a validated measure. Several limitations of the included studies were highlighted by the authors, including the variation in instruments used, the lack of validation of these instruments, and a reliance on parental or proxy reports of child OHRQoL.

Several papers have been published since this review which warranted systematic investigation and analysis to determine how well the limitations identified by Jankauskiene and Narbutaite have been addressed by more recent work. Their review was also limited to English language papers so may have also been subject to publication bias. In addition, as no quality assessment of included papers was carried out previously, it is unclear how robust the included studies in the previous review were.

This chapter presents a systematic review of the literature reporting change in OHRQoL following treatment under GA, to provide an updated appraisal of the body of recent research, regardless of the language of publication. It will also assess the quality of those studies seeking to measure changes in OHRQoL. In contrast to the review by Jankauskiene and Narbutaite, which included all studies reporting OHRQoL, this review will be limited to studies reporting a change in OHRQoL before and after treatment. An understanding of the current use of OHRQoL measures with children and an evaluation of the strengths and limitations of the studies to date will inform the study design for this PhD research project.

3.2 Aim and objectives

Aim

The aim of this stage of the thesis is to systematically appraise the literature to examine changes in OHRQoL in children undergoing dental treatment for the management of dental caries under GA.

Objectives

To fulfil this aim, the specific objectives are as follows:

1. To describe the study designs and methodology employed in studies assessing changes in OHRQoL in children following dental treatment under GA,
2. To describe the instruments used to measure OHRQoL in these studies,
3. To describe changes in OHRQoL reported in these studies,
4. To examine the quality of these studies using a validated quality assessment tool.

3.3 Methods

This review was conducted in accordance with published guidelines for undertaking a systematic review (Akers et al., 2009; Higgins and Green, 2011). A protocol was written and submitted to the International Prospective Register of Systematic Reviews (PROSPERO), which guided the review process, and any changes made to the methodology were documented and this protocol updated, to improve transparency and reproducibility of the review process.

3.3.1 Inclusion and exclusion criteria

Inclusion and exclusion criteria were determined by discussion and agreed by three investigators (RK, ZM and FG), based on the population, intervention, comparator, outcome and study design (PICOS) model (Higgins and Green, 2011) as follows:

- **Population:** a paper was considered suitable for inclusion if it involved children aged 16-years-old and under. The paper was excluded if the studies involved participants over the age of 16.
- **Intervention and comparators:** studies examining dental treatment under GA for dental caries were included. Studies where treatment was not carried out under GA or where treatment was under GA but for other clinical presentations (e.g. surgical removal of unerupted teeth, exploration of oral pathology) were excluded.
- **Outcomes:** The primary outcome was the change in OHRQoL following treatment, but studies exploring secondary outcomes in addition to this (e.g. anxiety, parental satisfaction) were included. Studies were excluded if OHRQoL was only measured at one time point, i.e. change post-treatment was not examined.
- **Study design:** all study designs were included.

3.3.2 Search strategy

A systematic search strategy was adopted to identify relevant articles. An attempt was made to identify all relevant studies, regardless of the year of publication or language to ensure the review was as comprehensive as possible, an. Database searches were carried out from the date of inception to present of MEDLINE (1946-), Scopus (1966-) and Web of Science (1900-) using free text and MeSH terms combined with Boolean operators. The following terms were included in the search strategies: oral health, quality of life, dental treatment, general anaesthesia, dental care for children. The Cochrane library and PROSPERO were searched to identify any other systematic reviews. Citation searching and reference list searching for included studies were carried out to identify additional articles. Duplicates were recorded and removed at this stage.

3.3.3 Study selection

Titles and abstracts were independently reviewed against inclusion criteria by two investigators (RK and FG) to ascertain whether they met the inclusion criteria. Where titles and abstracts met or appeared to meet the inclusion criteria, the full text was obtained and reviewed against the criteria to determine eligibility for inclusion in the review. This process of assessing the full texts for eligibility was carried out by two researchers independently. RK reviewed all full-text papers, with ZM, FG and HR each reviewing a third of the papers. Where there was disagreement between reviewers, they met to discuss and reach a conclusion. Where agreement could not be reached the opinion of a third reviewer was sought. Studies which did not meet the criteria at this stage and reason for exclusion were documented.

3.3.4 Data extraction

Data were extracted using a custom spreadsheet to record the following for each study:

1. **Study details:** the author and publication year were recorded, as well as study characteristics such as the study design, sampling and data collection methods.
2. **Population characteristics:** the number of participants, caries experience (recorded as dmft/DMFT) and demographic details were recorded, along with whether they received extractions only or comprehensive care. Follow up rates were also noted.
3. **Outcomes:** The primary outcome was the change in OHRQoL, and the instrument used was noted along with details of whether it was a validated instrument and

whether it was designed for self-report by children. Information was also recorded on whether the change was statistically significant, what the minimally important difference was and whether change scores had been correlated to a global transition judgement. In addition, information was recorded on the use of secondary measures, for example, parent satisfaction or child anxiety.

Initially, the data extraction spreadsheet was piloted using three articles, all reviewed by three investigators independently (RK, ZM, FG). This exercise gave the opportunity to refine the spreadsheet, and any disagreements in the extraction data were resolved by discussion. A final version of the data extraction sheet was produced following these discussions. Subsequently, three teams of two investigators (RK/ZM, RK/FG and RK/HR) independently carried out the data extraction for each paper. Where there were discrepancies, these were resolved by discussion. Where agreement could not be reached the opinion of a third reviewer was sought.

3.3.5 Quality assessment

The same teams of two reviewers then independently assessed the quality of included studies using the Quality Assessment Tool for Studies of Diverse Design (QATSDD), which has shown good reliability and validity for use with a range of study designs (Sirriyeh et al., 2012). This tool includes 16 items to assess quality, which are scored between 0 and 3. Two of the items were not evaluated as they were only relevant to qualitative studies, giving a total possible score of 42 from 14 criteria. Total scores for each paper were calculated. The mean score, out of a total possible score of 3, for each criterion met by the included papers was also calculated. Disagreements between the reviewers over

the quality assessment of studies were resolved by discussion, with a third reviewer invited to resolve issues where necessary.

3.4 Results

The search strategy yielded 325 records, of which 204 were duplicates, leaving a total of 121 abstracts. Following the screening of these titles and abstracts against the eligibility criteria, 28 full papers were retrieved which appeared to be eligible for inclusion in the review. Following the screening of the full papers against the inclusion criteria, by two reviewers independently, a further six full-text articles were excluded. In total, 20 studies, which had been reported in 22 different papers, were included in the final review, with reasons for exclusion of papers documented (*Figure 3*).

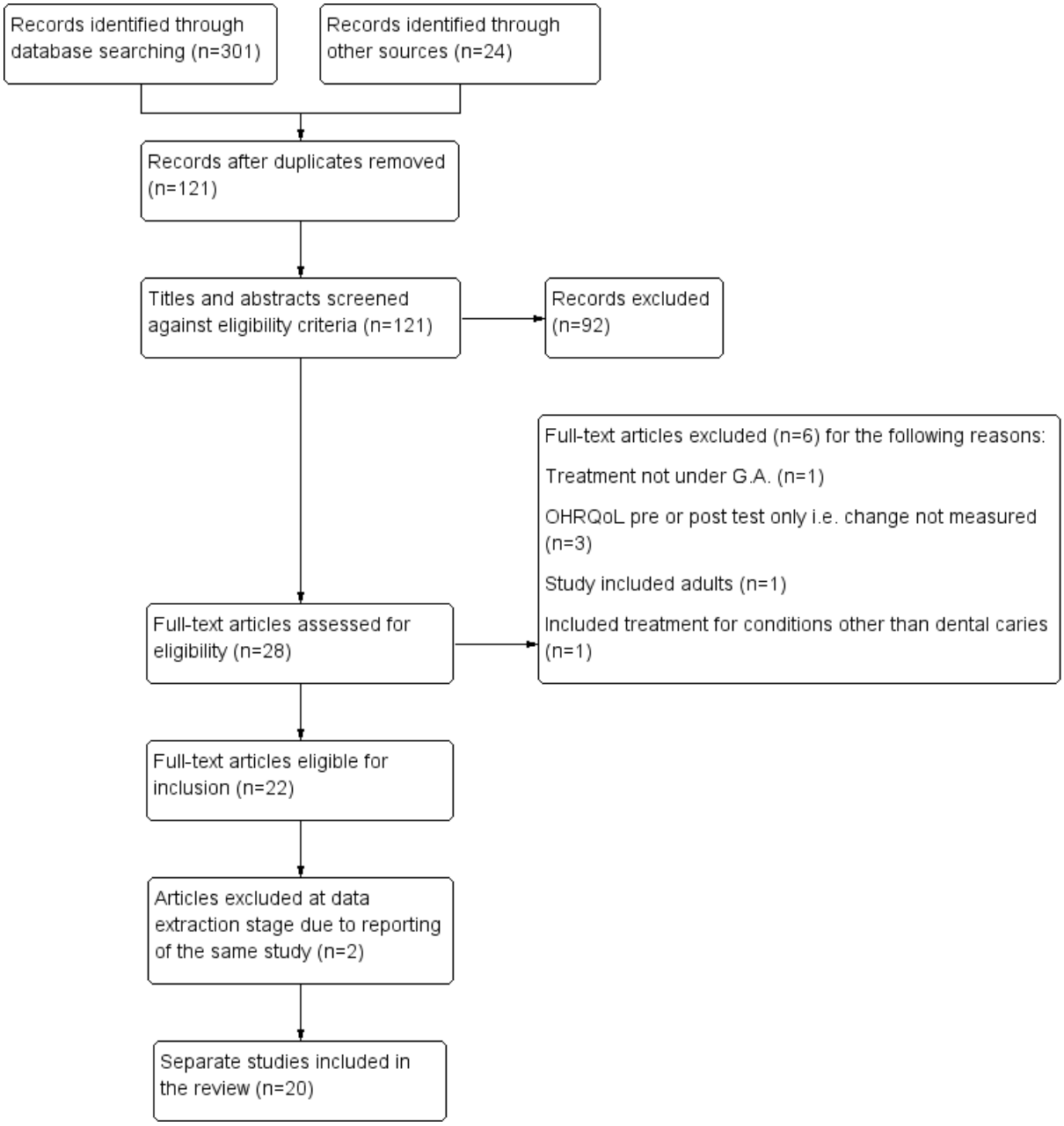

Figure 3: PRISMA diagram to show stages of systematic review and reasons for exclusion of papers

3.4.1 Description of study design and methodology (objective 1)

Study design

Most of the included studies employed a prospective longitudinal study design (n=18). One study was a randomised controlled trial, but rather than randomising to treatment, randomisation groups were created to measure the effect of administering a pre-test questionnaire (Klaassen et al., 2009). One study conducted a retrospective secondary analysis of data from previous research (Thomson et al., 2014). The majority of the prospective studies employed a single group pretest-posttest study design, with just one study including a cross-matched control group (Baghdadi, 2015). However, OHRQoL was only measured at one time point in this control group, limiting its value in allowing comparison with the intervention group, where a change in OHRQoL was measured.

Included studies were conducted in 14 different countries, with the majority based in a hospital setting and the remaining four studies conducted in a community clinic (Klaassen et al., 2008, 2009; Gaynor and Thomson, 2011; Thomson et al., 2014). Only one study (Klaassen et al., 2009) used random sampling, with the other studies using convenience sampling (n=9) or consecutive sampling (n=10). Nineteen studies were published in English, with just one study published Mandarin which was subsequently translated by a dental colleague (Xiao et al., 2014).

Data collection

The method of data collection varied across the studies and across time points within those studies, and these are detailed in *Table* 2 below.

Table 2: Methods of data collection used in the studies

Data collection method	**Number of studies using this method**	
	At baseline	At follow-up
Structured interview (face to face)	3	2
Structured interview (telephone)	1	2
Self-completed at clinic	12	7
Self-completed by post	0	1
Combination of methods	1	7
Not stated/ not clear	3	1

Most of the studies used self-completed questionnaires on the clinic as the primary method of data collection. For the post-test questionnaire, only seven studies used this method in isolation, with a further seven using a combination of methods. One study used a combination of self-completion on clinic and self-completion by post, depending on which arm of the study a participant had been randomly assigned to (Klaassen et al., 2009). In the remaining six studies, the researchers attempted to use self-completed questionnaires on clinic for the post-test time point, but then conducted structured interviews by telephone (Gaynor and Thomson, 2011; Thomson et al., 2014; de Souza et al., 2016) or self-completed questionnaires by post if participants failed to attend their follow up appointment (Malden et al., 2008; Jankauskiene et al., 2014; de Souza et al., 2016).

In most cases, studies did not report whether it was the same parent/caregiver that completed both the pre- and post-test questionnaires. Only five studies specifically documented that it was the same person in both cases. In three studies it was reported that

a percentage of the questionnaires were completed by different people, ranging from 1.6% (Jankauskiene et al., 2014) to 9.2% (Malden et al., 2008) of instances.

There were marked differences between the studies in the timing of the completion of questionnaires. In ten studies, the pre-test questionnaire was completed on the day of the GA itself (Low et al., 1999; Klaassen et al., 2008, 2009; Malden et al., 2008; Jabarifar et al., 2009; Gaynor and Thomson, 2011; Jankauskiene et al., 2014; Thomson et al., 2014; Yawary et al., 2015; de Souza et al., 2016). In one study (Lee et al., 2011) the questionnaire was completed the day before the GA, and in two studies (Anderson et al., 2004; Baghdadi, 2014) it was between one and two weeks prior to the GA. In the remaining seven studies it was unclear how far in advance of treatment the questionnaires were administered.

In the majority of studies, just one post-test questionnaire was administered. Seven studies carried this out four weeks after treatment (Jabarifar et al., 2009; Klaassen et al., 2009; Gaynor and Thomson, 2011; Almaz et al., 2014; Jankauskiene et al., 2014; Pakdaman et al., 2014; de Souza et al., 2016). Six studies administered the post-test questionnaire earlier than this, between one and four weeks (Anderson et al., 2004; Klaassen et al., 2008; Malden et al., 2008; Cantekin et al., 2014; Thomson et al., 2014; Yawary et al., 2015). Two studies collected data at between four and eight weeks (Low et al., 1999; Baghdadi, 2014), one study at three months (Lee et al., 2011), one study at six months (Xiao et al., 2014) and one study at between six and nine months (Baghdadi, 2015). In two studies it was unclear when the post-test questionnaire was carried out (Thomas and Primosch, 2002; El Batawi et al., 2014). Just two studies administered a second post-test questionnaire and in both cases this was three months after treatment (Pakdaman et al., 2014; Yawary et al., 2015).

Response Rates

The loss to follow-up was reported in fourteen of the studies, with figures ranging from 0% (Jabarifar et al., 2009; Baghdadi, 2015) to 47.8% (Yawary et al., 2015), and a mean loss to follow up of 18.8%. In only seven studies were the characteristics of those lost-to-follow-up participants considered, but these studies found no difference in characteristics between the groups (Malden et al., 2008; Gaynor and Thomson, 2011; Jankauskiene et al., 2014; Pakdaman et al., 2014; Baghdadi, 2015; Yawary et al., 2015; de Souza et al., 2016).

Participant characteristics

The number of participants in the individual studies ranged from 28 (Xiao et al., 2014) to 352 (El Batawi et al., 2014) (median: 88, interquartile range: 68, 140). The age of the children undergoing treatment in the included studies ranged from 2.3 years (El Batawi et al., 2014) to 15.1 years (Malden et al., 2008), with the mean age across all the studies being 4.6 years. In one study (de Souza et al., 2016) children received either comprehensive care or exodontia only treatment under GA, with the remaining studies all involving comprehensive care treatment only. Just ten studies recorded caries experience (Anderson et al., 2004; Klaassen et al., 2008; Lee et al., 2011; Cantekin et al., 2014; El Batawi et al., 2014; Jankauskiene et al., 2014; Pakdaman et al., 2014; Xiao et al., 2014; Baghdadi, 2015; Yawary et al., 2015). Caries experience was recorded as the total number of primary and secondary decayed, missing and filled teeth (dmft/DMFT). At baseline, mean dmft/DMFT in the studies ranged from 6.9 (Cantekin et al., 2014) to 13.3 (Lee et al., 2011). Within individual studies, the caries experience of individuals varied considerably. For example, in the study by Anderson and colleagues, the baseline dmft/DMFT in the study sample ranged from 1 to 18 (Anderson et al., 2004).

3.4.2 Description of oral health-related quality of life instruments used (objective 2)

Table 3 below details the instruments used to measure OHRQoL in each of the studies. Two studies (Low et al., 1999; Pakdaman et al., 2014) designed their own questionnaires with the remainder employing pre-existing questionnaires. However, of these, only nine of the included studies used instruments which had been previously validated in the study population, or included validation of the instrument as part of their study (Gaynor and Thomson, 2011; Lee et al., 2011; Almaz et al., 2014; Baghdadi, 2014, 2015; Cantekin et al., 2014; Pakdaman et al., 2014; Thomson et al., 2014; de Souza et al., 2016). The most commonly used instrument was the Early Childhood Oral Health Impact Scale (ECOHIS), used in nine of the studies. All the studies relied solely on parent/caregiver reported outcomes, most commonly in the form of the ECOHIS or the Parental-Caregiver Perceptions Questionnaire (P-CPQ).

3.4.3 Description of changes in OHRQoL reported in previous studies (objective 3)

There was significant heterogeneity in how the studies reported change in OHRQoL, and therefore a meta-analysis of the data was not possible. A summary of the findings of each paper, ordered by ascending year of publication, is given in *Table 3.* In all the included studies an overall improvement in OHRQoL was seen, however, improvements were not found across all subscales in some studies.

Table 3: Summary of OHRQL instruments and results of the included studies

Study	Instrument	Summary of change in OHRQoL following treatment
Low et al. (1999)	Designed own questionnaire	Reported change in presence of symptoms as follows: the presence of pain reduced from 48% to 3%, problems eating reduced from 43% to 3%, 59% of children began to eat more following treatment, 84% children reported improved sleeping. Number of children with behavioural issues reduced from 4 to 2. Significant changes in all but the 'behavioural issues' category, indicating an overall improvement in OHRQoL.
Thomas & Primosch (2002)	Designed own questionnaire	Overall improvement in OHRQoL reported in 90% of children. Reported reduction in percentage reporting symptoms as follows: complaints about teeth 56% (pre-test) to 2% (post-test), chewing problems 60% to 8%, eating less 52% to 4%, sleeping problems 30% to 4%, behavioural problems 32% to 0%. No statistical significance test carried out.
Anderson et al. (2004)	Designed own questionnaire	The study found a reduction in numbers reporting 'all the time/ often' for all questions post-test compared to pre-test, indicating improvement in all aspects of OHRQoL examined. All changes were statistically significant.
Klaassen et al. (2008)	P-CPQ & FIS	Statistically significant overall change in mean score from 0.73 pre-test to 0.44 post-test indicating improved OHRQoL. The change in the majority of individual subscales was a statistically significant decrease, except for 'emotional wellbeing' where the decrease was not significant, and 'social wellbeing' where there was actually a non-significant increase in score. Pre-test not found to affect results.

Study	Instrument	Summary of change in OHRQoL following treatment
Malden et al. (2008)	P-CPQ & FIS	Mean overall P-CPQ scores reduced from 25.9 to 11.8, mean FIS score reduced from 10.1 to 4.0, with decreases in all P-CPQ subscales, indicating improved OHRQoL. All results were statistically significant.
Jabarifar et al. (2009)	P-CPQ & FIS	Mean scores for P-CPQ decreased from 43.3 to 39.2 and FIS decreased from 8.0 to 3.7, indicating improved OHRQoL. Results were all statistically significant. Effect sizes were large for all subscales except 'emotional wellbeing' which had a moderate effect size.
Klaassen et al. (2009)	ECOHIS	Mean total ECOHIS reduced from 12.9 to 7.4, which was statistically significant and indicated improved OHRQoL. Pre-test was found to have no effect.
Gaynor & Thomson (2011)	P-CPQ & FIS	Decrease in mean overall P-CPQ score from 22.8 to 8.8 and mean overall FIS score from 8.7 to 4.4, indicating improved OHRQoL, which was statistically significant. Significant decreases were seen in all P-CPQ and FIS subscale scores also. Effect sizes were large for P-CPQ and moderate for FIS.
Lee et al. (2011)	ECOHIS	27.6% reduction in overall ECOHIS score which was statistically significant with large effect size, indicating improved OHRQoL overall. For the individual subscales, statistically significant reduction in scores was found with moderate effect sizes for all subscales except two. 'family function' had a non-significant decrease, and 'child self-image and social interaction' had a non-significant increase.
Almaz et al. (2014)	ECOHIS	54.7 % reduction in total score, 48.4% in CIS and 67.4% in FIS. The decrease in scores was seen in all subscales, and all changes were statistically significant. Effect sizes were large for all subscales except 'child psychology' and 'child self-image and social interaction' (small effect size) and 'family function' (moderate effect size).

Study	Instrument	Summary of change in OHRQoL following treatment
Baghdadi (2014)	Short form P-CPQ & FIS	Statistically significant decreases in overall and all individual subscale scores in P-CPQ and FIS following treatment, with mostly large effect sizes. The 'social wellbeing' and 'parental emotions' subscales showed moderate effect sizes.
Cantekin et al. (2014)	ECOHIS	Overall score decreased by 44%, CIS by 34%, FIS by 65%, indicating improved OHRQoL. Statistically significant decrease in mean scores was seen in all subscales, except the 'child self-image and social interaction subscale which showed a significant increase in score.
El Batawi et al. (2014)	Modified P-CPQ & FIS	Reduction in the percentage of individuals reporting all outcomes, indicating improved OHRQoL. No statistical test carried out.
Jankauskiene et al. (2014)	ECOHIS	Overall and all individual subscale scores decreased after treatment and all changes were statistically significant. Large effect sizes for all but the 'child self-image and social interaction' subscale where the effect size was small.
Pakdaman et al. (2014)	ECOHIS	Mean scores for both the child and parent subscales decreased at both the first (4 weeks) and second (3 months) follow up, and these changes were statistically significant compared to baseline. The change between 4 weeks and 3 months, however, was not statistically significant.
Thomson et al. (2014)	ECOHIS	Mean ECOHIS-child score decreased from 7.7 to 2.6 with large effect size and mean ECOHIS-family score decreased from 3.8 to 1.8 with moderate effect size, indicating improved OHRQoL. Both changes were statistically significant.
Xiao et al. (2014)	ECOHIS	Mean scores for ECOHIS overall and all subdomains showed statistically significant decreases.
Baghdadi (2015)	P-CPQ & FIS	Mean scores for the P-CPQ and FIS showed a statistically significant decrease, with large effect size, indicating improved OHRQoL.

Study	Instrument	Summary of change in OHRQoL following treatment
Yawary et al. (2015)	ECOHIS, P-CPQ and FIS	ECOHIS, P-CPQ and FIS overall and subscale mean scores all showed a statistically significant decrease at 2 weeks and 3 months, indicating improved OHRQoL. The decrease in mean scores between 2 weeks and 3 months, however, was not statistically significant. Effect sizes were large to moderate for all subscales and large overall.
de Souza et al. (2016)	P-CPQ & FIS	Statistically significant reduction in overall scores and all individual subscales with medium to large effect sizes, indicating improved OHRQoL. No significant difference was found between treatment groups (exodontia only versus comprehensive care).

All but one study (Thomas and Primosch, 2002) applied statistical tests to determine whether there were significant differences in OHRQoL following treatment. Most of the studies found a significant change in both overall and subscale scores, regardless of which instrument was used. However, some studies found that within the subscales there was not always a significant change in score (Klaassen et al., 2008; Lee et al., 2011).

Interestingly, some studies found an increase in some subscale scores, i.e. worsened OHRQoL, following the dental GA. For example, two studies (Lee et al., 2011; Cantekin et al., 2014) found an increase in mean score for the ECOHIS 'child self-image and social interaction' subscale and another study (Klaassen et al., 2008) found an increase in mean P-CPQ 'social wellbeing' subscale score, albeit not statistically significant.

As well as considering if the change was statistically significant, three studies (Malden et al., 2008; Gaynor and Thomson, 2011; de Souza et al., 2016) looked at whether

the change was clinically significant by calculating the minimally important difference (MID). Two of these found 63% of the population showed or exceeded the MID for the P-CPQ, but only 40% did so for the Family Impact Scale (FIS) (Malden et al., 2008; Gaynor and Thomson, 2011), whereas de Souza and colleagues found 54% of the population showing or exceeding the MID for the P-CPQ and 65% for the FIS (de Souza et al., 2016).

Eleven studies included a measure of effect size, the results of which are also given in *Table 3*. Large to moderate effect sizes were seen for overall changes and in all subscales, with the exceptions being the small effect sizes seen in the ECOHIS 'child psychology' (Almaz et al., 2014) and ECOHIS 'child self-image and social interaction' subscales (Almaz et al., 2014; Jankauskiene et al., 2014).

Half of the studies asked a global transition judgement (GTJ) question, which are patient-level assessments of how great or significant they perceived the overall change in oral health-related quality of life to be. However, of the ten studies which included a GTJ question, seven did not then correlate this to the change in OHRQoL scores as recommended by the COSMIN group in order to help assess the responsiveness of the measure to change over time.

Secondary outcomes

In addition to patient-reported outcomes, two studies also looked at clinical outcomes, in the form of ongoing caries experience (El Batawi et al., 2014; Xiao et al., 2014). El Batawi and colleagues (El Batawi et al., 2014) found 59% of participants had new carious lesions within two years of treatment and Xiao and co-workers (Xiao et al., 2014) found that 37% of participants had new carious lesions after six months.

Dental anxiety was assessed in three studies, which used the Dental Subscale of the Children's Fear Survey Schedule (CFSS-DS) (Klaassen et al., 2008, 2009; Cantekin et al., 2014). In the studies by Klaassen and co-workers, no significant difference was found between pre-test and post-test anxiety scores (Klaassen et al., 2008, 2009). In contrast, Cantekin and colleagues found a statistically significant decrease in CFSS-DS score post-treatment, indicating an increase in dental anxiety (Cantekin et al., 2014).

Four studies reported parental satisfaction following treatment, all of which developed their own questionnaires for this purpose. No statistical tests were applied to these data, but all four studies found high levels of parental satisfaction, with 80-100% of parents reported as being 'satisfied' with the treatment (Anderson et al., 2004; Almaz et al., 2014; El Batawi et al., 2014; Jankauskiene et al., 2014).

3.4.4 Quality assessment of the included studies (objective 4)

Study quality varied considerably, and out of a total possible QATSDD score of 42, scores for the individual studies ranged from 7 (Thomas and Primosch, 2002) to 32 (Gaynor and Thomson, 2011). The average score was 22(±7).

Table 4 shows the mean score for each of the 14 criteria of the quality assessment. A mean score of 0 indicates none of the papers met any of the components of the criteria, with a total possible score of 3 indicating all the papers fully met the criteria.

Table 4: Mean score, standard deviation (SD) and range for each quality criteria against which the papers were assessed (possible score range 0-3)

Quality criteria	**Mean score (SD, range)**
Evidence of user involvement in design	0 (0, 0)
Explicit theoretical framework	0.2 (0.5, 0-2)
Evidence of sample size considered in terms of analysis	1.1 (1.4, 0-3)
Statistical assessment of reliability and validity of measurement tools	1.2 (1.3, 0-3)
Strengths and limitations critically discussed	1.2 (1.0, 0-3)
Good justification for the method of analysis	1.3 (1.0, 0-2)
Clear description of research setting	1.8 (0.7, 1-3)
Description of the procedure for data collection	1.9 (0.6, 1-3)
Statement of aims/objectives in body of report	2.0 (0.6, 0-3)
Detailed recruitment data (no. approached, declined etc)	2.0 (0.8, 1-3)
A representative sample of reasonable size	2.1 (0.7, 1-3)
The rationale for the choice of data collection tool	2.3 (1.1, 0-3)
Fit between the research question and method of analysis	2.5 (0.8, 1-3)
Fit between the research question and method of data collection	2.7 (0.7, 1-3)

Some quality criteria were well addressed by the included studies, in particular, the fit between the research question and method of data collection and analysis. However, none of the included papers had evidence of user involvement in the design and there was a lack of explicit theoretical framework underpinning the majority of the studies. Two other

areas less well addressed were the approaches taken to estimate the sample size and assessment of reliability and validity of the measurement tools used.

3.5 Inter-reviewer reliability

The two independent reviewers agreed on 115/121 (95%) abstracts when screened against the inclusion and exclusion criteria, and the disagreements for selection/omission were resolved by discussion. Independent reviewers then agreed on 26/28 (93%) full-text articles screened against the inclusion and exclusion criteria, and again the disagreements were resolved by discussion.

At the data extraction stage, independent reviewers agreed on 605/684 (88.5%) data extraction criteria, with the areas of disagreement resolved through discussion. For the quality assessment, there was an overall agreement between independent reviewers of 176/280 (62.9%) of quality criteria scores. However, because the quality assessment was an ordered variable, a weighted kappa was also carried out to establish relative concordance between reviewers. It was assumed that the differences between individual quality scores were equal. The inter-rater agreement (kappa with linear weighting) was 0.65 (95% CI, 0.59- 0.72) indicating substantial agreement.

3.6 Discussion

This systematic review examined 20 studies, reported across 22 papers. It was clear that all of the studies reported an overall improvement in OHRQoL in children following dental treatment under GA. Within studies, however, there were differences in the change score for individual subscales. Interestingly, in some cases, results suggested that some aspects of OHRQoL may worsen following dental treatment under GA. There could be several reasons for this. Many of the studies carried out the post-test questionnaire within four weeks following treatment, at which point children may still be experiencing discomfort from extraction sites or difficulty eating due to the number or difficulty of extractions. It is unclear what the longer-term impacts of treatment might be, and whether these aspects of the OHRQoL measures would improve over time. Further research is indicated to add to this body of evidence. Future work should explore whether the actual number of extractions impacts on OHRQoL; one might expect that children who have higher numbers of extractions are more likely to experience the negative side effects of post-treatment discomfort or impaired function compared to those who have single or few extractions. One might also need to consider the difficulty of extraction or whether it was primary teeth or permanent teeth removed. For example, one might expect that removal of first permanent molars, which necessitated the use of elevators or post-treatment sutures may be associated with greater post-morbidity than simple forceps extractions of first primary molars.

In contrast with the previous systematic review (Jankauskiene and Narbutaite, 2010), the majority of studies involved instruments which had been used in other studies, with just three studies developing their own questionnaires (Low et al., 1999; Pakdaman et

al., 2014). However, less than half of the included studies used instruments that had been previously validated for the study population or included validation of the instrument as part of their study. Importantly, it has been shown that the properties of quality of life instruments should be evaluated when used in a different context to the one in which they were developed (Aaronson et al., 2002). There is, therefore, still a need for further research using validated instruments to evaluate the change in OHRQoL following a dental GA, and for longitudinal validation of OHRQoL instruments.

Despite recommendations made in previous studies (Klaassen et al., 2008; Malden et al., 2008; Jabarifar et al., 2009; Jankauskiene and Narbutaite, 2010) none of the studies to date had employed a child-reported measure of OHRQoL, instead relying on proxy reports of OHRQoL. Caution should, therefore, be exercised when interpreting some of the findings as it has been shown that parents/caregivers generally have a low to moderate overall agreement with their child's ratings (Achenbach et al., 1987; Wilson-Genderson et al., 2007). A systematic review of parent and child reports of health-related quality of life (HRQoL) by Eiser and Morse (2001) revealed greater agreement between proxy and child ratings in some subscales (e.g. physical HRQoL) than other, less-observable, subscales (e.g. emotional or social HRQoL). This highlights the need for child-reported measures to be used in future OHRQoL research.

Another limitation of some of the included studies was that, in some instances, different individuals completed the pre-test and post-test questionnaires. These change scores were included in the final analysis, despite this potentially impacting on the scores. Future studies should ensure consistency in respondents for all time points in the study.

The use of convenience samples and lack of controls needs consideration. A consecutive sample would be preferable as it would better represent the whole population. The majority of studies highlight the issues in obtaining a suitable control for this population, as withholding treatment for a control group would be unethical. Where random allocation is not possible, it may be possible to improve the validity of inferences by using statistical techniques to adjust for potential confounders (Bonell et al., 2011). The disadvantage of this option is that to adequately adjust for confounders, potential confounding variables must be identified and accurately measured. Inadequate identification and measurement of confounding factors have been identified as a deficit in observational studies (von Elm et al., 2014). A clear framework underpinning the research is important for understanding which factors may impact the outcome of interest; something which is lacking in all the included studies. To improve the quality of future research, the underpinning theoretical framework should be clearly stated.

While all the studies stated that children were undergoing a GA for the treatment of dental caries alone, only ten studies recorded the level of caries experience of their sample. Recording caries experience and number of decayed teeth, e.g. as dmft/DMFT, would have been useful to give an indication of the burden of disease in the study population, and also because this may influence changes in OHRQoL following treatment. Interestingly, even in those studies which reported caries experience using dmft/DMFT, there was no reference to this in the subsequent analysis or discussions. It might have been useful for caries experience to have been taken into account and perhaps correlated against pre-test OHRQoL scores.

Only one study considered the difference in change in OHRQoL according to treatment approach and found no significant difference between extraction only and comprehensive care (i.e. including restorations) groups (de Souza et al., 2016). However, the sample size was relatively small and more extensive studies should be carried out to validate this finding.

3.7 Study strengths and limitations

3.7.1 Strengths

The present review employed a comprehensive search strategy and should therefore fully represent the current literature base. The inter-reviewer reliability assessments show a substantial level of agreement, adding to the reliability of the findings. This is the first time a quality assessment of the included studies has been carried out, which has shown there is significant variability in the quality of the studies reporting OHRQoL changes. This assessment also highlighted key areas for improvement in quality, which is of use to those planning future research. The findings have also highlighted areas of discrepancy in the current literature, for example, variation within subscales of the measures following the intervention.

3.7.2 Limitations

There was significant heterogeneity between the studies so it was not possible to carry out a meta-analysis of the findings, which also limits the conclusions that can be drawn. During the literature search, hand-searching of journals was not undertaken thereby

potentially omitting some articles. However, it was felt that through thorough database searching, citation searching and reference searching that the search strategy should have been exhaustive, without the need for hand searching. The inclusion of all papers, regardless of language, presented a difficulty with obtaining an accurate translation. Although it was possible to extract all of the relevant data for inclusion in the review itself, it was not possible to calibrate the translator to use the QATSDD tool and therefore the quality assessment for this paper was not completed. Quality assessment may have an element of subjectivity and the QATSDD tool does not weight individual criteria by importance or degree of impact on quality. The total scores should, therefore, be used with caution. Subjectivity was, however, reduced by having more than one reviewer complete the quality assessment for all the studies.

3.7.3 Novel aspects

This systematic review presents an updated appraisal of the evidence presented by Jankauskiene and Narbutaite (Jankauskiene and Narbutaite, 2010), and while building on this previous body of work,some novel aspects have emerged. This review focused exclusively on studies reporting a change in OHRQoL, whereas the previous review also considered OHRQoL reports at a single time point. This review also included all papers regardless of language, although this approach only resulted in the inclusion of one additional study. The systematic review in this chapter is the first to examine the quality of papers presenting data on OHRQoL in children undergoing treatment for dental caries under GA.

3.8 Summary

The included studies provide evidence which would justify the use of GA in the treatment of dental caries due to overall proxy-reported improvements in OHRQoL. However, there is a lack of evidence on the impact of dental caries and treatment under GA from a child's perspective. The results also highlight the need for future research to compare the impact of different treatment approaches and to examine the long-term impact of treatment, which in turn will better inform clinical practice and provide justification for treatment options. The quality assessment of the included studies demonstrated variable quality in articles reporting a change in OHRQoL in children undergoing treatment for dental caries under GA, highlighting areas for improvement in the design of future studies.

3.9 Implications for this thesis

The findings of this systematic review have highlighted gaps in the current knowledge base, which have implications for this PhD research project. The first of these is the lack of data arising from children themselves, with most of the work relying on proxy-reported measures of OHRQoL. Therefore, these findings suggest the need for research involving a child-reported measure and, preferably, one which involved children in the design, and which is also disease (caries) specific. There is also a scarcity of research examining the effect of comprehensive care under GA and exodontia only, and the impact of treatment in the mid- to long-term.

In addition, there were quality issues with the previous studies which should be addressed in future work. Firstly, there has been a lack of a clear theoretical framework

underpinning the research to date. As already discussed in *Chapter 2*, this is a common limitation with the quality of life literature more generally. While there are currently no models available specifically relevant for research with children, the Wilson and Cleary model of health-related quality of life has been chosen as an underlying framework for this work and adapted accordingly (see *Chapter 4*). Ideally, a suitable sample size should be recruited which would then allow testing of this model against the data.

3.10 Update to the systematic review

It is widely acknowledged that findings and conclusions from systematic reviews may be superseded by new evidence, even before publication of the review (Beller et al., 2013). An update to this systematic review was therefore conducted in June 2019, to establish whether there had been any significant change in the literature since completion of this PhD research study. The repeat literature search was performed as described for the initial review (see *Section 3.3.2*), to identify papers from January 2016 onwards, and the results of this search are given below.

3.10.1 Results from updated search

The search strategy yielded an additional 18 records. Two articles were excluded as duplicates from the original systematic review, which were print publications following earlier e-publication (Yawary et al., 2016; de Souza et al., 2017). One article was excluded as it was a summary review of this published systematic review (Martins-Junior, 2017). Following the screening of the remaining titles and abstracts against the eligibility criteria, nine full papers where retrieved which appeared to be eligible for inclusion in the update.

Following the screening of the full papers against the inclusion criteria, two further papers were excluded. Therefore, the literature search identified seven additional papers which were eligible for inclusion.

Description of study design and methodology

Six of the studies were prospective longitudinal studies and the remaining article was a meta-analysis of previous studies (Park et al., 2018). The findings from the meta-analysis are considered separately below.

The prospective studies were conducted in six different countries, all in a hospital setting. All the studies used convenience sampling. As before, the data collection method varied across the studies (*Table* 5). In most cases, it was not clear at what time-point prior to treatment the pretest questionnaire was completed, with just two studies stating it was administered at the time of the GA itself (Jankauskiene et al. 2017; Hashim et al. 2019). As before, most studies used a short-term follow-up period of 1-month post-treatment, with just one study looking at the longer term impacts of treatment at both the 1-month and 6-month recall period (Jankauskiene et al. 2017).

Table 5: Methods of data collection used in the studies

Data collection method	**Number of studies using this method**	
	At baseline	At follow-up
Structured interview (face to face)	2	2
Self-completed at clinic	2	2
Not stated/ not clear	2	2

Oral health-related quality of life findings

The most commonly employed OHRQoL questionnaire was the ECOHIS, used in five of the six studies. Only one study employed a child-reported measure, using the CPQ_{11-14} (Brondani et al., 2018).

All of the studies reported statistically significant reduction in mean total scores from completed questionnaires (i.e. improvement in OHRQoL) at one month following treatment under GA, with moderate to large effect sizes, where these were reported (Jankauskien et al., 2017; Brondani et al., 2018; Hashim et al., 2019). Jankauskiene and colleagues (2017) found that ECOHIS scores remained significantly lower at six months following GA, suggesting the improvement in OHRQoL was maintained over time.

A number of studies reflected on the treatment approach (i.e. GA versus other management technique, or restorative versus exodontia treatment) in their findings. However, there were some limitations to these discussions. Rane and colleagues (2017) compared treatment under GA with treatment under LA and found no significant difference in change in overall ECOHIS scores between the two treatment approaches. However, no information was provided as to what specific treatment items were carried out in each group. The only information available was that all participants had 'at least five deeply carious teeth, which required pulpotomy or pulpectomy, followed by stainless steel crowns' at assessment. It is not clear whether these teeth were always restored, or in some cases whether they were extracted. Therefore, while this study explored the effect of method of treatment delivery, it did not explore the treatments given in detail.

In contrast, Guney and colleagues (2018) compared treatment carried out for GA and Intravenous Sedation (IVS) groups, including numbers of teeth extracted and restored. They found that there was no statistically significant difference in the number of extractions between the two groups but, on average, those having comprehensive care under GA had more teeth operated on than those in the IVS group. No significant difference was found in mean ECOHIS scores between the groups, although it is worth noting that OHRQoL scores were not directly correlated with numbers of restorations or extractions carried out.

One further study recorded treatment approach, but only presented the results for 'all participants' and those who 'only received restorations'; no separate analyses were given for the exodontia only or combined care groups (Brondani et al., 2018). The authors found that both groups (all participants versus those who only received restorations) reported statistically significant improvements in OHRQoL, but no comparison of change scores for each group was given. Furthermore, it was not possible to determine from the results given how many teeth had been restored, nor how many extractions were carried out in the extraction only group, or whether there was any difference in change in OHRQoL in the extraction only group, limiting the usefulness of these findings.

Other findings

Guney and colleagues (2018) also examined the effect of treatment on anxiety, using the Venham Picture Test (VPT) for children aged 3-5 years and the children's fear survey schedule- dental subscale (CFSS-DS) for children aged 6-12 years. They found a significant decrease in VPT and CFSS-DS scores following treatment under GA, and a significant decrease in CFSS-DS following IVS. There was no statistically significant change in VPT scores following IVS. However, it was unclear at what point before and

after treatment the questionnaires were administered, so it is difficult to draw many conclusions from the data. Dental anxiety may be one of the main reasons for children being unable to accept treatment under LA and thus been offered GA or sedation. It may be that pharmacological interventions do not reduce anxiety *per se*, but rather manage it. The wider topic of using dental anxiety as an outcome measure for children receiving dental treatment under GA will therefore be considered in more detail in the general discussion in *Chapter 6*.

Updated meta-analysis findings

Park and colleagues (2018) included 22 research articles in their recent review, all of which used either the ECOHIS or C-OHRQoL measures. The full details of these have been explored earlier in this chapter. The meta-analysis revealed statistically significant improvements in OHRQoL following treatment under GA, with moderate to large effect sizes for both the ECOHIS and C-OHRQoL measure groups. However, there were significant clinical and methodological differences between the studies, so the conclusions that can be drawn from this are limited. For example, there were differences in the time period of follow-up, protocols for how the questionnaires weere administered, and the caries experience of the study populations. The authors drew similar conclusions, however, that there remained a need for research investigating the longer-term impacts of treatment, and for appropriate theoretical models to guide future studies.

Quality of the additonal studies

Study quality was assessed using the QATSDD assessment tool. *Table 6* shows the mean score for each of the 14 criteria of the quality assessment.

Table 6: Mean score, standard deviation (SD) and range for each quality criteria against which the papers were assessed (possible score range 0-3)

Quality criteria	**Mean score (SD, range)**
Explicit theoretical framework	0 (0, 0)
Evidence of user involvement in design	0.2 (0.4, 0-1)
Statistical assessment of reliability and validity of measurement tools	0.5 (1.2, 0-3)
Evidence of sample size considered in terms of analysis	0.8 (1.3, 0-3)
The rationale for the choice of data collection tool	0.8 (1.2, 0-3)
Clear description of research setting	1.5 (0.5, 1-2)
Detailed recruitment data (no. approached, declined etc)	1.5 (0.8, 1-3)
A representative sample of reasonable size	1.5 (0.8, 1-3)
Good justification for the method of analysis	1.6 (0.6, 0-3)
Strengths and limitations critically discussed	1.7 (0.5, 1-3)
Description of the procedure for data collection	1.8 (0.9, 1-3)
Statement of aims/objectives in body of report	2.3 (0.6, 1-3)
Fit between the research question and method of analysis	2.5 (0.6, 1-3)
Fit between the research question and method of data collection	2.7 (0.7, 1-3)

The updated review revealed very similar quality assessment results to those found in the earlier systematic review, with study quality varying considerably. Overall quality scores from the studies ranged from 11 (Li *et al.*, 2017) to 32 (Hashim et al., 2019). The average score was 17.8 (±7.9). As found previously, the quality criteria which were least well addressed were the lack of an explicit theoretical framework underpinning the research and evidence of user involvement in the study design.

3.10.2 Discussion

The findings from the update did not differ significantly from those in the original systematic review. The majority of the studies were prospective, longitudinal studies, which used a variety of measures to collect data on OHRQoL. Significantly, since the

publication of the original systematic review, only one study sought to obtain the views of children themselves (Brondani et al., 2018) and the majority of studies relied on proxy-reported measures of OHRQoL.

Studies which looked at treatment approach did not investigate the type of treatment carried out in detail (e.g. numbers of extractions), and only one paper related treatment approach (GA versus IVS) to OHRQoL outcomes (Guney et al., 2018). None of the studies explored the impact of different treatment approaches on OHRQoL from a child's perspective, reinforcing the importance of the work in this thesis to explore the potential impact of treatment on the everyday lives of children, from their own perspective.

The findings of this updated review have confirmed that, since publication of the 2016 systematic review, the same gaps have remained in the knowledge base. These gaps, namely, the lack of data arising from children themselves, the limited number of longer-term follow-up studies (greater than one month) and the limited investigation into the effect of treatment approach, have all been addressed in this PhD. The update has, therefore, confirmed the novel aspects of this PhD which will contribute to the field. In addition, none of the previous studies had identified a clear theoretical framework underpinning their research. This thesis, therefore, adds to the body of work to date by proposing a theoretical framework and exploring the impact of clinical, individual and environmental factors on OHRQoL following treatment under GA. It is acknowledged, however, that this is an area of ongoing interest, and new studies are continually being added to the knowledge base.

Chapter 4: Materials and methods

4.1 Overview

This study employed a pretest-posttest design. Potential participants were assessed against eligibility criteria. Eligible participants, aged 5-16 years old who were having treatment for dental caries under general anaesthetic (GA), were recruited from new patient clinics at the Charles Clifford Dental Hospital, Sheffield. Oral Health-Related Quality of Life (OHRQoL) was measured before and three months after treatment under GA using the Caries Impacts and Experiences Questionnaire for Children (CARIES-QC). Overall child reported quality of life was measured at baseline and follow-up using the Child Health Utility 9D (CHU9D). Additionally, the parents/caregivers of children participating in the study were asked to complete the Family Impact Scale (FIS) questionnaire at two separate time points, baseline and at the three-months following their child's GA. All questionnaires were anonymised. Results from the questionnaires were inputted into an electronic database (IBM SPSS Statistics, version 24) and subject to appropriate statistical analysis, which included exploring changes after treatment for each measure using Wilcoxon tests. Path analysis was conducted using appropriate software (StataCorp STATA, version 15), to investigate the relationships between clinical, individual and environmental factors and quality of life outcomes, guided by the theoretical model.

4.2 Rationale

As highlighted in the preceding chapters, dental caries is a significant public health problem worldwide (Kassebaum et al., 2015). The negative impact of dental caries on children and their families is well documented, and is associated with a number of factors which affect quality of life in children, including pain, impaired function and loss of school days (Alsumait et al., 2015; Baghdadi, 2015; Gilchrist et al., 2015). However, what is less well understood is the subjective impact of caries from the view of children themselves. Many paediatric patients with dental caries receive treatment under general anaesthesia (GA). In the UK, the approach for the management of caries under GA varies across the country. In some instances, exodontia (extraction) only is offered, while other providers may offer comprehensive care (a combination of extractions and restorations) where appropriate. Waiting times for a dental GA are also variable across the UK.

Over recent years there has been a move beyond the evaluation of clinical outcomes alone to also evaluate health interventions using patient reported outcome measures (PROMs). This transition reflects changing views of health, from a medical model to multidimensional models which view health more holistically. Oral health-related quality of life (OHRQoL) is one such PROM, which aims to evaluate the impact of oral health on everyday life.

The systematic review, presented in *Chapter 3*, revealed that a range of measures exist to assess OHRQoL in children, but the majority of these rely on parent or caregiver proxy reports (Knapp et al., 2017). To date, there has been limited research involving children themselves, furthermore, it is recognised that proxy assessment may not always

match the child perceptions (Filstrup et al., 2003). In addition, the majority of measures of OHRQoL have not included children in their development and therefore may not be valid in this population (Gilchrist et al., 2014). Further investigation into the impact of dental caries from the child's perspective using OHRQoL instruments which have been developed with, and are completed by, children themselves is therefore warranted.

In addition, while previous studies have used generic measures to assess the impacts of caries in children, these may not be sensitive enough to measure impacts related to caries specifically. The use of a caries-specific measure to investigate the impact of caries and its treatment would be of benefit to ensure the impacts of dental caries on children's daily lives are fully assessed. The systematic review also revealed that there has been little research to assess long-term changes in OHRQoL in children following a dental GA, or if the changes are affected by the treatment approach. Further investigation to explore these key variables was warranted to better inform future dental GA services for children.

4.3 Theoretical framework

As discussed in *Chapter 2*, the Wilson and Cleary (1995) conceptual model of health-related quality of life was used to underpin the proposed research. This provided the theoretical basis for developing a specific model of OHRQoL in order to guide the study design and identify variables for inclusion in the final path analyses. This model incorporates five aspects, namely, biological or physiological factors, symptom status, functional status, general health perceptions and overall quality of life. It also incorporates individual and environmental characteristics.

Variables were identified from the literature which might impact on OHRQoL and these were incorporated into an adapted version of the Wilson and Cleary model. Although in the original model 'symptom' and 'function' status are assessed separately, it was felt that children are less able to make the distinction between the two. This decision was based on findings during the development of CARIES-QC, where children would combine impacts to access how they were affected by their teeth, for example, how often food gets stuck in their teeth combined with how much it hurts when it does (Gilchrist, 2015). Therefore, in the adapted model, symptom and function status were assessed concurrently using the CARIES-QC measure of OHRQoL. Other measures of child reported OHRQoL were discounted as less suitable, as discussed in *Chapter 2*. The final proposed theoretical model is shown in *Figure 4*.

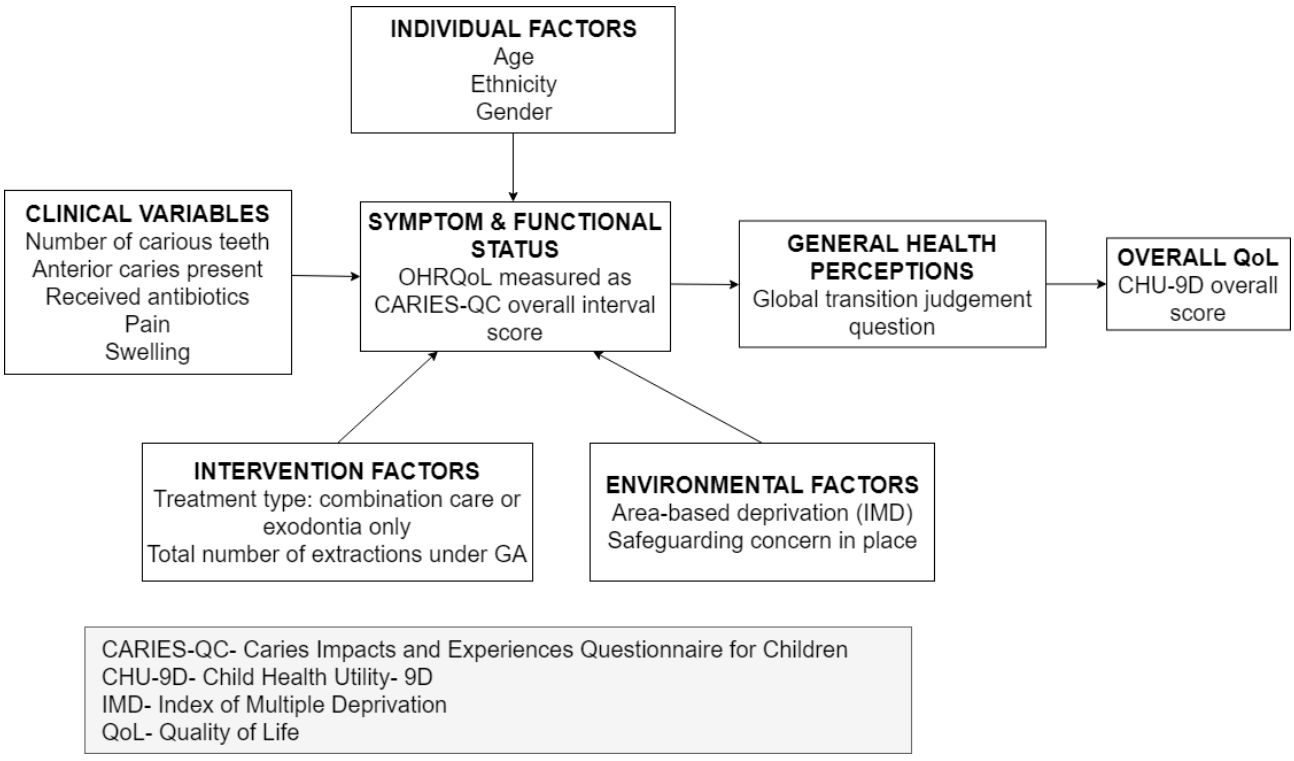

Figure 4: Proposed theoretical framework, adapted from Wilson and Cleary (1995) model of HRQoL

In this model, clinical variables included the caries experience of the child, measured as the number of carious teeth, whether the child had received antibiotics for their dental problem, whether there was a swelling present and whether the child was in pain. Intervention factors included the type of treatment, either extraction only or combination care involving restorations and extractions, and the total number of extractions carried out. Environmental factors in this model were deprivation, measured by Index of Multiple Deprivation scores derived from the postcode and whether the child was a safeguarding concern. Further information on these variables, including how they were measured, is given in *Section 4.4.7* below.

4.4 Methods

4.4.1 Study design

As in previous studies, an observational approach was adopted and a prospective longitudinal pretest-posttest study design employed, which is the most commonly used study design for this type of research (Yawary et al., 2015; de Souza et al., 2016).

While complete control for confounding is not possible in a non-randomised study, measured characteristics that differ between individuals (e.g. child age or deprivation) were considered in path analyses to examine their potential impact on the outcomes. Further details of the variables included in the final path analyses are given in *Section 4.7.7.2* below.

The study was designed to test the following hypotheses:

1. Treatment for the management of dental caries under GA results in improved OHRQoL, as reported by children.
2. Treatment for the management of dental caries under GA has a positive effect on the family.
3. Participants receiving restorations as well as extractions report greater improvements in OHRQoL change scores than those receiving extractions only.

4.4.2 Setting for the project

Participants were recruited from consultant-led new patient clinics at the Charles Clifford Dental Hospital paediatric department, Sheffield. This unit has seen a year on year increase in the number of referrals received, which exceeds agreed capacity and places considerable demands on the service. In the last financial year (2018/9) this department conducted over 4500 new patient assessments. Around 45% (n=2039) of those who attended such assessments received treatment under GA for dental caries, of which there were 1205 cases of exodontia only and 468 of comprehensive care. This department receives referrals from a wide geographical area, including South Yorkshire, Lincolnshire, Nottinghamshire and Derbyshire.

4.4.3 Ethical approval

This study was approved by the South East Scotland Research Ethics Committee (reference: 16/SS/0178) in December 2016. The documentation regarding this approval is given in *Appendix 1*. This approval was based on the submission of the following documents to be used during the study, which are included in the appendices listed below:

- Age-appropriate child information sheets, for children aged 5-7, 8-11 and 12-16 (*Appendix 2*)
- Parent/ caregiver information sheet (*Appendix 2*)
- Child initial and follow-up consent and questionnaires (*Appendix 3*)
- Parent/ caregiver initial and follow-up consent and questionnaires (*Appendix 4*)
- Letter to parent/ caregivers three months following treatment: (*Appendix 5*)
- Letter to parent/ caregivers if questionnaire not returned (*Appendix 5*)
- Completion of the study thank you letter (*Appendix 5*)

Due to limited numbers of children undergoing comprehensive care and the lengthy waiting times for comprehensive care treatment following assessment, a subsequent amendment was made to the study to allow direct recruitment from the GA waiting list. This amendment was approved in March 2018, and a copy of the letter sent to parents of children who were about to undergo comprehensive care treatment under GA can be found in *Appendix 6*. The documentation regarding this amendment approval is also found in *Appendix 6*. However, unfortunately, recruitment via letter from the waiting list did not result in any additional participants being recruited to the study, and therefore this approach is not reported on further. Difficulties with recruitment are discussed in more detail in *Chapter 6*.

4.4.4 Eligibility

The following inclusion and exclusion criteria were used in deciding whether to approach potential participants for the study:

Inclusion criteria

- Children aged 5 to 16 years
- Children with active dental caries
- Children who are otherwise medically fit and well (i.e those who were recorded as American Society of Anaesthesiologists classification (ASA) 1 in the patient record)
- Children able to understand spoken English, i.e. able to understand and undertake the research with support
- Children with parents/ caregivers who could understand spoken English, i.e. able to understand and undertake the research with support

Exclusion criteria

- Children with caries in conjunction with other dental conditions such as a traumatic dental injury or a dental anomaly such as molar incisor hypomineralisation, amelogenesis imperfecta or dentinogenesis imperfecta
- Children having treatment under GA for other dental reasons (e.g. surgical removal of unerupted teeth)
- Children with other medical conditions (ASA 2 or above)
- Children who are unable to understand and undertake the research even with support

- Children with parents/ caregivers who are unable to understand and undertake the research even with support

4.4.5 Sample size

4.4.5.1 Sample size to detect effect of treatment

The systematic review conducted prior to study commencement revealed a range of sample sizes in previous studies of OHRQoL from ranged from 28 (Xiao et al., 2014) to 352 (El Batawi et al., 2014) (median: 88, interquartile range: 68, 140) with a mean sample size of 116. A sample size calculation was carried out using G*Power that revealed a sample size of 42 would be needed to detect an effect size of 0.8 at 5% level of significance and 80% power in order to test the proposed hypotheses. The Wilcoxon test, used to analyse change in total score for each of the measures, was used for this calculation

4.4.5.2 Sample size required for path analysis

There are relatively few studies which have employed path analysis, and limited guidelines available to inform calculation of adequate sample sizes when conducting path analysis. Therefore, for the sample size calculation, the recommendations for structural equation modelling (SEM) have been employed, which is considered suitable based on the fact that path analysis is a subtype of structural equation modelling (Stage, Carter, and Nora 2004). One classical rule of thumb in SEM is for between 5 and 10 participants per variable in the model (Bentler and Chou, 1987), although more recently Kline (2005) recommended a minimum of 10 cases per variable. Based on these simple rules of thumb, a minimum sample size of 60-120 would be required to test the proposed model, with the consensus

being that the greater the sample size, the greater the power. However, it has been noted that smaller sample sizes may be applicable where there are no latent variables in the model (Kenny and McCoach, 2003), as is the case in this study.

Another key aspect of sample size determination is whether the sample size is sufficiently powered for the calculation of Chi square (χ^2) and fit indices (Kim, 2005). The recommended sample size for χ^2 is 75-100. This upper limit of 100 is set because χ^2 is highly sensitive to sample size change and will almost always be significant (indicating a poor fit) with higher sample sizes (Iacobucci 2010). Three additional fit indices were employed to examine model fit to the data in this study, namely Root Mean Square Error of Approximation (RMSEA), Comparative Fit Index (CFI) and Tucker-Lewis Index (TLI). For the RMSEA fit index, the sample size calculator revealed a minimum sample size of 62 was required to test the full model (power=0.8, alpha=0.05) (Maccallum et al., 1996). CFI and TLI indices are not as sensitive to sample size changes, hence the decision to include them in this study. In fact, studies suggest that a minimum sample size of 50 is required for calculation of these indices, after which any increases in sample size have little impact (Iacobucci, 2010).

Based on all these considerations, the required minimum sample size was set at 75, based on the χ^2 requirements, with the recognition that an absolute minimum sample size of 62 would be required for most of the model fit analyses to be performed.

4.4.5.3 Overall recruitment requirements

Based on findings from the systematic review, and previous research conducted in the department, it was estimated that of those who complete treatment under GA, the worst-case follow-up response rate could be around 40% (Knapp et al., 2017). In line with current figures in the department, it was estimated that of all those potentially eligible for this study who came to be assessed in the department, only 45% would end up completing treatment under GA, as some may have treatment completed by other means or may not return for treatment. This suggested that 416 potential participants would need to be approached to allow for those who subsequently do not have a GA or who are lost to follow-up during the study. It was decided to continue recruitment until this number had been approached or the sample size of 75 was reached, whichever came sooner.

4.4.6 Study recruitment and process

A convenience sample of potential participants, who met the eligibility criteria, were approached at new patient clinics at Charles Clifford Dental Hospital (CCDH) paediatric department between January 2017 and January 2019. Potential participants were approached during the new patient assessment by a member of the direct care team in the first instance. The purpose of the study was explained verbally in simple terms by a member of the direct care team or by the PhD student (RK), and potential participants were then invited to find out more and consider if they wanted to take part in the study. RK was present at all the clinics and available to answer any questions potential participants had about the study. *Figure 2* below shows the path of the participants through the study.

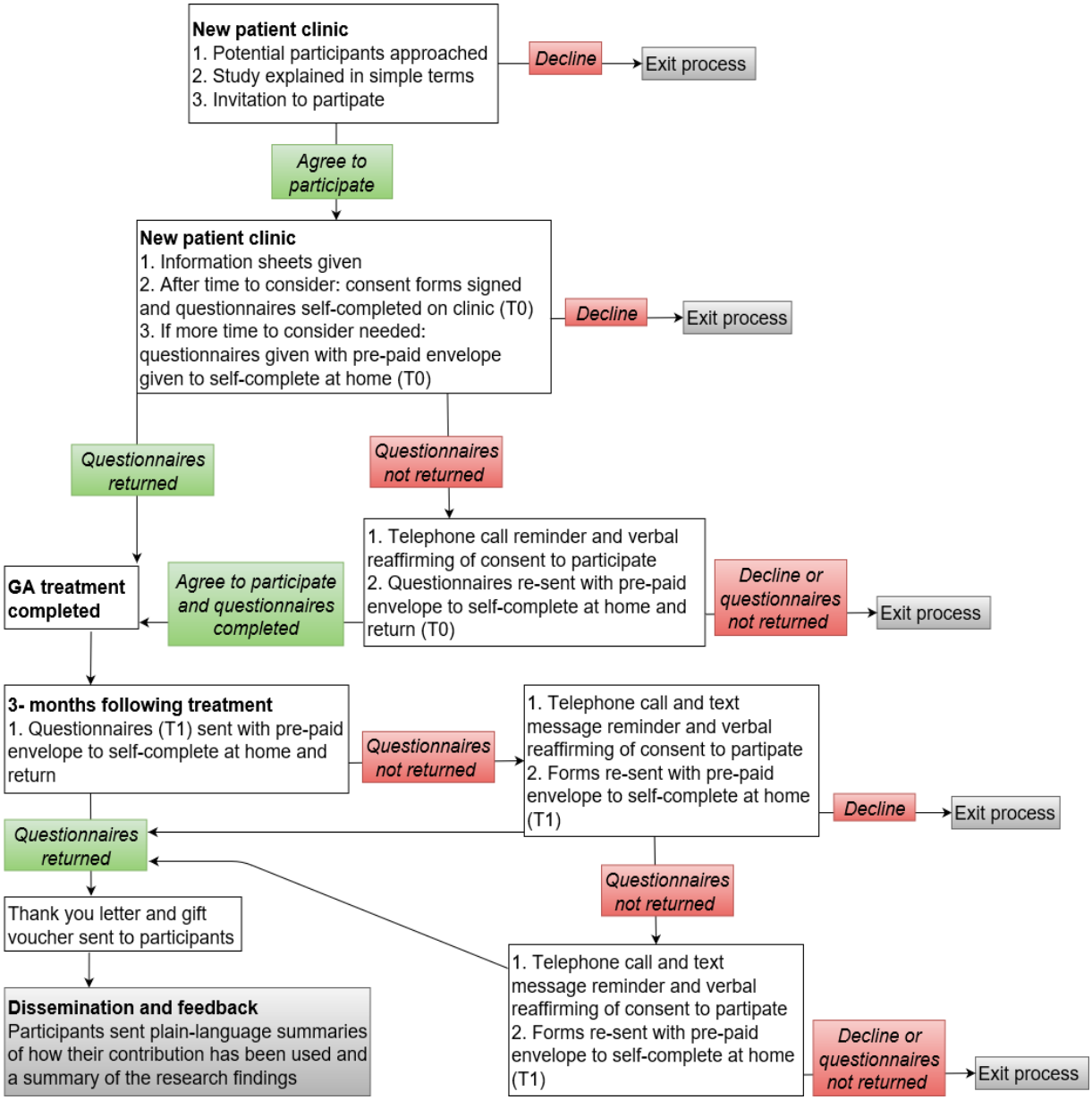

Figure 2: Flowchart to show the participant pathway through the study

If they were interested in taking part, the child and parent/ caregiver were given separate written information sheets, outlining the study in more detail, to read while waiting for radiographs to be taken. These age-appropriate sheets detailed the purpose of the study, what participating would entail and informed potential participants that if they completed both questionnaires in the study, they would receive a £10 gift voucher in recognition of their time and commitment. These sheets also explained that completion of the study meant

completing the questionnaires three months following their dental GA. Participants were informed they were free to withdraw at any point without having to justify this decision and without consequence. Questionnaires were therefore completed at the following time points:

1. Pretest (T0) at the new patient clinic appointment
2. Posttest (T1) at 3 months post-treatment (dental GA)

Where the child and parent/ caregiver felt they had had sufficient time to consider their participation, and consented to participate, they completed the first questionnaires (T0) at their new patient assessment visit. Previous experience in this setting had suggested that most participants were happy to provide consent at the initial appointment, without requiring additional time. However, if they felt they needed more time to consider their involvement, potential participants were given the questionnaires to complete at home, with a self-addressed envelope for their return.

After completion of treatment under GA, follow-up (T1) questionnaires were sent by post to participants, along with a covering letter and pre-paid return envelope. Return of the questionnaires was taken as confirmation of their willingness to continue to participate in the study. Following completion of the follow-up questionnaires, participants were sent a thank you letter and their gift voucher in acknowledgement of their time and commitment.

If participants failed to return the T1 questionnaires, they were telephoned or sent a text message to reaffirm their consent to participate in the study and invited again to complete the final questionnaires by post. If they were unable to be contacted by telephone,

then the questionnaires were sent again by post. If questionnaires were not returned on this occasion, the process was repeated once more. In cases where the questionnaires were not returned after both reminders then participants were considered ‘lost to follow-up' and were removed from the study.

4.4.7 Instruments and additional data collection

Data were collected via child and parental questionnaires and from the patient record as follows:

4.4.7.1 Main outcomes

Child questionnaire

Children were given a questionnaire which contained the consent form, instructions for completing the questionnaire and then the following instruments to measure OHRQoL and HRQoL respectively: The Caries Impacts and Experiences Questionnaire for Children (CARIES-QC) and the Child Health Utility 9D (CHU9D). Further information on these instruments is given below.

Children were asked to complete the questionnaire themselves, but younger children were given assistance if needed e.g. having the questions read to them, but with no additional guidance on the answers. A copy of the children’s questionnaire, which includes both CARIES-QC and CHU9D is given in *Appendix 3*.

i) The Caries Impacts and Experiences Questionnaire for Children (CARIES-QC)

CARIES-QC was used to gather data from children about the impact of their oral health on their everyday life. In contrast to previous studies, where proxy-reported outcome measures have been used, CARIES-QC was chosen to assess the impact on OHRQoL from the child's perspective. This questionnaire has been previously validated for use in this population (Gilchrist et al., 2018). This is also the only disease-specific measure available to assess the impact of caries, and as such may be more sensitive to treatment-related changes than generic measures of OHRQoL.

CARIES-QC contains 12-items with a 3-point response format, where children can rate whether they are affected 'not at all', 'a bit' or 'a lot'. There is also a single global question, which asks children to rate how much of a problem their teeth are to them, on the same three-point response format. In the follow-up questionnaire, to be completed three months following treatment under GA, a global transition judgement question (GTJ) was included. The aim of this single question was to ask child participants to rate the extent to which their oral health had improved or deteriorated since treatment, on a similar three-point Likert scale (i.e. 'better', 'the same' or 'worse'). The results from this question were to be correlated with change scores in an anchor-based assessment of clinical significance, i.e. to assess what change in score was considered to be clinically meaningful (Masood et al., 2014).

Completion of the CARIES-QC questionnaire results in possible raw scores of 0-24 obtained by simple addition of the individual item scores as follows: as follows: 'Not at all'=0, 'A bit'=1 and 'A lot'=2. The global score at baseline and follow-up is presented separately, as are the results from the global transition judgement question. To calculate

change following treatment, the raw scores are converted to an interval scale score using a conversion table (*Table 7*). This allows for the more accurate calculation of change at all points along the scale (Gilchrist, 2015).

Table 7: Conversion table for converting CARIES-QC ordinal raw scores to interval scale scores.

Raw score	Interval score
0	0
1	2.63
2	4.50
3	5.84
4	6.90
5	7.80
6	8.60
7	9.32
8	10.00
9	10.64
10	11.26
11	11.86
12	12.45

Raw score	Interval score
13	13.03
14	13.62
15	14.22
16	14.84
17	15.48
18	16.17
19	16.92
20	17.76
21	18.75
22	19.96
23	21.65
24	24.00

ii) Child Health Utility 9D (CHU9D)

The CHU9D is a well-established health-related quality of life (HRQoL) instrument, used in over 150 studies worldwide, including the UK. Children were involved throughout its development to ensure that it is child-centred (Stevens and Ratcliffe, 2012). The CHU9D was developed for use in 7- to 11-year-old children to identify health dimensions that are important to them, but has successfully been used in studies involving older and younger children, ranging from 5- to 17- years (Stevens and Ratcliffe, 2012; Canaway and Frew, 2013; Foster-Page et al., 2015). Unlike CARIES-QC, this is a generic measure of HRQoL. Although a previous study using CHU9D found it to be unresponsive

to changes in caries experience, the authors of this study suggested that further research was needed using CHU9D in populations with higher rates of dental caries (Foster-Page et al., 2015). As the child population in this study has higher overall caries experience and associated impacts than the general population, CHU9D was more likely to be able to detect changes following treatment.

The CHU9D consists of nine items, each with five ordinal response options (scored 1 to 5) that assess the child's functioning across domains of worry, sadness, pain, tiredness, annoyance, school, sleep, daily routine and activities. The overall score for CHU9D, therefore, ranges from 9 to 45, where increasing score implies greater impact on health-related quality of life.

Parental/ guardian questionnaire

The parental questionnaire consisted of a consent form, the Family Impact Scale questionnaire and additional questions about their child's dental history to determine:

- Whether their child had previously received a dental GA
- Whether any of the child's siblings had previously received a dental GA
- Whether the child had received antibiotics for their dental problem, and if so, how many courses

A copy of the parental questionnaire participants is given in *Appendix 4*.

i) Family Impact Scale (FIS)

The FIS was used to gather data from parents/caregivers on the impact of the child's oral health on the family. This is a 14- item questionnaire, designed to measure the family

impact of a condition/ intervention and comprises of four domains: parental family activity, parental emotions, family conflict and economic impact (Jokovic et al., 2002). This instrument has been used in a number of previous studies and is validated for use in this population (de Souza et al., 2016). Answers to the questions are scored using a 5-point Likert scale (response options: 'Never'=0; 'Once or twice'=1; 'Sometimes'=2; 'Often'=3; 'Every day or almost every day'=4). A 'Don't know' response option was also provided and scored as 0. Total scores for the FIS can, therefore, range from 0 to 56, where 56 indicates the greatest (worst) impact on the family. In the follow-up questionnaire, as with CARIES-QC, global transition judgement questions were also included, scored using a 5-point Likert scale to access whether parents perceived an overall change in the impact on the family, and their child, following treatment.

4.7.7.2 Other data

Patient characteristics

Data were also collected from patient records on variables which may impact OHRQoL, as identified in our adapted Wilson and Cleary model, and for descriptive purposes. These included:

i) Individual factors

- Age, recorded in years, taken from the patient record.
- Ethnicity recorded as either 'White British' or 'Black or Ethnic Minority Group (BME)', taken from the patient record.

ii) Environmental factors

- Safeguarding concerns as indicated by the child's placement on a historic or current care protection plan, whether they had a paediatric liaison letter or whether there was social care involvement (a named social worker, as detailed in the new patient proforma).
- Deprivation, which was assessed using a composite measure of area-based deprivation, the Index of Multiple Deprivation 2015 (IMD) score derived from their house postcode. The IMD is the official measure of relative deprivation for neighbourhoods in England and is based on seven different domains of deprivation:
    - Income Deprivation
    - Employment Deprivation
    - Education, Skills and Training Deprivation
    - Health Deprivation and Disability
    - Crime
    - Barriers to Housing and Services
    - Living Environment Deprivation

  The IMD scores rank every area from 1 (most deprived area) to 32,844 (least deprived area). It is common for IMD scores to be presented in quintiles; quintile 1 (Q1) being the most advantaged, and quintile 5 (Q5) being the most disadvantaged quintile, although there is no official cut-off at which an area is described as deprived. Both the overall IMD rank score and quintile were recorded, derived from the postcode in the patient's record.

iii) Clinical factors

- Caries experience recorded as total number of decayed, missing and filled teeth (dmft/DMFT) and a separate record of the total number of carious teeth, taken from the patient record. This usually included caries recorded following radiographic assessment, and therefore early carious lesions as well as cavitated lesions, unless the child was unable to tolerate radiographs being taken.
- Type of treatment recorded as exodontia only (EXO) or combination care (CC). For the CC group, it was recorded whether restorative treatment was carried out on clinic followed by exodontia under GA, or whether the child received comprehensive care (restorations and extractions) under GA.
- Number of teeth extracted and, where applicable, number of teeth restored, taken from the patient record.
- Waiting time, in weeks, before completion of treatment under GA

4.4.8 Outcome measures

Primary outcome

The primary outcome measure was child-reported OHRQoL, before and after treatment under GA. Change in OHRQoL following treatment was recorded as an overall change in CARIES-QC interval score. Change in the score for individual items of the questionnaire was also recorded.

Secondary outcomes

In addition, the following outcomes were measured:

- Change in overall child-reported quality of life recorded as an overall change in CHU9D score and change in score for the individual items of the questionnaire
- Change in the impact on the family recorded as an overall change in FIS score and change in score in the individual domains of the questionnaire

4.4.9 Statistical analysis

Data were entered into an electronic database (IBM SPSS Statistics, version 24) and descriptive statistics were calculated and reported. Statistical analysis results were considered significant at $p<0.05$.

To check for accuracy and consistency of data entry, intra-rater reliability was assessed by randomly re-testing 10% of the sample (9 patients) and by calculating the Intraclass Correlation Coefficient (ICC). ICC values less than 0.5 are indicative of poor reliability, values between 0.5 and 0.75 indicate moderate reliability, values between 0.75 and 0.9 indicate good reliability, and values greater than 0.90 indicate excellent reliability (Koo and Li, 2016).

Descriptive statistics for demographic and clinical factors (including treatment carried out) were calculated, to include means, standard deviation and ranges. For categorical variables, numbers and percentages were calculated. Differences in baseline characteristics between those who completed the study and those who were lost to follow-up were analysed using Mann-Whitney U tests for continuous data (as data were not normally distributed) and chi-square tests for categorical data to assess if there is any difference between those who completed the study and those who did not.

Differences in baseline characteristics and treatment carried out between treatment groups to see if there were statistically significant differences between the groups. Independent t-tests (or non-parametric equivalent) were used to test for differences between continuous variables and the Pearson's chi-squared test was used for differences in categorical variables.

Total CARIES-QC, CHU9D and FIS scores were calculated. Percentages of those reporting the highest and lowest scores in each scale were calculated, to establish whether floor and ceiling effects were in the acceptable range, i.e. less than 15% of cases (Terwee et al., 2007).

CARIES-QC raw scores were converted to their equivalent interval scores, as recommended for calculation of change scores (Gilchrist, 2015). Total scores for each of the measures at baseline and follow-up were analysed to see if the change in score was statistically significant, using the Wilcoxon signed rank test as the scores were not normally distributed. Change scores for each measure were calculated by subtracting follow-up scores from baseline scores, so a positive change indicated improvement in QoL, and a negative one represented deterioration.

The effect size was calculated for each of the measures to assess the magnitude of change. Cohen's-d effect sizes were calculated by dividing the mean change scores by the standard deviation of the pre-treatment scores, to give a dimensionless measure of effect. By convention, an effect size of 0.2 indicates a small magnitude of change, 0.2-0.7 a moderate change and greater than 0.7 a large change (Sawilowsky, 2009).

Internal consistency of each of the measures was assessed using the Cronbach's alpha test. By convention, the alpha cut-off value of 0.70 or higher was considered as being acceptable, with larger alpha values indicating greater internal consistency (Kline, 1999).

The longitudinal construct validity and responsiveness of CARIES-QC and FIS were assessed by comparing mean change scores for each measure with the global transition question responses. Good longitudinal construct validity was indicated if those reporting deterioration had negative mean change scores, those reporting stability had change scores close to zero, and those reporting improvements had positive change scores of increasing magnitudes. Responsiveness of CARIES-QC and FIS were assessed using Mann-Whitney U tests to examine the differences between baseline and follow-up scores for each change in response to the global transition question: for participants where their global rating 'stayed the same' and where participant moved at least one category; i.e. 'got better' or 'got worse'. If a measure is responsive, the first change score should be non-significant and the other two should be significant (Lee et al., 2011; Wright et al., 2012).

The minimal important difference (MID), i.e. the smallest difference in the score which is considered clinically meaningful and which patients perceive as beneficial (Masood et al., 2014), of the CARIES-QC and FIS scores was calculated using the mean change scores of participants who reported 'improvement' on the global rating.

Construct validity was evaluated by correlation between total CARIES-QC, CHU9D and FIS scores and ordinal categories of caries experience and clinical presentation at baseline. Where ordinal categories did not exist, i.e. number of carious teeth, then ordinal categories were created using the mean value as a cut-off point for each category. These

analyses were also used to identify which variables were potentially impacting on the quality of life outcomes, for inclusion in the path analyses.

Finally, path analysis was used to analyse the direct and indirect relationships between clinical factors, individual factors and environmental factors and child-reported quality of life outcomes in the proposed Wilson and Cleary model (*Figure 4, Section 4.3*). The standardised coefficients of each path were compared to identify which factors had greater effects within the model. Full details of the methodology for the path analysis are given below (*Section 4.4.11*).

4.4.10 Missing data

An acknowledged problem with self-complete questionnaires is the potential for missing data. In order to reduce the likelihood of item non-response, the measures in this study were selected, in part, because they have been shown to have high levels of complete response (Gilchrist et al., 2014; Gilchrist, 2015). Where greater than one-fifth of questions were unanswered in a questionnaire then the participant was eliminated from further analysis (Shrive et al., 2006). Where fewer than one-fifth of questions were unanswered, the missing values were replaced using individual mean imputation, where the imputed value is the calculated mean of a participant's responses to the other questions (Shrive et al., 2006).

4.4.11 Data handling and governance

Data handling

Participants were anonymised, and only participant identification numbers were used on the questionnaires. No personally identifiable information was stored on the computer data sets. Hard copies of questionnaires were stored in a locked filing cabinet, in a secure room in the department, and were only accessible by the research team. Data were transferred onto a password-protected desktop computer in a secure room within the department. All data files on the computer were password protected. These files were only accessible to members of the research team. The data generated by this study will be kept for five years then destroyed.

Confidentiality

To ensure participant confidentiality, all participants were anonymised through assignment of a participant identifier number. No participant identifiable information was recorded on the questionnaires.

Protection from harm

All researchers in contact with participants had the appropriate level of disclosure obtained from the Criminal Records Bureau and had received safeguarding training.

Ensuring rigour

Steps to ensure quality of the research are in place at all stages of the research process as follows:

- **Project protocol**
    - The project protocol was developed collaboratively, drawing on the range of experience of the supervisors and other advisors e.g. statisticians.
    - During development, the protocol was subject to the independent scientific review process, which assessed the quality of the proposal and was amended accordingly.
- **Monitoring and oversight during the study**
    - The PhD student (RK) was supported by a highly experienced supervisory team, who monitored progress during the study. This took the form of monthly supervisory meetings and ad-hoc advice as required.
- **Sharing early findings**
    - Early findings were presented at research group meetings, national and international conferences, which provided opportunity for comments and suggestions on how the project should develop.
- **Formal publication**
    - Quality assurance of results and findings through publication in a relevant peer review journal.
    - Dissemination of key findings via the website will open the research to comment and review more widely.

4.4.12 Path analysis

One of the novel aspects of this research was the decision to explore the effects of different factors on child OHRQoL and QoL, before and after treatment under GA for dental caries, based on the theoretical model.

Path analysis was used to investigate the relationships between the variables in the proposed theoretical model, based on the Wilson and Cleary model of HRQoL (See *Chapter 2*). Path analysis, a subset of structural equation modelling, is an extension of regression analysis to enable a deeper understanding of the direct and indirect relationships between the variables being examined (Stage et al., 2004). All aspects of the path analysis were carried out using appropriate statistical software (StataCorp STATA, version 15). The process of path analysis was conducted as follows, as recommended by Stage et al. (2004):

1. A full theoretical model to be tested was identified
2. Refining of this full model, including fit indices for all examined models
3. Report of fit indices for the final model
4. Illustration of the final model
5. Discussion of the findings in relation to theory

Each of these aspects of the path analysis is described in more detail below. The results from items 1-4 are presented in *Chapter 5* (Results), with item 5 explored in *Chapter 6* (Discussion).

1. Theoretical model

Path analysis begins with a full theoretical model, derived from the literature, which is tested to see how well it fits the experimental data. In order to carry out the path analysis, an 'input' model is generated based on the proposed full theoretical model. This 'input' model contains the same variables as the full theoretical model, but they are positioned chronologically, i.e. those variables towards the right of the model are considered to have

occurred following those on the left. The model shows hypothesised paths between variables, indicated by arrows which point in the expected direction of influence.

As discussed earlier in this thesis, the theoretical model for this research was based on the Wilson and Cleary model (See *Chapters 2*). Analysis of the paths in the full model was carried out to provide information about the magnitude and significance of the hypothesised relationships between variables, as well as information about the overall fit of the data to the theorised model.

2. Refining the model

The process of developing and refining the model was based on a parsimonious attempt to build a more concise, yet still coherent, model, a method that has been employed in similar studies (e.g. Vettore et al. 2019). The modification of the model involved the removal of paths which were deemed non-significant. The initial significance level of p=0.1 was chosen based on recommendations by Olobatuyi (2006). Fit analyses for the parsimonious model were carried out to examine whether it was still a good fit to the data. The parsimonious model was then tested against the full theoretical model using a Chi-square (χ^2) difference test to examine whether there was a statistically significant difference between the two. The goal was to find the most parsimonious model with good fit to the data and which did not differ significantly from the full theoretical model. If the fit to the data or comparison to the full model does not meet these requirements, the parsimonious model can be refined and re-tested until these criteria are met. One method of refinement is to adjust the significance level at which paths are removed, e.g. to 0.2 rather than 0.1. However, in this case both criteria were met so no further model adaptation was necessary.

3. Fit indices

Model fit was evaluated using a range of indices as recommended by Kline (2005):

- Model Chi-square (χ^2) to assess the overall fit of the data to the model. This tests the null hypothesis that there is no difference between the model and the data. Where $p>0.05$, it indicates the null hypothesis is not rejected (i.e. the data fit the model well).
- Root Mean Square Error of Approximation (RMSEA), where values closer to zero represent a better fit of the data to the model. RMSEA values less than 0.06 indicate a good fit.
- Comparative Fit Index (CFI) and Tucker-Lewis Index (TLI) were assessed as they are both preferred fit indices for smaller sample sizes as they are less sensitive to change in sample size. Values greater than 0.9 indicate a good fit of the data.

As the data for the variables in the model were not all normally distributed, Satorra-Bentler corrections were applied to all analyses as recommended in the literature (Satorra et al., 1999).

4. Illustration of the final model

The final parsimonious model is reported, along with descriptions of the significant direct and indirect relationships identified.

5. Discussion of the findings

Direct and indirect relationships are discussed in Chapter 6, with reference to the literature.

Chapter 5: Results

5.1 Introduction

The results in this chapter are presented in three main sections, as follows:

Section 5.2 gives the results of the descriptive analysis. This includes an overview of the sample population, including individual, environmental and clinical characteristics. This section also includes information on those lost to follow-up and results of the analyses to test whether this group differed from those who completed the study.

Section 5.3 presents the changes in child-reported oral health-related quality of life (OHRQoL), overall quality of life (QoL) and family impact, following treatment under general anaesthetic (GA). The results of individual items and domains (where applicable) are also given. Clinical significance, or meaningful magnitude of change, of each measure is reported as Cohen's d effect size statistics. Internal consistency, as Cronbach's alpha, is reported. For CARIES-QC and FIS, which both included a global question, the cross-sectional construct validity is evaluated by examining the association of the global rating and the mean scale scores. The minimal important difference for the measures is given, based on these responses to the global questions. Finally, construct validity of the measures was evaluated by correlation of total scores for each measure with categories of caries experience and clinical presentation.

Section 5.4 presents the results from the path analysis and describes the direct and indirect relationships identified between variables in the proposed theoretical model, including the effect of treatment approach.

5.2 Descriptive analysis

5.2.1 Recruitment and loss to follow-up

Recruitment to this study ran from January 2017 to January 2019. Of the 273 potential participants approached to participate in the study, 106 declined to participate. In total, 167 child and parent dyads were recruited and completed the questionnaires at baseline (T0), giving a 61.2% response rate.

Of those recruited, subsequently, 35 were withdrawn from the study as they failed to attend further appointments, either for initial treatment or for the GA appointment itself. A further 47 were withdrawn because they did not respond to requests to complete the final questionnaire. Overall, 82 participants were lost to follow-up. In total, therefore, 85 parent-child dyads completed the follow-up questionnaires (T1) and were included in the final analyses (completion rate = 49.1%). Of those completing the study, just over half (n=46) required a telephone call or text message reminder to complete the final questionnaire.

5.2.2 Intra-examiner reliability

Based on the nine records selected, there was absolute agreement (100%) on repeat data entry of the measures. This represented an Intraclass Correlation Coefficient (ICC) of 1.00 (95% CI, 1.00-1.0), or excellent reliability (Koo and Li, 2016).

5.2.3 Sample characteristics

Demographic details

Of those who completed the study, there were 38 (44.7%) males and 47 (55.3%) females. Their ages ranged from 5 to 11 years, with a mean ± standard deviation (SD) age of 6.5 (± 1.5) years. Most children were reported to be white British (n=62, 72.9%). Index of Multiple Deprivation (IMD) scores, based on demographic information from the participant's postcode, ranged from 4.33-75.23 (mean=36.49 ± 18.92), with 72.9% (n=62) of participants living in the most deprived areas of England. The numbers of participants in each IMD quintile are shown in *Table 8*. Over a quarter (27.1%) of children had a sibling who had previously received treatment under GA, while 2.4% (n=2) of children had previously had dental treatment under GA themselves. In addition, 8 (9.4%) children had safeguarding concerns in place, in the form of either a Paediatric Liaison Letter (letter to the safeguarding team to highlight where a clinician may have concerns about a child, to be shared with a network of services including health visitors and schools as required), Care Protection Plan or social worker involvement.

The results of the Mann-Whitney U and Pearson's chi-square tests, conducted to see if there were any differences with respect to these sample characteristics between those

lost to follow-up and those who completed the study, are given in *Table 8*. There was no statistically significant difference between the two groups for any of the demographic variables.

Table 8: Comparison of demographic characteristics at baseline, of those followed up and those lost to follow-up. Numbers, with percentages in brackets, are given unless otherwise stated.

Variable		**All (n=167)**	**Followed up (n=85)**	**Lost to follow-up (n=82)**	**p-value**
Age, years	Range	5-14	5-11	5-14	0.14
	Mean (±SD)	6.70 (±1.69)	6.49 (±1.53)	6.91 (±1.83)	
Gender	Male	79 (47.3%)	38 (44.7%)	41 (50.0%)	0.49
	Female	88 (52.7%)	47 (55.3%)	41 (50.0%)	
Ethnicity	White British	121 (72.5%)	62 (72.9%)	59 (72.0%)	0.89
	BME	46 (27.5%)	23 (27.1%)	23 (28.0%)	
Deprivation (based on IMD score)	Least deprived	10 (6.0%)	6 (7.1%)	4 (4.9%)	0.46
	Less deprived	19 (11.4%)	10 (11.8%)	9 (11.0%)	
	Average	16 (9.6%)	7 (8.2%)	9 (11.0%)	
	More deprived	36 (21.5%)	14 (16.5%)	22 (26.8%)	
	Most deprived	86 (51.5%)	48 (56.5%)	38 (46.3%)	
Safeguarding concern	No	138 (82.6%)	77 (90.6%)	61 (74.4%)	0.19
	Yes	20 (12.0%)	8 (9.4%)	12 (14.6%)	
	Data missing	9 (5.4%)	0	9 (11.0%)	

Notes: SD= standard deviation; p-values are for comparisons between the followed-up and lost to follow-up groups. As the data were not normally distributed, the Mann-Whitney U test was used to test for significant difference between the groups. Pearson's chi-squared test was used to test for difference in categorical variables. There were no statistically significant results. BME= Black or minority ethnic group.

Clinical variables

The mean (±SD) total number of decayed, missing and filled primary and permanent teeth (dmft/DMFT) of child participants was 6.9 (±3.0), with total numbers ranging from 1-16. The number of carious teeth ranged from 1-15 (mean=6.6, SD=2.9). Anterior caries was present in 11.8% (n=10) of the participants. Swelling was present in 43.5% (n=37) of cases and pain was reported at initial assessment by 70.6% (n=60) of participants. Antibiotics had been received by 43.5% (n=37) of children prior to their assessment visit. Full details of these characteristics are given in *Table 9*.

Dental treatment provided

In total, 62 (72.9%) children received exodontia only (EXO) treatment under GA, while 23 (27.1%) received combination care (CC) involving restorations and extractions. The CC group is comprised of those who received comprehensive care under GA, and those who had restorations on clinic prior to a GA for extractions. Information on the baseline demographic characteristics, caries experience and clinical presentation of children in each treatment group are shown in *Table 9*. There was no statistically significant difference in numbers of carious teeth between the groups (p=0.89), or in any of the other baseline demographic and clinical factors analysed.

Table 9: Overall and comparison of caries experience and clinical presentation of children in each treatment group. Numbers (%) are given, unless otherwise stated.

Variable		**Overall (n=85)**	**EXO group (n=62)**	**CC group (n=23)**	**p-value**
Age, years	Range	5-11	5-11	5-9	0.10
	Mean (±SD)	6.49 (±1.53)	6.66 (±1.62)	6.04 (±1.15)	
Gender	Male	38 (44.7%)	27 (43.5%)	11 (47.8%)	0.73
	Female	47 (55.3%)	35 (56.5%)	12 (52.2%)	
Ethnicity	White British	62 (72.9%)	47 (75.8%)	15 (65.2%)	0.32
	BME	23 (27.1%)	15 (24.2%)	8 (34.8%)	
Deprivation (based on IMD score)	Least deprived	6 (7.1%)	6 (9.7%)	0	0.19
	Less deprived	10 (11.8%)	5 (8.1%)	5 (21.7%)	
	Average	7 (8.2%)	4 (6.5%)	3 (13.0%)	
	More deprived	14 (16.5%)	11 (17.7%)	3 (13.0%)	
	Most deprived	48 (56.5%)	36 (58.1%)	12 (52.2%)	
DMFT/dmft	Mean (±SD)	6.9 (±3.0)	6.9 (±3.2)	6.8 (±2.5)	0.83
	Range	1-16	1-16	2-12	
Number of carious teeth	Mean (±SD)	6.6 (±2.9)	6.7 (±3.0)	6.6 (±2.4)	0.89
	Range	1-15	1-15	2-12	
Anterior caries present	Yes	10 (11.8%)	9 (14.5%)	1 (4.3%)	0.20
	No	75 (88.2%)	53 (85.5%)	22 (95.7%)	
Pain reported at assessment	Yes	60 (70.6%)	46 (74.2%)	14 (60.9%)	0.25
	No	25 (29.4%)	16 (25.8%)	9 (39.1%)	
Swelling present	Yes	37 (43.5%)	27 (43.5%)	10 (43.5%)	0.99
	No	48 (56.5%)	35 (56.5%)	13 (56.5%)	
Received antibiotics	Yes	37 (43.5%)	27 (43.5%)	10 (43.5%)	0.99
	No	48 (56.5%)	35 (56.5%)	13 (56.5%)	
Notes: SD= standard deviation; p-values are for comparisons between the exodontia only (EXO) and combination care (CC) group. Independent t-tests were used to test for differences between continuous variables and the Pearson's chi-squared test was used for categorical variables. There were no statistically significant results.					

The mean waiting time for completion of treatment under GA was 9.4 (±8.0) weeks for the EXO group, compared to 16.7 (±9.6) weeks for the CC group. This difference was tested for significance using the Mann-Whitney U test and was found to be statistically significant (p=0.001).

The treatment received in each group is summarized in *Table 10.* The mean number of extractions in the EXO group was 6.7 (± 3.0) and in the CC group, it was 5.1 (± 2.4). This difference was statistically significant (p<0.05).

Table 10: Summary of dental treatment provided.

Variable		**Overall (n=85)**	**EXO group (n=62)**	**CC group (n=23)**	**p-value**
Number of extractions	Mean (±SD)	6.3 (±2.9)	6.7 (±3.0)	5.1 (±2.4)	0.03*
	Range	1-15	1-15	2-12	
Number of restorations	Mean (±SD)	2.3 (±1.8)	0	2.3 (±1.8)	<0.001*
	Range	0-9	0	1-9	
Notes: SD= standard deviation; p-values are for comparisons between the exodontia only (EXO) and combination care (CC) groups. Independent samples t-tests were used to assess for statistical significance. * indicates a significant difference between the groups at p<0.05.					

5.3 Quality of life results

5.3.1 Missing data

A total of three (3.5%) participants had missing values for CARIES-QC at either baseline (T0) or follow-up (T1). All three had just one missing value, and so these values were replaced using individual mean imputation, where the imputed value is the calculated mean of a participant's responses to the other questions (Shrive et al., 2006). No participants had more than one missing value, and so all participants were included in the final analyses. All participants completed the global question at baseline and follow-up, and transition judgement question at follow-up, therefore all participants were included in analyses involving these scores.

The same approach was used for both the CHU9D and the FIS questionnaires. A total of four (4.7%) participants had missing values for CHU9D and two (2.4%) for the FIS. For the CHU9D questionnaire, all four participants had just one missing value, and so these missing values were replaced using individual mean imputation. In the FIS, both participants had missed two questions, so these values were again replaced by the individual's mean value. No participants had more than two missing values in CHU9D or FIS, and so none were excluded from further analyses. All participants completed the global transition FIS questions and therefore all participants were included in analyses involving the global score.

There were a few instances of participants supplying two responses to a question in the questionnaires. However, in all instances, it was clear which answer was in error and

which was the correct value and so this is the one that was included in the analysis. Therefore, as no participants had to be removed due to missing values, a total of 85 participants were included in subsequent analyses.

5.3.2 CARIES-QC

5.3.2.1 Overall raw and interval scores

At baseline, the mean (± SD) raw score was 7.69 (± 5.24) with a range of 0-22. Just two (2.4%) participants scored the lowest possible score of 0, with no individuals scoring the highest possible score of 24. These figures are within the acceptable range for floor and ceiling effects, i.e. less than 15% of participants (Terwee et al., 2007). At follow-up, the mean (± SD) raw score was 3.22 (± 3.28), with a range of 0-18.

The raw CARIES-QC scores were then converted to their equivalent interval scores as recommended in the guidance, to allow more accurate calculation of change at all points along the scale (Gilchrist, 2015). The mean CARIES-QC interval score (CIS) at baseline was 8.99 (± 4.29), with a range from 0-19.96. The mean CIS at follow-up was 4.55 (± 3.75) and scores ranged from 0-16.17. Overall, there was a mean change in CIS score of 4.47 (±5.58), indicating improved OHRQoL. This result was statistically significant ($p<0.001$). These change score findings are summarised alongside those of the other outcome measures in *Table 17*, (*Section 5.3.5*, Effect sizes).

5.3.2.2 Individual item scores

The impact most commonly reported by children at baseline was 'food getting stuck in their teeth' (n=73, 85.9%) with 'finding it hard to do schoolwork' being the least reported (n=17, 20.0%). At follow-up, the most commonly reported impact was still 'food getting stuck in their teeth', reported by 58.8% of children. *Table 11* shows the number and proportion of children reporting each impact in the CARIES-QC questionnaire at baseline and follow-up. There was a reduction in the numbers of children reporting all the impacts at T1, but the percentage reduction from baseline shows that treatment had greater effect on some impacts than others. Percentage reduction in individual impacts ranged from 31.5% (food getting stuck) to 91.0% (being kept awake).

Table 11: Number and proportion of children responding positively ('a bit' or 'a lot') to each item at baseline (T0) and follow-up (T1).

Item	Number (%) at T0	Number (%) at T1	% reduction at T1
Food stuck	73 (85.9%)	50 (58.8%)	31.5%
Feel cross	61 (71.8%)	13 (15.3%)	78.7%
Cried	61 (71.8%)	27 (31.8%)	55.7%
Eat more carefully	52 (61.2%)	22 (25.9%)	60.0%
Eating on one side	50 (58.8%)	32 (37.6%)	36.0%
Teeth hurt	48 (56.5%)	15 (17.6%)	68.8%
Hard to eat some foods	47 (55.3%)	27 (31.8%)	42.6%
Annoyed	45 (52.9%)	19 (22.4%)	57.8%
Eat more slowly	35 (41.2%)	17 (20.0%)	51.4%
Kept awake	33 (38.8%)	3 (3.5%)	91.0%
Hurt when brushing	33 (38.8%)	14 (16.5%)	57.6%
Hard to do schoolwork	17 (20.0%)	3 (3.5%)	82.4%

5.3.2.4 Global question responses

At baseline, n=46 (54.2%) children reported that their teeth were 'a bit' or 'a lot' of a problem for them. This compares to n=20 (23.6%) reporting problems at follow-up. The CARIES-QC interval scores at baseline and follow-up for these groups are shown in *Table 12*. The overall scores increased as the global response worsened. A Kruskal-Wallis H test showed that there was a statistically significant difference in CARIES-QC score between the different global question responses at baseline ($\chi 2(2) = 26.50$, $p<0.001$) and at follow-up ($\chi 2(2) = 7.61$, $p=0.02$).

Table 12: Numbers and proportions reporting each global response, with mean CARIES-QC scores for each category

Global question response	**Baseline**		**Follow-up**	
	Number (%)	**CARIES-QC interval score**	**Number (%)**	**CARIES-QC interval score**
'not at all'	39 (45.9%)	6.00 ± 3.48 (0-13.03)	65 (76.5%)	3.57 ± 3.11 (0-10.64)
'a bit'	36 (42.4%)	10.70 ± 2.80 (6.90-16.92)	18 (21.2%)	7.23 ± 3.62 (2.63-11.86)
'a lot'	10 (11.8%)	14.46 ± 2.61 (10.00 – 19.96)	2 (2.4%)	11.98 ± 5.91 (7.80 – 16.17)
p-value		<0.001*		0.02*
Notes: CARIES-QC interval scores show mean ± SD (range). The Kruskal-Wallis H test was used to check for clinical significance between CARIES-QC score between the groups. * indicates a significant difference in score between the groups at p<0.05.				

5.3.2.4 Global transitional judgement responses

The responses to the global transition judgement question at follow-up is summarized in *Table 13*. Nearly all the children (n= 91.8%) rated their teeth improved at

follow-up, with the rest claiming they were the same. No children rated their teeth as worse overall since they last completed the measure.

Table 13: Number and proportion (%) of children giving each response to the CARIES-QC global transition question

Response to global question: 'Since you last filled in these questions, are your teeth…'	**Number (%)**
Better	78 (91.8%)
The same	7 (8.2%)
Worse	0

5.3.3 CHU9D

5.3.3.1 Overall scores

At baseline, the mean (± SD) overall score was 13.58 (± 4.96) with a range of 9-31. None of the participants scored the lowest possible score of five and none scored the highest possible score of 45, making these figures within the acceptable range for floor and ceiling effects. At follow-up, the mean (± SD) overall score was 11.09 (± 3.07) with a range of 9-24. Overall, there was a mean reduction in CHU9D score of 2.48 (±5.29), indicating improved QoL. This change in score between baseline and follow-up was statistically significant ($p<0.001$). These change score findings are summarised alongside those of the other outcome measures in *Table 17*, (*Section 5.3.5*, Effect sizes).

5.3.3.2 Individual item scores

The results from the individual dimensions are given in *Table 14* below. The most common impacts reported by children at baseline were related to being tired (n=51, 60.0%) and worried (n=49, 57.6%). The least reported impact was daily activities being affected (n=11, 12.9%). All impacts reduced 3-months following treatment under GA, but the percentage reduction from baseline shows that treatment had greater effect on some impacts than others. Percentage reduction in individual impacts ranged from 5.9% (difficulty with schoolwork) to 73.5% (feeling worried).

Table 14: Number and proportion of children responding positively to each item at baseline (T0) and follow-up (T1).

Dimension	Number affected (%) at T0	Number affected (%) at T1	% reduction at T1
Tired	51 (60.0%)	44 (51.8%)	13.7%
Worried	49 (57.6%)	13 (15.3%)	73.5%
Pain	23 (27.1%)	7 (8.3%)	69.6%
Sad	22 (25.9%)	6 (7.0%)	72.7%
Sleep	19 (22.4%)	13 (15.3%)	31.6%
Schoolwork	17 (20.0%)	16 (18.8%)	5.9%
Daily routine	16 (18.8%)	9 (10.6%)	43.8%
Annoyed	13 (15.3%)	9 (10.6%)	30.8%
Activities	11 (12.9%)	10 (11.8%)	9.1%

5.3.4 Family Impact Scale (FIS)

5.3.4.1 Overall scores

At baseline, the mean (± SD) overall score was 9.21 (± 7.31) with a range of 0-35. Six (7.1%) participants scored the lowest possible score of zero, with no participants

scoring the highest possible score of 56. These figures are within the acceptable range for floor and ceiling effects.

At follow-up, the mean (± SD) overall score was 7.02 (± 6.40) with a range of 0-28. Overall, there was a mean change in FIS score of 2.19 (±7.83), indicating improved family impacts at follow-up. This change in score between baseline and follow-up was statistically significant (p=0.03). These change score findings are summarised alongside those of the other outcome measures in Table 17, (*Section 5.3.5*, Effect sizes).

5.3.4.2 Individual domain scores

The results from each of the individual domains are given in *Table 15* below. Only the 'parental and family activities' domain saw a statistically significant change in score between baseline and follow-up.

Table 15: Mean scores for FIS individual domains at baseline and follow-up.

Domain (total possible score)	**Score at baseline**	**Score at follow-up**	**p-value**
Parental and family activities (20)	5.48 ± 3.63 (0-15)	4.11 ± 3.28 (0-12)	0.003*
Parental emotions (16)	1.12 ±1.92 (0-10)	0.86 ± 1.50 (0-8)	0.44
Family conflict (16)	2.60 ± 3.09 (0-13)	1.88 ± 2.33 (0-9)	0.12
Family finances (4)	0.02 ± 0.22 (0-2)	0.07 ± 0.30 (0-2)	0.41
Notes: FIS domain scores show mean ± SD (range). p-values are for Wilcoxon test for difference between baseline and follow-up scores. * Statistically significant result between scores at baseline and follow-up (p<0.05).			

5.3.4.3 Global transitional judgement responses

The responses to the global transition judgement questions at follow-up are summarized in *Table 16*. Nearly all the parents (n=81, 95.3%) rated their child's oral health as improved at follow-up. Approximately three-quarters of parents felt the overall quality of life of their children had improved (n=63), and none felt it had worsened. Just over half the parents (n=46) reported that the impact on the family had improved, with the remaining parents reporting it had stayed the same.

Table 16: Number and proportion (%) of parents giving each response to the FIS global questions

Global transition judgement question	**Response**		
	Improved	**The same**	**Worsened**
Has your child's dental health improved, stayed the same or worsened?	81 (95.3%)	3 (3.5%)	1 (1.2%)
Has your child's overall quality of life improved, stayed the same or worsened?	63 (74.1%)	22 (25.9%)	0
Has the change to your family's life improved, stayed the same or worsened?	46 (54.1%)	39 (45.9%)	0

5.3.5 Effect sizes (magnitude of change)

Information on baseline, follow-up and change scores for each of the measures are presented alongside the effect size statistics in *Table 17* below. The effect size for CARIES-QC demonstrated a large meaningful change, associated with a statistically significant reduction in overall scores at follow-up ($p<0.001$). Effect sizes for CHU9D and FIS showed

moderate meaningful changes, associated with statistically significant reductions in scores ($p<0.001$ and $p=0.03$ respectively).

Table 17: Mean overall scores at baseline and follow-up, with effect sizes.

Measure	Baseline	Follow-up	Change	p-value	Cohen's d effect size	Effect size description
CARIES-QC interval	8.99 ± 4.29 (0-19.96)	4.47 ± 5.58 (0-16.17)	4.43 ± 4.92 (-8.63-16.92)	<0.001*	0.91	Large
CHU9D	13.58 ± 4.96 (9-31)	11.09 ± 3.07 (9-24)	2.48 ± 5.29 (-14-20)	<0.001*	0.60	Moderate
FIS	9.21 ± 7.31 (0-35)	7.02 ± 6.40 (0-28)	2.19 ± 7.84 (-17-24)	0.03*	0.32	Moderate
Notes: Scores show mean ± SD (range). p-values are for Wilcoxon test for difference between baseline and follow-up scores. * Statistically significant result (p<0.05).						

5.3.6 Internal consistency (Cronbach's alpha)

Cronbach's alpha was calculated using baseline questionnaire data for each of the measures. Cronbach's alpha for CARIES-QC and CHU9D was 0.9 and 0.8 respectively, indicating good internal consistency. Results for FIS revealed good overall consistency (alpha=0.8), but sub-domain analysis revealed individual domains had poor to acceptable internal consistency (*Table 18*).

Table 18: Internal constancy data for CARIES-QC, CHU9D, FIS and sub-domains.

Measure		**Cronbach's alpha**	**Internal consistency**
CARIES-QC		0.9	Excellent
CHU9D		0.8	Good
FIS		0.8	Good
FIS domains	Parental/ family activities	0.7	Acceptable
	Parental emotions	0.5	Poor
	Family conflict	0.7	Acceptable

5.3.7 Longitudinal construct validity and responsiveness of CARIES-QC and FIS

5.3.7.1 Longitudinal construct validity

The mean scores for CARIES-QC among those whose global response 'improved', and 'stayed the same' demonstrated good longitudinal validity, having positive mean change scores and mean change scores closer to zero mean respectively (*Table 19*). The mean change scores for FIS did not demonstrate longitudinal validity for any of the global responses, as the mean change score for those who 'improved' was not significant, and closer to zero than those whose global response 'stayed the same'.

However, for both CARIES-QC and FIS, there was considerable variation in the overall scores for each global response, where some individual scores were higher and lower than at baseline for each response (*Table 19*). The relevance of these findings will be explored in *Chapter 6* (Discussion).

5.3.7.2 Responsiveness

CARIES-QC demonstrated good responsiveness overall. That is, for participants whose global ratings improved, on average they had statistically significant lower CARIES-QC scores than at baseline i.e. a positive change score ($p<0.001$). Equally, for those whose global response stayed the same, demonstrated by no statistically significant change in CARIES-QC score at follow-up ($p=0.49$) indicating acceptable responsiveness.

In contrast, the results for the FIS suggested that the measure had poor responsiveness overall. The mean scores for the FIS among those who rated the impact on the family as 'improved' did not change significantly ($p=0.46$). For those who responded that the impact on the family 'stayed the same' there was a greater mean change FIS mean score of 2.82, and this result was statistically significant ($p=0.009$). These findings are summarized in *Table 19*.

Table 19: Mean change in quality of life scores by response to the global transition question

Response to GTJ	**CARIES-QC Interval**			**FIS**		
	Number (%)	**Change score**	**p-value**	**Number (%)**	**Change score**	**p-value**
Improved	78 (91.8%)	4.68 ± 4.81 (-7.8-16.92)	<0.001*	46 (54.1%)	1.65 ± 9.23 (-17-24)	0.46
Stayed the same	7 (8.2%)	1.69 ± 5.61 (-8.63-6.69)	0.49	39 (45.9%)	2.82 ± 5.83 (-7-20)	0.009
Got worse	0	n/a	n/a	0	n/a	n/a
Notes: CARIES-QC interval scores show mean ± SD (range). P-values are for Wilcoxon test for difference between baseline and follow-up scores for each group. * Statistically significant result between scores at baseline and follow-up (p<0.05)						

5.3.8 Minimal important difference for CARIES-QC and FIS

The minimal important difference (calculated as the mean change score for those who reported an improvement) was 4.68 for CARIES-QC interval score and 1.65 for FIS. Overall, therefore, 40 participants (47.1%) exceeded the MID for CARIES-QC and 24 (28.2%) did so for the FIS.

5.3.9 Subgroup analysis

The results of the subgroup analyses are given in *Table 20*. Continuous variables were divided into two subgroups around the mean value. Only CARIES-QC scores had statistically significant correlations with any clinical presentation subgroup analyses, which was for individuals who had pain at their initial assessment visit ($p<0.001$). Those who reported pain had higher CARIES-QC scores on average (mean= 9.24 ± 5.01) than those who did not report having pain (mean 4.24 ± 3.89). The subgroup analysis of demographic variables revealed that those from the BME group had higher CARIES-QC scores on average (mean= 9.87 ± 5.33) than those from the White British group (mean 6.89 ± 5.01). No other statistically significant correlations were identified.

Due to the sample size requirements for the path analysis, it was decided to only include clinical variables which demonstrated a correlation with quality of life scores. The subgroup analyses suggested that, of all the clinical factors, only pain was statistically significantly associated with change in quality of life outcomes. Therefore, this was the only factor related to clinical presentation to be included in the final path analyses.

Table 20: Mean (±SD) baseline scores for subgroups based on demographic and clinical variables, with correlations between variables and quality of life scores

Variable		N	CARIES-QC (raw)		CHU9D		FIS	
			Mean score ±SD	r (p-value)	Mean score ±SD	r (p-value)	Mean score ±SD	r (p-value)
Age	<6.5 years	47	7.43 ± 5.45	0.83 (0.45)	13.32 ± 5.36	0.15 (0.16)	8.66 ± 6.99	0.17 (0.12)
	≥6.5 years	38	8.03 ± 5.02		13.89 ± 4.48		9.89 ± 7.74	
Gender	Male	38	8.50 ± 5.68	-0.12 (0.27)	13.58 ± 4.43	-0.08 (0.48)	9.68 ± 8.34	-0.02 (0.88)
	Female	47	7.04 ± 4.82		13.57 ± 5.40		8.83 ± 6.44	
Ethnicity	White British	63	6.89 ± 5.01	0.26 (0.02)*	13.32 ± 4.55	0.09 (0.40)	8.60 ± 7.02	0.13 (0.25)
	BME	23	9.87 ±5.33		14.52 ± 5.94		10.87 ± 7.99	
Deprivation (based on IMD score)	High (Q1-3)	23	7.00 ± 5.21	0.09 (0.44)	13.13 ± 4.85	0.10 (0.34)	8.65 ± 5.69	0.002 (0.98)
	Low (Q4-5)	62	7.95 ± 5.27		13.74 ± 5.03		9.42 ± 7.86	
Number of carious teeth	≤6.5	42	7.57 ± 5.22	0.16 (0.33)	13.21 ± 3.84	-0.11 (0.30)	8.31 ± 7.17	0.13 (0.23)
	>6.5	43	7.81 ± 5.32		13.93 ± 5.87		10.09 ± 7.41	
Anterior caries present	Yes	10	7.72 ± 5.38	0.02 (0.84)	13.77 ± 5.21	-0.04 (0.72)	9.20 ± 7.33	-0.008 (0.94)
	No	75	7.50 ± 4.28		12.10 ± 1.70		9.30 ± 7.58	
Pain reported at assessment	Yes	60	9.24 ± 5.01	0.46 (<0.001*)	14.03 ± 5.43	0.10 (0.37)	9.51 ± 6.59	0.15 (0.16)
	No	25	4.24 ± 3.89		12.60 ± 3.62		8.60 ± 9.02	
Swelling present	Yes	37	8.14 ± 5.21	0.09 (0.44)	14.00 ± 5.97	0.008 (0.94)	9.89 ± 7.11	0.14 (0.20)
	No	48	7.35 ± 5.29		13.25 ± 4.06		8.69 ± 7.50	

Variable		N	CARIES-QC (raw)		CHU9D		FIS	
			Mean score ±SD	**r (p-value)**	**Mean score ±SD**	**r (p-value)**	**Mean score ±SD**	**r (p-value)**
Received antibiotics	Yes	36	8.28 ± 5.31	0.11 (0.31)	14.00 ± 5.63	0.02 (0.83)	10.67 ± 7.85	0.18 (0.10)
	No	48	7.21 ± 5.25		13.23 ± 4.48		8.10 ± 6.84	

Notes: SD= standard deviation; N= number. BME= Black and ethnic minority group. Q= quintile. r= correlation coefficient (Spearman's rank for continuous variables, point-biserial for dichotomous variables). P-values are for correlations. *=Statistically significant correlation (p<0.05).

5.4 Path analysis

5.4.1 Full theoretical model

The resulting full theoretical model that was tested is shown in *Figure 5.* The lines show the hypothesised paths, or relationships, between variables, with the arrows indicating the direction of influence. As the model included data from variables which were not normally distributed, Satorra-Bentler corrections were applied to all path analyses, and all indices in the following results refer to Satorra-Bentler scaled versions of each index.

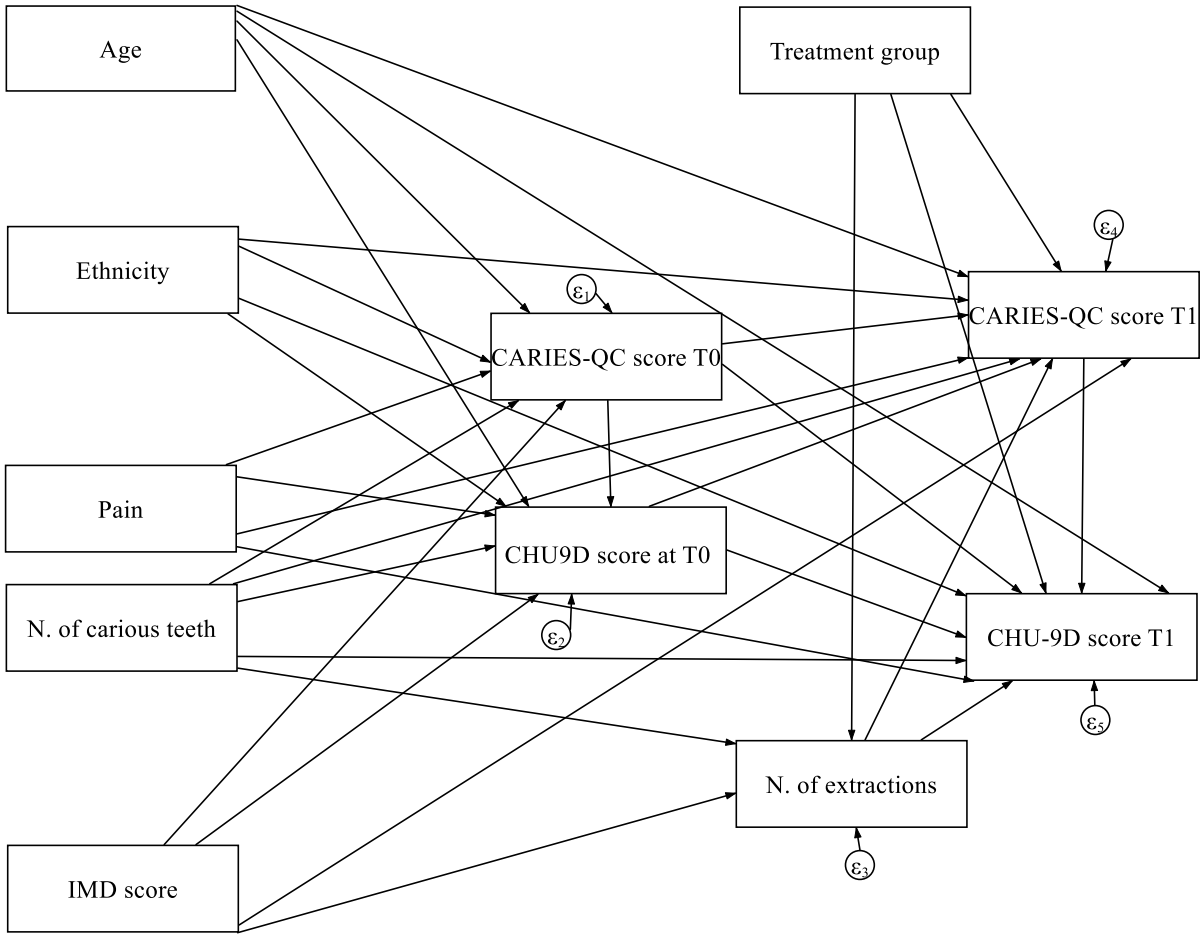

Figure 5: Full theoretical model, showing hypothesised paths between variables. Notes: Ɛ= additional causes or causes outside the model (Olobatuyi, 2006). N= Number. IMD= Index of Multiple Deprivation. T0=baseline. T1= follow-up. CARIES-QC scores are interval scores.

5.4.2 Refining the model

Analysis of the full theoretical model showed a Chi-square (χ^2) of 6.88 (df=8, p=0.55), Log-likelihood= -1931.1695, RMSEA= <0.001, TLI= 1.04 and CFI = 1.00. These results indicated a good fit of the data to the theoretical model. The full results from this initial path analysis are given in *Appendix 7*.

In order to create a parsimonious or 'trimmed' model, non-significant paths were removed from the model. Paths were deemed non-significant if p>0.1 (Olobatuyi, 2006). These removed paths are highlighted in the full results given in *Appendix 7*. *Figure 6* below shows the resultant parsimonious model.

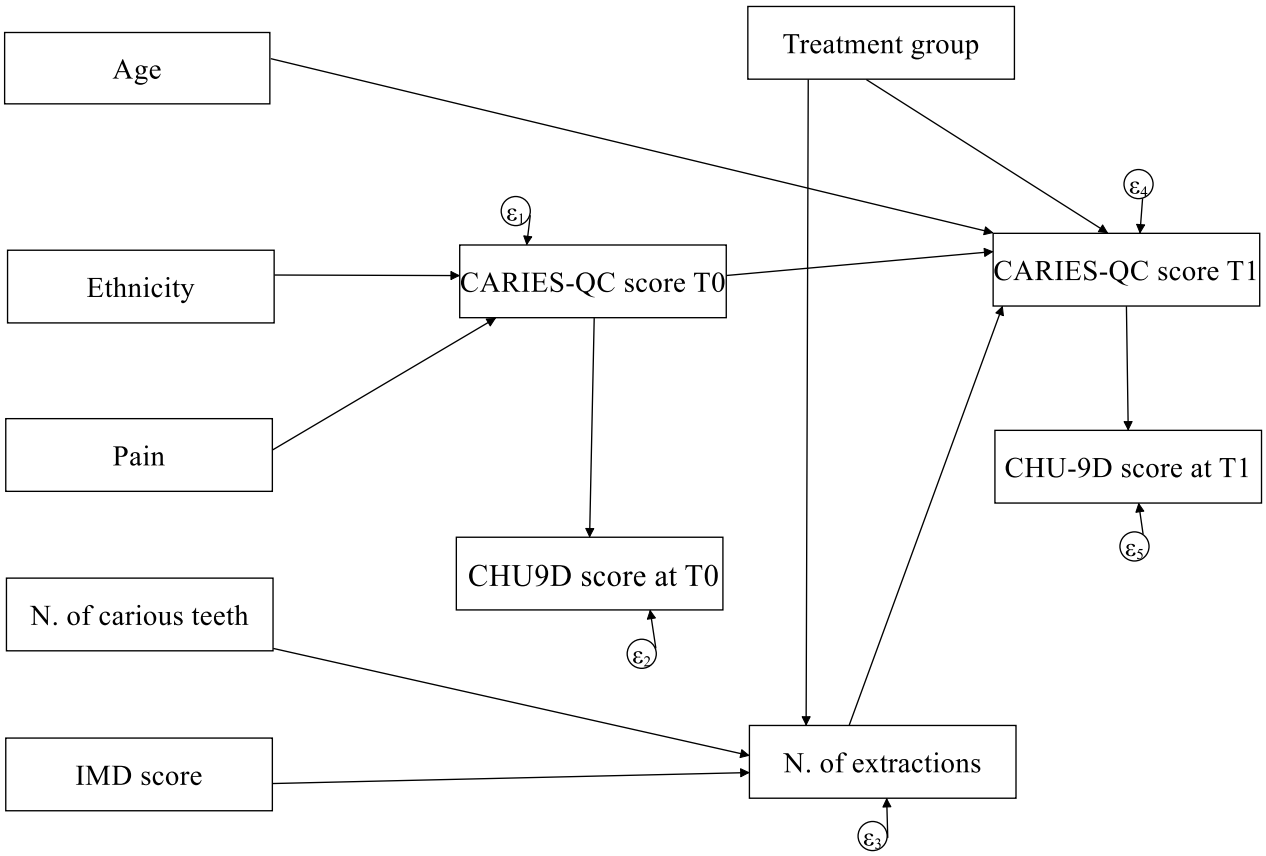

Figure 6: Parsimonious model, showing hypothesised paths between variables. Notes: Ɛ= additional causes or causes outside the model. N=Number. IMD= Index of Multiple Deprivation. T0=baseline. T1=follow-up. CARIES-QC scores are interval scores.

Analysis of the parsimonious model showed a Chi-square (χ^2) of 16.09 (df=28, p=0.96), Log-likelihood= -1936.0644, RMSEA=<0.001, TLI= 1.11 and CFI = 1.00. These results indicated a good fit of the data to the parsimonious model.

Comparison of the full and parsimonious models

The parsimonious model was compared to the full theoretical model using the Chi-square test for a difference between the models and showed p=0.97. This non-significant result at p<0.05 indicated that there was no statistically significant difference between the full and parsimonious models. The parsimonious model was therefore accepted. The results of the path analysis for this final model are given below.

5.4.3 Report of fit indices for the final model

The statistical fit indices for the full and parsimonious models are presented in *Table 21*. The full and parsimonious models both had good fits to the data, meeting all four *a priori* criteria.

Table 21: Summary of fit indices for full and parsimonious models.

Model	**χ^2 (df, p-value)**	**RMSEA**	**CFI**	**TLI**
Full theoretical model	6.88 (8, p=0.55)*	<0.001*	1.00*	1.04*
Parsimonious model	16.09 (28, p=0.96)*	<0.001*	1.00*	1.11*
Ideal value	**p > 0.05**	**< 0.06**	**> 0.9**	**> 0.9**
Notes: χ^2 = Chi-square test of model fit; df= degrees of freedom, RMSEA= Root Mean Square Error of Approximation, CFI= Comparative Fit Index; TLI= Tucker-Lewis Index; * =good model fit.				

5.4.4 Illustration of the final model

The final parsimonious model, detailing the total effects of variables, is given in *Figure 7*. This model shows the standardized β-coefficient and statistical significance of each path in the model.

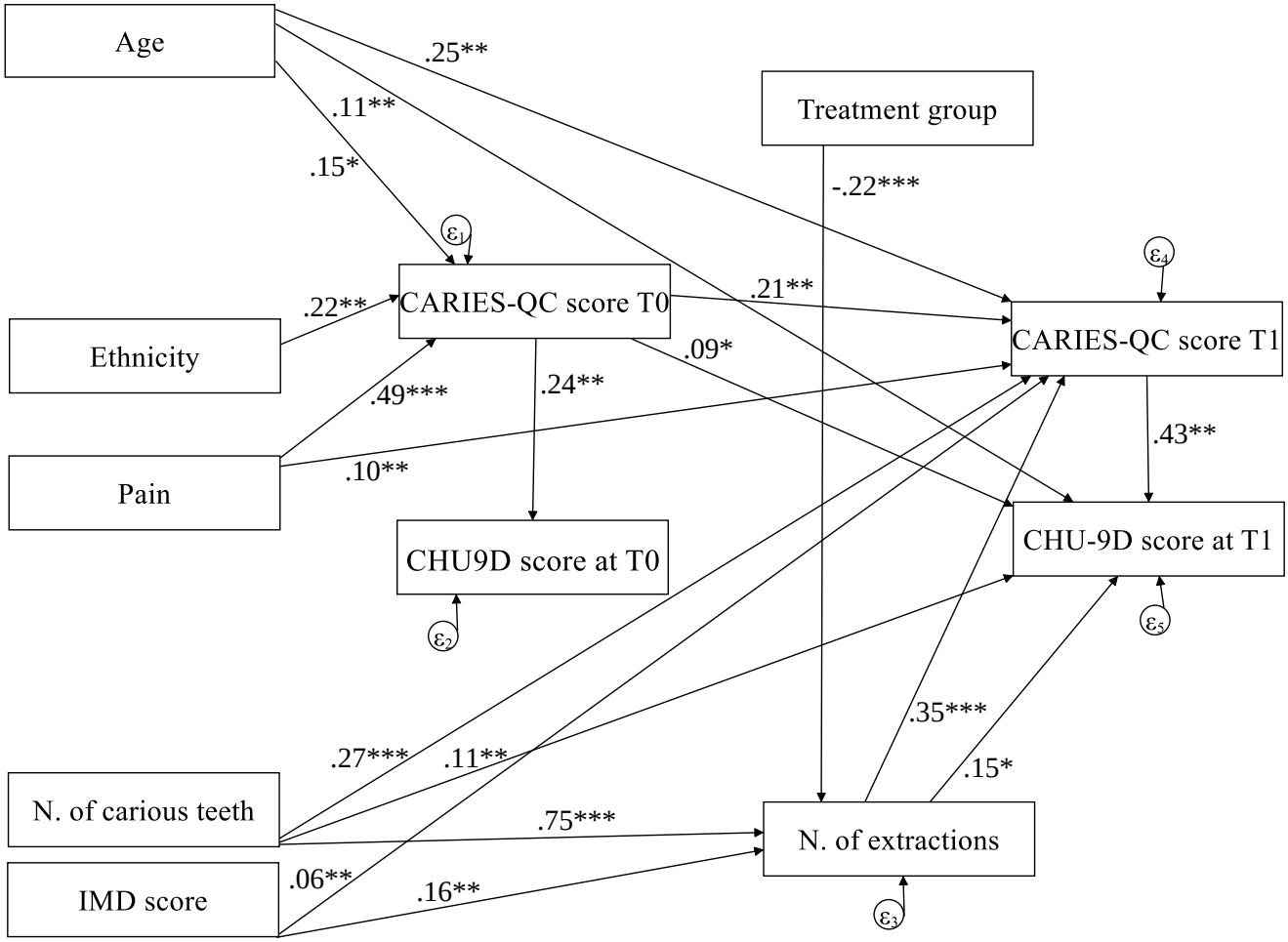

Figure 7: Final model, showing total effects paths between variables and corresponding, standardized, β-coefficients. Notes: ***p<0.01, **p<0.05, *p<0.10. N= Number. IMD= Index of Multiple Deprivation. T0=baseline. T1=follow-up. CARIES-QC scores are interval scores

The full path analysis results for the total effects are available in *Appendix 8*. The standardized β-coefficient of each path shows the relative strength of the effect of each factor in the model. The greatest impact on OHRQoL (CARIES-QC score) at baseline was from pain (β=0.49, p<0.001). At follow-up, the total number of extractions was the most significant factor affecting OHRQoL (β=0.35, p<0.001). The greatest impact on overall QoL (CHU9D) score was OHRQoL, at both baseline (β=0.24, p=0.02) and follow-up

(β=0.43, p=0.002). These total effects comprise of both direct and indirect effects, the full details of which are given below.

Equation-level goodness of fit

In the final model, 51.8% (R^2=0.518) and 24.2% (R^2=0.242) of the variance in CARIES-QC and CHU9D scores, respectively, were accounted for by the variables in the model.

5.4.4.1 Direct effects

Figure 8 and *Table 22* show the direct effects within the model. Statistically significant (p<0.05) effects are identified and are described in detail below.

The standardised β-coefficient for each path helps to identify the comparative magnitude of the impact of each factor. Ethnicity and pain both influenced child OHRQoL at baseline, but pain had a greater effect (β=0.49, p<0.001) than ethnicity (β=0.22, p=0.02). Increased pain and black and minority ethnic group (BME) were associated with higher CARIES-QC scores i.e. worse OHRQoL at baseline. At follow-up, CARIES-QC scores were directly affected by the total number of extractions received and the baseline CARIES-QC score. Increased numbers of extractions were associated with higher CARIES-QC scores, or worse OHRQoL (β=0.46, p<0.001). Higher baseline CARIES-QC scores were associated with higher scores at follow-up but had less of an effect than the number of extractions (β=0.18, p<0.03). Higher CARIES-QC scores at baseline and follow-up were associated with worse overall QoL, i.e. higher CHU9D scores, at baseline (β=0.28, p=0.02) and follow-up (β=0.36, p=0.02) respectively. The total number of extractions

carried out was most strongly affected by the number of carious teeth (β=0.28, p=0.02) and the treatment type (β=0.28, p=0.02), but level of deprivation, as indicated by the IMD score, also had a significant effect (β=0.03, p=0.02). Increased numbers of extractions were associated with exodontia only treatment and higher levels of deprivation.

Table 22: Direct effects of different factors on variables in the final model

Variable	**Factor**	**Satorra-Bentler**				
		β-Coef.	**Std. Err**	**z**	**p-value**	**Standardized β-Coef.**
CARIES-QC score at T0 (Baseline)	Pain	4.57	0.84	5.46	0.00*	0.49
	Ethnicity	2.12	0.91	2.34	0.02*	0.22
	Age	0.42	0.24	0.08	0.08	0.15
CARIES-QC score at T1 (Follow-up)	Number of extractions	0.46	0.11	4.02	0.00*	0.35
	Age	0.54	0.28	1.96	0.05	0.22
	CARIES-QC score at T0 (Baseline)	0.18	0.08	2.15	0.03*	0.21
	Treatment group	1.41	0.77	1.83	0.07	0.17
CHU9D score at T0 (Baseline)	CARIES-QC score at T0 (Baseline)	0.28	0.12	2.37	0.02*	0.24
CHU9D score at T1 (Follow-up)	CARIES-QC score at T1 (Follow-up)	0.36	0.11	3.12	0.00*	0.43
Number of extractions	Number of carious teeth	0.76	0.05	14.35	0.00*	0.75
	Treatment group	-1.39	0.38	-3.63	0.00*	-0.22
	IMD score	0.02	0.01	2.36	0.02*	0.16
Notes: z= standardised value based on standard error (Std. Err.) *= significant result at p<0.05						

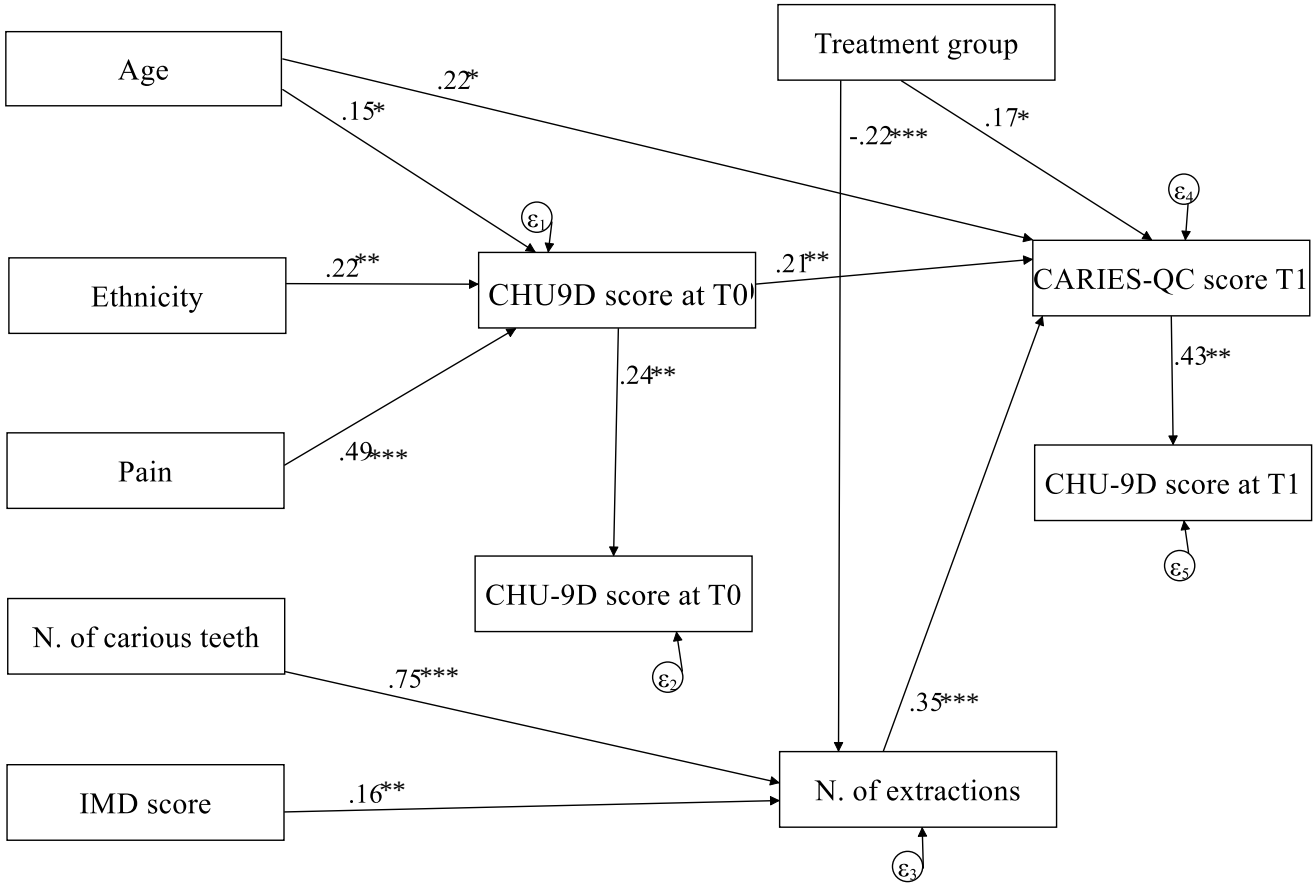

Figure 8: Statistically significant direct relationships in the final model, including standardized β- coefficients for each path. Notes: *p<0.1, **p<0.05, ***p<0.01. N= Number. IMD= Index of Multiple Deprivation. T0=baseline. T1=follow-up. CARIES-QC scores are interval scores. Ɛ= additional causes or causes outside the model.

5.4.4.2 Indirect effects

In addition to the direct effects outlined above, statistically significant indirect effects were also found within the model (See *Figure 9* and *Table 23*). The number of carious teeth, treatment group, pain, and level of deprivation predicted the change in CARIES-QC interval score indirectly. The total number of extractions, the number of carious teeth and age of the child all predicted the change in CHU9D score indirectly. Indirect pathways are as follows, with arrows indicating the direction of effect:

N. of carious teeth → N. of extractions → CARIES-QC score T1 (β=0.27, p<0.001)

Treatment group → N. of extractions → CARIES-QC score T1 (β=-0.08, p=0.005)

Pain → CARIES-QC score T0 → CARIES-QC score T1 (β=0.10, p=0.03)

IMD score → N. of extractions → CARIES-QC score T1 (β=0.06, p=0.04)

Pain → CARIES-QC score T0 → CHU9D score T0 (β=0.12, p=0.02)

N. of extractions → CARIES-QC score T1 → CHU9D score T1 (β=0.15, p=0.02)

N. of carious teeth → N. of extractions → CARIES-QC score T1 → CHU9D score T1 (β=0.11, p=0.02)

Age → CARIES-QC score T1 → CHU9D score T1 (β=0.11, p=0.05)

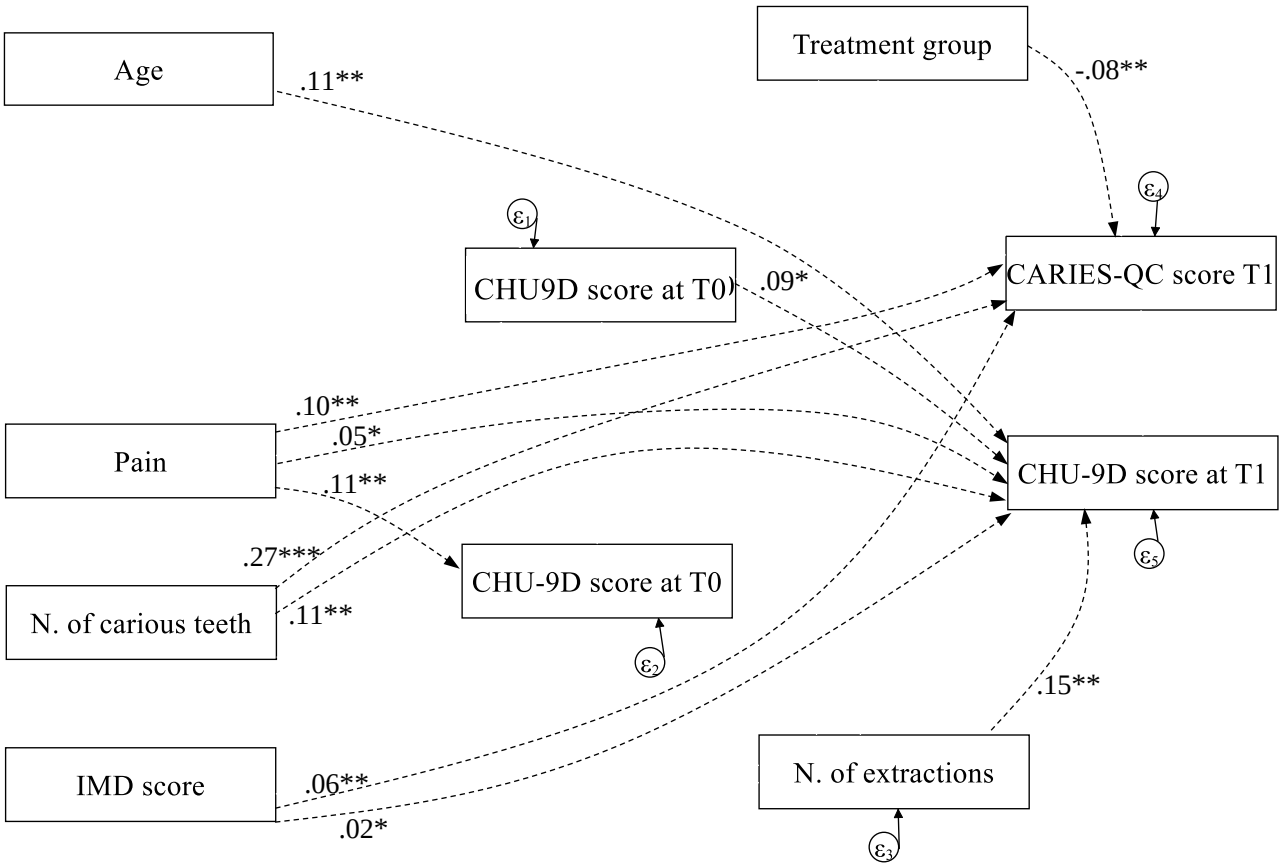

Figure 9: Statistically significant indirect relationships in the final model, including standardised β- coefficients for each path. Notes: *p<0.1, **p<0.05, ***p<0.01. N= Number. IMD= Index of Multiple Deprivation. T0=baseline. T1=follow-up. CARIES-QC scores are interval scores. Ɛ= additional causes or causes outside the model.

Table 23: Indirect effects of different factors on variables in the final model

Variable	Indirect factor	Satorra-Bentler				
		β Coef.	Std. Err.	z	p-value	Standardized β Coef.
CARIES-QC score at T1 (Follow-up)	Number of carious teeth	0.35	0.10	3.62	0.00 *	0.27
	Pain	0.83	0.39	2.13	0.03 *	0.10
	Treatment group	-0.64	0.23	-2.78	0.01 *	-0.08
	IMD score	0.01	0.01	2.01	0.04 *	0.06
	Ethnicity	0.38	0.25	1.56	0.12	0.05
	Age	0.08	0.06	1.34	0.18	0.03
CHU9D score at T0 (Baseline)	Pain	1.27	0.55	2.31	0.02 *	0.12
	Ethnicity	0.59	0.36	1.61	0.11	0.05
	Age	0.12	0.08	1.50	0.14	0.04
CHU9D score at T1 (Follow-up)	Number of extractions	0.16	0.07	2.40	0.02 *	0.15
	Number of carious teeth	0.12	0.05	2.33	0.02 *	0.11
	Age	0.22	0.11	1.98	0.05 *	0.11
	CARIES-QC score at T0 (Baseline)	0.06	0.03	1.86	0.06	0.09
	Pain	0.29	0.16	1.85	0.06	0.04
	Treatment group	0.28	0.31	0.90	0.37	0.04
	IMD score	0.00	0.00	1.72	0.09	0.02
	Ethnicity	0.14	0.10	1.40	0.16	0.02
Notes: z= standardised value based on standard error (Std. Err.) *= significant result at p<0.05						

5.5 Summary of results

The results from this study have demonstrated that dental caries has a significant impact on children and their families. The most common impacts reported by children were related to eating and their teeth causing them to cry; the most common impacts on the family of these children were having sleep disrupted and having to take time off work. The findings have shown that, overall, child OHRQoL and QoL are improved following treatment for dental caries under GA, as demonstrated by statistically significant reduction in CARIES-QC and CHU9D scores ($p<0.001$) with large to moderate effect sizes. Path analysis revealed that the most significant impact on child OHRQoL scores prior to treatment was from pain. Treatment type, and more specifically, total number of extractions was found to have significant impact on scores following treatment. The results have also demonstrated that treatment had a positive effect on the everyday lives of the family, with statistically significant reductions in overall FIS scores ($p=0.03$). Other key results from this study were the proportion of children receiving antibiotics prior to treatment, and that nearly three quarters of the children receiving treatment from caries were from the most deprived areas of the country. These findings are explored in more detail and in relation to the wider literature in the following chapter.

Chapter 6: Discussion

6.1 Introduction

This thesis has presented a systematic review and subsequent investigation of the impact of dental caries and treatment under general anaesthetic (GA) on children and their families. This discussion chapter will now outline the main findings from this study and explore the strengths and limitations of this research. Important aspects of the study design and challenges with the research will also be discussed. The implication of these findings for future research, clinical practice and health policy will be explored.

6.2 Overview

The narrative review in *Chapter 2* highlighted the public health significance of dental caries, and the cost of dental caries and its treatment under GA to individuals and society. While it was found that some aspects of the impact of dental caries were well documented, the majority of reports were from the perspective of professionals and parents rather than children themselves. The systematic review, presented in *Chapter 3*, identified gaps in the current literature, namely the impacts of dental caries and treatment under GA from the child's perspective and what effects are experienced according to different treatment approaches. It was also noted that all studies used generic measures of OHRQoL, which may not be sensitive enough to measure the impacts of caries. This study aimed to address the gaps in the literature by exploring impacts from the child's perspective, using

a disease-specific measure, and by examining the effect of different treatment approaches. This chapter will discuss the key findings from this research and implications for the future.

6.3 Key findings

The aim of this research was to examine the impact of dental caries and its treatment under GA on the everyday lives of children and their families, and this was achieved by fulfilling the objectives below. Key findings are presented in relation to each objective.

Objective 1: Conduct a systematic review of the current literature on the effect of dental treatment for caries under GA on OHRQoL

This objective was met by performing the systematic review described in *Chapter 3*. This systematic review identified key gaps in the current knowledge base and was the first systematic review to analyse the quality of papers reporting on child OHRQoL following treatment under GA.

The systematic review, and subsequent update in 2019, identified that all but one of the studies to date had relied on proxy-reported measures rather than seeking the views of children themselves, and the only study to seek the views of children had focused on older children aged over 11-years-old. There was also a lack of information regarding the impact of different treatment approaches and the longer-term impacts following treatment under GA, i.e. over one-month post-treatment. Quality issues with previous studies were identified, most notably the lack of a clear theoretical framework, no clear calculation of sample size and limited assessment of the reliability and validity of the measures used.

The key findings from this review were the gaps in the current literature, for example, most of the reports were short term (within four weeks of treatment) and relied on proxy/ caregiver reports of child-OHRQoL rather than seeking the views of children themselves. Furthermore, only one study to date had attempted to report on the impact of treatment type carried out under GA and had recommended further work in this area due to the small sample size in the study. The review also highlighted the issues around quality of the included studies, especially a lack of theoretical underpinning to the reported studies, limited evidence of calculation of sample sizes and few statistical assessments of the reliability and validity of measurement tools used. Another key finding from this review was that while there were overall improvements in OHRQoL following treatment under GA, there were some aspects of OHRQoL that worsened following treatment.

The main limitation of the systematic review was that a meta-analysis of the studies was not carried out. The rationale for this was that there was significant heterogeneity between the included studies in terms of methodology, especially with regard to the follow-up period. Since the original systematic review contained within this thesis was published, Park and colleagues (2018) conducted a meta-analysis of those studies employing the ECOHIS or C-OHRQoL measures. The meta-analysis revealed statistically significant improvements in OHRQoL following treatment under GA, with moderate to large effect sizes for both the ECOHIS and C-OHRQoL measure groups. However, there were significant clinical and methodological differences between the studies, so the conclusions that can be drawn from this are limited.

A further limitation with the systematic review conducted was that while papers were included regardless of the original language, it was not possible to calibrate the translator

in order for a quality assessment to be carried out. However, there was only one paper which had to be excluded from the quality analysis for this reason, and therefore the results from the quality assessment should still represent the body of research to date.

Objective 2: Investigate the impact of dental caries and its treatment under GA on children's everyday lives, using a child-centred measure of OHRQoL

Objective 3: Investigate the impact of dental caries and treatment under GA on the families of these children.

Objective 4: Examine the relationships between individual, clinical and environmental factors on children's OHRQoL and QoL, with respect to treatment for caries under GA, using path analysis.

These objectives were met through the study reported in *Chapters 4 and 5*, which sought to address some of the gaps in the literature identified in the systematic review. The impact on children was measured before and after treatment under GA using a caries-specific, child-reported measure of OHRQoL (CARIES-QC) and a child-reported measure of QoL (CHU9D). The impact on the family was measured using the FIS. Overall change, as well as change in individual items or domains in the questionnaires, were analysed for significance using Wilcoxon tests as data were not normally distributed. The clinical significance of the change scores was considered by calculation of effect sizes and minimal important difference. Path analysis was conducted to investigate the relationships between clinical, individual and environmental factors and child-reported quality of life outcomes, guided by a theoretical model.

The key findings from this study were that treatment under GA was associated with statistically significant overall improvements in QoL and OHRQoL, as reported by children, with moderate to large effect sizes. Prior to any intervention, the main impacts of dental caries on children were food getting stuck in their teeth, having to eat on one side and their teeth causing them to cry. All these impacts reduced following treatment under GA. Path analysis suggested that treatment type, but especially the number of extractions, had a significant impact on child OHRQoL following treatment under GA. Path analysis also revealed that pain was the most significant factor affecting child OHRQoL at baseline. The number of carious teeth had no significant direct effects, but indirectly affected child-OHRQoL at follow-up via number of extractions. Child OHRQoL also had significant effects on overall QoL at baseline and follow-up. This was the first time that path analysis had been used to investigate the impact of clinical, individual and environmental factors on child reported OHRQoL and QoL before and after treatment for caries under GA. It was interesting to note that while number of carious teeth did not have significant impact, total number of extractions did.

The overall impact on the family was reduced following treatment with moderate effect size. The main impacts on the family were in the parental and family activity domain, for example, having sleep disrupted or having to take time off work. There was no significant change in scores for the other individual domains, namely parental emotions, family conflict and family finances.

Key aspects of the study design and the main findings are discussed in more detail below, including the extent to which they reinforce or refute other bodies of work. Considerations of the ethical issues, recruitment and difficulties with retention to the study

are discussed, as well as a reflection on the demogrpahic and clinical profile of the participants. The clinical and potential policy implications of the findings are explored. The strengths and limitations of this research are presented, and the implications and recommendations for future research are discussed.

6.4 Ethical considerations

Informed consent

Potential participants were informed, using age-appropriate methods, of the potential benefits and obligations or inconvenience that was associated with agreeing to participate in the study. However, due to the young age of the children to be included in this study, where it was assessed that the child lacked the capacity to give consent, initial consent was obtained from the parent/ caregiver before the child was invited to assent to the research. Therefore, there were some occasions where children were not given the option to be involved because it was against their parents' wishes. Equally, where the child agreed but the parent did not, they were also excluded.

It became apparent during the study that a number of children were very keen to take part but were prevented from doing so because the parents did not wish to be involved. This raises the question about whether, if research is to be truly child-centred, a child should be able to participate without their parent acting as a 'gatekeeper'. This might require future research to have a clear way to assess a child's understanding and ability to consent for themselves. Methods to improve research communication with this group could be explored in future work. Indeed, Parsons and colleagues (2016) identified that, while child-centred research methods have developed in recent years, child-centred consent processes

have not been developed to the same extent. They highlighted the need for innovation in informed consent practices, for example using technologies such as video to explain research to children. They drew on the work of Flewitt (2005) which demonstrated that video can be a useful tool in effectively communicating research to children and their parents, especially in reassuring them about what participation would involve. The use of such tools comes with its own challenges, for example actually producing the videos, but their use could be tested in future studies for the effectiveness in contrast to the paper-based consent process more commonly used at the moment.

Although individuals were not asked why they did not wish to participate, one potential scenario is that parents did not wish to complete a questionnaire themselves but would have been happy for the child to do so. On reflection, given that the family impact was not analysed in relation to the impacts on the child in this study, it would have been possible to just include children's responses, i.e. recruit children if their parents were happy for them to participate, without the parent having to complete a questionnaire themselves.

Power

There is the potential for the power adults have over children in society to be transferred into, and have an effect in, research (Harden et al., 2000). There could have been an effect of power in this study, with children answering questions differently because of the presence of the researcher or dental staff. However, to minimise the effect of this power relationship, children were reminded that the decision to participate was entirely theirs. Where the treating dentist was involved in recruitment, they made it clear that it was a colleague conducting the research, and that the decision to participate had no bearing on treatment. It was made explicit that at any point they may withdraw from the study, and

that this wouldn't make the researcher upset with them or change any treatment they were due to have (Hurley and Underwood, 2015). In addition, those involved in the research were advised to be aware of the body language of participants which may indicate that they are not entirely happy or comfortable with the research or some element of it (Kirk, 2007).

6.5 Participants

6.5.1 Sample size and diversity

The demographics of the patients included in the study sample reflected the overall population of those attending the dental hospital for new patient assessments, and despite significant loss to follow-up in the study, comparison between those lost to follow-up and those completing the study confirmed that there was no difference in baseline characteristics between the two groups. While the study population reflected those typically referred to the dental hospital in Sheffield for treatment, there were important differences with the general population. Nearly three quarters of participants in this study (73%) were living in areas which were within the most deprived two fifths in England, based on Index of Multiple Deprivation (IMD) data derived from their postcode. This is higher than figures for Sheffield as a whole, where around 50% reside in the most deprived areas (Public Health England, 2018). Around 27% of the participants were from black and minority ethnic groups (BME), which slightly exceeds the figure reported for Sheffield as a whole (16%), based on the most recent census data (Office For National Statistics, 2012). These findings support those of previous studies which have shown dental caries disproportionally affects those living in deprived areas and from BME groups (Marcenes et al., 2013; Gilchrist et

al., 2018). The impact of deprivation and ethnicity are considered in more detail later in this discussion.

Importantly, one strength of this study is that sample size calculations were carried out prior to the data collection, ensuring enough patients were recruited to the study for the results to have statistical power. This calculation of sample size was lacking in previous work, as identified in the systematic review.

One limitation of this study was that from the outset it was decided to only recruit those individuals who were able to understand the questionnaire, given some support. This decision was made as, at the time of commencing the study, CARIES-QC was only validated for use in an English-speaking population. This requirement to understand English meant that some individuals were unable to participate in the study. Indeed, where a translator was booked, some potential participants were not approached about whether they would be interested in participating. This was primarily because translation of the questions may have affected their validity, as we could not be sure the translation would be accurate to the meaning of the question, but there was also a practical problem with the use of translation: translators would often have limited time with those attending the new patient clinics, and priority had to be given to ensuring their assessment could be carried out. This may have introduced selection bias into the study, which could only be overcome by ensuring that future work seeks to produce resources in additional languages, which would then need testing for validity and reliability before use.

6.5.2 Recruitment and loss to follow-up

Recruitment to this study took place between January 2017 and January 2019. In total, of the 273 potential participants approached, 61% agreed to participate. It is difficult to determine the reasons for individuals declining to participate, as part of the consent process was assuring participants that they did not have to give a reason should they not want to take part, or at a later stage leave the study. Anecdotally, from observations made by RK and as mentioned above, a number of those approached were unable to participate in the study due to the requirement to understand English. In addition, as mentioned above, the requirement for both parent and child to consent to the study meant that where only one party wished to participate, they could not do so.

One of the problems encountered with recruitment was not being able to get enough participants for each treatment group. It became clear over time that it was difficult to recruit enough children who would receive comprehensive care treatment under GA. One of the reasons for this was that many patients undergoing such treatment also had additional medical needs. The decision was made to only recruit those who were fit and well (ASA 1), as general health would have been a confounding factor in the study and would have increased the required sample size in order for it to be taken into account in the analyses. In addition, some children with complex medical or special needs would have required more support to complete the questionnaires than was available in this setting. Ideally future work would seek to include this group of patients, as well as having a larger sample in order for medical status to be taken into account in path analyses.

Interestingly, the amendment made to the study (see *Chapter 4*) to try and recruit additional comprehensive care patients from the waiting list proved unsuccessful in recruiting any other participants. This suggests that approaching potential participants in person, talking them through the study and addressing any questions, has real value in encouraging participation in research.

While the completion rate of 49% in this study was comparable to similar studies e.g. Yawary et al., 2015 (52%), there was still a significant loss to follow-up. It is worth noting that if participants had not been followed up with telephone calls and text messages then the number not completing the study would have potentially been even greater. Indeed, previous studies have noted that follow-up and additional communication can increase response rates (Huntington et al., 2017). It is well documented that individuals from low-income families are less likely to participate in research studies than those from higher income backgrounds (Heinrichs et al., 2005). While the reasons for this are complex, this work highlights the need to consider how best to reach this group of patients. The inconvenience of completing and returning a paper questionnaire could be considerable. Although participants were given a gift voucher to try and compensate them for their time, it may have been that this was insufficient reimbursement. A systematic review of strategies to improve retention in clinical trials found that the most effective method for increasing response rates to postal questionnaires was to give a monetary incentive, and the higher the incentive the higher the response rates (Brueton et al., 2014). It might be possible in future to make use of other methods, for example, by including other participatory approaches, such as drawings or activities, to encourage the involvement of children (Marshman and Hall 2008).

Analysis was carried out to assess if there was any difference between those lost to follow-up and those who completed the study. If the groups had been significantly different, then the results might have been subject to attrition bias. However, as no difference was found between the two groups it is assumed that the results were not subject to attrition bias from loss to follow-up.

6.5.3 Caries experience

As expected, this population had high levels of caries experience. The most recent survey of five-year old children in England found that the mean dmft in 5-year-olds in Sheffield was 1.1, and the mean dmft for Sheffield children who had caries experience was 3.5 (Public Health England, 2017b). The dmft of children included in this study was considerably higher than this at 6.9, although the figure is similar to that in other studies investigating the impact of treatment under GA in children (e.g. Gilchrist et al., 2018). The decision was made for the analyses to use the number of carious teeth only, as a simple measure of active caries; although this mean figure was only slightly lower than the dmft figure at 6.6. For most of the children this was the number of primary teeth with dental caries, although some of the children participating in the study were in the mixed dentition, therefore their caries experience was determined by combining the number of decayed primary and permanent teeth.

The path analysis found no significant impact of caries experience (as number of carious teeth) and children's OHRQoL. This is in contrast to some previous studies , which found high caries experience was associated with worse OHRQoL (e.g Alsumait et al., (2015) and Foster Page et al., (2019)). However, in these studies, caries experience was

measured as number of decayed, missing and filled teeth; and individuals were grouped into high and no or low experience, where high caries experience was a dmft of four or more. In comparison, this is a high caries experience group and all children had some experience of dental caries, so perhaps it could be expected that there would not be much difference in OHRQoL within the sample. Indeed, Gilchrist and co-workers (2018) found there was only weak association between dmft and CARIES-QC scores in a similar population, with dmft of 6.2. It makes sense that children with caries, rather than being caries free, would report worse quality of life; but it also makes sense that number of decayed teeth might not have an impact. For example, a single carious tooth which is causing pain and keeping a child awake at night is likely to have greater impact on their everyday life than a child with several early carious teeth which aren't causing pain.

Interestingly, in contrast to the work by Gilchrist and team (2018), no significant association was found between anterior caries and OHRQoL. One explanation for this could be the age range of the children. In this study, children had a mean age of 6.5, and only 18% were over 7 years old, whereas in the study by Gilchrist and co-workers 42% of children were over 7 years old (mean=8.1 years). It could be that increased age results in increasing awareness of aesthetic impacts of dental caries, or perhaps increased awareness of how their day to day lives have been changed by their oral health. Indeed, the path analysis showed that age was a significant factor impacting on OHRQoL at follow-up and weakly significant factor at baseline.

Finally, it was clear from the path analyses that pain was the most significant factor associated with dental caries to impact on child reported OHRQoL. This perhaps is an indicator that severity of disease is a more important predictor of OHRQoL than extent of

disease. However, no measure of severity of disease was recorded in this study as the information was not always obvious in the patient records. It would have been useful to have had further information about the severity of disease e.g. whether there was pulpal involvement, to see if this had a significant effect on OHRQoL. These findings are also interesting given that, in most epidemiological studies, dmft/DMFT is used as the indicator for burden of disease in populations. Future studies could perhaps attempt to define caries severity and extent in more detail to better understand the effect of caries on OHRQoL. One possible way to take into account the extent or severity of dental caries is the International Caries Detection and Assessment System (ICDAS). Whereas the dmft/ DMFT index allows recording of the number of carious teeth only, the ICDAS allows recording of dental caries along a continuum of progression from enamel, to dentine, then pulp. It also differentiates between teeth with a distinct cavity (with visible dentine) and those with a more extensive cavity involving more than half of the tooth surface (Gugnani et al., 2011).

6.5.4 Deprivation

This study has added to the body of literature which suggests that dental caries affects children from all sections of society, but those from the most deprived areas carry the bulk of the disease burden, as nearly three quarters of those requiring treatment under GA in this sample were living in the most deprived areas of England. The epidemiological data from the most recent child dental health survey in the UK found that children from more deprived backgrounds were disproportionately affected by dental caries. In five-year-olds, 41% of those eligible for free school meals, which is linked to deprivation, had caries experience compared to 29% of other children (Steele et al., 2015). These findings support

those in other studies where children receiving treatment under GA tended to be from socially deprived backgrounds (Hariharan et al., 2017). It is therefore perhaps not surprising that a significant number of children in this population were from more deprived areas.

6.5.5 Safeguarding concerns

There were several children in this study (n=8, 9.4%) who had safeguarding concerns in place. Perhaps more worryingly, a greater number (n=12, 14.6%) of children with safeguarding concerns in place were lost to follow-up, some of whom will have done so because they failed to return for further treatment. These children would have been processed through the departmental protocol for children who are not brought to appointments; which would have included communication with the safeguarding teams to ensure they were seen again at a later stage. However, this process means that there would have been a delay in these children receiving the treatment they required.

Another concerning finding in this study was that in 27% of cases the family had another child who had also required treatment for caries under GA. There are several important implications of these findings. Firstly, and perhaps most important, there is the impact of dental caries on these children, and the potential of further impact if they are then not brought to further appointments for treatment. Second, is the implication for the involvement of dental services in both recognising when dental caries or failure to attend for treatment might be an indicator that the child is at risk, or at very least, the family need more support.

Previous studies have reported on higher than average levels of caries in various vulnerable groups, including children in who are looked after, have a history of maltreatment or adverse childhood experiences and children who have substance-using parents (Harris, 2018). In some cases, this gives rise to dental neglect, which is defined as 'the persistent failure to meet a child's basic oral health needs, likely to result in the serious impairment of a child's oral or general health or development' (Harris et al., 2018). As discussed in *Chapter 2*, and as the results from this study support, untreated dental caries can have considerable impact on children, and may lead to pain, impaired quality of life and occasionally even life-threatening spreading infection.

Failure or delay in seeking dental care, including for dental caries, is a cause for concern, all the more so in the UK where child dental care is available free-of-charge on the NHS and cost is not a barrier to access. It is only more recently that missed appointments for dentistry have been considered from a safeguarding perspective (Harris, 2018). Studies have shown that even among paediatric dentists' referral in response to concerns is rare. One study found that despite 67% of those questioned having recognised signs of dental neglect, only 29% had ever referred these concerns (Harris et al., 2009). The results of this study support the literature highlighting dental caries as a potential indicator of dental neglect, and the importance that these children are followed up when they fail to attend. Further work is needed to help dental professionals grow in their knowledge and confidence surrounding safeguarding issues in children, and also to help identify and remove barriers to acting on their concerns.

6.5.5 Antibiotic use

The results from the study also show that significant numbers of children (43.5%) had received antibiotics prior to their initial assessment for a GA, with some children receiving several courses. These findings are concerning in the light of growing antibiotic resistance. It brings into question the prescribing practices of dentists, and whether these antibiotics are always indicated. Certainly, anecdotally, there were cases of children with no history of systemic spread of infection who had received antibiotics, and very few children had received analgesics to manage pain from toothache. It may be that antibiotics are viewed as a course of 'treatment' in the interim period from referral to assessment, where a dentist is unable to carry out treatment in the dental chair. Indeed, a survey of prescribing practices in the North of England found that there was only evidence of spreading infection or systemic involvement in approximately half of the cases, and that other reasons such as patient expectations, time pressure and lack of co-operation were impacting on the decision to prescribe antimicrobials (Sturrock et al., 2018). Although antibiotic prescribing in dentistry has reduced in recent years, 5.2% of all antibiotics are prescribed in dentistry (Public Health England, 2018); suggesting more needs to be done. Common approaches with medical colleagues to educate the wider public about the need to reduce antibiotic use and why antibiotics might not be appropriate is advocated, alongside support for dentists to have the confidence and time to do the right thing. This has implications for practice, such as ensuring urgent care appointments are available and appropriately remunerated to allow time for appropriate treatment to be carried out.

6.6 Impacts on children

The study was designed to test the following hypothesis: 'Treatment for the management of dental caries under GA results in the improved oral health-related quality of life, as reported by children.' The findings in relation to this hypothesis are discussed below.

6.6.1 CARIES-QC

CARIES-QC was chosen as it was the only disease-specific, child reported measure of OHRQoL available at the time of the study, and as such on reflection this choice is still valid. This study has supported previous work which showed CARIES-QC was suitable for use with children as young as 5 years of age (Gilchrist et al., 2018). It is the first study to investigate the effect of treatment under GA on children's everyday lives using a child-reported measure, building on the work carried out by Gilchrist and colleagues (2018) who explored change following treatment for caries as part of psychometric analysis of CARIES-QC in a smaller sample.

Overall, children had statistically significant improvements in OHRQoL following treatment under GA, with large effect sizes (d=0.91). These findings are consistent with previous work summarised in the systematic review (*Chapter 3*) which found overall improvements in OHRQoL with moderate to large effect sizes, albeit over shorter follow-up periods and using generic, proxy-reported, measures of OHRQoL. At baseline, 55% of children reported their teeth were a problem at baseline, compared to 24% at follow-up.

Despite some children still reporting their teeth were a problem at follow-up, the response to the global transitional judgment question revealed that nearly all children (92%) rated their teeth as 'better' following treatment. No children thought their teeth were 'worse' following the GA. These findings support the literature that reports overall improvements in children's quality of life following treatment for caries under GA (Gaynor and Thomson, 2011; de Souza et al., 2016; Knapp et al., 2017). The findings also suggest that initial improvements following treatment under GA for caries noted in previous studies, which commonly had follow-up times of up to one month, are maintained in the longer term i.e. three-month period.

In contrast to previous studies, however, CARIES-QC has provided insights into the aspects of a child's daily life which are most improved following treatment, from their own perspective rather than relying on a proxy report. Analysis of individual question responses revealed that the most common impacts reported by children prior to treatment were food getting stuck (reported by 86% of children) and feeling cross (72%), with all impacts reducing following treatment, supporting the findings by Gilchrist and team (2018). The findings showed that different impacts were reduced to varying degrees, with the greatest reduction in those reporting their sleep being affected (91.0% reduction in children reporting this impact at follow-up). The percentage reduction in impact was smaller for questions related to eating such as food getting stuck or having to eat on one side. This finding can perhaps be explained in part by the treatment being carried out. Where children have received extractions, especially multiple extractions, is it conceivable that this may result in continued, or even increased, difficulty eating. These findings reveal that overall improvements may mask the effect in individual impacts, but that treatment for

caries under GA results in improvements on all aspects of children's everyday lives to varying degrees.

It was interesting to note that even when children rated their teeth 'better' or 'the same' following treatment some individual overall CARIES-QC scores did not follow the expected pattern e.g. some CARIES-QC total scores increased even when a child rated their teeth better overall. This finding is perhaps supported by previous work which highlighted worse proxy-reported child OHRQoL in some subscales following dental GA. For example, two studies (Lee et al., 2011; Cantekin et al., 2014) found an increase in mean score for the ECOHIS 'child self-image and social interaction' subscale and another study found an increase in mean P-CPQ 'social wellbeing' subscale score (Klaassen et al., 2008).

It may be that, while a child may rate an improvement in their teeth overall, they may experience additional impacts from having treatment. It is conceivable that, for example, if a child had several carious teeth removed, treatment may have reduced previous impacts but generated new ones, for example when eating. A video-diary study by Rodd and team (2014) found that the most negative impacts reported by children themselves immediately following a GA were disturbed eating and hunger. A limitation of this study is that the reason why individual OHRQoL deteriorated was not investigated. It would be useful to explore whether worse OHRQoL scores is related to other factors, such as normal development changes, i.e. exfoliating primary teeth or eruption of permanent teeth. Future work should seek to explore the underlying reasons for children reporting worse OHRQoL scores following treatment, potentially to include a qualitative approach to gain depth of insight in this area.

6.6.2 CHU9D

This is the first study to use the CHU9D to explore the impact on a child's overall QoL following treatment for caries under GA. The study has added to the evidence that CHU9D is acceptable for use in children as young as 5 years of age, with support from their parents (Stevens and Ratcliffe, 2012; Canaway and Frew, 2013; Foster-Page et al., 2015). It has shown that children's overall QoL improved following treatment for dental caries under GA, but also demonstrated an important relationship between OHRQoL and overall QoL.

Overall CHU9D scores statistically significantly decreased following treatment, with moderate effect sizes. Previous work had questioned whether the CHU9D was suitable for use as an outcome measure for child oral health after finding there was not a significant difference in scores between children with and without dental caries (Foster Page et al., 2014). However, this was in a low caries population (mean dmft= 2.4). Our findings show that CHU9D may be useful in longitudinal studies exploring the impact of different treatment types for dental caries. As well as evaluating QoL, use of CHU9D in child oral health research will allow calculation of quality-adjusted life years (QALYs) for use in economic evaluation (Stevens and Ratcliffe, 2012).

The findings also showed that there were reductions in all individual domains following treatment under GA. At baseline, the greatest impacts reported by children were being tired (60%) and worried (52%), both of which reduced following treatment. Percentage reduction in individual impacts ranged from 5.9% (difficulty with schoolwork) to 73.5% (feeling worried). It was interesting to note that while there was a 91% reduction

in children reporting they had been kept awake by their teeth in their responses to CARIES-QC, there was only a 32% reduction in children reporting they had problems sleeping in their responses to CHU9D. This could reflect the wording of the question; or could reflect that the CHU9D is a generic measure. Whereas CARIES-QC is asking, specifically, if the child is kept awake by their teeth, CHU9D is asking whether they have problems sleeping generally, and therefore responses may be impacted by other factors.

This perhaps brings into question what other factors are impacting on the children's overall quality of life. Indeed, the path analysis revealed that only 24% of the variation in CHU9D score was accounted for by the variables in the model. This suggests that other factors are having a significant effect on overall quality of life in these children, and further research is needed to explore what these might be.

One important finding from the path analysis was that OHRQoL had a statistically significant effect on overall QoL at baseline ($p<0.05$) and follow-up ($p<0.001$); i.e. those children with higher CARIES-QC scores had corresponding higher CHU9D scores overall. This adds to the literature which has discussed the impact of oral health, and specifically dental caries, on children's overall quality of life.

6.6.3 Other impacts

Several papers in the systematic review looked at whether treatment under GA impacted on child anxiety in relation to dental treatment. There were conflicting results in the literature, in some studies anxiety was less in the short term (e.g. Cantekin *et al.*, 2014), but in others, treatment under GA had no effect on anxiety (e.g. Klaassen *et al.*, 2008 and

2009). The results from the individual domains in CHU9D would suggest that children felt less 'worried' overall following treatment, but it is not clear from the findings how this relates to dental anxiety. In this study, dental anxiety was not explored as the completion of another measure would have increased the response burden for the children. In addition, it would be difficult to establish the effect of GA on dental-related anxiety as children would not be returning for further treatment. Research into reducing dental anxiety, and whether treatment under GA has a positive or negative impact, is important given that anxiety is a key reason for children to receive treatment under GA (Savanheimo and Vehkalahti, 2014).

6.6.4 Conclusion

The overall results for change in CARIES-QC and CHU9D scores following treatment mean that the null hypothesis can be rejected, and that the results show that overall child-reported OHRQoL was improved three-months following treatment under GA. There is further support for the hypothesis in that all individual impacts reduced in incidence following treatment. However, as discussed above, the overall results can hide some of the important differences between individuals and further research is required to better understand the factors which may result in worse outcomes following treatment.

6.7 Impacts on the family

The study was designed to test the hypothesis: 'Treatment for the management of dental caries under GA has a positive effect on the family.' The findings in relation to this hypothesis are discussed below.

6.7.1 FIS

This study has added knowledge about the impact of caries, and subsequent treatment under GA, on the family of those children receiving treatment. The overall scores from the FIS following treatment showed a statistically significant reduction in score, i.e. improved quality of life, with moderate effect sizes. These findings support those in similar studies, which found significant improvements in the FIS scores one month following GA treatment (e.g. de Souza et al., 2016; Yawary et al., 2016), and suggests the improvement seen in these studies is maintained in the longer term. The global question results showed that overall, most parents felt their child's oral health and quality of life improved following treatment, and just over half noted an improvement in the everyday life of their family. These findings support the literature base which has identified high parental satisfaction rates with treatment under GA (Anderson et al., 2004), and adds weight to the justification of providing treatment under GA to children with dental caries.

Only one individual FIS domain showed no significant change, namely the parental and family activities domain. In contrast to other studies, the economic domain showed no significant change. However, this may reflect the public funding of UK healthcare systems, as highlighted in similar studies in the UK (de Souza et al., 2016). In addition, the importance of loss of earnings through time off work may not have been so great if parents and caregivers were not currently working. Unfortunately, information was not gathered on the parental employment status, so it is not possible to determine if this was the case in this sample (see limitations, below).

This study did not investigate the family impact in more detail, i.e. through path analysis as it was decided to focus on the child impacts due to sample size requirements. It was therefore not possible to assess the impact of additional factors on the FIS results. For example, family structure and functioning are thought to influence family impact results, but it was not possible to control for these in this study (Thomson et al., 2013). As mentioned above, some family factors would be especially important to ascertain to understand the individual domain effects in more detail, e.g. parental employment status on questions regarding economic impact. This investigation of family factors would have increased the participant response burden, but also the time required for analysis and interpretation, so they were not included in this study. However, these factors are important considerations for future work seeking to explore family impacts in more detail.

6.7.2 Conclusion

Statistically significant reduction in overall FIS scores suggest that the null hypothesis can be rejected, and that there is evidence to support the alternative hypothesis; namely that treatment for dental caries under GA has a positive effect on the family'. Only the parental and family activities domain showed statistically significant improvements following treatment. Further work is needed to explore the impact of additional factors, such as family structure, on FIS results in order to try to account for the variation seen in the results.

6.8 Psychometric properties of the measures

A limitation of previous work was the limited psychometric testing of measures used. Therefore, in this study, analysis of the validity and responsiveness of the measures was carried out to address this quality issue with previous studies. The main findings in relation to each measure are discussed below.

CARIES-QC

CARIES-QC demonstrated good overall internal consistency (Cronbach alpha= 0.9), and had few floor and ceiling effects, which supports findings in previous work. For example, Gilchrist and co-workers (2018) also found CARIES-QC had an alpha value of 0.9 in a similar population. This value is slightly higher than that found in work by Foster-Page and colleagues (2019), who obtained an alpha of 0.8; although this figure still represented good overall consistency. This difference in figures may be due to the different levels of caries in the populations in the studies. This study adds to the evidence from other studies that CARIES-QC has good longitudinal construct validity and responsiveness, as the mean scores for CARIES-QC among those whose global response 'improved', and 'stayed the same' had positive mean change scores and scores closer to zero, respectively (Gilchrist et al., 2018; Foster Page et al., 2019). Interestingly, the large effect sizes seen for change in CARIES-QC scores in this study were greater than those previously reported by Foster Page and colleagues (2019), who found moderate effect sizes. This may be because all the child participants in this study were requiring treatment under GA, which may reflect a greater treatment need than in the other study populations. These previous studies also found that for individuals whose global rating worsened the measure was not as responsive. However, in this study, none of the children felt that their teeth were worse at follow-up,

so it was not possible to test the responsiveness of CARIES-QC further for this global response. Further work using CARIES-QC in different populations, including those with different treatment needs, is required to establish its reliability and validity in other situations. This should include evaluation of the use of the measure in longitudinal studies, with those with different rates of dental caries, but also with those with different demographics such as older children and those from other ethnic and socio-economic backgrounds.

CHU9D

Only limited psychometric testing of CHU9D was possible in this study. As with CARIES-QC, CHU9D demonstrated acceptable consistency, as revealed by the Cronbach alpha of 0.8, which was an improvement on the level of alpha of 0.66 found by Foster Page and team (2014) and similar to findings by Furber and Segal (2015), suggesting the items are demonstrating better consistency in a dental population with higher rates of dental caries. Given this difference in consistency of results in the literature, further testing of the measure, perhaps by factor or Rasch analysis, is perhaps indicated to establish which items might not be performing as well.

Although a previous study using CHU9D found it to be unresponsive to changes following caries treatment, the authors suggested that further research was needed using CHU9D in populations with higher rates of dental caries (Foster-Page et al., 2015). Unfortunately, as no global transition judgement question was used for CHU9D it was not possible to further test the measure in terms of longitudinal construct validity and responsiveness. Further psychometric testing of this measure is therefore warranted, to fully assess its suitability for use in longitudinal studies.

FIS

The FIS also showed good overall internal consistency, as revealed by the Cronbach alpha of 0.8. However, the results showed that for the individual domains within the scale there was poor internal consistency for the parental emotions domain. This may indicate that further refinement of the measure is required for use in this population. The FIS measure is a generic measure, and as such may not be sensitive enough to detect caries-related impacts on the family.

The results showed that the FIS demonstrated poor longitudinal validity and poor responsiveness to change over time. This means that the results need to be interpreted with some caution, but also brings into question the usefulness of the FIS in longitudinal studies. Previous studies have demonstrated good internal validity and reliability of the measure in cross-sectional studies but suggested that further work is needed in longitudinal studies (Barbosa and Gavião, 2009; Thomson et al., 2013). The exact reasons for this finding are not clear at this stage. This lack of responsiveness to change over time may reflect the fact that some aspects of family life remain unchanged following treatment (e.g. feeling guilty) or that, as previously mentioned, the measure is not sensitive enough to detect changes in relation to dental caries. It is also possible that the lack of responsiveness could also relate to the wording of the global transition judgement question i.e. the global transition question is itself not valid. Further research is therefore indicated to further test the longitudinal validity and responsiveness of the FIS in other populations and should include testing of other global transition questions to establish which is most suitable.

Conclusion

All three measures showed good internal consistency and had few floor and ceiling effects. CARIES-QC demonstrated good internal consistency, longitudinal construct validity and responsiveness, supporting its use to evaluate change following treatment for dental caries in children. In contrast, the FIS demonstrated poor validity and responsiveness in this population; and variable internal consistency. This may be because it is a generic rather than caries-specific measure, unable to detect changes in caries-related impacts on the family. Further psychometric testing of all measures, especially in larger samples is warranted. Factor and Rasch analysis may be helpful to establish which questions may not be performing as well.

6.9 Effect of treatment approach and other factors

This study was also designed to test the hypothesis: 'Participants receiving restorations as well as extractions report greater improvements in OHRQoL change scores than those receiving extractions only.' The findings in relation to this hypothesis are discussed below.

6.9.1 Treatment approach

In contrast to the study by de Souza and co-workers (2016), who found no difference between comprehensive care and exodontia only treatment approaches on OHRQoL score in children, the direct effect of treatment group on child OHRQoL in this study was significant at $p<0.05$. This difference may be because the sample size in the de Souza may have been too small to detect any between-group differences, or that the measure of OHRQoL used (P-CPQ) was not detecting differences; perhaps because it was

proxy-reported or because it was a generic measure which may not have been sensitive enough to capture caries specific impacts. The path analysis in this research also allowed for control of other confounding factors which may have hidden an effect.

Another key difference between this study and that by de Souza and colleagues was that the analysis of treatment approach looked at combined care and exodontia only under GA, whereas their research compared comprehensive care under GA and exodontia only under GA. The combined care group in this study composed of those who received treatment on clinic followed by exodontia under GA and those who had comprehensive care under GA. Unfortunately, there were insufficient numbers in each group to examine these separately, and that is a limitation with these findings. Further work in larger samples should seek to explore these different approaches in more detail.

Interestingly, the direct effect of combined care treatment was higher CARIES-QC scores at follow-up, or worse OHRQoL, than those who had extractions only ($p=0.01$). However, this effect was less than that of the number of extractions, which meant that overall, those who had exodontia only (and therefore more extractions, see below) were likely to have worse OHRQoL scores at follow-up. It is not clear from the results why the direct effect of combined care was worse OHRQoL at follow-up than those for exodontia only, and further research would need to explore the differences between the groups in more detail to understand this finding. It might be that the increased waiting time for combined care treatment is having an impact on the results.

The waiting times for those completing exodontia only treatment in this study were 9.4 weeks, compared to 16.7 in the combined care group. Previous research has found that

parents reported concerns over how long their child had to wait for GA treatment and the negative effect these waiting times had, including continuing or increased pain and sleepless nights (Goodwin, Pretty and Sanders, 2015). Sample size requirements meant that waiting time could not be considered in the path analyses in this study. Further work is therefore needed to explore the impact of other factors in relation to treatment approach which may impact on outcomes, such as waiting times, to understand the effect of treatment approach more thoroughly.

6.9.2 Number of extractions

The results of the study identified a significant difference in the number of extractions being carried out between those in the exodontia only group and those who received combination care. Those in the exodontia group were on average having 1.5 more extractions than those in the combined care group. The path analysis revealed that it was this number of extractions, rather than treatment approach *per-se*, that was having the greatest impact on child OHRQoL following treatment. Previous studies (Lee et al., 2011; Cantekin et al., 2014) have noted that some OHRQoL domains worsen following treatment, and it would make sense that if prior to treatment the children did not have significant impacts from their carious teeth that the sudden loss of teeth could affect both function, in relation to eating, and appearance.

Previous work has noted that in centres were exodontia only is available, children may have more teeth extracted, to remove all signs of caries and hopefully prevent the need for future GA treatment (Harrison and Nutting, 2000; Goodwin et al., 2015). In many ways, this approach makes a lot of sense. However, the impact of this treatment approach has not

been explored from the child's perspective before. The findings from this study would suggest that increased number of extractions is having a significant effect on the quality of life of the children. While this is a limited sample, the findings suggest there may be a case for clinicians to explain the potential impact of choosing extractions (over restorations) to parents and children, where there is an option. This novel finding could have important implications for treatment planning and provision of care to those with high rates of dental caries, although further work in larger samples is needed to test these findings further.

6.9.3 Other clinical factors

Overwhelmingly, the greatest impact on OHRQoL, and indirectly on overall QoL, was from pain with those children who were experiencing pain as a result of dental caries having statistically significantly higher CARIES-QC scores. It was interesting that from the subgroup analyses that other factors, namely the presence of swelling, receipt of antibiotics and the presence of anterior caries did not have a significant effect on scores. These findings are perhaps understandable but are interesting to consider in relation to the severity of disease, revealing that pain is the most important factor for children in terms of the impact on their day to day lives. The results highlight the need for urgent treatment to reduce the impact of pain on children with dental caries.

6.9.4 Demographic factors

Previous work has highlighted that those from more deprived backgrounds are more likely to experience dental caries, and the findings from this study support this, with nearly three-quarters of children living in areas which are amongst the most deprived areas of

England. What this study adds, however, is that children living in more deprived areas had significantly more extractions than those living in less deprived areas, and that this is impacting on their quality of life. Path analysis revealed that level of deprivation was not impacting directly on OHRQoL and QoL in children. However, it was having a statistically significant effect on the number of extractions received, and therefore indirectly impacting on both CARIES-QC and CHU9D scores at follow-up.

The exact reason for this difference in number of extractions is unclear. It may be that children living in more deprived areas present later and therefore teeth are less likely to be restorable and require extracting. It may be that the parents or children would prefer teeth to be extracted than restored. Certainly, there was a trend within families, with over a quarter of children in the study having a sibling who had also had dental treatment under GA, perhaps indicating that oral health advice offered with previous children had not had the desired effect. Although this study did not explore parental attitudes, previous work has found that 53% of parents did not understand the importance of primary teeth (Akhlaghi et al., 2017). Further research would need to be carried out to explore the relationship in more detail. It is well documented in the literature that many parents view GA as an acceptable method of addressing their child's oral health needs, allowing them to interact socially soon afterwards (Goodwin, Pretty and Sanders, 2015) and complete treatment in a single session (Anderson et al., 2004). It might be that parents, and for that matter clinicians, view extractions as the best way to complete all treatment and reduce the need for future treatment. Whatever the reason, however, these findings could have important implications for targeted oral health promotion programmes in these areas, such as the 'Childsmile' programme in Scotland (See *Chapter 2*), to reduce inequalities surrounding caries in this population.

6.9.5 Other findings

The path analysis confirmed that individual factors, namely ethnicity and age, were impacting on OHRQoL, but were having far less impact than other factors. Older age and black and minority ethnic group were associated with worse OHRQoL at baseline and follow-up, respectively. Ethnicity was only a significant factor at baseline, and age at follow-up. This finding, i.e. the effect was not present at both time points, is interesting and somewhat difficult to explain. It may be there is an effect at both timepoints but that it is too small to be captured by the sample size in this study. Other studies have hypothesised that worse OHRQoL in some ethnic groups may not be purely related to ethnicity but to associated factors such as cultural differences in oral health practices, socioeconomic status or parental education (Çolak et al., 2013). Further work would be needed to explore the factors related to ethnicity which may impact on OHRQoL. Regarding age, there are conflicting results in the literature, which perhaps explains why age is only having a minor impact in this study. It might be that increasing age results in increased awareness of oral problems, or more especially changes following treatment, and therefore age had an effect at follow-up but not at baseline.

6.9.6 Conclusions

The path analysis is an important novel aspect of this research, which gives further detail about the relative impact of different factors on OHRQoL and QoL. Overall, there was a good fit of the data to the model. The study was designed to test the hypothesis: 'Participants receiving restorations as well as extractions report greater improvements in OHRQoL change scores than those receiving extractions only.' The results from the study

suggest this could be the case, but further work is needed to explore the effect in more detail. Certainly, path analysis reveals that increased extractions results in worse OHRQoL at follow-up; but other factors (such as waiting times) could also be impacting on outcomes.

A limitation with this work was that the number of factors which could be analysed was constrained by the sample size. The proposed theoretical model explained 52% of the variance in CARIES-QC scores, but only 24% of the variance in CHU9D scores. This is useful, as it implies that further work is needed to fully explain which factors are impacting on OHRQoL, but more especially QoL, in children with dental caries undergoing treatment under GA.

6.10 Study strengths

Child-centred approach

One of the strengths of this study is that it sought to obtain the views of children themselves, rather than relying on parental or caregiver responses. This addressed a gap in the literature but was also important from an ethical standpoint. Much dental research has been carried out *on* children, rather than *with* and *for* them. This work has added to the work of others which seeks to address this imbalance. As recommended by Marshman and team (2015), in this research children were viewed as active participants. Measures were chosen which had been developed with children, and which had been validated for children to complete themselves. It adds evidence to the body of work which shows children are capable and willing to discuss the oral health impacts they have experienced (Rodd et al., 2013; Gilchrist, 2015; Gilchrist et al., 2015). This study has added knowledge about the

impact of caries, and subsequent treatment under GA, on children's everyday lives from their own perspective.

Methodology

Although the ideal study design when investigating treatment effect would have been a randomised controlled trial, in this instance it would be impossible to randomise to treatment. It would be unethical to withhold treatment from some individuals to form a suitable control group (i.e. for no intervention) (Mann, 2003). Therefore, as in previous studies (Yawary et al., 2015; de Souza et al., 2016), an observational approach was adopted and a prospective longitudinal pretest-posttest study design employed, which is the most commonly used study design for this type of research. However, the novel use of path analysis in this study is one of the important benefits of this work. While complete control for confounding is not possible in a non-randomised study, measured characteristics that differed between individuals (e.g. age and deprivation) were considered in path analyses to examine their potential impact on the quality of life outcomes. This methodology should be considered for use in future work exploring treatment impacts where randomisation is not possible.

Use of a theoretical model

One of the main limitations identified in the quality assessment of papers included in the systematic review was that studies were not underpinned by an explicit theoretical framework. One of the strengths of this work is that it was driven by the Wilson and Cleary model for HRQoL, which has been widely used in other dental research, and items included in the model were selected on the basis of previous studies exploring their role in OHRQoL (Baker et al., 2010; Gururatana et al., 2014; Vettore et al., 2019).

The study has made several important additions to the literature. This is the first study to explore the effects of clinical, environmental and individual factors on OHRQoL following treatment under G.A. It has identified the impact of treatment approach, and more specifically number of extractions, on child OHRQoL. It is also the first study to demonstrate the impact of child reported OHRQoL on their overall quality of life following treatment for caries under GA.

6.11 Study limitations

Lack of a control group

The main limitation of an observational study design is that without a cross-matched control group it is not possible to confirm whether the change in child reported HRQoL and QoL is entirely due to the treatment for caries. As mentioned, however, it would be unethical to recruit children and then withhold treatment from them. It would have been interesting to recruit a cross-matched caries-free control group, to allow for comparison. However, practically this would be very difficult to do. It would be difficult in terms of where to recruit this cross-matched group from, and also in terms of matching the group for other factors, such as deprivation and ethnicity.

Changes over time and response shift

One of the other limitations with the methodology of this study is that it is not possible to determine whether the quality of life outcomes changed between initial assessment and the GA itself. Ideally, children and parents would have been asked to complete the questionnaires again just before their GA. However, given the difficulties with follow-up in this group, and due to practical considerations, it was not possible for this to

be carried out. It was felt that asking families to complete questionnaires on the day of the GA itself would have been impractical but also the results could have been affected by the fact they were receiving treatment that day, e.g. they may have been more anxious than usual.

Related to this problem of change over time is the fact that this study did not attempt to calculate or analyse response shift. Response shift is the term used to describe how individuals change over time and how the basis on which they make judgements about their health-related quality of life may also change. For example, an individual may have a change in their internal standards or values (Ring et al., 2005). Future studies should, therefore, seek to explore the effect of response shift, to see if any such effect is present when exploring the effect of treatment on OHRQoL. In order for this effect to be investigated, it would be necessary to see children on more occasions e.g. just prior to their GA and just after, which comes with the additional burden to participants and potential loss to follow-up. This is particularly difficult in this population, as previously discussed, and therefore a study to explore response shift may need to be conducted in a different population, or alongside courses of treatment where individuals are expected to return on more than one occasion to the clinics.

Limited depth of insights into child perspectives and family impacts

This study adopted a purely quantitative methodology, which has some limitations. While it allowed generation of significant amounts of data, and allowed comparison with previous studies, this approach meant that there was no generation of deeper insights into the effects on children and their families. The study identified an overall improvement in QoL outcomes for children and their families but was unable to explain in detail how this

manifested in their day to day lives. In addition, the study was not able to identify the reasons why some individual scores worsened, even when their global responses were 'better' overall.

Impact of other factors

As mentioned, path analysis was used to examine the impact of potential confounding factors on the child-reported quality of life outcomes. The main limitation of this work is that it was not possible to include more sample characteristics due to the sample size requirements and so as not to increase the response burden for participants. Future work in larger samples could allow for more sophisticated analysis of potential confounding factors such as anxiety, sense of coherence and family parenting styles, which have previously been shown to impact on OHRQoL (e.g. Baker et al., 2010). Future research could explore these factors to add to our understanding of the impact of other individual and environmental factors on child OHRQoL. In order for the larger sample sizes to be reached, it may be necessary for future studies to adopt a multi-centre approach.

Another issue with the sample size requirements is the potential for type II errors to occur where there is a small sample size. A type II error occurs when the effect being investigated is found to be insignificant, when in fact there is an effect i.e. there may be significant effects, but the sample is too small to detect it. The sample size calculation in this study were based on detecting a large effect size of 0.8 at 5% level of significance and 80% power. That means that in all the statistical analyses an effect was only detected as significant if it was a large effect (0.8). One benefit of this is that that any effects which are detected are unlikely to be in error. However, it does mean that some of the other factors

examined in this study may have an effect, but it was too small to detect. Therefore, further work using a larger sample is warranted.

6.12 Implications for clinical care

This study has demonstrated that CARIES-QC is suitable for investigating OHRQoL in children receiving dental treatment. Consideration should be given to the use of CARIES-QC routinely for Paediatric Dentistry. This would allow additional testing of the measure, but also provide information on the impacts of a range of oral health conditions on the everyday lives of children.

In addition, some children had experienced impacts which may not have come up in discussion with the dentist, or which parents may not be aware of. Identifying impacts, such as difficulties eating or sleeping, may help clinicians to more accurately assess a child's need for treatment, as while they might not report pain, caries may be having other effects on their daily lives which may improve following treatment. Therefore, the routine use of CARIES-QC in the clinical environment may help clinicians better understand the impacts that children are experiencing, allowing treatment to be tailored to reducing these.

These findings may have important implications for treatment planning for dental caries. If removal of additional teeth is likely to have a negative impact on OHRQoL, there may be a case for trying to restore as many teeth as possible. This is a complex discussion, as a number of competing factors are at play. In a high-risk group, the rationale to remove rather than restore teeth may result in preventing further treatment under GA in the future.

Further work is needed to confirm the effect of number of extractions, and comprehensive care versus exodontia only, to ensure the best treatment approach is adopted.

6.13 Implications for policy

This study has shown that dental caries has significant effects on the everyday lives of children and their families, which appear to improve following treatment under GA. This would imply that, nationally, child oral health needs to be a priority, and policies developed which seek to prevent the disease but also to minimise the impacts experienced by children. It is already well documented that dental caries has a number of common risk factors with obesity and Type II diabetes, therefore thought should be given to a common risk factor approach to reduce the impact of all these diseases (Sheiham and Watt, 2000).

The study has highlighted the link between deprivation and dental caries, and identified a link between deprivation and increased number of extractions, impacting on OHRQoL. These findings suggest that targeted interventions to more deprived populations are important in reducing oral health inequalities. Such nationwide interventions have been used elsewhere in the UK with success in reducing inequalities, for example, 'Childsmile' in Scotland (McMahon et al., 2011) and 'Designed to Smile' in Wales (Morgan, 2018). There is currently no national funded oral health promotion programme in England. Another important factor in policymaking is to ensure there is remuneration for dentists to carry out preventative activity and to provide restorative care before extractions are required (Watt et al., 2019). A recent government green paper, published in the UK, has recognised the need for modernisation of prevention services but also to provide ways to make good health, and healthy choices, easier for the population. This included a section

on prioritising child oral health, and within this prioritising tooth brushing schemes in schools and an investigation into removing barriers to water fluoridation (Department of Health and Social Care, 2019).

6.14 Implications for future research

This study has highlighted some important areas for future research. Several of the limitations with the present study can only be addressed if more data, i.e. larger sample sizes are available. For this reason, it would be valuable for a multi-centre study to take place, to further examine the effect of different treatment approaches in children having a GA for dental caries. Further work is needed to explore aspects of comprehensive care and combined care, e.g. waiting times, which may impact on outcomes. Work exploring the impact of numbers of extractions is also warranted, to add to the evidence provided in this study. Larger samples would also allow further psychometric testing of CARIES-QC, CHU9D and the FIS in other populations and in longitudinal studies to confirm their suitability of use.

As well as an increase in the number of participants, future work could also focus on gaining greater depth of insight than was possible in the present study. For example, qualitative studies to explore the impacts of treatment under GA for caries in children, to investigate why some children report worse OHRQoL following treatment or to capture in-depth views of parents in relation to the impacts experienced by their family. In addition, future work should consider exploring the effect of response shift following treatment under GA, to determine whether this impacting on the results.

Chapter 7: Conclusions and recommendations

7.1 Introduction

The work within this thesis aimed to examine the impact of dental caries and its treatment under general anaesthetic (GA) on the everyday lives of children and their families. The specific objectives were to:

1. Conduct a systematic review of the current literature on the effect of dental treatment for caries under GA on OHRQoL.
2. Investigate the impact of dental caries and its treatment under GA on children's everyday lives, using a child-centred measure of OHRQoL.
3. Investigate the impact of dental caries and its treatment under GA on the families of these children.
4. Examine the relationships between individual, clinical and environmental factors on children's OHRQoL and QoL, with respect to treatment for caries under GA, using path analysis.

These objectives were met through a systematic review and observational study. The systematic review identified key gaps in the current literature, which were then addressed in the research. The research presented provides additional knowledge regarding

the impacts of dental caries and subsequent treatment under GA on the everyday lives of children and their specific families. In particular, the research sought the views of children themselves, used a disease- measure of OHRQoL and looked at the longer-term impact of treatment. In addition, path analysis was carried out to investigate which factors in the proposed theoretical model were impacting on OHRQoL outcomes.

7.2 Summary of findings

Previous studies exploring the impact of dental caries and treatment under GA have largely relied on proxy-reported measures of OHRQoL. Very few studies to date have had a clear theoretical framework, and other quality issues were evident such as limited psychometric testing of the measures used and lack of sample size calculations to ensure analyses were suitably powered. This study sought to address those gaps in the knowledge base.

Treatment under GA was associated with significant improvement in QoL and OHRQoL, as reported by children, as well as overall improvements on the family. The main impacts of caries reported by children were related to eating and their teeth causing them to cry. All impacts reported by children were reduced following treatment, albeit to varying degrees. A further notable finding was the fact that 44% of the children had been given antibiotics for their dental problem prior to attending the dental hospital, providing some insight into the proportion of children with dental caries who are prescribed antibiotics, which potentially has worrying implications in the fight against antimicrobial resistance. Post-treatment, there were statistically significant improvements in overall child- reported OHRQoL scores ($p<0.001$), child-reported QoL scores ($p<0.001$) and

family impact scores (p=0.03). The change in scores represented moderate to large effect sizes. Path analyses suggest the data were a good fit to the proposed theoretical model. The path analysis revealed that treatment type, but specifically, the total number of extractions had a significant effect on OHRQoL following treatment. Increasing numbers of extractions was associated with worse OHRQoL outcomes. Further work in larger samples is needed to explore the impact of other factors, such as waiting times for treatment, in more detail.

7.3 Recommendations for clinical care and policy

- Routine use of patient reported outcome measures (such as CARIES-QC) in Paediatric Dentistry to allow further testing of the measure and greater understanding of child-reported impacts from oral conditions.
- Consideration of these findings in treatment planning and communication with children and their families of the potential benefits and risks of treatment in relation to the impacts on their everyday lives.
- Prioritisation of prevention of dental caries and a common risk-factor approach to reduce impacts on children, with consideration to targeted schemes to help reduce socio-economic inequalities.

7.4 Recommendations for future research

- Investigate the impacts of other factors in relation to treatment approaches for caries, including the impact of waiting times, in larger samples.

- Further psychometric testing of CARIES-QC, CHU9D and the Family Impact scale in other populations and in longitudinal studies to confirm their suitability of use.
- Investigate the response shift following treatment under GA
- Qualitative studies to explore the impacts of treatment under GA for caries in children, to investigate why some children report worse OHRQoL following treatment.
- Qualitative studies to capture in-depth views of parents in relation to OHRQoL.

7.5 Conclusion

Treatment under GA was associated with significant improvement in QoL and OHRQoL as reported by both children and their parents. Path analysis suggests that treatment type, via number of extractions may impact on child OHRQoL and QoL following treatment under GA. A number of areas for recommended further research have been identified, with a priority towards investigating the findings in larger samples and different populations. Such studies should seek to investigate the effect of treatment approach, and in particular number of extractions, in more detail. The results have implications for treatment planning and the provision and commissioning of dental services.

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Appendices

Appendix 1: Ethical approval letter

Lothian NHS Board

South East Scotland Research
Ethics Committee 02

Waverley Gate
2-4 Waterloo Place
Edinburgh
EH1 3EG
Telephone 0131 536 9000

www.nhslothian.scot.nhs.uk

Date 25 October 2016
Your Ref
Our Ref

Enquiries to: Joyce Clearie
Extension: 35674
Direct Line: 0131 465 5674
Email: Joyce.Clearie@nhslothian.scot.nhs.uk

Please note: This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

25 October 2016

Mrs Rebecca Knapp
Oral Health & Development PG Office,
School of Clinical Dentistry, Claremont Crescent
Sheffield
S10 2TA

Dear Mrs Knapp

Study title:	**The effect of dental treatment for the management of dental caries under general anaesthetic on the oral health-related quality of life of children**
REC reference:	**16/SS/0187**
IRAS project ID:	**211730**

The Proportionate Review Sub-committee of the South East Scotland REC 02 reviewed the above application on 25 October 2016.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this favourable opinion letter. The expectation is that this information will be published for all studies that receive an ethical opinion but should you wish to provide a substitute contact point, wish to make a request to defer, or require further information, please contact the REC Manager Ms Joyce Clearie, joyce.clearie@nhslothian.scot.nhs.uk. Under very limited circumstances (e.g. for student research which has received an unfavourable opinion), it may be possible to grant an exemption to the publication of the study.

Ethical opinion

On behalf of the Committee, the sub-committee gave a favourable ethical opinion of the above research on the basis described in the application form, protocol and supporting documentation, subject to the conditions specified below.

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Participant information Sheet

- Please consider rewording the statement "As part of my PhD" as this may not mean a lot to some people – perhaps explain what a PhD is?

You should notify the REC once all conditions have been met (except for site approvals from host organisations) and provide copies of any revised documentation with updated version numbers. Revised documents should be submitted to the REC electronically from IRAS. The REC will acknowledge receipt and provide a final list of the approved documentation for the study, which you can make available to host organisations to facilitate their permission for the study. Failure to provide the final versions to the REC may cause delay in obtaining permissions.

Management permission must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for HRA Approval (England)/ NHS permission for research is available in the Integrated Research Application System, www.hra.nhs.uk or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations.

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database. This should be before the first participant is recruited but no later than 6 weeks after recruitment of the first participant.

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact hra.studyregistration@nhs.net. The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from the HRA. Guidance on where to register is provided on the HRA website.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion").

Summary of discussion at the meeting

- **Social or scientific value; scientific design and conduct of the study**

This was considered a worthwhile project and overall the Committee had no significant concerns with the social or scientific value, scientific design and conduct of the study.

There were some concerns raised that there is a need for a control group – otherwise conclusions could be drawn which are not valid as they are present/not present in a control group as well. Suggest in further research consider whether could they recruit a group of children who for example are having orthodontics rather than being treated for tooth decay.

- **Informed consent process and the adequacy and completeness of participant information**

Over all this was considered acceptable. Minor rewording of the participant information sheet was suggested.

Overall considered acceptable in following areas or no further comments:-

- **Recruitment arrangements and access to health information, and fair participant selection**
- **Favourable risk benefit ratio; anticipated benefit/risks for research participants (present and future)**
- **Care and protection of research participants; respect for potential and enrolled participants' welfare and dignity**
- **Suitability of the applicant and supporting staff**
- **Independent review**
- **Suitability of supporting information**
- **Other general comments**
- **Suitability of research summary**

Approved documents

The documents reviewed and approved were:

Document	*Version*	*Date*
Copies of advertisement materials for research participants [poster]	2	28 September 2016
IRAS Application Form [IRAS_Form_18102016]		18 October 2016
Other [Email confirming academic supervisor IRAS authorisation]		18 October 2016
Participant information sheet (PIS) [PIS_age5-7]	4	26 September 2016

Participant information sheet (PIS) [PIS_age8-11]	4	26 September 2016
Participant information sheet (PIS) [PIS_age12-16]	4	26 September 2016
Participant information sheet (PIS) [PIS_parent]	4	26 September 2016
Research protocol or project proposal [protocol]	3	20 October 2016
Summary CV for Chief Investigator (CI) [CV_knapp]	2	11 October 2016
Summary CV for supervisor (student research) [cv_zm]		29 May 2016
Summary CV for supervisor (student research) [CV_fg]		19 July 2016
Summary, synopsis or diagram (flowchart) of protocol in non technical language [plain language summary]	2	03 October 2016
Validated questionnaire [questionnaire_child_initial (incl. consent form)]	2	23 August 2016
Validated questionnaire [questionnaire_child_followup (incl. assent form)]	2	23 August 2016
Validated questionnaire [questionnaire_parent_initial (incl. consent form)]	2	23 August 2016
Validated questionnaire [questionnaire_parent_followup (incl. consent form)]	2	23 August 2016

Membership of the Proportionate Review Sub-Committee

The members of the Sub-Committee who took part in the review are listed on the attached sheet.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- ☐ Notifying substantial amendments
- ☐ Adding new sites and investigators
- ☐ Notification of serious breaches of the protocol
- ☐ Progress and safety reports
- ☐ Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/

HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at http://www.hra.nhs.uk/hra-training/

With the Committee's best wishes for the success of this project.

16/SS/0187	**Please quote this number on all correspondence**

Yours sincerely

Ms Joanne Mair
Chair

Email: joyce.clearie@nhslothian.scot.nhs.uk

Enclosures: *List of names and professions of members who took part in the review*

"After ethical review – guidance for researchers" [SL-AR2]

Copy to: *Mrs Samantha Walmsley*

NHS Lothian

South East Scotland REC 02

Attendance at PRS Sub-Committee of the REC meeting on 25 October 2016

Committee Members:

Name	*Profession*	*Present*	*Notes*
Dr Yann Maidment	General Dental Practitioner	Yes	
Ms Joanne Mair	Research Facilitator	Yes	
Dr Hester Ward	Public Health Consultant	Yes	

Also in attendance:

Name	*Position (or reason for attending)*
Dr Alex Bailey	Scientific Officer
Ms Joyce Clearie	REC Manager

Appendix 2: Age-appropriate child and parent/caregiver study information sheets

Age appropriate information sheets were provided as follows, for children aged 5-7 years, 8-11 years and 12-16 years, and to parent/ caregivers.

Participant information leaflet (Children aged 5-7)

The University Of Sheffield.

CREATE

Teeth and dentists: what do you think?

Who am I?

My name is Becky and I work at the University.

This is me

What am I doing?

I want to find out what children think about their teeth

Can you help me?

I would like you to fill in a quiz for me. I will ask you to fill in the same quiz next time you come to the dentist.

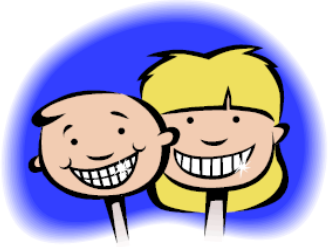
There are no right and wrong answers. It should be fun!

What happens after?
I will write a story to tell other people what I've found out.

After you do both quizzes, I will give you a small gift to say thank you for helping me.

What if you don't want to join in anymore?
If you don't want to do it anymore, you can stop at any time.

No one will be cross.



What do you do now?
There is a sheet to fill in. If you would like to join in, please but the first letter of your first and last names in the boxes on the sheet. You can get an adult to help you with this. Then fill in the quiz and give the sheet back to me.

If you have any questions or are unhappy, you can phone me on 0114 271 7877 or email becky.knapp@sheffield.ac.uk or the Patient Services Team on 0114 271 2400 or email PST@sth.nhs.uk

Thank you for reading this. I hope to meet you soon.

Becky

Participant information leaflet (Children aged 8-11)

The University Of Sheffield.

CREATE

Teeth and dentists: what do you think?

Who am I?

My name is Becky Knapp and I work at the University of Sheffield.

This is me

What am I doing?

Research tries to find an answer to an important question. I am doing research with children who have holes in their teeth. I want to find out how you feel about the holes in your teeth and about the work you've had done to your teeth. I hope this will make coming to the dentist easier for children.

Can you help me?

I would like you to answer some questions for me. I have a sheet for you to fill in. It will take about 20 minutes. There are no right or wrong answers. I will ask you to fill in the same sheet after you have had work done to your teeth.

Do I have to join in?
No. I hope that you will want to join in but you don't have to. It is up to you.

What happens after?
The details about you and your answers will be kept private so no-one outside of the research will know it is you. Afterwards I will write about what I have found out from the research. You will get a letter to tell you what I have found. After you complete your treatment and the other set of questions your family will be given a £10 gift voucher to say thank you for helping me.

What if something goes wrong?
We can't see anything going wrong during this project. If you or your parents feel unhappy about anything, we are very happy to talk to you at any time.

What if you don't want to join in anymore?
If you want to stop doing the research at any time, you can stop without giving a reason. Just tell me or your parent/ guardian. No one will be cross.

What do you do now?
There is a sheet to fill in. If you would like to join in, please check you agree with what is written on the sheet and put your initials in the boxes to say you agree. You can then answer the questions. If you don't want to join in, that is fine.

You might have some questions to ask me about the research. That is fine, you can ask me anything.

What should I do if I have questions or a problem?
You can phone me on 0114 271 7877 or email becky.knapp@sheffield.ac.uk

If you have a problem you can also contact Mrs Tracey Plant, Clinical Hospital Manager, Charles Clifford Dental Hospital, Sheffield S10 2SZ or the Patient Services Team on 0114 271 2400 or email PST@sth.nhs.uk

Thank you for taking the time to read this. Please feel free to ask any questions if you need to.

Teeth and dentists: what do you think?

Participant information sheet (Children aged 12-16)

Part 1: To give you an introduction to the study

What is this research and why is it being done?
Hello, thank you for reading this information sheet. This will give you information about the study I am doing and which I would like to invite you to take part in. My name is Becky Knapp and I am a researcher at the University of Sheffield Dental School. Before you decide whether you are happy to take part, it is important that you know why the research is being done and what it will involve. Please take time to read this leaflet and talk with others if you wish.

Why am I doing this research?
The purpose of my project is to find out how having holes in your teeth affects you and how you feel about any treatment you have had.

Why have you been asked to take part?
You have been invited because you attend the clinic here. We don't know very much about young people's experience of treatment, so I'm hoping to find out more from people like you. I hope that about 150 young people will fill in the questionnaire, so you're not the only one.

Do you need to take part?
No! It is up to you. If you do, you:

- Will be asked to initial boxes to give your consent
- Will be given a copy of your information sheet to keep
- **Will be free at any time to stop taking part without giving a reason. If you decide to stop, it will not affect the care you receive.**

What will I be asked to do?
You will be asked to fill in the questionnaire and then asked to repeat it again next time you come to the clinic, 3 months after you have had treatment. The questionnaire will take about 20 minutes to do.

I will also need access to information about you in your medical records, such as your age and what treatment you have done. We will keep your personal information private, so no-one will be able to identify you from the information we use.

What do I do now?
There is a sheet to fill in if you do want to take part. Please put your initials in the boxes on the sheet to say you are happy to take part. If you don't want to join in, that's fine.

Is there anything to be worried about if I part?
There are no known risks to you or your parents from taking part in the study. You don't have to tell us about anything you don't want to.

What are the possible benefits of taking part?
The study will not change the care or treatment you receive at the dentist. The study will not benefit you directly, but we hope that the study will help children in the future. After you complete the second questionnaire, your family will be given a £10 gift voucher as a thank you for your time.

Contact details
If you have any questions or want to find out more, please contact me by telephone: 0114 271 7877 or email becky.knapp@sheffield.ac.uk

Thank you for reading so far, if you're interested in taking part please read on.

Part 2: More detail, information you need to know if you want to take part

What happens when the research stops?
When the study is finished I will look at all the information that I have gained from you and other young people. I will then write a report on my findings and send you and your parents a copy. You will continue your regular dental care as normal. If you would like to take part with a further questionnaire in the future, you have the option to do so- just tick the box on the questionnaire to say you are happy for us to contact you again. We will only store your personal

Teeth and dentists: what do you think?

Participant information sheet (Parent/Guardian)

Part 1: To let you know a little about the project

What is this research and why is it being done?

Hello, thank you for reading this information sheet. This will give you information about the study I am doing and which I would like to invite your child to take part in. My name is Becky Knapp and I am a researcher in the University of Sheffield Dental School. Before you decide whether you are happy for yourself and your child to take part, it is important that you know why the research is being done and what it will involve. Please take time to read this leaflet and talk with others if you wish.

What are you researching?

As part of my PhD, I am carrying out research to find out how tooth decay and its treatment can affect children and young people and their families. To help me do this, I have questionnaires for children and their parents to fill in.

Why are you doing this research?

We don't know much about how children and young people feel about dental decay and its treatment or how this impacts on their families. We hope this questionnaire will help us find out more.

Why do you want to talk to my child and me?

I have approached you and your child as your child has attended the dental clinic for treatment. I am hoping to get about 200 children of different ages to take part.

Please turn the page

Do we have to take part?

No! It is up to you and your child. If you both decide to take part, you:

- Will be asked to initial boxes to give your consent to take part
- Will be given a copy of your information sheet to keep
- **Will be free at any time to stop taking part without giving a reason. If you or your child decides to stop, it will not affect the care your child receives.**

What will happen if we agree to take part?

I would like you and your child to fill in a questionnaire before treatment on their teeth to find out more about how tooth decay affects them. The questionnaire will take about 20 minutes to complete.

If you child needs treatment under general anaesthetic, then I would like you and your child to repeat the same questionnaires 3 months after your child has completed their treatment. You can complete the questionnaires when you come back to the clinic after treatment, or the questionnaire can be sent to your home and it can be returned to us in the stamped addressed envelope provided.

I will also require your permission to access your child's clinical records. The purpose of this is to record details about your child's teeth and any treatment they have received. This information will be kept anonymous so your child cannot be identified from the research.

Is there anything to be worried about if my child takes part?

There are no known risks to you or your child from taking part in the study. Your names will not appear in any report written about the study, so you do not need to worry that other people will know what you've said

What are the possible benefits of taking part?

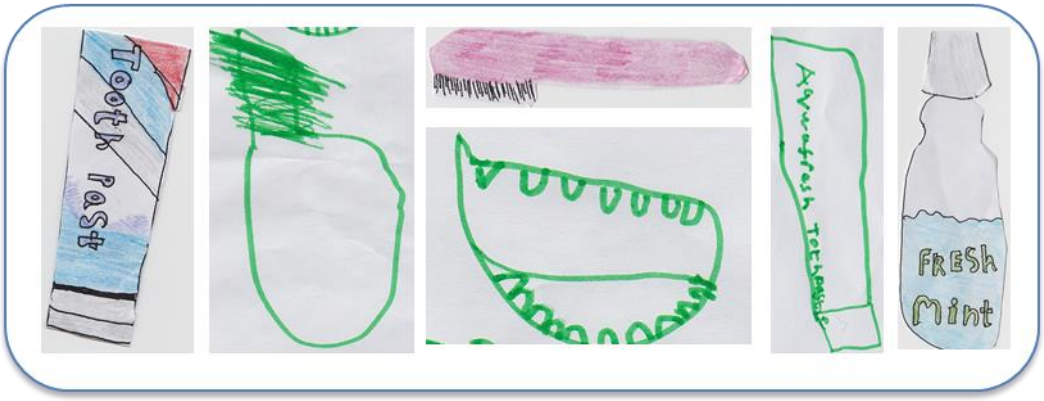
The study will not change the care or treatment your child receives at the dentist. The study will not benefit your child directly, but we hope that the study will help children with dental decay in the future.

If your child has treatment under general anaesthetic and you both complete the questionnaires again, your child will receive a £10 gift voucher to say thank you for continuing in the study and for the time you have given.

Please turn the page

Appendix 3: Child consent and questionnaires

Questionnaire for children about how you feel about your teeth and the dentist



Hello

Thank you for agreeing to help us with our study.

This study is being done so we can find out more about how young people feel about their teeth and work they have done at the dentist. By answering the questions, you will help us to find ways to make young people feel happier about visiting a dentist.

In this booklet, you will find some sets of questions about you, how you feel about your teeth and the dentist.

We would be very grateful if you could answer all the questions using the instructions.

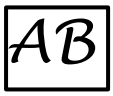
There are no right or wrong answers. Some of the questions may seem to be asking the same thing but each question tells us about something slightly different that we would like to find out.

Please ask your parent or guardian if you need help with this.

On the next page you can agree to take part in the study.

Please go to the next page →

Section 1 Are you happy to take part?

It is up to you if you want to take part. Before you start, can you **put your initials** in the boxes below **if you agree** with each statement like this: 

Please ask an adult to help you if you need to:

	Initial here
I am happy to answer the questions and for you to use my answers in your research	☐
I am happy for you to get details about me and my treatment, as long as these are kept private so no-one outside of the research will know it is me	☐
During the study, I am happy to be contacted by phone	☐
During the study, I am happy to be contacted by post	☐
I am happy to be contacted in the future about similar research (you do not have to agree to this to take part in this study)	☐

Please write the date here:

Thank you for agreeing to take part

Please go to the next page to start the questions! →

Section 2: How do you feel?

These questions ask about how you are **today.** They are <u>not</u> about going to the dentist. For each question, read all the choices and decide which one is most like you **today**. Then put a **tick** ☑ in the box next to it. Only tick **one** box for each question.

1. Worried	
☐ 1	I don't feel worried today
☐ 2	I feel a little bit worried today
☐ 3	I feel a bit worried today
☐ 4	I feel quite worried today
☐ 5	I feel very worried today

2. Sad	
☐ 1	I don't feel sad today
☐ 2	I feel a little bit sad today
☐ 3	I feel a bit sad today
☐ 4	I feel quite sad today
☐ 5	I feel very sad today

3. Pain	
☐ 1	I don't have any pain today
☐ 2	I have a little bit of pain today
☐ 3	I have a bit of pain today
☐ 4	I have quite a lot of pain today
☐ 5	I have a lot of pain today

4. Tired	
☐ 1	I don't feel tired today
☐ 2	I feel a little bit tired today
☐ 3	I feel a bit tired today
☐ 4	I feel quite tired today
☐ 5	I feel very tired today

Please go to the next page

For each question, read all the choices and decide which one is most like you **today**. Then put a **tick** ☑ in the box next to it. Only tick **one** box for each question.

5.Annoyed	
☐ 1	I don't feel annoyed today
☐ 2	I feel a little bit annoyed today
☐ 3	I feel a bit annoyed today
☐ 4	I feel quite annoyed today
☐ 5	I feel very annoyed today

6. School work/homework (such as reading, writing, doing lessons)	
☐ 1	I have no problems with my schoolwork/homework today
☐ 2	I have a few problems with my schoolwork/homework today
☐ 3	I have some problems with my schoolwork/homework today
☐ 4	I have many problems with my schoolwork/homework today
☐ 5	I can't do my schoolwork/homework today

7. Sleep	
☐ 1	Last night I had no problems sleeping
☐ 2	Last night I had a few problems sleeping
☐ 3	Last night I had some problems sleeping
☐ 4	Last night I had many problems sleeping
☐ 5	Last night I couldn't sleep at all

8. Daily routine (things like eating, having a bath/shower, getting dressed)	
☐ 1	I have no problems with my daily routine today
☐ 2	I have a few problems with my daily routine today
☐ 3	I have some problems with my daily routine today
☐ 4	I have many problems with my daily routine today
☐ 5	I can't do my daily routine today

Please go to the next page

For each question, read all the choices and decide which one is most like you **today**. Then put a **tick** ☑ in the box next to it. Only tick **one** box for each question.

9. Able to join in activities (things like playing out with your friends, doing sports, joining in things)	
☐ 1	I can join in with any activities today
☐ 2	I can join in with most activities today
☐ 3	I can join in with some activities today
☐ 4	I can join in with a few activities today
☐ 5	I can join in with no activities today

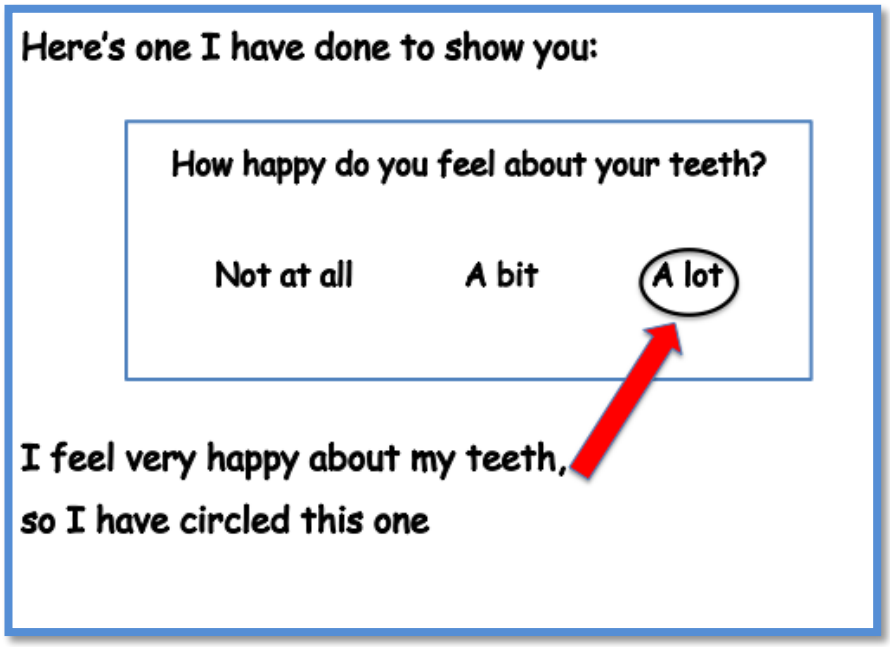
Thank you for answering those questions. We now want to find out a bit more about how you feel about your **teeth**.

Please go to the next page →

Section 3: How do you feel about your teeth?

These questions ask how you feel about your **teeth**. Read all the answers and see which one is most like you.

Please put a **circle** round the answer like this ⬭ . Only make **one** circle for each question.



Now please think about your teeth and answer the questions on the next pages.

Please go to the next page →

Please **circle one answer** for each question.

1. How much do your teeth hurt you?

Not at all A bit A lot

2. Do your teeth make it hard to eat some foods?

Not at all A bit A lot

3. Do you have to eat on one side of your mouth because of your teeth?

Not at all A bit A lot

4. Do you get food stuck in your teeth?

Not at all A bit A lot

5. How much do you get kept awake by your teeth?

Not at all A bit A lot

Please go to the next page →

Please **circle one answer** for each question.

6. How much do your teeth annoy you?

Not at all A bit A lot

7. How much do your teeth hurt when you brush them?

Not at all A bit A lot

8. Do you have to eat more carefully because of your teeth?

Not at all A bit A lot

9. Do you have to eat more slowly because of your teeth?

Not at all A bit A lot

10. Do you feel cross because of your teeth?

Not at all A bit A lot

Please go to the next page →

Please **circle one answer** for each question.

11. How much have you cried because of your teeth?

Not at all A bit A lot

12. Do your teeth make it hard to do your schoolwork?

Not at all A bit A lot

13. How much of a problem are your teeth for you?

Not at all A bit A lot

Thank you for answering all the questions!

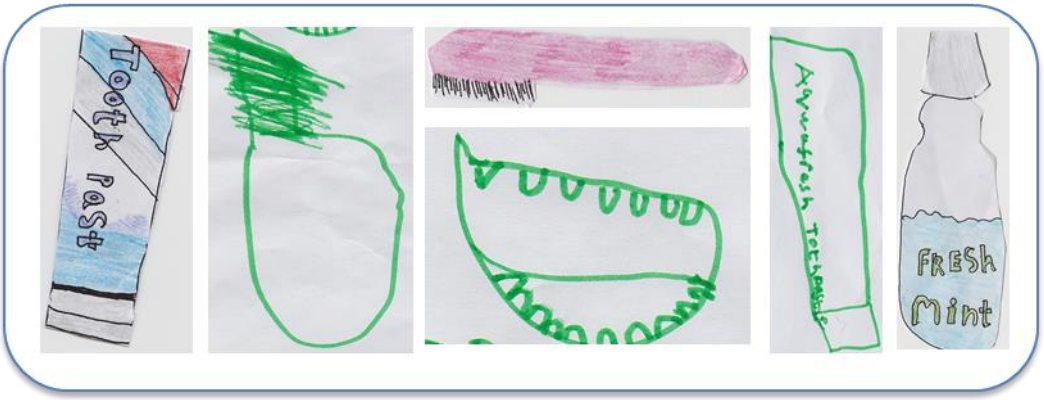
The child follow-up questionnaire has the following additional question:

14. Since the last time you answered these questions, do you think your teeth are:

Better The same Worse

Appendix 4: Parent/ caregiver consent and questionnaires

Questionnaire for parents/ guardians about how their child's teeth affect the family



Hello

Thank you for agreeing to help us with our study.

This study is being done so we can find out more about how young people feel about their teeth and work they have done at the dentist. We also want to know how children's teeth affect their family. By answering the questions, you will help us to find ways to make young people feel happier about visiting a dentist.

You child will be given questions to answer themselves. In this booklet, you will find some sets of questions about how your child's teeth have affected the family.

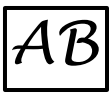
We would be very grateful if you could answer all the questions using the instructions.

There are no right or wrong answers. Some of the questions may seem to be asking the same thing but each question tells us about something slightly different that we would like to find out.

Please go to the next page →

1. Are you happy to take part?

It is up to you if you want you and your child to take part.

Before you start, please **initial** the boxes below **if you agree** with each statement like this: 

	Initial here
I am happy for the information given in this questionnaire to be used for research purposes	☐
I am happy for information about my child and their treatment to be collected anonymously to help with the research project only	☐
During the study, I am happy to be contacted by phone	☐
During the study, I am happy to be contacted by post	☐
I am happy for my child to be contacted in the future about similar research (you do not have to agree to this to take part in this study)	☐

Please write the date here: ☐

Thank you for agreeing for you and your child to take part.

Please go to the next page →

2. Information about yourself and your child's previous treatment

Please answer the following questions. For each question, read all the choices and put a **tick** ☑ in the box next to the correct answer. Some questions give you space to write in your own answer if none of the options are correct. If you are unsure about an answer, please tick 'Don't know'. Thank you.

1. What is your relationship to your child?

☐$_0$ Mum

☐$_1$ Dad

☐$_2$ Carer

☐$_3$ Other: please write your answer in the box:

2. Has your child had a general anaesthetic for dental treatment before? A general anaesthetic is given to a child in hospital so they are fully asleep for their dental treatment to be carried out.

☐$_0$ No

☐$_1$ Yes

☐$_{dk}$ Don't know

3. Have any of their brothers or sisters had a general anaesthetic for dental treatment before?

☐$_{na}$ This is an only child (has no brothers or sisters)

☐$_0$ No

☐$_1$ Yes

☐$_{dk}$ Don't know

Please go to the next page →

4. Before coming to the dental hospital, how many courses of antibiotics has your child had for their dental problem?

☐$_0$ None

☐$_1$ 1

☐$_2$ 2

☐ Other: please write your answer in the box:

Please go to the next page →

3. Instructions for the questionnaire

1. This questionnaire is about the effects of oral conditions on children's wellbeing and the effects on their families. There are **14** questions in total. **Please answer all the questions**.

2. To answer the question please put a tick ☑ **in the box by the answer you want to give**.

3. Please give the answer that **best describes your child's experience**. If the question does not apply to your child, please answer with "Never".

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

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

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

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

Please go to the next page →

4. Questions

The following questions ask about effects that a <u>child's oral condition</u> may have on <u>parents and other family members.</u>

During the <u>last 3 months</u>, because of your child's <u>teeth, lips, mouth or</u>

1. Been upset?

☐ 0 Never | ☐ 1 Once or twice | ☐ 2 Sometimes | ☐ 3 Often | ☐ 4 Everyday or almost everyday | ☐ dk Don't know

2. Had sleep disrupted?

☐ 0 Never | ☐ 1 Once or twice | ☐ 2 Sometimes | ☐ 3 Often | ☐ 4 Everyday or almost everyday | ☐ dk Don't know

3. Felt guilty?

☐ 0 Never | ☐ 1 Once or twice | ☐ 2 Sometimes | ☐ 3 Often | ☐ 4 Everyday or almost everyday | ☐ dk Don't know

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

☐ 0 Never | ☐ 1 Once or twice | ☐ 2 Sometimes | ☐ 3 Often | ☐ 4 Everyday or almost everyday | ☐ dk Don't know

During the last 3 months, because of your child's teeth, lips, mouth or

5. Had less time for yourself or the family?

☐ 0	☐ 1	☐ 2	☐ 3	☐ 4	☐ dk
Never	Once or twice	Sometimes	Often	Everyday or almost everyday	Don't know

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

☐ 0	☐ 1	☐ 2	☐ 3	☐ 4	☐ dk
Never	Once or twice	Sometimes	Often	Everyday or almost everyday	Don't know

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

☐ 0	☐ 1	☐ 2	☐ 3	☐ 4	☐ dk
Never	Once or twice	Sometimes	Often	Everyday or almost everyday	Don't know

During the last 3 months, because of his/her teeth, lips, mouth, or jaws, how

8. Been jealous of you or others in the family?

☐ 0	☐ 1	☐ 2	☐ 3	☐ 4	☐ dk
Never	Once or twice	Sometimes	Often	Everyday or almost everyday	Don't know

9. Blamed you or another person in the family?

☐ 0	☐ 1	☐ 2	☐ 3	☐ 4	☐ dk
Never	Once or twice	Sometimes	Often	Everyday or almost everyday	Don't know

10. Argued with you or others in the family?

☐ 0	☐ 1	☐ 2	☐ 3	☐ 4	☐ dk
Never	Once or twice	Sometimes	Often	Everyday or almost everyday	Don't know

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

☐ 0	☐ 1	☐ 2	☐ 3	☐ 4	☐ dk
Never	Once or twice	Sometimes	Often	Everyday or almost everyday	Don't know

During the last 3 months, how often has the condition of your child's teeth,

12. Interfered with family activities at home or elsewhere?

☐ 0	☐ 1	☐ 2	☐ 3	☐ 4	☐ dk
Never	Once or twice	Sometimes	Often	Everyday or almost everyday	Don't know

Please go to the next page →

During the last 3 months, because of his/her teeth, lips, mouth, or jaws, how

13. Caused disagreement or conflict in your family?

☐ 0	☐ 1	☐ 2	☐ 3	☐ 4	☐ dk
Never	Once or twice	Sometimes	Often	Everyday or almost everyday	Don't know

14. Caused financial difficulties for your family?

☐ 0	☐ 1	☐ 2	☐ 3	☐ 4	☐ dk
Never	Once or twice	Sometimes	Often	Everyday or almost everyday	Don't know

Thank you for answering all the questions!

The follow-up questionnaire also included the questions:

Since the treatment on your child's teeth:

15. Has your child's <u>dental health</u> been…?

☐ 0	☐ 1	☐ 2	☐ 3	☐ 4
Much improved	A little improved	The same	A little worse	Much worse

16. Has your child's <u>overall quality of life</u> been…?

☐ 0	☐ 1	☐ 2	☐ 3	☐ 4
Much improved	A little improved	The same	A little worse	Much worse

17. Has the change to your <u>family's life</u>…?

☐ 0	☐ 1	☐ 2	☐ 3	☐ 4
Much improved	A little improved	The same	A little worse	Much worse

Appendix 5: Letters sent to parents during the study

Letters sent to parents/carers during the study are given in this appendix. This includes the letter sent to parents 3-months following their child's GA, a follow-up letter sent if questionnaires were not received and the letter sent upon completion of the study.

School of Clinical Dentistry
Claremont Crescent
Sheffield
S10 2TA

Parent/Carer of XXXX
XXXXXXXXX
X
X

11/09/2019

Dear Parent/Carer,

This letter is being sent to you as your child is taking part in our study "Teeth and dentists- what do you think?" conducted by the University of Sheffield.

I have been informed that it has been 3 months since your child completed their dental treatment following their referral to Charles Clifford Dental Hospital. **We would like you and your child to complete the final questionnaires in this study.** Please find enclosed one questionnaire for yourself to complete (white cover), and one for your child to complete (purple cover). Also enclosed is a pre-paid envelope to return the questionnaires to us.

Once you have completed the questionnaires, please post them back to us using the pre-paid envelope enclosed. **You do not need a stamp.** Once we receive your questionnaires, your **£10 gift voucher will be sent to you by post as a thank you.**

If you have any questions, or no longer wish to take part, please contact me by email becky.knapp@sheffield.ac.uk or phone on 0114 2717877.

Many thanks for your help in this study.

Yours sincerely,

Becky Knapp
PhD Research Student

XXXX

School of Clinical Dentistry
Claremont Crescent
Sheffield
S10 2TA

Parent/Carer of XX
XX
XX
XX

11/09/2019

Dear Parent/Carer,

This letter is being sent to you as your child is taking part in our study "Teeth and dentists- what do you think?" conducted by the University of Sheffield.

We recently sent you a letter asking you and your child to complete the final questionnaires in this study. If you have already returned the questionnaires to us, we would like to thank you for your time and please ignore this letter.

If you have not yet returned your questionnaires, please can you do so as soon as possible. This information is really important to us, and as soon as we receive them we will be able to send you your **£10 gift voucher as a thank you for taking part.**

In case you have lost the originals, please find enclosed one questionnaire for yourself to complete (white cover), and one for your child to complete (purple cover). Also enclosed is a pre-paid envelope to return the questionnaires to us. **You do not need a stamp.**

Once we receive your questionnaires, your **£10 gift voucher will be sent to you by post as a thank you.**

If you have any questions, or no longer wish to take part, please contact me by email becky.knapp@sheffield.ac.uk or phone on 0114 2717877.

Many thanks for your help in this study.

Yours sincerely,

Becky Knapp
PhD Research Student

CREATE
Children's Oral Health Research

0XXX

School of Clinical Dentistry
Claremont Crescent
Sheffield
S10 2ZZ

Parent/Carer of XXX
XXX
XXX
XXX
XXX

11/09/2019

Dear Parent/Carer,

This letter is being sent to you as your child has been taking part in our study "Teeth and dentists- what do you think?" conducted by the University of Sheffield.

Please find enclosed their £10 gift voucher and certificate as a thank you. We are really grateful that you have completed our questionnaires as it is important for us to find out more about how children feel about their teeth and dentists so we can hopefully improve dental care for them.

Please sign and date the enclosed receipt and send it back to us in the pre-paid envelope (no stamp needed).

We will send you a final letter at the end of the study to tell you about what we have found out and what we think it means.

If you have any questions please contact me by email becky.knapp@sheffield.ac.uk or phone on 0114 2717877.

Many thanks for your help in this study.

Yours sincerely,

CREATE
Children's Oral Health Research

Becky Knapp
PhD Research Student

Appendix 6: Study amendment documentation

A list of amendments to the original IRAS application, letter to be sent to potential participants and subsequent ethical approval are included in this section.

IRAS form amendments

A-6-1: *After 'Parents and children who come to CCDH will be asked if they want to take part at their first appointment'* a line will be added: **"or they will be sent a letter inviting them to take part two weeks before their appointment"**

A13: After *'Recruitment of participants at new patient clinics'* a line will be added **"and by letter"**

A18: For reading the patient information sheet, after "Self-completed on clinic", a line will be added "**or at home**"

A30-1: We will add: "Alternatively, potential participants will be sent the information sheets, consent and questionnaires to their home, at least two weeks prior to their appointment. This will give them opportunity to consider all the information and their involvement. If they wish to take part, they will be asked to return the completed consent and questionnaires using the pre-paid envelope provided to them".

A31: Where participants have been contacted by post, they will have at least two weeks to consider their involvement.

The University Of Sheffield.

School of Clinical Dentistry
Claremont Crescent
Sheffield
S10 2ZZ

Parent/Carer of xx
Address xx
Xxx
Xxx

14/02/2018

Dear Parent/Carer,

This letter is being sent to you as your child is expected to have dental treatment carried out under general anaesthetic.

One of our PhD students, Becky Knapp, is doing a study called "Teeth and dentists- what do you think?", and we would like you and your child to take part. We would like to know how their dental problem affects them and your family. It is completely up to you if you take part, and it won't affect your child's treatment if you do not.

If you are interested in taking part, please read the enclosed information sheets, which give you the full details of the study. **If you are happy to take part, please complete the enclosed questionnaires and send them back to us using the pre-paid envelope.**

Please keep the information sheets for your records in case you need to contact us about the study.

We will send you the questionnaires again 3 months after their treatment is complete. Once you return these questionnaires to us, you will be sent your **£10 gift voucher** by post as a thank you.

If you have any questions, please contact Becky using the details on the enclosed information sheets.

Many thanks for your help in this study.

Yours sincerely,

CREATE
Children's Oral Health Research

Prof. Helen Rodd
Professor/Honorary Consultant in Paediatric Dentistry

Lothian NHS Board

Please note: This is an acknowledgement letter from the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

South East Scotland Research Ethics Committee 2
Waverley Gate
2 - 4 Waterloo Place
Edinburgh
EH1 3EG
Telephone: 0131 465 5674
Joyce.clearie@nhslothian.scot.nhs.uk
www.nhslothian.scot.nhs.uk

NHS Lothian

13 March 2018

Mrs Rebecca Knapp
Oral Health & Development PG Office,
School of Clinical Dentistry
Claremont Crescent
Sheffield
S10 2TA

Dear Mrs Knapp

Study title:	**The effect of dental treatment for the management of dental caries under general anaesthetic on the oral health-related quality of life of children**
REC reference:	**16/SS/0187**
Amendment number:	**16/SS/0187/AM02**
Amendment date:	**02 February 2018**
IRAS project ID:	**211730**

The above amendment was reviewed by the Sub-Committee in correspondence.

Ethical opinion

The members of the Committee taking part in the review gave a favourable ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

The Sub-Committee had no significant ethical concerns regarding the amendment.

Approved documents

The documents reviewed and approved at the meeting were:

Document	*Version*	*Date*
Letters of invitation to participant [Letter to Parents]	2	14 February 2018
Notice of Substantial Amendment (non-CTIMP)		02 February 2018
Other [IRAS form amendments]		07 February 2018
Research protocol or project proposal	4	07 February 2018

Headquarters
Waverley Gate
2-4 Waterloo Place
Edinburgh EH1 3EG

Chair Brian G. Houston
Chief Executive Tim Davison
Lothian NHS Board is the common name of Lothian Health Board

NHS
Lothian

Membership of the Committee

The members of the Committee who took part in the review are listed on the attached sheet.

Working with NHS Care Organisations

Sponsors should ensure that they notify the R&D office for the relevant NHS care organisation of this amendment in line with the terms detailed in the categorisation email issued by the lead nation for the study.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

We are pleased to welcome researchers and R & D staff at our Research Ethics Committee members' training days – see details at http://www.hra.nhs.uk/hra-training/

16/SS/0187:	**Please quote this number on all correspondence**

Yours sincerely

pp
Dr Yann Maidment
Chair

E-mail: joyce.clearie@nhslothian.scot.nhs.uk

Enclosures: *List of names and professions of members who took part in the review*

Copy to: *Mrs Samantha Walmsley*
Mrs Rebecca Knapp

NHS Lothian

South East Scotland REC 02

Sub-Committee of the REC meeting on 12 March 2018

Committee Members:

Name	*Profession*	*Present*	*Notes*
Dr Yann Maidment	General Dental Practitioner	Yes	In the Chair
Mr Lindsay Murray	Health & Safety Manager	Yes	
Professor Lindsay Sawyer	Professor Emeritus	Yes	

Also in attendance:

Name	*Position (or reason for attending)*
Ms Joyce Clearie	REC Manager
Miss Pavlina Yaneva	Ethics Administrative Officer

Appendix 7: Full path analysis results

Results from the full theoretical model path analysis. Non-significant paths (p<0.1) which were removed to create the parsimonious model are highlighted in grey.

	Satorra-Bentler Coef.	Std. Err.	z	P>|z|	[95% Conf.	Interval]
Structural						
CQCintT0						
Age	.431663	.2325622	1.86	0.063	-.0241505	.8874765
pain	4.509918	.8320085	5.42	0.000	2.879211	6.140625
cariousTeeth	.1108091	.1381044	0.80	0.422	-.1598707	.3814888
Ethnicity	1.976278	.9266764	2.13	0.033	.1600259	3.792531
IMDscore	.0030536	.0194407	0.16	0.875	-.0350496	.0411567
_cons	1.737182	1.920065	0.90	0.366	-2.026077	5.50044
C9DtotalT0						
CQCintT0	.2348676	.1411853	1.66	0.096	-.0418505	.5115856
Age	.0951981	.2869884	0.33	0.740	-.467289	.6576851
pain	.321541	1.034604	0.31	0.756	-1.706247	2.349328
cariousTeeth	-.1508513	.1462662	-1.03	0.302	-.4375279	.1358252
Ethnicity	1.111529	1.448018	0.77	0.443	-1.726534	3.949591
IMDscore	.0379584	.0311202	1.22	0.223	-.0230361	.0989528
_cons	9.969145	2.510785	3.97	0.000	5.048097	14.89019
CQCintT1						
CQCintT0	.1736013	.0984067	1.76	0.078	-.0192724	.3664749
C9DtotalT0	.0679016	.0818974	0.83	0.407	-.0926143	.2284176
totalXns	.6077897	.2257892	2.69	0.007	.165251	1.050328
Age	.4810821	.2910436	1.65	0.098	-.0893528	1.051517
pain	-.1363767	.8780589	-0.16	0.877	-1.85734	1.584587
cariousTeeth	-.1491446	.2263673	-0.66	0.510	-.5928163	.2945271
treatmentgroup	1.38791	.8536972	1.63	0.104	-.2853057	3.061126
Ethnicity	.1381487	.8329658	0.17	0.868	-1.494434	1.770732
IMDscore	-.01818	.0190258	-0.96	0.339	-.0554699	.0191099
_cons	-3.511833	2.287638	-1.54	0.125	-7.995521	.9718548
C9DtotalT1						
CQCintT0	.0138922	.0961337	0.14	0.885	-.1745265	.2023109
C9DtotalT0	.0849158	.0694026	1.22	0.221	-.0511108	.2209424
CQCintT1	.331688	.1325843	2.50	0.012	.0718276	.5915483
totalXns	-.1081396	.1685673	-0.64	0.521	-.4385255	.2222462
Age	.2201136	.2118242	1.04	0.299	-.1950543	.6352815
pain	-.4980761	.8295409	-0.60	0.548	-2.123946	1.127794
cariousTeeth	.0794416	.1584563	0.50	0.616	-.2311271	.3900102
treatmentgroup	-.2075236	.6453431	-0.32	0.748	-1.472373	1.057326
Ethnicity	.219038	.8636284	0.25	0.800	-1.473643	1.911718
_cons	7.394262	1.410628	5.24	0.000	4.629482	10.15904
totalXns						
cariousTeeth	.7560914	.0548516	13.78	0.000	.6485841	.8635986
treatmentgroup	-1.390447	.3875974	-3.59	0.000	-2.150124	-.6307698
IMDscore	.0246841	.0108557	2.27	0.023	.0034073	.045961
_cons	.7321945	.5513406	1.33	0.184	-.3484133	1.812802

Appendix 8: Total effects in the final model

	Satorra-Bentler Coef.	Std. Err.	z	P>|z|	[95% Conf.	Interval]
Structural						
CQCintT0						
Age	.415424	.2394146	1.74	0.083	-.0538199	.8846679
pain	4.570578	.837477	5.46	0.000	2.929153	6.212003
Ethnicity	2.120011	.9073658	2.34	0.019	.3416065	3.898415
_cons	2.606131	1.838638	1.42	0.156	-.9975335	6.209795
CQCintT1						
CQCintT0	.1814323	.084318	2.15	0.031	.0161721	.3466925
totalXns	.457273	.1136293	4.02	0.000	.2345636	.6799824
Age	.5386713	.2752891	1.96	0.050	-.0008855	1.078228
treatmentgroup	1.412208	.7728543	1.83	0.068	-.1025586	2.926975
_cons	-3.802909	2.099195	-1.81	0.070	-7.917255	.3114369
totalXns						
cariousTeeth	.7560914	.052698	14.35	0.000	.6528052	.8593775
treatmentgroup	-1.390447	.3834217	-3.63	0.000	-2.141939	-.6389539
IMDscore	.0246841	.0104764	2.36	0.018	.0041508	.0452175
_cons	.7321945	.5338581	1.37	0.170	-.3141482	1.778537
C9DtotalT0						
CQCintT0	.2771716	.1169707	2.37	0.018	.0479133	.5064299
_cons	11.09541	1.070421	10.37	0.000	8.997421	13.19339
C9DtotalT1						
CQCintT1	.3557227	.1141481	3.12	0.002	.1319965	.579449
_cons	9.491828	.4369791	21.72	0.000	8.635365	10.34829