Dental ExtraCtion versus filling of adult teeth In chilDren: an Economic evaluation (DECIDE)

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Abstract

Introduction

This thesis identifies the most efficient pathway for children with compromised first permanent molar (cFPM) adult teeth, comparing extraction versus restoration, including establishing and incorporating public willingness to pay (WTP). Adolescents' and adults' views and experiences of managing cFPM were qualitatively established. cFPM, most commonly due to dental caries and molar-incisor hypomineralisation, cause pain and impact children across England. Clear guidance on whether to restore or extract cFPM in children does not exist.

Methods

This thesis comprises three complementary studies. Online semi-structured interviews were undertaken with adolescents and adults. Transcripts were thematically analysed. A discrete choice experiment was designed and disseminated to 430 members of the public. Conditional (fixed effects) logistic regression established public preferences, and WTP. An individual patient-level microsimulation model was built, and parameterised, to compare the relative efficiency of initial cFPM strategies over the life course of a child.

Results

Several internal and external factors influence adolescents' and adults' management of cFPM. Any decision should be made in a shared-care approach, ensuring active involvement of adolescents. There is no clear public preference to restore or extract cFPM in a child, providing the resultant space was closed spontaneously or orthodontically. A preference exists for management by a general dental practitioner or a specialist. The public prefer decisions to be made by a parent, or in conjunction with the dentist. Base-case analysis, and scenario analyses, suggests the most efficient approach is to extract cFPM between the age of seven and ten. Definitively restoring is an efficient option but is less so than extraction. In the scenario modelled, active monitoring and temporary filling, followed by extraction at the optimum time, are dominated strategies and should not be offered.

Discussion

Clinical and policy implications of these findings, limitations of the methods used and recommendations for future research are discussed.

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Finally, I dedicate this thesis to Sienna.

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List of Abbreviations

ADHS - Adult Dental Health Survey

AIC - Akaike Information Criterion

ASC - Alternative Specific Constant

BIC - Bayesian Information Criterion

BWS – Best Worst Scaling

CARIES-QC - Caries Impacts and Experiences Questionnaire for Children

CARIES-QC-U - Caries Impacts and Experiences Questionnaire for Children (for Utility)

CBA – Cost-Beneifit Analysis

CCA - Cost-Consequence Analysis

CDHS – Child Dental Health Survey

CEA – Cost-Effectiveness Analysis

CEAC – Cost-Effectiveness Acceptability Curve

cFPM – Compromised First Permanent Molar

CHEERS - Consolidated Health Economic Evaluation Reporting Standards

CI – Chief Investigator

CMA – Cost-Minimisation Analysis

CPQ – Child Perceptions Questionnaire

COHIP - Child Oral Health Impact Profile

COREQ - Consolidated Criteria for Reporting Qualitative Research

CUA – Cost-Utility Analysis

C-OIDP - Child Oral Impacts on Daily Performances

CVM – Contingent Valuation Method

DAM – Decision Analytical Model

DCE – Discrete Choice Experiment

DI – Dental Implant

DMFT - Decayed, Missing and Filled Teeth

DMHDS - Dunedin Multidisciplinary Health and Development Study

DSA – Deterministic Sensitivity Analysis

EVPI - Expected Value of Perfect Information

EVPPI - Expected Value of Perfect Parameter Information

FB – Fixed Bridge

FPM – First Permanent Molar

GA – General Anaesthetic

GBP – Great British Pound

GDP – General Dental Practitioner

ICDAS - International Caries Detection and Assessment System

ICER – Incremental Cost-Effectiveness Ration

INMB – Incremental Net-Monetary Benefit

ISPOR - International Society for Pharmacoeconomics and Outcomes Research

MCN – Managed Clinical Network

MDAS – Modified Dental Anxiety Scale

MIH – Molar-Incisor Hypomineralisation

MNL - Multinomial logit

MRS – Marginal Rates of Substitution

NaF – Sodium Fluoride

NaOCl – Sodium Hypochlorite

NATCEN – National Centre for Social Research

NGT – Nominal Group Technique

NHS – National Health Service

NMB – Net-Monetary Benefit

NICE - National Institute for Health and Clinical Excellence

OHRQoL – Oral Health Related Quality of Life

OR – Odds Ratio

PIL – Patient Information Leaflet

PSA – Probabilistic Sensitivity Analysis

PUFA - Pulpal Involvement, Ulceration, Fistula or Abscess

QALY – Quality-Adjusted Life Years

QATY – Quality-Adjusted Tooth Years

RCT – Root Canal Treatment

RPD – Removable Partial Denture

RR – Relative Risk

SDF – Silver Diamine Fluoride

SES - Socioeconomic Status

SPM – Second Permanent Molar

TTO – Time-Trade Off

UDA – Unit of Dental Activity

UK – United Kingdom

UOA – Unit of Orthodontic Activity

VAS – Visual Analogue Scale

VOI – Volume of Information Analysis

WTP – Willingness to Pay

List of Appendices

Chapter 1. Compromised first permanent molars

1.1 Introduction

Teeth are subject to many internal and external factors that can lead to compromised longevity and can result in them requiring treatment to remediate this or to lead to their early loss. In this chapter, the issue of compromised first permanent (adult) molar teeth (cFPM) in children, including the uncertainties that exist about how best to manage these teeth, will be explored. Contemporary evidence will be outlined, and along with Chapter 2, Chapter 1 will detail how an economic evaluation approach can begin to address some of the management concerns, in particular the uncertainty of the future impact of early childhood decisions, in the context of the United Kingdom (UK) healthcare system. Section 1.2 will provide a brief overview of the problem of cFPM. Section 1.3 and 1.4 will summarise the aetiology, epidemiology, and inequalities of dental caries and molar incisor hypomineralisation (MIH) respectively. Section 1.5 will focus on how these two conditions relate specifically to first permanent molars. Section 1.6 will explain the direct and indirect impacts of compromised first permanent molars. Section 1.7 will detail how dental services in the UK are set-up to provide dental care for children. Finally, section 1.8 will provide an overview of management strategies available for cFPM, what is currently being done in the UK, and elsewhere, before discussing the confusion noted by patients, parents, and professionals on whether to fill or extract these teeth.

1.1 The problem - compromised first permanent molar teeth

First permanent molar (FPM) teeth begin to erupt around the age of six years, however, embryonic development begins around the twentieth week *in-utero* with hard tissue formation occurring some eight to twelve weeks later. Hard tissue formation continues beyond gestation with crown formation often completing around the age of two to three years old. These teeth can become compromised if subjected to unfavourable circumstances and can be referred to as cFPM. There are many causes of cFPM, however, dental caries (decay) and/or a developmental anomaly, known as MIH, in childhood are often regarded as the most common. The aetiology and epidemiology, with a focus on inequalities, relating to both dental caries and MIH in general will be discussed, before focussing specifically on cFPM.

1.2 Dental caries

1.2.1 Aetiology of dental caries

As shown in **Figure 1.1**, caries aetiology is multifactorial.

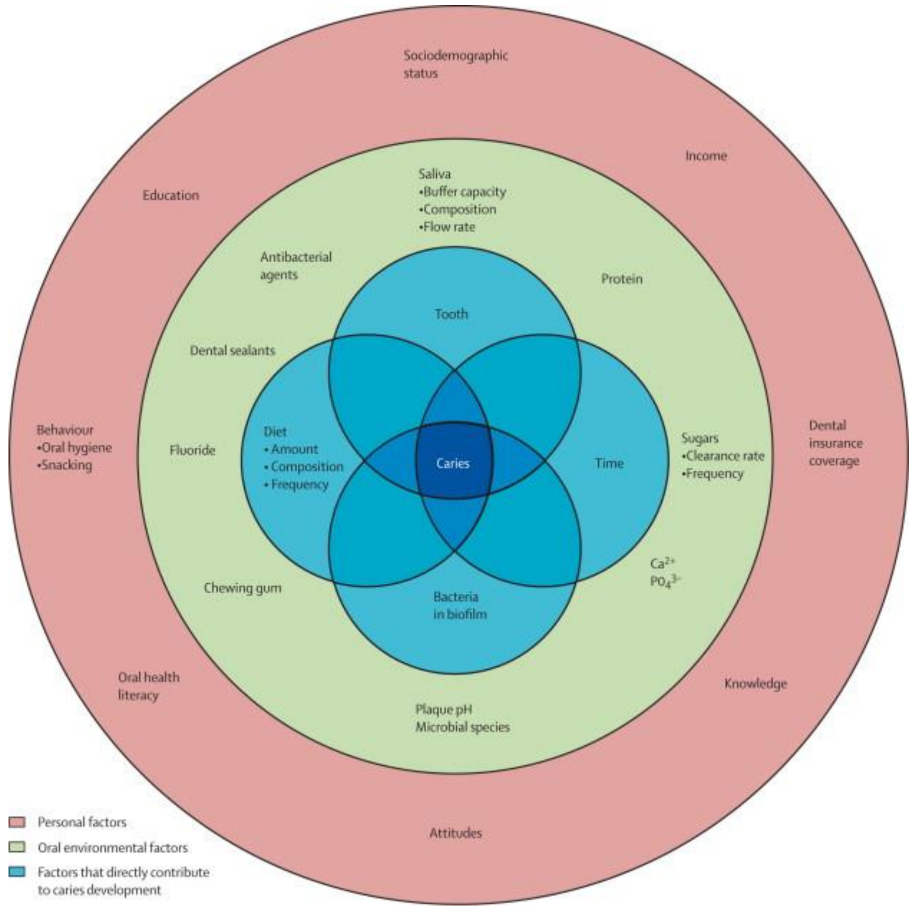


Figure 1.1 Diagrammatic representation of the complex interactions that are required for the initiation and progression of dental caries (reproduced from (Selwitz et al., 2007))

The dynamics of carious lesions are dependent on bacteria fermenting sugars, producing acids and dissolving tooth mineral (Kidd & Fejerskov, 2004; Featherstone, 2008). The microflora of the oral cavity is diverse with more than 700 bacterial species present. In addition to other microorganisms, such as fungi, viruses etc., these bacterial species form a biofilm. A biofilm is a dense aggregate of microorganisms. Oral biofilms adhere to all surfaces in the mouth in an organised fashion. In the case of dental caries, the biofilm that promotes mineral loss and leads to the disease is attached to a tooth surface (Kidd & Fejerskov, 2004).

Dental caries is therefore acknowledged as a biofilm-dependent disease but is driven by patient behaviours. Bacterial species, such as *Streptococcus mutan*s, in the presence of fermentable sugars produce (acidogenic) and tolerate (aciduric) high concentrations of lactic acid, and therefore obtain a selective ecological advantage over other species (Philip et al., 2018). The main component of teeth, a crystalline mineral structure known as hydroxyapatite, is readily dissolved by lactic acid forming a sub-surface non cavitated lesion at an ultrastructural (or sub-clinical) level. This loss of mineral from the tooth when the pH is dropping, is known as demineralisation. However, this demineralised lesion has the potential to remineralise and repair itself, or gain minerals, when the pH is increased due to the buffering capacity of the saliva (Featherstone, 2008).

In health, remineralisation/demineralisation processes are in homeostasis, with no net mineral loss and therefore no detriment to the tooth. However, if an ecological shift in the biofilm environment is observed, particularly in areas that are sheltered micro niches e.g., pits and fissures of molars, then the biofilm can remain undisturbed, mature over time, and generate acids and proteolytic enzymes. This results in the homeostatic imbalance of the plaque biofilm becoming acidic, with a net loss of mineral, which, if it continues, ultimately leads to the formation of a cavitated carious lesion (Philip et al., 2018).

Regular removal of the biofilm is necessary to prevent it maturing into an acidogenic environment and slow down or stop establishment or progression of carious lesions (Kidd & Fejerskov, 2004). Improving plaque control will disrupt and remove the biofilm and help reduce the risk of dental caries for that individual. Similarly, managing the diet to reduce the exposure to fermentable sugars will help reduce the risk (Albino & Tiwari, 2020).

It is well established that younger children rely on their parents to instil, support and maintain oral-health behaviours to reduce the risk of dental caries. Parental oral-health behaviours, and knowledge, influence a child's risk of developing dental caries (Agarwal et al., 2011; Hooley et al., 2012). Altering patient- and parent-related behaviours reduces the risk of dental caries. Whilst behaviour-related factors are the major influence on the development of dental caries, the aetiology extends beyond these factors.

It has been proposed that alterations in an individual's genetics could influence the development of dental caries (Shaffer et al., 2015). A single gene that directly regulates dental caries has yet to be identified (Shaffer et al., 2012); however, variations in the genetic expression of the AMELX gene have been suggested (Ferraro & Vieira, 2010). AMELX is particularly important during enamel development and therefore it could be argued that what Ferraro and Viera (2010) are suggesting, is differing composition of individual's enamel makes their teeth more susceptible to developing dental caries rather than the genes regulating the carious process. There is some suggestion instead, that epigenetic factors should be considered as a contributory factor in propensity to develop dental caries (Barros & Offenbacher, 2009; Fernando et al., 2015). Epigenetic changes occur when environmental influences, such as an individual's diet or exposure to certain pollutants in the surrounding areas, cause a change in the chemical compounds that regulate DNA activity (Egger et al., 2004). At present, the evidence-base to support genetic and epigenetic changes for the initiation and progression of dental caries remains weak (Shaffer et al., 2012).

1.2.2 Epidemiology of dental caries

Untreated dental caries, affecting permanent teeth, is the most prevalent overall health condition in the world (Bernabe et al., 2020) with a global prevalence of 29.4% (for all ages combined). In comparison to other oral health conditions, it eclipses the next most common condition, severe periodontitis, which has a global prevalence of 9.8% (Bernabe et al., 2020).

Epidemiologically, the two main indices used to assess dental caries detection and assessment are Decayed, Missing and Filled Teeth (DMFT) and the International Caries Detection and Assessment System (ICDAS). DMFT is a well-established oral health index used as a measure of caries severity. The numbers signify the summation of the total decayed (D), missing due to caries (M) and filled (F) teeth (T) for an individual or, presented as a mean of the total population (Broadbent & Thomson, 2005). Scores range from 0-28 (or 0-32 if you include third permeant molars) in the permanent dentition. In contrast, ICDAS allows detection at various threshold levels, ranging from non-cavitated early visual changes in enamel (Code 1) to extensive distinct cavity with visible dentin (Code 6). Only ICDAS codes 5 and 6 are used for dental caries that has extended into the dentine, causing distinct cavitation. To distinguish between the two, ICDAS code 5 involves obvious loss of tooth structure; whereas, ICDAS 6 relates to an extensive cavity that involves at least half of the tooth (Ismail et al., 2007).

Observing how dental caries changes over time, epidemiologically, can be established using sequential cross-sectional national surveys or cohort studies.   However, sequential cross-sectional national surveys are limited in that each sequential survey will examine a different cohort of children each time. Variations in the those sampled, in addition to changes in survey methodologies, will only give an estimation of how dental caries prevalence and/or experiences change over time (Broadbent et al., 2013). To overcome this, prospective longitudinal studies provide accurate representations of the rate of dental carious lesion development within individuals over time, as the same cohort are examined at sequential time points throughout their lives. Currently worldwide, only one long-standing prospective study, the Dunedin Multidisciplinary Health and Development Study (DMHDS), exists (Hong et al., 2020).

DMHDS has followed a birth cohort (n=1037) born in Dunedin (New Zealand) in 1972/1973 (Hong et al., 2020). Several other long-standing birth-cohort studies, which examine oral health at points, do exist (Pearce et al., 2009; Hallal et al., 2018), however, currently, there are no UK-based dental cohort studies that have examined the oral health of children from birth into adulthood. DMHDS is the only longitudinal study in the world to have clinically investigated dental health from birth to the 5th decade (Broadbent et al., 2013). Across this cohort, mean DMFT has been used to describe trends (rather than prevalence). The DMHDS study reports a mean DMFT (SD) of 1.0 (1.3) at age 9, increasing to 8.1 (5.0) at age 38 (Broadbent et al., 2013). Other birth cohorts exist elsewhere worldwide (Mongkolchati et al., 2010; Hallal et al., 2018), but currently have much shorter follow-up than the DMHDS.

In the UK, prospective birth cohorts exist (Raynor & Born in Bradford Collaborative Group, 2008; Pearce et al., 2009; Connelly & Platt, 2014) but are limited to the extent oral health data (in terms of measures and in terms of measurement points) is included. Instead, only a series of sequential cross-sectional studies exist for oral health. Despite not being as good as a prospective birth cohort study, their findings are still useful.

In the United Kingdom (UK), the most recent Children's Dental Health Survey (CDHS) (England, Wales, and Northern Ireland) in 2013 reported that 46% of 15-year-olds and 34% of 12-year-olds had obvious decay experience in permanent teeth. The term 'obvious decay experience' includes untreated dental caries into dentine (cavitated and visual dental caries into dentine), and dental caries that has previously been restored or has resulted in the tooth being extracted (Pitts et al., 2015). In Scotland, the 2019 National Dental Inspection Programme (NDIP) reported 20% of children, aged 11-12 years old, had obvious dental caries experience in their permanent teeth (Information Services Division, 2019). These reported figures clearly show that despite being a preventable condition, high levels of dental caries amongst UK children still exists.

Despite continuing high levels of dental caries, the UK prevalence in UK children has declined over the last fifty years. Obvious caries prevalence, in 15-year-olds, has reduced by 51 percentage points, from 97% to 46%, since the introduction of regular national CDHS survey in 1973 (Murray et al., 2015). Although recent evidence suggests this is happening quicker in Scotland, compared to the rest of the UK (Pitts et al., 2015; Information Services Division, 2019). This difference may be attributed to a national dental prevention programme in Scotland, implemented in 2008, known as Childsmile (Kidd et al., 2020).

It is possible to understand trajectories of dental caries for younger children, adolescents, and adults. Hall-Scullin *et al*., (2017) have shown that children in the UK, who started aged 7-9 years old, with dental caries in their primary teeth were more likely to have dental caries in their permanent teeth, compared to children who were caries-free at the beginning of the study, after seven years. Although seven years is relatively short follow up, there was a 35 per cent difference between these groups, illustrating the consequences of developing caries in early childhood.

Despite overall prevalence rates declining, dental caries remains a major problem for children in the UK. It appears from national surveys, and longitudinal cohort studies that dental caries prevalence is consistent as children move through adolescence and into adulthood (Broadbent et al., 2013). However, the rate of the increase in the number of carious lesions has been shown to slow down as age increases (Mejàre et al., 2004; Broadbent et al., 2013). A complicating factor in this interpretation is that the distribution of the presence/ absence of caries as a disease as well as the number of lesions per individual, amongst children in the UK is not equal. These inequalities will be explored in the next section.

1.2.3 Inequalities associated with dental caries

Oral health inequalities exist in children across the UK (Rouxel & Chandola, 2018). There are apparent differences, in dental caries, in relation to factors such as socioeconomic status, gender and ethnicity. These will now be explored, in turn, in more detail.

Socioeconomic status

There is an association between dental caries, in children, and socioeconomic status (SES). Schwendicke *et al*., (2014) reported in their meta-analysis that 83 of the 86 selected studies including primary and permanent teeth in children and adults had at least one measure of dental caries that was higher amongst individuals with low SES, thus suggesting a higher risk of developing dental caries. The parameter with the strongest association of an increased risk of developing dental caries children was parents with lower levels of reported parental education (Schwendicke et al., 2014). Other studies have reported similar findings (Li et al., 2011; Stormon et al., 2020).

In 2013, the UK CDHS reported obvious dental caries experience was greater amongst children who were from lower income families, using free school meals eligibility as a marker of deprivation (which as a binary marker, has limitations (Taylor, 2018)), compared to other children of the same age. At age 15, 26% of patients who were eligible for free school meals had severe or extensive tooth dental caries, compared to 12% who were not (Pitts et al., 2015).

Despite a decline in dental caries prevalence, an increase in socioeconomic inequalities in oral health in children has been reported. The change has not been evenly distributed, with the prevalence of dental caries having remained unchanged for those children living in the most deprived areas, despite sustained equality in dental attendance (Ravaghi et al., 2020).

Gender

When dental caries rates are reported by gender in the literature, females are typically found to exhibit higher prevalence rates than males across all age categories (Lukacs & Largaespada, 2006; Ferraro & Vieira, 2010; Shaffer et al., 2015). The most recent CDHS highlighted that 15-year old girls were more likely to experience obvious dental caries, compared to boys, although this difference was not apparent at age 12 (Pitts et al., 2015). Shaffer *et al*., (2015) summarised possible explanations for gender differences, such as earlier tooth eruption, difference in dietary behaviours, hormonal and/or physiological changes and altered phenotypical changes to tooth enamel and salivary composition with, Ferraro and Viera (2010) suggesting genetic variations. However, there remains little evidence to support why females have higher caries rates than males.

Ethnicity

There are high levels of caries experience amongst UK school children from Pakistani, Bangladeshi, Chinese and East European backgrounds, even after controlling for levels of socioeconomic deprivation. Ethnic variations in dental caries rates are more profound in younger children. However, by age 15, differences had reduced considerably for each ethnic group and were comparable to White British/Irish children (Anderson et al., 2015).

Cultural variations, relating to self-care oral health practices, and peer-to-peer interaction within the school environment contributing to social equalisation amongst different ethnic groups have been postulated as explanations for these results. However, the link between dental caries and ethnicity is complex, and is unlikely to be explained at an individual level only (Hooley et al., 2012; Rouxel & Chandola, 2018).

However, it could be that for these groups, other attributes such as poverty, low income etc. may intersect with ethnicity. This intersectionality of inequalities, rather than just cultural variations in oral-health related practices, are likely to cumulatively contribute to oral health issues (Freeman et al., 2020).

In summary, inequalities in dental caries clearly remain an issue for children within the UK. In addition to these, dental caries is likely to place a significant burden on the child and the wider family. This will now be explored in the following sub-section.

1.2.4 Impact and burden associated with dental caries

In this context, impact refers to both positive and negative changes that are attributable to a particular dental condition. Pain is most common associated feature with dental diseases (including cFPM) (Rodd et al., 2007; Selwitz et al., 2007; Gilchrist et al., 2015; Tsakos et al., 2015; Taylor et al., 2018), and is likely to impact individuals, by causing sleep loss, interfering with eating/drinking or limiting participation in regular daily activities (Sheiham, 2006; Bernabé et al., 2007; Taylor et al., 2018).

These changes to an individual's quality of life, and oral health related quality of life (OHRQoL). Changes can be identified and measured through child specific oral health related quality of life (OHRQoL) tools.

Oral Health Related Quality of Life (OHRQoL)

OHRQoL is as a multidimensional construct describing how oral and dental disease disrupts an individual's normal ability to function (Baker, 2007). Understanding the psycho-social consequences of oral conditions, in particular how oral health affects aspects of social life, including self-esteem, social interaction, school and job performance, is regarded as important given it is the direct clinical consequences of a particular disease (Allen, 2003).
Locker (1988) developed a conceptual model of oral health to help explain how oral diseases and conditions affect a person's quality of life. This conceptual framework includes five domains - impairment, functional limitation, pain/discomfort, disability, and handicap – and these are sequentially related (Baker, 2007). The model is supported by empirical evidence, although, OHRQoL in reality is likely to be more complex than originally described as factors relating to the individual and environmental were not included (Baker, 2007).

Several validated paediatric measures to assess OHRQoL are available, for example, the Child Perceptions Questionnaire (CPQ), the Child Oral Impacts on Daily Performances (C-OIDP), the Child Oral Health Impact Profile (COHIP) (Genderson et al., 2013).

The 2013 Child Dental Health Survey used the C-OIDP tool to elicit the extent to which specific oral conditions negatively affected the daily lives of children. From the CDHS, it was reported that two thirds of 12- and 15-year-olds (68% and 66% respectively) self-reported experiencing at least one problem with their dental health over the previous three months, which affected their quality of life. Sensitivity was reported by 32% and 34% of 12- and 15-year-olds respectively whilst toothache, which can be associated with MIH and/or dental caries, was reported at 18% and 15% by both groups with a higher prevalence of up to 30% in younger children from deprived area. Interestingly, the likelihood of reporting did not differ by sex or deprivation status, however, more girls self-reported having experienced toothache compared to boys at both ages, which could be related to differences in dental caries between boys and girls (Tsakos et al., 2015). However, these results are for all 12- and 15-year-old children in the UK, and not just those FPM affected by dental caries. To date, there is only one paediatric condition-specific measure to assess OHRQoL. The Caries Impacts and Experiences Questionnaire for Children (CARIES-QC) is a valid and reliable child-centred caries-specific quality of life measure in children with dental caries (Gilchrist et al., 2018).

In addition to quality of life, and OHRQoL, further impacts can be noted. Rebelo *et al*., (2018) reported that children and adolescents with dental caries, especially those reporting worse oral health experience, had poorer school performance and attendance than caries-free children. Poor performance and absenteeism can lead to missed opportunities for learning and academic advancement amongst these children which may lead to future economic deprivation and social, occupational, and marital problems in adulthood (Hibbett et al., 1990; Hibbett & Fogelamn, 1990)

Untreated dental caries in children has been shown to impact the growth of a child. Sheiham (2006) reported that pre-school children with caries often weighed less, when compared to unaffected children, and after receiving dental treatment would show an increase in growth. Alkarimi *et al*., (2012) agreed with these findings by showing children with untreated caries had significantly poorer appetites compared to treated children. It was suggested that pain attributed to caries impeded both the quantity and variety of food consumed (Sheiham, 2006; Alkarimi et al., 2012). Another suggestion was that chronic inflammation, associated with chronic pulpitis and dental abscesses, suppresses growth as a result of depressed erythrocyte production (Sheiham, 2006).

Indirectly, dental caries will have an impact on the parents and families of affected children. Abed *et al*., (2019) reported that parents of children with severe dental caries, as reported in the 2013 CDHS, felt guilty, felt stressed , had their normal activities, and sleep disrupted. In addition, a significant negative impact on the wider family was evident, as those children with severe dental caries often needed more attention compared to their siblings (Abed et al., 2019). In addition, parents of children with severe dental caries, had greater chance of taking time off work (Abed et al., 2019). It has been indicated that approximately 160 million work hours a year are lost due to oral disorders (Reisine, 1984; Gift et al., 1992).

Section 1.3 has shown that dental caries is a significant problem for children across the UK. Despite being a preventable condition, it remains prevalent, and affects different groups in different ways, thus driving oral health inequalities. As a disease, it has a direct impact on the child and their wider family. The next section will focus on MIH, the other most common reason for permanent teeth to be compromised in children.

1.3 Molar-incisor hypomineralisation (MIH)

1.3.1 Aetiology of molar-incisor hypomineralisation

Molar-incisor hypomineralisation (MIH) was initially described by Weerheijm *et al*., (2001) as a qualitative defect of 1-4 first permanent molars with or without the maxillary and mandibular permanent incisors. MIH was then further defined as a developmental, qualitative enamel defect caused by a reduction in the mineralisation of the enamel (Weerheijm et al., 2003).

The exact aetiology is unknown, but a recent meta-analysis reported that peri- and post-natal aetiological factors were associated with increased chance of developing MIH, whereas pre-natal factors were not. Overall, the quality of included studies varied; however, those at critical risk of bias were excluded prior to the meta-analysis (Garot et al., 2021).

Children who suffered from hypoxia were found to be at increased odds of developing MIH. This was true also of a caesarean- and premature-delivery as they are both hypoxia-inducing events (Garot et al., 2021). These results corroborate findings of earlier studies (Lygidakis et al., 2008; Alaluusua, 2010; Silva et al., 2016), suggesting hypoxia is a key factor. In addition, hypoxic exposures in the post-natal phase similarly appear to be significant in the development of MIH; however, the extent remains unknown (Garot et al., 2021).

In this recent review, several post-natal conditions that commonly affect children, such as measles, otitis media and bronchitis amongst others were reported to be associated with increased odds of developing MIH (Garot et al., 2021). The reported aetiological factors are consistent with previous reviews (Alaluusua, 2010; Silva et al., 2016), however, the exact causative mechanisms of MIH have yet to be established (Crombie et al., 2009). Several studies comment on the difficulties in explaining the pathological/physiological mechanisms that lead to the development of MIH (Alaluusua, 2010; Silva et al., 2016; Garot et al., 2021). Ascertaining whether MIH is caused directly by the pathological process of the disease, its associated symptoms (e.g., fever) or because of how these were managed (e.g., antibiotic use) is challenging.

Existing research would suggest body temperature interferes with enamel development. One *in-vitro* study reported that rats placed in a febrile state for 57 hours exhibited disruption to the enamel formation of their teeth when compared to controls (Tung et al., 2006). A later study demonstrated that the expression of genes responsible for enamel development were altered when the temperature of cultured mouse molars was increased by 2ºC, from 37ºC to 39ºC (Ryynänen et al., 2014). These *in-vitro* studies did suggest that fever, associated with post-natal diseases, is highly like to cause disruption to the amelogenesis process, leading to enamel defects. Similarly, several *in-vitro* studies with rats and piglets suggest that antibiotic use might have a role to play in enamel development (Laisi et al., 2009; Kuscu et al., 2013; Kameli et al., 2019). However, the dose and regime of antibiotics used in these studies are not comparable to those used in children. Thus, they may have demonstrated a possible mechanism, but the relevance is unclear.

Recent evidence suggests that genetics and/or epigenetics may have a role in MIH. Teixeira *et al*., (2017) reported a greater concordance amongst monozygotic twins in the diagnosis of MIH. In isolation, this could suggest a strong genetic influence; however, recent evidence supports the idea that epigenetic changes in the genome, due to the influence of environmental factors, is more likely to be the case in MIH (Vieira & Manton, 2019).

In general, therefore, it seems that the development of MIH can be attributed to peri- and post-natal factors. As explained earlier, there appears to be no single aetiology for the manifestation of a disruption in tooth formation. Information elicitation in recent studies has been based on relying on recall from the mothers, thus introducing a recall bias (Alaluusua, 2010; Garot et al., 2021; Silva et al., 2016). In future studies, objectively identifying medical diagnoses, problems, hospital admissions and/or medication use, supplemented by parental recall, is likely to be more accurate (Garot et al., 2021).

1.3.2 Epidemiology of molar-incisor hypomineralisation

MIH prevalence rates vary widely with a range of 2.4% - 40.2% reported (Lygidakis et al., 2022). Despite clear assessment criteria, until recently, there was no specific index used to assess and grade MIH, which could explain the wide range reported across these studies. Furthermore, a definitive diagnosis of MIH, particularly in young children, can be challenging as permanent teeth are still erupting, and the lesions observed in MIH can be misinterpreted as dental caries or other enamel defect disorders e.g., amelogenesis imperfecta, dental fluorosis. An MIH training manual, for use in epidemiological studies and clinical assessment, was developed (Ghanim et al., 2017) and validated (Ghanim et al., 2019) for use to allow meaningful comparisons between future studies. A recent global meta-analysis, based on 99 observational studies (including 113,144 participants from 43 countries) reported a mean global MIH prevalence of 12.9% (11.7–14.3%) (Schwendicke et al., 2018, 2019). To date, there is only one UK MIH prevalence study, reporting a prevalence of 15.9% (Balmer et al., 2012).

There are no sequential national surveys that demonstrate how the prevalence of MIH varies across time. Despite being a developmental condition, understanding trends in large epidemiological surveys could be helpful, especially if supplementary information relating to aetiological factors was obtained from the individual, their parents, or ideally external healthcare data sources.

1.3.3 Inequalities associated with molar-incisor hypomineralisation

Like dental caries, differences in oral health inequalities in children such as sociodemographic status, gender and ethnicity exist. These will now be explored, in turn, in more detail.

Socioeconomic Status

There is some evidence to suggest a link between MIH and SES in the UK. Balmer et al., (2012) reported that children from a higher SES were more likely to have MIH. It is somewhat surprising that high SES was linked to MIH, as it would be expected that the commonly suggested aetiologies for MIH are noted more so in children from a low SES. However, the authors conclude that a confounding factor is likely to explain this, as children in their sample could have been born from mothers of a higher maternal age (which is known to be associated with a higher SES) and as such increases the risk of pre-, and peri-natal issues that are known to increase the risk of MIH. This concept that higher SES and MIH are linked was corroborated by a Finnish study (Wuollet et al., 2014) and a later Brazilian study (Tourino et al., 2016).

Gender

Balmer *et al*., (2012) reported there were no gender differences in children with MIH in their UK-based study. Some studies have shown a slightly higher prevalence in males (Preusser et al., 2007; Garcia-Margarit et al., 2014), whilst others have shown a higher prevalence in females (Lygidakis et al., 2008; Zawaideh et al., 2011). A recent meta-analysis by Zhao *et al*., (2018) concluded that no sex predilection can be established in MIH. Thus, the evidence is equivocal as to whether differences in MIH exist based on gender.

Ethnicity

There is no UK-based study, or meta-analysis which demonstrates whether there is an increased prevalence of MIH amongst different ethnicities. Some studies have reported no difference amongst various ethnic groups within the same country (Mahoney et al., 2006; Zawaideh et al., 2011).

It appears that some inequalities exist in relation to SES and MIH, however there is insufficient evidence to show whether gender and ethnicity are associated with the development of MIH.

1.3.4 Impact and burden associated with molar-incisor hypomineralisation

The impact and burden for teeth associated with MIH are similar to those described for dental caries, in section 1.3.4. However, some nuances do exist.

The 2013 Child Dental Health Survey noted that sensitivity was reported by 32% and 34% of 12- and 15-year-olds respectively (Tsakos et al., 2015). This condition is particularly common in MIH, whilst toothache, reported at 18% and 15% at both ages, can be associated with both MIH and/or dental caries (Tsakos et al., 2015). However, these results are for all 12- and 15-year-old children in the UK, and not just those FPM affected by MIH. Regarding sensivity, hypomineralised enamel is a poor insulator and is less able to protect the pulp from external thermal stimuli (Rodd et al., 2007). Over time, this leads to sub-clinical inflammatory and pH changes in pulpal tissues, which makes the pulp hypersensitive (Rodd et al., 2007).

Individuals' with MIH are likely to have an altered OHRQoL (Portella et al., 2019; Elhennawy et al., 2022; Joshi et al., 2022; Jälevik et al., 2022). However, these studies have used generic measures that identify OHRQoL. As previously discussed, CARIES-QC can be used to elicit quality of life in children with dental caries (Gilchrist et al., 2018). No studies have specifically addressed the OHRQoL of children with MIH as no MIH-condition-specific measure exists.

Sections 1.3 and 1.4 have shown that dental caries and MIH compromise permanent teeth in children across the UK. However, the disease burden in permanent teeth in children is uneven in its distribution, with the FPM teeth often being far more compromised than any others. This next section will detail the aetiological reasons for increased susceptibility of this tooth, as well as the epidemiology of conditions of FPM teeth in children.

1.4 Compromised first permanent molar teeth in children due to dental caries and MIH

In this chapter so far, dental caries and MIH have been discussed. These are the most common aetiologies that would render a FPM compromised, although other less common aetiologies e.g., amelogenesis imperfecta, do exist. As a result, for the purposes of the empirical work in thesis, a cFPM was defined as a restorable first permanent molar that has either distinct cavitation (ICDAS Codes 5 & 6) due to dental caries, or post-eruptive breakdown due to MIH, with an absence of any signs or symptoms of loss of pulp vitality (toothache). This section will firstly focus on the increased susceptibility, to dental caries and MIH, of the FPM that renders it compromised. This will be followed by the epidemiology of the cFPM.

1.4.1 Increased susceptibility of the first permanent molar teeth

Caries

FPM teeth are often compromised due to dental caries as shown in **Figure 1.2**. The notion of increased susceptibility for FPM teeth due to caries has been derived from cross-sectional (Macek et al., 2003; Batchelor & Sheiham, 2004; Pitts et al., 2015), short-term (Chestnutt et al., 1996) and long-term observational studies (Broadbent *et al*. 2013). In fact, the FPM tooth retains its status as the most susceptible tooth to dental caries across the life-course (Broadbent et al., 2013). Morphological variations of these teeth, compared to other permanent teeth, are likely to contribute to an increased susceptibility (Macek et al., 2003; Batchelor & Sheiham, 2004). An alternative explanation could be that these teeth are the first permanent teeth to erupt, and therefore spend the longest time in the mouth. Although anecdotal, the additional years, in comparison to adjacent permanent teeth, of being exposed to mediators of the biofilm that cause dental caries could increase the susceptibility of these teeth to dental caries.

Figure 1.2 FPM tooth compromised by dental caries in child aged 8

MIH

FPM teeth are often compromised due to MIH, as shown in Figure 1.3. The susceptibility as a result of MIH is due to an inherent lack of structural integrity (Weerheijm et al., 2001; Balmer et al., 2012) which is compounded by normal biting forces causing further mechanical destruction, known as post-eruptive breakdown (see Figure 1.4). This affects the macroscopic appearance of these teeth which: a) makes them harder to treat; and b) increases their susceptibility to dental caries due to greater plaque accumulation (Americano et al., 2017) and bacterial penetration (Rodd & Boissonade, 2006).

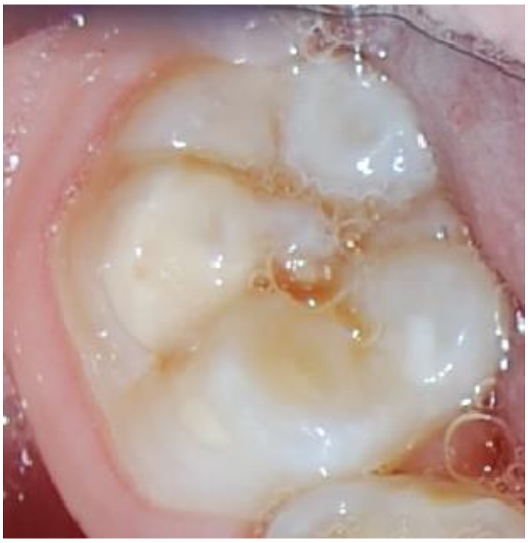


Figure 1.3: FPM tooth compromised by Molar-incisor hypomineralisation (MIH) in child aged 7

Figure 1.4 FPM tooth compromised by MIH and showing post-eruptive breakdown in a child aged 9

1.4.2 Epidemiology of the compromised first permanent molar teeth

Caries

By age 15 across England, Wales and Northern Ireland, 25% of FPM teeth will have obvious dental caries experience (Pitts et al., 2015). By comparison, the next most prevalent permanent tooth to be affected is the second permanent molar at 9% (Pitts et al., 2015). A study including 20,052 US children, regardless of age, ethnicity or water fluoridation, concluded the most susceptible tooth surfaces to develop dental caries were the buccal pits of the lower FPM tooth, and occlusal fissures of maxillary (upper) and mandibular (lower) FPM teeth (Batchelor & Sheiham, 2004).

These findings corroborate an earlier study which showed that in 4,294 Scottish adolescent teeth, the tooth surface most susceptible to dental caries was the occlusal surfaces of FPM teeth. It was noted that just over one third of these teeth, which were clinically sound at baseline, had evidence of dental caries three years later. The next most susceptible surfaces were the buccal and palatal pits of the FPM teeth, whereby 8.8% of these developed caries over the three year period (Chestnutt et al., 1996). Although relevant, it is worth noting that the study was conducted over 25 years ago and in a single Scottish Health Board; which is known to have had greater than average levels of dental caries when compared to the national average (Information Services Division, 2019).

MIH

A recent meta-analysis reported that in 135,181 children, across 116 included observational studies, the estimated cases that involve one affected MIH-molar was 24.3%, with 26.7%, 18.1% and 26.8% reported for two, three and four respectively (Lopes et al., 2021). However, due to factors discussed earlier in Section 1.4.2, the heterogeneity amongst the studies was significant. In addition to the number of affected molars, Weerhijm (2004) reported that the extent and severity of the defects vary from molar to molar within individual patients.

Section 1.5 has shown that FPM teeth in children are susceptible to being compromised by dental caries and MIH. Given the scale of this problem, it is important to appreciate the impact associated with these teeth. The next section will address some of the direct and indirect impacts associated with cFPM.

1.5 Impact of compromised first permanent molar teeth

As discussed in sections 1.3.4 and 1.4.4, dental caries and MIH impact children and their families. The following sections will focus on the direct and indirect impacts, due to dental caries and MIH, specifically for cFPM.

1.5.1 Direct impacts associated with compromised first permanent molar teeth

Direct impacts associated with cFPM are expected to have an effect throughout life, and not just in childhood (Selwitz et al., 2007). Unlike certain chronic medical conditions, where the impacts can be cumulative and constant throughout life (Sampogna, 2013), impacts associated with cFPM are more likely to occur intermittently. Intermittent impacts are quite common with dental conditions as often most impacts are acute and short-term (Tsakos et al., 2015). The frequency however will vary greatly.

A recent systematic review, investigating the relationship between MIH and OHRQoL, reported potential for MIH to cause a negative impact on OHRQoL in children, but this was not consistent across all included studies. Where there was a significant relationship, notable variations were observed in terms of which domains of the CPQ (oral symptoms, functional limitations, emotional well-being, and social well-being) contributed to the negative OHRQoL. Two studies showed a statistically significant impact of MIH across all domains, whilst in one study, it was only the oral symptoms domain. The remaining four studies showed no statistically significant impact on any domain. Interestingly, one study showed statistically significant results in all domains, irrespective of severity, whereas two others only demonstrated this in the functional limitations and/or oral symptoms domain (Jälevik et al., 2022). It does appear that OHRQoL is negatively affected in some children. Unfortunately, the review did not report the teeth affected by MIH, therefore definitive conclusions for cFPM are unable to be drawn from this.

In an attempt to establish direct impacts associated with cFPM in children, Taylor *et al*., (2018) undertook a small pilot study for those attending a specialist paediatric dental department in the north east of England. As shown in **Table 1.1**, the four most commonly reported impacts (pain, eating, sleep loss and daily activities) were chosen. Questions to address these impacts were taken from C-OHIP (Broder & Wilson-Genderson, 2007) questionnaire and amended, with children and adolescents, to be more age-appropriate. Additional amendments were made to the response scale (Likert scale 1-6 and 1-10 for 6-9- and 9–12-year-olds respectively; simple yes/no for 13–16-year-olds) as this was felt to better reflect how children in these age ranges would prefer to mark the impact. Sixty children (20 children in each age group: 6-9 years old; 9-12 years old; 13-16 years old) with affected cFPM only (dental caries n=35; MIH n=25) completed the relevant age-appropriate questionnaire. It was apparent that pain, directly associated with cFPM had the most impact on children across all age groups, with eating difficulties, sleep loss and disruption of daily activities being evident, but to a lesser degree (See **Table 1.1**) (Taylor et al., 2018). However, only 20 children in each age group were included, and the sample was local to the northeast of England.

To draw some stronger conclusions, a larger UK wide sample with stricter inclusion criteria would be of benefit

Age	Median impact for 6-9 years old (n=20)	Median impact for 9-12 years old (n=20)	Impact for 13-16 years old (n=20)
Scale	*(1=no impact. 6=most impact)*	*(1= no impact; 10=most impact)*	*(% who reported this over last 3 months)*
Pain	4	8	85%
Eating	4	8	75%
Sleep Loss	3	7	60%
Daily Activities	4	8	70%

Table 1.1: Impact associated with children with compromised first permanent molars (reproduced from (Taylor et al., 2018))

In addition to pain and its impact on quality of life, infection and subsequent sepsis is not uncommon for cFPM (Leal et al., 2012; Grund et al., 2015). Odontogenic infection, as a consequence of untreated dental caries, has been reported to be one of the most frequent reasons for the hospitalisation of young children in a German study (Grund et al., 2015). Parten *et al*., (2019) corroborated this by reporting that odontogenic infections was amongst one of the most common reasons for attendance, for children aged 6 – 16, to one UK medical emergency department.

Specifically for cFPM with MIH, Schwendicke (2018) reported that 27.4% (95% CI: 23.5% - 31.7%) of patients with MIH were, or will be in need of, treatment due to pain, sensitivity and post-eruptive breakdown. Compared to unaffected control children, those with MIH are approximately 10 times more likely to have undergone dental treatment on their first permanent molar, with each affected tooth likely to have been treated on average twice (Schwendicke et al., 2018)

Although there is evidence that dental disease directly affects a child's OHRQoL, the evidence is more limited for children with cFPM. However, indirect impacts with cFPM should not be overlooked.

1.5.2 Indirect impacts associated with compromised first permanent molar teeth

cFPM will indirectly impact potential economic growth as loss of productivity, due to depletion in labour, capital and other production costs (Listl et al., 2015) by parents of children with cFPM is likely. Furthermore, the economic burden on the wider society from cFPM is apparent. Dental caries remains the most common cause for admission to hospital for children aged 5-9 years old, which is more than double the second most common condition, acute tonsillitis (NHS Digital, 2020a).

Sections 1.2 – 1.6 have demonstrated how significant a problem compromised first permanent molars are for children across the UK. Given the prevalence of cFPM is likely around 1 in 4 children, the stark impact, and the cost to society is not insignificant, it is essential to understand how to manage these teeth effectively to maximise resources and patient benefit.

Section 1.7 will detail how dental services are set-up to provide dental care for children in the UK, whilst Section 1.8 will provide an overview of management strategies, and the confusion surrounding which strategies are best chosen to manage cFPM.

1.6 Dental services in the UK

1.6.1 Provision of dental services in the UK

The National Health Service (NHS) in England currently spends around £3.7billion per year or 3.5% of the NHS budget on dental services (NHS Commissioning Board, 2013). Fundamental changes to the provision of dental services in England and Wales were made in 2006. In dissolving the universal national contract and introducing locally commissioned primary dental services, the intention was to eliminate the fee-for-item system (remuneration based on the quantity of treatment) whilst improving preventive practices and patient access. Under these new arrangements dental contracts were set up by local commissioners, under the auspices of NHS England, for dental care providers at a local level. In addition to this major change in commissioning of care, a more simplified system of how dental care was to be remunerated was also introduced (Chestnutt et al., 2009; NHS Commissioning Board, 2013).

Primary care contracts are currently based on a standardised national contract of remuneration, which specifies a level of activity, measured in so-called Units of Dental Activity (UDA), at a negotiated price. Three different bands of treatment generate either one, three or 12 UDAs with more complex courses of dental treatment generating more UDAs (Vernazza et al., 2019). The monetary value of each UDA varies by practice, however, an average value has been estimated at around £25 Great British Pounds per UDA (British Dental Association, 2020). The patient (where they are not exempt) pay £23.80, £65.20, and £282.20 towards a course of treatment that will accrue one, three and 12 UDAs respectively. The system however been highly criticised as it bases remuneration purely on activity (Chestnutt et al., 2009), and includes problems such as lack of incentivisation for prevention, perverse incentives to undertake certain treatments and not undertake high quality dentistry (Steele et al., 2009; Health Select Committee, 2009). Recent changes to the dental contract indicate that as of 1st October 2022, five different bands of treatment generate either one, three, five, seven or 12 UDAs are available with a minimum UDA value of £23 being noted (NHS England, 2022a, 2022b).

In contrast, Scotland and Northern Ireland have continued to remunerate their dental providers nationally on a fee-per item basis. Each item of possible dental intervention available to NHS patients is given a fixed fee. The patient (where they are not exempt) pays 80% of the cost for each individual treatment item received on a treatment plan. The maximum any patient can pay is capped at £384, irrespective if the total cost of the treatment plan exceeds £480[1]. However, the fee-per item system has been criticised as being too complex, as over 400 separate charges for individual items of treatment exist. Furthermore, as the system is based on quantity alone, this may incentivise over-treatment, or increase the likelihood that the highest paid option of care will always be provided, when it may not be in the patients best interests (NHS Scotland, 2021; Business Services Organisation, 2021)

1.6.2 Provision of paediatric dental services in the UK

Across the UK, most children access care and treatment through general dental practitioners (GDPs). GDPs commonly provide this care under the UK National Health Service (NHS), where the treatment provided for all children under the age of 18 years is free, with no direct out of pocket costs or co-payments borne by the patient or their family. The provision of private care does exist with the treatment costs being paid in full, out of pocket, or on an insurance basis, by the child's parents/guardians.

[1] The maximum a patient can pay is £384 (80%). The 100% fee for this level of patient contribution equates to £480.

Sub-groups of children may present or be referred to a dentist with additional skills and experience, most commonly a specialist or consultant in paediatric dentistry. Specialists and consultants may work in the community dental service or in secondary (hospital) dental care facilities. Specialists provide oral healthcare, for children from birth to their 16th birthday, whose needs cannot be managed by their GDP. These children may have extensive oral disease, developmental disorders of the teeth and mouth, be too young or anxious to accept routine dental treatment or have intellectual, medical, physical, social, psychological and/or emotional factors/disability (NHS England, 2018). However, it has recently been highlighted that access to such specialists, and consultants, is limited and inequitable across England, and indeed the UK (Mills, 2020).

1.6.3 Implications for compromised first permanent molars in the UK

Anecdotally, it is likely that the 2006 changes have impacted the way cFPM are managed across England and Wales. Despite no specific evidence being available for cFPM, it has been reported that the new dental contract led dentists to make different decisions compared to pre-2006. In some situations, treatment plans and referral patterns were altered to avoid disadvantage to the dentist's business. Complex treatments were deemed financially unviable with such treatments, often dealt with by GDPs previously, more likely to be referred (Davies & Macfarlane, 2010). An alternative, anecdotal explanation could be that children with cFPM are referred more frequently as managing children in general is potentially more complex and 'time-demanding' than managing an adult, especially when the same treatments attract the same UDA value. The UDA value for a simple filling and extraction, the most common treatment options chosen for cFPM in children, are the same; however, the costs of providing these are likely to differ. Therefore, it is unknown whether these changes incentivise GDPs to choose one over another.

In England, a commissioning guide for paediatric dentistry highlighted that the management of cFPM can come under a GDP, those with a special interest or a specialist depending on the severity of the condition and need for additional services that only specialists are able to provide, e.g. general anaesthesia. Although the commissioning guide is an attempt to help practitioners, specialists, and commissioners, it adds to the confusion as all treatment options in managing cFPM, other than general anaesthesia, can be provided by any dental provider, irrespective of severity (NHS England, 2018; Taylor et al., 2019).

Section 1.7 has discussed the provision of dental care, in general and for children, in England. The implications on how the provision of care affects cFPM have been briefly highlighted. Section 1.8 will discuss the management strategies for cFPM, confusion over who is best to manage these teeth.

1.7 Management of compromised first permanent molars

Overall, there is limited evidence to support the optimal management for cFPM in children (Keightley & Surendran, 2021; Somani et al., 2022). As previously discussed, for the empirical work undertaken in this thesis, a cFPM has been defined as a restorable first permanent molar that has either distinct cavitation (ICDAS Codes 5 & 6) due to dental caries, or post-eruptive breakdown due to MIH, with an absence of any signs or symptoms of loss of pulp vitality (toothache). How these specific cFPM are best managed, where and who by, upon initial presentation, and in future, will be discussed in the rest of this section.

1.7.1 General overview of management strategies available for compromised first permanent molar teeth

When considering the treatment options for cFPM, there are three general management strategies available:

1. Active monitoring, accepting the tooth is likely to worsen without intervention
2. Restoring (filling) the cFPM (with the potential need for endodontic (root canal) treatment in some cases)
3. Extraction (removal) of tooth (with potential for orthodontic (braces) or prosthetic (false tooth) treatment in some cases)

A full assessment of the developing dentition and the patient is required before presenting and discussing available treatment options for cFPM. Factors such as the presence of dental pain, how affected the tooth is, condition of the remaining teeth, future orthodontic treatment need and the patient’s oral health values need to be considered (Keightley & Surendran, 2021; Somani et al., 2022).

1.7.2 Management strategy: active monitoring

Dental Caries

Active monitoring can be used in circumstances when the parent and/or patient declines treatment or wishes time to consider their options. Self-care approaches, such as flossing or targeted toothbrushing, can be encouraged to stop dental caries progressing (Public Health England, 2021).

Other approaches that do not involve restorative management of caries lesions include those where the dentist uses prevention methods “in office” such as those involving ‘site-specific prevention’, whereby application of 38% silver diamine fluoride (SDF) (Seifo et al., 2019) and/or a 5% sodium fluoride (NaF) varnish therapy (Public Health England, 2021) is undertaken to halt the dental caries process. For non-cavitated lesions, these can be non-operative such as the placement of fissure sealants or resin infiltration.

MIH

Self-care approaches, such as flossing or targeted toothbrushing, are not feasible options for MIH-affected cFPM as the pathology can’t be halted or reversed. Attempts to reduce hypersensitivity and increase mineral content have been proposed when active monitoring is chosen. Somani *et al*., (2022) reported a reduction in hypersensivity, for both mild and severe MIH-affected molars, using techniques such as topical fluoride varnish application and low-level laser therapy. The long-term efficacy is unknown, and generalisability reduced for these interventions as short follow-up periods and small sample sizes were evident across the studies.

A range of topical medicaments demonstrated success in remineralising MIH-affected teeth. Serious methodological flaws relating to the validity in measuring the mineral density reduces the generalisability of the results (Somani et al., 2022). Increasing the mineral content could improve the physical strength of the affected enamel (Farah et al., 2010), however, how this translates clinically requires further investigation.

If the decision is to actively monitor a cFPM, and not actively manage the tooth by restoring or extracting it, then the risk of dental pain and subsequent infection and/or sepsis is high (Leal et al., 2012; Grund et al., 2015; Taylor et al., 2018). The carious lesion will progress, whilst the risk of further post-eruptive breakdown, in an MIH-affected cFPM, is likely. It is likely if the tooth remains untreated, any future treatment to deal with the broken/painful/abscessed cFPM would be more difficult, more expensive and have less certain results, with ultimately the only feasible option being removal.

1.7.3 Management strategy: restoration (filling)

Dental Caries

In 2016, the 'International Caries Consensus Collaboration' presented clinical recommendations for carious tissue removal. Overall, options include non-intervention (sealing in dental caries), minimally invasive approaches or conventional restoration. When restoring a tooth, to maximise restoration success and longevity, attempts should be made to preserve as much healthy and remineralisable tissue as possible but remove sufficient caries to achieve a seal. Not all bacterially contaminated tissue is required to be removed, with carious tissue removal to firm dentine being proposed for shallow lesions and to soft dentine for deep lesions (Schwendicke et al., 2016). These recommendations are supported elsewhere (Duncan et al., 2019; Featherstone et al., 2021); however, inconsistent application clinically is apparent (Innes & Schwendicke, 2017; Edwards et al., 2021) as these cavities are likely being over invasively managed (Innes et al., 2016).

Historically, non-selective caries removal has been advocated. This technique involves the removal of all carious dentine and enamel until only sound enamel and hard dentine remain. The cavity is subsequently restored. However, more minimally invasive approaches such as selective removal, stepwise removal and sealing in dentinal caries have been proposed. Selective removal is the complete removal of the dental caries around the circumference or edges of the cavity, whilst leaving softened dentine at the base before placing a definitive filling. In contrast, stepwise removal is undertaken, most often, over two visits. On the first visit, selective carious dentine is removed to soft dentine, before being restored with a restoration designed to function for 3 to 12 months. During this time, the caries arrests, as sealed bacteria are inactivated, and dentine remineralises, becoming hardened and dried. On the second visit, selective carious tissue removal to firm dentine can be then completed, prior to a definitive filling. Finally, cavitated dental caries can be sealed either by placing a thin coating of dental material over the dental caries, or by placing a preformed metal crown that encompasses the entire tooth (Schwendicke et al., 2016, 2021).

A recent Cochrane review addressed the evidence of failure was lowest for selective carious removal, compared to complete removal and a stepwise approach (Schwendicke et al., 2021). For deep lesions, a network meta-analysis showed complete removal had a higher probability of failure compared to selective- and stepwise removal. Interestingly, there was an absence of evidence where performance of preformed metal crowns, using the Hall Technique, in the permanent dentition was compared to conventional restorations (Schwendicke et al., 2021). It could be hypothesised that either there is a lack of research into their use, or that in the permanent dentition, preformed metal crowns are not routinely used for caries management. This may be partially explained by a UK-based study, using clinical vignettes, confirmed this as very few GDPs and specialists in paediatric dentistry used a preformed metal crown to manage a cFPM in a 9-year old patient (Taylor et al., 2019).

For each of the intervention techniques to manage the carious lesion, the restorative material needs to be considered. The most common dental filling materials are dental amalgam and resin composite. Dental amalgam is inexpensive, with predictable outcomes (Burke & Lucarotti, 2018a). However, it is unaesthetic and does not bond to the tooth, thus needing mechanical retention which often involves unnecessary tooth tissue removal (Worthington et al., 2021). In addition, a recent European directive (Article 10 (2) of Regulation (EU) 2017/852 on Mercury), as a result of the Minamata Convention (United Nations Environment Programme, 2013), advised against the use of amalgam in children under the age of 15, unless strictly necessary. In contrast, resin composites provide a tooth-coloured alternative to amalgam. They utilise adhesive technology by bonding directly to the tooth; thus, reducing the amount of tooth tissue that needs removing (Opdam et al., 2014). Concerns have been raised about resin composite toxicity; however, advances in material science and regulation demonstrate this material is safe for use (Gupta et al., 2012; Hatton et al., 2022; German, 2022).

Worthington *et al*., (2021) reported that composite resins were more likely to fail and develop caries at the restoration/tooth interface compared to amalgam fillings. In contrast, composite resin restorations were less likely to fracture, than amalgam although this was statistically non-significant.

One study (Bernardo et al., 2007) reported failure rates of composite and amalgam across different teeth, in children aged 8-12. They reported that, of the 1,545 restorations placed in molars (765 amalgam; 780 composite), at a seven-year follow-up, 94.4% (n=722) amalgam, and 85.5% (n=667) composite restorations survived. Caries developing at the tooth/restoration interface was the most common reason for failure for both materials, but greater for composite resin. Conversely, there was no increased risk of fracture between composite and amalgam restorations in molar teeth (Bernardo et al., 2007). If a filling becomes defective, then replacement or repair are both viable options, despite there being no evidence to support one approach over the other for either material (Sharif et al., 2014)

Despite the evidence supporting the use of amalgam over composite resin, amalgam use is being phased out. Interestingly, five of the eight primary studies included in the Worthington et al., (2021) review were all undertaken over 20 years ago. Advances in composite resin technology have improved since then. A recent study, that used a dataset including over 25 million courses of treatment in primary care in Eastbourne, Sussex, UK, reported similar survival rates, that being no reintervention was needed, for composite (34% at 15 years) compared to amalgam restorations (41% at 15 years) (Burke & Lucarotti, 2018c, 2018a). Although, it should be noted that these treatments were completed between 1990 and 2006.

MIH

In addition to the concerns previously raised about amalgam toxicity, it is not recommended that MIH-affected cFPM are restored with amalgam, as lack of adhesion, and atypically shaped cavities, are likely to promote breakdown at the margins (Ghanim et al., 2017; Lygidakis et al., 2022).

As MIH is a developmental condition, the alterations in the microstructure and mineral content of these teeth makes restoring them challenging. An increase in porosity, reduction in hardness and elasticity and a change in carbon–carbonate ratios are observed in hypomineralised enamel (Elhennawy, Manton, et al., 2017). Increased protein content, within hypomineralised teeth, inhibit crystal growth, reducing the mineral quantity and overall quality of MIH-affected enamel (Farah et al., 2010). These changes mean that although adhesion to hypomineralised enamel is possible, decreased bond strengths and higher failure rates, compared to sound enamel, have been reported (Krämer et al., 2018; Lagarde et al., 2020). Anecdotally, advances in bonding techniques and/or a pre-restoration rinse with sodium hypochlorite (NaOCl), by altering the protein-rich structures, have been suggested as ways to improve adhesion to hypomineralised enamel (Lagarde et al., 2020). In their systematic review, which set out to assess strategies to optimise bonding of adhesive materials to MIH-affected enamel, Lagarde *et al.,* (2020) reported that two of the four included laboratory studies showed increased bond strengths after application of NaOCl, whereas the other demonstrated no additional benefit. One clinical study suggested a higher survival rate at 24 months of composite fillings when NaOCl was applied to MIH-affected enamel (Sönmez & Saat, 2017).

In a recent systematic review focussing on treatment modalities for MIH-affected teeth, Somani *et al.,* (2022) included eight studies where MIH-affected molars were restored with composite. Significant heterogeneity, in particular the variation in the primary outcome measures, precluded meta-analysis. One study, which restored the greatest number of MIH-affected molars, reported a success rate of 96.8% (n=316) after 24 months (Gatón-Hernandéz et al., 2020).

An apparent difference, noted across these studies, was how much MIH-affected enamel was removed prior to restoration. There are two approaches: total hypomineralised enamel removal and partial hypomineralised enamel removal. Total removal is where all visibly defective enamel is removed, which may require significant tooth tissue to be removed to achieve this, and in theory permits bonding to completely sound enamel; whereas partial removal is removable of hypomineralised enamel until resistance to the probe or bur is felt. In five out of eight studies in this review, all participants had total removal of enamel, with success rates ranging from 54% to 100%. In one study, all participants had partial removal with reported success rates of 62.3% and 80.8% (depending on pre-bonding enamel preparation) being reported. In the remaining two included studies, a combination of approaches was compared. The first of which reported 93.7% success, for total removal, compared to 80.7% success after 24 months. The second observed that 76.2% of composites placed survived after total removal, compared to 29.9% after partial removal, at an average follow-up period of 42.9 (SD 35.1) months (Somani et al., 2022).

For MIH-affected molars, alternative direct restorative materials that can be used are preformed metal crowns or glass ionomer. Preformed metal crowns are most beneficial when cFPM are severely affected, as they restore the structural integrity of the tooth and alleviate symptoms of sensitivity. Success rates ranged from 86% to 100%, in the three included studies in a recent review (Somani et al., 2022). In the recent review (Somani et al., 2022), five studies that used glass ionomer cements, reported success rates ranging from 7% to 98%, with only one study having a follow-up period that was greater than 24 months. Recent guidelines suggest that glass-ionomer cements should not be used as a definitive restoration, and thus are often placed in molars that are due to be extracted, as interim restorations, or for those children who might find it difficult to cope with the invasive procedure, or for whom moisture control is challenging (Lygidakis et al., 2022). Interestingly, a recent UK-based survey of practitioners corroborated this, as they were more commonly used as a temporary measure, prior to making a final decision on the whether to restore or extract cFPM (Taylor et al., 2019). Although, advances in modified glass ionomer cements are showing some promise, and might be an option as a more definitive restoration in the future, more evidence is required (Hill, 2022).

Laboratory-made restorations, specifically for MIH-affected cFPM, are most commonly available in three main categories: metal alloys, indirect composite, and ceramic restorations. Wear resistance, strength, durability, and aesthetics must be taken into consideration when deciding which material to use; however, no one material has been shown to be more successful than the others in any clinical study. Four studies, included in the most recent review (Somani et al., 2022), reported success rates ranging from 85% to 100%, with the shortest mean follow-up period being 34.8 months. Despite reported success, they are rarely used by primary care, or specialist, practitioners in the UK (Taylor et al., 2019).

Endodontic Therapy

Endodontic therapies, such as vital-pulp therapies (partial and coronal pulpotomy) and complete root canal treatment[2] (American Academy of Pediatric Dentistry, 2020) are required when there is irreversible inflammation in the pulp, or when there is loss of vitality. Despite this, their use in children with cFPM in the UK appears to be limited (Taylor et al., 2019).

Taylor *et al*., (2020) systematically reviewed the clinical success of endodontic therapies used on cFPM in children. In this review, they reported that vital pulp therapies have overall success rates

[2] A vital-pulp therapy is a procedure that treats inflamed and/or infected dental pulp tissue by only removing affected pulp tissue and leaving some healthy pulp tissue behind.

of at 91.3% (range 78.5%-100%) and 90.5% (range 70%-100%), for partial and coronal pulpotomies respectively. Factors such as rubber dam use, material choice, and maturity of the tooth did not influence the success rates of these techniques. Only one study included in this review reported a 36% success rate for complete root canal treatments in cFPM. It appears from this review that if cFPM are symptomatic, or the pulp is exposed during caries removal, then vital pulp therapies can be used with a high degree of confidence in their success and may provide an alternative to an elective extraction of the tooth.

1.7.4 Management strategy: extraction and replacement

Extracting cFPM can be carried out at any time. However, removing them at the ideal stage of dental developmental stage (usually occurring at a chronological age of eight to ten years) allows forward movement ('mesial migration') of the unaffected and unerupted second permanent molar into the space left by the extracted first molar (Cobourne et al., 2014).

A systematic review carried out by Eichenberger *et al*., (2015) showed that overall spontaneous closure was estimated to be good to perfect in 72% (95% CI: 63%; 82%) and 48% (95% CI: 39%; 58%) of cases after extraction of maxillary and mandibular cFPM respectively. Timing does appear to be critical, with this review suggesting extractions between 8-10 years old for an upper cFPM resulted in 80% spontaneous good to perfect closure, compared with 55% where removal was at 10.5 - 11.5 years and 56% where removal is beyond 11.5 years old. For lower cFPM, extractions carried out when the child was between 8 – 11.5 years old were statistically significantly more likely to have good to perfect spontaneous closure, when compared removal when they were <8 years-old. These findings may be somewhat limited by the fact that probabilities were based on data for only 38 upper cFPM and 489 lower cFPM respectively. Similarly, these studies were of low to moderate levels of evidence. These results need to be interpreted with caution as age of extraction alone is not a perfect predictor of spontaneous closure following an extraction.

A prognostic study quantified what radiographic features were likely to lead to successful spontaneous space closure outcome after loss of the permanent first molar. This study retrospectively assessed dental age, second molar developmental stage, second premolar and second molar angulations, and presence or absence of the third molar on pre-extraction radiographs of 148 maxillary and 153 mandibular cFPM, extracted from 81 participants. It was reported that 89.9% and 49.0% of spaces were closed in the maxillary and mandibular areas, respectively, using visual examination, study models, and/or radiographs as methods to assess closure. In addition, the presence of the third molar and a mesially angulated second molar were both clinically and statistically significant factors that predict favourable space closure (Patel et al., 2017).

To prevent overeruption, removal of the opposing upper cFPM, even if unaffected, may be considered when an affected lower cFPM is extracted. In contrast, extraction of an unaffected lower cFPM should never be considered when removing an affected upper cFPM. There is little evidence to support this practice (Innes et al., 2013; Cobourne et al., 2014; Noar et al., 2023), with the most recent UK national guidance suggesting that removal of an unaffected upper FPM should not be routinely carried out (Noar et al., 2023). Despite an absence of evidence, in cases where there is uncertainty, a specialist orthodontic opinion could be sought to support this decision (Ong & Bleakley, 2010; Cobourne et al., 2014; Lygidakis et al., 2022; Noar et al., 2023).

Despite planning, removing a cFPM at the 'optimal' stage of development may not always be feasible due to patient choice or more likely, the presence of symptoms. If a cFPM is extracted too early, there may be distal drifting and rotation of the unerupted second premolar and conversely, if too late, extraction will often result in unsatisfactory space closure with mesial tipping of the second permanent molar (Ong & Bleakley, 2010; Cobourne et al., 2014). In such cases, where partial or no spontaneous space closure is noted, the options available for future management of the space are to accept it, orthodontically close it or fill it with a false tooth.

Orthodontic Therapy

Following extraction of the cFPM, closing the space orthodontically is a feasible option, that often results in a positive outcome. It is most often done using fixed orthodontic (metal braces). Closing an maxillary cFPM space is straightforward; whereas, closing the lower cFPM space can be more challenging, given the tendency for the lower second permanent molar to tilt mesially and roll lingually when mesial-directed orthodontics forces are applied (Sandler et al., 2000; Chua & Felicita, 2015). Closing the space by mesially moving the second permanent molar was reported to take 10.1 ± 2.6 months for an upper tooth and 11.9 ± 4.2 months for a lower (Jacobs et al., 2011).

Orthodontic space closure of extracted cFPM sites appears to be more predictable when carried out as close in time to the extraction (Ong & Bleakley, 2010), although these findings were based on a total of 35 adolescents only. This outcome is contrary to that of Jacobs et al., (2011) who found no statistically significant difference in the amount of closure, or time required to close, between cFPM extracted before, or during orthodontic treatment. A recent systematic review concluded that no statistically or clinically significant differences were found in the duration of fixed orthodontic treatment between adults and adolescents (Abbing et al., 2020). Therefore, in summary, post cFPM extraction space can be closed at any age, with the chance of a successful outcome likely to be similar.

Prosthodontic Therapy

There was very little information in the literature on the question of replacing a cFPM space with a prosthetic tooth. Several options exist to replace a single missing molar tooth: removable partial dentures (RPD), fixed bridges (FB), or dental implants (DI). RPD are relatively quick to construct and do not require tooth preparation. However, they are removable, and require a lot of dental material to retain just one tooth, thus introducing oral hygiene challenges. In contrast, FB are fixed in the mouth, and may require preparation of the adjacent teeth to aid retention. DI are titanium screws placed directly into jaw and are used to support crowns. They are fixed and do not require any preparation of the adjacent teeth; though, they do require invasive surgery as well as sufficient quality and quantity of bone to retain them (Al-Quran et al., 2011; Bohner et al., 2019).

However, DI placement to replace a space left post extraction of a cFPM in children is not recommended. As the DI is fixed into the jaw, the DI is incapable of following the continuous eruption of adjacent natural teeth during growth of the jaw bones, thus clinically manifesting as a discrepancy in the height of the occlusal plane (biting surface), compared to adjacent teeth. Therefore, FB or RPD are the only available options for use in children and until the patient has ceased growth.

1.7.5 Confusion relating to current management strategies available for cFPM

As demonstrated in sections 1.8.2, 1.8.3 and 1.8.4, cFPM can be managed in many different ways. Upon first presentation in childhood, the most routine options available are restoration or extraction. Thereafter, strategies will depend on whether there is a preference to retain teeth or replace the space, following extraction. At present there is a lack of understanding as to what is the optimal decision for managing cFPM (Ashley & Noar, 2019; Taylor et al., 2019; Alkhalaf et al., 2020; Lygidakis et al., 2022).

Restoration vs. Extraction

The key question of whether to restore or extract cFPM in children remains unanswered (Taylor et al., 2019; Keightley & Surendran, 2021; Somani et al., 2022).

Restorative options will allow the tooth to remain in-situ. Restoration is a less invasive procedure than extraction of the tooth and, given the advances in restorative techniques, it is a viable option for carious- and MIH-affected cFPM, being recommended as a viable option for mild to moderately MIH-affected teeth in the most recent management guidelines (Lygidakis et al., 2022). There appears to be a desire amongst most adults interviewed in an earlier study about retaining teeth for as long as possible, with some interviewees describing tooth loss as being devastating and disruptive (Rousseau et al., 2014). However, there are consequences to restoring a tooth in a child. It means entering the 'restorative cycle' at an early stage in life where any filling placed will eventually fail, requiring larger replacements each time, until such a point that there is no more tooth substance left to restore, and the tooth is extracted (Elderton, 1998). It is suggested that should a large restoration be required to repair the cFPM, then it is likely to have a poorer long-term prognosis and shorten the overall restorative cycle of that tooth (Blum & Özcan, 2018).

The alternative, extraction, prevents the need to maintain a restoration throughout life, despite being more invasive. A recent study attempted to demonstrate the effect extracting a lower cFPM in children (aged 8–11 years) had on the position/angle of the developing third molar (Murphy et al., 2022). Using radiographs, at two different time points, it was noted that the third molars moved significantly more mesial in the extraction group compared to the non-extraction group (Murphy et al., 2022). Thus, suggesting that early removal will improve eruption of the third molar into a more favourable position and could reduce the impaction, and associated morbidities, that third permanent molars (Murphy et al., 2022). Therefore, if removal is chosen, then spontaneous closure of the space, if carried out at the correct time, could occur, negating the need for orthodontic space closure, or prosthetic replacement. However, as discussed earlier, these outcomes are not guaranteed, and unfavourable tooth movement, or tipping of adjacent teeth, may occur (Eichenberger et al., 2015).

Despite the advantages and disadvantages of these two main approaches, there is an absence of evidence that guides patients and professionals with what option they should choose. In reviewing the literature, no data was found on the views and opinions of children/adolescents on how they should manage cFPM. In a recent study, parents of children with cFPM, reported satisfaction with having cFPM extracted; however, some reported they would be accepting of spaces, if restoration had been placed initially and then failed at some point in the future (Agel et al., 2021).

Similar to patient and parent views, it is important to consider what management options are currently being offered for these teeth, in the UK, in addition to what evidence is available to support professionals in offering either restoration or extraction for children presenting with cFPM.

In a UK study, 52% (n=206) of children with MIH had at least one MIH-affected molar extracted over a one-year period (Humphreys and Albadri 2020). These results were corroborated by the findings of a recently published study that reported 81% (n = 201) of children with cFPM had extraction of at least one (AlKhalaf et al. 2022). These studies were based in hospital-based specialist-led paediatric dental services, which biases the results as these children are more likely to have additional complexities, given the need to be referred to a specialist. In addition, these results do not reflect what management is currently being done in primary care.

In contrast, a UK-based cross-sectional study of GDPs and specialists, observed that decision making to manage cFPM is complex, with substantial differences noted between, and within, these professional groups (Taylor et al., 2019). One of the clinical vignettes used in this study focussed on a 9-year-old child, with cFPMs that could be managed with either a restoration or extraction. Responses to this vignette created greatest diversity in the results, with GDPs favouring restoration of cFPM, whilst the specialists in paediatric dentistry were mainly unequivocal, there was a slight preference towards extraction. Furthermore, a strong positive correlation was observed in GDP responses to questions about having a responsibility to treat cFPM and confidence in doing so. Interestingly, over half of GDPs still preferred to have a specialist orthodontic opinion for treatment planning children with cFPM. This could suggest that GDPs were confident to make the decision and manage the cFPM, based on the clinical findings presented to them, but lacked confidence about the impact their decision may have in the longer-term. Similarly, specialists reported that GDPs had a responsibility to treat children with cFPM in practice, whilst also suggesting referring to a paediatric specialist for management. This seems to be contradictory, however, perhaps illustrates that some specialists are unsure where these teeth are best managed or that children with milder cFPM can be treated by GDPs alone, but for more severe cases, specialist input is required (Taylor et al., 2019).

It could be argued that compared to other countries, extraction of cFPM in the UK may be more common than restoration (Taylor et al., 2019; Humphreys & Albadri, 2020; Sanghvi et al., 2022) as there were only a few European studies that reported extractions of cFPM (Eichenberger et al., 2015). In contrast, several European studies have reported a preference towards restoration, primarily with composite (Kotsanos et al., 2005; Kopperud et al., 2016; Wall & Leith, 2020) whilst others advocate the use of indirect restorations (Gaardmand et al., 2013; Dhareula et al., 2019). Several anecdotal reasons may explain these geographical differences. There may be variations in societal preferences for retaining versus removing teeth in children between countries. Alternatively, there may be a lack of availability of adjunctive services such as sedation and general anaesthetic available to dentists in Europe may drive the preference to restore cFPM, as under local anaesthetic alone, this is a less invasive option. Instead, it could be that the provision of children's dentistry in Europe may drive decisions. In Europe, dental care for children is predominately provided on a private basis, although some do provide subsidised dental coverage. Thus, if a tooth is restored, then future option of teeth restored (e.g., root canal treatment etc.) will provide further payment. Whereas, if a tooth is removed, then there are no further payments available to be made for that tooth, as any future orthodontic movement or prosthetic replacement would be provided by another practitioner.

In summary, deciding how best to manage cFPM in children presents challenges. The most pertinent question is how to manage these teeth upon early presentation: Should they be restored, and maintained, or should they be extracted at the ideal time to allow for spontaneous space closure? Individual clinical and patient factors may suggest a preferrable option, but it is not as simple as what is regarded to be the most successful option as several factors, including patient and parent values and opinions, in addition to economic and societal implications have to be considered when making this decision. Unfortunately, an absence of evidence exists as to which of these is the 'optimal' option, taking all these factors into consideration, for children presenting with a cFPM (Taylor et al., 2019; Ashley & Noar, 2019; Keightley & Surendran, 2021; Somani et al., 2022).

1.8 Conclusion

This chapter has explored the problem that is cFPM teeth in children. A brief overview of the literature pertaining to the problem, the aetiology, epidemiology, inequalities, and direct and indirect impacts associated with the most common dental conditions to contribute to this problem, dental caries, and MIH, have been presented alongside how these teeth are currently managed for under the provision of dental care for children in the UK.

Chapter 2 will complement the contemporary evidence that has been outlined in this chapter. It will detail the basic principles of economic evaluations, including a summary of their use in health and dentistry, before focussing on the two proposed methodologies used in this PhD, decision analytic modelling and preference elicitation.

Chapter 2. Economic evaluations and their application in oral health

2.1 Introduction

The argument that cFPM are a significant oral health issue for children in the UK has been set out in Chapter 1. In addition, the confusion surrounding the management strategies and deficiencies in conventional research methodologies to understand the long-term impact of early childhood decisions were explored in that chapter. As alluded to in Chapter 1, economic evaluations can be used to understand both the short and long-term implications of such early childhood decisions for cFPM.

In this chapter, section 2.2 will provide a brief introduction of the general principles of economic evaluation in health. Following section 2.2, the focus is on three related issues that represent challenges and opportunities for the economic evaluation. Section 2.3 briefly explores how health is valued for economic evaluations. Thereafter, section 2.4 details how stated preferences are elicited, primarily focusing on the main approaches before discussing methods used in children, whose preferences to elicit (patient or parents) and those used in oral health. Section 2.5 provides a brief overview of trial-based economic evaluations and decision analytical modelling. Finally, a brief review of published dental economic evaluations, including those in child oral health, is explored in section 2.6.

2.2 Economic evaluation – general principles in health

2.2.1 Principles of economic evaluations

Economics is concerned with maximising benefits from the resources that are available to be used. It is based on three key principles: scarcity, choice and opportunity cost (Drummond et al., 2015). Fixed health budgets exist in all healthcare systems, for example the National Health Service (NHS) in the UK, or privately funded care (e.g., as the number of staff and facilities available are fixed, at least in the short term). Choices are therefore necessary e.g., which interventions to provide, drugs to fund etc., as the resources available are scarce relative to the needs for care. That is, there will never be enough resources to be able to produce all the healthcare that is needed. As a result, decisions need to be made, between different health-care interventions or health-care programmes, to ensure that neither too many nor too few resources are being used (i.e. the use of resources is 'optimal') to produce maximum benefit (Eddama & Coast, 2008). When resources are allocated to one intervention/programme it means that the opportunity to spend these resources on alternative intervention/programmes is forgone. This concept is more commonly referred to as opportunity cost, that is, the:

"…benefit that would have been derived from using a resource in its next best alternative use (Elliot & Payne, 2005, p.14)".

In deciding between different options for using resources, both costs and benefits need to be considered. An economic evaluation in healthcare is defined as the:

"…*comparative analysis of alternative courses of action in terms of both their costs and benefits* (Drummond et al., 2015, p.4)".

In this definition, alternative courses of action must be mutually exclusive, that is a patient can only receive one and not both interventions at the same time. Analysing these alternative courses relies on establishing the relevant costs and benefits, or outcomes, for the alternative healthcare interventions and/or programmes being examined. It is worth noting, the term consequence can be interchanged with terms such as effects, benefits, and outcomes. For the purposes of the remainder of this thesis, the term benefit will be used. In order for an economic evaluation to be conducted there are several key concepts that need to be considered. These are perspective, discounting, time horizon, sensitivity analysis, and technical/allocative efficiencies are now discussed each, in turn.

Perspective

Perspective is the point of view that is adopted for any given economic evaluation. Choosing the appropriate perspective(s), must be decided early, as it will determine what costs and benefits require to be taken into consideration (Ternent et al., 2022). If only a patient perspective was chosen, then only costs that are borne by patients directly with the disease process being investigated are included (e.g., direct cost of healthcare as paid by the patient (or the child's family) but also other indirect costs such as travel, childcare etc). Alternatively, an evaluation could take a healthcare provider perspective (e.g., NHS, or a hospital trust etc.), whereby the only costs included are those borne by the healthcare provider. This would exclude out of pocket expenses that are borne by the patient. If a societal perspective is adopted, this would include costs borne by the patient, the health system, and wider society. An example of a cost to the wider society could be the costs and harms that fall on the environment from medical waste. Other perspectives, such as technology manufacturers, exist. However, common approaches are health care provider, health care payer, or combinations that combine provider/payer perspective plus patient perspectives (Garrison et al., 2018).

Discounting

Costs and benefits related to an intervention in an economic evaluation will occur at different points in time, some in the present and in the future. Those outcomes that present in the future are commonly valued less than they would at present (known as time preference). This reflects the fact, for example, that individuals would prefer to have money now, rather than later, as they can benefit from having that money in the interim. As such, costs and benefits are recommended to be discounted in any economic evaluation (Claxton et al., 2011).

In England, the National Institute for Health and Clinical Excellence (NICE) suggest that the results of any economic evaluation should reflect the present value of the costs and benefits that accrue over the length, or time horizon, of the analysis. For health care technologies, an annual discount rate of 3.5%, which is based on the recommendations of the UK Treasury, should currently be used for both costs and benefits (NICE, 2022a).

It is important to consider discounted future costs, to reflect that the amount spent or saved in the future should not weigh as heavily in decisions as those spent or saved today. This positive rate of time preference for costs is argued to be appropriate because of positive economic growth over time, and therefore in economic evaluations, all future costs need to be reduced to reflect the lower cost compared to costs incurred today (Rudmik & Drummond, 2013).

Similarly, benefits may be discounted as a healthcare intervention, or programme implemented may provide immediate health benefits, or losses, for a given population, as well as in the future. However, there is a debate whether benefits should be discounted in the same way that costs are.

One argument for discounting costs relates to returns to investments in the marketplace, whilst discounting benefits is linked to strength of preferences for consumption of a good now rather than later (Nord, 2011; Drummond et al., 2015).

Time Horizon

The time horizon is the duration over which costs and benefits are considered. It should be long enough to reflect key differences in costs and benefits. The same time horizon should be used for both costs and benefits. The choice will often depend on the nature of the disease and intervention under consideration and the purpose of the analysis (Drummond et al., 2015; NICE, 2022a).

In some economic evaluations, the measurement of benefits will be based directly on the clinical evidence established from an empirical study, such as is the case in clinical trials. However, the follow-up period of the trial may not be long enough to fully reflect all the key differences between the benefits, and the associated costs, of the alternative courses of action involved. However, a longer time horizon such as an estimated life time requires assumptions and modelling in order to extrapolate costs and benefit data, as it is unlikely that data from a single clinical trial or cohort will exist to provide these answers (Drummond et al., 2015; Bojke et al., 2017).

Sensitivity analysis

Every economic evaluation will have a certain level of uncertainty associated with it. Briggs et al., (2012) proposed several different types of uncertainty that can be either related to the different sources of data to be used, or uncertainty in the methodological assumptions. A summary of these concepts is shown in Table 2.1.

Term	Concept
Stochastic Uncertainty	Random variability in benefits identified between individual patients, as individuals facing the same probabilities and outcomes will experience the effects of a disease or intervention differently
Parameter Uncertainty	Parameter uncertainty, or data required to be input into the economic evaluation, can be linked to methodological uncertainty of the studies that are used to estimate the parameter. Uncertainty could arise from multiple studies that provide conflicting estimates of the same parameter. In addition, there could be problems with the internal or external validity of a study that is providing data. Finally, there may be parameter uncertainty because there is a lack of empirical data, and expert opinion may be required.
Heterogeneity	Variability between patients, of the intervention being investigated, due to the characteristics of those patients
Structural Uncertainty	Structural uncertainty focusses on the uncertain functional form of any decision model. Scientific judgements are made when constructing a model (e.g., assumptions used to extrapolate costs and benefits over time) which affect the model structure and will introduce uncertainty.

Table 2.1 A summary of uncertainties that may present in an economic evaluation (adapted from (Briggs et al., 2012))

The most common sensitivity analytical approaches for model-based analyses are deterministic sensitivity analysis (DSA) and probabilistic sensitivity analysis (PSA), with both being used in this thesis. The two are not mutually exclusive and can be used in combination as described below.

Deterministic sensitivity analysis

This method can be used to investigate the sensitivity in variations of a specific input parameter or set of parameters. These can be manually changed, across a pre-specified range, and the results are analysed to see to what extent the change has made on the outcome. The range will correspond to the uncertainty that was reported for the parameter in the source study, that is the point estimate of each input parameter value is used. Alternatively, it may reflect a legitimate difference e.g., the use of an alternative value for a cost, the impact of a change in discount rate, etc. In a one-way DSA, only one parameter is changed at a time, whilst in multi-variate DSA, several parameters are varied simultaneously.

Petrou and Gray (2011b) report that using DSA to address aspects of parameter uncertainty does not properly reflect the role of joint uncertainty or indeed the possible correlation between the variables. Furthermore, varying more than five parameters at the same time using a DSA approach is often not feasible. DSA can be used to explore model validity – that being changing parameter values and looking for counter-intuitive results. Furthermore, if conclusions of the model are unchanged when extreme value DSA is used, then PSA may not be necessary. Finally, not all parameters can have distributions e.g., discount rates. In these situations, as noted above, DSA can be combined with PSA.

Probabilistic sensitivity analysis

This technique can be used to understand several parameter uncertainties at one time. In a probabilistic analysis, parameters are represented as distributions. These distributions are supported by evidence from a range of source studies, such as empirical data, the literature and expert opinion.

In PSA, a set of input parameter values are drawn by random sampling from each distribution and the model is 'run' to generate both cost and benefits outputs. This is repeated many times, typically 1000 to 10,000 resulting in a distribution of outputs. From this an average result with a 95% credible intervals, sometimes known as an uncertainty interval can be produced (Petrou & Gray, 2011b).

The key principles of economic evaluations have been described. It is also important to consider the role economic evaluations have in estimating efficiency. Efficiency measures whether healthcare resources are used to achieve the best value for money, thereby aiding health care decision-makers on how best to allocate scarce resources in a way that maximises efficiency and reduces opportunity costs (Palmer & Torgerson, 1999; Eddama & Coast, 2008; Drummond et al., 2015). Weinstein and Stason (1977) propose that economic efficiency relates to society making choices to maximise the health outcomes gained from the available resources. An economic evaluation will thereby provide information to aid judgements about efficiency. They do this by considering how inputs (resources) are converted into outputs (health gain, improving quality of life, etc.). This enables judgements to be made about whether the outputs obtained in using resource this way are worth the benefits that could have been obtained (i.e., the opportunity cost) had they been used in another desirable way. Evaluating efficiency, together with efficacy (the benefit of a treatment under ideal conditions), effectiveness (the benefit of a treatment as measured in routine clinical practice), and equity (a distribution of outcomes such as health or wealth, that is considered just and fair) are important when addressing the quality of healthcare (Drummond et al., 2015).

Efficiency is categorised into two types, technical and allocative efficiency which will now each be described in turn.

Technical Efficiency

Technical efficiency is about how to provide care. It can either be what is the least costly way of obtaining a given output, or how to maximise outputs using a set of given resources (or inputs). When addressing technical efficiency, it has already been decided that the intervention(s) or healthcare programme needs to be done, but how best that is done given the resources available is yet to be established (Palmer & Torgerson, 1999; Elliot & Payne, 2005).

Allocative Efficiency

Allocative efficiency is about what, or how much of a healthcare intervention(s) or healthcare programmes to provide. It is broader than technical efficiency as it considers which programmes resources should be allocated to in order to maximise social welfare (Palmer & Torgerson, 1999). Wiseman and Jan (2011) suggest that attempting to achieve allocative efficiency is similar to judging which healthcare intervention/programme is worth pursuing.

Therefore, economic evaluations are used to address whether efficiency, either or both of technical or allocative efficiency, can be improved in addition to informing judgements on the provision of health care (Elliot & Payne, 2005). The next section will focus on the most common types of economic evaluations, and how they differ from one another.

2.2.2 Types of economic evaluation

There five major types of economic evaluation are:

- Cost-minimisation analysis (CMA)
- Cost-effectiveness analysis (CEA)
- Cost-consequence analysis (CCA)
- Cost-utility analysis (CUA)
- Cost-benefit analysis (CBA)

Each type of economic evaluation will need to identify, measure and to a lesser or greater extent value both costs and benefits. The difference in outcome characteristics, of each type of economic evaluation, are shown in **Table 2.2** (Drummond et al., 2015).

Type of Economic Evaluation	Measurement/ valuation of Costs	Identification of benefits	Measurement of benefits
Cost-Minimisation Analysis	Monetary Units	Common to both alternative courses of action and are shown or assumed to be the same	No measurement needed as benefits are gained to the same level
Cost-Effectiveness Analysis	Monetary Units	Single benefit of interest, common to both alternative courses of action, but gained to different levels	Typically, natural units are measured
Cost-Consequence Analysis	Monetary Units	Any number of benefits of interest, common to both alternative courses of actions, that must not overlap one another	Different aspects of benefits valued in natural units, healthy years, typically measured as QALY's or in monetary units. No one method of valuation is used for all aspects of benefits
Cost-Utility Analysis	Monetary Units	Single or multiple benefits that are not necessarily common to both alternatives, but which can be valued using a common metric	Healthy years, typically measured as QALY's
Cost-Benefit Analysis	Monetary Units	Single or multiple benefits that are not necessarily common to both alternatives, but can be values in a common metric that is commensurate to the valuation of costs	Commensurate with costs, typically, monetary units

Table 2.2 Different outcome characteristics in each type of Economic Evaluation (adapted from (Drummond et al., 2015))

Considering costs first, in strictest terms, the costs of the resources should generally be valued on an opportunity cost basis. However, it is often the case that these opportunity costs are proxied by using monetary units. Using this proxied approach, consideration must be given to the market prices of the resources, although it could be that these prices are a poor proxy for the opportunity cost. This proxied approach first requires identification of all resources that are required for each course of action. Next, quantifying the total number of each identified resource is undertaken. Finally, a monetary value which reflects the opportunity cost (which is not a fixed value but depends on the context), is appropriately assigned.

Now considering benefits, understanding which benefits to include follows a similar approach to that of costs. Firstly, identification of which benefits, both health and non-health, will be gained is undertaken. Next, ascertaining how the benefits gained will be measured e.g., clinically, or using quality of life tools is undertaken. Finally, these benefits are assigned an appropriate value e.g., Quality Adjusted Life Years (QALYs) (Drummond et al., 2015).

Whilst economic evaluations are usually defined by the type of benefit included, types of economic evaluations can also be defined practically on the type of efficiency that is being addressed. CMAs only address technical efficiency. CCAs can address both technical and allocative efficiencies in health care and the wider economy. However, CCAs may not deemed to be a full economic evaluation as the benefits are not fully valued. CEAs are best suited to addressing technical efficiency but can provide some limited information on allocative efficiency if the results are presented as an incremental-cost effectiveness analysis (ICER). CUA can be used to address technical and allocative efficiencies provided the perspective is limited to the health care sector and the only outcome we are interested in is health. Similarly, CBA address both efficiencies in the health care sector and the wider economy as a CBA is defined as including all costs, and all benefits no matter on who they fall.

Having shown variations in how the benefits are identified and how they are measured/valued for each type of economic evaluation, the following sub-sections will discuss each of the five main types of economic evaluation in more detail.

2.2.3 Cost minimisation analysis (CMA)

A CMA is used when the benefits of two or more alternative actions have no difference. Therefore, it is a direct comparison of the costs that is of interest, with the less costly option being recommended. In reality, it is very unlikely that two alternative course of actions will have exactly the same benefits, or have no uncertainties associated with either the clinical or economic parameters (Elliot & Payne, 2005). For these reasons, CMA is rarely justified as a form of economic evaluation (Drummond et al., 2015).

2.2.4 Cost-effectiveness analysis (CEA)

In CEAs, the benefits relate to a single, common effect which occurs to a different extent between alternative courses of actions, measured in unidimensional natural units. Examples of a natural unit could be the number of episodes of pain avoided for different type of materials used to fill teeth, or millimetres of mercury reduced in patients with high blood pressure. Most commonly, when one intervention is more effective but more costly than another, the results are presented in terms of an ICER. That is how much extra cost is incurred to obtain an extra unit of that outcome e.g., an episode of pain avoided.

The ICER from a CEA provides some limited information to guide decisions about allocative efficiency (Drummond et al., 2015). This occurs when an explicit or implicit threshold value is set for the given outcome, with results sometimes being phrased as 'Intervention x is cost-effective compared to intervention y if we are willing to pay £z for an additional unit of [effectiveness measure]'.

However, there are several limitations of CEAs. The natural unit of effect chosen may not necessarily be the most appropriate to use and the natural unit will not necessarily be relevant to other disease/conditions, even within the same health area.

Another drawback of CEAs is that natural units may not fully reflect the impact on the patient, in the widest sense of health or wellbeing. Any single clinical or natural measure is unlikely to reflect all the impact e.g., both benefits and harms or in terms of a clinical measure may have a more complicated interpretation. Therefore, incorporating a wider sense of health or wellbeing, including patient values, as well as assessing the opportunity cost across different disease/conditions and/or interventions/healthcare programmes, may be better done using a CUA, with a generic measure of benefit, or by undertaking a CBA (Drummond et al., 2015).

2.2.5 Cost consequence analysis (CCA)

CCAs is an economic evaluation that lies somewhere between CEA and CUA (if the benefits are restricted to health) or between CUA and CBA (if wider benefits are considered). They are not regarded as full economic evaluations as the outcomes are not fully valued. However, a recent medical research council framework for developing and evaluating complex interventions suggest that CCAs may often be more suitable for an economic evaluation of a complex intervention than narrower approaches, such as cost effectiveness or cost utility analysis (Skivington et al., 2021). The benefits in CCAs are usually presented in a disaggregated manner, that is the benefits from all viewpoints e.g., patient, health service, society, etc. of two or more alternative interventions. The costs can be presented in a similar way (e.g., patient costs, intervention cost, NHS costs, etc), or just from one viewpoint. Presenting in such a way allows all decision-makers (e.g. a clinician, a commissioner of services) to form their own opinion on the relative importance of the costs and benefits from each viewpoint of each intervention that is most relevant to their context (Drummond et al., 2015). Despite being easy to understand, there is risk that with CCAs, investigators and/or decision makers may choose to only select and report the most positive results (Drummond et al., 2015); although in a study evaluating a complex intervention, to do so may be appropriate (Skivington et al., 2021).

2.2.6 Cost-utility analysis (CUA)

CUA's measures benefits in terms of both quantity (life years) and quality of life, which, are combined into a single measure of health most commonly QALYs. Alternative measures such as the healthy years equivalent, disability-adjusted life years and saved young life equivalent have been proposed as alternatives to QALYs. However, healthy years equivalent and saved young life equivalent are rarely used, whilst disability adjusted life years are mainly used in evaluations conducted in low and middle income countries (Drummond et al., 2015). To establish a quantifiable measure of quality of life, health state utilities can be used to measure how strongly a person values a certain state of health.

Health State Utilities

A health state utility value is a score, or preference weight, that is most often between 0 (a state equivalent to dead) and 1 (full health) (Torrance, 1986; Whitehead & Ali, 2010; Devlin & Brooks, 2017). Negative values, or values less than 0 can exist in some frameworks for measuring health utility, and represent health states considered to be worse than death (Whitehead & Ali, 2010; Devlin & Brooks, 2017).

Health state utilities can be established using various direct elicitation methods, such as visual analogue scale, time-trade off or standard gamble, although using these methods can be costly and time consuming (Whitehead & Ali, 2010; Drummond et al., 2015). The more common approach is to derive values from existing HRQoL life measures, such as EQ-5D, using indirect elicitation methods (Whitehead & Ali, 2010).

Deriving QALYs from EQ-5D, and derivatives

The most commonly accepted generic measure used to establish health states is the EQ-5D (Rabin & de Charro, 2001). One version, EQ-5D-3L, measure contains five domains: mobility; self-care; usual activities; pain/discomfort; and anxiety/depression. Each domain has a single question with three levels of severity and is scored as 1 (no problem) to 3 (severe problems). Therefore, a respondent who gives a response scored as 11111 would have the best health state possible. Using the EQ-5D-3L, there are 243 possible health states. Unique health states, reported by patients using the EQ-5D-3L instrument, have been mapped to provide a value set of utility values using general population surveys. This can be summarised as a single value and used to help calculate QALYs (Kind et al., 1999; Devlin & Brooks, 2017). Since the development of the EQ-5D-3L further variations to the EQ-5D tool have been developed including the EQ-5D-5L (Devlin & Brooks, 2017).

The EQ-5D-3L and EQ-5D-5L are only validated for use in people aged 12 years of age or older. A child-friendly version, EQ-5D-Y was developed in 2010 to measure HRQoL in young people in an age-appropriate manner. An international valuation protocol, to allow EQ-5D-Y value sets, was only published in 2020 (Ramos-Goñi et al., 2020) with only a handful of countries to date publishing value sets (Prevolnik Rupel et al., 2021; Shiroiwa et al., 2021; Ramos-Goñi et al., 2021). At present, there is no UK value set of the EQ-5D-Y tool.

Utility values (quality) from measures such as EQ-5D-3L are combined with the time spent (quantity) in a particular health state. This gives a total number QALYs, where 1 QALY is equivalent to 1 year in full health. For example, if an individual is in a health state for 10 years, where the health state has an associated utility of 0.8, this would generate eight undiscounted QALYs (i.e., 0.8 multiplied by 10 years).

Use of QALYs in CUA

In England, it is an official requirement of economic evaluations undertaken by/for NICE to report QALYs as the measure of benefit (NICE, 2022a). The cost-effectiveness threshold for use in CUAs adopted by NICE ranges from £20,000 and £30,000 per QALY gained. However, thresholds are not fixed and do vary according to the context e.g., higher thresholds are adopted for end-of-life treatments, or rare diseases (NICE, 2022a).

CUAs have several advantages compared with CEA. Utility values provide a summary measure of health gain in terms of both quantity and a judgement of quality of life, rather than just describing a change in a natural or clinical unit. Furthermore, CUAs can compare the incremental cost per QALY for healthcare interventions/programmes in either the same or different health areas. This comparison enables judgements about opportunity costs to be more readily established across various health areas.

However, limitations of CUAs do exist. Claxton et al., (2015) argued that the current NICE threshold used for CUAs is set too high, was based on arbitrary figures and hasn't changed in over ten years. In most cases, CUAs will use utilities derived from generic measures of HRQoL, such as the EQ-5D. However, such measures might be insensitive to particular diseases, or unable to detect any discernible differences (Whitehead & Ali, 2010). In contrast, condition-specific outcome measures or preference based condition specific utility tools (e.g., glaucoma utility index (Burr et al., 2007)) can be used instead. Brennan and Spencer (2006) attempted to map a specific oral health measure, the Oral Health Impact Profile (OHIP) to a generic health state measure, the EQ-5D, in order to estimate health state values based on OHIP data. Despite Brennan and Spencer (2006) concluding health state values can be derived from OHIP scores, it could be argued that the two measures used in their mapping process were not relatable, and thus renders the process futile. In an attempt to overcome the need for mapping, two caries-specific utility measures currently exist, CARIES-QC-U (Rogers et al., 2020) and Dental Caries utility index (Hettiarachchi et al., 2020), although to date, their routine use for dental CUAs remains limited.

2.2.7 Cost-benefit analysis (CBA)

CBAs use commensurate units to value costs and benefits. This is most commonly a monetary valuation of health, and non-health, outcomes that can be interpreted alongside the costs of the programme (Birch et al., 1999; Brent, 2003; Drummond et al., 2015).

The results of CBAs can be presented as a simple sum representing the net monetary benefit, or loss, or the ratio of benefits to costs to allow a direct interpretation. The healthcare intervention/programme with the lowest costs to benefits ratio is the most efficient. That is the additional benefits will exceed the increased costs of the chosen option compared to its alternative (Birch, 1987; Drummond et al., 2015).

CBAs are the most comprehensive method of economic evaluation as they usually adopt a societal perspective. This allows comparison across other areas of the economy, such as transport (Elliot & Payne, 2005). However, it is possible to measure costs and benefits in a CBA for a narrower set of outcomes, using a health impact perspective instead.

In comparison to CUAs and CEAs, CBAs have the advantage that they can include both health and non-health benefits. Like CUAs, CBAs can be used to inform resource allocations across different healthcare settings or be compared across different sectors of the economy.

Limitations of CBAs do exist. Firstly, the values assigned to health and non-health benefits, in monetary terms, are likely to differ significantly between individuals as WTP is a function of ability to pay. Furthermore, eliciting WTP requires individuals to be fully informed of the benefits, which can be challenging when outcomes are unknown, or, even when they are, it may be too cognitively demanding to take in all the information to establish this (Ryan et al., 2008). In addition, inaccurate WTP elicitation may occur if an unrepresentative sample of the population is obtained, as preferences, as well as risk taking, have been shown to vary over time (Albert & Duffy, 2012). Collecting data on individuals' willingness to pay for a health gain is also not straightforward. There is considerable debate about the most appropriate tools to do this along with concerns about the acceptability of valuing 'health' in monetary terms. As a result, this is often why alternative economic evaluation methods, such as CEAs and CUAs, are often used in the health care sector.

Section 2.2 has summarised the main principles that underpin economic evaluations as well as discussing the main types of economic evaluations used in health. The next section will focus more in depth on how health and oral health is valued, and then measured in monetary terms.

2.3 The valuation of health

2.3.1 Valuing health

Valuing health is critical to determine how best to allocate resources, within a fixed healthcare budget. In the UK, individuals' valuations are most often used to calculate QALYs, and the QALY still remains the preferred measure of benefit to be used in economic evaluations conducted for NICE (NICE, 2022a). Dolan et al., (2009) noted concerns with the recommended approach by NICE (NICE, 2022a) because hypothetical preferences, often based on generic assessment of HRQoL, bear little resemblance to the real-life experiences of those who live in those health states (Dolan et al., 2009).

Karimi et al. (2017) reported that individuals will align their valuation of health with their personal interests, circumstances and environment that surrounds them and this of course aligns with a central tenant of classical economic theory. However, methods that seek to capture influences beyond health are not recommended in the methods guidance for the conduct of technology assessment reviews in England (NICE, 2022a).

2.3.2 Valuation of health and non-health outcomes

Where functioning markets exist, monetary valuation of the outcomes can be easily undertaken using prices revealed in the market. However, when markets do not exist, or depart significantly from perfect competition, prices do not reflect the opportunity cost of resources used to produce the outcomes of interest. In these circumstances it is necessary to either use the human capital approach or estimate the WTP for a healthcare intervention or programme.

Using the human capital approach, productive output can be obtained by potential value of production loss due to illness using actual or proxy wage rates (Lensberg et al., 2013). However, there are some disadvantages of taking this approach due to the undervaluing of children and leisure and retirement time. Also, this approach does not capture impacts such as pain, grief etc. except in so far as they affect a person's capacity to work (Lensberg et al., 2013).
An alternative method is based on individual's observed (revealed) or stated willingness to pay. An observed willingness to pay would be based upon what someone pays for a good or service. A stated preference attempts to assess values by asking an individual how much they would be prepared, or willing, to pay in order to obtain the benefits for a good or service (Ryan, 2004). Adopting this approach assumes that individuals seek to maximises their utility and get the most

value for their money when making choices, known as consumer theory. That is an individual's decisions will be rational and reflect the value they place on the intervention, fitting it into their own budget constraints (Liljas & Lindgren, 2001; Ryan, 2004; Lancsar & Louviere, 2008).

The next section provides an overview of common approaches to estimate stated preferences.

2.4 Eliciting stated preferences

Stated preference elicitation methods allow trade-offs to be made two or more alternatives, which reveal the value individuals associate with health programmes or attributes of health services and products. They are often obtained from surveys which can be controlled to determine what preferences are elicited. As such, individuals state their choices in hypothetical markets to help quantify preferences for various attributes of an intervention and/or directly elicit the monetary value (e.g., WTP) of such an intervention. Common methodologies, such as contingent valuation methods (CVM) or discrete choice experiments (DCE) are used to elicit preferences and/or establish a WTP (Ryan et al., 2008; Ali & Ronaldson, 2012).

2.4.1 Contingent valuation

Contingent valuation methods are often survey-based approaches to elicit preferences and establish a WTP for a given intervention/programme. A hypothetical market, where the intervention can be traded, is described. In addition to this, the context in which the good would be provided, and the way it would be financially funded is provided (Klose, 1999).

Theoretically, CVMs are consistent with consumer theory, assuming that an individual is fully informed and utility maximising. As such, stated WTP values would consistently relate to respondents' underlying preferences (Hanley et al., 2001; Ali & Ronaldson, 2012). In essence, a higher WTP value (constrained by a given level of income) suggests a greater benefit is derived by the respondent. Establishing respondent's WTP can be done using either directly obtained- or discrete indicator methods. Directly obtained methods allow measurement of the maximal WTP from respondents, whilst discrete indicator methods allow a range to be established that would include their WTP (Klose 1999; Carson 2000).

Directly obtained approaches, using single questions, are often regarded as the simplest form of CVM. One such approach is the open-ended approach, here participants are asked what the maximum they would be willing to pay for a good, or service (See **Figure 2.1**). Despite being straightforward, this method can be cognitively challenging, especially if respondents have no context of the health disease in question (O'Brien & Gafni, 1996) and may lead to large numbers of non-responses (Klose, 1999). In comparison, a closed question involves respondents being asked if they would be willing to pay a single given total, as shown in **Figure 2.2**. This approach is simple to comprehend; however, it provides much less information on overall WTP and requires a much larger sample size at a group level to estimate WTP than open ended approach. Bias is introduced based on the amount presented (Klose, 1999).

What is the maximum you would be willing to pay for the treatment option described? £______________________

Figure 2.1 Example of direct open-ended question

Would you be willing to pay <u>£100</u> to have the treatment option described? (Please circle) Yes No

Figure 2.2 Example of direct closed question

More advanced CVM have been developed that use a series of closed questions either in a systematic or random fashion to narrow down respondents WTP to a range, which can then be followed up with a further open-ended question.

In summary, CVM elicit WTP, with direct methods may be of greater value, and preferred, as they establish an exact WTP value, rather than a range (Klose, 1999). CVM strengths is they can value welfare implications, in the absence of a market, as well as being overall less complex and burdensome, compared to other stated measures. However, CVM are criticised when used to establish stated WTP as it may be a poor indicator of actual WTP. This is due to the potential that what the respondent states they are willing to pay may not correlate with them actually having enough money to be able to pay for it (Ryan & Watson, 2009). To overcome these issues, an alternative method to eliciting preferences, and WTP, is to undertake a DCE (Ryan et al., 2008) as outlined below.

2.4.2 Discrete choice experiments

DCEs are multi-attribute elicitation experiments which collect individual's stated preferences. DCEs create hypothetical markets, to suit relevant research questions, and involve generation and analysis of choice. Thus, DCEs can elicit preferences and values for goods and/or services for which markets do not exist.

DCEs are often used in two ways: a) elicit preferences, quantifying trade-offs and predicting uptake of good/services to inform policy development and, b) estimating outcomes for inclusion in economic evaluation, such as WTP (Lancsar & Louviere, 2008; Ryan et al., 2008).

DCEs are theoretically underpinned using the characteristics theory of demand (Lancaster, 1966) and random-utility theory (McFadden, 1974). The characteristics theory of demand assumes that the value of a good or service depends on its component attributes or characteristics. By extension, the value placed on a good or service thus depends on the value placed on each component attribute level. It is assumed that individuals will consider all information provided and then select the alternative with the highest utility (Lancaster, 1966).

DCEs have their theoretical foundations in random-utility theory. McFadden (1974) developed this theory into economics by previous work in psychology (Thurstone, 1927). This theory assumes that respondents will make trade-offs between these attributes and levels in a manner that maximises their level of satisfaction based on the level of the attributes in the DCE. These choices can be analysed to estimate the contribution of the attributes/levels to overall utility, using econometric methods which are compatible with random utility theory. McFadden (1974) derived the utility function, which can be shown in the following equation:

$$Ui = Vi + \varepsilon i$$

where,

Ui is the latent utility of an individual for the alternative i

Vi is the systematic component of utility

εi is the random and unobservable component of utility

In a DCE, Vi is defined by the attributes and levels of the alternatives presented. The εi is the random error term which represents the unobservable factors influencing choices made rational individuals. This random error infers that individuals' true utility function cannot be observed; therefore, a probabilistic utility function can be used to estimate the likelihood of an individual choosing a particular alternative from a set of alternatives to choose from (McFadden, 1974). Hence, the probability that an individual will choose alternative i over another alternative j is given by the following equation:

$$P_i = Prob(U_i > U_j) = Prob(V_i + \varepsilon_i > V_j + \varepsilon_j) = Prob(V_i - V_j > \varepsilon_i - \varepsilon_j)$$

This equation shows that the higher the probability of choosing alternative i, the larger the difference in utility between alternative i and alternative j will be. An increase in the quality of attributes in alternative i relative to quality of attributes in alternative j (e.g., the difference in estimated utility between alternatives increases) means the probability of choosing alternative i tends towards 1 (McFadden, 1974).

In a DCE, each respondent makes discrete choices for a set of alternatives presented to them. The proportion of respondents choosing alternative i is interpreted as the probability of choosing alternative i. As such, if the attributes significantly relate to respondents' choices, then the data analysis should determine how the average respondent's utility (or WTP) is affected by a change in the level of attribute (Lancsar & Louviere, 2008; Ryan et al., 2008).

Typically constructed as a survey, DCEs are comprised of several sequential scenarios (choice sets), each containing hypothetical options composed of two or more competing alternatives that vary along several common attributes. For each choice set the respondents chooses between the interventions presented. As alluded to above each alternative can be described by several attributes, with each attribute having numerous levels. One of these attributes may be the price of the alternative or some approximation for it. The levels describe the range of options specifically for that attribute. For example, when choosing between dental care practitioners, a key attribute could be the travel time required to access care, with levels ranging from 10, 30 or 60 minutes. When faced by a given choice set the respondents are assumed to make decisions on which choice set they would choose by looking at the level of each attribute and then making trade-offs between attributes (Lancsar & Louviere, 2008; Ryan et al., 2008).

DCEs can demonstrate the incremental benefit consumers derive from various individual attributes of health care interventions under consideration. As DCEs describe an individual's underlying utility function, based on theory of demand and random-utility theory, compared to just utility maximisation, it can be of more benefit to policymakers. It is possible to predict how choices might differ across interventions (defined in terms of the DCE attributes) and support how services are reconfigured. This requires the assumption that the DCE provides a more realistic version of the choices faced by individuals on a daily basis (Hanley et al., 2001).

Ryan (2004) queried this assumption that individuals behave the same in reality as they state in a hypothetical DCE context. Cookson (2003) agreed, suggesting that individual's responses are far from well-behaved, especially when DCEs are choice tasks which ask about unfamiliar outcomes, small probabilities and long-time-horizons. As such, respondents state their preferences, often on the spot when posed with context-specific stimuli. Responses can be susceptible to psychosocial influences and heuristics, such as generalisation, rules of thumbs and societal norms, often resulting in inaccurate conclusions.

Determining whether a DCE is the most appropriate stated preference approach depends on how much detail is required about the characteristics of the healthcare intervention being valued (Lancsar & Louviere, 2008; Ryan et al., 2008). In healthcare, DCEs have been argued as being more desirable as some individuals may find it difficult, or indeed refuse, to place a monetary value on health when using CVM which thus, increases the incidence of protest zero bids (respondent choose £0 as their value in protest to placing a value on the health state) or implausibly high valuations (Pennington et al., 2017). A cost attribute can be included in a DCE, which can allow a monetary measure of benefit (e.g., WTP) to be estimated indirectly for a unit change in an attribute level rather than explicitly pricing the good. From this it is possible to quantify how individuals' trade-off between them. This is done by establishing the marginal WTP estimates for the attributes in addition to the total WTP for a combination of attribute levels that represent a given intervention of interest (Ryan et al., 2001).

However, there are issues with using DCEs to elicit WTP. Estimating WTP from DCEs may be sensitive to the range of levels presented within the 'cost' attribute, which is a criticism of CVM as described previously. Additionally, if relevant non-cost attributes are not identified, then the element of the total value will be missing. Costs must be realistic to encourage trading, but not so high a respondent is likely to automatically make the choice set based on price alone (Clark et al., 2014). In addition, a 'budget constraint bias' may be introduced, which is when individuals are asked to consider an intervention in isolation, they are often willing to pay far greater that they would be willing to pay when asked to consider the same intervention in relation to a range of other interventions (Cookson, 2003). There remains the concern that participants may be willing to pay a certain amount, but whether they can pay is a different question. The hypothetical nature of DCEs means participants are not duty bound by the choices they make, so an over-estimation of WTP could be obtained (Clark et al., 2014). However, training materials e.g.*,* animated videos, cheap talk scripts etc. have been used to lessen this by attempting to contextualise the hypothetical nature of DCEs (Vass et al., 2020)). Interestingly, it could be assumed that differences in WTP and ability to pay exist between high-income and low-income groups exist. Though, directions of preferences often appear to be similar for both high-income and low-income groups, and therefore it may not seem as problematic as would be expected (Tan et al., 2017). How the attributes are framed in the DCE questionnaire may impact on WTP estimation (as even subtle changes in framing can change responses (Boyce et al., 2014)). Finally, when estimating marginal WTP, from DCEs, it is assumed that the marginal utility of money is constant, with the price attribute being continuous and linear (e.g.*,* total value is equal to the sum of individual parts), however, this may overstate monetary values obtained (Ryan, 2004).

In summary, CVM and DCEs described in the last two sub-sections are well established in adult populations, whereas their use in child and adolescent populations remains limited (Rowen et al., 2020). The next sections will briefly discuss the use of preference elicitation methods in children, followed by whose preferences should be valued – patient or public, child or parent?

2.4.3 Methods for use in children

Eliciting preferences using techniques such as DCEs are theoretically plausible in children and adolescents. An alternative, best-worst scaling (BWS) has gained some traction in eliciting preferences (Stevens, 2015; Rowen et al., 2020).

BWS methods elicit respondents' priorities, by asking participants to identify the best and worst items among sets. In contrast to DCEs, participants evaluate and compare utilities of all the attribute levels on offer and pick the level that is most important (for which they derive the highest utility (best)) and least important (for which the lowest utility (worse)) to them. The respondent has then chosen a pair of attribute levels that they consider to be furthest apart on the latent utility scale. Analytical techniques can then be used to derive utilities at a respondent level or at a sample level (Flynn et al., 2007; Severin et al., 2013).

Rowen et al., (2020) critically reviewed elicitation methods used to obtain valuations from preference-based outcome measures in adolescent populations. They report that techniques such as DCEs, best-worst scaling (BWS), standard gamble etc. have been used to good effect; however, ethical, and practical concerns around the acceptability and appropriateness for using these methods in adolescents do exist. It appears that DCEs (and BWS) are considered more cognitively demanding. However, they often do not require consideration of being dead as is required in a normal standard gamble exercise, which can be conceptually problematic for many dental procedures where death is exceedingly rare.

Irrespective of which method is used, appropriate framing, use of language, number of choice tasks and piloting are all essential to ensure applicability in the child and adolescent population (Rowen et al., 2020). However, no formal comparison was made between age groups in the review (Rowen et al., 2020). Rogers et al., (2021) explored and compared the acceptability of DCEs and BWS approaches in children and young people, using qualitative methodologies. It was reported that BWS tasks were easier to understand and complete than DCEs. This was due to young people reporting difficulty in comprehending the amount of text required to explain and introduce DCEs, in addition to the superior levels of compromise which was required to make between choice sets in completing DCEs. A previous study, using qualitative methodologies also, reported that DCEs were understood and completed by children aged 14 and above; BWS as young as ten (Stevens, 2015).

In summary, preference elicitation tasks with children and adolescents are plausible. However, key uncertainties around comprehension, completion and ethical concerns still exist. Sufficient evidence to suggest that one technique is preferable over the other is lacking (Prosser et al., 2007; Rowen et al., 2020). Therefore, irrespective of which approach is chosen, it is most important to ensure a series of piloting exercises are undertaken to ensure appropriateness for the population being evaluated.

2.4.4 Whose preferences to elicit – patient or public, parent or child?

The choice of whose preferences to obtain is important to consider as different populations will elicit different preferences, when posed the same question (Brazier et al., 2005; Helgesson et al., 2020; Rowen et al., 2020).

Obtaining patient preferences from those with disease experiences may make them better placed to value their own health condition compared to the general public trying to imagine them (Brazier et al., 2005). These values will be based on current experience and previous personal experiences, in addition to experiences of another health state similar to the one being valued, or experience based on relatives' or other peoples' ill-health (Cubi-Molla et al., 2018). The public are not necessarily as well informed, nor do they have accurate understanding of the impact of ill-health, compared to patients which may lead to uninformed preferences (Karimi et al., 2017; Helgesson et al., 2020). In general, patients tend to give lower utility scores for their health states compared to those given for the same health states by the general population, due to misinterpreting the health state or incorporating differing levels of disease, based on their experiences, into their valuations (Brazier et al., 2005; McTaggart-Cowan, 2011; Helgesson et al., 2020). Impacts are likely to be exaggerated as biased judgements are likely if only patient views are obtained.

It has been argued that public preferences may be more appropriate to obtain, especially when the healthcare interventions under scrutiny are publicly funded, as is the case in the UK. A representative sample of the public, which may include patients with the disease, are more likely to provide an unbiased judgement of values, compared to patients alone, which is important when re-allocation of resources occurs on the back of the evaluation (Brazier et al., 2005; Helgesson et al., 2020).

The debate on whose preferences to obtain continues, with the choice often depending on the perspective and decision-making context of the underlying evaluation (Brazier et al., 2005). NICE (2022a) recommend that public preferences should be used. However, when conditions, or indeed evaluations, are pertinent to a child and adolescent's health, consideration must be given eliciting preferences directly from young people. Yet, tools such as the EQ-5D-Y are only considered valid for young people aged 12 and over and currently for the UK there is no actual value set that can be used to score EQ-5D-Y survey responses. Therefore, thought should be given to parents eliciting their preferences as a proxy instead (Prosser et al., 2007; Rowen et al., 2020; Powell et al., 2021).

It could be argued that when children and adolescents experience the health state being evaluated, eliciting their preferences is an important consideration, if not more important than their parent or caregiver as they have the lived experience. However, consideration as to whether children and adolescents can understand, and complete preference elicitation tasks is required. It is noted that age is most likely to impact completion as younger children aged around 7–10 years may not fully understand the tasks so may not be able to express a 'rational' choice (Prosser et al., 2007; Stevens, 2015; Rowen et al., 2020; Powell et al., 2021). Other factors such as educational ability, experience of ill health and socio-demographic characteristics influence capability of completing tasks, thus, some younger children may be more able to undertake these tasks, whilst some older children may not (Rowen et al., 2020). In such circumstances when a child or adolescent is unable to complete the tasks, then they may require the support of an adult to make them. Consideration however must be given to the extent to which the adult is likely to influence health valuations (Prosser et al., 2007; Powell et al., 2021). Proxy versions of the EQ-5D-Y exist, and either ask the proxy's opinion directly, or how they think their child/adolescent would value their quality of life, it if they were able to communicate themselves (EuroQol Research Foundation, 2022).

Parental proxies may be unavoidable when children are too young to comprehend, or complete elicitation tasks. When interpreting such valuations, it is likely these will be made not just on the child undergoing that healthcare intervention, but other competing priorities such as their own parental HRQoL, the influence of other children in the family, parental guilt, or other beliefs.

Furthermore, they may misinterpret the impact of a health condition on the child/young person, as well as biasing preferences towards their own viewpoints (Prosser et al., 2007; Powell et al., 2021). Thus, using parents as proxies may cause an over- or under-estimation of health state values, or uninformed preferences. In contrast, it could be argued that adult preferences should be obtained, on the basis that they fund the vast majority of health care through direct and indirect taxation and hence, their preferences should be used to determine re-allocation of healthcare resources (NICE, 2022a). Eliciting preferences from a representative sample of the population may overcome some of the issues raised by proxies. Although, any population sample will still include a high proportion of parents. It is safe to assume that most parents included would be less likely to exhibit some of the potential biases than a sample of parents of affected children (Prosser et al., 2007; Rowen et al., 2020).

Although not an economic evaluation, findings from a recent study demonstrated that end of life preferences of adolescents with cancer were mainly congruent with their parents' preferences (Jacobs et al., 2015). Whose preferences to elicit will very much depend on the evaluation being undertaken and implications of such decisions. The final sub-section will briefly review the use of DCEs (and WTP) in oral health research.

2.4.5 DCEs in dentistry

Despite DCEs being widely adopted in health (de Bekker-Grob et al., 2012; Clark et al., 2014), there use in dentistry remains limited (Barber et al., 2018).

Barber et al., (2018) systematically reviewed the literature and found only 12 examples where DCEs had been used in dentistry, with the first being published in 1999 and the last in 2015, based on the search conducted in 2017. These studies focussed on service delivery, treatment, or oral health states from the perspective of the patients, dentists, or the public. Description of the methods used were comprehensive across all studies, despite a lack of uniformity in experimental design. Across the 12 studies, a range of two to seven attributes were chosen, with little justification given for the number selected. The most common attribute was related to cost (92%, n=11). It appears that determining what attributes to include was variable across studies, with little information provided. In most cases it was poorly done, with only 3 out of 12 studies directly involving participants from the target population in the identification stage. In regards to overall quality of reporting, the studies were compared to the International Society for Pharmacoeconomics and Outcomes Research (ISPOR) checklist (Bridges et al., 2011) with a range of 53% to 100% compliance was noted. The authors concluded that reporting of design features was variable across the studies, although, the rationale underpinning decision on task construction and survey presentation and details on selection and recruitment of the sample were often missing, the latter being of importance to ensure transparency and support external validity between the study sample and target population.

Therefore, despite there being few DCEs in dentistry, what there is, is of relatively good quality, although clearer justifications and better reporting are necessary (Barber et al., 2018). There may be a lack of awareness in dentistry, which may explain the scarcity, however, further studies (Barber et al., 2019; Boyers et al., 2021; Fenton et al., 2022) have been published since this review.

In summary, it appears that clinical measures and measures such as QALY may fail to capture impacts of care or, in the case of QALYs, there is no suitable method suitable for use in dentistry is available to value health. In contrast, CVM and DCEs can be used to elicit preferences, and value health, by estimating a WTP.

The next section will now look at how economic evaluations can be used to extrapolate outcomes beyond a single study, or over a longer time horizon, using decision analytical modelling technique.

2.5 Economic Evaluations – extrapolating outcomes

Economic evaluations in healthcare often take one of two stylised forms. They can be conducted in conjunction with an empirical study such as a randomised controlled trial (RCT), obtaining cost and benefit data as part of the trial. Alternatively, they can be conducted as an isolated decision analytical model, by either basing it on a single study, with a set of assumptions, or more commonly, incorporating synthesised data from a wide range of sources (Sculpher et al., 2006; Petrou & Gray, 2011a, 2011b). Most frequently, economic evaluations include elements of both.

2.5.1 Trial-based economic evaluations

When conducted alongside RCTs, economic evaluations can use individual patient data to estimate cost effectiveness. Researchers using this method tend to focus on estimating cost and effect differences, before assessing the likelihood that an intervention is cost effective, or not, rather than testing a particular hypothesis (Petrou & Gray, 2011a).

There are advantages to using trial-based economic evaluations. Firstly, they provide an opportunity to produce reliable estimates of cost effectiveness at a low marginal cost above the costs of obtaining effectiveness estimates. Secondly, using individual patient level data allows a wide range of statistical and econometric techniques to be used, including exploring sub-group variations, assuming the sub-group sample is large enough. Finally, relevant costs and outcome data can be directly collected as part of the trial, which can be challenge in models where some data may not exist (Petrou & Gray, 2011a).

One of the main limitations of trial-based economic evaluations is the truncated time horizon of the analysis. The usual short-term follow-up of patients in a trial means long-term outcomes that extend beyond the trial period will not be captured. Extrapolation of this data over an extended period could be undertaken using survival analysis models, such as the Cox proportional hazard model or Weibull model. However, such methods come with a degree of uncertainty as it involves assuming the behaviour of the quantities of interest beyond the time horizon are supported by the clinical evidence from the original trial (Bojke et al., 2017).

Other limitations of trial-based economic evaluations also exist. Basing clinical- or policy decisions on just one trial, may ignore relevant evidence, may lead to inappropriate implementation of healthcare interventions, or allocation of resources. Additionally, a single trial is unlikely to compare all relevant decision options for a particular disease. This can be demonstrated by a recent cost-effectiveness analysis run alongside a trial (Homer et al., 2020) which compared only three alternative dental caries management strategies for primary (baby) teeth. Although these were felt to be the most relevant at the time of the study design, other strategies existed but could not sensibly be included as trial comparators. Finally, the external validity of a single empirical study may limit the generalisability to different settings or countries as some of the trial data cannot readily be adapted to another setting, although the study may be repeated. This is not just an issue with trial-based economic evaluations, as a model may also not be transferable; however, it is easier to adapt to reflect local care practices, costs and preference values (Petrou & Gray, 2011a).

Despite these limitations, trial-based economic evaluations still have an important role in producing reliable estimates of cost effectiveness. Considering the limitations of economic evaluations based on a single empirical study, though, decision analytic modelling could be used as a complementary framework for economic evaluations.

2.5.2 Decision analytical modelling

There are several advantages to economic evaluations based on decision analytical models (DAMs). They allow expected costs and benefits, of decision options under uncertainty, to be explored beyond the trial-follow up period, which is conventionally three years. They are often more reflective of reality, that being several comparators, can be included in a model. Furthermore, rather than focussing on trial-based individual patient level data, DAMs synthesise evidence from range of sources, including expert opinion where evidence does not exist, across different patient groups and health care settings (Petrou & Gray, 2011b; Drummond et al., 2015).

Despite models being able to synthesise information from a wide range of sources, it is not possible to include every piece of information, as this increases the complexity of the model, to a point where it is unmanageable. Therefore, decisions must be made on which health states and pathways to include, and what assumptions must be made (Buxton et al., 1997; Caro et al., 2012).

Making assumptions in a model raises concerns (Buxton et al., 1997; Caro et al., 2012). Firstly, a model is only as good as the data that is input into it, that being if the costs or benefit data is of poor quality, or too heterogeneous, then the model is likely to incorrectly estimate the economic benefit of the intervention being evaluated. Secondly, models cannot reflect complete reality as they are by definition models and therefore involve inevitable simplifications and assumptions to reduce the computational burden. This makes conceptualising the model challenging, especially if similar models do not exist. However, if these decision and assumptions are transparent and clearly reported, then decision-makers can account for this in making clinical and policy decisions. Thirdly, extrapolating results beyond a clinical trial is also challenging (Bojke et al., 2017), and even more so when incorporating results from various trials, often with different follow-up periods, variations in outcomes and other sources of heterogeneity. Finally, when data is used in the model, even that from trials, then biases (e.g., attrition bias, selection bias etc.) in collecting this data will naturally be transferred into the model. Biases can be addressed using sensitivity analyses, however, knowing identifying biases is challenging (Briggs et al., 2006; Drummond et al., 2015). Additionally, a value of information analysis could be completed. This approach, alongside an economic evaluation, can value the expected gain by assessing the cost effectiveness of alternative research projects that could done in the future, and help reduce areas of uncertainty within the model (Wilson, 2015).

Deciding which model structure to choose depends on the decision problem and healthcare context being evaluated. Decision makers need to identify the perspective, time horizon, relevant outcome measures, and scope/boundaries of the model. Policy implications will greatly depend on explicit and implicit assumptions of the model chosen (Brennan et al., 2006; Petrou & Gray, 2011b).

Model structures are broadly categorised into aggregate cohort models or individual patient level models. An overview of decision analytical models is shown in Table 2.3.

		Cohort level		Individual level	
		Expected value/ continuous state/ deterministic	*Markovian/ discrete state/ stochastic*	*Markovian /discrete state/ individuals*	*Non-Markovian/discrete state/individuals*
No interaction Allowed	*Untimed*	Decision tree rollback	Simulated decision tree	Individual sampling model; Simulated patient-level decision tree	
	Timed	Markov Model (evaluated deterministically)	Simulated Markov model	Individual sampling model; Simulated patient-level Markov model	
Interaction allowed	*Discrete time*	Systems dynamics (finite difference equations)	Discrete time Markov chain model	Discrete time individual event history model	Discrete individual simulation
	Continuous time	Systems dynamics (ordinal differential equations)	Continuous time Markov chain model	Continuous time individual event history model	Continuous individual simulation

Table 2.3 Taxonomy of model structures (adapted from (Brennan et al., 2006; Jin et al., 2021))

A cohort model examines costs and benefits of an average patient from a population undergoing different events. The cohort modelled is identical, however, cohort characteristics for example, gender, stages of natural history of disease etc. can be chosen probabilistically, this then becomes an individual patient level model. In an individual patient level models, the progress of individual patients is followed, accounting for variation between patients, over time. The most common model structures used, and those that will be discussed in this section, are decision trees, Markov and individual patient level micro-simulation models (Brennan et al., 2006; Petrou & Gray, 2011b).

Decision Trees

The simplest model is the decision tree (**See Figure 2.3**). Alternative options are represented by a series of pathways or branches. The pathways that follow each option represent a series of logically ordered sequence of alternative events. Each alternative, at each chance node, in the logical order must be mutually exclusive, and the probabilities associated with these options must equal exactly one. The terminal end point of each pathway has a cost and benefits attached to it. Once the probabilities and values have been entered, the tree is 'folded-back' by summing up the costs and effects weighted against probabilities of each pathway, allowing the expected values for each original option to be calculated (Petrou & Gray, 2011b).

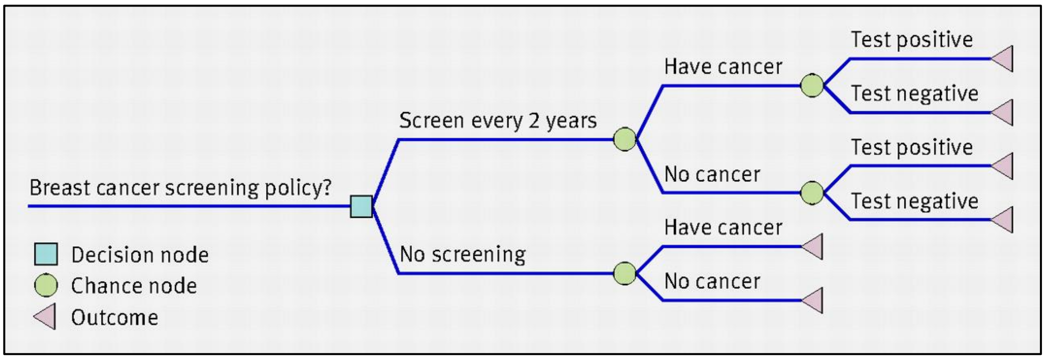
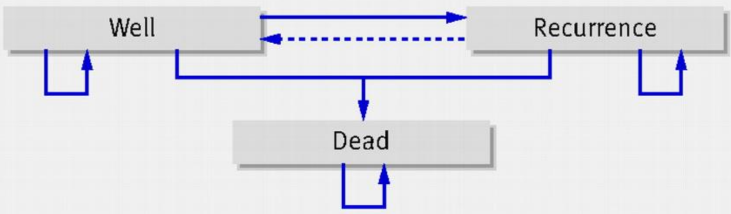


Figure 2.3 Example of a decision tree (reproduced from (Petrou & Gray, 2011b))

Decision trees can be simplistic and transparent, regularly being used for economic evaluations that have a short-time horizon but can be complex and may be combined with other model types to deal with longer term horizons. However, decision trees are not best suited to deal with time dependent elements of an evaluation. Furthermore, they are not ideal for chronic diseases that have recurring events e.g., exacerbations of chronic obstructive pulmonary disease, as the tree will become overly complex with several lengthy pathways (Briggs et al., 2006; Petrou & Gray, 2011b).

Markov Models

Markov models allows an easier representation of complex and repeated events over time. Any disease or care process to be modelled can be split into a set of discrete states each of which has an equal duration. The main premise of Markov models is that patients reside in any given health or treatment state, relevant to the health context, for a given period (**See Figure 2.4**).



State at time t	**State at t + 1**		
	Well	**Recurrence**	**Dead**
Well	1-(0.3 + 0.1)	0.3	0.1
Recurrence	0.1	1-(0.1 + 0.2)	0.2
Dead	0	0	1

Figure 2.4 Example of a Markov model (reproduced from ((Petrou & Gray, 2011b))

Transitions can be made to other health states after a discrete time interval, or cycle, throughout the chosen time-horizon. The cycle length should be short enough to represent the intervention being assessed. The definition of each health state, and the duration of the cycles, will be determined by the decision problem undergoing evaluation. The probability of staying in a particular health state or transitioning to another is determined by transition probabilities. Costs and benefits are assigned to each health state in the model, as a point estimate for each state per cycle. In most situations, Markov models simulate how a hypothetical cohort of patients transition through the model over time, allowing an estimate of expected costs and outcomes to be obtained. In each cycle, the costs, and benefits across health states, weighted by the proportion of the cohort expected to be in each state, are summed up. Then the totals for each cycle are calculated together. One key consideration is to ensure that models extending beyond one year, costs and benefits are discounted to consistently to generate the present value of those future costs and benefits (Briggs et al., 2006; Petrou & Gray, 2011b; Caro et al., 2012)

Markov models have one main limitation; that is the assumption that the transition probabilities depend only on the current health state and are independent of historical experience (the Markovian assumption). As such there is no 'memory' in the model. This results in all patients being treated as the same, irrespective of the time spent in each state, or indeed which states they have been in previously. This lack of memory can be overcome by introducing states that a person can only enter for one cycle or a series of states that must be completed in a fixed sequence and/or including time dependences. However, their addition will increase the computational burden of the model (Briggs et al., 2006; Petrou & Gray, 2011b).

Individual patient micro-simulation models

Individual patient micro-simulation models are far more flexible than Markov models as they enable the progression of individuals, rather than a hypothetical cohort, through the model to be calculated. Therefore, estimates of the mean costs and benefits for a group of people are estimated by calculating the costs and benefits of each person making up that group. Furthermore, using this model permits the progression of individuals that are different from one another, and whose future progression through the model can be determined by their previous history and how they have journeyed through the model up to that point. As such, a set number of hypothetical patients will be simulated through the model. Like Markov models, health states, cycle length and transition probabilities will be determined by the decision problem being evaluated. Costs and benefits, which may vary according to the individual, will be assigned to each health state, for each cycle, and discounted appropriately. The sum of the costs, and benefits are calculated for everyone, before providing a mean estimate to permit analysis (Briggs et al., 2006; Petrou & Gray, 2011b; Caro et al., 2012).

Patient level simulation models have limitations. They are more complex and require greater amounts of data. Furthermore, these models are computationally far more demanding, which makes assessing the uncertainty in the model harder and much more time consuming. Finally, interaction between individuals is not permitted in these models. The journey for each patient going through the model has no effect on any of the other patients e.g., if some people need specialist dental treatment it assumes that one person accessing this sort of care will not prevent or delay another person accessing this care. However, they are not ideal when there might be capacity constraints, e.g., using the previous example, specialist dental treatment does not have a limitless supply (Briggs et al., 2006). This restriction can be overcome with other forms of model (Jin et al., 2021) but these are not considered in this thesis.

In summary, trial-based economic evaluations use individual patient data to estimate cost effectiveness. They use the power of a trial to provide, for the data collected an unbiased comparison. However, individual randomised trials do not always provide a sufficient basis to inform regulatory and allocation decisions as they may not represent the totality of relevant available evidence and often cannot compare all available relevant alternatives. In comparison, DAM theoretically enable all relevant decision options to be compared although there may be some practical limitations with this. It is possible for them to include a wide range of costs and benefits, from various sources, and synthesised within a model framework to generate the overall effectiveness of a given healthcare intervention. However, models can be hard to conceptualise, they can be computationally burdensome. In addition, the data required to populate may be either non-existent, be of poor quality, too heterogenous, or fraught with bias inherited from the studies that generated that data, which will reduce both the internal and external validity of the findings.

The use of these methods is commonplace in healthcare, however, their use in dentistry remains limited. The following section will provide an overview of published dental economic evaluations.

2.6 Economic evaluations – review of published dental economic evaluations

Economic evaluations remain limited in a dental context (Tonmukayakul et al., 2015; Eow et al., 2019). This section will describe the evidence that has been systematically reviewed for dentistry in general followed by that specifically for child oral health.

2.6.1 Economic evaluation in dentistry

Several systematic reviews of economic evaluations in dentistry exist. The majority focus on specific areas of dental disease (Källestål et al., 2003; Mariño et al., 2013; Qu et al., 2019; Rogers et al., 2019; Anopa et al., 2020; Taylor et al., 2021; Mariño et al., 2020, 2020) or methodologies (Tan et al., 2017; Hettiarachchi et al., 2018; Barber et al., 2018), rather than all dental publications (Tonmukayakul et al., 2015).

Tonmukayakul et al. (2015) reported that as of 2013, 114 peer reviewed studies were identified that examined costs and/or outcomes in dentistry. Of these, 79 were full economic evaluations, that is a comparison of costs and benefits of two or more alternative healthcare interventions/programmes. A more recent, and less comprehensive, scoping review conducted by Eow et al. (2019) included 91 full economic evaluations, as of the end of 2017. Using recent guidance on critically appraising systematic reviews with costs and cost-effectiveness outcomes (Mandrik et al., 2021), it was apparent that despite having similar inclusion criteria, Tonmukayakul et al. (2015) used a more detailed search strategy and included a more comprehensive database search, that included MEDLINE, Cochrane Library and the NHS Economic Evaluation Database. Whereas, Eow et al. (2019) only searched MEDLINE, and this most likely explains why only 12 more full economic evaluations were identified in the four years between reviews. Despite this, both reviews reported that economic evaluations are becoming more common practice in dentistry, with a significant growth in the number of publications after the year 2000 - approximately threefold compared to those before 2000 (Tonmukayakul et al., 2015; Eow et al., 2019).

In the Tonmukayakul et al. (2015) review, CEAs were the most common form of economic evaluation (n=63). Most of the publications were population-based studies, assessing cost-effectiveness alongside a cross-sectional study or clinical trial. Most studies were based on dental caries prevention, with the remaining being spread over numerous other dental conditions. The next most common were cost description (n=17) and cost-outcome description studies (n=15), which at best could be described as 'partial economic evaluation (Drummond et al., 2015)'. The authors report that some cost-description and cost-outcome description studies were labelled as either CEA or CBA, however, on review they were re-classified as not being full economic evaluations. Only eight CBAs and eight CUAs were included in this review, which is understandable given the pertinent challenges conducting these in dentistry.

In this review, most of the CBAs (n=7) assessed the value of water fluoridation, with the benefits being presented in monetary terms (Tonmukayakul et al., 2015). Three studies (Griffin et al., 2001; O'Connell et al., 2005; Campain et al., 2010) expressed outcomes as net savings, as derived from the difference between water fluoridation programme costs and costs of treating of dental caries and productivity losses averted. Two studies (Doessel, 1985; O'Rourke et al., 1988) merely attached costs to reductions in DMFT/dmfs scores, and compared them to capital costs of water fluoridation programmes, which makes these cost-analyses not CBAs, as the comparison is between the costs incurred only. Similarly, the remaining two labelled CBAs were in fact with a CEA (number of averted newly decayed tooth surface in a permanent tooth) (Wright et al., 2001) and a cost-analysis of costs of caries treatments linked to fluoride prevention regimes for two time horizons, annual and lifetime (Splieth & Flessa, 2008). Interestingly, none of the studies in this review used a DCE to elicit a WTP, however, a recently published study has done this (Boyers et al., 2021) and is likely to be included in any updated review.

Four of the CUAs reported in Tonmukayakul et al., (2015) used QALYs as their outcome measure (Tsevat et al., 1989; Cunningham et al., 2003; Speight et al., 2006; Dedhia et al., 2011). Two studies (Ciketic et al., 2010; Cobiac & Vos, 2012) used disability-adjusted life year (DALYs) whereas one study (Bhuridej et al., 2007) used quality adjusted tooth year (QATY). Finally, one study (Heydecke et al., 2005) appears to have been mislabelled as a CUA as they report an ICER based on a change in the OHIP-20 (oral health impact profile - a quality of life measure) scores and in-fact should be labelled as a CEA.

Quality assessment of the included studies was performed using the Drummond's 10-item checklist for papers conducting full economic evaluations (Higgins et al., 2022) and the consolidated health economic evaluation reporting standards (CHEERS) checklist to assess the reporting quality of economic evaluations (Husereau et al., 2013). Common methodological limitations (e.g. absence of sensitivity analysis, discounting, and insufficient information on how costs and outcomes were measured and valued) were apparent (Tonmukayakul et al., 2015).

In summary, the Tonmukayakul et al., (2015) review is the most comprehensive systematic review of economic evaluations in dentistry to date. It suggested that economic evaluations are lacking. However, this review did not report, or extract data, to specifically detail whether any of these economic evaluations were pertinent to children, or whether they involved children as a sub-set. This next sub-section summarises two systematic reviews on the quality and scope of economic evaluations in child oral health (Rogers et al., 2019), and those that use decision modelling studies, with horizons extending into adulthood (Taylor et al., 2021)

2.6.2 Economic evaluations in child oral health

Rogers et al. (2019) was the first systematic review to evaluate the scope and quality of economic evaluations in child oral health. In addition, this review also considered the extent of children's involvement in these evaluations.

Of the 46 included studies, 38 were CEAs which mostly focussed on the prevention or management of dental caries. Two studies were CUAs, with one using QATYs (Bhuridej et al., 2007) and the other QALYs (Koh et al., 2015). QALYs were derived using the Child Health Utility (CHU-9D), a paediatric quality of life multi-attribute instrument, which is validated to provide a utility score used to measure QALYs (Stevens, 2012). One study described using a CBA framework (Kowash et al., 2006), however, it was in fact a form of cost-analysis, as the cost-benefit ratio presented was the cost of implementing a dental health education programme divided by the savings in costs of treatment that would be avoided if the carious tooth didn't require treatment. The value of the health intervention, as elicited by a WTP, was not undertaken.
A major flaw noted within these included studies was the lack of involvement of children in obtaining their valuation of dental health. Only one study used the CHU-9D (Koh et al., 2015), with the authors of this study reporting the measure was slightly adapted to allow parents to proxy where appropriate. However, the CHU-9D appears to be insensitive to assessing changes in dental caries experience, especially in populations where the caries experience, either based on clinical progression or symptoms, is not that substantial (Foster Page et al., 2015). Nonetheless, in not asking children directly, it is possible that issues relating to oral health which would be of direct importance to children themselves, are overlooked (Rogers et al., 2019). The omission of involving children in these evaluations is unfortunate, however, this is likely to change as the involvement of children in research, audit and service evaluation increases in future studies (Gilchrist et al., 2013).

Rogers et al. (2019) excluded modelling studies that extended beyond the age of 18. This was deliberate to focus on the benefits gained during childhood by interventions. However, these benefits will often extend beyond this time point.

As such, Taylor et al. (2021) undertook a systematic review to consider the scope and quality of decision modelling studies within the field of child oral health that had time-horizons extending beyond the age of 18. Nine studies were included in their narrative synthesis, with most of them focussing on the prevention or management of dental caries. Eight were CEAs and one was a cost-analysis. No CUAs or CBAs were included, despite a very broad inclusion criterion. Most studies used individual patient-level microsimulation models; however, justification of model type was often omitted in the reporting. Tooth retention years or caries increment were mainly used as outcome measures, both of which appear relevant for modelling studies, as they can capture long-term outcomes associated with the interventions. Two thirds of the studies used a lifetime horizon, whilst other used pre-determined age ranges (nine years, ten years, sixty-three and half years) with little justification provided (Taylor et al., 2021).

Although inherent challenges exist in using CUAs in dentistry (Hettiarachchi et al., 2018), the use of preference elicitation, namely DCEs, (Barber et al., 2018) and WTP tasks (Tan et al., 2017) is increasing, which might result in an increase in CBAs in any future reviews. What is apparent from these reviews (Rogers et al., 2019; Taylor et al., 2021) is the paucity of economic evaluations, such as CBAs, that use analytical modelling techniques to understand the longer term impacts of dental interventions undertaken in children.

2.7 Conclusion

This chapter has explored the general principles of economic evaluations, valuing health, preference elicitation and decision analytical modelling approaches. This was followed by a brief overview of economic evaluations used in dentistry and child-oral health. Chapters 5 and 6 will discuss in more depth the health economic methodologies used in this thesis.

Chapter 3. Thesis overview

3.1 Addressing the lack of evidence for managing compromised first permanent molar teeth - purpose of this thesis

As explored in Chapter 1, a lack of evidence and direction as to whether cFPM should be restored or removed in childhood exists. Furthermore, direct, and indirect impacts are apparent. Thus, better understanding of decisions about how to manage these teeth, in a UK context, is clearly important for patients, parents, dental professionals and the commissioning of dental services. As demonstrated in Chapter 2, addressing a paucity of evidence needs to not just focus on clinical efficacy from a trial, but can and should, include views and opinions, preferences for certain treatments and the longer-term implications of decisions made in childhood. The three complementary studies used in this thesis will aim to answer some of these queries:

- Chapter 4 will aim to establish adolescents' and adults' views and experiences around managing cFPM using qualitative methodologies
- Chapter 5 will aim to elicit the public's preferences for managing cFPM, including determining societal willingness to pay (WTP), using a discrete choice experiment
- Chapter 6 will aim to determine the most efficient way of managing cFPM over the lifetime of a patient, using a decision analytical model, by comparing initial management strategies
- Chapter 7 will include a discussion and bring together the findings of the 3 studies to create some recommendations

Chapter 4. Adolescents' and adults' views and experiences around managing cFPM

4.1 Introduction

To understand managing cFPM better, exploring adolescents’ and adults’ views and experiences of managing these teeth is required. In this thesis, a qualitative study was undertaken to contextualise and make sense of these views and experiences. In addition, early analyses helped inform attributes that were used in the public DCE (more in-depth discussion can be found in Chapter 5).

Qualitative research is a naturalistic, inquisitive approach that explores how people's behaviour and opinions are shaped within their natural setting. It focuses on the ‘what’, ‘why’ and ‘how’ questions of healthcare. In comparison, quantitative research methods, such as randomised control trials, often focus on the effectiveness and efficacy of a healthcare processes or interventions. Depending on the research questions, these methods can be used in isolation, or conjunction with one another (Berwick, 2008; Ritchie et al., 2014; Busetto et al., 2020).

In managing cFPM, establishing views and experiences are important as they provide complementary understanding to mutually informative data established through preference elicitation (see Chapter 5) and modelling of the natural history of the disease (see Chapter6). In section 4.2, the philosophical assumptions will be briefly discussed. Qualitative methodology will be discussed in Section 4.3. Section 4.4 will explore data collection and analytical methods with a focus on the approaches chosen for this thesis. Section 4.5 will provide the aim of the qualitative component of this thesis. Section 4.6 will describe the methods. Finally, section 4.7 will discuss the results and discussions, in addition to the strengths and limitations, of this chapter. These findings will be discussed in combination with the findings of the DCE (see Chapter 5) and decision analytical model (see Chapter 6), in Chapter 7, to consider the management of cFPM in a wider context.

4.2 Philosophical assumptions – ontology & epistemology

All methodological approaches are informed by the researcher’s ontological and epistemological beliefs. These will now be explored in turn.

Ontology

Ontology relates to the nature of reality and what there is to know about the world. This is an attempt to understand whether or not social reality exists independently of human interpretations or preconceptions (Ritchie et al., 2014).

The two main contrasting ontological positions are realism and idealism. Realism puts forward that an external reality exists, independent of human beliefs or understandings. That is, there is a difference between the way the world is, and the meaning of that world held by individuals (Bhaskar, 1982; Hammersley, 1998; Madill et al., 2000; Fletcher, 2017). Idealism suggests the opposite whereby no external reality exists independent of human beliefs or understandings. In other words, idealism suggests that social reality is only comprehensible through the human mind and socially constructed meanings (Engel & Kuzel, 1992; Madill et al., 2000; Barnett-Page & Thomas, 2009)

Several nuanced perspectives exist between these two extremes. One perspective, known as ‘critical’ or ‘subtle’ realism is the viewpoint which will be adopted in the thesis. Subtle realism proposes that external reality is intricate and layered, and this approach attempts to capture that reality, in all its complexity and depth. This viewpoint aligns with realism whilst recognising the critical importance researchers’ own subjective understandings are likely to have on the interpretation of the research (Madill et al., 2000; Fletcher, 2017).

Epistemology

Epistemology relates to the way we seek to know about the world, focussing mainly on how we can learn about reality and the foundations of one’s knowledge. One main epistemological issue is the way knowledge is acquired (Carson et al., 2001). It can be classified as inductive (‘bottom-up’), built through observations of the world or deductive (‘top-down), testing observations against logically derived hypotheses. It is often thought that qualitative research is an ‘inductive’ process (Barnett-Page & Thomas, 2009; Ritchie et al., 2014), although others argue that it cannot be purely inductive or deductive (Blaikie, 2007; Charmaz & Belgrave, 2015). That is, data generated and analysed through qualitative observations must have been based on assumptions relating to the field of work that have deductively been derived from previous work (Blaikie, 2007).

The two main epistemological stances that exist are positivism and interpretivism. Positivists believe that there is one objective reality, and this can be measured and understood whilst being unaffected by the researcher. Approaches taken by positivists are often deductive and associated with quantitative research in testing hypotheses (Silverman, 2013). In contrast, interpretivism acknowledges that people are different, and likely to experience the world in different ways. Thus, knowledge is primarily obtained inductively by exploring the social world of the people being studied and forms the basis of qualitative research. Furthermore, interpretivism acknowledge the influence researchers have on the research being carried out (Carson et al., 2001; Ritchie et al., 2014). For this component of the thesis, an interpretive stance is adopted.

Another epistemological issue which must be considered relates to the relationship between the researcher and the research participants. In qualitative research, the researcher is the research instrument, and the participants may be affected or unaffected by the researcher's behaviour when being studied. It is therefore suggested that researchers adopt a reflexive position, and transparently acknowledge that their assumptions and biases are likely to affect the findings (Arber, 2006; Ritchie et al., 2014; Hiller & Vears, 2016). Matters are further complicated when a qualitative researcher has an additional professional role, as is the case in this thesis. In these cases, the researcher must consider how their dual role affects participant consent, data collection, and analysis. Furthermore, considerations on the impact on authenticity, trust and overall transparency are required when deciding whether to disclose clinician status to participants or not (Arber, 2006; Geddis-Regan et al., 2022).

4.3 Methodology

In general, qualitative research will naturally be focused on either theoretical or applied contexts. Theoretical qualitative research is formed by generating thinking within a specific subject, and in some cases attempting to create new theories. In contrast, applied approaches use knowledge acquired to support the understanding of a specific issue(s) (Green & Thorogood, 2018; Ritchie et al., 2014). Several methodological approaches exist (Barbour, 2014; Ritchie et al., 2014). The remainder of this section will briefly explore two alternative conventional approaches, grounded theory, and interpretative phenomenological analysis, before focussing on the chosen approach for this thesis, generic qualitative research.

Grounded theory enables the researcher to generate a substantive theory grounded in empirical data (Glaser & Strauss, 1967; Walker & Myrick, 2006). As an approach, grounded theory is well suited to investigate social processes that have little prior research, or where previous research lacks breadth and/or depth (Milliken & Schreiber, 2012; Charmaz & Belgrave, 2015). However, it has been criticised as it produces a large amount of data, that can be difficult to manage, in addition to failing to recognise how the researcher is embedded within data creation and interpretation (Charmaz & Belgrave, 2015).

As an alternative, interpretative phenomenological analysis attempts to provide detailed accounts of personal lived experiences, through exploration of participants' experiences, understandings, perceptions and views (Brocki & Wearden, 2006; Smith et al., 2009) It addresses a respondent's account of the processes rather than formulating objective accounts (Smith et al., 2009; Tuffour, 2017). However, interpretative phenomenological analysis is mostly descriptive, and insufficiently interpretative, as it understands the lived experiences but fails to explain why they occur (Brocki & Wearden, 2006; Hefferon & Gil-Rodriguez, 2011). A sense of vagueness and lack of standardisation are major criticisms of this approach (Tuffour, 2017).

Committing to one ideology may not be suitable for all subject matters, or indeed research questions (Caelli et al., 2003; Ritchie et al., 2014). Persistent tension between a need for both methodological flexibility and structure, in following traditional approaches, such as grounded theory and phenomenology has been suggested (Holloway & Todres, 2003). A generic qualitative research approach can mitigate such tensions by permitting flexibility whilst encompassing the structure of more conventional approaches (Patton, 1990). This methodological approach has been used in this thesis and will now be described in more detail.

Generic qualitative research is a broad, flexible, pragmatic and adaptable approach that allows the correct method to be chosen for the research question (Patton, 1990, 2002). It permits the combination of data collection methods which fit with researchers' epistemological stance, discipline, and particular research questions (Caelli et al., 2003; Merriam & Tisdell, 2009; Kahlke, 2014; Ritchie et al., 2014). It is not guided by an established set of philosophic assumptions, which are associated with traditional qualitative approaches (Caelli et al., 2003; Kahlke, 2014). It is best used when the research question tries to:

"*...simply seek to discover and understand a phenomenon, a process, or the perspectives and worldviews of the people involved*." (Merriam, 1998, p.11)

As an approach, it is gaining traction in healthcare research (Cooper & Endacott, 2007; Auta et al., 2017; Gazarian et al., 2022) and offers the opportunity to utilise and combine, what each established approach has to offer, especially in cases when the research question may not fit within the confines of a single traditional approach. Concerns about it lacking rigour, and being atheoretical, have been disputed in the literature (Kahlke, 2014); however, achieving quality is more likely to be associated with choosing the right data collection/analytical methods, rather than using methods that are bound to specific traditional and theoretical approaches (Ritchie et al., 2014).

This approach is highly interpretative, as it focuses on how people interpret their experiences, construct their worlds, and what meaning they attribute to their experiences (Merriam, 2002; Lim, 2011). As such, highly inductive techniques, such as the use of open codes, categories, constant-comparison and/or thematic analysis are all feasible common with this approach (Lim, 2011). Furthermore, choosing the correct data collection method has to fit the research question, whilst allowing adequate interpretation (Caelli et al., 2003; Merriam & Tisdell, 2009).

Sections 4.2 briefly covered conventional philosophical assumptions – ontology & epistemology – that underpin qualitative research. This has been followed by a brief overview of a qualitative methods, with a focus on a generic qualitative research approach, in Section 4.3. The next section will explore data collection and analytical methods that are used in conjunction with a generic qualitative research approach, in more depth.

4.4 Qualitative data collection and analytical methods

Choosing the correct data collection and analytical method, whilst following a generic research approach has to be considered (Caelli et al., 2003; Merriam & Tisdell, 2009). This section will explore data collection, focussing mainly on interviewing, and the challenges pertinent to this thesis. This will be followed by a discussion on analytical methods, focussing mainly on thematic analysis.

4.4.1 Data collection

Interviews and focus groups are the most widely used methods in healthcare (Barbour, 2014; Ritchie et al., 2014; Busetto et al., 2020) and can be used whilst following a generic research approach (Caelli et al., 2003; Merriam & Tisdell, 2009).

Focus groups are the method of choice when the overall purpose of research is to study group norms, meanings or processes (Bloor et al., 2001). Participants who may or may not know each other are encouraged to explain, comment, and share their views/experiences collectively to generate ideas and knowledge as a group. In comparison, qualitative interviews have defined topics/questions to cover but are flexible enough to allow issues raised by respondents to be explored. Interviews are often shaped by the way the respondent replies. They permit a conversation with a purpose, so that each participant's views and experiences can be explored and made sense of (Kahn & Cannell, 1957; Green & Thorogood, 2018). The two most common types of qualitative interviews are semi-structured and in-depth.

Semi-structured interviews mainly consist of several open-ended questions as part of a topic guide. Using such questions enables data generation that is naturally evolving, which allows unexpected new areas not covered in the topic guide to be explored as the study progresses. However, semi-structured interviews may only reveal superficial knowledge, rather than delving deep into the psychological and sociological reasons behind respondent's answers. Furthermore, they can be time-consuming, requiring greater numbers of participants (in comparison to in-depth interviews) to draw significant conclusions (Barbour, 2014; Ritchie et al., 2014).

In comparison, in-depth interviews only ask one or two questions, in an attempt to elicit far more focussed detail on an overall topic. Thereafter, questions are purely based on what the respondents have said, most often to seek clarification, but if required can be used to probe for details to expand, explore, challenge, and understand underlying values, views, and experiences. In-depth interviews provide more natural, rich, and detailed data on a topic. However, they are the most time consuming and are prone to bias as certain probing questions might influence data generation (Barbour, 2014; Ritchie et al., 2014).

Despite their subtle differences, it is likely that in-depth/semi-structured approaches will be used interchangeably within an interview, but to what extent will depend on the topic being discussed and individual being interviewed. Unpicking the construct of an interview has long been discussed. Cicourel (1964) originally explained that interviewing was merely an artful social interaction, and to be successful as a method to collect information, it relied upon the speaker to utilise everyday conversational approaches. This theory was expanded more recently, with Rapley (2001) suggesting that both the interviewer and interviewee co-construct a series of accounts during an interview, rather than it being a purely a direct report of attitudes or perceptions of just the interviewee. Seale (2004) similarly felt that interview data generated was either reflective of a reality that was jointly constructed by the interviewee and interviewer.

The location of an interview has to be considered as this is likely to affect the content of the interview (Elwood & Martin, 2000; Barbour, 2014). This could be more problematic for adolescents, with it being recommended that interviews should be conducted outdoors, or in a familiar environment to the young person, such as their home (Einarsdóttir, 2007; Alderson & Morrow, 2020). Location may be of less importance for interviews given that online interviews in healthcare are becoming more attractive (Davies et al., 2020). A recent scoping review, comparing face-to-face with online interviews of health and illness experiences, reported that online approaches were able to recruit the required sample but may risk less contextual information being obtained (Davies et al., 2020). Relational satisfaction was lower with online interviews, although, all included studies were pre-COVID-19 (Davies et al., 2020). However, online methods are not inferior as they are able to support rigorous, qualitative research in a virtual format, that is more equitable (Roberts et al., 2021).

Finally, children have the right to participate in decisions affecting their lives and communicate their own views. As defined in Article 12 of the UN convention on the rights of the child, state that any child who is capable of forming his or her own view should have the right to express these views freely on all matters affecting the child (United Nations, 1989). Therefore, when enquiring about how cFPM should be managed in children, using qualitative interviews, it is not only right, but important, to obtain the views of the child, rather than using their parents as a proxy. However, it could be anticipated that the interview process could be complex and challenging for some adolescents. In these cases, the presence of a parent or guardian may help. However, the interviewer must be aware of the inherently greater power that adults hold in their relationships with children, especially younger children (Alderson & Morrow, 2020), which could result in children being told what to say, whether they have an opinion or not (Einarsdóttir, 2007; Ponizovsky-Bergelson et al., 2019).

4.4.2 Data Analysis

Choosing the correct method of analysis, whilst following a generic research approach, requires consideration (Caelli et al., 2003; Merriam & Tisdell, 2009). In general, analysis of qualitative data seeks to organise and condense data into themes, or categories, which can be fed into descriptions, patterns, or theories (Ritchie et al., 2014; Green & Thorogood, 2018). One way of undertaking this is to label data that is interesting and merits exploration, whilst seeking contrasting similarities and differences within these sections of data (Rapley, 2013). This labelled data is then compared with other data and placed into themes, based on similarity. Labelling of data is an iterative, inductive, and reductive process (Patton, 2002; Rapley, 2013). A common analytical approach used in generic qualitative research, thematic analysis, uses this process and will now be described. (Caelli et al., 2003; Kahlke, 2014).

Thematic analysis (TA) can be thought of as a component of other types of qualitative data analysis (Ryan & Bernard, 2000); however, Braun and Clark (2006) argue that it is a widely used and flexible analytical process, that sits as an analytical method in its own right. TA is independent of theory and epistemology, being compatible with essentialist, constructionist and contextualist paradigms. An essentialist method reports the experiences, meaning and realities of participants, whist a constructionist paradigm examines the ways in which events, realities, experiences are the effects of a range of discourses operating within society. A contextualist method acknowledges the ways participants make meaning of their experiences, and how broader social contexts impact on such meanings, while retaining focus with the realms of ‘reality’ (Braun & Clarke, 2006). TA develops themes that run through the data, in an attempt to understand patterns (Braun & Clarke, 2006).

In using TA, it is important not to just label the data, but more to understand the process. The steps involved in TA, as described by Braun and Clarke (2006) are outlined in **Table 4.1**.

Phase	Description of the Process
1. Familiarising yourself with your data	Reading and re-reading the data, making note of initial ideas and interesting things
2. Generating initial codes	Coding interesting features of the data, systematically, across the entire data set collating data relevant to each code.
3. Searching for themes	Collating codes into potential themes, gathering all data relevant to each potential theme. identifying patterns and considering relationships between codes so that common themes may be identified
4. Reviewing themes	Checking that the themes work in relation to the coded extracts and the entire data set
5. Defining and naming themes	Clear definitions and names for each theme is undertaken. Ongoing analysis, refining the specifics of each theme, and the overall story that is being told
6. Producing the report	A selection of vivid and compelling extract examples, relating back research question and literature, to help produced a report of the analysis.

Table 4.1 Phases of Thematic Analysis (adapted from (Braun & Clarke, 2006))

A major criticism of TA is that coding can be too superficial, especially when trying to establish initial themes. In attempts to try to make sense of the data, it is possible to miss the depth and meaning within (Braun & Clarke, 2006). As a method to partially overcome this, a 'constant comparative method' can be adopted (Glaser, 1965). This approach allows theoretical notions and concepts highlighted in earlier interviews to be re-integrated into future interviews for further exploration, understanding and review (Glaser, 1965). This ensure that anticipated themes (those that the researcher thinks are important) and emergent themes (those raised by the participants) are fully explored.

This section has summarised the methodology and the remainder of the chapter will cover the empirical qualitative work.

4.5 Aim

To establish adolescents' and adults' views and experiences around managing cFPM.

4.6 Methods

This section will report ethical considerations, sample, recruitment and conduct of the interviews, detailing pertinent factors, and their relevance. Reporting in this chapter is in line with consolidated criteria for reporting qualitative research (COREQ) recommendations and the checklist is included as **Appendix A** (Tong et al., 2007).

4.6.1 Ethical approval and considerations

Ethical Approval

The conduct of this project was carried out in accordance with the ethical principles set out in the Declaration of Helsinki (World Medical Association, 2013). A favourable ethical opinion was obtained from North of Scotland Research Ethics Service (REC Reference 20/NS/0124, Date: 22/10/2020) prior to commencement of the study (see **Appendix B**). As the project progressed, one non-substantial amendment was made to add participant identification centres to support recruitment of the qualitative interviews. This, unfortunately, led to a considerable delay but approval was obtained (see **Appendix C**). Local research and development, and Caldicott approvals were obtained from the Newcastle Upon Tyne Hospital NHS Foundation Trust.

Ethical considerations

Several ethical issues were anticipated with this qualitative study. These mainly related to consenting and interviewing children aged 12-16 and managing any safeguarding concerns that may be raised. These ethical concerns were mitigated by development of adult (**see Appendix D**) and adolescent's (see **Appendix E**) participant information leaflets (PIL) and assent/consent forms. All documents used for adolescents were developed in conjunction with a young person's patient representative panel to make it age appropriate.

It was expected that young adolescents (>12 years old) would have the capacity to consent themselves. However, after discussion with the ethics committee, it was agreed that the assent should be taken from the young person and consent from the parent. After recruitment, adolescents were invited to do the interview by themselves or have a parent/guardian sit beside them. However, if a parent/guardian was present, it was stressed that the questions needed to be mainly answered by the adolescent as it was their views that were important. Written informed consent was obtained. However, due to COVID-19, these were emailed to participants, with an electronically signed copy being emailed back to the chief investigator (CI).

Any safeguarding concerns identified or raised were managed in line with the Safeguarding Policy and Procedures as set out by Newcastle upon Tyne NHS Foundation trust with whom the CI and main supervisor have honorary contracts.

As a token of generosity, research participants (adults and adolescents) who completed a qualitative interview were given a £20 gift voucher. This approach was approved by the ethics committee.

4.6.2 The influence of the interviewer on the interview

The possible influence of the CI's professional background and gender on the interview process was considered (Geddis-Regan et al., 2022). This study focusses on the experiences of managing cFPM, for which the CI provides care to both children and adults as a specialist paediatric dental trainee and out of hours emergency dentist. The CI took the approach to present himself as a researcher to participants, assuming respondents may want to talk more freely about managing cFPM without being distracted by the knowledge they were talking to a clinician. However, the CI was willing to disclose being a dentist, if asked, as proactively withholding professional status would not be ethically justifiable.

4.6.3 Sample

Purposive sampling was used in this study. This enabled participants to be recruited with a range of characteristics that helped understand the central ideas of managing cFPM, rather than using a representative sample of the general population (Suri, 2011). The CI was able to actively choose participants, targeting those with characteristics of interest (age, gender, oral health experiences and oral health treatment experiences) in later interviews, if they had not been included to that point (see Table 4.2).

Characteristics	Variation within characteristics
Age	Adolescent Interview: 12 – 16 years old Adult interview: 17 - 65 years old
Gender	As described by the participant
Oral health experiences	Those who access and do not access dental care regularly, as described by the participant
Oral health treatment experiences	Those who have had experience of a filling, an extraction, both, and no treatment on first permanent molar

Table 4.2 Characteristics of selected sample

Attempts were made to ensure all characteristics, and their variations, were included in the sample. Thus, maximising the diversity of the data likely to be generated to allow the research question to be answered.

4.6.4 Recruitment

Initially, adults and adolescents were recruited from outpatient clinics from one tertiary dental hospital and an out-of-hours emergency clinic, both based in the north of England. Recruitment posters, placed in the respective waiting rooms, were used to aid recruitment. Potential participants were identified by the dental staff working in each clinic, and if they met the inclusion criteria, were given a PIL to read whilst attending the radiology department. Upon return, if the participant wished to take part, the staff member completed a consent-to-contact form and shared it with the CI. Once recruited, adults, and parents of the adolescents, were contacted via email to arrange a convenient date and time to be interviewed. Upon completion, electronic PILs were sent, along with a consent form, which was to be completed and electronically sent back to the CI, prior to the interview.

As a result of the COVID-19 pandemic, recruitment of participants was slow. As an initial additional strategy to increase recruitment, three primary dental care clinics were set up to act as participation identification centres. Due to primary care services re-establishing post COVID-19, these clinics only identified a few potential participants. Therefore, a further addition to the recruitment strategy was undertaken and direct recruitment completed by the CI in his clinical role. This approach enabled adequate recruitment. Recruitment was ceased when data saturation was reached, that is, stopping when further interviewing generated no additional new information (Saunders et al., 2018).

4.6.5 Interviews

Semi-structured interviews were undertaken and conducted by the CI only. The CI had relevant training and previous experience of conducting interviews. Two adolescent interviews were completed in the presence of an adult.

Location

Due to the concerns of COVID-19, and varying government restrictions on travel, all interviews were completed virtually using the online platform Zoom.

In this study, online interviews relied on the participant having access to a smart phone, tablet, laptop, or computer, given that sharing of an image as part of the interview was required. This precluded a telephone interview. All online interviews were conducted with the participants in their own home, without the CI being there.

Topic Guide

A topic guide was initially developed, with questions being derived based on literature with an overall focus on the overall aims of this thesis. This was then discussed with the supervisory team. The initial version of the topic guide (**see Appendix F**) included two sections:

- 'Broad Areas for Questions' - focussing on dental attendance, general past dental experiences, and expectations of what to receive when visiting the dentist
- 'Focussed Areas for Questions' - focussing on identifying the first permanent molar, experiences specifically with these teeth, for those who had treatment and those who had not.

The topic guide had several iterations and developed over the interviews, based on areas of interest generated using a constant comparative analytical approach (see section 4.4.2). The final version (**see Appendix G**) included sections:

- Section 1 - attendance, general experiences, and expectations.

- Section 2 - identifying the first permanent molar and exploring experiences specifically with these teeth
- Section 3a & 3b – How decisions were made for those who have had treatment and have not had treatment
- Section 4 (for adult interviews only) – How to manage these teeth in your child or a hypothetical child

To ensure the interview focussed on the first permanent molar, participants were asked to identify the FPM using a staged questioning approach. First, participants were asked a direct question - "Are you aware that humans have two sets of teeth?" – which opened discussions about tooth identification. A diagram to support the identification of the FPM was shared (see **Figure 4.1**). At this point, participants were asked a) Which colour represents the molar teeth? And b) Which letter was the first permanent molar? This method helped to ensure the participants knew the focus of the interview was on the first permanent molar. Participants were advised that all future questions and discussions were about these teeth only. To support this, **Figure 4.1** remained on the screen throughout and was referred to when required.

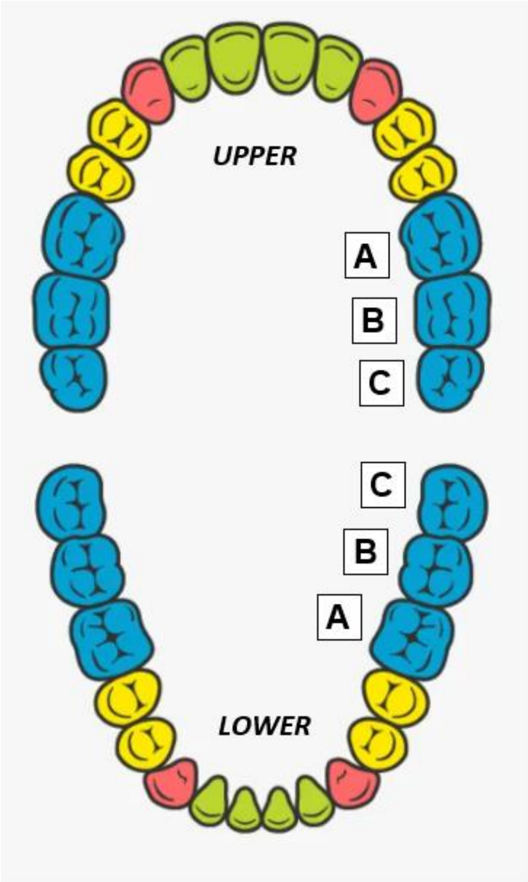


Figure 4.1 Diagram to support the identification of the first permanent molar for use in the interviews

As the interviews with adults progressed, it became apparent including a section on “managing your child” was required to establish the role the parent had in the decision making of their child’s tooth. In some cases, the parent’s children had not had treatment of this first permanent molar, or their children were not old enough to have these teeth present yet and others did not have children. In these circumstances, the concept of a ‘hypothetical’ 9-year-old child was used for this section. Including this hypothetical ‘vignette’ allowed for exploration of the future parent and non-parents views.

Pilot

Prior to commencing the main qualitative study, two pilot interviews (one adult and one adolescent) were conducted using two volunteers: one familiar and one unfamiliar to the CI. The main purpose was to test the suitability of the topic guide. None of the questions were seen as potentially sensitive. The topic guide was re-organised to improve the natural flow of conversation.

Additionally, these pilot interviews allowed identification of practical issues with the proposed study set up as well as allowing interviewing techniques to be practiced and developed.

4.6.6 Data handling

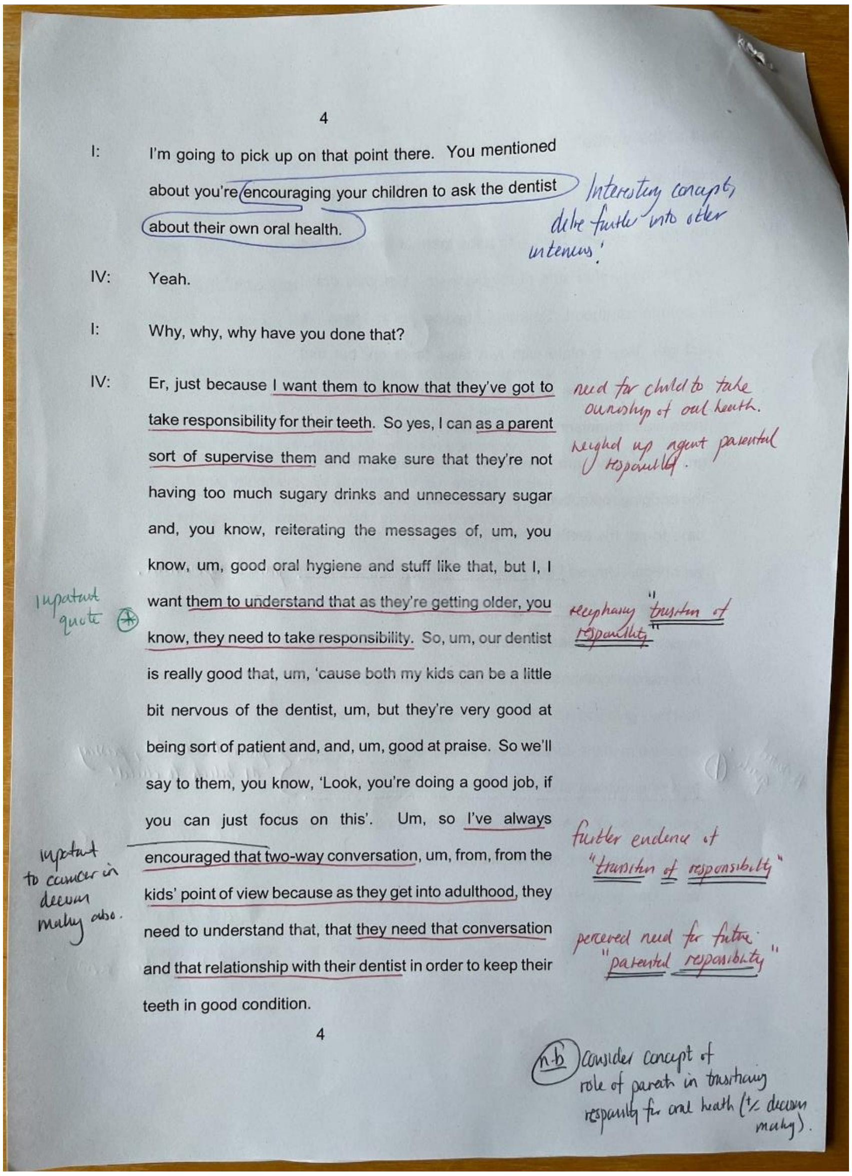
The audio from the semi-structured interviews were recorded using a digital recorder. Field notes were completed in addition to support these recordings. This file was uploaded to a secure server before being sent for professional online transcription. Interviews were transcribed verbatim and anonymised. The transcribed data were entered into NVivo version 12© (QSR International Pty Ltd 2012), and to assure accuracy, transcripts were re-read whilst listening to the sound file. The digital recordings were stored on a Newcastle University password-protected computer.

4.6.7 Data analysis

Thematic analysis (Braun & Clarke, 2006) was used to analyse the data, adopting a constant comparative approach (Glaser, 1965).

After careful examination of the transcripts, labelling sections of dialogue with interesting ideas was undertaken (Stage 1). Labelling data starts with the indexing of sections, which permits retrieval later. These pieces of data (initial codes) were organised in a meaningful and systematic way to reduce large volumes of data into small chunks of meaning (Stage 2). NVivo™ (version 1.7) (QSR International, 2021) was initially used to support analyses of seven adolescents and three adult transcripts. However, the CI felt more distanced from the data. A more helpful approach to allow understanding of the data better was to revert to a more traditional paper-based approach, as shown in **Figure 4.2**. This approach was used for the remaining adolescent and adult interview transcripts.

These processes allowed themes to be searched (Stage 3) and reviewed (Stage 4). To mitigate superficial coding of data initially and support the process of developing the themes, regular discussion within the study team, and separately with the qualitative expert on the supervisory team was undertaken. Prior to refining the themes (stage 5), initial themes that had been searched and reviewed during earlier analyses were re-analysed to ensure the validity of their analyses. Continually reviewing the dataset ensured that no potentially significant information was overlooked. This process was continued until new interviews failed to produce any new themes, at which point the data was considered saturated. Finally, themes were refined and named, in discussion with the supervisory team.



4

I: I'm going to pick up on that point there. You mentioned about you're encouraging your children to ask the dentist about their own oral health.

IV: Yeah.

I: Why, why, why have you done that?

IV: Er, just because I want them to know that they've got to take responsibility for their teeth. So yes, I can as a parent sort of supervise them and make sure that they're not having too much sugary drinks and unnecessary sugar and, you know, reiterating the messages of, um, you know, um, good oral hygiene and stuff like that, but I, I want them to understand that as they're getting older, you know, they need to take responsibility. So, um, our dentist is really good that, um, 'cause both my kids can be a little bit nervous of the dentist, um, but they're very good at being sort of patient and, and, um, good at praise. So we'll say to them, you know, 'Look, you're doing a good job, if you can just focus on this'. Um, so I've always encouraged that two-way conversation, um, from, from the kids' point of view because as they get into adulthood, they need to understand that, that they need that conversation and that relationship with their dentist in order to keep their teeth in good condition.

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Figure 4.2 Excerpt of data analysis from transcript of interview with adult_6

4.7 Results

This section will report the findings from adolescents' and adults' interviews, the aim of which were to understand and explore the views and experiences around managing cFPM.

In total, nine adolescent and thirteen adult interviews were undertaken. Adolescents' characteristics are described in **Table 4.3**. Interviews ranged from 22min 14secs to 39mins 9secs. Eight of the nine adolescents were able to identify the cFPM when **Figure 4.1** was shared with them.

Participant	Age	Gender	Oral Health Experiences	Treatment Experience
Adolescent 1	13	Male	Regular	Nil
Adolescent 2	12	Female	Regular	Nil
Adolescent 3	16	Male	Regular	Fill & Extract
Adolescent 4	12	Male	Regular	Fill
Adolescent 5	13	Male	Irregular	Extract
Adolescent 6	15	Female	Regular	Nil
Adolescent 7	14	Female	Regular	Fill
Adolescent 8	14	Male	Regular	Nil
Adolescent 9	12	Female	Regular	Fill & Extract

Table 4.3 Characteristics of adolescents (n=9)

Adults' characteristics are described in **Table 4.4**. Interviews ranged in duration from 27min 36secs to 43mins 17secs. Twelve out of the thirteen adults were able to identify the cFPM when **Figure 4.1** was shared with them.

Participant	Age	Gender	Oral Health Experiences	Treatment Experience
Adult 1	17	Male	Regular	Nil
Adult 2	25	Male	Regular	Fill
Adult 3	32	Female	Irregular	Extraction/Fill
Adult 4	20	Male	Irregular	Nil
Adult 5	43	Male	Regular	Extraction
Adult 6	37	Male	Regular	Fill
Adult 7	39	Female	Regular	Extraction
Adult 8	24	Female	Regular	Nil
Adult 9	48	Female	Regular	Extraction/Fill
Adult 10	53	Female	Regular	Fill
Adult 11	46	Male	Regular	Extraction/Fill
Adult 12	47	Male	Regular	Extraction/Fill
Adult 13	37	Female	Regular	Extraction/Fill

Table 4.4 Characteristics of adults (n=13)

In the remainder of the results, quotations will be labelled with the group (adolescent or adult), their ID number and then pertinent characteristics (gender etc.). The results will be presented alongside the context in terms of other literature, combining aspects of the results and discussion sections.

4.7.1 Adolescents

Three major themes were generated from adolescents' interviews: i) influencing factors; ii) long-term considerations; iii) shared decision making. The themes and sub-themes are shown in **Table 4.5**.

Final Themes (and sub-themes)		
INFLUENCING FACTORS • Professional • External • Personal • Acquired vs Developmental	LONG-TERM CONSIDERATIONS • Preference for retention • Recurrent pain leads to extraction	SHARED DECISION-MAKING • Provision of Information • Asserting Autonomy • Trust Professional Opinion

Table 4.5 Themes (and sub-themes) of adolescent interviews (n=9)

Theme 1: Influencing factors

The data showed that adolescents are influenced by several factors when deciding how they want to manage cFPM. All adolescents interviewed explained that they always considered their dentist's opinion when deciding how they should manage a cFPM:

> *"I'd probably want my dentist's advice, probably ask them which one they thought was better, and then probably make a decision off that…I feel like a dentist would know a lot more about like what I should do for my teeth"*
>
> Adolescent 6 (15, F, Regular, Nil)

> *"I think they [dentist] would like tell us which one is the best option for us…It's like if you say you want a certain option, then they [dentist] say it's not the best option and if you keep going for that it could make you worse, because you do not know what's best for you. So, what I am saying is I would kind of just go with what they say"*
>
> Adolescent 5 (13, M, Irregular, Extract)

For some adolescents, a dental professional's opinion outweighed their parents:

> *"They know more than your parents about teeth, so I'd listen more to what they suggested about how to treat these teeth…"*
>
> Adolescent 2 (12, F, Regular, Nil)

Whereas, for others, parental opinion was equivalent to that of a dentist:

> *"Yeah, I would ask my parents for their advice, and listen to the what the dentist has to say…"*
>
> Adolescent 7 (14, F, Regular, Fill)

These data support findings of other studies which have demonstrated the importance of the patient-dentist relationship, and the impact that it has on decision-making (Muirhead et al., 2014; Waylen et al., 2015; Armfield et al., 2017). An adolescent that has a dentist who is friendly, explains options and involves them in the decision-making process is more likely to feel satisfied (Gilchrist et al., 2013; Waylen et al., 2015; Birkeland et al., 2022).

It is known that due to their knowledge and experience healthcare professionals are likely to hold a position of power in any patient-healthcare professional relationship (Isaacs, 2019). This perception that a dentist knows how best to treat these teeth come across strongly in these interviews and explains why adolescents valued their opinions so much when formulating their decision.

Despite not always taking their parent's advice when deciding how to manage cFPM, it appears that their parent's treatment experiences and philosophies influenced their decision-making:

> *"…my parents have always taught me to like keep my teeth, just like they have…"*
>
> Adolescent 4 (12, M, Regular, Fill)

> *"I've had an extraction because and a filling because my parents have had both before"*

Adolescent 3 (16, M, Regular, Fill/Extract)

When it came to peer influence, some adolescents suggested that what their friends said would not affect their decision:

> *"If I had to, I'd ask my friends what they [have] had done. I'm sure if I had to have a filling or extraction they wouldn't be fussed or mean to me about it, as if they did, then they're not your friends, are they?"*

Adolescent 1 (13, M, Regular, Nil)

It is known that children develop most of their values and behaviours from their parents (Knafo & Galansky, 2008), although this influence lessens with age (Ward et al., 2014). The sample interviewed were aged between 12 and 16 years. It was originally suggested that by the age of 15, most adolescents would have the cognitive competence to make their own decisions, and not require input from their parents (Mann et al., 1989). However, more recent reviews have suggested that children much younger have the cognition to derive a decision without any parental influence (Boyer, 2006; Coughlin, 2018). As such, these subconscious thoughts are likely to form part of their processing (Erb et al., 2002) when deciding how to manage cFPM. This is likely to be the same for factors such as functional impact, psychosocial impact, anxiety, and aesthetic implications, despite not specifically featuring in the data.

It is known that besides family, peers are also known to influence adolescents' behaviour and decision-making (Ragelienė & Grønhøj, 2020). However, adolescents interviewed did not refer to peer influence influencing their decisions regarding managing a CFPM. It could be that dental treatments, in general, are not hugely influenced by adolescent peers. Alternatively, it could be that management of cFPM have limited aesthetic implications. It is known that adolescent peers are more likely to make social judgements based on having ideal anterior aesthetics (Henson et al., 2011). Therefore, it could be argued that adolescents' decision-making is less susceptible to peer influence as either restoring or extracting the cFPM are unlikely alter anterior aesthetics.
In addition to professional, parent and peer influences, prior treatment experiences helped shape what adolescents would want done with their cFPM, now or in the future:

"Erm, I've had them [filling and extraction] both once done before and its erm, like I haven't died from it…yes, because erm, like you never know what will happen when you do it the first time but once you've done it the first time, you've actually done it, like a roller coaster, when you do not know what's going to happen, you do not know what the tracks are but when you're on it, you already know like, done it before and then when you do it again, you're like, oh, well I know where it's going and what speed it is."

Adolescent 3 (16, M, Regular, Fill/Extract)

"Having a tooth yanked was canny [very/really] sore, but I'd still have it done again if that what I chose to have done"

Adolescent 5 (13, M, Irregular, Extract)

It appears that adolescents' past experiences of managing cFPM and knowledge of dental procedures influence their decision-making ability. Indeed, having prior experiences is likely to permit sufficient understanding of the issues when faced with making a decision, both in health (Coughlin, 2018) and dentistry (Adewumi et al., 2001; Gilchrist et al., 2015).

Another factor was the physical size of the defect, with the consensus being that the larger the defect, the more complex the work to save it could be, so removal may be best:

"…if it's bigger then it will take a bit more time to fill and could hurt more, so take it out, as if it was a bit bigger then it would be harder to treat…"

Adolescent 8 (14, M, Regular, Nil)

"…if it was really small, the filling yeah but if it was really big, I'd probably want it out as it'll be harder…"

Adolescent 1 (13, M, Regular, Nil)

"…if it's just this massive hole as in like in my mouth, there's no point them just saving that massive hole, may as well just get rid of it."

Adolescent 7 (14, F, Regular, Fill)

Although, defining a threshold of how big the defect was appeared to be a more challenging concept:

> *"…it is hard to say what the size of the hole would be to tip me into having the tooth removed…"*
>
> Adolescent 3 (16, M, Regular, Fill/Extract)

> *"It just depends on how like really big it is, but what I mean by big I do not really know…"*
>
> Adolescent 4 (12, M, Regular, Fill)

It also appeared that for some adolescents, if the cFPM was due to a developmental condition, rather than an acquired condition, then there was a preference to try and retain this tooth:

> *"I'd not be as worried because I hadn't caused it, but this would make me want to try and save it, as I was born with it…"*
>
> Adolescent 2 (12, F, Regular, Nil)

Some felt that it did not matter what caused it, or how much of the tooth was affected, they would manage them the same way:

> *"…no matter whether the tooth's, er, is decayed…or whether because I've got some sort of developmental problem, I'd want to hold on to it…"*
>
> Adolescent 8 (14, M, Regular, Nil)

> *"…it doesn't really matter whether I caused this, or [if] I was born with it, if the hole is too big I'd have it extracted."*
>
> Adolescent 5 (13, M, Irregular, Extract)

One interviewee thought that a tooth with a developmental defect meant it was poorer quality, and should always be removed:

"…if it did not develop and was going to form in a different way then it is not really worth trying to salvage the tooth with that. I would say get rid of it"

Adolescent 6 (15, F, Regular, Nil)

Children are able to act as agents of their own health (Davó-Blanes & La Parra, 2013) and competently able to discuss their experiences of dental disease (Parekh et al., 2014; Gilchrist et al., 2015). It appears that adolescents in this study have divergent views on how they would want to manage cFPM with a developmental condition. Previous qualitative work on children with amelogenesis imperfecta, another developmental condition, showed they had an awareness of the colour, shape and size of affected teeth (Parekh et al., 2014). Similar awareness has been noted in children with MIH (Hasmun et al., 2018), however, this study focussed on anterior teeth only.

It is interesting that adolescents perceive the size of the defect to be a major contributor to deciding how to manage these cFPM. The general concept that the larger the defect, the more complex and harder to treat was apparent. Restoring a tooth that requires a small filling one surface is no more challenging than one that require a large filling on one surface. Therefore, further work is required to ascertain whether adolescents correlate defect size as the amount of surface that is affected, or the actual three-dimensional size of the cavity.

Theme 2: Long-term considerations

Most adolescents interviewed preferred retention of the tooth over removal, when asked about initially managing a cFPM:

"If asked to choose between filling or extraction, I would prefer to keep hold of the tooth and retain it…"

Adolescent 8 (14, M, Regular, Nil)

"I'd have the filling as I wouldn't want to like go through the long process of taking it out…at least I'd still have my tooth there."

Adolescent 2 (12, F, Regular, Nil)

However, if the tooth was to become sore later in life, they expressed a preference for extraction:

"…if I get the filling, and if it's gonna keep giving us pain there's no point keeping the filling in. So, I may as well just get it taken out…"

Adolescent 1 (13, M, Regular, Nil)

"Erm if it was a tooth with a filling that was painful when I chewed. So, if that would kind of not go away with another filling in, then I would definitely want an extraction…"

Adolescent 3 (16, M, Regular, Fill/Extract)

This remained true, even if the option of endodontic management was offered:

I'm not sure I'd want to have the nerve removed, even if I could hold onto the tooth that's there…"

Adolescent 4 (12, M, Regular, Fill)

"…I think if the root canal treatment and kind of keeping up the maintenance of the tooth had to happen a lot of times over kind of multiple years then I would say it's not worth it. Erm just get the extraction."

Adolescent 9 (12, F, Regular, Fill/Extract)

When adolescents are initially faced with having to decide whether to fill or extract a cFPM, the filling is the preferred option. One explanation could be that previous experience of fillings, or observation of their parents having these treatments drove this this decision. An alternative could be that the asymptomatic nature of the teeth prompted a restorative decision. To date, several cross-sectional studies have focussed on how clinician's decide how to manage cFPM (Kopperud et al., 2016; Taylor et al., 2019; Alkadhimi et al., 2022; Wall & Leith, 2020), whereas no studies have actually asked children themselves. Another explanation could be that the sample were more regular attenders, and therefore were more likely to choose the conservative option. Although based in adults, a recent study showed that the odds (OR 1.07 95%CI: 1.04-1.07) of visiting the dentist for problematic teeth increased as the OHRQoL reduced (Gaewkhiew et al., 2017).

If the cFPM were to be become painful, then on the whole, adolescents preferred this to be extracted (Versloot et al. 2004; Alohali et al. 2019). A possible explanation for this might be that pain is something adolescents' want to avoid. The uncertainty of complete pain removal and risk of future pain are disadvantages of a root canal treatment. Therefore, choosing a treatment option, such as extraction, that avoids prolonged, or indeed future pain is something that adolescents potentially value (Versloot et al., 2004; Alohali et al., 2019).

Theme 3: Shared decision-making

Asserting autonomy of their healthcare decisions was important to all adolescents interviewed:

> *"…what happens with my teeth is my choice, but how often I get to make that choice I am not sure"*
>
> Adolescent 2 (12, F, Regular, Nil)
>
> *"I agreed with erm the extractions and the braces…I felt like I had a choice in the matter, rather than my parents…"*
>
> Adolescent 3 (16, M, Regular, Fill/Extract)

Involving adolescents in a shared decision-making is vitally important (Coyne & Harder, 2011; NICE, 2021), as adolescents report high satisfaction when involved (Wogden et al., 2019; Davison et al., 2021). Adolescents can be seen as competent social actors, who are actively involved in shaping their own social worlds, rather than incomplete adults who do not have a say (Gilchrist et al., 2013). In addition, the United Nations Conventions on the Rights of the Child 1989 (United Nations, 1989) and Children Act 1989 (Children Act, 1989) both advocate that children, including adolescents, have the right to assert their autonomy and have their views heard as part of a shared-decision making dynamic. However, some children prefer not to engage in this dynamic (Kelly et al., 2017). It may be the case that always involving children in their treatment decisions may not ideal. However, it would be prudent not to overlook adolescents' opinions, as ultimately it is them who have to endure the dental procedure, and the associated consequences. As a minimum, acknowledging opinions is important as it is accepted as a key developmental step in the transition into adulthood (Colver et al. 2020).

However, the frequency or how this is experienced in practice is difficult to ascertain. Only one adolescent interviewed reported having total autonomy in deciding about what treatment they would have (for brace treatment). Some interviewees reported varying levels of involvement in deciding, whilst most adolescents reported not being involved at all. This suggests a need to educate parents and clinicians on the importance of shared decision-making in dentistry.

In this shared decision-making idiom, adolescents certainly felt that professionals had a role to play. There was a sense that their opinion was trusted:

> *"Because they're like professional and they're probably like doing it for the good of me."*
>
> Adolescent 8 (14, M, Regular, Nil)

> *"Yeah, cos what's the point in going if you're not gonna trust them[dentist]?"*
>
> Adolescent 9 (12, F, Regular, Fill/Extract)

Certainly, trust in a professional's opinion was very much associated with how this opinion, or indeed information as part of the process of SDM, was provided:

> *"I think it's important to have information and like they should have like a pros and cons list, basically, [ok] of what would happen and what could happen erm in the future."*
>
> Adolescent 9 (12, F, Regular, Fill/Extract)

> *"I think lay out all of – it's always good to lay out all of the options. Erm explain the pros and cons of both maybe explain the process which would happen in both cases. At the end it might help them feel more decided to say, 'Okay, this is definitely the wrong option, this is definitely the right option."*
>
> Adolescent 6 (15, F, Regular, Nil)

Trusting professionals' opinions was evidently important to adolescents. Favourable experiences are noted when trusting relationships are formed with healthcare professionals (Davison et al., 2021). Personable, wise, sincere and relatable were characteristics that describe what makes a trusting healthcare professional (Davison et al., 2021). In adolescents' opinions, it seems that being confidential, not withholding information, and engaging in small talk to show concern are all essential to gain trust (Klostermann et al., 2005). A recent Australian study demonstrated that in adults, the majority of people appeared to exhibit trust in dentists, although, it could be compromised if patients were unhappy with the care provide, were in pain, or had negative past experiences of treatment (Armfield et al., 2017).

Age appropriate information supports children in making competent and meaningful decisions about their own care (Mårtenson & Fägerskiöld, 2008). Therefore, when supporting adolescents to make decisions on managing their cFPM, clinicians need to be trustworthy, and provide information in a way that is well received, accepted, and more likely considered.

4.7.2 Adults

Three major themes were generated from adults' interviews: i) influences on patient's decision; ii) perceptions of specialist's role; iii) Importance of shared decision-making for cFPM in children. The themes and sub-themes are shown in **Table 4.6**.

Final Themes (and sub-themes)		
INFLUENCES ON PATIENT'S DECISION • Lived experiences • Parental effect • Societal constructs	PERCEPTION OF SPECALIST'S ROLE	IMPORTANCE OF SHARED-DECISION MAKING FOR cFPM IN CHILDREN • Reality vs. abstract • Empowering children

Table 4.6 Themes (sub-themes) of adult interviews (n=13)

Theme 1: Influences on patient's decision

Deciding whether to restore or remove cFPM is a decision that many adults are likely to have to make at some point in their lives. Those adults interviewed recalled making decisions, based on internal and external influencing factors.

A common view amongst interviewees was that their previous lived experiences of dental treatment would influence any future management of cFPM

> *"I've had fillings and extractions in the past, extractions were a lot worse than the fillings, so that would influence what I do …"*
>
> Adult 3 (32, F, Irregular, Ext/Fill)

When a participant only had previous experience of a filling or an extraction of a cFPM, in all cases, this would make them choose this option again if required:

> *"I've had one of these molars removed before, but never a filling, and it [the extraction] solved my issues, so I'd chose this option over a filling again."*
>
> Adult 5 (43, M, Regular, Ext)
>
> *"I've only experienced a filling, and it has worked, so I'm pretty content with having those done again..."*
>
> Adult 2 (25, M, Regular, Fill)

Management strategies such as root canal treatment, or prosthetic replacement post extraction are used. A small number of those interviewed had lived experiences of these additional treatment, and similarly echoed the views noted prior. For example, one interviewee said:

> *"My previous experiences of root canal treatment prompted me to try it out again. However, I knew that if it did not work, like it did previously, then I was happy to get it [tooth] out."*
>
> Adult 11 (46, M, Regular, Ext/Fill)

In addition to internal lived experiences, external influencing factors were referred to in interviews. Exploring the influence their own parents had on decision-making found that personal experience, including observation, was important in their decision making for any future cFPM that would need managed:

> *"I know that my parents have had fillings in the past erm, as I saw them have them, so I knew it was sort of the right thing to get done."*
>
> Adult 2 (25, M, Regular, Fill)

> *"I'm always inclined to do whatever my parents did with their teeth…"*
>
> Adult 7 (39, F, Regular, Extraction)

Only a small number of respondents indicated that their parent's previous treatment was not something that was considered:

> *"I recall my parents mainly having teeth extracted, but I cannot say this fits with how I make decisions about my own teeth."*
>
> Adult 9 (48, F, Regular, Ext/Fill)

Overall, supporting previous research (Vernazza et al., 2015), it was apparent that when adults decided how to manage cFPM now, or in the future, their decisions are driven by previous lived experiences of treatment. In this study, those who had previous experience of extraction, with the reference case being no experience, increased the odds of choosing an extraction over retaining the tooth (Vernazza et al., 2015). In contrast, experience of root canal treatment and crowns reduced the odds of an extraction being chosen (Vernazza et al., 2015).

Other internal factors such as functional impact, psychosocial impact, anxiety, aesthetic implications, or long-term considerations did not specifically feature in people's accounts of their intended future decision making for cFPM. However, it could be argued that these factors contribute towards the preconscious processing that patients undergo, before determining which preference they choose (Erb et al., 2002). Therefore, non-disclosure is unlikely to mean they do not exist, but instead are unconscious influences on decision making (Newell & Shanks, 2014).

Parental values and behaviours are known to transfer from one generation to another, with socialisation theories suggesting that a child's development is influenced by their parents (Maitre et al., 2021). The extent of these influences are shaped by interactions, and processes such as observation and modelling (Maitre et al., 2021). There are no reported studies which discuss observation and live parent modelling for deciding oral health treatments of cFPM. Previous evidence has shown that live parental modelling is likely to alter a child's behaviour, allowing them to accept dental treatment when they previously wouldn't have (Farhat-McHayleh et al., 2009). It could therefore be hypothesised that observations and parental modelling would similarly be successful in influencing a child's ability to decide. Although, it would appear that these approaches would only be pertinent for younger children, as in general, parental influences on a child declines after reaching young adulthood (Ward et al., 2014).

A common view amongst interviewees was that 'accepted' societal construct of the term filling and extraction would likely influence future decision-making for cFPM:

> *"Fillings seem like quite normal, so I do not think I would think of it as a large, a big thing really…and I think most other adults would think the same."*
>
> Adult 12 (47, M, Regular, Ext/Fill)

> *"My filling came out, I'm gonna have to have that sorted out next week, but I wouldn't wait until next week to have my tooth taken out next week. You know they're very different things in most people's eyes."*
>
> Adult 13 (37, F, Regular, Ext/Fill)

There were some suggestions that procedural complexities were linked to the societal construct of the term fillings and extractions:

> *"I tend to take fillings more in my stride 'cause yes, it's a little bit weird for about half an hour afterwards, but, but generally speaking, you know, it's a fairly short-lived experience…whereas the extraction was more invasive, took longer and is linked with more problems after…"*
>
> Adult 9 (48, F, Regular, Ext/Fill)

> *"I'd imagine most people would think having a tooth taken out is more difficult than having a filling"*
>
> Adult 1 (17, M, Regular, Nil)

Societal, or social, constructs are ideas that have been created and accepted by the people in a society (Burke et al., 2009). It is known that when faced with a decision, people will seek information from wider social contexts to help them (Zikmund-Fisher et al., 2011). They like to know what others would do when faced with the same decision, and have been shown to use social norms that are subjective, doing what others think they ought to do, and descriptive, actually doing what others do (Zikmund-Fisher et al., 2011; Brabers et al., 2016).

Recent studies have established that there are knowledge disparities existing around dental terminology (Hayes et al., 2017; Hamilton et al., 2021) and more specifically, to the terms decay and cavities (Claiborne et al., 2021). Despite these disparities around terminologies, those interviewed appeared to understand what fillings and extractions were. Some interviewees felt that the accepted societal norm of the term filling related to a less complex procedure, which would not require much thought about deciding whether to do it or not. If this holds true, then fillings could be seen as a conservative social norm (Brabers et al., 2016). Brabers et al., (2016) demonstrated that, in a Dutch population, the more conservative the social norms were, the less likely people were willing to be involved in medical decision-making, and more likely to just accept what was told. However, it could be argued that for some, extractions are considered to be a conservative social norm. Ultimately, fillings are not the panacea for all cFPM, and for some, extraction may be the more appropriate choice (Ashley & Noar, 2019; Lygidakis et al., 2022). These findings may suggest that adults use social norms, coupled with personal experiences, to help them make their decision. It is therefore important that patients are informed of all available options to minimise the potential mis-use of social constructs (Brabers et al., 2016).

Theme 2: Perceptions of specialist's role

Data from the adults interviewed suggest that they perceive the role of a specialist to be different from their general dentist, meaning the situation is more challenging, and perhaps something to be concerned about:

> *"Being sent to a specialist means it's more complex, so therefore that worries me a bit."*
>
> Adult 13 (37, F, Regular, Ext/Fill)
>
> *"If a problem was bad enough that I had to go to see a specialist, I, I guess that would be worrying"*
>
> Adult 8 (24, F, Regular, Nil)

If a child was required to see a specialist, some interviewees felt that something was wrong with the child while others suggested this was not the case:

"Does it mean my child's a bit abnormal because they have to go to a specialist?"

Adult 13 (37, F, Regular, Ext/Fill)

"I wouldn't say my child, or say my niece and nephews, were abnormal because they had to get sent to a specialist"

Adult 10 (53, F, Regular, Fill)

A common view amongst interviewees was that of acceptance should their general dentist suggest a referral to a specialist for themselves, or a child:

"Like our dentist said, 'This is the right place to go. You're going with my blessing. These people will be lovely'."

Adult 6 (37, M, Regular, Fill)

"…[if] you need to go and see and specialist, you go and see a specialist"

Adult 4 (20, M, Irregular, Nil)

Respondents felt that a referral to a specialist automatically meant the case was more challenging and complex. National guidance details the levels of complexity that general dentist and specialists and consultants should treat (NHS England, 2015). Referral to specialist services routinely involves more complex cases, with reasons such as disparity in knowledge, competence in providing care and challenges of coping with more complex patients being reported (Tzartzas et al., 2019). However, it has also been suggested that a referral may be made due to the increased time and resources required to provide optimal care for these complex cases (Tonelli et al., 2018), an attempt to seek validation (Tzartzas et al., 2019) or to merely provide guidance, rather than treatment (Taylor et al., 2019). Therefore, despite participants thinking that referral meant the treatment was more complex, this may not always be the case.

Parental acceptance of a referral, when required, supported a paternalistic approach, in that if a child needed specialist care, parents thought they should get it. Despite being focussed on adults, patients who attended a specialist pain clinic for management of chronic orofacial pain were provided with more accurate and appropriate treatments, compared to being managed by their general dentist (Bonathan et al., 2014). It could be argued that chronic orofacial pain is a niche diagnosis, that almost always requires specialist input. However, observations from this study could be considered for cFPM in children, especially when a multidisciplinary approach is required (Taylor et al., 2019; Alkadhimi et al., 2022).

The concept, however, that for some, a sense of their child being abnormal due to the need for a referral was an interesting finding. There is scarce literature to support this concept in health, let alone dentistry, and this merits further investigation. Patient complexity is likely to extend beyond just clinical need, with socioeconomic, cultural, behavioural, and environmental factors notably contributing to the notion of being 'complex' (Safford et al., 2007). As previously discussed, if there is a clinical need, then all adult participants supported referral. However, as referrals are usually made for more complex patients (Tonelli et al., 2018; Tzartzas et al., 2019), then these external factors may provide a very tenuous rationale as to why some adults felt the child was abnormal.

Theme 3: Importance of shared decision-making for cFPM in children

There was a sense that choosing to restore a child's cFPM was the right choice amongst adult interviewees. For most, this was irrespective of their previous experiences or beliefs, as restoring the tooth was seen to be a reversible choice:

> *"I've only had them [compromised first adult permanent molars] pulled, but for a child, it can only be a filling, because obviously that, getting extracted is the worst scenario really, you know its gone forever. Whereas if you have a filling, you can get the filling and then you can, you know erm, carry on and then make sure it doesn't happen again."*
>
> Adult 5 (43, M, Regular, Ext)
>
> *"…my first thought is I'd rather preserve the tooth and have a filling and then improve what we were doing as a family to then try and preserve that tooth and hopefully, so they do not need more fillings, or the tooth extracted, in the future"*
>
> Adult 9 (48, F, Regular, Ext/Fill)

However, one adult, who had no experience of filling or extraction, suggested removal was preferred:

"…why bother with a filling in someone so young. If removal is an option, and it would prevent them from future issues, then it needs to be removed, and I would still feel like I'd be acting in their best interests."

Adult 4 (20, M, Irregular, Nil)

It was a common practice that respondents agreed/discussed that these decisions should not be made in isolation by a parent or guardian, but as a shared decision which included parental views as well as the views and opinions of the dentist and the child:

"I would try and explain to them what was going on…but ultimately my decision would be, you know, i-, in, in partnership with the sort of dentist and my children…"

Adult 10 (53, F, Regular, Fill)

"I would definitely endeavour to make sure that my child has a more active role in that decision-making process."

Adult 6 (37, M, Regular, Fill)

In some case, involving a child's opinion is something they have experienced, or have the potential to do so, as they are a parent:

"…it's up to him what he has done to his body [yeah]. Obviously, we – I'll, I'll influence him as much as I can but I'm not going to tell him – make him do something he doesn't want to do because I wouldn't."

Adult 11 (46, M, Regular, Ext/Fill)

"…obviously I'd have to decide with her, it's her mouth at the end of the day that we're dealing with"

Adult 13 (37, F, Regular, Ext/Fill)

For some, involving a child's opinion was very much an abstract concept. They too felt involving the child was important, but demonstrated a child's involvement by either creating a hypothetical scenario, or, considering what they would have wanted as a child:

"Yeah, yeah, I mean like erm, I think it would be good if a dentist could tell them this happened to a person when they were younger if they had a filling, and this happened if they had an extraction. Maybe making it personal, by it being their parent or another child they knew…"

Adult 5 (43, M, Regular, Ext)

"…[I'd] try and think what they're going to be thinking about in the future. And they feel about it and how I would feel about it if it was me later on in the future, what I would have wanted to have done back when I was a kid..."

Adult 2 (25, M, Regular, Fill)

Whether involvement was a reality, or in the abstract, all respondents agreed that it was important to involve and empower children to be involved in treatment decision-making:

"[I've] always encouraged that two-way conversation from the kids' point of view because as they get into adulthood, they need to understand that, that they need that conversation and that relationship with their dentist in order to keep their teeth in good condition and make decisions about their dental health."

Adult 9 (48, F, Regular, Ext/Fill)

The importance of shared decision-making in health is the accepted gold standard for healthcare (NICE, 2021). A shared decision-making process has to involve the person and their healthcare professional(s), detailing risks, benefits and consequences, culminating in reaching a joint decision about what care to receive (NICE, 2021). This moves away from the previous method where the healthcare provider decides who gets what treatment, even it is deviates from the patient's own preferences (Asa'ad, 2019). It is very clear from these data, that deciding how to manage cFPM in children requires the opinions of the children themselves. This requires adults to involve children in the decision making process, whilst supporting them to make their own decisions, and respecting their autonomy (Buldur, 2021; NICE, 2021). Wogden et al., (2019) highlighted that adolescents with cleft lip and/or palate felt with increasing age, they should become more involved in decision-making, as it was important for adolescents to have a voice. In addition, adolescents reported overall high satisfaction at being involved in decision-making but on occasions felt as if they were ignored by professionals, or under pressure to decide by their parents.

Coyne and Harder (2011) propose that involving young people in decision-making is necessary, but can be quite complex. They suggest that children may not have the competence and capacity to understand the implications of the decisions they are making, and that the decision-making capacity of a young child will be significantly different to that of someone who is approaching adulthood. In addition, they propose that in some cases, parents and health professionals may prefer to take a protective stance towards children, to act in their best interests.

The concept of 'shared decision-making' was challenged by asking interviewees to recall a time they actively involved a child in a decision about the child's dental health. Those who were parents of adolescents suggested they involved, or would involve, their children in healthcare decisions. Understanding this dynamic from an adolescent point of view is important to establish. Even those adults who were not parents, or who had children that were too young to contribute to a shared decision-making process, concurred that involving a child in the decision-making process was the right thing to do. This finding provides a sense that adults want to empower children to be accountable for their own healthcare decisions, which is necessary as they transition into adulthood (Colver et al., 2020). Parents should be actively encouraged to empower their children by professionals, and supported in providing their child with the emotional support and guidance they need as they begin to take control of their own healthcare decisions (Keller & Whiston, 2008; Coyne & Harder, 2011). However, difficulties can arise in using this approach. These respondents' discourse may have focussed on their ideas, rather than taking the fictional parent perspective in the vignette. In addition, distinguishing between the respondents own opinions and the discourse that represents their understandings of social norms, that is their responses may not reflect what they would do but rather what is acceptable to say (Sampson & Johannessen, 2020).

4.7.3 Strength and limitations

The strengths and limitations of this qualitative study are discussed in this section.

Sample

A key strength was adolescents and adults with and without experience of cFPM were sampled. Identifying and understanding a diverse range of experiences allowed concepts to be discussed both in the reality, and in the hypothetical abstract, thus understanding both stated and revealed notions and ideas.

Younger children (<12 years old) were not included, which is a limitation. On reflection, conducting an interview with adolescents (>12 years old) was not as challenging as anticipated. It could be the concepts being discussed were of importance to these participants, hence their engagement, or that as a paediatric dentist, the CI had developed adequate communication skills to engage with this cohort. Therefore, including children from aged 6 upwards would have been pertinent to consider. Such inclusion would have captured those who had been through this decision around age 9, or to obtain opinions not based on prior experiences.

In addition, other key characteristics such as being a parent, having a particular disease profile or ethnicities were not purposively selected for during the sampling. It was felt these were not to be of importance for the research question at the outset. The recruited participants fortuitously were a range of parents and non-parents, which allowed concepts to be discussed in reality and the abstract.

Participants were only recruited from England. This was deliberate as early qualitative findings were going to be used to inform the DCE, based on a representative sample of the English public. However, given the distinct differences in how dental treatments are funded across devolved nations of the UK, it may have been prudent to include participants from across the UK.

Online Interviewing

On reflection, given the implications of COVID-19, changing to online interviewing was a real strength. The quality of the interactions did not appear to be hampered by using this method. Using this approach may have unintentionally excluded potential participants due to their technological circumstances, those who had poor internet connection or those who did not feel competent in using technology. However, conversely, it allowed participants to be reached, irrespective of their circumstances or geography, a potential issue had these remained in-person face to face (Carter et al., 2021).

Developing and validating the use of Figure 4.1

Using **Figure 4.1** as a vignette to support the interviews was a real strength. The results of the content analysis for each group demonstrate this is a good method to ensure the interviews remain focussed on cFPM. Certainly, this was an anticipated challenge, with several options to overcome this being discussed prior to commencing the interviews. One option considered was to ask the participant to identify the tooth in their own mouth, and this would be confirmed by the CI. However, this was felt to be potentially intrusive, especially in an online interview. Another was to show a picture of a real mouth and ask the participants to identify the first permanent molar; this was less invasive, however, felt to be cognitively challenging. In the end, a vignette (see **Figure 4.1**) was developed and appropriately piloted before use.

Recruitment

Unfortunately, but not surprisingly, recruitment was negatively impacted as the increased burden on services due to COVID-19 meant recruiting to a research study was not a priority. Despite making an ethical amendment to include three primary care dental practices for recruitment, only four participants were identified, and one recruited from these sites. On reflection, it was a time when all dental services were recovering from the effects of COVID-19, and thus, asking these sites to aid recruitment was always going to be challenging.

4.8 Conclusion

In summary, this chapter has described the qualitative processes and established adults' and adolescents' views and experiences around managing cFPM. It is apparent that for both adolescents and adults, several factors are likely to influence how they decide to manage cFPM, for themselves, and in the case of adults, for children. The relevance of shared decision-making is clearly supported by the current findings in both datasets. Overall, this study strengthens the idea that adolescents should be actively involved in as part of a shared decision-making dynamic for cFPM.

The early findings of the adult qualitative interviews were used, in part, to inform the DCE design. The next chapter will discuss the methodology, methods, results, and discussion of the DCE to elicit public preferences of managing cFPM in children.

Chapter 5. Public Discrete Choice Experiment

5.1 Introduction

An overview of preference elicitation methods was provided in Chapter 2, highlighting the importance of one method, the DCE. This chapter will report the development, analysis, and findings of a public DCE. This explores public preferences for managing cFPM in a 9-year-old child. The main objectives of the public DCE were to: a) establish the preferences and trade-offs between attributes of how to manage cFPM in addition to observing how respondents' characteristics influence their choice; and b) use the marginal WTP values, derived from the DCE, as inputs into an economic model of the management of cFPM over the life-course (discussed in Chapter 6).

Section 5.2 will discuss additional methodological considerations, in conjunction with those discussed in section 2.4.2 of Chapter 2, for the development and design of a DCE. Section 5.3 will provide the aim of the DCE of this thesis. Section 5.4 focusses on the development of the public DCE, including the samples, attributes and levels, survey design and analytical methods. Section 5.5 will present the results of the public DCE, prior to section 5.6 discussing the interpretation and implications of the public DCE, in addition to the strengths and limitations. These findings will be discussed in combination with the findings of the qualitative interviews (see Chapter 4) and decision analytical model (see Chapter 6), in Chapter 7, to consider the management of cFPM in a wider context.

5.2 Methodology

DCEs are a method to elicit preferences in a healthcare setting and section 2.4.2 of Chapter 2 discusses their underpinning theoretical assumptions (Lancsar & Louviere, 2008; Ryan et al., 2008; de Bekker-Grob et al., 2012). The methodology has mainly been discussed in Chapter 2 and so the main emphasis of this sub-section is on the selection of attributes.

In deciding on attributes and levels for a DCE, various sources of information should be included. A combination of evidence synthesis, expert (clinical, non-clinical and patient) opinion and qualitative research data can ensure the potential range of preferences and values that people may hold are included whilst including what is most relevant to policy- and decision-making (Bridges et al., 2011; Coast et al., 2012).

In this thesis, an expert panel was created. The main remit of this panel was to ensure the study reflected reality and provide expert opinion about aspects of model and DCE where evidence does not exist. Relevant stakeholders were purposefully recruited, using pre-existing contacts, to form a panel that would provide a divergent range of relevant and pertinent views. The following stakeholders were included:

- Three parents/members of the public who provided patient perspectives relating to cFPM
- Two academic paediatric dental consultants, both of whom, have extensively published on cFPM
- One NHS paediatric dental and orthodontic consultants who gave specialist paediatric and orthodontic input, from an NHS secondary care perspective
- Two general dental practitioners who provided insight from an NHS general dental practice perspective
- One consultant in dental public health, who supplied expert knowledge of public health strategies and commissioning of children's dental services
- One commissioner of dental services, who, as a non-clinician gave expertise on how established pathways fit into the current commissioning of children's dental services

Obtaining a collective expert opinion can be complicated when clinicians, non-clinicians and members of the public are involved (McMillan et al., 2016; Arakawa & Bader, 2022). Establishing a consensus agreement of these opinions can be obtained using an approach known as nominal group technique (NGT) (Harvey and Holmes 2012). This formal consensus development method ensures equal representation, where a small number of stakeholders are involved (as opposed to the Delphi Technique which determines consensus from a larger group) and reduces the potential for power differentials that can exist between clinicians and members of the public. The first stage of NGT is known as silent generation, where participants work individually on issues relating to a particular topic. The second stage involves everyone, one at a time, discussing their ideas in a 'round robin' fashion until all ideas are stated. The final stage is where participants clarify and elaborate the generated ideas as a collective group. The stage can involve ranking the ideas if this is appropriate. Therefore, utilising NGT allows clinicians, non-clinicians, and members of the public to work together to create an environment conducive to discussion. (Harvey and Holmes 2012; McMillan et al. 2016).

As previously explained in Chapter 2, a DCE can provide a value of health, and measure WTP, by the inclusion of a cost-attribute (Lancsar and Louviere 2008; Ryan et al. 2008). Given the inherent challenges of establishing appropriate utilities in dentistry, (see sections 1.2.6 and 1.6.1), estimating a WTP, through a DCE, allows a clear decision rule to be applied. As such, a DCE was used in this thesis to establish public preferences in managing cFPM, whilst eliciting societal WTP values that can be embedded into a model (see Chapter 6)

5.3 Aim

To design the DCE, to elicit the public's preferences for each pathway including determining societal willingness to pay (WTP).

5.4 Methods

The key stages involved in the development of the public DCE are described in the next sections. Available guidance for best practice were closely followed in designing the DCE described in this chapter (Bridges et al., 2011; Reed-Johnson et al., 2013) . Discussions regarding ethical approval and considerations for this thesis are reported in Chapter 4 (section 4.6.1). Reporting in this chapter is in line with ISPOR recommendations and a checklist is included as **Appendix H** (Bridges et al., 2011)

5.4.1 Early expert panel input

The first expert panel meeting took place virtually, at the very outset of the thesis. The primary focus of this meeting was to externally validate the conceptual model (see Chapter 6). In preparation for the second meeting, as described later in this chapter, members of the panel were asked to consider potential attributes for the DCE and email their thoughts to the CI. Some initial concepts regarding shared decision and providing information were proposed by members of this panel. A summary of the feedback, decision and rationale based on this exercise is available in **Appendix I**.

5.4.2 Establishing attributes and levels

The first stage in design of a DCE is to establish the attributes, which are relevant to the decision context, and their subsequent levels (Bridges et al., 2011).

Using qualitative data collection methods should be adopted when determining attribute selection (Coast et al., 2012). Initial concepts were established from five semi-structured qualitative interviews with adolescents and five members of the public (for more detailed information on the qualitative component of this study, see Chapter 4). These ideas focussed on the management (treatment options and who provided the treatment) and decision-making (who made the decision) of cFPM in children. These initial ideas were supplemented by UK practitioner survey data (Taylor et al., 2019) and evidence synthesis (Lygidakis et al., 2010; Cobourne et al., 2014; Burke & Lucarotti, 2018c; Kanzow et al., 2018; Zahran et al., 2018; Ashley & Noar, 2019; Colver et al., 2019; Schwendicke et al., 2021; Fenton et al., 2022) relating to the decision-making and management of cFPM. The attributes (and their levels) were chosen to be as congruent with the modelled health states in the model (see Chapter 6). Four attributes were chosen to ensure the DCE would be manageable by respondents. These attributes were discussed and agreed with the supervisory team. The initial attributes that were generated, and selected, from this process were:

- Provider of Dental Care
- Information needed to help decision
- Number of future appointments avoided
- Gap left post extraction

A cost attribute was included in the DCE, to ensure marginal WTP could be established and allow adequate mapping of WTP tariffs to the model. To cross-validate the decision on attributes, and support the assignment of levels, expert opinion was obtained.

Confirming the attributes, and subsequently assigning levels for each, were undertaken with the expert panel, using the principles of NGT to achieve consensus where required (Harvey & Holmes, 2012). An online expert panel meeting, with eleven members, was held. Panel members were split into two groups. Initially, each group were asked to discuss the chosen attributes, and comment on their relevance and importance to the context of managing cFPM. Additionally, they were asked to consider if any key attributes had been omitted. Each group fed back to one another, leading to an agreement of the final list of attributes to be included in the DCE.

The second task was to obtain the levels for each of the chosen attributes. The first stage of the second task, asked each member of the panel to give specific levels for each attribute. Members were cautioned against choosing too many attribute levels, and advised to try and limit levels to three or four per attribute (Bridges et al., 2011). If they were struggling to come up with specific levels, then general concepts/ideas about what they felt the levels should be was suffice. This stage was completed in individual breakout rooms, with each member recording their thoughts on an individual online whiteboard, known as Jamboard (See **Figure 5.1**).

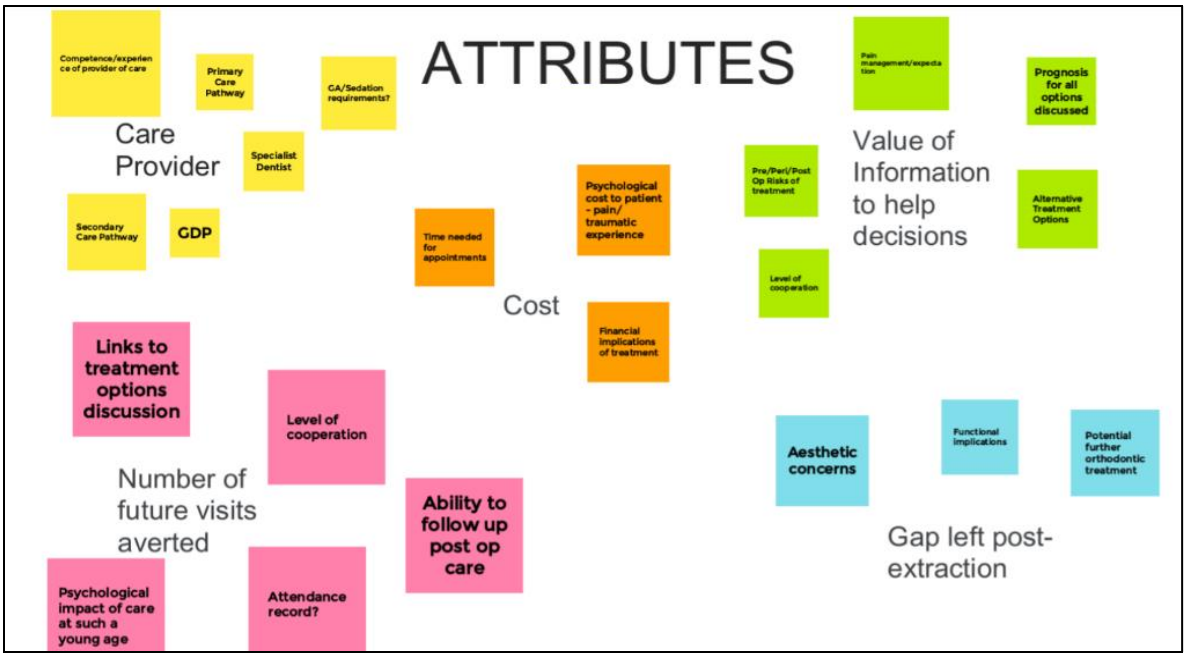

Figure 5.1 Example of Jamboard for levels exercise (individual)

The second stage of the second task involved each member discussing their levels, initially in small groups of three/four, before coming together as one large group. Once in the final group, one member of the panel was tasked to document the group consensus on a Jamboard (See **Figure 5.2**).

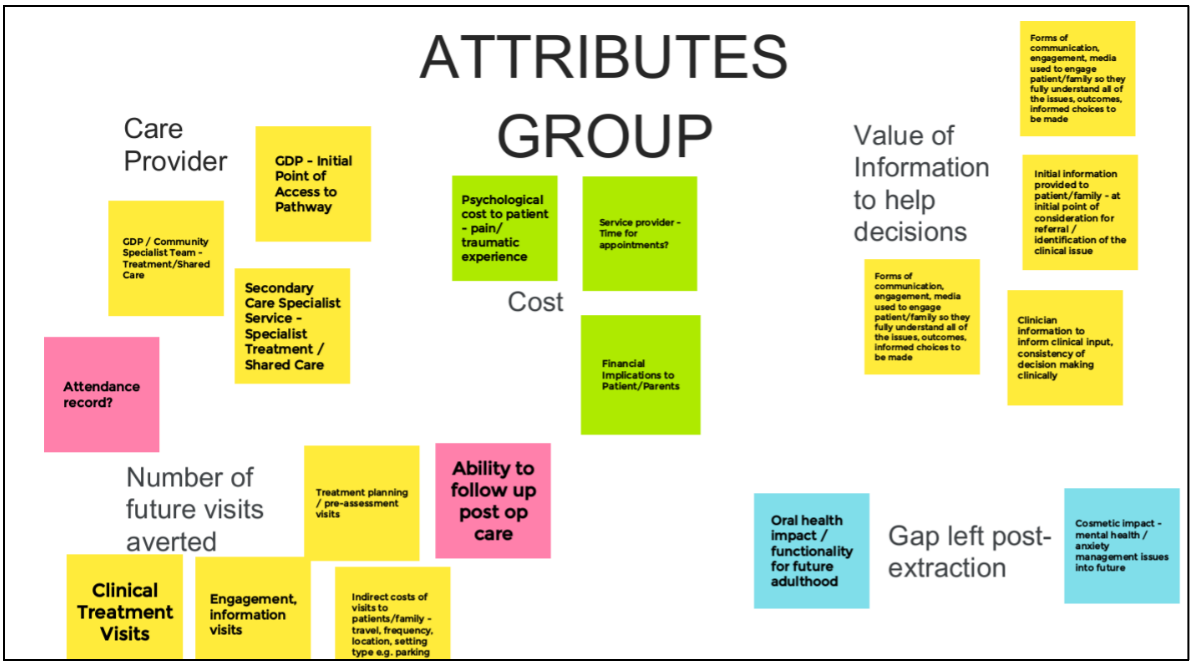

Figure 5.2 Example of Jamboard for levels exercise (whole expert panel)

Refinement of these levels was undertaken with the supervisory team before a final agreement was reached.

Establishing levels for the cost attribute was undertaken by utilising data from various different sources: a previous PhD thesis, on 'willingness to pay' for non-vital molar tooth in an adult (Vernazza, 2011) (see **Table 5.1**); Unit Costs of Health & Social Care 2021 (Jones & Burns, 2021) and expert opinion.

Initial choice	Prosthetic replacement	Minimum	Lower Quartile	Median	Upper Quartile	Maximum
Save tooth (Root Canal Treatment)	N/A	0	80	175	250	10000
Extract tooth	None (Leave gap)	0	40	67.5	100	750
	Removable denture	30	58	163	200	1750
	Fixed partial denture	5	118	200	331	3000
	Implant	10	168.8	250	500	2500

Table 5.1: Median WTP values with ranges and quartile values by initial preference. Adapted from (Vernazza 2011)

Expert opinion, from the clinical members of the expert panel, was sought to obtain the maximum price that would be paid privately for extracting a tooth and placing a bridge at age 20. The following calculations were used to establish the three cost attributes:

- Cost of extraction and bridge placement at age 20 = £2000; discounted back to age 9 (discounting rate 3.5% (NICE, 2013)) = £1351.54 (£1350)
- Cost of Extraction/Leave Gap = median WTP £67.5 in 2011 (Vernazza, 2011); inflating to 2020 (Gross domestic product annual % increase (Jones & Burns, 2021))) = £76.24 (£80)
- Cost of retaining tooth = median WTP £175 in 2011 (Vernazza, 2011); inflating to 2020 (Gross domestic product annual % increase (Jones & Burns, 2021))) = £201.83 (£200)

The final attributes, their levels, and the relevant evidence used for the public DCE are shown in **Table 5.2**.

Attribute	Levels	Evidence Used
Gap	1. No gap as filling is undertaken and the so the tooth is still in place 2. Full tooth gap present, with no intervention to close 3. Gap partially closed with no dental intervention (tooth behind moves forward some of the way) 4. Gap closed with no dental intervention (tooth behind moves forward completely) 5. Gap closed orthodontically (with braces) 6. Gap closed with prosthesis (with bridge)	Expert opinion and evidence synthesis (Lygidakis et al., 2010; Cobourne et al., 2014; Ashley & Noar, 2019; Taylor et al., 2019; Schwendicke et al., 2021)
Provider of Dental Care	1. General Dental Practitioner 2. Dentist with enhanced skills in Paediatric Dentistry 3. Specialist in Paediatric Dentistry	Expert opinion and evidence synthesis (Taylor et al., 2019; Fenton et al., 2022)
Cost	1. £80 2. £200 3. £1350	Expert opinion and evidence synthesis (Vernazza, 2011; Jones & Burns, 2021)
Decisions about a child's care	1. Dentist discusses options with me and my child, but the dentist makes the decisions 2. Dentist discusses options with me and my child, and we come to an agreement between us 3. Dentist discusses options with me and my child, but I make the decision on my own	Expert opinion and evidence synthesis (Colver et al., 2019)
Number of future appointments avoided	1. Extraction + leave gap – 2 2. Filling - 15 3. Extraction + orthodontic closure - 33	Expert opinion and evidence synthesis (Burke & Lucarotti, 2018c; Kanzow et al., 2018; Zahran et al., 2018)

Table 5.2: Attributes (& Levels) used in the public discrete choice experiment

5.4.3 DCE experimental design

The attributes and their levels (as shown in **Table 5.2**) were used to develop an experimental design of choice scenarios using Ngene™ version 1.1.1 (ChoiceMetrics, 2012). A full factorial design which incorporates all possible combinations of attributes and levels (Lancsar & Louviere, 2008) would have resulted in 117,855 possible scenarios[3]. Given the task complexity, a full factorial design was considered too large. Instead, a fractional factorial design was employed, thus reducing the number of choice scenarios to a more manageable number whilst enabling the exploration of all main effects (effects of each attribute) of interest (Lancsar & Louviere, 2008). In experimental designs, it is desirable to seek orthogonality (i.e., the attribute and levels are evenly distributed across all the pairs of scenarios presented), however, strict orthogonality may not be always feasible to achieve and in such cases the design should be as mathematically efficient as possible (Reed-Johnson et al., 2013). A D-efficient design was used to maximise efficiency and to ensure that an optimal set of choice scenarios are used. Generating an efficient design requires knowledge of prior co-efficient values (Rose & Bliemer, 2009; Reed-Johnson et al., 2013). However, prior coefficient values, and directions of these values, for all attributes, were not readily available; thus, these were assumed to be zero and instead, dummy variable coding was used. An overview of the Ngene coding can be found in **Appendix J.**

The efficient design generated 18 choice sets after the 24,094th iteration, with a D error 0.673905. It has been suggested that respondents can manage multiple choice tasks. However, choice variance and heavy cognitive burden are likely to increase with increased number of choice sets (Bech et al., 2011). The balance between maximising statistical efficiency and response efficiency is challenging. Deviating from the most statistically efficient design, compromising on statistical precision, may be desirable as it can improve relevance and realism. In doing so, less precise model parameter estimates are obtained, but these are more likely to be reliable, consistent, and valid choices (Mühlbacher & Johnson, 2016). Thus, to minimise this, the 18 choice sets were blocked into two, with 9 choice sets per block with attribute and level balance being assessed to check for orthogonality (See **Appendix K**) (Reed Johnson et al., 2013). The number of choice sets used were considered manageable considering the existing evidence (Clark et al., 2014).

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[3] $6 \times 3^4 = 486$; $(486 \times 485)/2 = 117{,}855$ unique choice sets

The inclusion of an ‘opt-out’ alternative has been debated. Including an ‘opt-out’ may generate smaller attribute coefficients compared to those obtained from a forced choice. In addition, it may lead to unnecessary loss of efficiency if a higher number of respondents choose to opt out (Veldwijk et al., 2014). However, considering the nature of the decision problem faced by respondents, an opt-out alternative of ‘no treatment’ was included, as forcing a choice to manage cFPM did not reflect reality.

Respondents were informed that choosing the ‘no treatment’ option meant the tooth would get worse, and it would likely to break and become painful and/or infected, potentially causing a swelling or an abscess. It was assumed, and inferred, that this would happen within five years, and any future treatment to deal with the broken/painful/abscessed tooth would be more difficult, more expensive and have less certain results

Two internal validity checks were included in the survey, taking the choice tasks to 11 per block. The first check was to repeat a choice scenario (choice set 3 in each block was repeated as choice set 10) to assess the reliability of participant responses. The test-test reliability was included as consumer theory specifies individuals should have a preference between any two or more possible alternatives i.e., if choice scenario A is preferred to B, then if presented with this same choice scenario at any future point, they mostly should again prefer A to B. The second check was to included a within set-dominated pairs scenario, whereby when one alternative is unambiguously better, across all attributes, to check whether respondents choose the dominated alternative within the set (Johnson et al., 2019). Including this test allows potential issues, for example, participants not paying sufficient attention to the choice tasks, to be ascertained. Respondents who violate these tests may be deleted and excluded from the main analysis (Ryan et al., 2001). However, failure of these tests does not necessarily mean that respondent was irrational in their choice (Lancsar & Louviere, 2006). In addition, respondents may have valid reasons for failing validity tests, such as they may rate particular attributes very highly, and fail the dominance test as a result (Ryan et al., 2009). Removal of these valid preferences may bias the results (Lancsar & Louviere, 2006). As such, the choice data analysed in this DCE included all respondent data, irrespective if they failed the validity tests.

Respondent demographics were obtained to help understand the characteristics of the sample studied. Existing information held about respondents by the National Centre for Social Research (NATCEN), who provided the online panel of respondents for the final DCE, was used to determine gender, age, ethnicity, political party identification, main economic activity, and income. Person identifiable information such as name, date of birth, full postal address and NHS number were not collected to comply with data governance protocols.

Direct questioning of the respondents included specific questions relating to respondents' parental status (Did they have children? If so, how many were currently under the age of 16? Had any of their children ever had fixed orthodontic treatment?).
Further questions on respondents' current oral health experiences included:

- Do you have any of your own teeth left in your mouth?
- Have you ever had fixed orthodontic treatment (metal braces attached to your teeth)?
- Which, if any, of the following types of dentists have you ever received dental treatment from?
    - General Dental Practitioner
    - Dentist with enhanced skills
    - Specialist dentist
    - None – I have never received dental treatment
- Which of the following best represents how your dental care is provided?
    - Private: You pay the full costs of treatment
    - Paid-for NHS dental care: You pay the NHS 'band' charges
    - Free NHS dental care: You do not pay and are exempt from NHS charges
    - NHS and private care (Mixed of some NHS banded and some private treatments)
    - Insurance-based: You pay a fee which covers all treatment except laboratory bills
    - Insurance-based: You pay a fee which gives discounts on any NHS or private treatment received

In addition, questions focussing on respondents' dental anxieties were included, as it was assumed anxiety could likely influence preferences. Questions, and scaled responses for each question (1 – not anxious, 2 – slightly anxious, 3 – fairly anxious, 4- very anxious and 5 – extremely anxious), were adapted from the modified dental anxiety scale (MDAS) (Humphris et al., 1995):

- How do you feel generally about going to the dentist?
- How anxious are you about having a local anaesthetic injection in your gums for dental treatment?
- How anxious are you about having a filling?
- How anxious are you about having a tooth taken out?

Finally, respondents were then asked questions specifically about their first permanent adult molar teeth. A picture (see **Figure 5.3**) was included to support these questions:

- Have you ever had a filling (metal or white filling) carried out on any of your first permanent adult molar teeth?
- Have you ever had an extraction (removal of tooth) carried out on any of your first permanent adult molar teeth?
- How important is it for you to keep your first permanent adult molar teeth?
    - Important, neither important nor unimportant or unimportant

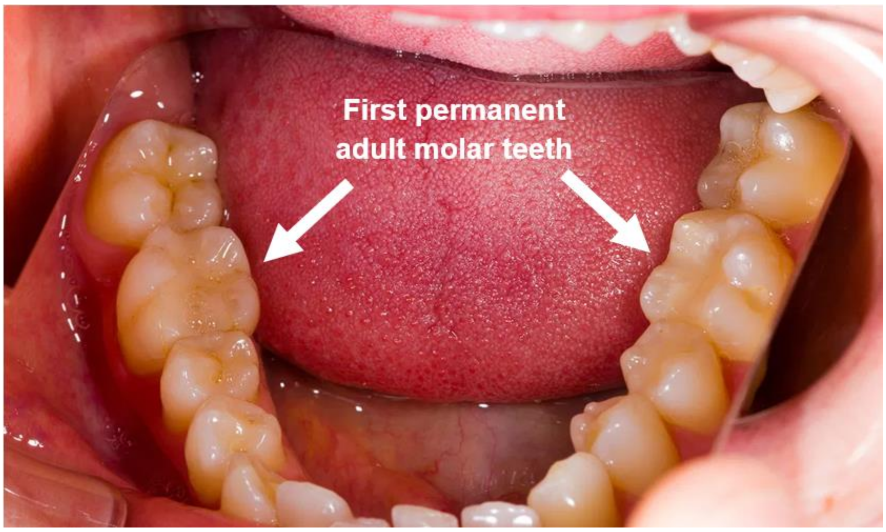

Figure 5.3 Picture included to support respondents in answering questions about first adult molar teeth

After demographic questions, the nature of the problem being considered was described. It was important to emphasise to respondents that this cFPM was not causing any pain (as per the agreed definition – see Section 1.8). As such, this preparatory information read:

Imagine you have a 9-year-old child who has a compromised first permanent adult molar, as shown in the picture below. The tooth is not causing your child pain but does cause occasional sensitivity.

Any treatment options (aside from no treatment) would require a local anaesthetic to numb the mouth. There should therefore be no pain associated with any dental procedure.

You should therefore assume the level of pain is equal across all treatment options. For all the treatment options, your child would receive local anaesthetic to numb the mouth – so, please assume there is the no pain associated with any of the procedures.

Following this, an introduction to the choice tasks, including a detailed description of each of the attributes, and their levels, was given. Respondents were given further information about costs, to avoid confusion, as in England, there are no costs to the parent for the management of children's teeth. Respondents were advised that in many other countries' parents would have to pay for their child's dental treatment, so they were to assume they lived in such a country where you must pay, out of your pocket, for the option they chose.

Prior to completing choice tasks, respondents were given a completed example of a choice task, rather than a 'warm up' exercise, (See **Figure 5.4**) to aid completion (Veldwijk et al., 2016). This scenario was chosen at random and did not include any of the choice sets in the efficient design. A description of the implications of choosing the scenario was given to help the respondents understand what it meant to choose that scenario.

An example of a complete choice set…

Example A	Scenario 1	Scenario 2	No treatment
Treatment Options	No Gap 	Partial Tooth Gap Present 	
Provider of Care	Dentist with enhanced skills in Paediatric Dentistry	General Dental Practitioner	
Decisions about a child's care	Dentist discusses options with me and my child, but **I** make the decision on my own	Dentist discusses options with me and my child, but the **dentist** makes the decision	
Number of future treatment appointments avoided	33 visits	15 visits	
Cost	£1350	£80	
Your choice (tick one box only)	☑	☐	☐

The participant has chosen scenario 1. They are willing to pay significantly more money for a 9-year-old child to have a filling done, and leave no gap, by a dentist with enhanced skills rather than leave a partial gap following tooth removal by a general dental practitioner. The participant is prepared to pay more for them to decide they want to make the decision, rather than having the dentist make it for them. Also, they are wanting to pay more because choosing scenario 1 will avoid their child having 33 future treatment visits compared to 15.

Figure 5.4 Example of completed choice task included in the survey

Demographics, preparatory information, an example choice task, and the 11 choice tasks were compiled into a paper format for pre-testing and piloting.

5.4.4 Pre-testing

The paper format of the initial survey was pretested, among four members of staff of the School of Dental Sciences, Newcastle University, primarily for content and face validity. Following feedback, minor amendments were made to simplify the wording and layout of how the levels and attributes were described.

5.4.5 Piloting

After the pre-test, the paper format of revised questionnaire was piloted on a sample of 30 purposively selected clinical and non-clinical participants working across various departments in Newcastle Dental Hospital. The main participant demographics are shown in **Table 5.3**. This survey pilot was run on 25th – 27th August 2021. In addition to completing the survey, further feedback was sought on the wording, layout, quality of pictures and whether they had any difficulties in understanding and completing the survey. This feedback was obtained either by annotating the paper questionnaires, or through informal verbal feedback. All 30 respondents completed the survey accurately, with none being excluded from analysis of the pilot data. Of the 30 analysed, 5 (16.67%) failed the test-retest internal validity check and 2 (6.67%) failed the dominance test. The level of accurate and complete responses was considered acceptable. This suggested that the attributes, and levels used in the survey were appropriate and did not result in cognitive overload for respondents.

Characteristics	N (%)
Sample	30 (100%)
Age	
23-30	4 (13.3%)
30-40	11 (36.7%)
40-50	9 (30.0%)
50-60	4 (13.3%)
60+	2 (6.7%)
Gender	
Male	11 (36.7%)
Female	19 (63.3%)
Parent	
Yes	22 (73.4%)
No	8 (26.6%)

Table 5.3: Characteristics of the participants in the pilot test

Most (n=21) reported no difficulty in completing the choice sets. The remaining (n=9) explained that the cost attribute, in most cases, was the main reason for deciding which choice set was chosen. The example choice task was helpful for most (n=27), however, the high-cost attribute in this chosen example may have influenced responses. Given the apparent influence of the cost attribute in the example choice task used in this piloting phase, it could be hypothesised that if an example choice task with a higher cost attribute was used then respondents should be more concerned by cost. As such, a nested methodological experiment within the DCE was created to test this hypothesis and see whether the cost attribute that was chosen in the example influenced respondents' decisions. As such, Figure 5.4 was duplicated, with one example having the cost attribute as high, £1350, and the other as low, £80. Across the sample (and each block - as described in section 5.4.3), half of the respondents were randomised to be shown the example choice task with the low-cost attribute and the other the high-cost attribute.

Annotated comments suggested minor amendments to the text, the layout of the treatment option attribute and quality of the picture used to describe the partial gap. These were carried out making some of the text bold to highlight important difference in attribute levels, improving the quality of all pictures and ensuring the table is all on the same page.

Completed pilot responses were input and analysed in STATA™ Version 17 (2021) to primarily develop the coding framework for the full dataset. In addition, theoretical validity was assessed by checking whether the parameters moved in expected directions, by looking at the estimated signs of the parameter coefficients. For example, it was expected that alternative specific constant (ASC), which captures the variation in choices not explained by the attributes, should be positive, suggesting that respondents preferred receiving a package of care compared to no care, which it was.

5.4.6 Online DCE

The paper format of the questionnaire was developed into a web version of survey, in collaboration with NATCEN. This was tested, and compared to the paper copy, to ensure consistency. There were no reports of technical issues in completing the web version of the survey. Timers were included in the online DCE to ensure respondents spent sufficient time reading each page. The complete online questionnaire can be found in **Appendix L**.

5.4.7 Sample

The survey was completed online by a representative sample of the UK adult population (>18 years of age) using an existing web-panel provider, NATCEN. NATCEN have a random-probability sample with additional offline coverage, to ensure representativeness. Originally, offline coverage was planned, however, given the COVID-19 implications, it was not attempted. Intermittent checking of predominately the age ranges of completed responses was done by NATCEN in an attempt to ensure the sample remained representative. Targeted efforts were made during subsequent invitations to these increase responses in under-represented age profiles. Ethical approval (see Chapter 4) permitted the use of NATCEN to collect this data, and for NATCEN to provide basic patient characteristics data e.g., gender, household income etc. Informed consent was assumed by completion of the online DCE. Electronic PILs and contact details were available for any participant, upon request, from NATCEN.

The survey was administered between October 2021 and November 2021, with NATCEN guaranteeing completed surveys from 430 respondents. Each respondent was incentivised with a £10 voucher upon completion.

With regards to sample size, in a review of 69 healthcare DCE studies published in 2012, it was found that only 30% of studies reported their sample size calculation method (de Bekker-Grob et al., 2015). Most existing studies calculated sample sizes by the rule of thumb (Orme, 2010), with a range usually between 100 to 300 respondents being noted (de Bekker-Grob et al., 2015). The use of parametric approaches to calculate sample sizes has been proposed (Rose & Bliemer, 2013). However these suffer from the limitation of requiring prior knowledge about the parameter estimates (de Bekker-Grob et al., 2015), which is not readily available for many healthcare, or dental, studies. Another suggested approach is a calculation based on an inferred asymptotic variance-covariance matrix informed by required significance level, statistical power, choice of analysis model, some prior information and the DCE design (de Bekker-Grob et al., 2015). However, similar issues are concerned with lack of prior information. The sample for this study was estimated using Orme's (2010) rule of thumb approach, using the following equation:

$$N > 500\frac{L}{TA}$$

where N is sample size required, L is the largest number of levels for any of the choice attributes, T is the number of choice sets, and A is the number of alternatives assessed. Using this equation, the required minimum sample was estimated as 90 respondents for this DCE.

5.4.8 Data analysis

The completed dataset was obtained, cleaned, and provided by NATCEN prior to analysis.

Several logistic regression modelling approaches for analysing DCEs are available (Hauber et al., 2016). Multinomial logit (MNL) analyses and conditional logit analyses appear to be used most often for choice set data (Clark et al., 2014) as it is consistent with random-utility theory (McFadden, 1974). MNL are most used to describe models that compare the characteristics of the respondents making the choices to their choices, whereas conditional logit relates choices to the elements that define the alternatives among which respondents choose (Hauber et al., 2016). As such, a simple conditional (fixed-effects) logistic regression was performed in STATA 17 (2021) within a random utility framework to estimate preferences for different processes and outcomes of care in the population. Despite being the most common analytical approach, conditional logit can lead to unrealistic predictions as well as not being able to account for preference heterogeneity (Hauber et al., 2016). To relax the assumptions of the conditional logit model, a mixed logit (random errors) model was performed in STATA 17 (2021) to check if similar predictions were made. In addition, preference heterogeneity was able to be assessed due to the mixed-logit allowing one or more of the parameters in the model to be randomly distributed (Hauber et al., 2016). Akaike information criterion (AIC) and Bayesian information criterion (BIC) were used as measures of the model fit (Hauber et al., 2016).

The signs (negative or positive) of coefficients indicate the direction of an attribute in terms of utility. However, these attributes are not directly comparable against each other because of differences in the scale of measurement in each attribute. In addition, the size of the coefficient provides little meaningful information. To overcome this, and compare coefficient results, the marginal rates of substitution (MRS) were estimated. Unit WTP for each attribute was derived by dividing the coefficient of one attribute by coefficient of the cost attribute (Mott et al., 2020). Conversion from dummy coding to effects coding was undertaken to allow WTP values for all levels within each attribute (Bech & Gyrd-Hansen, 2005). These WTP values were then used as estimates for the benefit parameters in the economic evaluation model (see Chapter 6). Similar methods of interpreting the mean parameters are noted for the mixed logit model. In terms of preference heterogeneity, the sign of the co-efficient is meaningless, however, significant SDs might be indicative of preference heterogeneity within the sample for that particular attribute level.

The initial plan was to perform subgroup analysis. However, small sample sizes within the subgroups of interest were observed and meant meaningful results could not be produced. Instead, interaction analyses were performed to see how certain attributes within the DCE were influenced by specific patient characteristics e.g., age, gender etc. Interaction analyses were primarily driven by the relevant patient characteristics of interest. As a result, interaction analyses for each characteristic were only completed for the attributes which were felt to be the most likely influenced by the said characteristics of interest. In **Table 5.4**, each of the interaction analysis are shown, with information relating to the respondent characteristics of interest and the anticipated outcome for the relevant attribute chosen. All interactions shown in **Table 5.4** were agreed prior to analysis to reduce the risk of Type I error.

For certain interaction analyses, responses to certain characteristics (anxiety, how care was provided and income) were combined to provide two dichotomous groups. For anxiety, anyone who responded as slightly, fairly, very, or extremely anxious, were classified as being 'anxious' and combined into one group. For those who responded as having care provided private only, insurance based - all costs and insurance based - discounted costs, were combined as one group and classified as being paying for them treatment 'privately'. Finally, those whose responded with a monthly income >£2560 (selected either £2,561 - 4,350 or £4,351 or more) were combined and defined as a 'high-income' group.

Respondent Characteristic	Anticipated outcome of interaction analyses
Gender	Expect female respondents to be more concerned about having a gap therefore the co-efficient on the full gap and partial gap attribute should be higher
Age	Expect older respondents to be less concerned by cost and less concerned by having a gap, as they may have more disposable income and have most likely lost teeth already. Therefore, the co-efficient on the cost and full gap and partial gap attribute should be smaller
Have children	Respondents who have children may be more protective of retaining the tooth, therefore expect to see a higher negative co-efficient full gap and partial gap attributes
Income (monthly)	Expect respondents with higher monthly income to be less concerned by cost, therefore the co-efficient on the cost attribute should be smaller
Political Party Identification	Those identifying as a voter of the conservative party (in the main, the more right-wing party in the UK) would be less concerned by cost but more concerned about having a gap, as members of this party tend anecdotally tend to be more affluent based on the ideologies of the Conversative agenda, therefore the co-efficient on the cost attribute should be smaller and the full gap/partial gap greater
Previous orthodontic treatment	Expect those who have had orthodontic treatment would have higher preference for orthodontic closure of the gap and having treatment done by specialist (based on their previous experiences with a specialist provider), therefore these coefficients should be larger
Anxiety - having a filling?	Those who are anxious about having a filling would be more likely to want a treatment outcome that is not a filling. Therefore, the full gap, partial gap and no gap attribute would have a smaller co-efficient
Anxiety - having an extraction?	Those who are anxious about having an extraction would be more likely to want this tooth filled. Therefore, the full gap, partial gap and no gap attribute would have greater co-efficient
How dental care provided	Those pay privately for treatment (private only, or insurance based) are less concerned by cost but more concerned about having a gap (often more private treatment are linked with aesthetics treatment), therefore the co-efficient on the cost attribute should be smaller and the full gap/partial gap greater
Previous filling of cFPM	Those who have had a previous filling would be more likely to want this tooth filled, given the minimal impact fillings often have on people. Therefore, the full gap, partial gap and no gap attribute would have greater co-efficient
Previous extraction of cFPM	Those who have had a previous extraction would be more likely to want this tooth extracted, and accept a gap, given extraction is often linked to managing pain. Therefore, the full gap, partial gap and no gap attribute would have smaller co-efficient, still negative
Example given to help with DCE	Expect respondents with example where higher cost attribute us chosen should be more concerned by cost, therefore the co-efficient on the cost attribute should be higher

Table 5.4 Interactions - Hypotheses testing, including the nested methodological experiment

5.5 Results

5.5.1 Descriptive statistics

A total of 430 respondents, from the NATCEN UK public panel, completed the online survey. It was difficult to assess a total response rate as there was no method of identifying how many respondents from the panel viewed the invitation but declined to take part. **Table 5.5** presents a summary of the key characteristics of the respondents, whilst **Tables 5.6 - 5.8** describe responses to the general dental questions, dental anxiety, and previous management of first adult molar teeth respectively.

The sample was mostly representative of the UK population, as age, gender, ethnicity, household composition and main economic activity were similar to the most recently available and comparable census data (Office for National Statistics, 2011). The same was found for the total average monthly income per household data (Office for National Statistics, 2021). Unfortunately, there is no robust UK based data source which confirms political party identification.

Characteristics	N	%
Sample	430	100
Age		
18-29	35	8.1
30-39	63	14.7
40-49	77	17.9
50-59	67	15.6
60-69	106	24.6
70+	82	19.1
Gender		
Male	250	58.1
Female	180	41.9
Ethnicity		
Prefer not to answer	2	0.5
Don't know	4	1.0
White British	358	83.2
Any other White	29	6.7
Mixed or multiple ethnic groups	8	1.9
Asian or Asian British	17	3.9
Black or Black British	10	2.3
Other	2	0.5
Any child (U-18) living in household		
Prefer not to answer	1	0.2
Don't know	0	0
Single person household	89	20.7
One adult (with children)	20	4.7
2 adults (no children)	162	37.7
2 adults (with children)	97	22.6
3+ adults (no children)	44	10.2
3+ adults (with children)	17	3.9
Main Economic Activity		
Prefer not to answer	0	0
Don't know	0	0
Full time education	12	2.8
Paid work	210	48.8
Unemployed	7	1.7
Retired	129	30.0
Other	72	16.7
Total Household Income (per month)		
Prefer not to answer	16	3.7
Don't know	1	0.2
Less than £1,410	90	20.9
£1,411 - 2,560	105	24.5
£2,561 - 4,350	120	27.9
£4,351 or more	98	22.8
Political Party identification		
Prefer not to answer	2	0.5
Don't know	1	0.2
Conservative	131	30.5
Labour	122	28.4
Liberal Democrat	33	7.7
Other	26	6.0
None	115	26.7

Table 5.5 Characteristics of the respondents

In terms of dental health, the sample is mostly representative of the population. When compared to the 2009 ADHS (England, Wales and Northern Ireland), the sample had a similar level of dentate respondents (Fuller et al., 2011). The number of sample respondents who had received a filling on their cFPM was similar to those who had at least one filled tooth in the 2009 ADHS, albeit not directly comparable as individual tooth level data was not available (Steele et al., 2011). Comparison of the sample to the population in terms of extraction of cFPM was unable to be ascertained, due to no data being available. However, in the sample, there was a relatively even split. The type of care received by the sample was similar to the 2009 ADHS, for those who paid for NHS and private care, although the sample had lower representation of those who received free NHS care and higher for mixed NHS/private care (Morris et al., 2011). Levels of general dental anxiety amongst the sample were like those reported in the 2009 ADHS, although when looking at individual components of the MDAS scale used, the sample was less anxious about having a filling/local anaesthetic injection and more anxious about an extraction, which is the opposite from the 2009 ADHS (Nuttall et al., 2011). There was a higher percentage of respondents children having had orthodontic treatment, compared to the 2013 CDHS (Rolland et al., 2016). This is likely due to the sample being asked if any of their children had had braces, whereas the 2013 CDHS was a representation of children undergoing orthodontic treatment at the time of the survey.

Characteristics	N	%
Sample	430	100
Do you have any of your own teeth left in your mouth?		
Yes	416	96.7
No	14	3.3
Have you ever had fixed orthodontic treatment?		
Yes	92	21.4
No	338	78.6
Do you have any children (including adult children and child not living with you)?		
Yes	328	76.3
No	102	23.7
If yes (n=328), how many under-16 and have had braces?		
Child currently under-16		
Yes	122	37.2
No	206	62.8
Child had braces		
Yes	114	34.8
No	214	65.2
Which of the following best represents how your dental care is provided?		
Prefer not to answer	1	0.2
Don’t know	1	0.2
Private Care only	92	21.4
Paid-for NHS Dental Care	215	50.0
Free NHS Dental Care	58	13.5
Mixed NHS and Private Care	34	7.9
Insurance-based (all costs)	19	4.4
Insurance-based (discounted)	10	2.3
Which, if any, of the following types of dentists have you ever received dental treatment from?		
Prefer not to answer		
Don’t know	0	0
General Dental Practitioner	0	0
Dentist with enhanced skills	396	92.1
Specialist	59	13.7
Have never received dental treatment	106	24.7
	12	2.8

Table 5.6 Responses to questions on general dental information

Characteristics	N	%
Sample	430	100
How do you feel generally about going to the dentist?		
Don't Know	0	0
Not Anxious	155	36.0
Slightly Anxious	139	32.3
Fairly Anxious	61	14.3
Very Anxious	40	9.3
Extremely Anxious	35	8.1
How anxious are you about having a local anaesthetic injection in your gums for dental treatment?		
Don't Know		
Not Anxious	0	0
Slightly Anxious	132	30.7
Fairly Anxious	157	36.5
Very Anxious	65	15.1
Extremely Anxious	49	11.4
How anxious are you about having a filling?	27	6.3
Don't Know		
Not Anxious	1	0.2
Slightly Anxious	122	28.4
Fairly Anxious	156	36.3
Very Anxious	77	17.9
Extremely Anxious	50	11.6
How anxious are you about having a tooth taken out?	24	5.6
Don't Know		
Not Anxious	1	0.2
Slightly Anxious	48	11.2
Fairly Anxious	118	27.5
Very Anxious	105	24.4
Extremely Anxious	92	21.4
	66	15.3

Table 5.7 Responses to questions on dental anxiety

Characteristics	N	%
Sample	430	100
Have you ever had a filling (metal or white filling) carried out on any of your first permanent adult molar teeth?		
Don't Know	0	0
Yes	346	80.4
No	84	19.6
Have you ever had an extraction (removal of tooth) carried out on any of your first permanent adult molar teeth?		
Don't Know	1	0.2
Yes	251	58.4
No	178	41.4
How important is it for you to keep your first permanent adult molar teeth?		
Don't Know	0	0
Important	351	81.6
Neither Important nor unimportant	75	17.4
Unimportant	4	1.0

Table 5.8 Responses to questions on previous management of first permanent adult molar teeth

Of the 4730[4] responses, the majority opted for a treatment option (rather than opt out) with 42.39% (n=2005) and 54.95% (n=2599) choosing scenario 1 and 2 respectively. The no treatment choice was chosen by 1.64% (n=78). The remaining 1.01% (n=48) respondents preferred not to answer. Regarding the validity tests, 91.4% (n=393) and 75.6% (n=325) passed the dominance and test re-test reliability test respectively. Irrespective if they passed or not, all respondents were included in the final analysis.

5.5.2 Conditional logit model - dummy coding

The regression analysis using dummy coding (used for categorical data) is shown in Table 5.9. The negative and positive signs in the coefficients indicate preference of a lower level and higher level of an attribute respectively. The WTP values established from this dummy-coded model are displayed in Table 5.10.

[4] 430 participants x 11 choice tasks = 4730 responses

Conditional (fixed effects) logistic regression				Number of obs.	14,064	
(Iteration 4) Log likelihood = -2811.4599				LR chi2(12)	4677.67	
				Prob > chi2	0.0000	
				Pseudo R2	0.4541	
Choice	Coefficient	Std. err.	z	P>z	95% conf. interval	
ASC	4.273386	0.134738	31.72	0.000	4.009305	4.537467
Filling (reference)						
Full gap	-1.435166	0.085225	-16.84	0.000	-1.602203	-1.268129
Partial gap	-0.767303	0.080712	-9.51	0.000	-0.925494	-0.609111
No tooth gap	-0.022228	0.07928	-0.28	0.779	-0.177613	0.133158
Ortho gap	0.008178	0.087393	0.09	0.925	-0.163108	0.179465
False tooth gap	-1.054161	0.07295	-14.45	0.000	-1.197141	-0.911182
General Dental Practitioner (reference)						
Enhanced GDP	-0.165737	0.054858	-3.02	0.003	-0.273255	-0.058218
Specialist	0.074416	0.050971	1.46	0.144	-0.025486	0.174317
Dentist makes decision (reference)						
Shared	0.13918	0.053398	2.61	0.009	0.034522	0.243839
Patient	0.107982	0.053885	2	0.045	0.00237	0.213594
Tx Avoid (cont.)	-0.003038	0.001673	-1.82	0.069	-0.006317	0.000241
Cost	-0.0008432	0.0000395	-21.36	0.000	-0.000921	-0.000766
Model	N	ll (null)	ll (model)	df	AIC	BIC
m1	14,064	-5150.294	-2811.46	12	5646.92	5737.536

Table 5.9 Regression analyses (using dummy coding)

Choice	Coefficient	Std. err.	z	P>z	95% conf. interval	
ASC_wtp	5068.24	267.01	18.98	0.000	4544.92	5591.56
Filling (reference)						
Full gap_wtp	-1702.11	111.91	-15.21	0.000	-1921.46	-1482.76
Partial gap_wtp	-910.02	101.31	-8.98	0.000	-1108.58	-711.46
No tooth gap_wtp	-26.36	94.11	-0.28	0.779	-210.82	158.10
Ortho gap_wtp	9.70	103.66	0.09	0.925	-193.48	212.88
False tooth gap_wtp	-1250.24	105.80	-11.82	0.000	-1457.60	-1042.87
General Dental Practitioner (reference)						
Enhanced GDP_wtp	-196.56	65.51	-3.00	0.003	-324.96	-68.17
Specialist_wtp	88.26	60.14	1.47	0.142	-29.62	206.13
Dentist makes decision (reference)						
Shared_wtp	165.07	64.62	2.55	0.011	38.42	291.72
Patient_wtp	128.07	65.07	1.97	0.049	0.53	255.61
Tx Avoid (cont.)_wtp	-3.60	1.97	-1.82	0.068	-7.47	0.27

Table 5.10 WTP values established (using dummy coding)

Key valuation of care packages

There is preference amongst the sample to have a package of care, compared to no treatment, with a WTP £5068 (95% CI: 4545 – 5592; p<0.001). The negative cost co-efficient (-0.0008) suggests that in general, if all other attributes are held constant, then the public prefer packages of care that cost less.

5.5.3 Conditional logit model – effects coding

In **Table 5.11**, the results of the regression analysis for the effects coded conditional (fixed effects) logistic regression model are shown. WTP values established for this effects-coded model are reported in **Table 5.12**.

Conditional (fixed effects) logistic regression				Number of obs.	14,064	
(Iteration 4) Log likelihood = -2811.4599				LR chi2(12)	4677.67	
				Prob > chi2	0.0000	
				Pseudo R2	0.4541	
Choice	**Coefficient**	**Std. err.**	**z**	**P>z**	**95% conf. interval**	
ASC	3.697832	0.119631	30.91	0.000	3.46336	3.932305
Filling	0.545113	0.050495	10.8	0.000	0.4461453	0.6440811
Full gap	-0.890053	0.060428	-14.73	0.000	-1.008489	-0.771616
Partial gap	-0.222189	0.053666	-4.14	0.000	-0.327373	-0.117006
No tooth gap	0.522886	0.05185	10.08	0.000	0.4212623	0.6245089
Ortho gap	0.553292	0.055129	10.04	0.000	0.4452415	0.6613415
False tooth gap	-0.509048	0.05399	-9.43	0.000	-0.614867	-0.40323
GDP	0.03044	0.031396	0.97	0.332	-0.031094	0.0919747
Enhanced GDP	-0.135296	0.030015	-4.51	0.000	-0.194124	-0.076469
Specialist	0.104856	0.027636	3.79	0.000	0.0506896	0.1590225
Dentist	-0.031198	0.051714	-0.6	0.546	-0.132557	0.0701599
Shared	0.056793	0.030089	1.89	0.059	-0.00218	0.1157656
Patient	0.025595	0.030376	0.84	0.399	-0.033942	0.085131
Tx Avoid (cont.)	-0.003038	0.001673	-1.82	0.069	-0.006317	0.0002406
Cost	-0.000843	.0000395	-21.36	0.000	-0.000921	-0.000766
Model	**N**	**ll (null)**	**ll (model)**	**df**	**AIC**	**BIC**
m1	14,064	-5150.294	-2811.46	12	5646.92	5737.536

Table 5.11 Regression analyses (using effects coding)

Choice	Coefficient	Std. err.	z	P>z	95% conf. interval	
ASC_wtp	4385.64	233.27	18.80	0.000	3928.43	4842.84
Filling_wtp	646.51	64.91	9.96	0.000	519.28	773.73
Full gap_wtp	-1055.60	74.46	-14.18	0.000	-1201.54	-909.67
Partial gap_wtp	-263.52	64.14	-4.11	0.000	-389.23	-137.80
No tooth gap_wtp	620.14	63.20	9.81	0.000	496.26	744.02
Ortho gap_wtp	656.20	71.64	9.16	0.000	515.79	796.62
False tooth gap_wtp	-603.73	72.84	-8.29	0.000	-746.49	-460.97
GDP_wtp	36.10	37.35	0.97	0.334	-37.11	109.31
Enhanced GDP_wtp	-160.46	35.73	-4.49	0.000	-230.49	-90.44
Specialist_wtp	124.36	32.43	3.83	0.000	60.80	187.92
Dentist_wtp	-97.71	38.10	-2.56	0.010	-172.39	-23.04
Shared_wtp	67.36	35.94	1.87	0.061	-3.09	137.80
Patient_wtp	30.36	36.22	0.84	0.402	-40.63	101.34
Tx Avoid (cont.) _wtp	-3.60	1.97	-1.82	0.068	-7.47	0.27

Table 5.12 WTP values established (using effects coding)

5.5.4 Mixed logit model – effects coding

Across most results from the mixed-logit model, the parameters did not change substantially in terms of their coefficient sign, significance, and magnitude of WTP values, compared to the conditional logit model. The results can be found in **Appendix M.** There was only one attribute that became significant in the mixed-logit model – that being a preference amongst the sample in terms of the future number of visits avoided with a WTP -£5.76 (95% CI: -10.19 – -1.32; p=0.011). The negative cost co-efficient (-0.0074853) suggests that the public prefer to not avoid future treatment visits, which appears to be counter intuitive.

Significant SDs of the attributes (partial gap, ortho gap, number of future visits and cost) might be indicative of preference heterogeneity within the sample for that attribute level.

5.5.5 Interaction analyses

The results of the pre-specified interaction analyses (shown in **Table 5.4**), and their interpretations are shown in **Table 5.13**

Respondent Characteristic	Outcome
Gender	No evidence in the data to suggest that females are more concerned compared to male respondents (p>0.05) about being left with a full or partial gap
Age	No evidence in the data to suggest that older respondents are less concerned compared to younger respondents (p>0.05) about the cost attribute or being left with a full or partial gap
Have children	No evidence in the data to suggest that respondents who have children are less sensitive to respondents without children (p>0.05) about being left a full or partial gap
Income (monthly)	No evidence in the data to suggest that high income respondents (>£2560 per month) are less concerned compared to those with low-income respondents (p>0.05) about cost
Political Party Identification	No evidence in the data to suggest that those respondents who align in voting conservative respondents are less concerned compared to those respondents who vote for other political parties (p>0.05) about cost or being left with a full or partial gap
Previous orthodontic treatment	No evidence in the data to suggest that those respondents who have had previous orthodontic treatment are more concerned compared to those respondents who have not (p>0.05) about orthodontic closure of the gap or having treatment done by specialist
Anxiety - having a filling?	No evidence in the data to suggest that those respondents who were anxious about having a filling are more concerned compared to those respondents who are not (p>0.05) about having an outcome that was not a filling (e.g., full gap, partial gap, and no gap)
Anxiety - having an extraction?	No evidence in the data to suggest that those respondents who are anxious about having an extraction are more concerned compared to those respondents who are not about having an outcome that leaves a partial- and full gap outcome (p>0.05). There is evidence to suggest that those who are more anxious about having an extraction were more concerned about having an outcome that leaves no tooth gap (p=0.024)
How dental care provided	Evidence in the data to suggest that those respondents who pay privately are less concerned compared to those respondents who are not, in terms of cost (p=0.007). However, there is no evidence to suggest those who pay privately are more concerned about being left with a full or partial gap (p>0.05)
Previous filling of cFPM	No evidence in the data to suggest that those respondents who have had a previous filling in their cFPM compared to those to those who have not had a filling on cFPM would want this tooth to be filled (p>0.05).
Previous extraction of cFPM	No evidence in the data to suggest that those respondents who have had a previous extraction of their cFPM are less concerned compared to those who have not had an extraction on cFPM in terms of being left with a full-, partial- or no tooth gap (p>0.05).
Example given to help with DCE	No evidence in the data to suggest that those who were given the high-cost DCE example are more concerned compared to those who were given the low-cost example (p>0.05) about cost

Table 5.13 Results of interactions hypotheses testing, including the nested methodological experiment

During interaction analyses, an additional two ‘significant’ interactions, which were unexpected, nor based on a priori assumptions were observed:

• There is evidence in the data to suggest that respondents who have children prefer to have a filling compared to no tooth gap and are less sensitive to this compared to respondent who don’t have children (p=0.030)

• There is evidence in the data to suggest that respondents who have had a previous filling on their cFPM prefer to have a filling compared to replacing the gap with a bridge and are more sensitive to this compared to respondents who have not had a previous filling (p=0.006)

5.6 Discussion

The following section discusses the interpretation, and implication of the DCE findings, followed by the strengths and limitations of this study.

5.6.1 Interpretation and implication of DCE findings

In the context of managing cFPM, the public prefer to have a package of care, compared to no care, makes sense as the sequalae of choosing no treatment in this scenario (where child will likely suffer further pain and require future treatment that is more difficult, expensive and have less certain results) is likely to result in a demonstrable reduction in quality of life (Leal et al., 2012; Grund et al., 2015; Taylor et al., 2018).

It should be noted that both the effects- and dummy-coded model report the same results. The effects-coded model provides absolute WTP values for all levels within each attribute and can be used as a measure of benefit in the decision analytical model (see Chapter 6). To minimise confusion, and permit pragmatic and simpler interpretations of the DCE, the remaining discussion will primarily focus on the dummy-coded model results.

There is no evidence of a clear preference for a filling over the gap being closed, post extraction, naturally by mesial migration. This could be explained in that both treatment options result in the same outcome, that is a functioning unit with no gap. Anecdotally, it could be expected that a filling would be favoured over an extraction, as the former is a much less invasive, or anxiety-provoking procedure. In isolation, financially there is no difference to society or the NHS, as the UDA value accrued is the same for both options (British Dental Association, 2020). However, a micro-costing exercise used for the model (see Section 6.4.7) highlighted that on average, an extraction will cost the practice just over £20 more to undertake, compared to a filling, as this procedure is likely to require more time to complete.

In this study, as shown during interaction analyses, there was no evidence to suggest that previous treatment experience of filling and extracting a cFPM, as well being anxious about having a filling, influenced preferences for the gap attribute. However, there was evidence to support that being anxious about having an extraction influenced the need to have no gap, or perfect closure, as the outcome. A possible explanation is that those anxious about having an extraction may prefer to have the most favourable outcome, post extraction. If they had to have an extraction, despite their anxieties, they want the best outcome as a reward for overcoming their fears. Despite limited evidence existing to suggest statistical significance that previous treatment experiences influenced preferences, prior experiences or memories for some parents may explain why they would choose the less invasive option of a filling compared with the more invasive, and potentially painful option, of an extraction. Ultimately, despite the limited clinical evidence for perfect spontaneous closure (Eichenberger et al., 2015; Somani et al., 2022), if the cFPM can be extracted (developmentally at the correct time with favourable prognostic radiographic features (Patel et al., 2017)) or filled and retained, then the public appear unequivocal in their preferences.

Of course, patient factors such as cooperation of the child etc. should be taken into consideration. More recently, one key piece of information to help make the decision is acknowledging the presence or absence of the third permanent molar (TPM) (Ashley & Noar, 2019). If the TPM is present, then extraction of the cFPM may be the preferred option, as removal has been shown to reduce TPM impaction (Bayram et al., 2009). Alternatively, if the TPM is absent, then a more restorative approach could be adopted to prevent only leaving one standing molar in the quadrant.

A preference does exist for a filling over extraction, which leaves a full gap or a partial gap. Thus, the results suggest the public would be willing to pay £1,702 for a filling to avoid a full gap and £910 for a filling to avoid a partial gap. An unfavourable outcome of a full gap or partial gap is something that all members of public, and not just those with children, would not want a child to have. A recent critical review of existing literature reported that in adults, retention of teeth was associated with better OHRQoL (Tan et al., 2016). These findings are corroborated in an earlier qualitative study, which demonstrated that for some adults, the loss of a tooth was devastating and disruptive (Rousseau et al., 2014). This concept of disruption can likely be explained by the functional impairment of tooth loss (Tan et al., 2016). Alternatively, it could be that the negative aesthetic consequence of having a full tooth gap explains this preference. Although negative OHRQoL due to missing teeth is more prominent with anterior teeth, there may be some suggestion that this may also be the case for molar teeth (Tan et al., 2016). Despite a filling being preferred to a partial gap, those who had experience of having an extraction of their cFPM were less concerned by having a partial gap (p=0.024). It could be that this cohort acknowledge that function is not completely compromised, by losing one tooth (or molar) unit, a finding reported by some adults (Rousseau et al., 2014). Similar explanations to being anxious for an extraction, and preferring complete closure, can be proposed for those who were more sensitive to having a partial gap as an outcome.

There is preference for a filling over the gap being closed with a bridge following removal, with a WTP of -£1250.24. It could be that the 5-year delay in having to wait for a bridge and being left with a full tooth gap in the interim, explains this preference. Alternatively, it could be the public prefer for their child not to have a prosthesis in their mouth. Furthermore, as shown in the interaction analyses, those with previous experience of having a filling on their cFPM, compared to those who had not had a filling, preferred having this tooth filled over replacing the gap with a bridge. It could be that the respondent's prior experiences may have influenced this despite high levels of patient satisfaction (Durey et al., 2011) and good survival rates at 10 years reported (Burke & Lucarotti, 2012), although previous bridge experiences were not specifically obtained from the sample.

In contrast, there was no evidence of the public preferring closure with orthodontics over a filling. The DCE implied this treatment would be successful, although in reality, complete space closure following extraction of a cFPM orthodontically is not guaranteed (Cobourne et al., 2014; Chua & Felicita, 2015). Therefore, this lack of evidence for a preference could be explained by 'assumed' successful outcomes of prior orthodontic treatment of respondent's children (37.2% (n=122)) or indeed themselves (21.4% (n=92)). It is unlikely that for most of these respondents, they would have had a first molar space closed orthodontically: however, it was not within the remit of this thesis to specifically ask this question. It could be expected that those who have had previous orthodontic treatment would have higher preference for orthodontic closure of the gap, assuming it was a positive experience. However, there was no evidence to suggest this is the case. It could be argued that a more plausible explanation, is that respondents had no preference for orthodontically closing the space, in comparison to a filling, as it has the same eventual outcome as a space that is perfectly closed following the ideal removal of a cFPM. In fact, when comparing preferences between closing the gap spontaneously compared to orthodontics, there was no evidence of a difference in values. Significant preference heterogeneity existed for both of these attributes within the sample, which corroborates these findings. The implications of these preferences have on the overall decision making for cFPM are discussed in Chapter 7.

There is strong preference for attending a GDP with the results suggesting the public would be willing to pay £197 to avoid having the treatment carried out by a dentist with enhanced skills. It appears that despite having additional training, knowledge and skills, the public would prefer any treatment to be carried out by a GDP over a non-specialist dentist with enhanced skills. The most likely explanation is that the respondents, and indeed the public, value the relationship they have with have with their own GDP, when compared to an enhanced practitioner. It could be that they trust, and have greater confidence, in a GDP managing a child with a cFPM, based on their own experiences. An alternative explanation is that the concept of a dentist with enhanced skills is unknown to most respondents, with only 13.7% (n=59) having reported to receiving care from such a practitioner. This may be a true reflection of access, or it could have been confused as a specialist by some. Dentist with enhanced skills are an integral part of future dental commissioning, as they aim to provide patients with expedited access to more 'specialised services', closer to their home, thus reducing the need for unnecessary hospital referrals (NHS England, 2015). This finding has key policy implications, as if future policies are keen to utilise dentists with enhanced skills, then further education of the public is likely to be of help to mitigate any concerns, or confusions, they appear to have. The patient, profession and policy implications of these findings are discussed further in Chapter 6.

In contrast, there was no evidence of a preference in attending a specialist over a GDP. It is clear there is no strong preference between a GDP and specialist, when managing cFPM in children. It may be that the public recognise the extra value of a specialist whilst balancing against the trust/familiarity they have with their own GDP. It was noted that 24.7% (n=106) of respondents had accessed some form of specialist care, and potential positive experiences may account for this lack of preference. Alternatively, the extra value of a specialist may not be understood, and the public are equally happy to see either.

There seems to be a preference for shared decision making and the parent making the decision compared to the dentist making the decision. The WTP values for these are £165 and £128 respectively. It is likely that patient-centred care approaches to decision making are valued and should be adopted when making decisions regarding cFPM. Historically, paternalistic models of decisions about healthcare were made solely by the healthcare professional, on a basis that 'they know best' (Asa'ad, 2019). This was demonstrated in an earlier study, which reported that a patient will choose, and have treatment, based on whether the dentist recommends it or not (Gilmore et al., 2006). However, recent NICE shared-decision guidance promotes shared decision-making as the accepted standard in healthcare. This collaborative process should involve person and their healthcare professional(s), detailing risks, benefits and consequences, culminating in reaching a joint decision about what care to receive (NICE, 2021). The concept of shared decision-making, in the wider context of managing cFPM, is discussed further in Chapter 7.

There is no evidence for a preference for avoiding 2, 15 and 33 future treatment appointments in the conditional logit model. This could be explained that respondents were unable to visualise or conceptualise 33 future treatment visits, rather than just future visits, as anecdotally, it is likely only a small proportion of respondents will have had such a number of intervention visits. That being said, the prevalence of reported treatments is greater in older adults, as they likely to have longer treatment history, compared to younger members of the population (Morris et al., 2011). Alternatively, it could be that the assumptions of the conditional logit were too strict. When the data was analysed using the mixed logit model (and effects coding), the preference became significant. However, the negative co-efficient remained, suggesting the sample preferred to not avoid future visits. This is an interesting result, as anecdotally it would be expected that reducing the number of future intervention visits would be a positive. It could be that public are more concerned with the short-term outcome, i.e., the gap being closed or not, rather than the process of saving future appointments, thus not considering these outcomes when trading. Alternatively, it could be a genuine disinterest in the long-term appointments that could be saved. Interestingly, the preference for avoiding future visits avoided showed significant preference heterogeneity within sample. This suggests that within the sample, this attribute drove decision-making, with a proportion of respondents placing relative importance on this attribute during their decision-making.

It was assumed from the interaction analysis, those respondents who had higher monthly incomes would be less concerned about the cost attribute. In this DCE, there was insufficient evidence to suggest this was the case. This does contradict trends demonstrated in other WTP studies, which suggest higher income was associated with a higher WTP (Tan et al., 2017). Similar observations were noted in those who were older or aligned as a Conservative politically. As both of these attributes were assumed to link to higher levels of income, it could suggest that cost is not as important to the public, when considering an intervention in children, as other attributes, such as management etc. It has been shown that in health, household income has a mixed effect on child health outcomes, with the link between income and parental health-related behaviours such as decision making, are more uncertain (Cooper & Stewart, 2021). It is worth noting that in this review, none of the studies were dentally focussed. Alternatively, it could be that despite advising respondents that these would be out of pocket costs, the fact that in the UK parents do not pay for their child's treatment under the NHS, means personal income may not have been taken into consideration when making these choices. However, it is worth noting that those who pay privately for their care, were less concerned by the cost attribute (p=0.007), as they may have greater ability to pay. If out-off pockets payment system existed in the UK, in the DCE, there might be different results.

5.6.2 Strengths and limitations

The results should be interpreted taking into consideration strengths and limitations associated with this DCE. These are presented in the following sections.

Sample and responses

A real strength was the sampling framework used in this DCE. This was achieved by using a robust online panel, hosted by NATCEN, and a random probability sample attempts that ensured sufficient representativeness of the UK population (Hays et al., 2015). In general, the sample that responded was fairly representative of the UK population. NATCEN do usually offer offline representation; however, this was not possible for this study due to the impact of COVID-19. The difference this could have made will never be known. It is likely that specific population groups were excluded, or under-represented, as shown by high proportion of white respondents. This is not uncommon when using internet panels (Hays et al., 2015). Furthermore, using the 2009 ADHS survey, it is known that ethnic minority groups are statistically significantly less likely, compared to Caucasians, to have fillings and dental extractions (Arora et al., 2016).

Another key strength of using NATCEN was the sample obtained. As described in 4.4.6, the rule of thumb sample size collection suggested only 90 respondents were required. NATCEN were able to provide 430. It is worth noting that financial incentives (£10) were provided by NATCEN for each completed response. It has been shown incentives can boost recruitment whilst providing a token of appreciation for their time in taking to complete the study (Resnik, 2015). However, there is a concern that prospective participants may only participate in research if it is financially worth their while. In addition, it may be these participants might make poor choices because money has clouded their judgment (Resnik, 2015). In this study, the incentive was relatively low, and completion was voluntary. This is both a strength and a weakness, as all responses are likely not going to be influenced by financial incentive alone, however, there may be a response bias in that those who did not complete may have different oral health behaviours compared to those that did.

Despite the strengths of using the panel, it does bring some challenges. Members of these panels may be 'seasoned' survey completers, and thus answers may not reflect their preferences (Hays et al., 2015). To mitigate against this, timers were introduced into the survey to ensure that sufficient time was spent on each page, assuming that the information on that page was appropriately read. NATCEN were unable to give times for each completed DCE, and this is something that could be done in the future to ensure such surveys are completely with careful consideration by the respondent.

Attribute and level selection

A key strength was the use of qualitative interviews, expert opinion, and use of evidence to develop the attributes, and their levels, in the DCE. Using several approaches reduces the risk of not including attributes that important to respondents in the DCE. Involving qualitative methodologies in the DCE has been highlighted as an important component of the DCE design process (Vass et al., 2017). These interviews used were conducted appropriately (Vass et al., 2017) and analysed using recognised analytical approaches (Braun & Clarke, 2006), rather than being informal conversations, which strengthens their use in informing this DCE. Furthermore, including both adolescents and members of the public gave further clarity and contextualisation. The five members of the public were diverse in terms of age, gender, and previous dental experience. Despite not being respondents of the final public DCE, involving adolescents was important given the context of the choice task. This could be perceived as weakness, as their opinions on appropriate attributes may not be appropriate for a public DCE, as it has been suggested that involving just the target respondent group is likely to ensure a valid and acceptable DCE (de Bekker-Grob et al., 2012; Barber et al., 2019). This is discussed further in the discussion chapter (see Section 7.3).

Another key strength was using the expert panel in attribute selection. They transformed and shaped the 'information needed to help decision' attribute, so it was more meaningful.

The panel were concerned that level of pain was not included as attribute. This certainly could be a weakness, as differences in procedural pain between extraction and restorations are discussed in the literature (Ghanei et al., 2018) as well as being mentioned briefly in the interviews (see Section 4.7) . However, operationalising pain relating to the condition (i.e. symptoms of cFPM) in a meaningful manner, in the context of cFPM in this DCE would be challenging, as symptomatic teeth would almost certainly be removed (Taylor et al., 2019), thus, precluding a relevant option of restoration. There are examples of DCEs where pain has been incorporated into attributes (Poder et al., 2019; Shanahan et al., 2019), however, these did not focus directly on procedures, and more on how pain can be managed and reduced. To overcome this perceived weakness, pain (as sensitivity) was incorporated as part of this DCE in the description of the cFPM. In addition, procedural pain was equivalent across all options, as participants were informed that there should be pain at the injection site, as LA will be required for an active intervention, however, once anaesthetised, any procedural pain associated with filling or extraction should be equal. Furthermore, it was assumed that levels of pain could be inferred by the preference over number of future treatment visits needed, as each treatment needed would require an intervention that needs an injection. Therefore, less future visits mean less pain.

Regarding agreement on the levels, this was only done using the expert panel. There were three patient members on this panel, however, they were not fully representative of the choice task respondents (de Bekker-Grob et al., 2012). This process could have involved wider representative views, although, the piloting phase did not identify any concerns in terms of appropriateness of these levels.

Inclusion of a cost attribute

One of the key strengths was the inclusion of a cost attribute, which allowed the generation of marginal willingness to pay, to be used in a cost-benefit analysis (Lancsar & Louviere, 2008). Despite the inherent challenges faced in framing this cost-attribute within this DCE, its inclusion was preferred over other options, such a time to wait, which would then require conversion to a WTP. Furthermore, the piloting work demonstrated that for 9/30 respondents, the cost-attribute was the key driver in choosing the choice set, rather than trading off all the attributes provided, by always choosing the cheapest alternative or not making a choice at all. In trying to mitigate the concerns of the cost attribute, as detailed in the section 5.4.4, two completed example DCE choice tasks were included. It was hypothesised that those who received the example where higher cost attribute was selected should be more concerned by cost, however, there was no evidence to suggest this was the case. Concerns have been proposed that respondents living in a healthcare system that is publicly funded may ignore the cost attribute, as the cost are not borne by them directly (Ratcliffe, 2000; Genie et al., 2021). However, in dentistry, for most, treatment is only part funded by the NHS, and therefore the concept of out-of-pocket costs does exist. This concept however is not relevant children's oral health interventions, where there are not out of pocket costs. As such, it could be that values drawn from this DCE may not reflect individual's true preferences (Johnson et al., 2011).

The concept of ignoring the cost-attribute was recently observed in a study that focussed on patient preferences, using a DCE, for personalisation of chronic pain self-management (CPSM) programmes in the UK. The sample was split so that one half completed the DCE with a cost-attribute included, whereas the other half completed the exact same DCE with the cost-attribute removed. For those who had the cost-attribute included, respondents most commonly ignored this attribute. Interestingly, including a cost attribute did not alter the structure of preferences but it did result in a less consistent responses. Furthermore, respondents who had experience of paying for health services (e.g., in a private market) were less likely to ignore cost (Genie et al., 2021).

However, even in circumstances where individuals may be used to paying for all, or part, of their healthcare, including a cost-attribute to generate WTP values from DCEs, that is subsequently ignored, raises concerns. Respondents in a DCE may apply heuristics that involves them ignoring, or not considering all attributes that describe the alternatives. Ignoring non-cost attributes are unlikely to affect WTP values. However, if respondents choose to ignore the cost-attribute, this could lead to biased utility and overestimated WTP values, even if it reflects the true preferences of respondents (Hole et al., 2016; Sever et al., 2019).

A recent DCE study attempted to demonstrate the effects non-attendance to the cost attribute had on WTP values. The DCE, which estimated patients' WTP for dental care at the dental school clinic in Croatia, found that almost every second respondent appeared to have ignored the cost attribute. They found that co-efficient, after accounting for non-attendance to the cost attribute, were more than two times lower than welfare estimates from a traditional multinomial logit model and mixed logit model, which led to overestimated WTP values (Sever et al., 2019).

Validity Tests

In the DCE, 91.4% (n=393) of participants passed the dominance test. A recent study reported that across a random sample of 112 DCEs used in healthcare in the year 2015, 25% (n=28) of studies included a dominance test with a range of 0%-21% being reported for participants choosing the non-dominant choice (Tervonen et al., 2018). Failure does not necessarily indicate a lack of attention or irrationality, as it would be expected under the random utility theorem (McFadden, 1974) some participant will fail the dominance test (Tervonen et al., 2018). To date, there is no consensus on how to interpret dominance tests results, or whether their choices should be included in the final analysis (Tervonen et al., 2018). Therefore, it can be summarised that ‘failure’ rate of the dominance test 8.6% (n=37) in this DCE is within accepted levels.

Test-retest reliability, in the context of DCEs for health valuations, allows choice consistency to be derived. That is the consistency of respondents’ choices of health states from choice sets, when exactly the same choices sets are presented at two separate time point (Gamper et al., 2018). There appears to be a lack of consensus on an acceptable choice-consistency level DCEs (Gamper et al., 2018; Pearce et al., 2021). It is accepted that choice consistency in a DCE is very likely to be negatively influenced by high task complexity, linked to the number of attributes and alternatives (Swait & Adamowicz, 2001). Therefore, although 24.4% (n=105) failed the test re-test reliability tests in the thesis, it could be concluded that this level is acceptable given the high complexity of the DCE given the attribute and levels chosen.

5.7 Conclusion

In summary, this chapter has described the development of the DCE, explored and quantified the strength of individual public preferences towards managing cFPM.

The results confirmed that the UK public do not prefer to restore or extract cFPM in a child, providing the resultant space was closed spontaneously or orthodontically. In addition, the public prefer this management to be carried out by a GDP or referred to a specialist, with any decision being made by the parent alone, or in conjunction with the dentist. Clinical and policy changes for managing cFPM should not just focus on clinical effectiveness, or cost savings, but should incorporate the preferences of those who are users of the service, or indeed indirectly, through taxation, contribute to it.

The results of this DCE will be incorporated into the economic evaluation model, as a measure of benefit, described in Chapter 6. The next chapter will discuss the methodology, methods, results, and discussion of the decision analytical model, built for managing cFPM in children.

Chapter 6. Decision Analytical Modelling

6.1 Introduction

An overview of decision analytical modelling methods was presented in Chapter 2. Key principles of economic evaluations such as perspective, discounting, horizon, and deterministic/probabilistic sensitivity analyses were discussed. In addition, the concept of DAMs and the strengths and limitations of common model structures were explored.

In section 6.2 of this chapter, further methodological concepts relating to DAMs are explored. Section 6.4 and 6.5 report the methods and analyses used for a decision analytic model that explores the cost-effectiveness of an initial decision for a cFPM. Section 6.6 will present the findings, including the incremental net benefit of the options modelled over a life-time horizon, with marginal WTP values, derived from the DCE (discussed in Chapter 5) used as a measure of benefit. Sensitivity analyses will be explored. Finally, section 6.7 will discuss the findings and address strengths and limitations of this particular aspect of the thesis. These findings will be discussed in combination with the findings of the qualitative interviews (see Chapter 3) and discrete choice experiment (see Chapter 5), in Chapter 7, to consider the management of cFPM in a wider context.

6.2 Methodology

As discussed previously in Chapter 2, key principles of economic evaluations such as perspective, discounting, horizon, and deterministic/probabilistic sensitivity analyses are discussed. In addition, the concept of DAMs and the strengths and limitations of common model structures were explored. Model conceptualisation and development are discussed in a practical guide to decision modelling for health economic evaluation (Briggs et al., 2006) and good practice guidelines, outlined by ISPOR Task Forces (Briggs et al., 2012; Caro et al., 2012; Eddy et al., 2012; Roberts et al., 2012). How the model was conceptualised, built, and analysed is discussed in the methods section (Section 6.4) of this chapter. The following sub-sections provide a brief overview of the more nuanced methodological decisions pertinent to model development and analysis.

6.2.1 Parameter estimations

Transition probabilities, costs and measures of benefit are derived in many ways. The following sub-sections focus on issues relating to each of these parameters.

Transition probabilities

Transition probabilities are populated by synthesising evidence from range of sources across different patient groups and health care settings (Petrou & Gray, 2011b; Drummond et al., 2015). Sources of evidence can be derived from randomised controlled trials (RCTs), observational studies, databases, case series, expert opinion and/or secondary analyses (such as systematic reviews and meta-analyses) (Briggs et al., 2006; Cooper et al., 2007). It is understood that well-conducted RCTs offer the best source of primary data to evaluate relative effectiveness (Cooper et al., 2007; Briggs et al., 2012). If sufficient trials do exist, then combining data and undertaking meta-analyses to derive one pooled point of relative effects is possible (Higgins et al., 2022). It is accepted that there may be situations where trial data may not be readily available (e.g., due to its cost of collection, practicality, short-term follow up etc.) or that heterogeneity amongst studies precludes evidence synthesis. Lamont and Clarkson (2022) raise this issue, and propose that developing core outcome sets for dentistry, will in turn promote the success of high-quality trials, to overcome this concern. Developing core outcome sets is certainly a sensible approach and would reduce some of the problems discussed previously. However, developing core outcome sets is not necessarily the panacea, given the time it takes to design, conduct, and report a trial, meaning these studies will not be available for meta-analysis for several years. In addition, the timeframe will be lengthy for sufficient studies to accrue, during which time the outcome set may require further development and limit the ability to overcome some of the problems mentioned.

Developing systematic search strategies, to identify the most relevant piece of evidence, for each individual parameter input value could be considered good practice. However, this is time-consuming, and may not be hugely effective. As such, a more efficient approach is to undertake focused searches that focus on parameters expected to have the biggest influence on the model. Adopting this process considers the applicability of the parameter for the given decision problem and narrows the searching needed for some parameters. This may raise a question about cherry-picking certain evidence. However, transparency of decisions regarding model structure, equations and assumptions and parameter values must be clearly described to support transparency in understanding the model (Eddy et al., 2012; Caro et al., 2012; Briggs et al., 2012).

Time dependency is important to consider when identifying parameters for use in a decision model. It could be assumed that all transition probabilities are fixed with regards to time. This is a simplification of reality, and a decision is required to be made whether this assumption for these probabilities being constant reflects reality or is it likely to generate misleading results. Transition probabilities may: a) vary across the model, or b) vary within a health state, with both needing to be considered if time is not assumed to be constant. For example, the probability of a composite restoration requiring replacement might increase over time because each time a replacement is carried out, more tooth structure is removed, and making the chance of failure greater. Thus, the transition probability from a composite with a replacement to composite that has been replaced should be higher in the second cycle compared the first. However, sufficient data may not exist to accurately determine how the transition probability should change over time (Briggs et al., 2006).

Obtaining probabilities directly from evidence synthesis for use in models can be challenging, as rates are more routinely reported than probabilities in the literature (Sun & Faunce, 2008; Briggs et al., 2006). The time period used to estimate events rates are unlikely to be equal to the chosen cycle length of the model. Therefore, using rates directly from the literature in the model will bias the results from the model. To overcome this, converting a rate into a probability over time can be done. More complex methods can be used for multi-state models (Fleurence & Hollenbeak, 2007). If time-dependent probabilities are required, these can be derived from patient-level time to event data, often from some sort of longitudinal study, using survival analysis (Briggs et al., 2006; NICE, 2022a).

Costs

The two most common cost categories in economic evaluations are: direct costs and indirect costs.

Direct costs cover all goods, services and resources that are consumed in the provision of any given healthcare service or intervention, including its subsequent follow-up care. These costs depend on the perspective chosen (see Section 2.2.1) and should be relevant to the study stakeholders' interests. Indirect costs are sub-divided into structural (e.g., administrative support costs, overhead building costs, maintenance costs etc.) and productivity (e.g., productivity loss due to illness). Another indirect cost to consider is the capital costs, which are not intervention specific but may require to be costed. These costs are usually captured by annuitsing the initial outlay, accounting for purchasing, using, and maintaining an asset, over the expected lifespan. These assets may have residual value when traded in or sold. In these circumstances, the residual value would be included in the equivalent annual cost calculation. Direct and indirect costs can either be fixed (remain the same irrespective of the quantity) or variable (vary with the level of output). Combined, fixed and variable costs provide the average total cost considered in an analysis (Garrison et al., 2018; Ternent et al., 2022).

Two main approaches to obtain costs are the bottom-up (micro-costing) and the top-down. Micro-costing requires identification of each resource being used to provide that good, obtaining pertinent cost information for that resource, and then summing up all resources that would be required per patient. This approach provides an accurate estimation of costs, however, is time-consuming. In contrast, the top-down approach utilises cost data from routine data sources, which is based on national averages. This approach is less time-consuming but may not reflect the true costs of an intervention. Nevertheless, using a combination of both approaches for a model is common practice (Xu et al., 2014; Ternent et al., 2022).

Benefits

As previously explained in Chapters 2 and 5, a DCE can support the valuation of health, and can derive a WTP if a cost attribute is included. It has been shown that DCEs are able to provide reasonable predictions of health-related behaviours (Quaife et al., 2018) and can be incorporated into a model (Grosse et al., 2008; Pol et al., 2010; Tinelli et al., 2016) assuming the DCE and model structures are as congruent with one another as is feasibly possible. Marginal WTP values, estimated from a DCE, can be embedded as a measure of benefit and used in a model adopting a CBA framework (McIntosh, 2006; Ryan et al., 2008; Mott et al., 2020).

Despite best efforts, model outcomes are dependent on data input. Therefore, an important feature of any model is to address uncertainty regarding within model.

6.2.2 Dealing with uncertainty

The concepts of DSA and PSA have been discussed in section 2.2.1. As described, several parameter uncertainties can be undertaken at one time with a set of input parameter values being drawn by random sampling from each distribution.

Normal (Gaussian) distribution is the most common form of continuous distributions used to capture parameter uncertainties. A normal distribution, assuming the sample size of the mean parameter value is large enough to justify a normal assumption, can allow a random value between negative and positive infinity to be used, most often one standard deviation (Briggs et al., 2012).

If data do not follow a normal distribution, then estimating a distribution is based on parameter type and standard statistical methods of estimation. Probability parameters that range between zero and one are often estimated using beta distributions. If estimated from logistic regression, then a lognormal distribution would be more appropriate. If there are numerous categories which sum into 1, then a Dirichlet distribution would represent the uncertainty (Briggs et al., 2006, 2012).

Cost data can range from zero to infinity. They are based on counts of resource use weighted by resource unit costs. Therefore, it would be appropriate to use gamma or lognormal distribution. However, if costs are not highly skewed, and based on sufficiently large data, then fitting the normal distribution to the cost estimates could be applied (Briggs et al., 2012). Although care would be needed to check that implausible (e.g., negative values) are not incurred.

6.2.3 Validity and transparency

Modelling studies support policy decision makers in the allocation of scare healthcare resources. As such, transparency in decisions made regarding building the model and assessing its validity are key to ensure decisions are made with all available knowledge. Transparency is important regarding decisions made about the model structure, parameters, equations, and any assumptions used in the model. Validity refers to how well the model reflects reality and the real world (Eddy et al., 2012).

6.2.4 Presentation of results

The cost-effectiveness is normally summarised in terms of an ICER (total difference in cost of the intervention under consideration and the comparator divided by the difference in their effects). These can be displayed on a cost-effectiveness plane (See **Figure 6.1**), which is divided into four quadrants, with the incremental costs displayed on vertical axis and incremental effectiveness on horizontal axis. If the intervention is more effective and more costly (e.g., fall into north-east (NE) quadrant) or less effective and less costly (e.g., fall into south-west (SW) quadrant), then a decision needs to be made as to whether to adopt this or not. To help, the ICER can be gauged against the willingness to pay threshold line (λ) (the dotted sloped line on the cost-effectiveness plane).

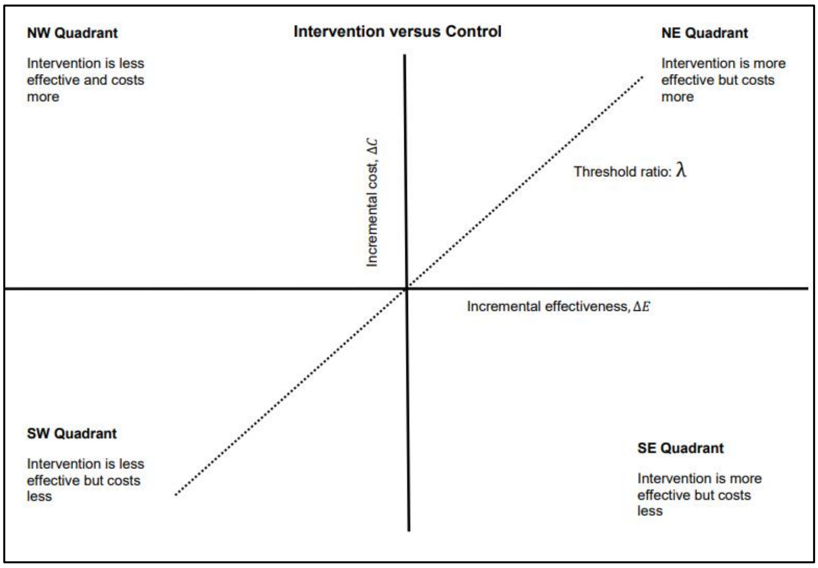

Figure 6.1: Cost-effectiveness plane (adapted from (Briggs et al., 2006))

For cost-benefit, the net-monetary benefit (NMB) can be established by subtracting the mean benefit from the mean cost for each strategy. The incremental NMB can be determined between alternative options. Incremental NMB is calculated using incremental benefit (ΔE – total difference in the benefits between two strategies), incremental costs (ΔC - total difference in the costs between two strategies) and a willingness to pay threshold (λ). Decisions can be made, in conjunction with the agreed willingness to pay threshold (λ), which in health usually takes the value of one. A value greater than one would mean the benefits obtained outweigh the costs, and less than one the opposite.

A benefit-cost ratio can be obtained by dividing incremental effectiveness (ΔE) by incremental costs (ΔC) between the two modelled strategies. A positive incremental NMB and/or incremental benefit-cost ratio indicates that the intervention is cost-effective, compared with the alternative option modelled, at the given willingness-to-pay threshold (Briggs et al., 2006; Drummond et al., 2015).

For a DSA, parameters that are changed often have no distribution e.g., the discount rate, or in changing parameter to a fundamentally different value etc. These results of each change can be presented as a scenario analysis.

For PSA, uncertainties can be represented in the cost-effectiveness plane (shown in **Figure 6.1**) as a cloud of points, with each point representing each iteration of the model during a Monte-carlo simulation. An additional method of reporting uncertainties obtained from a PSA in a CBA is to present a cost-effectiveness acceptability curve (CEAC) based on the comparison of two NMBs (Briggs et al., 2006). For each model iteration, NMB ($\lambda * \Delta E - \Delta C$) for intervention 1 and NMB ($\lambda * \Delta E - \Delta C$) for intervention 2 are compared. If the NMB for intervention 1 is higher, then it scores 1 and if it is lower it scores 0. This is then repeated in the PSA several times for each value of λ. At each value of λ, the probability (number of times it scores 1 / 1000) is calculated and plotted on the graph, as shown on **Figure 6.2**. This analysis assumes that the interventions are (i) perfectly divisible and (ii) mutually exclusive

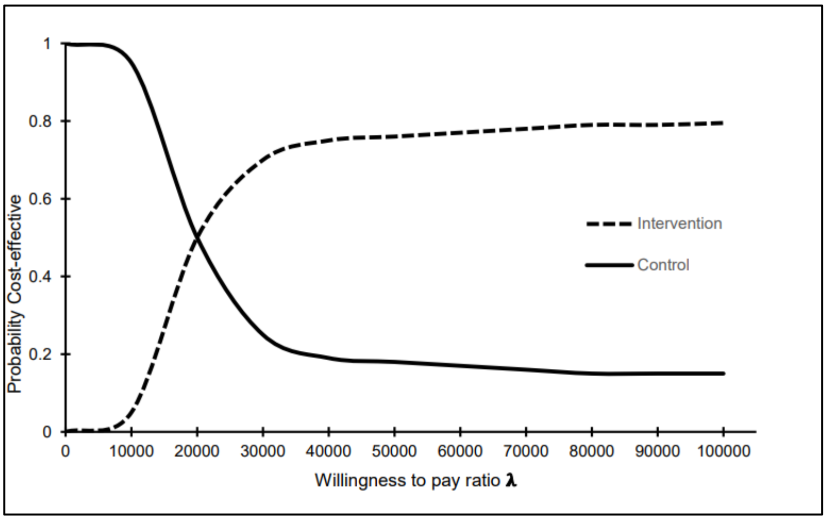

Figure 6.2: Cost-effectiveness acceptability curve (adapted from (Briggs et al., 2006))

In summary, section 6.2 has dealt with pertinent aspects of modelling that required more in-depth discussion. The remainder of this chapter will focus on the methods, including analysis (Section 6.4 & 6.5), results (Section 6.6), and subsequent discussion (Section 6.7).

6.3 Aim

To develop a decision analytical model to determine the incremental net-benefit of initial options to manage cFPM over a modelled life-time horizon.

6.4 Method

This section describes the building and analysis plan of the model, adopting the available practical guide (Briggs et al., 2006) and good practice guidelines (Briggs et al., 2012; Caro et al., 2012; Eddy et al., 2012; Roberts et al., 2012). Reporting in this chapter is in line with CHEERS 2022 recommendations and a checklist is included as **Appendix N** (Husereau et al., 2022).

6.4.1 Specifying the decision problem and conceptualising the model

As previously described in Chapter 1, the management of cFPM in childhood causes confusion, as several options are available both in the short- and long-term but with lifelong consequences. Comparing the management of cFPM does not lend itself well to trial methodologies and as such, can be better answered using a decision analytical model. Despite four cFPM being present in a child's mouth, this model will focus on a single lower cFPM only. The lower cFPM was chosen, as compared to the upper cFPM, there is more clinical uncertainty regarding the outcomes of treatment strategies that are available.

Conceptualising the model was undertaken by consulting with members of the research team and 'expert panel' (as described in Section 4.2). Initial panel feedback can be found in **Appendix O.** Ensuring the model would represent the disease process of cFPM appropriately and adequately address the decision problem (Roberts et al., 2012) were the key questions that were discussed.

6.4.2 Alternatives compared

The current main treatment strategies for cFPM are active monitoring, temporary filling followed by extraction, extraction, or restoration. Section 1.8.2 – 1.8.5 discuss these alternatives in more detail.

6.4.3 Choice of the model

Various model structures are available to support and inform decision-making under conditions of uncertainty. Their strengths and limitations are discussed in more detail in Section 2.5.2. The long-term costs and benefits of alternative interventions for managing cFPM remain unclear. Therefore, in the context of such uncertainties, a decision analytic model was determined to be the most appropriate approach to estimate the long-term costs and benefits of these decisions. A decision-tree structure was not appropriate given the complexity of the interventions being modelled. Children with cFPM were independent from each other and given no interaction between patients was feasible, sophisticated models like dynamic models and discrete event simulations with interaction were excluded.

A simple Markov (cohort) model was similarly ruled out as future transitions are dependent on previous states. Additionally, varying the age at which a patient enters the model is an important characteristic that not only reflects reality in managing cFPM but is likely to provide valuable clinical and policy information. Therefore, given the decision problem under investigation, an individual patient level micro-simulation model was determined to be more appropriate, despite being computationally more burdensome. This permits the progress of individual children, accounting for variations between children, over time and allows accumulation of individual child's history (or memory) to determine transitions between states, costs and health outcomes (Briggs et al., 2006).

6.4.4 Individual patient level microsimulation model structure

The microsimulation model seeks to reflect reality and represent the patient care pathways for the managing of cFPM. The conceptual model, as shown in **Figure 6.3**, consists of health states and events. These states and events, including assumptions regarding these, are described in **Table 6.1** and **Table 6.2**. In **Figure 6.3**, the health states are identified in purple, the events in blue and initial decision as orange. The arrows represent the possible transitions allowed from one state to another. Arrows point in one direction means they can't revert to that state. The arrow that loops back to the same state indicates the patient may remain in this state after each cycle.

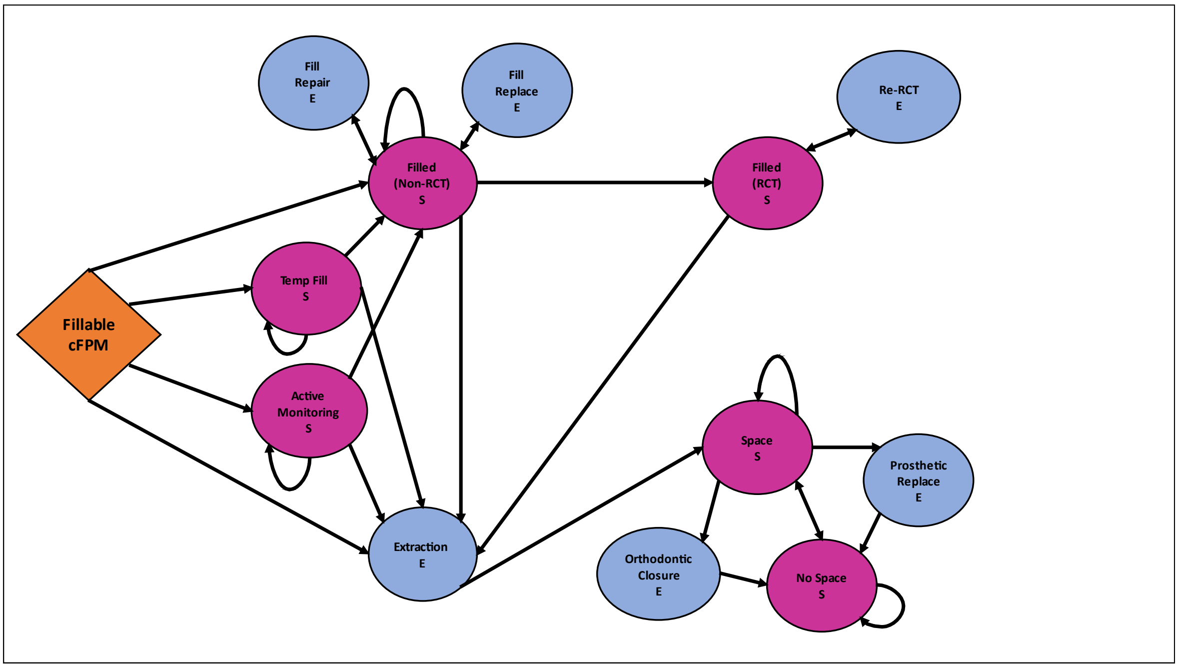

Figure 6.3 Conceptual individual patient-level microsimulation model for managing cFPM (Orange – original choice; blue – health events; purple – health states; arrows – transition probabilities)

These states are mutually exclusive, meaning a patient can only be in one state a one point in time. The absorbing state in this model was death. The start age of the model was 7 years-old to ensure the cFPM was allowed to be compromised. Ten thousand patients were sampled from a distribution that included 35% of 7-year-olds, 35% of 8-year-olds, 20% of 9-year-olds and 10% of 10-year-olds. This distribution was used to reflect the fact that patients with cFPM are more likely to present with these teeth at an earlier age. Once in the model, changes occurring based on the transition probabilities between the health states.

Initial validation of the conceptual model (see **Figure 6.3**) was based on expert opinion (as described in Section 5.2). Members of the panel amended and annotated an initial draft of the model, in addition to providing written and verbal feedback as part of a group discussion in a panel meeting. Refinements were made, and feedback was given to the panel. The final model was discussed and agreed within the supervisory team. The same model structure was used for an MIH and dental caries as the pathways for these different disease structures were assumed to be the same.

Health States	Description (and assumptions)
Active Monitoring	Patient has had the cFPM actively monitored. No interventions are undertaken when initially entering the model. However, at certain ages, the following percentage of patients will move out of this state: 8-years-old (10% will choose to have decision) 9-year-old (72% of those left will choose to make decision) 10-years-old (16% of those left will choose to make decision) 11-years-old (2% of those left will choose to make decision) 12-years-old) (Everyone has made the decision) Expert opinion determined that 40% of patients leaving active monitoring will have an extraction completed, and the rest will have a filling.
Temporary Filling	Patient has had the cFPM temporarily filled with glass ionomer cement with no active dental tissue removal. This material was assumed to be the same for carious and hypomineralised teeth. Preformed metal crown were not used as their placement is relatively uncommon for carious and hypomineralised teeth in the UK (Taylor et al., 2019). This will not be replaced or offered after 9.5 years old (maximum 5 cycles). All patients will thereafter have the tooth extracted, with the maximum chance of spontaneous alignment occurring at this point (Cobourne et al., 2014).
Filled (Non- RCT)	Patient has the cFPM filled with composite resin as a definitive restoration. This is assumed to be the same for carious and hypomineralised teeth. Amalgam is not permitted for use in children (Article 10 (2) of Regulation (EU) 2017/852 on Mercury). Preformed metal crowns, onlays, inlays and crowns are relatively uncommon as definitive management options for carious and hypomineralised teeth in the UK (Taylor et al., 2019).
Filled (RCT)	Patient has undergone conventional root canal treatment of the cFPM. As this is a single surface lesion, this is restored with a composite restoration (Mannocci et al., 2021).
Space	Patient has had the cFPM extracted and is left with a full unit space.
No Space	Patient has had the cFPM extracted, and has had the space closed either naturally, via orthodontics or using a resin-retained bridge.

Table 6.1 Description, and assumptions, for each health state in the model

Health Events	Description
Fill (Repair)	The restoration on the cFPM requires a repair. It is assumed this can be completed on a maximum of two separate occasions. This is the same for carious and hypomineralised teeth.
Fill (Replace)	The restoration on the cFPM requires a complete replacement. It is assumed this can be completed a maximum of one occasion before endodontic management is required. This is the same for carious and hypomineralised teeth. Once completed, it will be assumed to be restored to the same condition as the initial filling. For this reason, the same initial failure rate will be used.
Re-RCT	The cFPM has had a conventional root canal treatment for the cFPM; however, this has failed. A conventional non-surgical re-RCT is undertaken, with replacement composite restoration placed. This can be completed a maximum of one occasion. Thereafter the cFPM will be extracted.
Extraction	The cFPM has been extracted using a non-surgical extraction technique.
Orthodontic Closure	The resultant space left post-extraction of the cFPM has undergone orthodontic closure. This will be completed in a maximum of 2.5 years (5 cycles). Once the patient has reached the age of 19 (maximum 24 cycles) this treatment will no longer be offered as the provision of NHS orthodontic treatment is not routinely available for adults (NHS, 2020).
Prosthodontic Replacement	The resultant space left post-extraction of the cFPM has been replaced with a resin-retained bridge. The bridge can't be repaired and will only be replaced. This can be replaced a maximum of three times in their life, before they have been assumed to have a full space.

Table 6.2 Description, and assumptions, for each health event in the model

6.4.5 Time horizon

The model employed a lifetime horizon, following patients to the maximum age of 100-years-old (or death if this occurred earlier). It was deemed appropriate to consider a lifetime horizon for the base-case scenario as this would sufficiently capture relevant long-term costs and outcomes of initial decisions made for cFPM being modelled in this thesis. Routine all-cause mortality rates for 2019/20 were used (Office for National Statistics, 2019). Data from 2019/20 were used to reflect the remaining parameters that were modelled pre the COVID-19 pandemic.

6.4.6 Cycle length

The cycle length used was 0.5 of a year (or 6 months). This meant there were 186 monthly cycles in the model (start age 7-years-old to 100-years-old). The cycle length was chosen to reflect the accepted timeframe between dental check-ups. The corresponding model input parameters were adjusted to reflect this.

6.4.7 Perspective

An NHS perspective was adopted for the cost-benefit framework analysis. An NHS dental practice perspective was adopted for the cost-effectiveness analysis. Further details are provided in sections 6.4.8 and 6.4.9 regarding the costs and benefits included for each analysis.

6.4.8 Model parameters

The parameters used to populate individual patient-level microsimulation model were transition probabilities between health states, and the costs and benefits attributed to each state. Transition probabilities for dental caries and MIH were obtained separately for situations when the condition made a difference e.g*.,* restoration rate, repair rate etc. Once the tooth required endodontic management, or an extraction had taken place, transition probabilities were assumed to be the same for both conditions there on in. Transition probabilities used are reported in **Table 6.3** and **6.4**. The cost data considered in the models are reported in **Table 6.5**, **6.6**, and **6.7**. The measure of benefit, for the CBA, are in **Table 6.8** and the effectiveness measure, for the CEA, are in **Table 6.9**. The approaches used to derive these model parameters are described in the following sub-sections.

Transition Probabilities

Model input parameters were determined using a systematic structured approach, using Medical Subject Headings terms, in conjunction with the relevant health states that required to be populated (See **Appendix P**). This search was run-on 27th April 2022 in PubMed. This search provided 1227 records which were manually checked. These studies were analysed for methodological quality and risk of bias. Formal assessment using quality assessment tools were not carried out. Instead, each record was scrutinised regarding relevance, reliability, validity, and applicability for the decision problem being modelled.

One published meta-analysis was chosen to provide transition probability estimates for the model in this thesis (Eichenberger et al., 2015). However, the majority of estimates came from observational studies (Kotsanos et al., 2005; Ricucci et al., 2011; He et al., 2017; Burke & Lucarotti, 2018b, 2018c, 2018d; Gatón-Hernandéz et al., 2020; Kanzow & Wiegand, 2020; Durmus et al., 2021). These studies were chosen as they were the most up-to-date and had raw data available for cFPM specifically. In circumstances when evidence didn't exist, or it was not able to extrapolate data for a cFPM, then expert opinion was obtained using clinical members of the 'expert panel'. The probabilities obtained were averaged by the number of completed responses (n=4) and re-confirmed with the panel prior to use.

Transition probabilities were fixed with regards to time, as insufficient evidence existed specifically in relation to molar teeth, for all probabilities which were time dependent. Rates were converted to transition probabilities in Treeage Pro Healthcare 2022© (TreeAge Pro Healthcare, 2022). The probabilities applicable to the model are shown in **Table 6.3** and **6.4.** These are split into those that are caries- and MIH-specific. Once a carious and hypomineralised cFPM required endodontic management, it was assumed the initial disease was disregarded in terms of variations in transition probabilities.

Probabilities	Reference	Probability from study
Caries Specific		
Composite on affected molar	Burke & Lucarotti (2018c)	56% fail at 10 years
Composite Repair	Kanzow and Wiegand (2020)	5.25% repaired, after 5 years
Composite Replace	Kanzow and Wiegand (2020)	2.44% replaced, after 5 years.
Glass Ionomer Cement	Burke & Lucarotti (2018b)	19% fail @ 1 year
MIH Specific		
Composite on affected molar	Gatón-Hernandéz et al. (2020)	3.2% fail at 2 years
Composite Repair	*Expert Opinion*	45% replaced at 5 years.
Composite Replace	Kotsanos et al., (2005)	25.4% needs re-treated after 4 years
Glass Ionomer Cement	Durmus et al., (2021)	11.5% fail at 2 years
Applicable to Caries & MIH		
Spontaneous alignment (extraction at age 6)	Eichenberger *et al.,* (2015)	34%
Spontaneous alignment (extraction at age 9)	Eichenberger *et al.,* (2015)	50%
Spontaneous alignment (extraction at age 12)	Eichenberger *et al.,* (2015)	42.4%
RCT	Ricucci et al.,(2011)	19.6% failed at 5 years
Non-Surgical Re-RCT	(He et al., 2017)	9.6 failed at 2 years
Resin Retained Bridge	*Expert Opinion*	26% failed at 5 years
Orthodontic Closure (extraction at age 12)	*Expert Opinion*	80% complete closure after 2.5 years
Orthodontic Closure (extraction at age 15)	*Expert Opinion*	80% complete closure after 2.5 years
Orthodontic Closure (extraction at age 18)	*Expert Opinion*	70% complete closure after 2.5 years

Table 6.3 Probabilities used in model based on disease profiles

Transition probabilities	Reference	Probability
Chance of failed fillings that are subsequently extracted	*Expert Opinion*	65%
Chance of choosing an extraction after active monitoring (compared to a filling)	*Expert Opinion*	40%
Chance of failed root canal that have a re-root canal	*Expert Opinion*	15%
Chance of having orthodontic treatment to close the gap	*Expert Opinion*	60%

Table 6.4 Transition probabilities applicable to the model

Costs

Each health state was assigned a value that reflected the cost of being in that state in each cycle. Costs included in the model were gathered from routinely collected data. Costs are expressed in 2019/20 UK sterling (GBP, £). The year 2019/20 was chosen to be as reflective of costs prior to the impact of COVID-19 (Wilson, 2022).

Costs used in Cost-benefit analysis

For the cost-benefit framework analysis, the costs from an NHS perspective included those than fall on the NHS only. An overview of the cost data is available in **Table 6.5**.

As previously described in Section 1.7.1, UK dentists are reimbursed by the NHS for providing treatments to NHS patients. Given this model is being undertaken to reflect circumstances pre-COVID 19, existing NHS payments to dental providers for one, three and 12 UDA based on an average UDA value of £25 (British Dental Association, 2020) were used. Therefore, Band 1 (1 UDA) was £25, Band 2 (3 UDA) was £75, and Band 3 (12 UDA) was £300. NHS patient charges for Band 1, 2 and 3 respectively are £23.80, £65.20, and £282.20. However, patient charges are only applied after the age of 18, or 19 if the child remains in full-time education. In this model, it has been assumed that the children are in full time-education. Therefore, costs attached for each health state, before age 19, take the full UDA value (maximum 24 cycles). Thereafter, the total costs that fall on the NHS, and attached to each health state, are the UDA value less the patient charge.

Similar to the UDA, orthodontists are reimbursed for orthodontic care in terms of units of orthodontic activity (UOA). These vary depending on the age of the patient receiving orthodontic treatment. It is assumed in this model that all patient having orthodontic closure will be over the age of 12, which attracts 21 UOAs per course of treatment (NHS Business Services Authority, 2019). The average value for one UOA was calculated as £56.89, based on the average price of available UOA rates from contracts awarded in 2019 (NHS England - Contract Award Notice, 2019). No patient charges exist for orthodontic treatment provided by the NHS, irrespective of age; however, in this model, it was assumed that patients over 19-years-old will not receive orthodontic treatment as this is not routinely provided for adults (NHS, 2020).

Procedure	Patient charge (£) (<19 years old)	Number of UDAs/UOAs attributed	NHS UDA/UOA value (£)	Patient charge (£) (>19 years old)	Total costs fall on NHS (£) (> 19 years old)
Check-Up	£0	1	£25	£23.80	£1.20
Composite	£0	3	£75	£65.20	£9.80
Composite repair	£0	3	£75	£65.20	£9.80
Composite replacement	£0	3	£75	£65.20	£9.80
Temporary Filling (Glass ionomer cement)	£0	3	£75	n/a	n/a
Extraction	£0	3	£75	£65.20	£9.80
Root canal Treatment	£0	3	£75	£65.20	£9.80
Non-surgical re-root canal treatment	£0	3	£75	£65.20	£9.80
Resin retained bridge	£0	12	£300	£282.20	£17.80
Orthodontic closure	£0	21	£1194.69	n/a	n/a

Table 6.5 Costs used in the cost-benefit analysis framework

Costs used in cost-effectiveness analysis

For the cost-effectiveness analysis, an NHS dental practice perspective was adopted. The costs included staff and treatment costs, as these fall on the practice and not the NHS. Costs that fall directly on the patient and society were not included. Using data from the 'Unit Costs of Health and Social Care 2021' report (Jones & Burns, 2021), the average hour unit cost for a dental associate is £105, and a principal £136. This equates to an average of £120.50/hour.

This average unit cost of an hour of NHS dentistry is calculated by combining the average taxable income (average gross earnings less average total expenses) of self-employed primary care associates and principals, based on the average amount of NHS care provided[5]. The average gross earnings include the UDA payments. The average total expenses included direct staff, office and general business, premises, capital, equipment and miscellaneous (including resource costs such as dental materials, laboratory bills etc.) costs.

A primary care specialist orthodontist average taxable income of £85,100 (Talent.com, 2022) was obtained. Unlike general dentists, expenses data were not available, but it was assumed these would be like those for an NHS general dentist (e.g., direct staff costs, capital costs etc.) and included in this figure above. Using similar rates of provision of NHS care used in the dentist's calculation[5], the total hourly rate for an orthodontist was £120.14. This cost was only attributed to orthodontic closure.

The required number of visits, and time taken in minutes to complete each treatment was requested from the expert panel. These were averaged by the number of completed responses (n=4) and shown in **Table 6.6**. These were assumed to be the same for carious and hypomineralised cFPM. The cost per procedure was calculated by multiplying the total time for each procedure (as a fraction of an hour by dividing the average time/60) by the combined hourly rates of dentist and specialist orthodontist. The total costs for each treatment used in the cost-effectiveness analysis are shown in **Table 6.7**. These are assumed to be the same for caries and MIH.

[5] This is based on a dental working hours survey, which identified the average total number of weekly NHS hours worked was 25.9. On average, dentists took 5 days of sickness leave and 4.5 weeks annual leave. Unit costs are based on 1,535 hours (Jones & Burns, 2021; NHS Digital, 2020b).

Procedure	Average number of visits	Average total time per visit (mins)	Average total time per procedure (mins)
Check-up	1	10	10
Composite	1	24	24
Composite repair	1	25	25
Composite replacement	1	29	29
Temporary Filling (Glass ionomer cement)	1	15	15
Extraction	1	33	33
Root canal Treatment	2	80	160
Non-surgical re-root canal treatment	2	80	160
Resin retained bridge	2	50	100
Orthodontic closure	18	14	252

Table 6.6 Time taken per procedure

Procedure	Total costs per procedure (£)
Check-Up	£20.08
Composite	£48.20
Composite repair	£50.20
Composite replacement	£58.24
Temporary Filling (Glass ionomer cement)	£30.13
Extraction	£66.28
Root canal Treatment	£321.33
Non-surgical re-root canal treatment	£321.33
Resin retained bridge	£200.83
Orthodontic closure	£504.59

Table 6.7 Costs used in cost-effectiveness model

Benefits used in cost-benefit analysis

For the cost-benefit analysis, WTP values derived from the DCE in Chapter 5 (shown in **Table 6.8**), were attached to each health state. Active monitoring is congruent with the no treatment option in the DCE, and therefore takes a WTP value of £0. A WTP for a temporary glass ionomer filling was not obtained from the DCE (as it was not designed to capture this), nor available from the literature. Therefore, the value for temporary filling was taken as the mid-point between WTP values for a filling and active monitoring, as it was assumed the true value would lie between no treatment and a filing.

Health State	WTP Values
Active Monitoring	£0
Temporary Filling (Glass ionomer cement)	£332.25
Filling	£646.51
Space – full tooth gap	-£1055.60
No space – spontaneous closure	£620.14
Orthodontic Closure	£656.20
Resin Retained Bridge	-£603.73

Table 6.8 WTP values, derived from DCE in Chapter 4, used in the model

However, in the base-case scenario, decrement in terms of WTP for adverse events were not included. In the model, a composite repair and replacement, root-canal treatment and extraction were deemed to be adverse events, if they occurred after the initial decision point. As a result, for a scenario analysis, WTP values for adverse events were included. However, WTP for adverse events were not obtained in the DCE, as it was not designed to elicit these values. Rather than using arbitrary figures, WTP values were obtained from a DCE which estimated UK public respondents' WTP to avoid specific oral health problems in children (Lord et al., 2015). In this study, the WTP to avoid a permanent tooth for a child that has decay, but no pain, was £114.62. Similarly, WTP values were reported as £304.70, to avoid a permanent tooth having decay and pain, and £244.21, to avoid having a tooth removed (Lord et al., 2015).

As these WTPs were for avoiding adverse events, these were given a negative value for the purposes of the model as it was assumed these adverse events were required. Therefore, in the model, composite repair/replacement was given a WTP of -£114.62, root canal treatment -£209.66 (the average between the WTP for decay with no pain and decay with pain) and extraction -£244.21. The WTP value used for root canal treatment was an average of the two values, for a decayed permanent tooth with pain and no pain, as a root canal treatment does not always have to occur with pain. Despite not being specific for cFPM, it was accepted these available values were likely to be the most similar to the true valuation for just a molar.

Benefits used in cost-effectiveness analysis

For the cost-effectiveness analysis, the number of appointments required for each option was attached to a health state and/or event. These were derived from the expert panel during the micro costing exercise. To calculate the number of total avoided appointments, for each alternative option, the average number of appointments per patient in the model for each option was obtained, and then subtracted from one another. Given the model is addressing outcomes for original decisions made in children, using this as the effectiveness measure was felt to be the most pertinent for the decision problem of which choice to make. Given the additional impact on health and wellbeing of having to undergo care, it is assumed in this model, that health/wellbeing impact is inversely proportional to the number of appointments, for example, less future appointments are better.

Procedure	Average number of visits
Check-up	1
Composite	1
Composite repair	1
Composite replacement	1
Temporary Filling (Glass ionomer cement)	1
Extraction	1
Root canal Treatment	2
Non-surgical re-root canal treatment	2
Resin retained bridge	2
Orthodontic closure	18

Table 6.9 Number of appointments required for each option used as effectiveness measure in the model.

6.4.9 Model outcomes

The outcome considered in the model for the cost-benefit framework analysis is the incremental NMB. For the cost-effectiveness framework, the number of future appointments avoided, for each alternative option to manage cFPM, were used. The higher number of future visits was deemed to be a reduction in effectiveness.

6.4.10 Model validation

Model validity includes face, internal and external (Eddy et al., 2012; NICE, 2022a). For face validity, the members of the expert panel were asked to evaluate whether the model reflected reality, in terms of treatment options and clinical pathways. Amendments were made based on their feedback.

The focus in this thesis has been on the internal validity of the model. Steps were taken to check the model and included adjusting values used in model development and from the literature, to see if the observed change followed the predicted result e.g., changing all probabilities to 1 to see if all patients transition straight to the intended state. Additionally, patient flow calculations were checked for each arm of the model to ensure the model was working as expected. Similarly, uncertainty analyses also acted as an internal validity check, for example, changing the discount rate for costs and benefits at 0%, and observing the expected changes such as an increase in the total costs, or checking for consistency between the deterministic and probabilistic base case results. Further details of this analysis are reported in section 6.6.7 and discussed in section 6.7.1.

A further component of model validation is to compare model results with those obtained from other models, and with real world data and the expert panel. This external validation (Eddy et al., 2012; NICE, 2022a) is discussed in section 6.7.1.

6.5 Analysis

Cost-benefit analyses were reported as the net benefit of intervention and incremental net-benefit. Whereas the cost-effectiveness analysis was reported as the incremental cost per future appointment avoided for initial strategies. Details on how these are calculated are discussed in section 6.2.4 of this chapter. The base-case scenario was a child with a cFPM caused by dental caries, as part of a cost-benefit framework analysis. Ten thousand trials were simulated through the model using Treeage Pro Healthcare 2022© (TreeAge Pro Healthcare, 2022) through the individual patient-level microsimulation model. Consideration of discounting rates, half-cycle correction and sensitivity analyses are now described in the rest of this section.

6.5.1 Discounting

The costs and benefits (WTP and number of future appointments avoided) after the first year were discounted at the recommended rate of 3.5% (NICE, 2022a).

6.5.2 Half-cycle correction

There is uncertainty as to when the transition happens within each cycle of the model. Therefore, a half cycle correction was employed in the first and final cycles of the model (Caro et al., 2012).

6.5.3 Handling uncertainties and heterogeneity

Uncertainties and heterogeneity of the model parameters were assessed and included in sensitivity analyses.

Sensitivity Analysis

Deterministic sensitivity analyses were undertaken based on parameters and scenarios that may influence the interpretation of the results. Similar to the base-case scenario, each scenario analysis modelled 10,000 trials through the model using Treeage Pro Healthcare 2022© (TreeAge Pro Healthcare, 2022). The scenario analyses undertaken were:

- Scenario 1 – a child with a cFPM caused by dental caries, as part of a cost-effectiveness analysis, and included different costs
- Scenario 2 - a child with a cFPM caused by MIH, as part of a cost-effectiveness analysis
- Scenario 3 – change the discount rate from 3.5% to 0%
- Scenario 4 – altered the WTP value of temporary filling, from £332.25 to £161.63 (25% of the WTP value of a filling)
- Scenario 5 – altered the WTP value of temporary filling, from £332.25 to £493.88 (75% of the WTP value of a filling)
- Scenario 6 – Include WTP decrements, due to adverse events, into the model
- Scenario 7 – change the time horizon from lifetime to 18 years old
- Scenario 8 – using the WTP values obtained from the mixed-logit analysis

PSA over a lifetime was done using Monte-carlo simulation in Treeage Pro Healthcare 2022© (TreeAge Pro Healthcare, 2022), with 1000 2^{nd} order parameter values obtained randomly from the distributions fitted across all input parameters of the model. For each of the 1000 PSA, 10,000 simulations were run, with each simulation picking up random values from each distribution and generating the cost-effectiveness results. Transition probabilities derived as proportions were assigned the beta distributions. Costs were assigned gamma distributions. WTP values were assumed a normal (gaussian) distribution. These are summarised in **Table 6.10, 6.11** and **6.12**. The cost-effectiveness plane and the CEAC were used to report the results of the PSA.

Parameter	Parameter Value (distribution)	Distribution Type
WTP Values		
Active Monitoring	£0	n/a (fixed)
Temporary Filling	£332.25 (SD £32.45)	Normal
Filling	£646.51 (SD £64.91)	Normal
Space – full tooth gap	-£1055.60 (SD £74.45)	Normal
No space – spontaneous closure	£620.14 (SD £63.20)	Normal
Orthodontic Closure	£656.20 (SD £71.64)	Normal
Resin Retained Bridge	-£603.73 (SD £72.83)	Normal
Composite Repair – Adverse Event	-£114.62 (SD £22.84)	Normal
Root Canal Treatment – Adverse Event	-£209.66 (SD £41.93)	Normal
Extraction – Adverse Event	-£244.21 (SD £48.84)	Normal
ASC	£4385.65 (SD £233.27)	Normal

Table 6.10 WTP values, and their distributions

Parameter	Parameter Value (distribution)	Distribution Type
Costs for Cost-effectiveness analysis		
Check-Up	£20.08 (α = 25; λ = 1.245)	Gamma
Composite	£48.20 (α = 25; λ = 0.518)	Gamma
Composite repair	£50.20 (α = 25; λ = 0.498)	Gamma
Composite replacement	£58.24 (α = 25; λ = 0.429)	Gamma
Temporary Filling (Glass Ionomer)	£30.13 (α = 25; λ = 0.829)	Gamma
Extraction	£66.28 (α = 25; λ = 0.377)	Gamma
Root canal Treatment	£321.33 (α = 25; λ = 0.077)	Gamma
Re-root canal treatment	£321.33 (α = 25; λ = 0.077)	Gamma
Resin retained bridge	£200.83 (α = 25; λ = 0.124)	Gamma
Orthodontic closure	£504.59 (α = 25; λ = 0.049)	Gamma
Costs for Cost-benefit analysis		
Check-up (<19-years-old)	£25 (α = 625; λ = 25)	Gamma
Check-up (>19-years-old)	£1.20 (α = 1.44; λ = 1.2)	Gamma
Composite (<19-years-old)	£75 (α = 5625; λ = 75)	Gamma
Composite (>19-years-old)	£9.80 (α = 96.04; λ = 9.8)	Gamma
Composite Repair (<19-years-old)	£75 (α = 5625; λ = 75)	Gamma
Composite Repair (>19-years-old)	£9.80 (α = 96.04; λ = 9.8)	Gamma
Composite Replace (<19-years-old)	£75 (α = 5625; λ = 75)	Gamma
Composite Replace (>19-years-old)	£9.80 (α = 96.04; λ = 9.8)	Gamma
Temporary Filling (Glass ionomer cement) (<19-years old)	£75 (α = 5625; λ = 75)	Gamma
Extraction (<19-years-old)	£75 (α = 5625; λ = 75)	Gamma
Extraction (>19-years-old)	£9.80 (α = 96.04; λ = 9.8)	Gamma
Root Canal Treatment (<19-years-old)	£75 (α = 5625; λ = 75)	Gamma
Root Canal Treatment (>19-years-old)	£9.80 (α = 96.04; λ = 9.8)	Gamma
Re-Root Canal Treatment (<19-years-old)	£75 (α = 5625; λ = 75)	Gamma
Re- Root Canal Treatment (>19-years-old)	£9.80 (α = 96.04; λ = 9.8)	Gamma
Resin Retained Bridge (<19-years-old)	£300 (α = 90,000; λ = 300)	Gamma
Resin Retained Bridge (>19-years-old)	£17.80 (α = 316.84; λ = 17.8)	Gamma
Orthodontic Closure (<19 years old)	£1194.69 (α = 1427284.19; λ = 1194.69)	Gamma

Table 6.11 Costs, and their distributions

Parameter	Parameter Value (distribution)	Distribution Type
Failure of Composite (Caries)	0.040217887274674236 (α = 103265; β= 81137)	Beta
Composite Repair (Caries)	0.0053835076833236295 (α = 101; β= 1821)	Beta
Composite Replace (Caries)	0.0024727029337261452 (α = 47; β= 1875)	Beta
Failure of Glass Ionomer (Caries)	0.09999973064847723 (α = 58783; β= 250602)	Beta
Failure of Composite (MIH)	0.007758537903402418 (α = 10; β= 316)	Beta
Composite Repair (MIH)	0.05803174078617379 (α = 13.30; β= 16.25)	Beta
Composite Replace (MIH)	0.0024727029337261452 (α = 15; β= 44)	Beta
Failure of Glass Ionomer (MIH)	0.03334775068467233 (α = 17; β= 117)	Beta
Spontaneous alignment (extraction at age 6)	0.34042553191489366 (α = 16; β= 31)	Beta
Spontaneous alignment (extraction at age 9)	0.5 (α = 77; β= 77)	Beta
Spontaneous alignment (extraction at age 12)	0.42405063291139244 (α = 67; β= 91)	Beta
Failure of Root Canal Treatment	0.021197565697956056 (α = 27; β= 113)	Beta
Failure of Re-Root Canal Treatment	0.024957314768366934 (α = 5; β= 47)	Beta
Failure of Resin Retained Bridge	0.02966170376165511 (α = 18.24; β= 51.91)	Beta
Orthodontic Closure (extraction at age 12)	0.02966170376165511 (α = 4.2; β= 1.05)	Beta
Orthodontic Closure (extraction at age 15)	0.02966170376165511 (α = 4.2; β= 1.05)	Beta
Orthodontic Closure (extraction at age 18)	0.02966170376165511 (α = 6.80; β= 2.91)	Beta
Chance of failed fillings that are extracted (compared to having a root canal)	0.65 (α = 8.1; β= 4.36)	Beta
Chance of choosing an extraction after active monitoring (compared to a filling)	0.4 (α = 14.60; β= 21.9)	Beta
Chance of failed root canal that has a re-root canal (compared to having an extraction)	0.15 (α = 21.1; β= 119.56)	Beta
Chance of having orthodontic treatment to close the gap (compared to prosthetic replacement)	0.6 (α = 9.4; β= 6.27)	Beta

Table 6.12 Probabilities (adjusted for six monthly cycles based on source detailed in Table 6.2)

Sections 6.4 and 6.5 have described the methods and analytical plan for the model. The following section will describe the results, initially for the base-case scenario. This will be followed by scenario analyses, run as part of a DSA, and PSA.

6.6 Results

Despite the model running, it is plausible that some minor errors may still exist within the structure. Error checking is an iterative process (see section 6.4.10 for further details), and it is not uncommon for models to undergo several edits prior to running as expected. This is often the case for NICE appraisals. As a result, further error checks may be required to ensure the model is operationalising as expected. Therefore, the results presented in this chapter are based on the model as it stands.

The model was built in Treeage Pro Healthcare 2022© (TreeAge Pro Healthcare, 2022). A diagrammatic representation of the model is shown in **Figure 6.4**.

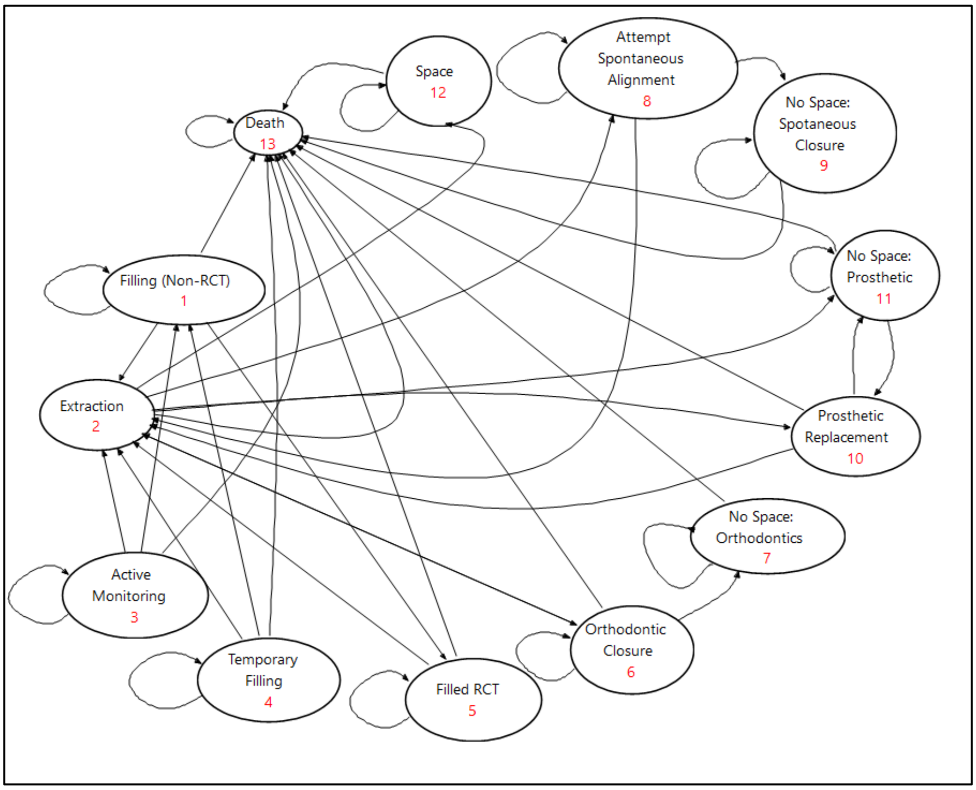

Figure 6.4 Diagrammatic representation of the individual patient-level microsimulation model

6.6.1 Base Case Scenario

The base-case scenario involved a child with a cFPM, caused by dental caries, as part of a cost-benefit analysis. The results from the base-case scenario are shown in **Tables 6.13, 6.14, 6.15** and **6.16** which summarise the costs, WTP and overall incremental analyses. In **Table 6.15** all initial strategies are presented in terms of ascending order of cost. Strategies which were more costly and less beneficial were strictly dominated. These were excluded from further incremental analysis as shown in **Table 6.16**. The cost-effectiveness plane for initial treatment strategies for cFPM, based on base-case scenario, are shown in **Figure 6.5.**

Statistic	Cost (Filling (Non-RCT))	Cost (Temp Filling)	Cost (Active monitoring)	Cost (Extraction)
Mean	648.24	1156.03	759.91	1125.55
Std Deviation	103.04	697.59	485.50	837.33
Minimum	410.96	214.28	310.72	210.38
2.5%	494.49	530.76	447.60	481.10
10%	536.12	566.55	491.78	515.10
Median	632.75	659.56	588.91	601.14
90%	778.58	2213.92	1605.35	2647.56
97.5%	896.26	2556.54	2405.31	3014.54
Maximum	2211.68	3025.95	3287.22	4155.34
95%% Lower Bound	646.22	1142.35	750.39	1109.14
95%% Upper Bound	650.26	1169.70	769.42	1141.96

Table 6.13 Base-case scenario results: costs associated with initial treatment strategies for cFPM

Statistic	WTP (Filling (Non-RCT))	WTP (Temp Filling)	WTP (Active monitoring)	WTP (Extraction)
Mean	3995.84	6461.65	333.71	7460.62
Std Deviation	2081.07	14656.82	8642.50	15636.41
Minimum	-16707.64	-23236.93	-29147.22	-27305.53
2.5%	-2023.76	-19216.82	-22650.46	-21115.11
10%	1350.73	-16720.05	-11297.73	-18916.97
Median	4849.07	16278.18	160.11	17298.55
90%	5279.27	19117.19	13165.50	20054.90
97.5%	5458.43	20374.79	14894.24	21218.28
Maximum	5899.79	22913.97	17777.99	24027.53
95%% Lower Bound	3955.06	6174.38	164.33	7154.15
95%% Upper Bound	4036.63	6748.92	503.10	7767.09

Table 6.14 Base-case scenario results: WTP associated with initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Effects	Incr. Eff	NMB	Incr. NMB
Filling (Non-RCT)	648.24	--	3995.84	--	3347.6	--
Active monitoring*	759.91	111.67	333.71	-3662.13	-426.2	-3773.8
Extraction	1125.55	365.64	7460.62	7126.91	6335.07	6761.27
Temp Filling*	1156.03	30.48	6461.65	-998.97	5305.62	-1029.45
n.b * - dominated strategy						

Table 6.15 Base-case scenario results: incremental analysis for all initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Eff.	Incr. Eff	NMB	Incr. NMB	Incr. B/C ratio
Filling (Non-RCT)	648.24	--	3995.84	--	3347.6	--	--
Extraction	1125.55	477.31	7460.62	3464.78	6335.07	2987.47	7.2590

Table 6.16 Base-case scenario results: incremental analysis, after excluding dominated strategies, associated with initial treatment strategies for cFPM

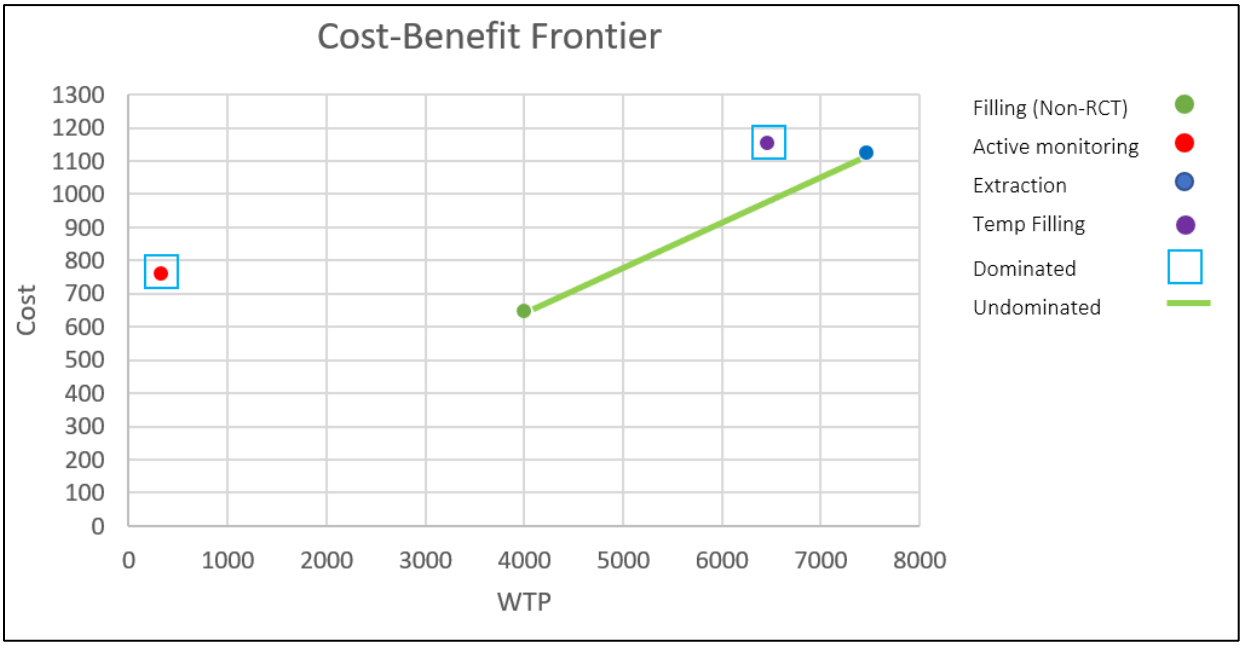

Figure 6.5 Base-case scenario results: Cost-effectiveness plane for initial treatment strategies for cFPM

Initial management strategies of temporary filling and active monitoring are dominated. Thus, these options should not be considered as the costs outweigh the benefits. The initial strategy of extraction has a higher net monetary benefit than filling.

6.6.2 Deterministic Sensitivity Analysis - Scenario Analysis 1

Scenario analysis 1 addressed a child with a cFPM caused by dental caries, as part of a cost-effectiveness analysis, including the micro costing from an NHS dental practice perspective. The results from scenario analysis 1 are shown in **Table 6.17, 6.18 and 6.19** which summarise the costs, effectiveness measure (number of future visits associated following each initial treatment strategy) and overall incremental analyses. In **Table 6.19** all initial strategies are presented in terms of ascending order of cost. The cost-effectiveness plane for initial treatment strategies for cFPM, based on scenario analysis 1, are shown in **Figure 6.6.**

Statistic	Cost (Filling (Non-RCT))	Cost (Temp Filling)	Cost (Active monitoring)	Cost (Extraction)
Mean	1388.41	1968.92	1482.64	1913.04
Std Deviation	576.38	1639.54	1085.60	1693.04
Minimum	41.32	31.06	19.73	57.22
2.5%	778.83	746.39	710.14	723.87
10%	920.92	891.71	854.60	869.40
Median	1235.04	1256.71	1169.31	1210.98
90%	2080.75	5047.63	2156.96	5215.53
97.5%	3065.38	6235.60	5453.94	6501.74
Maximum	6068.20	9155.53	9114.24	10659.66
95%% Lower Bound	1377.11	1936.79	1461.36	1879.85
95%% Upper Bound	1399.70	2001.06	1503.92	1946.22

Table 6.17 Scenario analysis 1 results: costs associated with initial treatment strategies for cFPM

Statistic	No. of Future Visits Avoided (Filling (Non-RCT))	No. of Future Visits Avoided (Temp Filling)	No. of Future Visits Avoided (Active monitoring)	No. of Future Visits Avoided (Extraction)
Mean	25.49	29.54	26.93	29.68
Std Deviation	1.67	6.63	4.42	6.76
Minimum	0.00	0.00	0.00	1.00
2.5%	21.52	22.75	21.99	23.25
10%	25.05	25.60	25.53	26.10
Median	25.60	26.00	26.06	26.10
90%	26.33	40.62	28.34	42.19
97.5%	27.09	41.92	40.88	43.46
Maximum	40.12	49.79	46.33	49.06
95%% Lower Bound	25.46	29.41	26.84	29.55
95%% Upper Bound	25.52	29.67	27.02	29.81

Table 6.18 Scenario analysis 1 results: effectiveness associated with initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Eff.	Incr. Eff	ICER
Filling (Non-RCT)	1388.41	--	-25.49	--	--
Active Monitoring	1482.64	Dominated	-29.54	Dominated	Dominated
Extraction	1913.04	Dominated	-29.68	Dominated	Dominated
Temporary Filling	1968.92	Dominated	-26.93	Dominated	Dominated

Table 6.19 Scenario analysis 1 results: incremental analysis associated with initial treatment strategies for cFPM

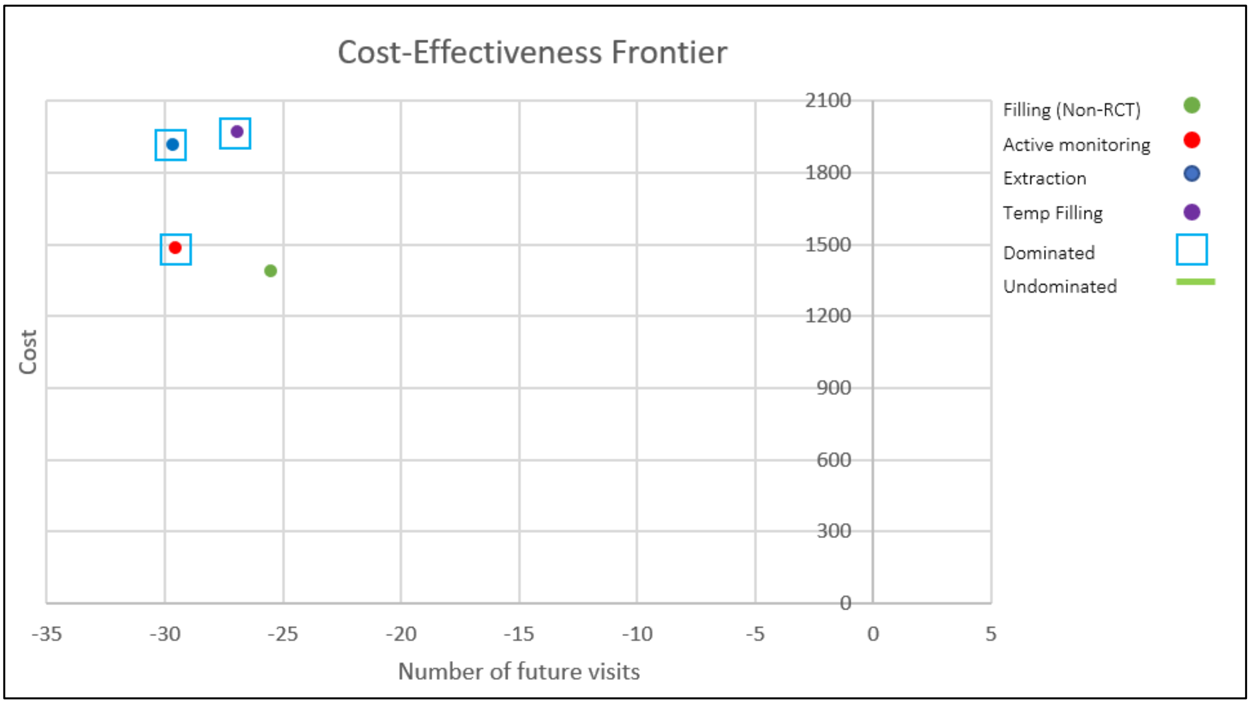

Figure 6.6 Scenario analysis 1 results: Cost-effectiveness plane for initial treatment strategies for cFPM

The higher the number of future visits is a worse outcome. Therefore, in this scenario analysis, the filling strategy dominated all other initial management strategies of extraction, temporary filling, and active monitoring. Thus, these options should not be considered as compared to filling, as they are all more costly and less effective.

6.6.3 Deterministic Sensitivity Analysis - Scenario Analysis 2

Scenario analysis 2 involved a child with a cFPM caused by MIH, as part of a cost-effectiveness analysis, using micro-costing. The results from the scenario analysis 2are shown in **Tables 6.20, 6.21 and 6.22** which summarise the costs, effectiveness measure (number of future visits associated following each initial treatment strategy) and overall incremental analyses. In **Table 6.22** all initial strategies are presented in terms of ascending order of cost. The cost-effectiveness plane for initial treatment strategies for cFPM, based on scenario analysis 2, are shown in **Figure 6.7.**

Statistic	Cost (Filling (Non-RCT))	Cost (Temp Filling)	Cost (Active monitoring)	Cost (Extraction)
Mean	1070.25	1959.17	1339.46	1908.97
Std Deviation	216.21	1634.52	1040.18	1685.73
Minimum	94.25	48.90	32.37	171.06
2.5%	683.72	748.74	677.91	728.66
10%	810.08	887.01	813.54	865.12
Median	1058.49	1253.87	1082.91	1207.58
90%	1352.41	5033.16	1564.14	5180.28
97.5%	1531.56	6213.22	5333.26	6513.62
Maximum	2510.25	8951.37	9125.52	8829.45
95%% Lower Bound	1066.02	1927.13	1319.07	1875.93
95%% Upper Bound	1074.49	1991.21	1359.84	1942.01

Table 6.20 Scenario analysis 2 results: costs associated with initial treatment strategies for cFPM

Statistic	No. of Future Visits Avoided (Filling (Non-RCT))	No. of Future Visits Avoided (Temp Filling)	No. of Future Visits Avoided (Active monitoring)	No. of Future Visits Avoided (Extraction)
Mean	25.23	29.45	26.76	29.69
Std Deviation	1.54	6.59	4.37	6.71
Minimum	0.50	0.50	0.50	3.37
2.5%	20.97	22.75	21.79	23.37
10%	24.97	25.60	25.40	26.10
Median	25.60	26.00	26.06	26.10
90%	25.60	40.62	28.01	42.19
97.5%	25.60	41.98	40.88	43.36
Maximum	26.40	47.30	46.97	49.12
95%% Lower Bound	25.20	29.32	26.68	29.56
95%% Upper Bound	25.26	29.58	26.85	29.82

Table 6.21 Scenario analysis 2 results: effectiveness associated with initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Eff.	Incr. Eff	ICER
Filling (Non-RCT)	1070.25	--	-25.23	--	--
Active Monitoring	1339.46	Dominated	-29.68	Dominated	Dominated
Extraction	1908.97	Dominated	-29.54	Dominated	Dominated
Temporary Filling	1968.92	Dominated	-26.93	Dominated	Dominated

Table 6.22 Scenario analysis 2 results: incremental analysis associated with initial treatment strategies for cFPM

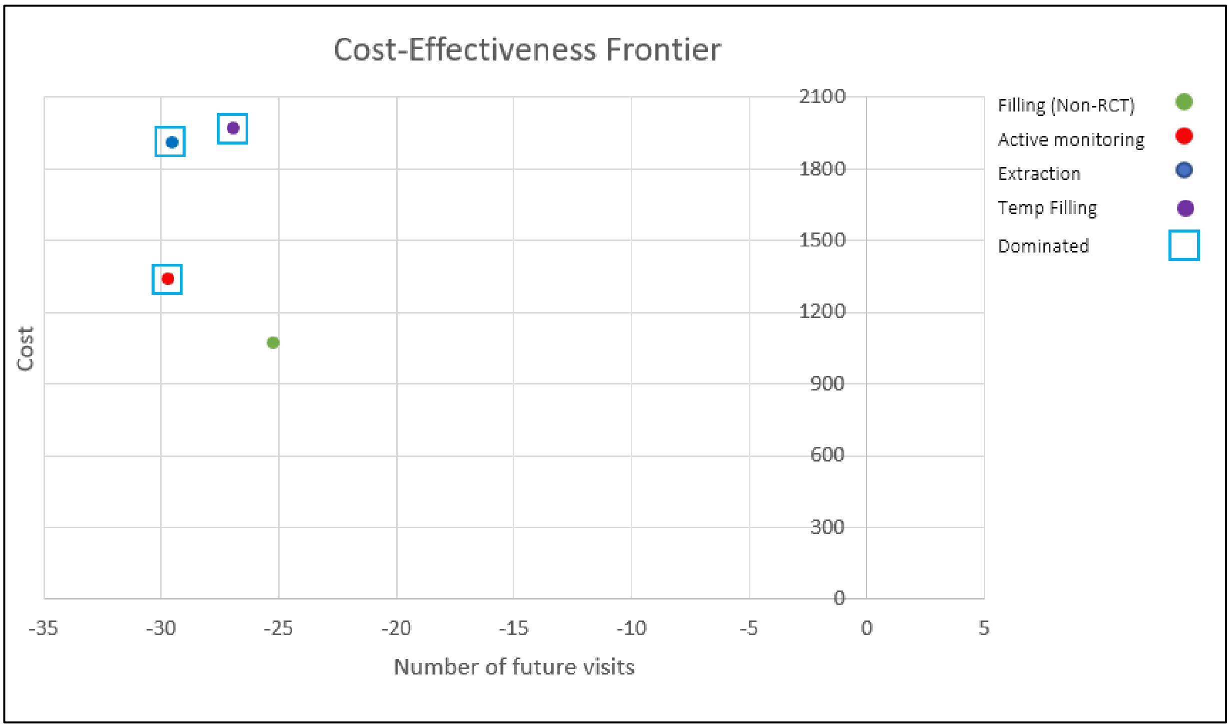

Figure 6.7 Scenario analysis 2 results: Cost-effectiveness plane for initial treatment strategies for cFPM

Similar to scenario analysis 1, the higher the number of future visits is a worse outcome. The filling strategy dominated all other initial management strategies of extraction, temporary filling, and active monitoring. Thus, these options should not be considered as compared to filling, as they are all more costly and less effective.

6.6.4 Deterministic Sensitivity Analysis - Scenario Analysis 3

Scenario analysis 3 involved the discount rate being changed from 3.5% to 0%. The results from scenario analysis 3 are shown in **Table 6.23, 6.24, 6.25** and **6.26** which summarise the costs, WTP and overall incremental analyses. In **Table 6.25** all initial strategies are presented in terms of ascending order of cost. Strategies which were more costly and less beneficial where strictly dominated. These were excluded from further incremental analysis as shown in **Table 6.26**. The cost-effectiveness plane for initial treatment strategies for cFPM, based on scenario analysis 3, are shown in **Figure 6.8.**

Statistic	Cost (Filling (Non-RCT))	Cost (Temp Filling)	Cost (Active monitoring)	Cost (Extraction)
Mean	913.72	1546.05	1036.60	1488.29
Std Deviation	233.53	1026.48	692.41	1184.71
Minimum	126.33	125.59	76.28	412.26
2.5%	618.70	625.04	554.49	575.71
10%	686.13	686.53	619.01	631.77
Median	853.70	884.21	789.68	800.28
90%	1239.94	3333.87	1833.31	3810.93
97.5%	1523.57	3578.69	3502.15	4230.70
Maximum	2919.68	3928.99	4341.21	4914.19
95%% Lower Bound	909.14	1525.93	1023.03	1465.07
95%% Upper Bound	918.30	1566.17	1050.17	1511.51

Table 6.23 Scenario analysis 3 results: costs associated with initial treatment strategies for cFPM

Statistic	WTP (Filling (Non-RCT))	WTP (Temp Filling)	WTP (Active monitoring)	WTP (Extraction)
Mean	-83.64	8266.95	-444.91	12229.09
Std Deviation	8850.68	38456.66	22808.00	38729.70
Minimum	-53957.82	-69397.54	-69730.40	-69532.40
2.5%	-25473.35	-57646.02	-58543.06	-59483.05
10%	-12617.59	-52073.06	-30090.25	-53495.97
Median	4794.95	34783.42	0.00	36732.11
90%	5274.44	42361.07	34280.87	43605.94
97.5%	5458.71	45171.58	38619.09	46475.98
Maximum	39197.56	52990.40	47745.38	53387.11
95%% Lower Bound	-257.11	7513.21	-891.93	11470.00
95%% Upper Bound	89.83	9020.69	2.12	12988.17

Table 6.24 Scenario analysis 3 results: WTP associated with initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Effects	Incr. Eff	NMB	Incr. NMB
Filling (Non-RCT)	913.72	--	-83.64	--	-997.36	--
Active monitoring*	1036.60	122.88	-444.91	-361.27	-1481.51	-484.15
Extraction	1488.29	451.69	12229.09	12674	10740.8	12222.31
Temp Filling*	1546.05	57.76	8266.95	-3962.14	6720.9	-4019.9
n.b * - dominated strategy						

Table 6.25 Scenario analysis 3 results: incremental analysis for all initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Eff.	Incr. Eff	NMB	Incr. NMB	Incr. B/C ratio
Filling (Non-RCT)	913.72	--	3995.84	--	-997.36	--	--
Extraction	1488.29	574.57	7460.62	12312.73	10740.8	11738.16	21.42947

Table 6.26 Scenario analysis 3 results: incremental analysis, after excluding dominated strategies, associated with initial treatment strategies for cFPM

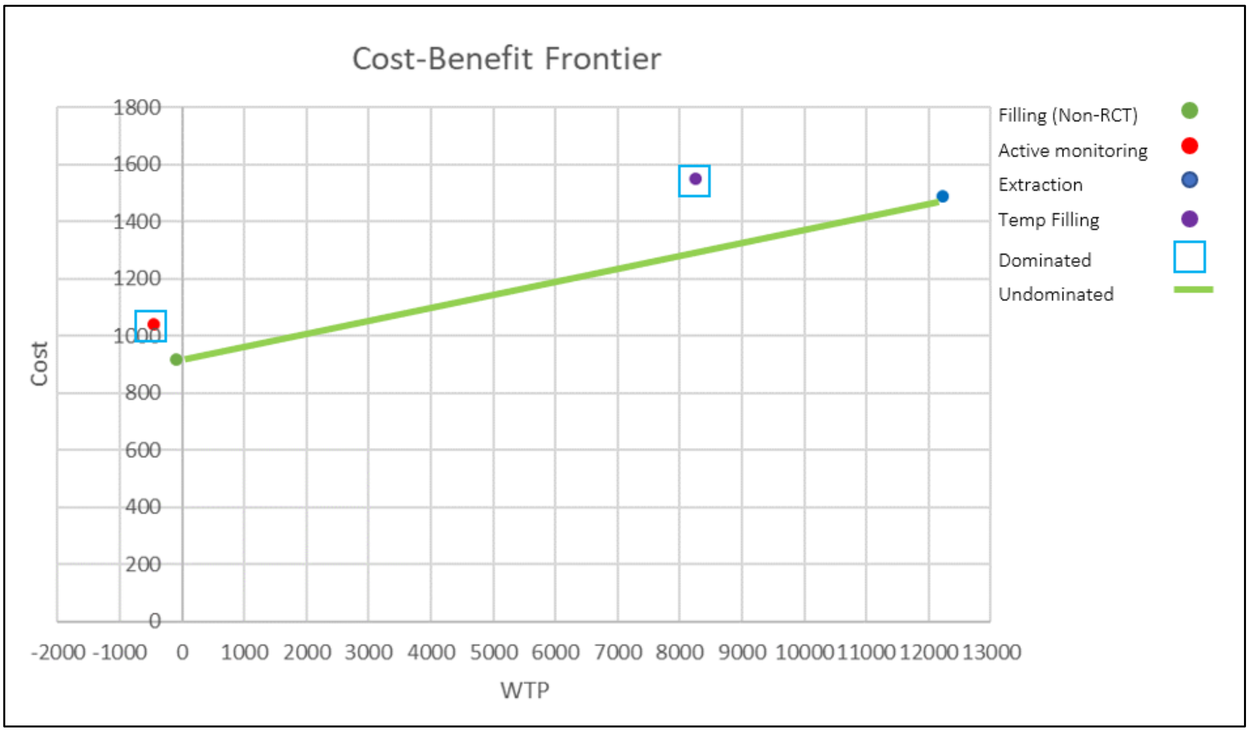

Figure 6.8 Scenario analysis 3 results: Cost-effectiveness plane for initial treatment strategies for cFPM

Initial management strategies of temporary filling and active monitoring are dominated. Thus, these options should not be considered as the costs outweigh the benefits. The initial strategy of extraction is more efficient than filling.

6.6.5 Deterministic Sensitivity Analysis - Scenario Analysis 4

Scenario analysis 4 involved changing the WTP value of temporary filling from £332.25 to £161.63 (25% of the WTP value of a filling). The results from scenario analysis 4 are shown in **Table 6.27, 6.28, 6.29** and **6.30** which summarise the costs, WTP and overall incremental analyses. In **Table 6.27** all initial strategies are presented in terms of ascending order of cost. Strategies which were more costly and less beneficial where strictly dominated. These were excluded from further incremental analysis as shown in **Table 6.30**. The cost-effectiveness plane for initial treatment strategies for cFPM, based on scenario analysis 4, are shown in **Figure 6.9.**

Statistic	Cost (Filling (Non-RCT))	Cost (Temp Filling)	Cost (Active monitoring)	Cost (Extraction)
Mean	648.84	1152.81	757.69	1109.95
Std Deviation	103.27	695.11	483.32	826.30
Minimum	73.55	75.88	26.32	75.15
2.5%	493.63	528.37	446.30	478.73
10%	538.45	565.79	489.97	514.39
Median	633.11	659.11	589.85	600.28
90%	779.68	2216.66	1604.29	2635.85
97.5%	893.44	2555.07	2389.89	2997.20
Maximum	2141.76	2998.13	3164.91	3526.41
95%% Lower Bound	646.81	1139.18	748.22	1093.75
95%% Upper Bound	650.86	1166.43	767.16	1126.14

Table 6.27 Scenario analysis 4 results: costs associated with initial treatment strategies for cFPM

Statistic	WTP (Filling (Non-RCT))	WTP (Temp Filling)	WTP (Active monitoring)	WTP (Extraction)
Mean	3986.41	6308.36	423.16	7688.43
Std Deviation	2045.02	14650.85	8423.61	15560.91
Minimum	-13777.05	-23509.00	-28488.25	-24857.03
2.5%	-2065.92	-19271.04	-22441.87	-21244.18
10%	1409.82	-16977.68	-10705.19	-18854.62
Median	4837.84	16046.46	230.83	17326.17
90%	5271.40	19002.33	13072.40	20029.61
97.5%	5454.30	20166.72	14751.56	21143.64
Maximum	5847.11	22671.66	17997.98	23618.02
95%% Lower Bound	3946.33	6021.21	258.06	7383.44
95%% Upper Bound	4026.49	6595.51	588.26	7993.42

Table 6.28 Scenario analysis 4 results: WTP associated with initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Effects	Incr. Eff	NMB	Incr. NMB
Filling (Non-RCT)	648.84	--	3986.41	--	3337.57	--
Active monitoring*	757.69	108.85	423.16	-3563.25	-334.53	-3672.1
Extraction	1109.95	352.26	7688.43	7265.27	6578.48	6913.01
Temp Filling*	1152.81	42.86	6308.36	-1380.07	5155.55	-1422.93
n.b * - dominated strategy						

Table 6.29 Scenario analysis 4 results: incremental analysis for all initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Eff.	Incr. Eff	NMB	Incr. NMB	Incr. B/C ratio
Filling (Non-RCT)	648.84	--	3986.41	--	3337.57	--	
Extraction	1109.95	461.11	7688.43	3702.02	6578.48	3240.91	8.028496

Table 6.30 Scenario analysis 4 results: incremental analysis, after excluding dominated strategies, associated with initial treatment strategies for cFPM

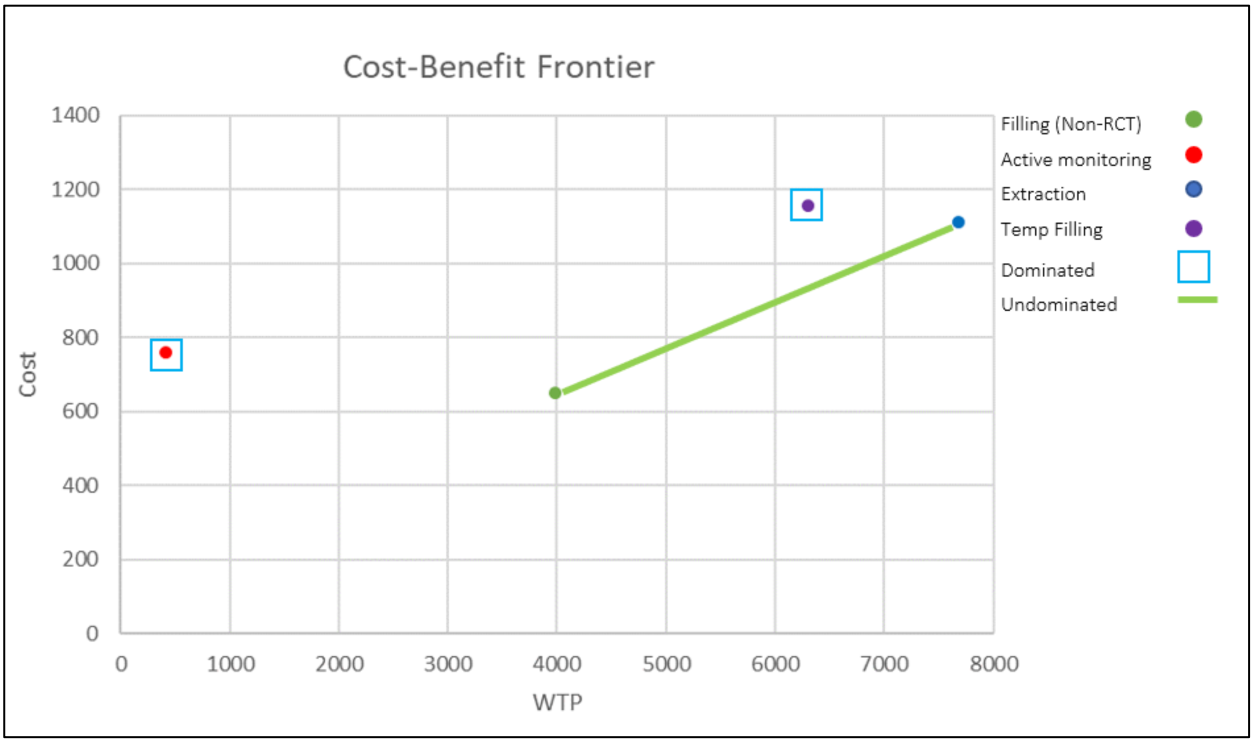

Figure 6.9 Scenario analysis 4 results: Cost-effectiveness plane for initial treatment strategies for cFPM

Despite reducing the WTP value for temporary filling, initial management strategies of temporary filling and active monitoring are dominated. The initial strategy of extraction is more efficient than filling.

6.6.6 Deterministic Sensitivity Analysis - Scenario Analysis 5

Scenario analysis 5 involved changing the WTP value of temporary filling from £332.25 to £493.88 (75% of the WTP value of a filling). Scenario analysis 5 involved changing the WTP value of temporary filling from £332.25 to £161.63 (25% of the WTP value of a filling). The results from scenario analysis 5 are shown in **Table 6.31, 6.32, 6.33** and **6.34** which summarise the costs, WTP and overall incremental analyses. In **Table 6.33** all initial strategies are presented in terms of ascending order of cost. Strategies which were more costly and less beneficial where strictly dominated. These were excluded from further incremental analysis as shown in **Table 6.34**. The cost-effectiveness plane for initial treatment strategies for cFPM, based on scenario analysis 5, are shown in **Figure 6.10.**

Statistic	Cost (Filling (Non-RCT))	Cost (Temp Filling)	Cost (Active monitoring)	Cost (Extraction)
Mean	652.49	1159.92	758.66	1144.48
Std Deviation	105.73	696.45	485.92	851.79
Minimum	285.03	199.58	191.96	245.95
2.5%	495.87	528.42	449.28	480.81
10%	539.76	569.22	493.43	516.58
Median	636.62	661.38	591.37	602.80
90%	784.93	2215.60	1597.99	2714.58
97.5%	896.18	2547.49	2432.97	3005.81
Maximum	650.41	1146.27	749.13	1127.79
95%% Lower Bound	654.56	1173.57	768.18	1161.17
95%% Upper Bound	652.49	1159.92	758.66	1144.48

Table 6.31 Scenario analysis 5 results: costs associated with initial treatment strategies for cFPM

Statistic	WTP (Filling (Non-RCT))	WTP (Temp Filling)	WTP (Active monitoring)	WTP (Extraction)
Mean	3938.87	6481.81	150.53	7380.99
Std Deviation	2142.55	14636.48	8539.85	15560.43
Minimum	-16935.73	-23528.47	-29599.31	-25653.40
2.5%	-2387.64	-19041.82	-22537.36	-21227.04
10%	1220.54	-16599.25	-11283.56	-18852.27
Median	4837.26	16335.83	96.81	17245.51
90%	5274.92	19218.73	13043.74	19995.61
97.5%	5460.31	20408.54	14746.64	21167.87
Maximum	3896.88	6194.94	-16.85	7076.01
95%% Lower Bound	3980.86	6768.68	317.91	7685.96
95%% Upper Bound	3938.87	6481.81	150.53	7380.99

Table 6.32 Scenario analysis 5 results: WTP associated with initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Effects	Incr. Eff	NMB	Incr. NMB
Filling (Non-RCT)	652.49	--	3938.87	--	3286.38	--
Active monitoring*	758.66	106.17	150.53	-3788.34	-608.13	-3894.51
Extraction	1144.48	385.82	7380.99	7230.46	6236.51	6844.64
Temp Filling*	1159.92	15.44	6481.81	-899.18	5321.89	-914.62
n.b * - dominated strategy						

Table 6.33 Scenario analysis 5 results: incremental analysis for all initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Eff.	Incr. Eff	NMB	Incr. NMB	Incr. B/C ratio
Filling (Non-RCT)	652.49	--	3938.87	--	3286.38	--	--
Extraction	1144.48	491.99	7380.99	3442.12	6236.51	2950.13	6.996321

Table 6.34 Scenario analysis 5 results: incremental analysis, after excluding dominated strategies, associated with initial treatment strategies for cFPM

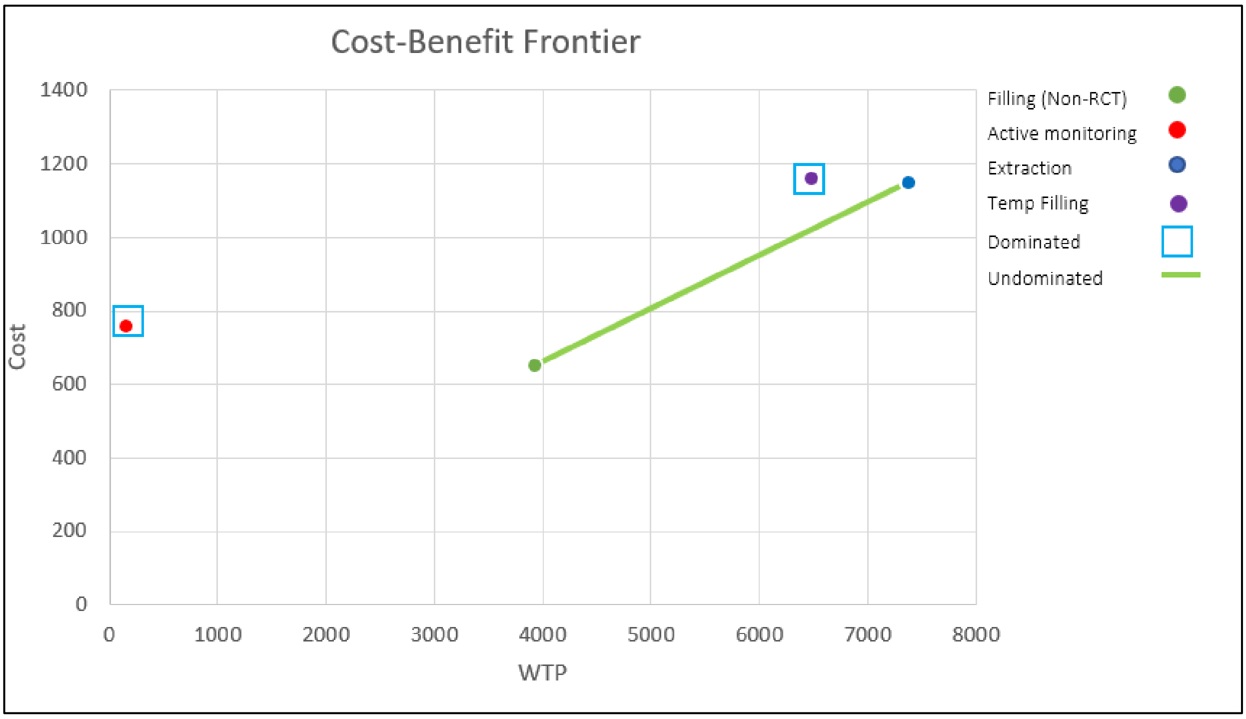

Figure 6.10 Scenario analysis 5 results: Cost-effectiveness plane for initial treatment strategies for cFPM

Despite increasing the WTP value for a temporary filling, initial management strategies of temporary filling and active monitoring are dominated. The initial strategy of extraction is more efficient than filling.

6.6.7 Deterministic Sensitivity Analysis - Scenario Analysis 6

Scenario analysis 6 involved including the WTP decrements, due to adverse events, into the model. The results from scenario analysis 6 are shown in **Table 6.35, 6.36, 6.37** and **6.38** which summarise the costs, WTP and overall incremental analyses. In **Table 6.37** all initial strategies are presented in terms of ascending order of cost. Strategies which were more costly and less beneficial where strictly dominated. These were excluded from further incremental analysis as shown in **Table 6.38**. The cost-effectiveness plane for initial treatment strategies for cFPM, based on scenario analysis 6, are shown in **Figure 6.11.**

Statistic	Cost (Filling (Non-RCT))	Cost (Temp Filling)	Cost (Active monitoring)	Cost (Extraction)
Mean	648.60	1160.25	771.51	1131.87
Std Deviation	103.99	700.11	498.59	838.26
Minimum	230.37	416.36	289.68	235.95
2.5%	496.50	531.28	452.08	481.40
10%	538.13	567.71	492.31	518.49
Median	633.29	657.29	590.90	601.00
90%	776.47	2215.58	1622.64	2651.54
97.5%	896.22	2552.31	2417.52	2999.25
Maximum	2144.95	2966.08	3384.87	3820.74
95%% Lower Bound	646.56	1146.52	761.74	1115.44
95%% Upper Bound	650.64	1173.97	781.28	1148.30

Table 6.35 Scenario analysis 6 results: costs associated with initial treatment strategies for cFPM

Statistic	WTP (Filling (Non-RCT))	WTP (Temp Filling)	WTP (Active monitoring)	WTP (Extraction)
Mean	3740.89	6098.87	242.04	7232.57
Std Deviation	2153.05	14730.07	8673.84	15710.38
Minimum	-15963.98	-23766.68	-28299.56	-25676.55
2.5%	-2535.81	-19488.89	-22807.54	-21406.82
10%	1034.97	-17039.96	-11444.66	-19033.77
Median	4642.73	16017.41	0.00	17187.07
90%	5105.86	18988.02	13016.96	20001.14
97.5%	5291.49	20198.08	14752.50	21171.36
Maximum	5820.55	22924.82	18351.56	24193.54
95%% Lower Bound	3698.69	5810.16	72.03	6924.65
95%% Upper Bound	3783.09	6387.57	412.04	7540.49

Table 6.36 Scenario analysis 6 results: WTP associated with initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Effects	Incr. Eff	NMB	Incr. NMB
Filling (Non-RCT)	648.60	--	3740.89	--	3092.29	--
Active monitoring*	771.51	122.91	242.04	-3498.85	-529.47	-3621.76
Extraction	1131.87	360.36	7232.57	6990.53	6100.7	6630.17
Temp Filling*	1160.25	28.38	6098.87	-1133.7	4938.62	-1162.08
n.b * - dominated strategy						

Table 6.37 Scenario analysis 6 results: incremental analysis for all initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Eff.	Incr. Eff	NMB	Incr. NMB	Incr. B/C ratio
Filling (Non-RCT)	648.6	--	3740.89	--	3092.29	--	--
Extraction	1131.87	483.27	7232.57	3491.68	6100.7	3008.41	7.225112

Table 6.38 Scenario analysis 6 results: incremental analysis, after excluding dominated strategies, associated with initial treatment strategies for cFPM

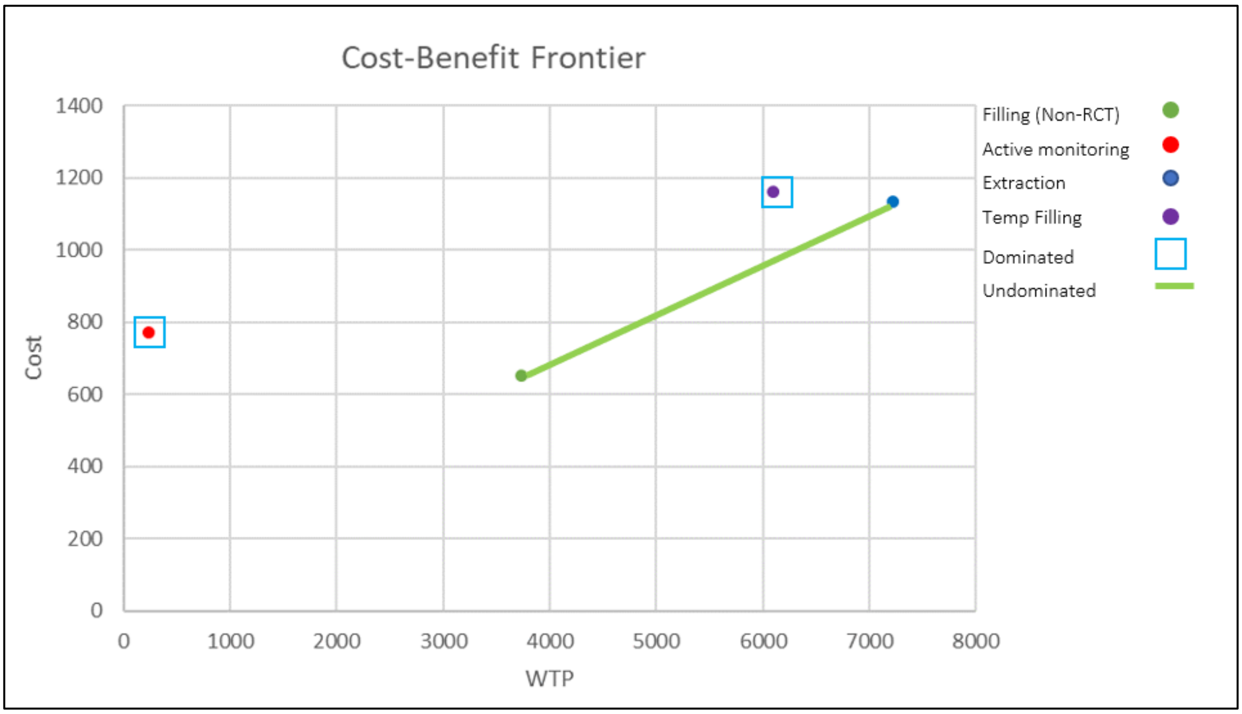

Figure 6.11 Scenario analysis 6 results: Cost-effectiveness plane for initial treatment strategies for cFPM

Despite including WTP for adverse events, the initial management strategies of temporary filling and active monitoring remain dominated. The initial strategy of extraction is still more efficient than filling.

6.6.8 Deterministic Sensitivity Analysis - Scenario Analysis 7

Scenario analysis 7 involved altering the time horizon from lifetime (100-years-old) to 18-years-old. The results from scenario analysis 7 are shown in **Table 6.39, 6.40, 6.41** and **6.42** which summarise the costs, WTP and overall incremental analyses. In **Table 6.41** all initial strategies are presented in terms of ascending order of cost. Strategies which were more costly and less beneficial where strictly dominated. These were excluded from further incremental analysis as shown in **Table 6.42**. The cost-effectiveness plane for initial treatment strategies for cFPM, based on scenario analysis 7, are shown in **Figure 6.12.**

Statistic	Cost (Filling (Non-RCT))	Cost (Temp Filling)	Cost (Active monitoring)	Cost (Extraction)
Mean	539.73	990.83	646.64	992.96
Std Deviation	77.76	554.67	404.70	696.01
Minimum	122.99	126.08	73.78	218.19
2.5%	413.69	466.04	381.73	416.49
10%	452.70	500.07	419.62	449.71
Median	532.06	579.11	503.73	525.81
90%	631.86	1640.34	1522.16	2084.30
97.5%	695.40	2015.29	1839.71	2449.03
Maximum	1898.15	2412.30	2680.23	3110.24
95%% Lower Bound	538.21	979.96	638.71	979.32
95%% Upper Bound	541.26	1001.70	654.58	1006.61

Table 6.39 Scenario analysis 7 results: costs associated with initial treatment strategies for cFPM

Statistic	WTP (Filling (Non-RCT))	WTP (Temp Filling)	WTP (Active monitoring)	WTP (Extraction)
Mean	5031.73	4980.03	264.58	5190.86
Std Deviation	243.21	2637.42	1668.97	3796.48
Minimum	2047.58	-923.40	-6869.96	-3743.96
2.5%	4547.86	215.92	-4615.77	-2459.51
10%	4725.18	707.14	-1690.03	-1180.33
Median	5030.44	5983.97	293.38	7117.68
90%	5340.24	7643.52	2599.06	8670.57
97.5%	5502.18	7967.33	3486.99	9195.73
Maximum	5989.57	8740.59	4732.44	10273.73
95%% Lower Bound	5026.96	4928.34	231.87	5116.45
95%% Upper Bound	5036.50	5031.73	297.29	5265.27

Table 6.40 Scenario analysis 7 results: WTP associated with initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Effects	Incr. Eff	NMB	Incr. NMB
Filling (Non-RCT)	539.73	--	5031.73	--	4492	--
Active monitoring*	646.64	122.91	264.58	-4767.15	-382.06	-4874.06
Temp Filling	990.83	360.36	4980.03	4715.45	3989.2	4371.26
Extraction	992.96	28.38	5190.86	210.83	4197.9	208.7
n.b * - dominated strategy						

Table 6.41 Scenario analysis 7 results: incremental analysis for all initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Eff.	Incr. Eff	NMB	Incr. NMB	Incr. B/C ratio
Filling (Non-RCT)	539.73	--	5031.73	--	4492	--	
Temp Filling	990.83	451.1	4980.03	-51.7	3989.2	-502.8	-0.11461
Extraction	992.96	2.13	5190.86	210.83	4197.9	208.7	98.98122

Table 6.42 Scenario analysis 7 results: incremental analysis, after excluding dominated strategies, associated with initial treatment strategies for cFPM

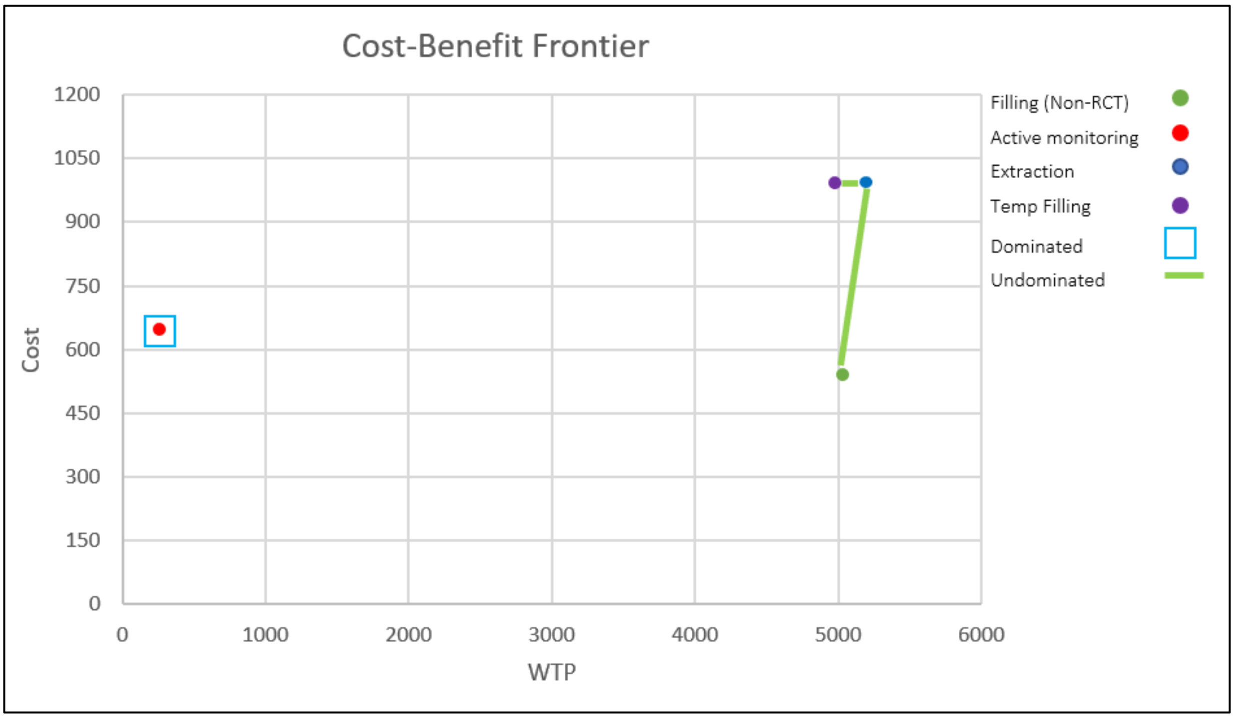

Figure 6.12 Scenario analysis 7 results: Cost-effectiveness plane for initial treatment strategies for cFPM

When reducing the time horizon from 100 years to 18 years, the initial management strategy of active monitoring is dominated. Extraction is the most efficient option. The negative incremental NMB between filling and temporary filling suggests that temporary filing should not be an option that is invested in.

6.6.9 Deterministic Sensitivity Analysis - Scenario Analysis 8

Scenario analysis 8 involved altering the WTP values in the model to reflect those from the mixed-logit analyses. The results from scenario analysis 8 are shown in **Table 6.43, 6.44, 6.45** and **6.46** which summarise the costs, WTP and overall incremental analyses. In **Table 6.45** all initial strategies are presented in terms of ascending order of cost. Strategies which were more costly and less beneficial where strictly dominated. These were excluded from further incremental analysis as shown in **Table 6.46**. The cost-effectiveness plane for initial treatment strategies for cFPM, based on scenario analysis 8, are shown in **Figure 6.13.**

Statistic	Cost (Filling (Non-RCT))	Cost (Temp Filling)	Cost (Active monitoring)	Cost (Extraction)
Mean	649.97	1169.84	755.35	1147.05
Std Deviation	106.65	700.04	473.90	854.29
Minimum	76.15	74.82	22.94	75.89
2.5%	495.52	529.50	448.74	481.38
10%	540.10	567.92	494.72	516.51
Median	633.35	661.68	591.22	602.55
90%	780.30	2218.04	1600.43	2690.90
97.5%	902.30	2558.37	2379.57	3015.38
Maximum	2379.42	3073.30	3056.74	3513.80
95%% Lower Bound	647.88	1156.12	746.06	1130.30
95%% Upper Bound	652.06	1183.56	764.64	1163.79

Table 6.43 Scenario analysis 8 results: costs associated with initial treatment strategies for cFPM

Statistic	WTP (Filling (Non-RCT))	WTP (Temp Filling)	WTP (Active monitoring)	WTP (Extraction)
Mean	4174.20	4978.32	-74.06	5352.40
Std Deviation	2254.50	13639.61	7923.64	14393.07
Minimum	-20420.84	-30805.35	-37755.17	-35563.76
2.5%	-2499.50	-21939.09	-22743.42	-24184.53
10%	1555.19	-16081.57	-10172.05	-17747.51
Median	5040.53	12825.78	244.03	13294.71
90%	5556.27	17741.22	10236.30	18134.09
97.5%	5771.12	20402.25	13638.50	20964.54
Maximum	18366.82	27605.17	20546.23	28673.48
95%% Lower Bound	4130.01	4710.99	-229.36	5070.31
95%% Upper Bound	4218.38	5245.65	81.24	5634.50

Table 6.44 Scenario analysis 8 results: WTP associated with initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Effects	Incr. Eff	NMB	Incr. NMB
Filling (Non-RCT)	649.97	--	4174.2	--	3524.23	--
Active monitoring*	755.35	105.38	-74.06	-4248.26	-829.41	-4353.64
Extraction	1147.05	391.7	5352.4	5426.46	4205.35	5034.76
Temp Filling*	1169.84	22.79	4978.32	-374.08	3808.48	-396.87
n.b * - dominated strategy						

Table 6.45 Scenario analysis 8 results: incremental analysis for all initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Eff.	Incr. Eff	NMB	Incr. NMB	Incr. B/C ratio
Filling (Non-RCT)	649.97	--	4174.2	--	3524.23	--	
Extraction	1147.05	497.08	5352.4	1178.2	4205.35	681.12	2.370242

Table 6.46 Scenario analysis 8 results: incremental analysis, after excluding dominated strategies, associated with initial treatment strategies for cFPM

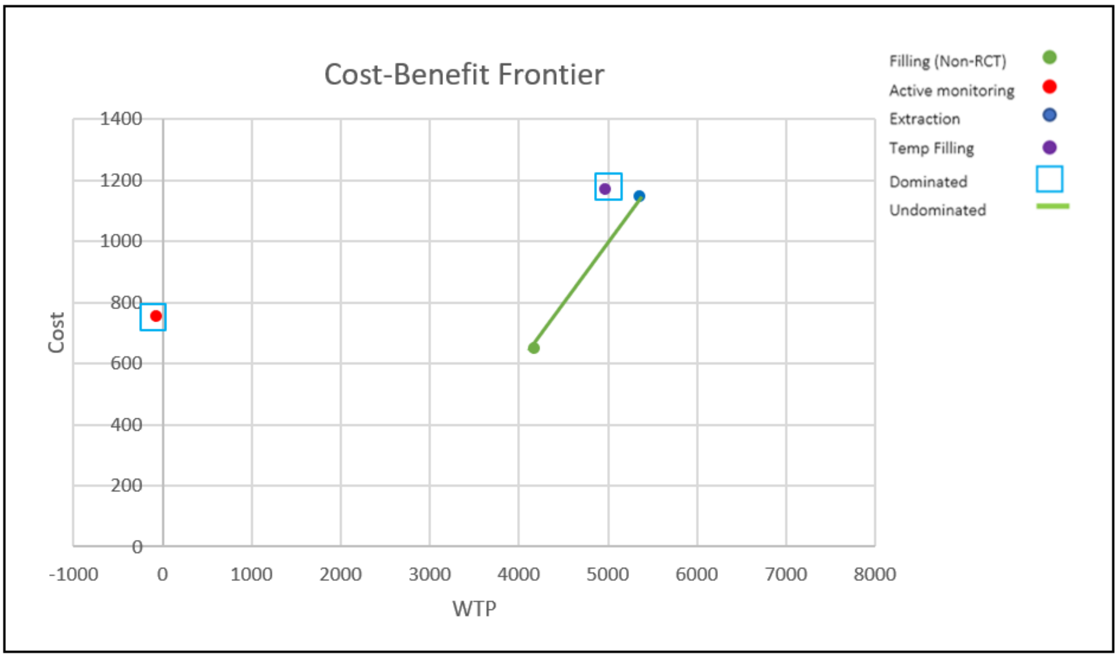

Figure 6.13 Scenario analysis 8 results: Cost-effectiveness plane for initial treatment strategies for cFPM

Using the WTP values obtained from the mixed-logit analysis, similar results are observed to that of the base-case scenario. Initial management strategies of temporary filling and active monitoring are dominated. Thus, these options should not be considered as the costs outweigh the benefits. The initial strategy of extraction has a higher net monetary benefit than filling.

6.6.10 Probabilistic Sensitivity Analysis – Base Case Scenario

The results from the PSA for the base-case scenario (a child with a cFPM, caused by dental caries, as part of a cost-benefit analysis) are shown in **Tables 6.47, 6.48, 6.49** and **6.50** which summarise the costs, WTP and overall incremental analyses. In **Table 6.49** all initial strategies are presented in terms of ascending order of cost. Strategies which were more costly and less beneficial where strictly dominated. These were excluded from further incremental analysis as shown in **Table 6.50**. The cost-effectiveness acceptability curve and cost-effectiveness plane for the PSA are shown in **Figure 6.13** and **Figure 6.14** respectively.

Statistic	Cost (Filling (Non-RCT))	Cost (Temp Filling)	Cost (Active monitoring)	Cost (Extraction)
Mean	649.77	1159.56	764.25	1137.84
Std Deviation	1.10	7.10	5.04	8.42
Minimum	646.57	1137.90	751.00	1114.71
2.5%	647.59	1145.78	754.56	1121.67
10%	648.39	1150.45	757.71	1126.60
Median	649.76	1159.64	764.13	1137.63
90%	651.19	1168.87	770.60	1149.30
97.5%	651.95	1173.75	774.32	1154.29
Maximum	653.87	1184.47	780.58	1164.31
95%% Lower Bound	649.71	1159.12	763.94	1137.32
95%% Upper Bound	649.84	1160.00	764.56	1138.36

Table 6.47 PSA base-case scenario results: costs associated with initial treatment strategies for cFPM

Statistic	WTP (Filling (Non-RCT))	WTP (Temp Filling)	WTP (Active monitoring)	WTP (Extraction)
Mean	3984.08	6308.41	276.99	7584.26
Std Deviation	20.54	146.06	87.11	150.42
Minimum	3913.58	5859.76	-1.27	7083.09
2.5%	3942.95	6015.12	99.18	7288.19
10%	3956.61	6124.28	163.16	7380.86
Median	3984.44	6304.55	280.65	7580.49
90%	4009.89	6505.24	385.84	7777.36
97.5%	4024.74	6580.72	443.21	7890.09
Maximum	4046.95	6803.94	526.41	8006.52
95%% Lower Bound	3982.80	6299.35	271.59	7574.93
95%% Upper Bound	3985.35	6317.46	282.38	7593.58

Table 6.48 PSA base-case scenario results: WTP associated with initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Effects	Incr. Eff	NMB	Incr. NMB
Filling (Non-RCT)	649.77	--	3984.08	--	3334.31	--
Active monitoring*	764.25	114.48	276.99	-3707.09	-487.26	-3821.57
Extraction	1137.84	373.59	7584.26	7307.27	6446.42	6933.68
Temp Filling*	1159.56	21.72	6308.41	-1275.85	5148.85	-1297.57
n.b * - dominated strategy						

Table 6.49 PSA base-case scenario results: incremental analysis for all initial treatment strategies for cFPM

Strategy	Cost	Incr. Cost	Eff.	Incr. Eff	NMB	Incr. NMB	Incr. B/C ratio
Filling (Non-RCT)	649.77	--	3984.08	--	3334.31	--	
Extraction	1137.84	488.07	7584.26	3600.18	6446.42	3112.11	7.37636

Table 6.50 PSA base-case scenario results: incremental analysis, after excluding dominated strategies, associated with initial treatment strategies for cFPM

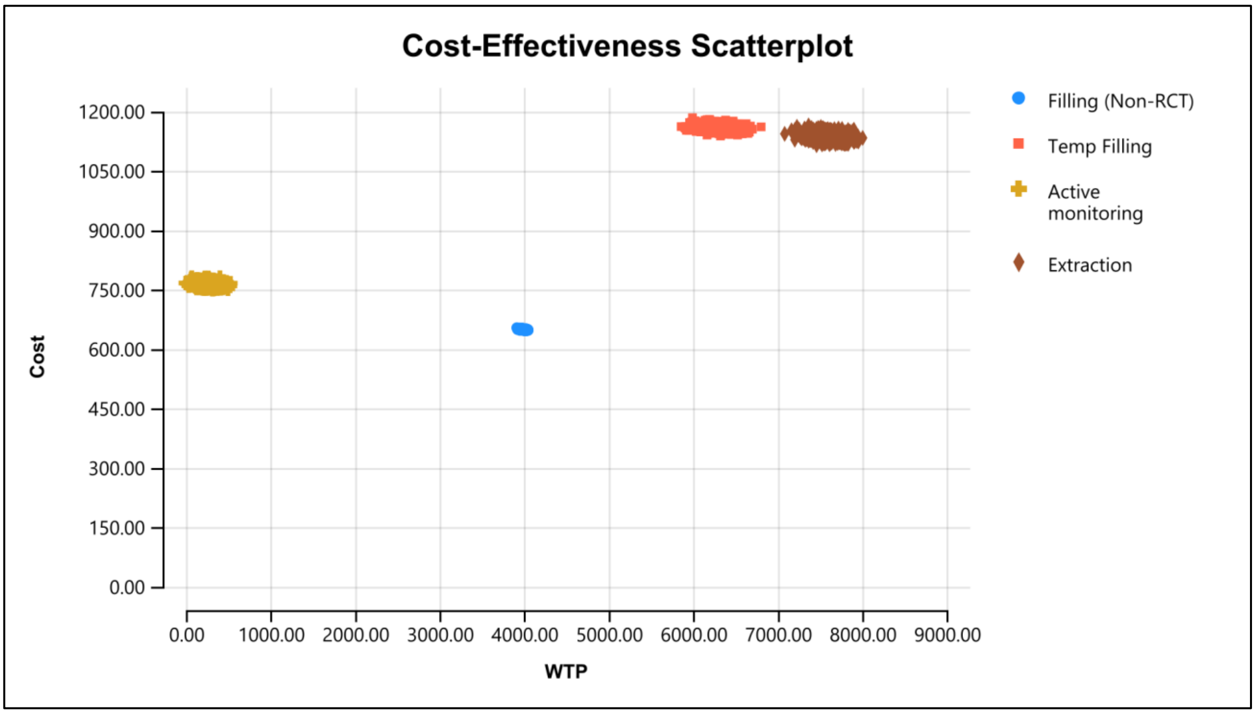

Figure 6.14 PSA base-case scenario results: cost-effectiveness plane associated with initial treatment strategies for cFPM

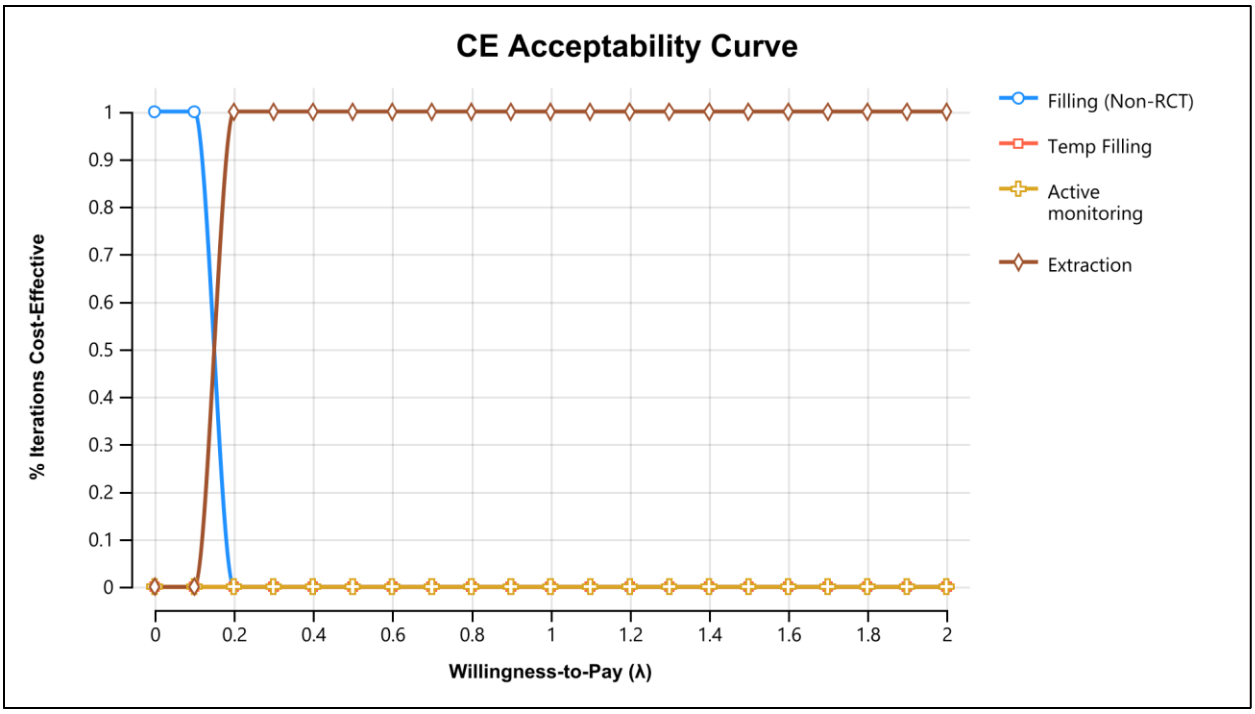

Figure 6.15 PSA base-case scenario results: cost-effectiveness acceptability curve associated with initial treatment strategies for cFPM

For the PSA of the base-case scenario, the initial management strategies of temporary filling and active monitoring are dominated. Extraction remains the most efficient option and has a higher net monetary benefit than filling.

6.7 Discussion

The following section will discuss the interpretation, and implication of the model findings, followed by the strengths and limitations of this study.

6.7.1 Interpretation and implication of model

Base Case Scenario

The base-case scenario involved a child with a cFPM, caused by dental caries, as part of a cost-benefit analysis. Initial management strategies of temporary filling and active monitoring are dominated. Thus, these options should not be considered as the costs outweigh the benefits. The initial strategy of extraction has a higher net monetary benefit than filling. This means that from an economic perspective having an extraction anytime between the ages of 7 and 10 is more efficient than having the filling completed. This is despite perfect spontaneous space closure occurring on less than 50% of lower cFPM (Eichenberger et al., 2015). Despite not being the most efficient, filling is still an option that could be deemed efficient. This is important because primary care practitioners (non-specialists) are more inclined to restore, than extract, these teeth (Taylor et al., 2019). It is re-assuring that active monitoring is dominated. Choosing this strategy will likely cause further problems such as pain, risk of infection etc. and should not be suggested. In the base case analysis, the temporary filling strategy is also dominated. This is important because previous evidence has suggested temporising and delaying extraction until the optimal development time (Cobourne et al., 2014; Ashley & Noar, 2019) yet the evidence from the analysis presented here is that this is inefficient.

Scenario 1 & 2

These scenario analyses addressed a child with a cFPM that was caused by dental caries or MIH respectively, as part of a cost-effectiveness analysis. Instead of using costs, and co-payments, that fell on the NHS, these scenarios included costs from an NHS dental practice perspective, based on micro costing. The most cost-effective option in both scenarios would be to fill the tooth. The alternative initial management strategies of extraction, temporary filling and active monitoring are dominated as they are all more costly and less effective. Although presented as a positive ICER, it is worth noting that a higher number of future visits of treatment should be considered a negative outcome. This makes interpretation slightly more challenging.

Based on these results, this analysis would suggest that for a carious cFPM it would cost the practice £403.13 per treatment visit avoided to actively monitor the cFPM. Similar, it would cost £22.49 per treatment visit avoided and £129.54 per treatment visit avoided for an extraction and temporary filling respectively. For an MIH-affected cFPM, it would cost the practice £210.64 per treatment visit avoided to actively monitor the cFPM. Similar, it would cost £175.95 per treatment visit avoided and £188.05 per treatment visit avoided for an extraction and temporary filling respectively.

This finding is important because primary care practitioners (non-specialists) prefer to restore than extract cFPM (Taylor et al., 2019). This would support their clinical decision as filling is the least costly (to them) and most effective strategy, in terms of having the least number of future treatments visited associated with it, over the life course of a patient. Similarly, for MIH, current evidence would suggest that MIH affected cFPM (as described in this thesis) can successfully be restored (Lygidakis et al., 2022; Somani et al., 2022). However, it is accepted that the severity and presentation of MIH will differ across patients. Thus, if these findings are to be adopted into clinical practice, then they should be done cautiously whilst educating primary care practitioners appropriately on the techniques required to achieve success (Lygidakis et al., 2022).

However, the results of this analysis need to be caveated by two points. Firstly, if a space were to occur and orthodontics were required, the costs and visits are unlikely to fall wholly on the non-specialist primary care practitioner's practice as these visits are likely to be completed externally by an orthodontist. Whereas, given the structure and assumptions (only available up to the age of 18) of the model, it is unlikely that any child initially having a filling will incur orthodontics; therefore, the number of future treatment visits is likely to fall completely on this practitioner. This is a nuance of dental treatment, as referral is often required for treatments that are out with the scope of non-specialist practitioner. Secondly, comparing to the filling strategy, the incremental difference in the mean number of future visits required are (for the carious cFPM) 1.44, 4.19 and 4.05 and (for the MIH-affected cFPM) 1.53, 4.22 and 4.46 for active monitoring, extraction, and temporary filling respectively. Therefore, it is worth considering whether saving on average four future treatment visits, over a lifetime, by filling the cFPM, compared to extracting it at any age between 7-10 years old or temporising and delaying extraction until the optimal time, is of value to patients.

Scenario 3

In this scenario analysis, both costs and benefits were undiscounted, e.g., the discount rate was set to 0%, which is a sensitivity analysis recommended by NICE for any economic evaluation (NICE, 2022a).

Discounting accounts for the fact that individuals would prefer to have both money and health benefits, now, rather than later. Undiscounted costs and benefits mean cost and benefits occurring in the future are weighted equally to the those cost and benefits that occur now (Nord, 2011; Drummond et al., 2015). As expected, costs of all strategies increased, whereas benefits reduced for filling and active monitoring, but increased substantially for extraction and temporary filling strategies. This is likely due to where the benefits are attached in the model design. WTP values for spontaneous space closure and orthodontic closure were obtained sooner than those WTP values obtained by having a filling as an initial strategy. A narrative comparison of the INB between the base-case, and this scenario, suggests having an extraction carried out between 7-10-years-old is more efficient than a filling when the costs and benefits are undiscounted.

Scenario 4 & 5

As described in section 6.4.8, the WTP value for a temporary filling was determined as the arbitrary mid-point cut off between £0 (WTP for active monitoring) and £647 (permanent filling) i.e., £332.35 in the base-case scenario. In scenario analysis 4, the WTP value of temporary filling was changed to £161.63 (25% of the WTP value of a filling) and increased to £493.88 (75% of the WTP value of a filling) for scenarios analysis 5.

As expected, in both scenarios, a lower or higher WTP value for the temporary filling resulted in an increase or reduction of the mean WTP for this strategy, compared with the base-case. Like the base-case scenario, for both scenario analyses, initial management strategies of temporary filing and active monitoring were dominated, therefore, suggesting the WTP value associated with a temporary filling made no overall effect on the most efficient strategies for managing cFPM.

A future iteration of the DCE completed in Chapter 5, that includes a temporary filling strategy, could provide a WTP value for use in a future version of this model. However, this raises an interesting point, as the temporary filling strategy is in essence an intermediary state as it leads to a future health state (extraction followed by closure) which is likely to be valued differently. Furthermore, it perhaps highlights the difficulty in valuing interventions versus health states. There remains debate in the literature about whether to value health profiles or health states (Devlin et al., 2019; Helgesson et al., 2020).

This dichotomy in valuing interventions vs. health states further supports the idea that including WTP, as a measure of benefit, in a model is challenging. Aligning a DCE and model pathways is feasibly possible but made harder when a combination of interventions and health states are included. The order in which of these to do first may have significance in achieving congruency. Building the model first would allow all health states and interventions to be incorporated into the DCE. However, it is likely, particular in the context of cFPM, that the DCE could be unmanageable. Iteratively designing the two has been suggested as a way to possibly overcome some of these issues (Vale et al., 2004). Additionally, it could be argued a DCE should be designed first, as this way it includes what patients value (Ryan et al., 2008; Tinelli et al., 2016). However, operationalising attributes and levels which are inconsistent with a model format make this challenging. Similar issues may be found if time horizons used in the DCE are inconsistent with the model structure. At present there is no agreed consensus as to which to do first, and therefore, challenges are likely to exist either way.

Scenario 6

In this scenario analysis, WTP decrements, due to adverse events of a repeat filling, root-canal treatment, and future extraction, were introduced into the model.

In the base-case scenario, WTPs values from the DCE were used on the assumption that the DCE respondents had fully understood the information provided and had understood the future ramifications of treatment options as was described to them e.g., filling will need repaired. However, for some respondents it is perfectly feasible that future adverse events were not included in their valuations, and therefore there may be a need to obtain separate WTP values for adverse events. However, it is acknowledged that using this approach risks double counting of WTP.

As expected, the mean WTP reduced for the initial strategies active monitoring, temporary filling, and permanent filling in this scenario analysis, as these pathways incur future adverse events. Whereas the mean WTP for extraction between this scenario analysis and the base-case were similar – as the initial extraction pathway had no additional future adverse event included. That is not to say those in the extraction arm did not get any WTP that was related to an adverse event, as a WTP of -£1055.60 was attached to patients if they were left with a full space, rather than having this space closed with orthodontics or a prosthetic. Like the base-case, the initial management strategies of temporary filling and active monitoring were dominated. Whilst the initial strategy of extraction was more efficient than filling.

Scenario 7

Scenario analysis 7 altered the time horizon from lifetime (100-years-old) to 18-years-old. This was chosen to reflect what happens when out of pocket costs (i.e., patient charges) are not included. As expected, the mean costs across all strategies reduced, as patients had less time to build up costs in the model. However, in comparison, these were much higher than anticipated, and suggests that most of the costs accrued for managing cFPM, and any meaningful subsequent managements, were likely to have been completed before this time horizon e.g., filling, orthodontic closure etc. An additional explanation is that discounting has quite an effect when costs are incurred in the long term. Interestingly, the costs associated with filling were lower than the temporary filling strategy, as the future costs of the filling arm e.g., filling repairs, filling replacements, root canal treatment and re-root canal treatments were less likely to be included, as they typically occurred in those aged older than 18 years of old.

In the scenario analysis, active monitoring is dominated by the other strategies. Extraction is the most efficient option, whereas the negative incremental NMB and incremental benefit-cost ratio would suggest that compared to filling, the temporary filling strategy should never be adopted. This analysis is an interesting finding, as it suggests that if costs were to entirely fall on the NHS (that is no patient charges) and the horizon of interest is short, then extracting would remain the most efficient strategy, however, filling the cFPM would still be an efficient option. However, it is unlikely that the provision of NHS dentistry is likely to abolish patient costs in the future.

Scenario 8

Subtle differences were noted in the magnitude, but not direction, of WTP values which were obtained from the mixed logit model (see section 5.6.1). Thus, it is unsurprisingly that like the base-case scenario, initial management strategies of temporary filling and active monitoring are dominated. Therefore, these options should not be considered as the costs outweigh the benefits. Furthermore, the initial strategy of extraction has a higher net monetary benefit than filling and means, from an economic perspective, that having an extraction anytime between the ages of 7 and 10 is more efficient than having the filling completed. Plausible explanations of this sensitivity analysis are similar to those found in the interpretation of the base-case scenario, earlier in this chapter.

Probabilistic Sensitivity Analysis

The PSA of the base-case scenario proposes that initial management strategies of temporary filling and active monitoring are dominated. Extraction remains the most efficient option and has a higher net monetary benefit, and positive benefit-cost ratio than filling. From an economic perspective, having an extraction anytime between the ages of 7 and 10 is more efficient than having the filling completed. In addition, the deterministic and probabilistic base case results support good internal validity are the results are consistent which re-affirms that the distributions have behaved as intended.

The cost-effectiveness plane, including the scatterplot, as shown in **Figure 5.13** confirm the dominated and undominated strategies but enable the amount of uncertainty surrounding each of these strategies to be observed. Through the 1000 simulations, the spread of the points along the horizontal plane of active monitoring, temporary filling and extraction indicates that there is some uncertainty regarding the magnitude of WTP. In contrast, there is less spread in the vertical plane, suggesting there is less uncertainty around the true costs for each of these options. The CEAC, shown in **Figure 5.14**, obtained from this PSA compares two NMBs of alternative options. Using the λ of 1, it is apparent that extraction is the optimal strategy across most iterations of the PSA. An observation from the PSA confirms that there appears to be very little uncertainty surrounding the most appropriate treatment. This, in part, could be explained by the large sample size used to inform the probability parameters, the certainty of the UDA system and the tight confidence intervals on the WTP data. Alternatively, it could suggest that the PSA has not captured the full range of parameter uncertainty that might be expected, based on the distributions that have been fitted. On the other hand, some structural decisions will not have been captured in the PSA (e.g., uncertainty in the number of fillings that might be undertaken before replacement).

These results confirm what has been discussed previously in this section, that despite perfect spontaneous space closure occurring on less than 50% of lower cFPM (Eichenberger et al. 2015), it is still the most efficient option from an economic perspective. Although less efficient than extraction, filling the cFPM remains an efficient option which is important as primary care practitioners (non-specialists) have been reported to prefer restoring, compared to extracting, these teeth (Taylor et al. 2019).

6.7.2 Strengths and limitations

These findings should be interpreted, and caveated, by the strengths and limitations associated with this model. These are discussed in the remainder of this sub-section.

Modelling cFPM

Currently, there is no accurate data that represents the real-life uptake of the various treatment options for managing children with cFPM in primary care dental services. In addition, data which is condition-specific, population-specific or healthcare system-specific does not exist. Having such data will be support service provision and resource planning for as long as accurate coding of interventions are undertaken (Getting It Right First Time, 2021). Having better data for the mutually exclusive strategies, from primary care dental services, in addition to developing longitudinal data sets that accurately predict success and failure over time, will provide more accurate estimates for future models.

Of course, this model only provides results for an isolated lower cFPM. However, there are four cFPM usually in the mouth. In some circumstances, decisions made for one cFPM could have implications for another e.g., a compensating extraction of an opposing healthy tooth to reduce over-eruption.

However, modelling four cFPM in one model is likely to be computationally challenging. This is discussed further in the section 7.5 of the discussion chapter

Input parameter Estimates

Several of the input parameters used in the thesis were from expert opinion. Iteratively utilising the expert panel member's expertise within the model (and DCE) design was a strength. Critical comments provided by these members helped with ensuring the model and DCE were as congruent as possible, whilst reflecting reality, giving it good external validity.

It was deliberate that as far as possible, parameter values were only obtained from data sources if data on cFPM could be extrapolated. Using other parameters that may have included cFPM data would likely alter the results of this model. However, these were unlikely to be reflect reality. However, in the absence of valid, reliable, and applicable parameter estimates, expert opinion is as an alternative (albeit so long as suitable levels of uncertainty around such estimates are built into the model). In some cases, values used within the model (e.g., adverse events) were taken from other sources. The derivation of parameter estimates from multiple sources is a common occurrence in decision-models. The key things are to explore the applicability of the identified data to the decision question posed and to explore the impact of using alternative values/sources of data.

A limitation in the model was assuming the WTP values for adverse events. Certainly using a comparable study (Lord et al., 2015) provided, in part, a solution to this issue. However, using data from other DCEs does raise issues about how generalisable the data is to the given decision context of cFPM. The WTP values obtained were for permanent teeth, and not specifically cFPM, however, in the absence of any better evidence, this provided a set of values whose use could be explored. Undertaking one-way deterministic sensitivity analyses and adjusting these parameters e.g., fundamentally lower, or fundamentally higher, may have proved beneficial, however, what direction and magnitude change to make would be based purely on anecdotal opinion. One way to overcome this issue would have been to design the DCE, used in this thesis, to include adverse events. However, the qualitative work that fed into the DCE design showed that future adverse events were not of notable importance by those interviewed, hence they were not included in the final design of the DCE. A future DCE could be designed to specifically address adverse events resulting from a failed filling or extraction of a cFPM and obtain WTP values for these. This would provide the necessary input parameters for the model.

Similarly, to including adverse events, not having an accurate WTP for the temporary filling may have limited the outcomes of this model, although, across all scenario analyses, temporary filling was a dominated strategy. In addition, how the model is structured meant glass ionomer was only used for a temporary filling and that after a maximum of 5 cycles (up to age 9.5), this tooth was then extracted. This assumption may not fully reflect reality, as it could be entirely feasible that a restoration was placed at this time point. In addition, the tooth could be temporised with an alternative material, such as a preformed metal crown (Keightley & Surendran, 2021; Lygidakis et al., 2022). Despite being relatively uncommon for carious and hypomineralised cFPM in the UK (Taylor et al., 2019), including these in future iterations of this model would provide different answers given that public valuations of having a preformed metal crown would likely be very different to a glass ionomer filling.

Despite already being a complex model, further complexity could have been added by including survival data within each parameter, or including events such as orthodontic relapse, or partial space closure. Time was assumed to be linear in this model, meaning that transition probabilities were fixed. This limits the outputs of this model, as in reality, failures and successes of treatments vary with time. There were studies used for input parameters that had survival data attached (Burke & Lucarotti, 2018b, 2018c, 2018d), however these rates were not adjusted in the model. This may be a limitation, but it was felt that as insufficient evidence existed for all time-dependent probabilities, specifically in relation to molar teeth, then no probability would vary over time.

Despite the initial strategies reflecting the management of cFPM, some future options, and variations of treatments, were not considered. It has been suggested that for any tooth that has been managed with a root canal treatment, a crown is used to restore the tooth (Mannocci et al., 2021). However, it was assumed in this model that a composite restoration was placed instead, as the cFPM had a single surface lesion. This could be considered a limitation of the model; however, it is likely to be more reflective of current practice in the UK (Gemmell et al., 2020; Edwards et al., 2021)

Another example of a limitation of a specific health state in this model was that for prosthetic closure, the only option was a resin retained bridge. Alternatives options such as implants and dentures exist, however, given the CBA framework was from an NHS perspective, the use of an implant would not be consistent with this perspective. Furthermore, it is unlikely these options would be used to replace a missing molar tooth in a child (Hjalmarsson et al., 2016; Taylor et al., 2019; Alkhalaf et al., 2020); a potential that the current model structure would allow. The structure of the model could have been adjusted to include a denture as part of the care pathway that someone could follow a certain age. This would have increased the complexity of an already computationally challenging model.

Chosen Cycle Length

The chosen length of six months was used to reflect routine dental check-ups for patients with current active dental disease (Public Health England, 2021). However, in the model, it is assumed that prevention will be implemented, and therefore, for some, the length of recall could be extended to as much as two years to reflect a reduction in risk of dental disease (Public Health England, 2021). However, it is not feasible to have a model that has two different cycle lengths. One method to overcome this could have been to include a table that indicates the management schedule indicating whether a patient has a recall in each cycle. As already explained, this would have increased the complexity of an already computationally challenging model. Similarly, it could be feasible that probabilities of events occurring are manipulated within the model, to reflect those who reduced their disease risk, likely meaning a reduction in the failure rate of a treatment over time. This would be a large assumption to make, but future iterations of this model could include this as a sensitivity analysis.

Similarly, the chosen cycle length dictates when someone transitions, as this could occur at the beginning or the end of each cycle. This doesn't reflect realty, as for example those in pain would likely seek treatment between their check-up visits. Half-cycle correcting costs and benefits within the model accounted for this. In addition, events such as filling repair and re-root canal treatments were carried out as in-cycle events (ensuring the costs and benefits for these were not half-cycle corrected). These changes somewhat reflect reality better than not including them.

Cost Data

A strength of this analysis was the inclusion of patient charges (that is the contribution paid by patients towards the costs of dental goods or services) (Ternent et al., 2022). Furthermore, adjusting the costs to factor different patient charges, and including this in the model, reflected reality within the England. However, it was assumed in this model that all patients pay for their treatment. In reality, this is not likely to be the case. In the year 2018/2019, it was reported that 23.7% of all adult claims in NHS primary dental care were made by those who were fee exempt (Shah & Wordley, 2021).

Given the two overall perspectives, different costings approaches were required to populate this model. For the CBA, it was relatively straightforward as the UDA values for each treatment are set (British Dental Association, 2020). However, the value of one UDA was estimated at £25 (British Dental Association, 2020). This is likely to be similar to the true average value, but further detailed costing analysis using routinely collected data from NHS Business Services authority would likely have provided a more accurate valuation of the current mean UDA value.

For the cost-effectiveness analysis, a simplified micro costing approach was used. Using data from the 'Unit Costs of Health and Social Care 2021' booklet (Jones & Burns, 2021) meant a true micro-costing exercise was not undertaken. An alternative approach would be to undertake detailed costings of all resources. As an example, cost data could have been obtained from online dental suppliers instead, and where necessary, broken down to estimate a unit cost per item if consumables were only available to be bought in bulk. Treatment resources can be split into consumables (single use) and reusable. When reusable items are concerned, the equivalent annual cost of the could have be used as the basis of a cost per use (Ternent et al., 2022).

In addition, this model has only included NHS costs and co-payments. Private options for managing cFPM in children and adults do exist, with costs likely be far greater than what have been proposed here. It is hard to predict what this change would make, as to allow a fair comparison, establish WTP values for these interventions privately are likely to differ to what has been established in the DCE in this thesis.

Chosen perspective

An NHS perspective was adopted for the cost-benefit analysis, and therefore included costs that fall on the NHS only. In a true CBA, all costs, and all benefits regardless of who they may fall should be included where costs and benefits are valued in commensurate units. This is done by adopting a societal perspective. Therefore, choosing an NHS perspective is consistent with the second part of the definition but not the first. As such, the base-case scenario could be regarded as a cost-benefit framework analysis, allowing commensurate units to be included, but not fully adopting a societal perspective.

Choosing an NHS perspective precluded the inclusion of costs that fall on society, such as productivity losses. Oral health conditions have been shown to lead to productivity loss due to absenteeism and presentism (Breckons et al., 2018), and by not including these, could be a limitation of this model, given the direct and indirect impact cFPM has on patients, sibling and their parents (Taylor et al., 2018). A more detailed costings study with patients could have established these costs, penitent to cFPM, and this is something that could be considered for the future.

Value of information (VOI) analysis

One further consideration could have been to undertake a VOI analysis. A VOI analysis considers whether the intervention being adopted (or rejected), based on the current available evidence, would benefit from additional information to support the decision. Expected value of perfect information (EVPI), a common measure used in VOI analysis, predicts the monetary value of undertaking additional research to remove all uncertainty across the model. In contrast, expected value of perfect parameter information (EVPPI) is used to identify certain parameters to which additional information would be the most valuable to reduce uncertainty. Thus EVPI (or EVPPI) is the maximum value of removing uncertainty in each given model. It is accepted that uncertainties may be reduced, if new, additional information was produced by further research. However, as an opportunity cost is created in undertaking this research, a decision on how much additional value having additional information will provide on reducing uncertainties requires to be made (Briggs et al., 2006; Rothery et al., 2020). A VOI analysis is likely to be included in further research once the model has been refined.

6.8 Conclusion

It appears from the current model's findings that managing a cFPM by extracting as an initial option at any age between 7-10 is the most efficient choice. Although not as efficient, restoring this tooth would also be worth due consideration. Despite the caveats noted, and the further validation checks required, this is the first model for cFPM and child oral health that has undertaken using a CBA framework and included WTP values derived from a public DCE.

The results of this model will be discussed, alongside the findings from the qualitative study (see Chapter 4) and DCE (See Chapter 5), in the next chapter. This chapter summarises the key findings from the three studies and implications of the findings of patients, healthcare professionals, service provision/policy and researchers.

Chapter 7. General Discussion

7.1 Introduction

Several cross-cutting, common concepts have arisen from the qualitative interviews, DCE and decision analytical model. Section 7.2 will summarise the findings of this thesis, placing these in the context of the existing literature. This will be followed by section 7.3 which will detail the overall limitations of the approaches chosen, in answering the question of managing cFPM in children. This will be followed by section 7.4 which will discuss the implications this thesis will have on the patient, public, dental (and healthcare) professionals, policy/service provision and finally researchers. The final section, 7.5, will briefly address future research opportunities that have arisen from this thesis.

7.2 Overall summary of results

Two main areas of discussion that arose from this thesis were the management of cFPM (restore or extract) and the importance of shared decision making. Each of which will now be described.

7.2.1 Management of compromised first permanent molar teeth – restoration or extraction?

Prior to this thesis, there was insufficient evidence to support which of the two main options for managing cFPM, restoration or extraction, were superior for either dental caries or MIH (Ashley & Noar, 2019; Keightley & Surendran, 2021; Lygidakis et al., 2022). In addition, different approaches were elicited across various professional groups (Taylor et al., 2019; Alkadhimi et al., 2022).

Qualitative interviews and DCE

The decision to restore or extract was not as clear cut as it was expected to be from the findings of the DCE and the qualitative interviews. This generally fits with the existing literature as disparities in current management are noted within the UK, and beyond. It could be argued that compared to other countries, extraction of cFPM may be more common than restoration in the UK (Sanghvi et al., 2022; Lygidakis et al., 2022). A recent one year retrospective study from 2020, conducted in the UK, observed that of the 397 patients diagnosed with MIH, 52% (n=206) had extractions of one of more cFPM teeth (Humphreys & Albadri, 2020). Another UK-based study reported that 81% (*n* = 201) of children with cFPM had extraction of at least one of them (AlKhalaf et al., 2022). However, these two studies reported data from patients referred to consultant-led services. In comparison, it has been reported that primary care dental professionals in the UK may be more inclined to restore, than extract, cFPM (Taylor et al., 2019). Although, this study has limitations as it was a vignette study (not a record of actual treatment) in addition to having a relatively small sample size (Taylor et al., 2019). In comparison to the UK, several European studies report a clearly delineated preference towards restoration, primarily with composite (Kotsanos et al., 2005; Kopperud et al., 2016; Wall & Leith, 2020) and/or indirect restorations (Gaardmand et al., 2013; Dhareula et al., 2019).

Despite these country-level variations, at present there is little data on the actual uptake of treatments being carried out in cFPM in primary care in the UK. Routinely collected data does not permit this detailed level of analysis, as it does not include this level of information in the coding of procedures. Future efforts should be made to collect routine primary care data that is tooth- and condition-specific (Getting It Right First Time, 2021).

Across the sample of the public responding to the DCE, there was no clear direction of preference as to whether cFPM were restored or extracted, providing the resultant space was closed spontaneously or orthodontically. This finding was certainly interesting, given the predilection for choosing an extraction of cFPM in prior studies (Humphreys & Albadri, 2020; AlKhalaf et al., 2022). It is worth noting that these studies were cross-sectional evaluation of current practice and did not include the long-term outcomes. A cross-sectional design is a more simplistic method of preference elicitation compared to the DCE used in this thesis and it is impossible to know how much of the revealed preference was due to child, patient, or professional preference.

Irrespective of this, it is apparent that the public value a child not having a gap. This preference is most likely explained in that both restoration and extraction (followed by guaranteed spontaneous or orthodontic space closure) result in the same outcome; that is a functioning unit with no gap. Indeed, the WTP values noted are high for these options and certainly suggest the public place a great value on retaining the tooth or having a gap that is spontaneously or orthodontically closed following extraction. If there had there been a preference for one over the other, then any differences could likely be explained by either variation in procedural complexities or short-term outcomes of the options.

In contrast, when the outcome of the extraction left an unfavourable space (a partial or full space), the public sample from the DCE showed a clear preference for restoring this tooth. The WTP values for these options corroborated this finding. Interestingly, these WTP valuations are made by the public for a 9-year-old child, and not for them deciding how to manage their own cFPM. If the public had valued management options for their own cFPM, then, it could have been expected that WTP value for having a gap would have been different, as the option of having a gap for an adult was not something that did not overly concern them when interviewed. A recent qualitative study that interviewed parents of children who were having cFPM extracted under general anaesthetic reported a different finding that some parents were likely to be accepting of subsequent spaces, should a restorative option have been chosen and failed in the future resulting in a space; however, these parents may be biased in their opinions as they had already chosen to have these teeth extracted (Agel et al., 2021). In addition, public preferences elicited from the DCE in this thesis expressed their preferences based on a hypothetical situation rather than a recent real-life decision.

Public preferences for space closure were dependent on it being complete, something that cannot be guaranteed (Eichenberger et al., 2015). However, it is apparent the public value orthodontics to close the space, as an alternative to spontaneous closure. Certainly, orthodontic space closure of extracted cFPM can be done (Jacobs et al., 2011), although if carried out at a later stage than the extraction, then prolonged closure times might be expected (Ong & Bleakley, 2010), which is something that can be frustrating (Sayers & Newton, 2007; Jopson et al., 2022). Despite patient satisfaction being noted for the placement of resin retained bridge (King et al., 2015) the public preferred that any resultant gap from a cFPM in a child was not closed by a resin retained bridge. One explanation could be that members of the public don't want a prosthesis and prefer to retain natural tooth in the gap. Preference for retaining natural teeth was expressed by adults who had lost teeth as to them having a false tooth (or teeth) on a denture were a marker of old age (Rousseau et al., 2014). It is worth noting these findings were pertinent to removable dentures, rather than a resin-retained bridge but these sentiments could have resonated with the DCE sample, as despite only 3.3% (n=14) having no teeth left, 43.7% (n=188) were over the age of 60, and based on UK epidemiological oral health data, would likely to have lost a tooth by this point (Fuller et al., 2011). Alternatively, it could be that the public did not see the added functional benefit of a resin-retained bridge, and instead felt having a shortened dental arch was acceptable.

It is worth noting that across 4370 choices in the DCE, only 1.64% (n=78) were for no treatment. It can be implied that the public highly value the need to make a choice and manage cFPM in children. A recent German study qualitatively explored reasons why patients (not specifically children) decide not to undergo dental treatments (Felgner et al., 2022). The most important reasons for not choosing dental treatment were: out-of-pocket payments; the perceived dentists' training, and ability to provide the treatment; and, the (lack of) trust that could exist between a patient and dentist (Felgner et al., 2022). Therefore, one reason that might explain why there were no treatment options chosen was that some may have misunderstood that no out-of-pocket costs were required. Alternatively, it could be a true representation participant choice, as they valued both mutually exclusive treatment options less than doing nothing for the cFPM.

Model Results

As described in the Chapter 6, for the base-case scenario, the initial management strategy of extracting the cFPM between the ages of seven and ten was the most efficient strategy. Definitively restoring the cFPM was similarly an efficient strategy, but incrementally less so than extraction. In addition, both of these strategies dominated temporary filling and active monitoring strategies. Despite the assumptions made in this model, the scenario analyses and PSA demonstrate that extraction at any age between seven and ten is the most efficient option for managing a cFPM. However, definitively filling the tooth remains an efficient strategy, which is supportive of previous research which has shown that primary care practitioners (non-specialists) would rather restore, than extract, cFPM (Taylor et al., 2019).

In comparison to the model presented in this thesis, a recently published UK cost-effectiveness model addressed a similar clinical problem (Sanghvi et al., 2022). The authors of this study used a Markov model to simulate and compare, over the lifetime of the tooth (62 years), two management strategies: extraction facilitating spontaneous space closure and maintenance of teeth with restorations. The outcome measure used was the QATY. One key problem in this study is the authors assumed that any participant who had an extraction completed between 8-12 years old were assumed to have perfect spontaneous closure as their outcome (Sanghvi et al., 2022). This does not fit with the most up-to-date evidence (Eichenberger et al., 2015) nor is likely to fully reflect the cost and benefits of this strategy. In comparison, the model used in this thesis accounted for variations in the success of spontaneous closure by varying the transition probabilities based on age having this treatment carried out. In addition, choosing a Markov model structure (Sanghvi et al., 2022) automatically meant no 'memory' was included in the model (Briggs et al., 2006). In the case of cFPM, previous restorations, for example, will impact how that tooth is managed in the future. Therefore, adopting a microsimulation model (as done in this thesis) supports the complexities that are observed in reality for patients with cFPM (Taylor et al., 2019; AlKhalaf et al., 2022).

Despite the critical assumption noted above, the key finding was that irrespective of the number of cFPMs affected, retention was always more effective than early removal, generating an additional 2.3 QATY's per cFPM. This result is not surprising, given the way in which the health state utilities used in this model were elicited (Sanghvi et al., 2022). In a separate study, valuations of each health state were elicited using ranking, visual analogue scale (VAS) and a time-trade-off (TTO) exercises. These were completed with 50 adults attending with a child to paediatric dental department in a dental hospital based in London (Cant et al., 2021). Interestingly, for the ranking and VAS exercises, participants were asked to rank/score the health states as if it was their cFPM, rather than their child's. For the TTO exercise, participants were asked for each management option, how many years (out of 10 years) with a 'perfect tooth' followed by a gap would be equivalent to the traditional treatment option (Cant et al., 2021). Despite producing health state utilities, this study had several limitations. The main limitation of this study was the failure to explain the anchoring values used for a QATY. In fact, the anchoring state values of 0 and 1 were not provided (Cant et al., 2021). Using the methods described above, it could be assumed that in estimating QATYs for these health states, a value of 1 would be for a healthy FPM and 0 is for an extracted FPM. However, the QATYs being established do not appear to be anchored by this assumed value of 0, as the lowest utility value reported was an extraction leaving an open gap at 0.3438 QATYs (Cant et al., 2021). In addition, these utility values are not generalisable to a UK population, given the small heterogenous sample, nor do they reflect children's valuations. As discussed in more depth in Chapter 2 (section 2.4.4), it is important to consider children valuations/preference in this context. Of course, the DCE in this model does not include or reflect children's valuations, as public values were needed for use in the cost-benefit model. However, a planned adolescent DCE (see section 7.5) will provide overall preferences for managing cFPM. Therefore, given the methodological limitations noted in obtaining these health state utility values

(Cant et al., 2021), any recommendations from the Sanghvi et al., (2022) model are interpreted with a degree of caution.

In contrast to these two UK models, a German microsimulation cost-effectiveness model was published in 2017. This model assessed what was the most cost-effective method of managing one to four severely affected MIH molars, from a mixed public-private-payer perspective, over a lifetime horizon. The outcome measure used was tooth retention years and three treatment strategies were compared: removal and orthodontic alignment of the second and third molars; composite restoration; indirect metal crown (initially using a preformed metal crown as a temporary, followed by non-precious metal crown at age 18). The authors from this study concluded that if the affected molar was removed at the optimal age, then the extraction/alignment strategy was always the least costly and most effective option; whereas the other strategies were more costly and less effective, thus dominated. These same conclusions were reported if more than one molar was removed, even if this extraction was not carried out at the ideal time for spontaneous closure. Whereas, if removal of only one molar was carried out, later than the optimal time for closure, then the composite restoration strategy was less costly than the other strategies but did not dominate the other strategies. Therefore, all of these options would still merit consideration if the patient presented later than the optimal time for spontaneous closure (Elhennawy, Jost-Brinkmann, et al., 2017).

Despite similar findings being noted, in that the most efficient and cost-effective method is to extract rather than restore an isolated cFPM, making direct comparisons between this model, and the model constructed in this thesis, is not sensible as several key differences exist. The German model uses costs and treatment strategies that do not necessarily reflect what happens in the UK. The German model focussed on severely affected MIH molars; whereas the model in this thesis was for a cFPM with dental caries or affected mildly-moderately by MIH (as described by the European Academy of Paediatric Dentistry index (Lygidakis et al., 2022)). The authors of the German model assumed that due to this increased MIH severity (despite not formally defining what they meant by severe), these teeth would be at an increased risk of restorative and pulpal complications. However, the probabilities of increased risk and subsequent complications were based on data for caries, and not MIH. In contrast to the model in this thesis, where the aligned second molar is assumed to remain healthy for the remainder of the individuals life, the German model reported the aligned second molar was subject to caries increment, upon alignment, at similar rate to the first permanent molar. In this regard, the German model greater reflects reality; however, the assumption that the first and second permanent molar are prone to developing caries at the same rate may not be true (Broadbent et al., 2013). As a result, this precludes a true and direct comparison between FPM teeth strategies over the horizon of the model. Despite this, including the impact on the second permanent molar is a strength of the German model, and is something that will be considered including in future iterations of this model if sufficient benefit data is available (see section 7.5).

A key distinction between the German model (Elhennawy, Jost-Brinkmann, et al., 2017), the model discussed earlier (Sanghvi et al., 2022), and the model used in this thesis is the number of teeth that were modelled. In this thesis, the model was only constructed to provide results for a mandibular cFPM. This tooth was chosen to be modelled as the restorative rates, used for the filling strategy within the model were very similar for both maxillary and mandibular cFPM (Burke & Lucarotti, 2018c; Somani et al., 2022). In contrast, there were distinct differences for the extraction strategy, as spontaneous closure was known to be much less certain within mandibular cFPMs (Eichenberger et al., 2015). In both published models (Elhennawy, Jost-Brinkmann, et al., 2017; Sanghvi et al., 2022), the outcomes were initially modelled for a single tooth and then cost and outcomes were scaled linearly up to four cFPMs, whilst the transition probabilities remained the same. As an approach, this is assuming that all cFPM in a mouth are regarded the same and any decisions are independent of one another. Given the uncertainty regarding the clinical evidence behind compensating extractions (removing an opposing tooth to prevent over-eruption) of cFPM (Cobourne et al., 2014), changing clinical practice based on these models which have modelled all four cFPM together would not be recommended, as the models had not intended to answer this question. Indeed, a mouth-level simulation that reflects the complex interaction of teeth, independent of one another, is likely to be possible but would require a level of modelling that has yet to be published. Certainly, this is something worth consideration in any future iteration of this model (see section 7.5). It can be summarised that deciding whether to restore or extract cFPM, in isolation, or at a mouth-level come with modelling alone is challenging. These three models have demonstrated these challenges, and the assumptions that have to be made to enable answers to be derived.

So far, the focus of this section has been deciding whether to restore or extract cFPM, based on the specific framing of this decision problem, across the three studies within this thesis. However, other considerations such as patient cooperation, including the need for adjunctive therapies, and who provides the care for cFPM needs to be considered. These will now be discussed in turn.

Patient Cooperation, including the need for adjunctive therapies

Options for managing cFPM are likely to be influenced by patient cooperation and/or anxiety with the use of adjunctive therapies of sedation and general anaesthesia possible although there is an absence of evidence to support superior clinical effectiveness (Ashley et al., 2015). Despite adjunctive therapies being explored during the interviews in this thesis, there was very little interest, from those adolescents interviewed about the influence it would have on their decision-making. It could be that these therapies were not considered important for managing cFPM specifically, or that these children had not experienced any of these adjunctive modalities. Alternatively, some uncertainties may have existed around the need to consider anxiety, or the need for adjunctive therapies, as the interview either used a hypothetical scenario, or based the discussion on historic prior experiences. Therefore, any potential negative consequences associated with either treatment option may have not been apparent, or anxiety provoking, and thus nullified the need to consider adjunctive therapies.

A recent UK hospital-based study reported that 20.8% (n=10) and 41.6% (n=20) of patients with MIH affected molars were managed under inhalation sedation and general anaesthetic respectively (Humphreys & Albadri, 2020). A similar UK-hospital based study reported that of the 249 children attending over a 4-month period, 201 had an extraction, of which 97.5% (n=196) had this treatment completed under general anaesthetic (AlKhalaf et al., 2022). These high rates are likely explained by the nature that those being managed in a hospital are likely to be highly anxious or have additional considerations, such as a significant medical history. Despite that, these figures are high, and concerning, particularly given the mortality associated with GA. However, the total sample size across the two studies is small, with it being likely that many cFPM extractions are being carried out in primary care, that don't involve general anaesthetic.

It appears that in hospital settings, the most common treatment choice is an extraction and adjunctive therapy used is GA. As noted in the model's findings from this thesis, the most efficient option is to extract these teeth. However, the model did not include adjunctive therapies, and only used costs and treatment strategies that were pertinent to primary care dental practices.

Therefore, the findings of this decision analytic model can't be generalised to secondary care settings. However, as no clear preference exists in the DCE, and most adolescents and adults interviewed preferred to retain these teeth, it could be suggested that hospital-based services may need to move towards a more restorative approach for cFPM, which in turn would likely reduce the number of general anaesthetics provided for cFPM. This paradigm shift would have several service provision and policy implications, which are discussed in section 7.4.4.

Who provides the care for cFPM?

As discussed in Chapter 1, most children will be managed by their own dentist, however, sub-groups of children may be referred to a dentist with additional skills and experience, most commonly a specialist in paediatric dentistry. Access to specialists, in paediatric dentistry, is limited and inequitable across England (Mills 2020). To combat these access issues, NHS England and the Office of the Chief Dental Officer approved accreditation of Tier 2 service providers in primary care. Tier 2 providers are not registered specialists but somewhere in between a qualified specialist and general dental practitioner (D'Cruz, 2018; NHS England, 2018). Unfortunately, a lack of evidence for a preference in the public DCE existed for enhanced non-specialist providers, when compared to a general dental practitioner. The implications of this are discussed in section 7.4.4.

It is understandable that the public are likely to value seeing a specialist whilst balancing against the trust/familiarity they have with their own GDP (Armfield et al., 2017). In some cases, adult participants interviewed perceived their child to be abnormal if they required a referral by their GDP to a specialist. Others felt it meant the case was more challenging and complex. This is a novel finding with no other published literature reporting these parental concerns in dentistry. It is likely other parental concerns on being referred to a specialist exist; however, these were not fully explored. Therefore, other concerns pertinent to children being referred to a specialist for a cFPM would merit further exploration.

In some cases, a child may need a specialist. It appears that non-specialist healthcare professionals tend to refer patients to a specialist because they deem them too complex (Taylor et al., 2019; Tzartzas et al., 2019). Other reasons, such as validation of a treatment plan, insufficient time and/or lack of knowledge/expertise have been reported (Tonelli et al., 2018; Taylor et al., 2019). Indeed, advice regarding treatment options for cFPM in childhood extend beyond just that which can be obtained from a paediatric dentist. An orthodontist has a significant role to play in treatment-planning for children with cFPM. A recent cross-sectional survey reported that less than half of the paediatric and orthodontic specialists agreed with a consensus panel's (made up of consultant in paediatric dentistry and orthodontics) proposed management options for cFPM affected by MIH (Alkadhimi et al., 2022). These findings suggest that even specialists in the two specialities which routinely encounter cFPM cannot agree on optimum strategies.

As a result, if referred to a specialist, then having several opinions on how best to manage complex cFPM may benefit patients and their parents. Joint paediatric/orthodontic clinics are commonplace in most dental hospitals; however, routine decisions for cFPM may not be routinely discussed on these clinics (Alkadhimi et al., 2022). Having a specific cFPM specialist multidisciplinary clinic would allow shared decisions to be made, regularly, for cFPM. However, it may not likely be efficient to run clinics just for cFPM in this way. Such a model has been shown to be effective for managing complex patients with orofacial pain (Bonathan et al., 2014), and therefore, could be given due consideration in future for cFPM.

In summary, this sub-section has discussed the considerations that have to be taken when deciding to restore or extract cFPM. However, decisions in healthcare should not be made in isolation by a healthcare professional. As this thesis has shown, deciding will depend on what perspective is being taken. Therefore, where appropriate, decisions for cFPM should be shared between all relevant stakeholders to ensure all viewpoints are included. The following sub-section will explore this in more detail.

7.2.2 Shared-Decision Making

The current findings from this thesis found that shared-decision making was an important consideration when looking to decide how to manage cFPM. The following sub-sections initially addresses adolescents' and adults' perceptions on shared decision making, including any additional considerations required to make the decision. The value placed on shared decision-making by the public will be discussed. Thereafter, how decisional uncertainties may exist in a shared decision-making model, and suggested ways that could overcome these, will be explored. This section will close with a brief discussion on the perceptions of professionals, and how they can support adolescent/adults in making shared decisions about cFPM.

Adolescents' and adults' perceptions on shared decision making

The importance of shared decision-making was a key finding, apparent in both adolescent and adult interviews, as well being a preferred method of decision-making by the public in the DCE. Shared decision-making requires a joint decision between the patient and healthcare professional(s) about what care they should receive, or in some situations not receive (NICE, 2021). However, across healthcare, the extent to which healthcare providers involve patients in decision making is reported consistently as low (Couët et al., 2015; Driever et al., 2022). It could be that professionals do not actively acknowledge, or seek patient views (Driever et al., 2022), or that parents, and/or adolescents, are unsure of a shared decision-making concept (Keij et al., 2022). In this thesis, how shared decision-making was interpreted subtly differed between adolescents and adults interviewed.

Adolescent's interview accounts suggested being included in a shared decision-making process is vital to allow them to assert their autonomy over their health, and healthcare decisions. Adults agreed that decisions regarding cFPM should not be made in isolation, and a shared decision, including their own views as well as the dentist and the child, should be commonplace. In fact, adult's interview accounts suggested for those who were parents, at present they would actively involve adolescents in these discussions. However, only one adolescent interviewed reported this to be the case. Not including adolescent views or recognising that their views need to be obtained (even if they declined to provide them), disregards the autonomy of the adolescents. Respecting adolescents' autonomy is their legal right, and they should be willing and able to express their views freely (United Nations, 1989). It has been documented that healthcare is one of the last social domains where children and young people start to express the will to make autonomous decisions (Smetana et al., 2004; Wray-Lake et al., 2010). This willingness to express and assert autonomy in healthcare gradually increases with age, ranging from age 9-20 (Wray-Lake et al., 2010). The potential negative consequences associated with healthcare interventions most likely explain why this social domain is the last to see adolescents asserting their autonomy (Smetana et al., 2004). An alternative explanation could be that limited exposure to health, and healthcare, as a child makes these environments unfamiliar, which could lead to a delay in autonomous decisions (Wray-Lake et al., 2010). Currently, there is no evidence to suggest whether these explanations stand true for adolescents in a dental context.

Despite the acknowledgement that respecting the autonomy of adolescents in oral healthcare decisions is important (Kopelman, 2001; Telford & O'Neill, 2012; Smith et al., 2018) it does not always mean that including, or seeking, their views in a decision making process is a requirement. It has been proposed that biologically, based on brain development, age 12 is the youngest age where adolescents are capable to make competent decisions about their health (Grootens-Wiegers et al., 2017). However, all children develop at different rates, and therefore their level of maturity must be taken into consideration in addition to age, when deciding whether to include their views (United Nations, 1989). To our knowledge, this is the first study that has explored autonomy of adolescents in the context of managing cFPM which fits with the findings that children's perspectives have largely been ignored in most dental research (Marshman et al., 2015). Recent dental research highlights that changes are being made in research (Gilchrist et al., 2015; Marshman & Rodd, 2015; Marshman et al., 2019) but whether this change can be translated to a clinical environment remains unknown. Further qualitative research, including an ethnographic study could help answer this uncertainty (see section 7.5 for more details).

Ultimately, respecting adolescents' and adults' autonomy should be core to clinical and research practices, both now and in the future. In addition to autonomy, other considerations will be made by adolescents and adults when making a shared decision.

Additional considerations to make a shared decision

As described, adults (whether parents or not) often make decisions regarding their own cFPM based on their internal lived experiences of prior dental treatments. Previous studies focussing on cFPM concur with these findings, suggesting that treatment decisions are heavily influenced by previous treatment experience (Azarpazhooh et al., 2013; Vernazza et al., 2015). In contrast, most adults interviewed in the thesis were highly supportive of the notion that a child (or their child) should have this tooth restored, irrespective of the adult's own treatment philosophies. A recent exploratory qualitative study obtained UK parental views of minimally invasive dentistry versus extractions under general anaesthesia for children with cFPM (Agel et al., 2021). The authors reported that extraction of cFPM under general anaesthesia was generally accepted by most parents; however, there appears to be an increasing acceptance of more minimally invasive approaches, instead of removing, cFPM (Agel et al., 2021). Different sampling approaches used between the interviews in this thesis and those interviewed in the Agel et al., (2021) study mean a narrative comparison of the findings can only be undertaken. Agel et al., (2021) interviewed parents/carer of children attending a teaching hospital in London for extraction of cFPMs under GA only; whereas, the adults (parents or not) interviewed in this thesis were recruited from a tertiary hospital and out of hour emergency services, both based in the north east of England, and were not attending as part of a consultation for managing cFPM. Given this specific inclusion criteria, the findings by Agel et al., (2021) are not surprising, as those interviewed had already accepted and agreed to an extraction as the preferred treatment option. In addition, all interviews were carried out post-treatment, which may have biased the accounts given by parents (Agel et al., 2021). In contrast, both parents, and those who were not parents, interviewed in this thesis were independent of children who had recently had a cFPM managed. Therefore, outcomes from the qualitative interviews in this thesis are unlikely to be biased by a recent clinical encounter.

Another preference noted amongst adults interviewed in this thesis was the influence their own parents had on decision making. This concept has significant impact and implications for the shared decision-making dynamic, especially if collaborative opinions are requested by a child from a parent (Miller, 2009; NICE, 2021). Parental values, and behaviours are transmitted from one generation to another, with children being shaped by processes such as observation and modelling (Maitre et al., 2021). Despite there being no literature to explore observation/modelling specifically for deciding how best to manage cFPM, parents have a responsibility to show their children how to consider options presented to them, and to make an informed choice. Similarly, parents have the responsibility to empower children to make their voices, and opinions heard. This not only makes children partly accountable for their own healthcare decisions (Colver et al., 2020), but supports the concept that adolescent decision-making competence will greatly from a multidisciplinary approach (Grootens-Wiegers et al., 2017). If, however, decisional uncertainty exists (disagreement as to whether to monitor, restore or extract a cFPM), then promoting an environment that enables positives discussion and understanding of each stakeholder's rationale, ultimately enables greater shared treatment decisions.

It appears that several considerations are likely to influence any shared decisions. However, as a concept, it was preferred, and valued, greatly by the public. The following sub-section will discuss this.

Value of shared decision-making

The public showed a clear preference for shared decision-making, compared to the dentist making the decision in isolation, as elicited using a DCE (see Chapter 5), and were willing to pay £165 to adopt this approach. Including 'decision-making' in the DCE was novel, with no previous dental DCE studies using this as an attribute (Barber et al., 2018). The public appear to value shared decision-making approaches for cFPM. This finding is reassuring as it fits with existing literature where parents support a shared-decision making approach (Miller, 2009; Légaré et al., 2018; Asa'ad, 2019) and conforms to UK guidance (NICE, 2021). It is hypothesised that adolescents would value the same approach, however, an adolescent DCE will help understand these preferences further (see section 7.3 and section 7.5). Although the concept of shared decision making is valued by the public, as a process, it is complex and may lead to decisional conflict. The following sub-sections will focus on these.

Potential for decisional conflict

In any shared decision-making model in health, understanding each stakeholder's role in a shared decision-making model is paramount (NICE, 2021). Appreciating different preferences, and examining interactions between multiple stakeholders, are necessary to recognise each other's perspective in what can be a complex dynamic.

Disputatious, or vague preferences from any stakeholder in any shared decision-making model may lead to decisional conflict (Légaré et al., 2018). Such decisional conflicts may lead to choices that are uninformed, resulting in dissatisfaction of the treatment outcome or process (Mulley et al., 2012). However, approaches such as asking for the other's opinion, or seeking information have been shown to be beneficial, by both children and their parents, in reducing these decisional conflicts (Miller, 2009). Understanding parent/family dynamics (e.g., previous parent-child conflict), child (e.g., level of maturity) and decision (e.g., urgency of the decision, if a child is suffering) factors are known to predict how successful shared decision-making process can be (Miller, 2009; NICE, 2021). It could be suggested that such changes are still lacking in a clinical dental environment; however, further research (see Section 7.5) is required.

As a result, to reduce the impact of these decisional uncertainties, additional decision-making supports such decision coaching or decision aids are worth due consideration.

Methods to support shared decision-making

Decision coaching, a non-directive approach that is delivered by healthcare providers to support, prepare, and encourage patients to actively participate in making decisions about their health (Jull et al., 2021). A recent Cochrane review identified that if decision coaching is used in isolation, there is little or no change in knowledge and/or understanding what they value better that can be used in shared-decision making process; whereas, if used in combination with evidence-based information, it may improve participants' knowledge. Unfortunately, only one of the included studies was used with parents, and none with children (Jull et al., 2021), which may reduce its generalisability to managing cFPM at present.

Decision aids support patients to make their decisions by providing information about treatment options, and associated benefits/harms of available options, when there is more than one option, and neither is clearly better. The most up to date Cochrane review (Stacey et al., 2017) reported that when compared to usual care, patients that used decision aids felt more knowledgeable, informed, and had a clearer understanding of their values, leading to a more active role in decision making. Furthermore, it was deduced that decision aids reduced the number of undecided participants, improved patient satisfaction, and had positive effects on patient-professional communication. Interestingly, no differences were observed in the improvement of participant knowledge and accurate risk perception for decision aids provided in preparation of the consultation compared to during the consultation. However, only three included studies related to oral health, none of which were focused on management within the paediatric population (Stacey et al., 2017). Since this review, two child focussed decision aids relating to oral health have been developed, and evaluated in the UK (Marshman et al., 2016; Hulin et al., 2017) but neither relate to cFPMs.

Therefore, it could be that developing a decision aid, to support patient and parents for making decisions around cFPM, is likely to be of benefit. This is more important to consider given the findings from the cost-effectiveness model, in terms of future number of visits avoided, and the lack of clear preference noted in the DCE. Irrespective of which shared-decision making support mechanism is adopted, ensuring this includes appropriately evidence-based information is likely to be important for adolescents.

Provision of information to support decision making

The provision of evidence-based, age appropriate information allows children to make competent and meaningful decisions about their own care (Mårtenson & Fägerskiöld, 2008). Adolescents are known to have the ability to demonstrate critical literacy, which is

> *"…the ability to assess the quality and relevance of information and advice to one's own situation"* (Harris et al., 2015, p.3).

However, it has been proposed that factors such as maturity, intelligence and complexity of the information provided interferes with critical literacy (Bröder et al., 2017; Muscat et al., 2020). Therefore, to help support an adolescent to decide how to manage cFPM, any information a professional provides has to be simple, evidence-based, and relevant.

It has been suggested that instead trying to understand what children and adolescents actually know about health, when asked to make a decision, focussing on how they construct meaning from health information would appear to be more beneficial (Fairbrother et al., 2016).

It is important that any information provided to adolescents, by professionals, need to consider what has been discussed previously. Adolescents interviewed suggest that professionals' opinions would more likely be trusted if evidence-based information on managing cFPM was given to them to support their ability to decide, as part of a shared decision-making approach. With regard to trust, children and adolescents have favourable experiences when trusting relationships were formed with healthcare professionals (Davison et al., 2021). This was particularly apparent for professionals who are confidential, actively provide information, and discuss any relevant concerns are more likely to be trusted (Klostermann et al., 2005). Despite no evidence on this for cFPM, the concepts are both relatable and transferrable, and something that dental care professional must consider. The perspectives of dental professionals on shared decision making are briefly discussed in the next section.

Dental Professional perspectives on shared decision making

For shared decision-making for cFPM, contextualised preferences of two out of the three main stakeholder groups (patients and parents) have been partly explored within this thesis. Professionals preferences, and interactions, in the UK have been predominately explored in observational studies (Taylor et al., 2019; Lee et al., 2021; Alkadhimi et al., 2022; AlKhalaf et al., 2022). A recent unpublished qualitative study of primary care practitioners accounts in managing MIH in the UK, led by Osborne et al., (2022) found that decision-making complexities and understanding of treatment options available for cFPM commonly exist. Another UK-based qualitative study focussing on general dental practitioners views of managing MIH reported similar findings, suggesting that managing uncertainty was the main over-arching theme (Humphreys et al., 2022). Implications for healthcare professionals, based on the findings of this thesis, are further discussed in section 7.4.2.

In summary, this section has covered the two key discussion points to come from this thesis, how best to management cFPM and the importance of shared decision making. The next section will focus on overall limitations of the methodological approaches used in thesis.

7.3 Overall limitations of methods

Despite best intentions, limitations of any chosen research methods will always be present. This section will focus on the overall limitations of methods chosen for this thesis, whereas internal validity issues relevant to each of the methods are discussed in more detail in their chapter (see Sections 4.7.3, 5.6.2 and 6.7.2).

Limitations of qualitative methods used

The qualitative interviews conducted were methodologically appropriate to establish adults' and adolescents' views and experiences around managing cFPM. At the outset of this thesis, interviews for both adolescents and adults were initially planned to inform the DCE only; however, during early qualitative data collection and analysis, it became apparent that a full qualitative study was required, prompting the change to include this in the thesis. Semi-structured interviews were chosen as they allowed flexibility to generate data, pertinent to cFPM, as it naturally evolved. Indeed, focus groups could have been used. Although trying to tease out individual responses in focus groups would have been challenging, especially as the variation in prior treatment experience may have dictated the flow of the conversation. Alternative qualitative methods could have been used to capture children's experiences (Fargas-Malet et al., 2010). Visual methods, such as drawings (Mitchell et al., 2011) and use of photographs (Darbyshire et al., 2005) have been documented as positive methods, that are fun and relaxing, to help trigger and discuss experiences. Video diaries (Buchwald et al., 2009) and participatory research (Fargas-Malet et al., 2010) can be used in isolation, or to supplement more conventional methods, as they are able to elicit data that may not be otherwise obtained by directly asking an adolescent (Fargas-Malet et al., 2010).

Another significant limitation of qualitative interviews is to consider the age range of those adolescents sampled. The age range of adolescents sampled in the interviews was 12-16 years old. This was deliberately chosen as it was the similar age range intended for the adolescent DCE. Including views and opinions from children younger than 12 would have been highly relevant given the key time to decide whether to extract or restore cFPM is often around 9-9.5 years old (Cobourne et al., 2014; Ashley & Noar, 2019). Therefore, if younger children were to be included, either changing the qualitative method from semi-structured interviews to focus groups (Adler et al., 2019), or utilising those methods described above (Darbyshire et al., 2005; Buchwald et al., 2009; Fargas-Malet et al., 2010; Mitchell et al., 2011) would likely be required to elucidate opinions from children aged 6-16.

Ethnography, an alternative qualitative approach, could have elicited an understanding of participants experiences, perspectives, and everyday practices (Barbour, 2014). This could have given a more in-depth insight into the ‘actual’ decision making of cFPM, rather than these being in the abstract or hypothetical, as was the case in the interviews. As an extension of this ethnography, observing clinicians would be beneficial.

Regarding professional’s opinion, a key limitation of the qualitative component of this thesis was that it only focussed on patients and the public: thus, not including the views of the profession. Understanding clinician’s viewpoints and their role in shared decision-making, via observation, as future pieces of research are merited and discussed further in section 7.5.

Limitations in eliciting preferences

Considerable debate exists about whose preferences to obtain in an economic evaluation involving children, but it usually is dictated by the perspective and decision-making context of the underlying evaluation (Brazier et al., 2005). In this thesis, obtaining public values, and WTP, were important to ascertain as managing cFPM is a societal issue as the relevant options in childhood in the UK are paid by the NHS, indirectly via taxation of society. Therefore, undertaking a cost-benefit model is relevant for policy, especially given the uncertainties surrounding the optimum treatment choice and provider of care.

Despite eliciting public preferences for use in the cost-benefit framework model, a proposed adolescent DCE was not completed due to recruitment issues and slow regulatory approvals as a result of COVID-19. Recruitment is currently ongoing; however, adolescent preferences have not been reported in this thesis. Therefore, it reduces how 'adolescent' viewpoints, and valuations, of managing cFPM fit into the wider context. Currently, only interview accounts can convey what adolescents value. As discussed earlier, it is important to consider and understand adolescent preferences in a shared decision-making dynamic (NICE, 2021).
It is likely that obtaining adolescents preferences, in a DCE, from those with cFPM may make them better placed to value their own health condition compared to a general adolescent trying to imagine them (Brazier et al. 2005). Values, and preferences, are likely to be based on current experience and previous personal experiences (Cubi-Molla et al., 2018). However, contrary to this, it could be that adopting this approach and only including those with cFPM may be heavily influenced by their previous experiences, which varies currently within the UK (Taylor et al., 2019; AlKhalaf et al., 2022). It could be argued that to fully explore all 'adolescent' opinions, and improve generalisability, a representative sample of a UK adolescent population should be invited to participate in a DCE.

Assessing adolescents' or members of the public's values in isolation does not permit elicitation of valuations that may come from a parent-child dyad interaction. In a recent DCE, on managing hypodontia, approximately half the dyads (parents and children) selected the same alternative when completing the choice tasks individually and together. Indeed, observations of this process revealed that if either adolescent or parent had a strong preference for a specific attribute, they were keen to promote this attribute; however, if deliberation was required, then preferences were not routinely changed (Barber, 2019).

An alternative to elicit WTP would have been to conduct a CVM. CVM are often less complex to complete and can elicit an exact WTP, or a range of WTP values, for particular health states (Klose, 1999). Having both could permit a distribution around the exact WTP, that could be used in a model, assuming what was being valued in the CVM was being modelled. It could be argued that stated WTP from CVM is a limitation of this approach as it is likely a poor indicator of actual WTP, with the concern being that do the respondents have the ability to pay, rather than what they say they will (Ryan & Watson, 2009). Additionally, DCEs are argued as being more desirable as some individuals may find it difficult, or indeed refuse, to place a monetary value on human health when using CVM (Ryan & Watson, 2009). The DCE, to an extent overcome this, however issues with the DCE, as described in more detail in Chapter 2, do exist. Furthermore, given the themes that were being generated, from the qualitative interviews used to inform the DCE, it became apparent that understanding overall preferences was just as important as establishing WTP, given the question this thesis was looking to address.

Limitations of using a decision model

An alternative method of answering whether to decide to extract or restore a cFPM could have been to conduct a clinical trial and incorporate an economic evaluation. This trial could have simply randomised patients to either have an extraction, or restoration, and follow them up over a time period of 3-5 years. Various primary outcome measure could have been used, such as episodes of pain, number of re-visits. In addition to accurately collecting cost data, measures such as the CARIES-QC-U (Rogers et al., 2020) could have been collected to obtain utility values, that could have been mapped to the EQ-5D-Y tariffs to obtain QALYs. Utilising a trial based economic evaluation permits individual patient data to accurately estimate cost effectiveness (Petrou & Gray, 2011a).

However, fundamentally a clinical trial is not the best method to establish how to decide whether to restore or extract a cFPM. In addition to the challenges faced in choosing the most appropriate primary outcome measure, the main limitations of a trial for cFPM are the truncated horizon and ignoring alternative options. As shown the results of this model, extraction at any age between seven and ten was the most efficient option, followed by definitively restoring the tooth. However, when the time horizon was altered to only include costs and benefits accrued up to the age of 18, it demonstrated that the efficient option of extraction was not as efficient as it was with the lifetime horizon, thus suggesting basing clinical or policy decisions on a single trial with limited follow-up would not reflect reality.

Alternative methodologies, such as a prospective longitudinal cohort studies (Hong et al., 2020) would be sensible to accurately follow-up early childhood decisions for cFPM. However, these methods take time and are extremely expensive. In addition, due to the protracted nature of data collection, in some cases every 5-10 years (Broadbent et al., 2013), there is a risk that adverse significant events, such as toothache, or multiple fill replacement, may be forgotten by participants or not picked up during the examination. Therefore, it appears that using a model may be the most predictable way to understand the long-term outcomes of extraction versus restoration of cFPM in children. Despite that, final sub-section will briefly review the limitations in using of DCEs (and WTP) in decision models.

Limitations of using DCEs (and WTP) in decision models

The decision analytical model has helped understand the long-term implications of initial decisions to manage cFPM in children. The completion of a CEA and CBA undertaken in thesis provided results from various perspectives. A decision can be made on resource allocations by comparing the CBA results with other CBAs across different healthcare settings and/or sectors of the economy. However, in England, most economic evaluations report QALYs, (NICE, 2022a) so the results from this thesis are not easily directly comparable to other health conditions. Instead, this thesis could have followed convention, and arranged to collect EQ-5D values. This would have permitted direct comparison across various health conditions and supported future policy decisions. However, it is acknowledged that the EQ-5D is not sensitive enough to elicit utility changes in dental health. Furthermore, latent periods exist in a dental condition such as cFPM where minimal changes can be noted over a long period of time, thus QALYs are likely to be very similar for all options.

As an alternative, this thesis could have utilised either of the two caries-specific utility measures that currently exist, CARIES-QC-U (Rogers et al., 2020) and Dental Caries utility index (Hettiarachchi et al., 2020). Utilising CARIES-QC-U (Rogers et al., 2020), as this was developed and validated for use in the UK, would allow a CUA to be completed, which may be of more relevance, compared to a

CBA, in terms of UK policy. However, this would not permit QALYs to be derived for cFPM affected by MIH. Furthermore, given the concerns about the current cost-effectiveness threshold estimates used by NICE (Claxton et al., 2015), and that it has to be assumed that for analysis is that all QALYs are of equal social value, irrespective of who accrues them, adopting a CBA, incorporating WTP values derived by a UK public DCE, could be of more relevance.

This section has discussed the overall limitations of the methodological approaches used in thesis. Despite these perceived limitations, the findings from this thesis have important implications for all relevant stakeholders. The following section, and sub-sections, will consider each relevant stakeholder in turn.

7.4 Implications

7.4.1 Adolescents/Parents

Ultimately, adolescents want to be involved in decisions made regarding management of dental issues, such as cFPM. Parents, and indeed members of the wider society, interviewed in this thesis acknowledge these adolescent concerns. It is widely accepted that parents impact the engagement of adolescents in making decisions (Fairbrother et al., 2016; Guassi Moreira et al., 2018). Therefore, substantial efforts should be made, by parents (and professionals) to ensure these views are not just heard but 'actively' included in any decision-making process

To combat these concerns, parents (or adults in the wider sense) need to create an environment that empowers adolescents to vocalise their thoughts and encourage them to make their own decisions. Adolescents who make decisions that result in positive outcomes are known to be satisfied, as they have made the decision for themselves instead of it being made by their parents (Wogden et al., 2019; Davison et al., 2021). Therefore, whether adolescents choose to restore (the apparent preferred option) or extract a cFPM, then either decision should be constituted as a 'good' decision by their parents. Ultimately, this makes adolescents accountable for their own healthcare decisions; a key transitional process for adolescents moving into adulthood (Colver et al., 2020).

However, expecting adults to transition from the monolithic paternalistic approach, to see adolescents as competent decision-makers, and include them in decision-making, will require significant support, at an individual level (Gilchrist et al., 2013; Carney et al., 2021), or require a larger societal attitudinal change.

Supported training does not have to be formalised; it could be the role of healthcare professionals. This support could be as simple as highlighting to parents that adolescents develop their general decision-making skills primarily through observation of how their parents make decisions (Davids et al., 2016). Therefore, suggesting adolescents directly observe the oral healthcare decisions of their parents, or actively encouraging parent-adolescent discussions surrounding oral healthcare decisions, at home, are strategies that could be used to support these individuals.

7.4.2 Dental care professionals

This thesis has focussed on cFPM, however, dental care professionals must ensure a holistic approach to oral health is maintained to reduce the overall dental disease burden to individuals, as for dental caries, evidence of disease activity is a risk factor for future disease (SDCEP, 2018; Public Health England, 2021) and for MIH, other teeth are likely to be affected (Lygidakis et al., 2022).

Dental professionals are active stakeholders in the shared decision-making dynamic (NICE, 2021). The outcome of the model, DCE and qualitative interviews, stresses the importance of dental care professionals considering all available options for managing cFPM. Therefore, conveying these options and appropriate risk and benefits for each to adolescents and parents is important for dental care professionals to consider. This permits the adolescents and/or parents to decide, based on clinical information whilst allowing them to incorporate their own values.

Supplemental information may be requested by general dental professionals to support optimal decision-making for cFPM. Therefore, general dental professionals should be encouraged to make a referral to a specialist for advice, or indeed management, if required. If orthodontic concerns are noted (Cobourne et al., 2014; Ashley & Noar, 2019), then obtaining an orthodontic opinion is appropriate to support a fully informed shared decision. Similarly, if specialist paediatric dental opinion, or management, is required for managing cFPM, then dental professionals should be encouraged to obtain this. It is key that bi-directional communication between professionals must be timely. Given the current climate of access to NHS dentistry, and the lasting effects of the COVID-19 pandemic, earlier recognition and referral should be made to ensure optimal care. In addition, dental professionals have a responsibility to alleviate any anxieties that may be raised when specialist input is required, especially if there is concern of a child being 'abnormal'.

Provision of information and trust in the profession are evidently important to patients. Therefore, professionals have a responsibility to build trusting relationships with both adolescents and adults, as favourable experiences are noted when trusting relationships were formed with healthcare professionals (Davison et al., 2021). Developing and maintaining good verbal and non-verbal communication skills such as being confidential, actively providing information, being honest and discussing relevant concerns when asked to do so instil trust (Klostermann et al., 2005; Smith et al., 2018). In terms of provision of information, it is accepted that children have the right to receive information, should they request it (Smith et al., 2018). Dental professionals therefore have to ensure they provide information that is pertinent, salient and age appropriate (Fairbrother et al., 2016). Consideration of the use of a chairside decision-aid, co-designed with children and their parents to include outcomes of the DCE and model, could benefit this (Stacey et al., 2017). Finally, being honest in the dental setting is important if professionals are to be trusted by patients, in particular adolescents (Welly et al., 2012; Smith et al., 2018). Therefore, when dental care professionals help support decision-making about cFPM, it is important that they are honest with information they provide relating to procedural complexities or uncertain short- and long-term outcomes, to enable a fully informed decision.

Therefore, professionals need to obtain all relevant and pertinent information to provide adolescents (and/or parents) or adults to allow them to make a decision that is based on their clinical presentations and incorporates their own values (Asa'ad, 2019; NICE, 2021).

7.4.3 Wider healthcare professionals

The option of active monitoring in the model was dominated by all strategies. If not managed, these individuals could attend elsewhere with problems from their cFPM. In the UK, patients (adolescent and adults) may consult their general medical practitioner (Cope et al., 2018; Currie et al., 2022) or attend the emergency department (Currie et al., 2017; Parten et al., 2019; Heggie et al., 2022) for dental issues, such as a cFPM. It has been suggested patients (adults) with a dental problems may attend their GP, instead of a dentist, due to having altered perceptions of the scope of practice of non-dental healthcare professionals, previous negative experiences of dental care, being unable to pay for dental care as well difficulty accessing dental services (Cope et al., 2018).

Therefore, wider healthcare professionals should be educated to appropriately signpost patients to dental professionals who present with dental issues, including those related to cFPM.

7.4.4 Policy and service provision

Modelling studies support policy decision makers in the allocation of scare healthcare resources. Although not adopting a true societal perspective, using a cost-benefit framework as the base-case scenario and scenario analyses three to seven permit some discussion regarding future policy and commissioning of services in primary care. As shown by the base-case, the initial strategy of extraction is the most efficient option and is incrementally more net-beneficial than filling over a 100-year time horizon. However, it could be argued that changes to policy should include valuations and preferences rather than just results from modelling studies (Vernazza et al., 2021). Thus, the lack of clear public preferences could make the argument that all options of restoring and extracting should be offered, irrespective of their efficiencies.

In England, CUAs can help policymakers support allocative decisions on which healthcare technologies based on the NHS fixed budget (NICE, 2022a), although it is worth noting that NICE has not typically become involved with allocative decisions in dentistry (NICE, 2022b). The economic evaluations undertaken in this thesis were CEA and CBA, therefore direct comparison to healthcare interventions evaluated using the method described above, is not permissible. This would be an issue if dental services were competing for resources within the same fixed NHS budget as other healthcare services; however, dental services are given their own fixed budget, which has remained at approximately £3.7billion per year (or 3.5% of the total NHS budget) (NHS Commissioning Board, 2013). Understanding how dental resources are best allocated, and commissioned, has been debated (Holmes et al., 2009; Vernazza et al., 2019). As of 1st July 2022, integrated care boards were established across England, on a statutory basis, to commission health services (NHS England, 2022c). However, for all dental services (including primary and secondary care, specialist, community and out of hours) the primary care commissioning team is responsible for commissioning under the banner of NHS England (NHS England, 2022d). A recent qualitative study with NHS England commissioners, preceding these commissioning changes, suggested that they recognised the need for resource allocation but underpinning a priority setting approach had to be implicit clinical advice and needs assessment (Vernazza et al., 2019). As a result, it is important that commissioners are made of aware of the findings of this thesis, in particular the model, as this forms part of the clinical advice that they need to support commissioning services pertinent to child oral health.

Currently, the standard dental contract for primary care dental services stipulates how dentists should use the resource that are available (NHS England, 2022d). However, contract reforms remains a national debate and is likely to be evolving incremental process (Harris & Foskett-Tharby, 2022). Dental remuneration, under the current dental contract in England, is likely to impact the way cFPM are managed.

Previously, simple fillings, extractions and molar endodontics all attracted three UDAs; however, the recent outcome of 2022/23 Dental Contract Negotiations may have some impact on service provision. Instead of three UDAs, a patient that requires filling or extraction of three or more teeth in a course of treatment would now accrue five UDAs. Therefore, if a child has more than one cFPM affected, then a dentist might see this as more financially feasible than before. Similarly, there may be an increase in molar endodontics as this treatment now attracts seven UDAS (rather than three) (NHS England, 2022e). Considering the implementation of these changes on the model would likely mean costs that fall on the NHS will increase for any of the endodontic procedures (conventional and re-root canal treatment), as to date, no proposed plans have been made to increase patient's charges to reflect the increase in UDAs available for these treatments. Thus, if the benefits remain the same, the costs of initially going down the filling arm would be higher, and therefore making the incremental costs wider, which could ultimately make filling less efficient or a dominated strategy. However, adolescent preference for retention, based on the interview accounts in this thesis, was displaced by removal as and when the cFPM became symptomatic. This could mean the uptake of molar endodontics in the adolescent population remains low, in spite of increased remuneration (NHS England, 2022e).

Access to NHS dental services has been identified as a key outcome for the new dental contract (NHS England, 2022a). However, access in England remains a real challenge for many patients (British Dental Association, 2022), exacerbated by COVID-19 (Watt, 2020). Anecdotally, the number of referrals made by primary care practitioners for children with cFPM may increase. This will be compounded by the results of thesis as they have left uncertainties around decision making for cFPM (Taylor et al., 2019). For some, the clinical protocol to support decisions may be enough (Ashley & Noar, 2019); however, there was still a large proportion who referred to both paediatric and orthodontic specialties (Taylor et al., 2019). Although, it is worth noting that further evolution of the contract could take these findings into account in not perversely incentivise one option over another.

The implications of the public not preferring a dentist with enhanced skills could be harmful, in terms of managing cFPM, as tier-2 providers have an important role to play, especially as access to specialists, and consultants in paediatric dentistry, is limited and inequitable across England (Mills 2020). It is hypothesised that a lack of knowledge, and appreciation, by the public regarding the role of an enhanced practioners explains this lack of preference. In a well-developed MCN, most children with cFPM can be managed by a tier-2 provider. In addition, the MCN set-up allows a formal network that includes a consultant-led service, as the hub of the MCN, so in theory, transition of more complex cases or those that require services that only specialists are able to provide e.g., general anaesthesia (Skipper, 2010; NHS England, 2018). if Tier-2 providers are going to support the management of patients with cFPM in the future, then education of the public should be encouraged to ensure policy changes are engrained into societal values and beliefs.

Ultimately clinical service provision and policy changes for managing cFPM should not just focus on clinical effectiveness, or cost savings, but should incorporate the preferences of those who are users of the service, or indeed indirectly, through taxation, contribute to it (Vernazza et al., 2021). Thus, any future research into managing cFPM, from an economic perspective, should bear this in mind.

7.4.5 Researchers – oral health and wider healthcare

It is clear that researchers must continue to actively recruit adolescents as participants in oral health research, as children are experts of their own lives (Marshman et al., 2015).

To date, there have been few qualitative studies focussing on cFPM. To our knowledge, the studies in this thesis are the first qualitative interview studies that explored adolescents' and adult views and experiences around managing cFPM.

It appears that DCEs have great potential as a method for many different oral health contexts and/or populations (adolescents, adults, and professionals). To date, there are small numbers of contingent valuations (Tan et al., 2017) and DCEs (Barber et al., 2018) in dentistry; however, there is a real paucity that are conducted with children as participants. Eliciting preferences using techniques such as DCEs are theoretically plausible in children and adolescents (Stevens, 2015; Rowen et al., 2020) and ongoing research (See Section 7.5) will permit adolescents valuations of managing cFPM, using a DCE, to be explored.

Using a decision analytical model to explore cFPM in this thesis has overcome the lack of information that would be obtained, should a clinical trial have been used instead. Models have a significant role to play in oral health research, as expected costs, and benefits, of dental decisions (such as restoration versus extraction) often extend beyond a typical trial-follow up period. In addition, as shown in this thesis, they can reflect reality greater by incorporating clinical nuances and multiple comparators, that a trial is unlikely to capture (Briggs et al., 2006; Drummond et al., 2015). However, dentistry is complex and the several nuances of decision making for cFPM, and for other conditions, are likely to make this area of oral health economics evaluations a real challenge.

This section has discussed the implications, and relevance, of the findings for each stakeholder group. The final section will briefly discuss what future research can be done to expand the findings of this thesis in addition to overcoming some of the limitations raised.

7.5 Future Research

This thesis has used a combination of qualitative and economic methods to understand the management of cFPM. A number of questions have arisen as a result of this thesis. This section will briefly describe further areas of research.

The decision analytical model has several assumptions, however, as a model it has reflected what currently happens with cFPM in the UK in primary care. Making these assumptions was a trade-off against increasing the computational burden of the model. However, the model complexity can be increased in future iterations. Expanding this model for secondary care services, or adjuncts such as sedation and general anaesthesia would be beneficial. The current structure could be expanded to include additional health events, such as denture, implant rather than just having a resin retained bridge. Complexity could be added to model, by including survival data within each parameter, or including events such as orthodontic relapse, or partial space closure. Additionally, decision trees could be embedded within health states, such including various options choosing different restorative materials, or reflect varying clinical presentations of severity. The model could be extended to reflect a mouth, extending to four molars independent of one another. This is likely to be computationally rather challenging, without making significant assumptions, but it is something worth exploring. Finally, it is evident from the model that further research is warranted. Uncertainties could be improved upon by having additional information. Undertaking a VOI analysis would be sensible and help identify data/information gaps exposed by the model. To obtain this information, further valuation studies and/or quantitative studies would help to provide these estimates. In addition, future work could aim to integrate uncertainty on those parameters that have relied on clinical expert opinion. For example, surveying a range of experts and creating a distribution around their views to better capture clinical heterogeneity or variability in practice within the PSA. Furthermore, the WTP valuations obtained from the DCE were only pertinent to dental caries, and therefore precluded a CBA for MIH. Future studies which ask the public to value health state for an MIH affected tooth would provide specific WTP values for these teeth. In addition, asking the public to value developmental vs acquired dental conditions health states would be worthwhile.

Interviewing adolescents and adults regarding cFPM raised several interesting points that could be extended beyond the focus of this thesis. The role of the specialist appeared to raise some uncertainties for those interviewed, and using further qualitative methods, understanding the role specialists have in paediatric and adult dental care pathways, or possibly exploring adolescents/publics' view of being referred to various different specialities. Anecdotally, the fear of abnormality discussed in this thesis could be limited to paediatric dentistry, a lesser-known speciality to the public, when compared to others, say orthodontics, which has a greater mainstream presence, and therefore understanding.

An alternative qualitative study would be to undertake an ethnography and observe behaviours within the shared decision-making dynamic of managing cFPM, or in general oral health. There are no ethnographic studies which have explored shared decision-making in a cFPM context, or wider in the paediatric dental context. However, examples in health have demonstrated that the method is feasible with children deciding for themselves (Daley, 2014), parents deciding for their children (Heinze & Nolan, 2012) and as a shared decision-making framework (Manhas et al., 2020). An extension of this ethnographic study could include formal analysis of taped consultations, using the validated OPTION (Observing Patient Involvement in Decision Making) instrument, which detects to what extent patients are involved in the decision-making process (Elwyn et al., 2003)

Another area worth exploring is addressing the diagnostic thresholds of adolescents, parents, and professionals in managing cFPM, to address at what point the tooth is perceived to be 'unrestorable'. This could be done using quantitively focussed methods, using sequential clinical vignettes or pictures of 2D-3D models of various levels of an affected tooth. Alternatively, a more novel method could be to use drawing as a research tool (Mitchell et al., 2011). As a method, drawings are often used to depict states of mind, often being used in psychology, or as a supportive method of describing qualitative interview data (Mitchell et al., 2011; Søndergaard & Reventlow, 2019). To elicit diagnostic thresholds, participants could be asked to draw what level they would deem to be 'unrestorable' and work out the diagnostic threshold from this.

Despite the most efficient option being extraction, a definitive restoration is still efficient as the benefits outweigh the costs. In addition, the lack of a clear preference in the DCE, and the clear preference for retention by adolescents interviewed, means a definitive answer of what option is best to choose for patients, and their parents is yet to be fully established. The most prominent step to build on this is the development of a decision aid to support shared decision-making between all relevant stakeholders (Stacey & Volk, 2021). Currently, a well-designed flowchart, depicting a clinical protocol, has been designed to support professionals whether to restore or extract cFPM (Ashley & Noar, 2019) Utilising co-design methods (Greenhalgh et al., 2016; Langley et al., 2018), and including the clinical protocol already published (Ashley & Noar, 2019), a decision aid could be developed prior to be piloted for use in routine clinical practice (Stacey & Volk, 2021). Beyond this, focussing on implementation science methods would be beneficial to support regular use (Clarkson et al., 2010; Bauer & Kirchner, 2020).

Finally, completion and analysis of the adolescent DCE, disrupted by COVID-19, will provide an understanding of adolescent's value when managing cFPM. These values will be an important starting point for including young people's views in the development of a decision aid. In addition, this will provide an opportunity to crudely observe any differences, by comparing odds ratios, of preferences between the two different populations who have completed identical DCEs (less the cost attribute for adolescents). Furthermore, a future DCE could be conducted on managing a child with a hypomineralised cFPM. There was some difference of opinions of adolescents interviewed about whether a developmental condition, against an acquired condition, was an influencing factor in deciding how to manage cFPM. This DCE would not only provide an understanding of preferences, but if done with members of the public and including a cost attribute, would allow WTP values to be established and used in a future cost-benefit analysis.

7.6 Conclusion

This chapter has summarised the key discussion points of this thesis and the implications of these findings in the context of patients, professionals, service provision, policymakers, and researchers. The overall limitations of the methodologies used have been discussed, before, discussing how future research may overcome some of these limitations, and further expand the evidence based around the management of cFPM.

In conclusion, the following provides a conclusion for each of the aims of the three complimentary studies within this thesis:

In Chapter 4, the aim of the qualitative study was to establish adolescents' and adults' views and experiences around managing cFPM. It can be concluded that several factors influence how adolescents' and adults decide to manage cFPM, for themselves, and in the case of adults, for children. Overall, the need to make shared decisions, and to actively involve adolescents as part of this process, regarding cFPM was abundantly clear.

In Chapter 5, the aim was to design the DCE, to elicit the public's preferences for each pathway including determining societal willingness to pay (WTP). It can be concluded that the UK public do not prefer to restore or extract cFPM in a child, providing the resultant space was closed spontaneously or orthodontically. In addition, the public prefer that any decisions made are done so by the parent alone (with the provision of information), or in conjunction with the dentist (as part of a shared decision-making process).

In Chapter 6, the aim was to develop a decision analytical model to determine the incremental net-benefit of initial options to manage cFPM over a modelled life-time horizon. It appears from the base-case scenario, and scenario analyses, the most efficient approach is to extract cFPM between the age of seven and ten. Definitively restoring is an efficient option but is incrementally less cost beneficial than extraction. Active monitoring and placing a temporary filling, prior to extracting at the ideal stage to maximise spontaneous closure, are dominated strategies, and should not be offered based on the CBA model findings alone.

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Appendices

Appendix A – COREQ checklist

COREQ (COnsolidated criteria for REporting Qualitative research) Checklist

A checklist of items that should be included in reports of qualitative research. You must report the page number in your manuscript where you consider each of the items listed in this checklist. If you have not included this information, either revise your manuscript accordingly before submitting or note N/A.

Topic	Item No.	Guide Questions/Description	Reported on Page No.
Domain 1: Research team and reflexivity			
Personal characteristics			
Interviewer/facilitator	1	Which author/s conducted the interview or focus group?	123
Credentials	2	What were the researcher's credentials? E.g. PhD, MD	121
Occupation	3	What was their occupation at the time of the study?	121
Gender	4	Was the researcher male or female?	121
Experience and training	5	What experience or training did the researcher have?	123
Relationship with participants			
Relationship established	6	Was a relationship established prior to study commencement?	122-123
Participant knowledge of the interviewer	7	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research	121
Interviewer characteristics	8	What characteristics were reported about the inter viewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	121
Domain 2: Study design			
Theoretical framework			
Methodological orientation and Theory	9	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	114-115
Participant selection			
Sampling	10	How were participants selected? e.g. purposive, convenience, consecutive, snowball	122
Method of approach	11	How were participants approached? e.g. face-to-face, telephone, mail, email	122-123
Sample size	12	How many participants were in the study?	128
Non-participation	13	How many people refused to participate or dropped out? Reasons?	N/A
Setting			
Setting of data collection	14	Where was the data collected? e.g. home, clinic, workplace	123
Presence of non-participants	15	Was anyone else present besides the participants and researchers?	123
Description of sample	16	What are the important characteristics of the sample? e.g. demographic data, date	128-129
Data collection			
Interview guide	17	Were questions, prompts, guides provided by the authors? Was it pilot tested?	123-124
Repeat interviews	18	Were repeat inter views carried out? If yes, how many?	N/A
Audio/visual recording	19	Did the research use audio or visual recording to collect the data?	126
Field notes	20	Were field notes made during and/or after the inter view or focus group?	126
Duration	21	What was the duration of the inter views or focus group?	128
Data saturation	22	Was data saturation discussed?	127
Transcripts returned	23	Were transcripts returned to participants for comment and/or	No

Topic	Item No.	Guide Questions/Description	Reported on Page No.
		correction?	
Domain 3: analysis and findings			
Data analysis			
Number of data coders	24	How many data coders coded the data?	127
Description of the coding tree	25	Did authors provide a description of the coding tree?	N/A
Derivation of themes	26	Were themes identified in advance or derived from the data?	129 & 139
Software	27	What software, if applicable, was used to manage the data?	126
Participant checking	28	Did participants provide feedback on the findings?	N/A
Reporting			
Quotations presented	29	Were participant quotations presented to illustrate the themes/findings? Was each quotation identified? e.g. participant number	129-148
Data and findings consistent	30	Was there consistency between the data presented and the findings?	129-148
Clarity of major themes	31	Were major themes clearly presented in the findings?	129 & 139
Clarity of minor themes	32	Is there a description of diverse cases or discussion of minor themes?	129 & 139

Developed from: Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*. 2007. Volume 19, Number 6: pp. 349 – 357

Once you have completed this checklist, please save a copy and upload it as part of your submission. DO NOT include this checklist as part of the main manuscript document. It must be uploaded as a separate file.

Appendix B – HRA and NHS Rec Approval

Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

NHS
Health Research Authority

Dr Greig Taylor
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25 November 2020

Dear Dr Taylor

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title:	**DECIDE study: Dental ExtraCtions versus fillings for adult teeth In children: a cost bEnefit/effectiveness analysis**
IRAS project ID:	**288198**
REC reference:	**20/NS/0124**
Sponsor	**Newcastle Univerity**

I am pleased to confirm that **HRA and Health and Care Research Wales (HCRW) Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the "Information to support study set up" section towards the end of this letter.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?
HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.

Appendix C – HRA and HCRW approval for non-substantial amendment

IRAS Project ID 288198. HRA and HCRW Approval for the Amendment

Reply | Reply All | Forward | ...

amendments@hra.nhs.uk <noreply@harp.org.uk>
To Chris Vernazza; Greig Taylor; Kay Howes

Thu 08/04/2021 11:42

You forwarded this message on 14/02/2022 16:28.
If there are problems with how this message is displayed, click here to view it in a web browser.

External sender. Take care when opening links or attachments. Do not provide your login details.

Dear Dr Taylor,

IRAS Project ID:	288198
Short Study Title:	DECIDE
Amendment No./Sponsor Ref:	201819 No25 Taylor Amendment 1
Amendment Date:	01 April 2021
Amendment Type:	Non Substantial Non-CTIMP

I am pleased to confirm **HRA and HCRW Approval** for the above referenced amendment.

You should implement this amendment at NHS organisations in England and Wales, in line with the guidance in the amendment tool.

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

Appendix D – Participant Information Leaflet for qualitative interviews (Adults)

The Newcastle upon Tyne Hospitals NHS Foundation Trust

DECIDE

Newcastle University

Participant Information Sheet

DECIDE: Dental ExtraCtions versus fillings for adult teeth In children: a cost bEnefit/effectiveness analysis

We invite you to take part in a research study

- Before you decide to take part in this study, it is important for you to understand why the research is being done and what it will involve
- Please take time to read this information sheet carefully as it will tell you the purpose of this study and what will happen if you take part.
- You are free to decide whether or not to take part in this study. If you choose not to take part in this study, it will not affect your regular care.
- If there is anything that is not clear from this information sheet or you would like more information, then please just ask a member of the research team

Important things you need to know

- We want to find out what aspects of treatment pathways for the management of damaged adult teeth in children, that are important to members of the general public.
- We will also be asking children aged 12 and over similar questions
- You can stop taking part in the study at any time and this will not affect the care you receive

How to contact us

If you have any questions, please contact the research team:

Mr Greig D Taylor
decide@newcastle.ac.uk

Full Title: Dental ExtraCtions versus fillings for adult teeth In children: a cost bEnefit/effectiveness analysis
Short Title: DECIDE
IRAS: *288198*
Researchers: Mr Greig D Taylor (PhD Student) &
Dr Chris Vernazza (PhD Supervisor)
Location: School of Dental Sciences, Newcastle University
Protocol Version: 1.4

Summary of the study

Why are we doing the study?

The most common adult tooth to cause children problems is the first molar (back) tooth. These are damaged by tooth decay and/or because they did not form properly. If the first molar is damaged it is more likely to further decay and unlikely to last a child's lifetime. A recent survey of UK dentists found some filled and some removed these teeth. Therefore, we do not know which is better for children, their families or the general public. We need to research whether these teeth should be: a) filled, with fillings needing replacements over time; or b) removed to allow the other teeth possibly to fill the space left behind.

What will the study involve?

- This study is looking at working out the best treatment pathway for children with damaged first molar teeth.
- We are looking to find out what parts of each treatment option is preferred by looking at the responses and how much they are valued.
- Participants in the study will be asked to take part in one face-to-face interview.
- Interviews will be undertaken using a virtual platform e.g. zoom.
- Each interview will be recorded.
- The interview will last between 30 and 60 minutes and can be done at a time that is convenient to you
- We can't pay you, however, a £20 Amazon gift voucher is available as a thank you for your time.

- Participants who take part in the interview will be invited to attend two workshops at the end of the study.
- You do not have to take part in the workshops if you do not want to.
- Workshops will include adolescents, members of the public, healthcare professionals and NHS managers to discuss:
  - a) if the project that has been undertaken reflects reality and
  - b) how best to use the results to change the way we fund and deliver treatments in the NHS
- Each workshop will take approximately 2-3 hours. Refreshments will be available.
- We can't pay you, however, a £20 Amazon gift voucher is available as a thank you for

What information will be collected?

- These audio recordings will be sent, securely, to a company who will type them up. This will allow the research team to understand, and check what everyone has said in these recordings
- Once this has been done, the audio recordings will be deleted
- Your identity will remain anonymous. This will be done by changing my name and disguising any details of my interview which may reveal my identity or the identity of people I speak about.

What are possible risk and benefits of taking part?

If you agree to take part in this study, there are no risks. The questions asked will be about your opinion on what you believe are the most important aspects on the management of damaged adult teeth in children. None of the questions will be sensitive or cause any distress.

If you agree to take part in this study, you are **unlikely to benefit directly**. However, you will be **adding to the scientific knowledge** and help us understand how to change the way these children are treated to benefit them, their family and the NHS.

Do I have to take part? Can I withdraw from the study?

Taking part in the study is voluntary and entirely up to you. You will not be forced to take part in the study.

We will describe the study and provide you with enough information for you to decide. If you agree to take part in the study, we will then ask you to sign a consent form.

If at any point during the study you decide to withdraw, you are free to do so without giving a reason for doing so. By withdrawing from the study, it will not affect the

What will happen to the results of the study?

I will write a report of the study results and publish this in a medical journal. I may also show these results at conferences I attend. Disguised extracts from my interview may be quoted. An example of how this will be disguised would be "46-year-old, female, has had a filling"

Any information presented will remain **anonymous**

A summary of the findings can be sent to you if you wish.

Who is organising and funding the study?

The lead researcher is Mr Greig D Taylor will be in charge of the day to day running of the study.

The study is being funded by an NIHR Doctoral Research Fellowship (NIHR300251).

How will we use information about you?

We will need to use information from you for this research project. This information will include your name and contact details. People will use this information to check your records to make sure that the research is being done properly.

People who do not need to know who you are will not be able to see your name or contact details. Your data will have a code number instead. We will keep all information about you safe and secure.

Once we have finished the study, we will keep some of the data so we can check the results. We will write our reports in a way that no-one can work out that you took part in the study.

What are your choices about how your information is used?

You can stop being part of the study at any time, without giving a reason, but we will keep information about you that we already have.

We need to manage your records in specific ways for the research to be reliable. This means that we won't be able to let you see or change the data we hold about you.

Where can you find out more about how your information is used?

You can find out more about how we use your information

- at https://www.hra.nhs.uk/planning-and-improving-research/policies-standards-legislation/dat a-protection-and-information-governance/gdpr-guidance/templates/template-wording-for-generic-information-document/
- by asking one of the research team
- by sending an email to decide@newcasle.ac.uk

Who has reviewed the study?

All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests.

This study has been reviewed by the **North of Scotland Research Ethics Committee** and given favorable opinion on 12th October 2020.

What do I do if I have a problem?

Any complaint you have about the way this study has been carried out or the way you have been dealt with in this study will be addressed.

If you have any concerns about this study, then please contact the researchers (Mr Greig D Taylor or Dr Chris Vernazza) who will be able to answer any questions. They can be contacted on decide@newcastle.ac.uk

If you remain unhappy and wish to complain formally, then you can do this via the NHS Complaints Procedure (https://nhsnss.org/contact-us/)

Appendix E – Participant Information Leaflet for qualitative interviews (Adolescents)

The Newcastle upon Tyne Hospitals NHS
NHS Foundation Trust

DECIDE

Newcastle University

Participant Information Sheet

DECIDE
EXTRACTION VS FILLING FOR ADULT TEETH IN CHILDREN

Dental ExtraCtions versus fillings for adult teeth In children: a cost bEnefit/effectiveness analysis

We would like to invite you to take part in a research study. It is important you know why this research is being done before you agree to take part. Please read this information sheet before you agree to take part. If anything is not clear, or you want to ask a question, then please ask one of the research team.

	Mr. Greig D Taylor *Study Lead*	Email address: DECIDE@newcastle.ac.uk Greig.taylor@newcastle.ac.uk Phone 0191-208-8188

DECIDE STUDY Information

Full Title: Dental ExtraCtions versus fillings for adult teeth In children: a cost bEnefit/effectiveness analysis
Short Title: DECIDE
IRAS: *288198*
Researchers: Mr Greig D Taylor (PhD Student) &
Dr Chris Vernazza (PhD Supervisor)
Location: School of Dental Sciences, Newcastle University
Protocol Version: 1.0

Pge **1** of **4**

Participant Information Sheet (young people). Version 1.4 (21st October 2020)
Study Title: DECIDE
IRAS: 288198

Why are we doing the study?

- The most common adult tooth to cause children problems is the first molar (back) tooth
- These can be damaged by tooth decay and/or because they did not grow properly
- Some dentists fill these damaged teeth, and some take them out
- We do not know which is the best option for children, their families or the general public.
- We need to research whether these teeth should be: a) filled, with fillings needing replacements over time; or b) removed to allow the other teeth possibly to fill the space left behind.
- As well as what parts of each treatment is preferred by the general public and adolescents.

Why would have you been asked to take part?

- You are a young person aged over 12 years old
- You can help us by taking part in an online survey to find out what parts of each treatment option (fill or remove) are preferred by young people whether or not you have had problems or treatment for your first molar teeth

What is the benefit of taking part?

- You will help us understand how to change the way how young people are treated to benefit them, their family and the NHS

What is the risk of taking part?

- There are no risks of taking part as the questions we ask will not be upsetting or personal.

What will you need to do if you want to take part?

- You would need to take part in an interview with a researcher called Greig.
- Several other young people will also be doing interviews with Greig.
- This will be done online using a programme called zoom. You can have the video on, or just the sound
- It will take up to 45 minutes
- The interview will be recorded. This will allow Greig and his research team to understand what has been said in more detail. Once Greig has done this, the recording will be deleted.
- You can do the interview yourself, or have a parent or guardian sit beside you. The questions need to be answered mainly by you as it is your views that is required.
- You will be given a £20 Amazon voucher for taking part

What information will be recorded?

- The recorded interviews will be sent, securely, to a company who will type them up.
- Once this has been done the recording will be deleted.
- When the final results are reported, something you said may be quoted. It will be impossible to know who said each quote as your identity will be disguised. An example of how this will be disguised would be "13-year-old, female, has had a filling"

Want to take part in further research?

- You may be invited to attend two separate workshops later in the study because you have taken part in the interview
- You do not have to take part in the workshop.
- Each workshop will include young people, members of the public, dentists and healthcare managers.
- One workshop will discuss the overall project findings and the other on how best to change the way dentistry is delivered for this problem
- Each workshop will last 2-3 hours. Refreshments will be available.
- You will be given a £20 Amazon voucher for taking part

Do I have to take part?

- Taking part is completely voluntary and you will not be forced to take part.
- You can decide to not take part at any point before, during or after the interview. This will not affect your dental care

- To help make sure this doesn't happen, we will describe the study and provide you with enough information for you to decide. If you agree to take part in the study, we will then ask you to sign a consent form. A parent or guardian will also be asked to sign a consent form.

How will we use information about you?

- We will need to use information from you for this research project. This information will include your name and contact details. People will use this information to check your records to make sure that the research is being done properly.

- People who do not need to know who you are will not be able to see your name or contact details. Your data will have a code number instead. We will keep all information about you safe and secure.

- Once we have finished the study, we will keep some of the data so we can check the results. We will write our reports in a way that no-one can work out that you took part in the study.

What are your choices about how your information is used?

- You can stop being part of the study at any time, without giving a reason, but we will keep information about you that we already have.
- We need to manage your records in specific ways for the research to be reliable. This means that we won't be able to let you see or change the data we hold about you.

What are your choices about how your information is used?

- You can find out more about how we use your information
    - at https://www.hra.nhs.uk/planning-and-improving-research/policies-standards-legislation/dat a-protection-and-information-governance/gdpr-guidance/templates/template-wording-for-generic-information-document/
    - by asking one of the research team
    - by sending an email to decide@newcasle.ac.uk

Who is organising the study?

- The lead researcher, Greig, will be in charge of the managing the study.
- The study is being funded by an NIHR Doctoral Research Fellowship (NIHR300251)
- This study has been reviewed **North of Scotland Research Ethics Committee** and given favorable opinion on (date to be entered).

What do I do if I have a problem?

- If you have a problem by the way the study has been run, or that you haven't been treated properly, then please let us know and we can see what has gone wrong.
- You, and/or your parents, can contact the researchers (Mr Greig D Taylor or Dr Chris Vernazza) who will be able to answer any questions. They can be contacted on decide@newcastle.ac.uk or 0191-208-8188
- If you remain unhappy and wish to complain formally, then you can do this via the NHS Complaints Procedure (https://nhsnss.org/contact-us/)

Appendix F – Initial version of the Topic Guide

DECIDE Study

WP2 - Qualitative Interviews

Adult/Adolescent Topic Guide

This interview will be recorded. Anything you say will be anonymised. Can you re-confirm you have read the Participant Information Sheet and consent to this interview?

START RECORDING

'Broad Areas for Questions'

- **Attendance**
  - How often do you attend the dentist?
  - If yes, how long have you had your current dentist for? What are your reasons for attending this dentist?
  - What would prompt you to visit your dentist?
  - Do you feel attending the dentist is important? If so why? What about your partner, or children's attendance?
  - Have you received dental treatment anywhere else, other than your normal dentist? If so, where? Why were you managed there?

- **Experiences**
  - What type of dental treatments have you required? (prompt with examples if need be)
  - When you've needed treatment in the past, do you feel an explanation about what treatment is needed and why it is being done help with the overall experience?
  - Tell me about a time when you've had to have treatment done?
    - What can you remember about it? How was it done? Did it take long? Was it painful? Did you feel emotional? How long until you felt like you 'normal'? is it what you expected to happen? (If not, why not?)

- **Expectations**
  - What do you expect to obtain in attending your dentist?
  - What do you expect your dentist to provide you?
  - Is this different for your children, or if you have none, for a child patient?

'Focused Areas for questions'

- **Identifying the 1st Permanent Molar**
    - Are you aware that humans have two sets of teeth? Are you aware of different types of teeth in the mouth? Can you name any?

 (Share a picture of a mouth with all teeth present – number or colours to help identify on zoom)

    - Could you identify where the molars are? Could you identify which is the 1st permanent molar?

I think at this point it would be good to show a picture of a compromised first permanent molar and explain the focus of the questions is now on this one tooth only.

- **Experiences with these teeth**
    - Have you ever had any problems with this particular tooth? If so, what problems have you had? (prompt if needed – pain, can't chew, can't sleep, etc)
    - Has your dentist ever mentioned if this tooth had a dental disease with it? Can you remember why they thought that? How did this make you feel, knowing you had a problem with this tooth? Were you worried about this tooth causing you problems now, and in the future?
    - Did you require treatment for that tooth? Can you explain what you have had done? Do you still have this tooth?

- **Experiences with these teeth – for those who have had treatment**
    - How did it feel having a filling and/or extraction of this tooth?
    - Was having a filling and/or extraction sore? Was it painful after the filling and/or extraction was completed?
    - Did you have this filling and/or extraction with an injection only? How was this? Why did you choose to have it done this way? Would you have preferred other options like happy air, or being put to sleep to have this done?
    - What made you want to have a filling/extraction done over the tooth taken out/filled in the first instance?
    - What helped you make that decision?
        - Did the type or colour of the filling material to be used influenced your choice?
        - Did you feel losing that tooth would make activities like eating harder?
        - How do you feel about having a gap? Does having a gap matter? If you haven't, would you want the gap to be closed? Why is this the case?
        - Did the dentist help with the decision? Was it important that your dentist did this treatment? Would you have preferred to be seen by a specialist? Why is this the case?
    - How much of a priority, to you, was keeping that tooth? If you had it filled, and when this tooth becomes sore, would you still want to keep it? At what point would you want to remove it?
    - Was it important that your dentist did this treatment? Would you have preferred to be seen by a specialist? Why is this the case?

- **Experiences with these teeth – for those who didn't require treatment**
    - How would you feel if you had to have a filling and/or extraction of this tooth?
    - Do you think it would be sore having the treatment done? What about afterwards?
    - Would you want to have a filling and/or extraction done with an injection only? Why is this? Would you have preferred other options like happy air, or being put to sleep to have this done?
    - Would you prefer to have this tooth filled or removed?
    - What factors helped you make that decision?
        - Would the type or colour of the filling material to be used influenced your choice?
        - Would you feel losing that tooth would make activities like eating harder?
        - Would you not want to have a gap? Does having a gap matter? Would you want the gap closed? How much would it matter if a fake tooth was needed to go instead?
    - How much of a priority, to you, would be keeping that tooth? What about your parents? If you had it filled, and when this tooth becomes sore, would you still want to keep it? At what point would you want to remove it?
    - Would it be important that your dentist did this treatment? Would you have preferred to be seen by a specialist? Why is this the case?

Is there anything else you would like to discuss, or feel that we haven't covered?

Appendix G – Final version of the Topic Guide

DECIDE Study
WP2 - Qualitative Interviews
Topic Guide

This interview will be recorded. Anything you say will be anonymised. Can you re-confirm you have read the Participant Information Sheet and consent to this interview?

START RECORDING

SECTION 1

Attendance

- Frequency of attendance?
- Reasons for attendance?
  - (Prompt: pain, values oral health, trust dentist's opinion)
- Attendance important for you? Family member? Child?
- Attendance other than dentist for dental care?
  - (Prompt: Why, where?)

Experiences

- Previous dental experience
  - (Prompt: types of treatments; Positive/negative experiences)
- Important to have proposed treatment fully explained for overall experience
- Tell me about a time when you've had to have treatment done?
  - (Prompt: What happened? Length? Painful? Emotional? Relief? Short- and long-term effects?)

- **Expectations**
- What do you expect to obtain in attending your dentist?
- What do you expect your dentist to provide you?
  - (Prompt: A child, or you child?)

PAGE 1

DECIDE
EXTRACTION VS FILLING FOR ADULT TEETH IN CHILDREN

Newcastle University

SECTION 2

Identifying the 1st Permanent Molar

- Are you aware that humans have two sets of teeth?

(Share a picture of a mouth with all teeth present – number or colours to help identify on zoom)

- Which colour represents the molar teeth?
- Which letter is the **first permanent molar**?

- Can you recall if you have ever required treatment on your **first permanent molar**?
    - (Prompt : If definitive yes, continue with experiences; if no, go to haven't had treatment)

Experiences with these teeth

- Problems with this **first permanent molar**?
    - (Prompt: pain, can't chew, can't sleep, etc)

- Dental disease on this **first permanent molar**?
    - (Prompt: Dentist ever mentioned specific disease? How did you feel knowing disease on this tooth? Worried? Future problems?)

- Treatment for this **first permanent molar**?
    - (Prompt: what treatment? Explain what happened? How did it feel? More than one episode of care? Still have the tooth?)

PAGE 2

THOSE WHO HAVE HAD TREATMENT

- Have you heard of treatments fillings and extractions? Do you know what they mean? What does mean to me?

- How did it feel having a filling and/or extraction of this **first permanent molar**?
    - (Prompt: painful during? After? Sensations? Worries?)
    - Are fillings "normalised"?

- Why that treatment over the alternative (e.g filling over extraction) for **first permanent molar**?
    - (Prompt: avoidance of pain? Previous experiences? Peer influence? Type or colour of the filling material? Impact on eating/chewing? Costs? Long term maintenance needs? Size of defect
    - **(Additional adolescent prompts: parental influence? Dental influence)**

- How did you decide to fill or extract for the **first permanent molar**?
    - (Prompt: help of dentist? Who did treatment help decide? Important for you to have your dentist do treatment? Seeking specialist care differ? Severity of defect? Expectation that treatment will work help decide?)
    - **(Additional adolescent prompts: help of parent?)**

- How much of a priority, to you, was keeping the **first permanent molar**?
    - (Prompt: If became sore after treatment, would you still want to keep it? What would change your mind? Would having a gap matter at the back? Different if at the front? Leave gap, or place false tooth – why?)
    - **(Additional adolescent prompts: felt it was important to your parent to keep tooth?)**

- Decision change between acquired and developmental conditions for compromised **first permanent molar**?

PAGE 3

THOSE WHO HAVEN'T HAD TREATMENT

(Imagine you visit dentist at they tell you that you have a dental problem with this first molar tooth, and you need something done)

- Have you heard of treatments fillings and extractions? Do you know what they mean?

- How would you feel if you had to have a filling of this **first permanent molar**? Extraction of this **first permanent molar**?
    - (Prompt: painful during? After? Sensations? Worries? Different if extraction?)

- Happy with Injection only?
    - (Prompt: why? Prefer Alternatives offered if treatment were to be done again – sedation, GA)

- What factors would help make that decision to either have your **first permanent molar** filled or removed?
    - (Prompt: Avoidance of pain? Severity/size of the defect? Previous experiences on other teeth? Peer influence? Dentist opinions? Type or colour of the filling material? Impact on eating/chewing? Costs? Long term maintenance needs? Clear information? Expectation that treatment will work help decide)
    - **(Additional adolescent prompts: parent help? Would parents influence your decision? Dentist vs. adult opinion? Have you been involved in decision making?)**

- Importance of your dentist doing treatment.
    - (Prompt: what about seeing a specialist?)

- How much of a priority, to you, was keeping that **first permanent molar**?
    - (Prompt: If became sore after filling, would you still want to keep it? What would change your mind to extraction? Would having a gap matter at the back? Different if at the front? Leave gap, or place false tooth – why?)
    - **(Additional adolescent prompts: felt it was important to your parent to keep tooth? Parental experiences of false teeth? Parental influence on when to lose tooth?)**

- Decision change between acquired and developmental conditions for compromised **first permanent molar**?

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Section 3

'Hypothetical child' (FOR ADULTS ONLY)

- Do you have any children?
- If so, what how many and what are their ages?

- How would you feel if your child had to have a filling of their **first permanent molar**?
  - (Prompt: painful during? After? Sensations? Worries? Different if extraction?)

- Would you be happy for your child to have an injection only?
  - (Prompt: why? Prefer Alternatives offered if treatment were to be done again – sedation, GA)

- What factors would help make that decision to either have your child's **first permanent molar** filled or removed?
  - (Prompt: How would your child feel? Blame parents? Social norms - how would their friends think of them? Severity of defect? Avoidance of pain? Previous experiences on other teeth? Peer influence? Dentist opinions? Type or colour of the filling material? Impact on eating/chewing? Costs? Long term maintenance needs?

- How important to you is it that your dentist does the treatment for your child's **first permanent molar?**
  - (Prompt: what about seeing a specialist?)

- How much of a priority is keeping your child's **first permanent molar**?
  - (Prompt: If became sore after filling, would you still want to them to keep it? Would mind them having a gap at the back? Different if at the front? Would you want them to have this gap left, filled or closed with braces?– why?)
  -
- Involvement of children in decision making? When you were a child, were you involve in decision making?

- **Is there anything else you would like to discuss, or feel that we haven't covered?**

- **End of Recording**

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Appendix H Completed DCE Checklist

A checklist for conjoint analysis applications in health care. *(Reproduced from: Bridges, J.F.P., Hauber, A.B., Marshall, D., Lloyd, A., Prosser, L.A., Regier, D.A., Johnson, F.R. & Mauskopf, J. (2011) 'Conjoint Analysis Applications in Health—a Checklist: A Report of the ISPOR Good Research Practices for Conjoint Analysis Task Force', Value in Health, 14(4), pp. 403–413.)*	
Was a well-defined research question stated and is conjoint analysis an appropriate method for answering it	
1.1 Were a well-defined research question and a testable hypothesis articulated?	Yes
1.2 Was the study perspective described, and was the study placed in a particular decision-making or policy context?	Yes
1.3 What is the rationale for using conjoint analysis to answer the research question?	Yes
Was the choice of attributes and levels supported by evidence?	
2.1 Was attribute identification supported by evidence (literature reviews, focus groups, or other scientific methods)?	Yes
2.2 Was attribute selection justified and consistent with theory?	Yes
2.3 Was level selection for each attribute justified by the evidence and consistent with the study perspective and hypothesis?	Yes
Was the construction of tasks appropriate?	
3.1 Was the number of attributes in each conjoint task justified (that is, full or partial profile)?	Yes
3.2 Was the number of profiles in each conjoint task justified?	Yes
3.3 Was (should) an opt-out or a status-quo alternative (be) included?	Yes
Was the choice of experimental design justified and evaluated	
4.1 Was the choice of experimental design justified? Were alternative experimental designs considered?	Yes
4.2 Were the properties of the experimental design evaluated?	Yes
4.3 Was the number of conjoint tasks included in the data-collection instrument appropriate?	Yes
Were preferences elicited appropriately, given the research question	
5.1 Was there sufficient motivation and explanation of conjoint tasks?	Yes
5.2 Was an appropriate elicitation format (that is, rating, ranking, or choice) used? Did (should) the elicitation format allow for indifference?	Yes
5.3 In addition to preference elicitation, did the conjoint tasks include other qualifying questions (for example, strength of preference, confidence in response, and other methods)?	Yes
Was the data collection instrument designed appropriately	
6.1 Was appropriate respondent information collected (such as sociodemographic, attitudinal, health history or status, and treatment experience)?	Yes
6.2 Were the attributes and levels defined, and was any contextual information provided?	Yes
6.3 Was the level of burden of the data-collection instrument appropriate? Were respondents encouraged and motivated?	Yes
Was the data-collection plan appropriate	
7.1 Was the sampling strategy justified (for example, sample size, stratification, and recruitment)?	Yes
7.2 Was the mode of administration justified and appropriate (for example, face-to-face, pen-and-paper, web-based)?	Yes
7.3 Were ethical considerations addressed (for example, recruitment, information and/or consent, compensation)?	Yes
Were statistical analyses and model estimations appropriate	
8.1 Were respondent characteristics examined and tested?	Yes
8.2 Was the quality of the responses examined (for example, rationality, validity, reliability)?	Yes
8.3 Was model estimation conducted appropriately? Were issues of clustering and subgroups handled appropriately?	Yes
Were the results and conclusions valid	
9.1 Did study results reflect testable hypotheses and account for statistical uncertainty?	Yes

9.2 Were study conclusions supported by the evidence and compared with existing findings in the literature?	Yes
9.3 Were study limitations and generalizability adequately discussed?	Yes
Was the study presentation clear, concise, and complete	
10.1 Was study importance and research context adequately motivated?	Yes
10.2 Were the study data-collection instrument and methods described?	Yes
10.3 Were the study implications clearly stated and understandable to a wide audience?	Yes

Appendix I Feedback on DCE from initial meeting with Expert Panel

Considerations of attributes for the DCE		
Summarised Feedback	**Decision**	**Rationale for decision**
Geographical locations – availability of specialists & waiting times	Consider including in DCE if qual worked supported	This could be a key attribute of the adolescent or public DCE. Preferences of local gdp vs. specialist, can link this into the CBA.
Preparedness of the treatment options, and how the treatment is likely to affect each child.	Consider Include in adolescent DCE, if supported by pre-DCE qualitative work	Unlikely to be useful for CBA model; but could be included in an adolescent DCE if qual work highlighted this.
The impact on the child/family short and longer term and involvement in the decision-making process are currently not reflected in the model.	Include in DCE if supported by pre-DCE qualitative work	If supported by Qual work, and the impact on the parent/family can be valued (in terms of WTP) then we can link to these decisions made in the model. However, if not, then impacts could be included in an adolescent DCE if supported by qual work
Is there anything around parental compliance?	Do not include	We need to assume that parents will comply with the treatment options suggested by the model. View the model as the ideal scenario. Therefore, we have to say, as a limitation to our model, parents and patients may pick and chose when to come back to different therapies at different points. Likely different methodologies needed to elicit reasons behind non-compliance with treatment options.
Patients vs parent decision of treatment options	Include in adult/adolescent DCE, if supported by pre-DCE qualitative work	This is an important aspect to consider, however, could be challenging to operatiionalise in the model; however, still likely to be a useful attribute in the public DCE, and an adolescent DCE
I would worry as a parent how much pain my child was in	Consider inclusion in DCE.	More important for preference elicitation, but could consider pain as a characteristic to include in the model? Seem to be recurring;? Or assume pain equal across all treatments?

Appendix J – Ngene coding

```
Design
;alts = alt1, alt2, alt3
;block = 2
;rows = 18
;eff = (mnl, d)
;model:
U(alt1) = b0 + b1.dummy[0|0|0|0|0] * TREATMENT [1,2,3,4,5,6] + b2.dummy[0|0] *
PROVIDER [1,2,3] + b3.dummy[0|0] * DECISION [1,2,3] + b4.dummy[0|0] * AVOID
[1,2,3] + b5.dummy[0|0] * COST [1,2,3]  /
U(alt2) = b0 + b1.dummy[0|0|0|0|0] * TREATMENT [1,2,3,4,5,6] + b2.dummy[0|0] *
PROVIDER [1,2,3] + b3.dummy[0|0] * DECISION [1,2,3] + b4.dummy[0|0] * AVOID
[1,2,3] + b5.dummy[0|0] * COST [1,2,3]  $
```

Appendix K– Experimental choice sets chosen for use in DCE

Choice situation	alt1.treatment	alt1.provider	alt1.decision	alt1.avoid	alt1.cost	alt2.treatment	alt2.provider	alt2.decision	alt2.avoid	alt2.cost	Block
1	6	3	2	3	3	2	2	1	2	1	1
2	3	2	3	2	3	2	3	2	1	1	2
3	6	2	1	3	1	5	3	3	2	3	2
4	6	3	3	2	1	1	1	2	3	2	2
5	3	2	2	3	3	1	1	1	2	2	2
6	4	2	2	2	1	6	1	3	1	3	1
7	4	3	1	2	3	5	2	3	3	1	1
8	4	1	1	1	3	3	3	2	2	1	1
9	5	2	1	1	2	6	1	2	2	3	2
10	2	1	1	3	3	4	3	3	1	2	2
11	1	3	1	1	1	4	2	2	3	2	2
12	3	1	3	1	2	5	3	1	3	3	2
13	1	2	3	2	2	3	1	1	1	1	2
14	5	1	2	2	2	2	2	3	1	3	1
15	5	1	2	1	1	3	3	1	3	2	1
16	1	3	3	3	1	6	2	1	2	2	1
17	2	3	2	1	2	4	1	3	3	1	1
18	2	1	3	3	2	1	2	2	1	3	1

Appendix L – Online DCE (as disseminated to the participants)

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Our next set of questions are about your teeth and how your dental care is provided, which are being asked on behalf of Newcastle University.

It is really important that your experiences are represented – as a thank you for your extra time, we will now send you a **£10 voucher** when you complete the questionnaire. Remember to check we have your correct home address at the end of the survey.

Please note that these questions will include images of tooth decay and other dental issues.

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Do you have any of your own teeth left in your mouth?

- Yes
- No

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NatCen Social Research

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Have you ever had fixed orthodontic treatment (metal braces attached to your teeth)?

- Yes
- No

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Do you have any children?

Please include adult children and children you do not currently live with.

- Yes
- No

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NatCen Social Research

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Which, if any, of the following types of dentist have you ever received dental treatment from?

Please select all that apply.

- ☐ Dentist with enhanced skills
- ☐ Specialist dentist
- ☐ General Dental Practitioner
- ☐ None – I have never received dental treatment

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Which of the following best represents how your dental care is provided?

- ◯ Private: You pay the full costs of treatment
- ◯ Paid-for NHS dental care: You pay the NHS ‘band’ charges
- ◯ Free NHS dental care: You do not pay and are exempt from NHS charges
- ◯ NHS and private care (Mixed of some NHS banded and some private treatments)
- ◯ Insurance-based: You pay a fee which covers all treatment except laboratory bills
- ◯ Insurance-based: You pay a fee which gives discounts on any NHS or private treatment received

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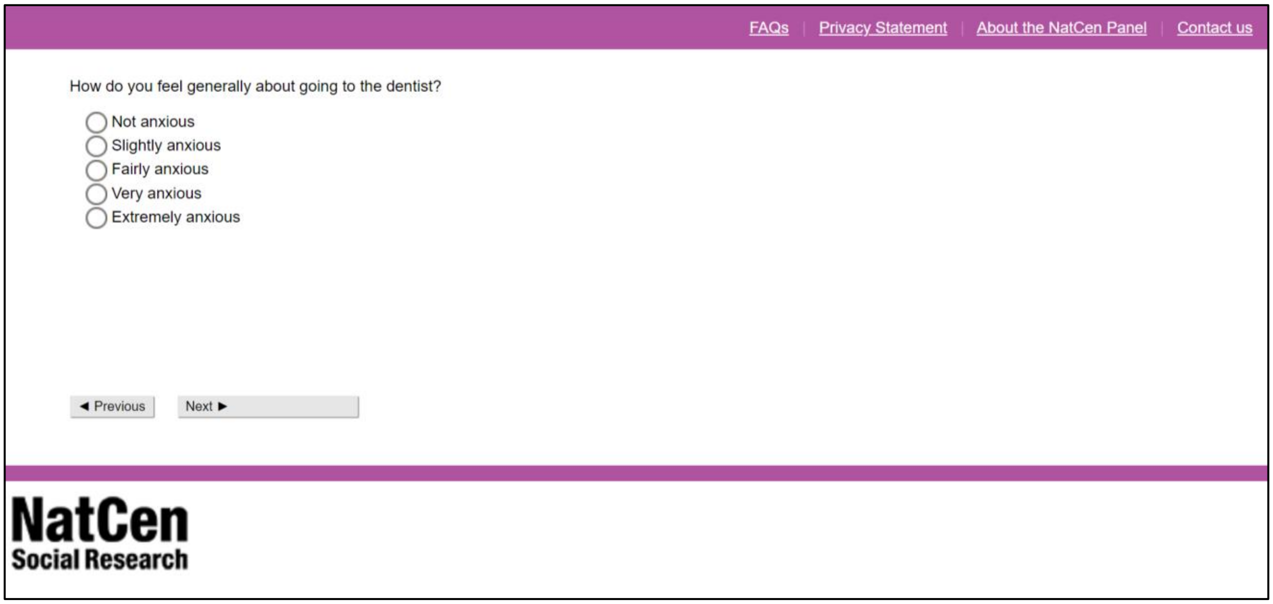
NatCen Social Research



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How do you feel generally about going to the dentist?

- Not anxious
- Slightly anxious
- Fairly anxious
- Very anxious
- Extremely anxious

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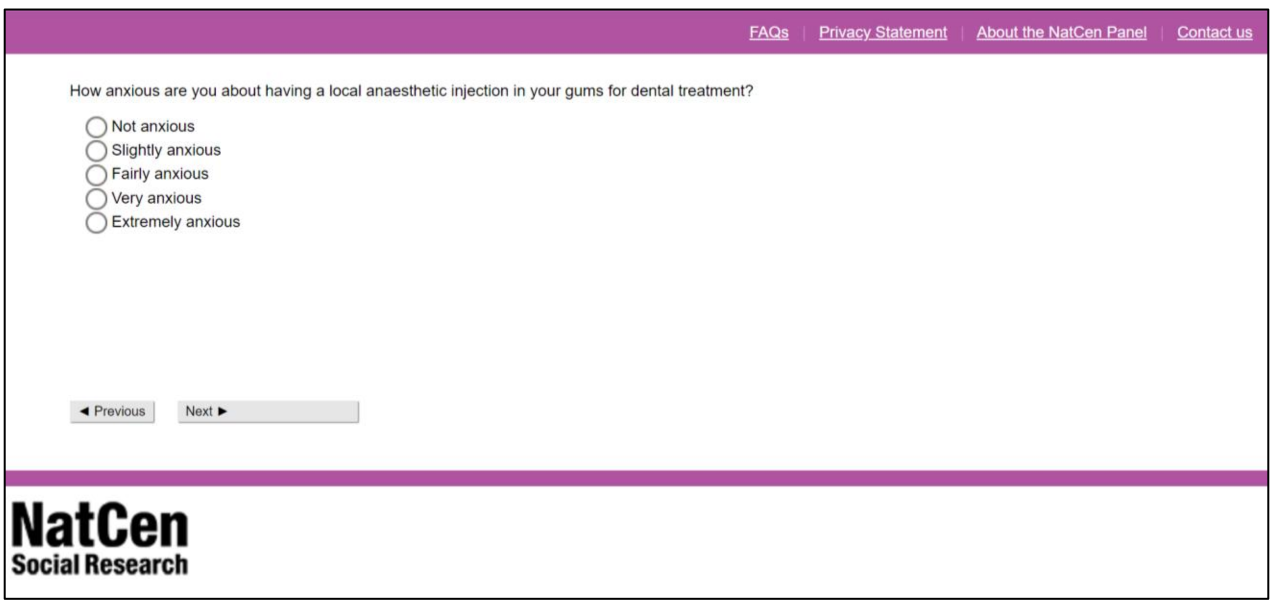
NatCen Social Research



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How anxious are you about having a local anaesthetic injection in your gums for dental treatment?

- Not anxious
- Slightly anxious
- Fairly anxious
- Very anxious
- Extremely anxious

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NatCen Social Research

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How anxious are you about having a filling?

- Not anxious
- Slightly anxious
- Fairly anxious
- Very anxious
- Extremely anxious

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How anxious are you about having a tooth taken out?

- Not anxious
- Slightly anxious
- Fairly anxious
- Very anxious
- Extremely anxious

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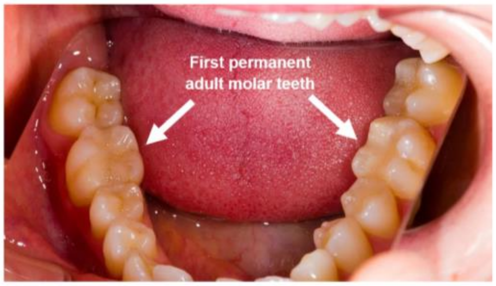
The next set of questions are about any of your first permanent adult molar teeth (indicated in the picture below).



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Have you ever had a filling (metal or white filling) carried out on any of your first permanent adult molar teeth?

- Yes
- No

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Have you ever had an extraction (removal of tooth) carried out on any of your first permanent adult molar teeth?

◯ Yes
◯ No

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How important is it for you to keep your first permanent adult molar teeth?

◯ Important
◯ Neither important nor unimportant
◯ Unimportant

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Often adult teeth can be damaged due to dental decay and/or they do not develop properly. Once the damage leads to a hole, these teeth are 'compromised'. In children, the most common adult tooth to be compromised is the first permanent adult molar

Our next set of questions are about different options for treating compromised permanent adult molars in a child who is aged 9. You will be provided with some information about the scenario, before being asked to choose the treatment you would pick.

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Imagine you have a 9-year-old child who has a compromised first permanent adult molar, as shown in the picture below. The tooth is not causing your child pain but does cause occasional sensitivity.

For all of the treatment options, your child would receive local anaesthetic to numb the mouth – so, please assume there is no pain associated with any of the procedures.

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There are different dental procedures available. Each of the scenarios you will be shown will include **five different aspects** of the treatment for you to compare: tooth treatment type, dental care provider, decision maker, number of future treatments avoided and the overall costs.

Scroll down to read the details.

Please read all of the information provided carefully before progressing with the questionnaire.

Treatment

This indicates how the tooth will be treated:

1. The tooth is filled (no gap is left)

The filling will last on average 7 years. A filling will need to be repaired or replaced in the future. Each time the filling is repaired/replaced the hole will get bigger.

2. The tooth is removed and the tooth behind it does not move forward on its own (a gap is left)

After the tooth is removed, a gap will be left. The tooth behind the gap will erupt into its normal position, in 3 years' time. The full gap will not be closed.

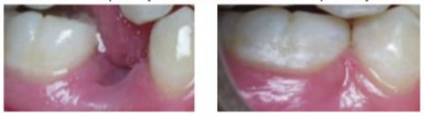
3. The tooth is removed and the tooth behind it partially moves forward on its own (a small gap is left)

After the tooth is removed, a gap will be left. The tooth behind will erupt and move forward on its own. The space will be partially closed. This will take up to 5 years.

4. The tooth is removed and the tooth behind it completely moves forward on its own (no gap is left)

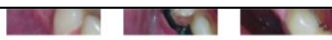
4. The tooth is removed and the tooth behind it completely moves forward on its own (no gap is left)

After the tooth is removed, a gap will be left. The tooth behind will erupt and move forward on its own. The space will be completely closed. This will take up to 5 years.



5. The tooth is removed and the tooth behind is completely moved forward using metal braces (no gap is left)

After the tooth is removed, a gap will be left. The space will be closed using metal braces fixed to the teeth. This takes approximately 2 years.



6. The tooth is removed and the gap is closed with a false tooth (no gap is left)

After the tooth is removed, a gap will be left. The gap will be closed with a dental bridge that is stuck to the teeth behind and in front. This will be completed when the child is 14 years old. There will be a gap for 5 years. The bridge will need to be maintained and replaced in the future.

Provider of dental care

This indicates who provides the dental care:

1. General Dental Practitioner

- Able to treat **most** children

Provider of dental care

This indicates who provides the dental care:

1. General Dental Practitioner • Able to treat **most** children • If the treatment is too complex or a patient is unable to cope with treatment, a referral to a dentist with enhanced skills, or a specialist will be made • Studied 5 years at university but have limited training in treating children
2. Dentist with enhanced skills in Paediatric Dentistry • Able to treat **most** children • If the treatment is too complex or a patient is unable to cope with treatment, a referral to a specialist will be made • Studied 5 years at university but have attended some additional short courses in treating children
3. Specialist in Paediatric Dentistry • Able to treat **any** children no matter how complex or whether the patient can cope with treatment • Studied 5 years at university and a further 3 years of additional training in treating children

Decision maker

This indicates who makes the overall decision for a child's care.

Number of future treatments avoided

This indicates how many **future 'treatment' visits** will be avoided over the lifetime of the patient. These do not include check-up visits. Each future visit will require some form of treatment.

Cost

This indicates the one-off, out-of-pocket cost you would have pay for the chosen scenario.

In England, there are no costs to the parent for the management of children's teeth. In many other countries, parents have to pay for their child's dental treatment. Please assume you must pay, out of your pocket, for the option you choose.

Please consider if you would be able and willing to pay the cost for each option you choose. Remember, if you spend money on this scenario, you may have less money available to spend on other things such as a meal in a restaurant or a trip to the cinema.

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Rather than choosing one of the two treatment options shown to you, you will also be able to choose not to have any treatment done. This would mean:

- The decay will get worse
- The tooth is likely to break and become painful and/or infected which may cause a swelling or an abscess
- This will happen within five years
- Any treatment to deal with the broken/painful/abscessed tooth in the future would be more difficult, more expensive and have less certain results

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Below is an **example** of a choice you might be asked to make.

In this **example**, 'Scenario 1' has been chosen (it was ticked/selected).
This means the parent of the 9-year-old child prefers:

- A filling, and no gap, to a partial gap
- A dentist with enhanced skills in paediatric dentistry to a general dental practitioner
- To make the decision themselves rather than the dentist make it for them
- Avoiding 33 future visits than 15 visits
- Their child to be treated than not have the treatment

They don't mind paying £1350 compared to £80 to secure the other aspects of this scenario.

EXAMPLE	Treatment scenario 1	Treatment scenario 2	No treatment
Treatment	The tooth is filled (no gap is left) 	The tooth is removed and the tooth behind it partially moves forward on its own (a small gap is left) 	No treatment (the decay will get worse) 
Provider of dental care	Dentist with enhanced skills in Paediatric Dentistry	General Dental Practitioner	No care provided
Decision maker	Dentist discusses options with me and my child, but I make the decision on my own	Dentist discusses options with me and my child, but the dentist makes the decision	Dentist discusses options with me and my child, but I make the decision on my own
Number of future treatments avoided	33 future treatment visits	15 future treatment visits	2 future treatment visits
Initial Cost	£1350	£80	£0
Your choice (tick one box only)	Which, if any, of these scenarios would you pick? ● Scenario 1 ○ Scenario 2 ○ No treatment		

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Imagine you had a 9-year-old child who has a compromised adult molar. Which, if any, of the following three scenarios would you pick?

	Treatment scenario 1	Treatment scenario 2	No treatment
Treatment	The tooth is removed and the gap is closed with a false tooth (no gap is left) 	The tooth is removed and the tooth behind it does not move forward on its own (a gap is left) 	No treatment (the decay will get worse) 
Provider of dental care	Specialist in Paediatric Dentistry	Dentist with enhanced skills in Paediatric Dentistry	No care provided
Decision maker	Dentist discusses options with me and my child, and we come to an agreement between us	Dentist discusses options with me and my child, but the dentist makes the decision	Dentist discusses options with me and my child, but I make the decision on my own
Number of future treatments avoided	33 future treatment visits	15 future treatment visits	2 future treatment visits
Initial Cost	£1350	£80	£0

Which, if any, of these scenarios would you pick?

- Scenario 1
- Scenario 2
- No treatment

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And still imagining you had a 9-year-old child who has a compromised adult molar. Which, if any, of the following three scenarios would you pick?

	Treatment scenario 1	Treatment scenario 2	No treatment
Treatment	The tooth is removed and the tooth behind it completely moves forward on its own (no gap is left) 	The tooth is removed and the gap is closed with a false tooth (no gap is left) 	No treatment (the decay will get worse) 
Provider of dental care	Dentist with enhanced skills in Paediatric Dentistry	General Dental Practitioner	No care provided
Decision maker	Dentist discusses options with me and my child, and we come to an agreement between us	Dentist discusses options with me and my child, but I make the decision on my own	Dentist discusses options with me and my child, but I make the decision on my own
Number of future treatments avoided	15 future treatment visits	2 future treatment visits	2 future treatment visits
Initial Cost	£80	£1350	£0

Which, if any, of these scenarios would you pick?

- Scenario 1
- Scenario 2
- No treatment

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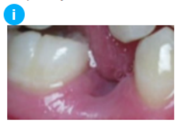
	Treatment scenario 1	Treatment scenario 2	No treatment
Treatment	The tooth is removed and the tooth behind it completely moves forward on its own (no gap is left)  	The tooth is removed and the tooth behind is completely moved forward using metal braces (no gap is left)   	No treatment (the decay will get worse) 
Provider of dental care	Specialist in Paediatric Dentistry	Dentist with enhanced skills in Paediatric Dentistry	No care provided
Decision maker	Dentist discusses options with me and my child, but the dentist makes the decision	Dentist discusses options with me and my child, but I make the decision on my own	Dentist discusses options with me and my child, but I make the decision on my own
Number of future treatments avoided	15 future treatment visits	33 future treatment visits	2 future treatment visits
Initial Cost	£1350	£80	£0

Which, if any, of these scenarios would you pick?

- Scenario 1
- Scenario 2
- No treatment

◄ Previous Next ►

And still imagining you had a 9-year-old child who has a compromised adult molar. Which, if any, of the following three scenarios would you pick?

	Treatment scenario 1	Treatment scenario 2	No treatment
Treatment	The tooth is removed and the tooth behind it completely moves forward on its own (no gap is left)  	The tooth is removed and the tooth behind it partially moves forward on its own (a small gap is left)  	No treatment (the decay will get worse) 
Provider of dental care	General Dental Practitioner	Specialist in Paediatric Dentistry	No care provided
Decision maker	Dentist discusses options with me and my child, but the dentist makes the decision	Dentist discusses options with me and my child, and we come to an agreement between us	Dentist discusses options with me and my child, but I make the decision on my own
Number of future treatments avoided	2 future treatment visits	15 future treatment visits	2 future treatment visits
Initial Cost	£1350	£80	£0

Which, if any, of these scenarios would you pick?

- Scenario 1
- Scenario 2
- No treatment

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And still imagining you had a 9-year-old child who has a compromised adult molar. Which, if any, of the following three scenarios would you pick?

	Treatment scenario 1	Treatment scenario 2	No treatment
Treatment	The tooth is removed and the tooth behind is completely moved forward using metal braces (no gap is left) 	The tooth is removed and the tooth behind it does not move forward on its own (a gap is left) 	No treatment (the decay will get worse) 
Provider of dental care	General Dental Practitioner	Dentist with enhanced skills in Paediatric Dentistry	No care provided
Decision maker	Dentist discusses options with me and my child, and we come to an agreement between us	Dentist discusses options with me and my child, but I make the decision on my own	Dentist discusses options with me and my child, but I make the decision on my own
Number of future treatments avoided	15 future treatment visits	2 future treatment visits	2 future treatment visits
Initial Cost	£200	£1350	£0

Which, if any, of these scenarios would you pick?

- Scenario 1
- Scenario 2
- No treatment

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And still imagining you had a 9-year-old child who has a compromised adult molar. Which, if any, of the following three scenarios would you pick?

	Treatment scenario 1	Treatment scenario 2	No treatment
Treatment	The tooth is removed and the tooth behind is completely moved forward using metal braces (no gap is left) 	The tooth is removed and the tooth behind it partially moves forward on its own (a small gap is left) 	No treatment (the decay will get worse) 
Provider of dental care	General Dental Practitioner	Specialist in Paediatric Dentistry	No care provided
Decision maker	Dentist discusses options with me and my child, and we come to an agreement between us	Dentist discusses options with me and my child, but the dentist makes the decision	Dentist discusses options with me and my child, but I make the decision on my own
Number of future treatments avoided	2 future treatment visits	33 future treatment visits	2 future treatment visits
Initial Cost	£80	£200	£0

Which, if any, of these scenarios would you pick?

- Scenario 1
- Scenario 2
- No treatment

◄ Previous Next ►

And still imagining you had a 9-year-old child who has a compromised adult molar. Which, if any, of the following three scenarios would you pick?

	Treatment scenario 1	Treatment scenario 2	No treatment
Treatment	The tooth is filled (no gap is left) 	The tooth is removed and the gap is closed with a false tooth (no gap is left) 	No treatment (the decay will get worse) 
Provider of dental care	Specialist in Paediatric Dentistry	Dentist with enhanced skills in Paediatric Dentistry	No care provided
Decision maker	Dentist discusses options with me and my child, but I make the decision on my own	Dentist discusses options with me and my child, but the dentist makes the decision	Dentist discusses options with me and my child, but I make the decision on my own
Number of future treatments avoided	33 future treatment visits	15 future treatment visits	2 future treatment visits
Initial Cost	£80	£200	£0

Which, if any, of these scenarios would you pick?

- ◯ Scenario 1
- ◯ Scenario 2
- ◯ No treatment

◄ Previous Next ►

And still imagining you had a 9-year-old child who has a compromised adult molar. Which, if any, of the following three scenarios would you pick?

	Treatment scenario 1	Treatment scenario 2	No treatment
Treatment	The tooth is removed and the tooth behind it does not move forward on its own (a gap is left) 	The tooth is removed and the tooth behind it completely moves forward on its own (no gap is left) 	No treatment (the decay will get worse) 
Provider of dental care	Specialist in Paediatric Dentistry	General Dental Practitioner	No care provided
Decision maker	Dentist discusses options with me and my child, and we come to an agreement between us	Dentist discusses options with me and my child, but I make the decision on my own	Dentist discusses options with me and my child, but I make the decision on my own
Number of future treatments avoided	2 future treatment visits	33 future treatment visits	2 future treatment visits
Initial Cost	£200	£80	£0

Which, if any, of these scenarios would you pick?

- ◯ Scenario 1
- ◯ Scenario 2
- ◯ No treatment

◄ Previous Next ►

And still imagining you had a 9-year-old child who has a compromised adult molar. Which, if any, of the following three scenarios would you pick?

	Treatment scenario 1	Treatment scenario 2	No treatment
Treatment	The tooth is removed and the tooth behind it does not move forward on its own (a gap is left) 	The tooth is filled (no gap is left) 	No treatment (the decay will get worse) 
Provider of dental care	General Dental Practitioner	Dentist with enhanced skills in Paediatric Dentistry	No care provided
Decision maker	Dentist discusses options with me and my child, but I make the decision on my own	Dentist discusses options with me and my child, and we come to an agreement between us	Dentist discusses options with me and my child, but I make the decision on my own
Number of future treatments avoided	33 future treatment visits	2 future treatment visits	2 future treatment visits
Initial Cost	£200	£1350	£0

Which, if any, of these scenarios would you pick?

- Scenario 1
- Scenario 2
- No treatment

◄ Previous Next ►

And still imagining you had a 9-year-old child who has a compromised adult molar. Which, if any, of the following three scenarios would you pick?

	Treatment scenario 1	Treatment scenario 2	No treatment
Treatment	The tooth is removed and the tooth behind it completely moves forward on its own (no gap is left) 	The tooth is removed and the tooth behind is completely moved forward using metal braces (no gap is left) 	No treatment (the decay will get worse) 
Provider of dental care	Specialist in Paediatric Dentistry	Dentist with enhanced skills in Paediatric Dentistry	No care provided
Decision maker	Dentist discusses options with me and my child, but the dentist makes the decision	Dentist discusses options with me and my child, but I make the decision on my own	Dentist discusses options with me and my child, but I make the decision on my own
Number of future treatments avoided	15 future treatment visits	33 future treatment visits	2 future treatment visits
Initial Cost	£1350	£80	£0

Which, if any, of these scenarios would you pick?

- Scenario 1
- Scenario 2
- No treatment

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And still imagining you had a 9-year-old child who has a compromised adult molar. Which, if any, of the following three scenarios would you pick?

	Treatment scenario 1	Treatment scenario 2	No treatment
Treatment	The tooth is removed and the tooth behind it does not move forward on its own (a gap is left)  	The tooth is removed and the tooth behind it completely moves forward on its own (no gap is left)  	No treatment (the decay will get worse) 
Provider of dental care	Dentist with enhanced skills in Paediatric Dentistry	Specialist in Paediatric Dentistry	No care provided
Decision maker	Dentist discusses options with me and my child, but the dentist makes the decision	Dentist discusses options with me and my child, and we come to an agreement between us	Dentist discusses options with me and my child, but I make the decision on my own
Number of future treatments avoided	2 future treatment visits	33 future treatment visits	2 future treatment visits
Initial Cost	£1350	£80	£0

Which, if any, of these scenarios would you pick?

- Scenario 1
- Scenario 2
- No treatment

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Below is an **example** of a choice you might be asked to make.

In this **example**, 'Scenario 2' has been chosen (it was ticked/selected).
This means the parent of the 9-year-old child prefers:

- A partial gap to a filling and no gap
- A general dental practitioner to a dentist with enhanced skills in paediatric dentistry
- The dentist to make the decision rather than they themselves make it
- Avoiding 15 future visits than 33 visits
- Their child to be treated than not have the treatment

They are only willing to pay £80 and not £1350 to secure the other aspects of this scenario.

EXAMPLE	Treatment scenario 1	Treatment scenario 2	No treatment
Treatment	The tooth is filled (no gap is left)  	The tooth is removed and the tooth behind it partially moves forward on its own (a small gap is left)  	No treatment (the decay will get worse) 
Provider of dental care	Dentist with enhanced skills in Paediatric Dentistry	General Dental Practitioner	No care provided
Decision maker	Dentist discusses options with me and my child, but I make the decision on my own	Dentist discusses options with me and my child, but the dentist makes the decision	Dentist discusses options with me and my child, but I make the decision on my own
Number of future treatments avoided	33 future treatment visits	15 future treatment visits	2 future treatment visits
Initial Cost	£1350	£80	£0
Your choice **(tick one box only)**	Which, if any, of these scenarios would you pick? ○ Scenario 1 ● Scenario 2 ○ No treatment		

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(Alternative nested methodological sample scenario)

Appendix M – Mixed-Logit Analyses

Mixed (random) logistic regression				Number of obs.	14,064	
(Iteration 12) Log likelihood = -2801.4112				**LR chi2(12)**	20.10	
				Prob > chi2	0.0456	
				Pseudo R2	0.4432	
Mean						
Choice	**Coefficient**	**Std. err.**	**z**	**P>z**	**95% conf. interval**	
ASC	5.015951	0.6800717	7.38	0.000	3.683035	6.348867
Filling	1.143781	.2641839	4.33	0.000	0.625990	1.661572
Full gap	-1.369656	.2098664	-6.53	0.000	-1.780986	-0.958325
Partial gap	-0.460548	0.1422993	-3.24	0.001	-0.739449	-0.181646
No tooth gap	0.5762641	0.1059992	5.44	0.000	0.368509	0.784018
Ortho gap	0.8812233	0.1637926	5.38	0.000	0.5601957	1.202251
False tooth gap	-0.7710644	0.1317117	-5.85	0.000	-1.029215	-0.512914
GDP	0.1015524	0.061822	1.64	0.100	-0.019616	0.222721
Enhanced GDP	-0.2143514	0.0605665	-3.54	0.000	-0.333059	-0.095643
Specialist	0.112799	0.0510067	2.21	0.027	0.0128276	0.212770
Dentist	0.012943	0.0898047	0.14	0.885	-0.1630709	0. 188956
Shared	0.0649139	0.0503417	1.29	0.197	-0.033754	0.163581
Patient	0.0778569	0.0572727	1.36	0.174	-0.034395	0.190109
Tx Avoid (cont.)	-0.0074853	0.0032218	-2.32	0.020	-0.013800	-0.001170
Cost	-0.0074853	0.0001914	-6.80	0.000	-0.0016757	-0.000925

Standard Deviation						
ASC	1.101169	.6952977	1.58	0.113	-.2615896	2.463927
Filling	1.372934	.2641839	4.39	0.003	0.645456	1.676764
Full gap	0.148596	.3593718	0.41	0.679	-0.555759	0.852952
Partial gap	1.119611	.4682647	2.39	0.017	0.201828	2.037393
No tooth gap	-0.591605	.4594708	-1.29	0.198	-1.492152	0.308940
Ortho gap	1.975452	.5384124	3.67	0.000	0.920182	3.03072
False tooth gap	-0.020818	.3518166	-0.06	0.953	-0.710366	0.668729
GDP	0.245768	0.061822	1.64	0.235	-0.019616	0.450304
Enhanced GDP	-0.239939	.1986536	-1.21	0.227	-0.629293	0.149414
Specialist	-0.051389	.1768388	-0.29	0.771	-0.397987	0.295207
Dentist	0.007382	0.0898047	0.14	0.634	-0.143543	0.344737
Shared	0.229284	0.1866897	1.23	0.219	-0.136620	0.595189
Patient	0.005570	0.1511179	0.04	0.971	-0.290615	0.301756
Tx Avoid (cont.)	-0.037382	0.0125167	-2.99	0.003	-0.061914	-0.012850
Cost	0.000760	0.000281	2.70	0.007	0.000207	0.001312

Choice	Coefficient	Std. err.	z	P>z	95% conf. interval	
ASC_wtp	3856.57	577.88	6.67	0.000	2723.94	4989.19
Filling_wtp	879.41	120.88	7.28	0.000	642.49	1116.33
Full gap_wtp	-1053.08	85.24	-12.35	0.000	-1220.14	-886.01
Partial gap_wtp	-354.10	86.19	-4.11	0.000	-523.08	-185.18
No tooth gap_wtp	443.07	87.25	5.08	0.000	272.07	614.07
Ortho gap_wtp	677.54	99.35	6.82	0.000	482.82	872.26
False tooth gap_wtp	-592.84	80.76	-7.34	0.000	-751.12	-434.56
GDP_wtp	78.08	45.70392	1.71	0.088	-11.49836	167.6577
Enhanced GDP_wtp	-164.81	40.58	-4.06	0.000	-244.35	-85.26
Specialist_wtp	86.73	38.12	2.28	0.023	12.02	161.44
Dentist_wtp	-109.77	44.36	-2.47	0.013	-196.72	-22.82
Shared_wtp	49.91	39.09	1.28	0.202	-26.71	126.53
Patient_wtp	59.86	42.75	1.40	0.161	-23.92	143.64
Tx Avoid (cont.) _wtp	-5.76	2.26	-2.55	0.011	-10.19	-1.32

Appendix N – Completed Cheers-2022 Checklist

CHEERS 2022 Checklist

Topic	No.	Item	Location where item is reported
Title			
	1	Identify the study as an economic evaluation and specify the interventions being compared.	200
Abstract			
	2	Provide a structured summary that highlights context, key methods, results, and alternative analyses.	I
Introduction			
Background and objectives	3	Give the context for the study, the study question, and its practical relevance for decision making in policy or practice.	200
Methods			
Health economic analysis plan	4	Indicate whether a health economic analysis plan was developed and where available.	p67
Study population	5	Describe characteristics of the study population (such as age range, demographics, socioeconomic, or clinical characteristics).	p201
Setting and location	6	Provide relevant contextual information that may influence findings.	p202
Comparators	7	Describe the interventions or strategies being compared and why chosen.	p201
Perspective	8	State the perspective(s) adopted by the study and why chosen.	p205
Time horizon	9	State the time horizon for the study and why appropriate.	p204
Discount rate	10	Report the discount rate(s) and reason chosen.	p214
Selection of outcomes	11	Describe what outcomes were used as the measure(s) of benefit(s) and harm(s).	p213
Measurement of outcomes	12	Describe how outcomes used to capture benefit(s) and harm(s) were measured.	p205 -p213

Topic	No.	Item	Location where item is reported
Valuation of outcomes	13	Describe the population and methods used to measure and value outcomes.	p205 -p213
Measurement and valuation of resources and costs	14	Describe how costs were valued.	p205 -p213
Currency, price date, and conversion	15	Report the dates of the estimated resource quantities and unit costs, plus the currency and year of conversion.	p205 -p213
Rationale and description of model	16	If modelling is used, describe in detail and why used. Report if the model is publicly available and where it can be accessed.	p201 -p204
Analytics and assumptions	17	Describe any methods for analysing or statistically transforming data, any extrapolation methods, and approaches for validating any model used.	p213 -p214
Characterising heterogeneity	18	Describe any methods used for estimating how the results of the study vary for subgroups.	N/A
Characterising distributional effects	19	Describe how impacts are distributed across different individuals or adjustments made to reflect priority populations.	p216 -p218
Characterising uncertainty	20	Describe methods to characterise any sources of uncertainty in the analysis.	p215-216
Approach to engagement with patients and others affected by the study	21	Describe any approaches to engage patients or service recipients, the general public, communities, or stakeholders (such as clinicians or payers) in the design of the study.	p201 & Appendix M
Results			
Study parameters	22	Report all analytic inputs (such as values, ranges, references) including uncertainty or distributional assumptions.	p219 -p239
Summary of main results	23	Report the mean values for the main categories of costs and outcomes of interest and summarise them in the most appropriate overall measure.	p219 -p239
Effect of uncertainty	24	Describe how uncertainty about analytic judgments, inputs, or projections affect findings. Report the effect of choice of discount rate and time horizon, if applicable.	p219 -p239

Topic	No.	Item	Location where item is reported
Effect of engagement with patients and others affected by the study	25	Report on any difference patient/service recipient, general public, community, or stakeholder involvement made to the approach or findings of the study	p201 & Appendix M
Discussion			
Study findings, limitations, generalisability, and current knowledge	26	Report key findings, limitations, ethical or equity considerations not captured, and how these could affect patients, policy, or practice.	p245 -p251
Other relevant information			
Source of funding	27	Describe how the study was funded and any role of the funder in the identification, design, conduct, and reporting of the analysis	N/A
Conflicts of interest	28	Report authors conflicts of interest according to journal or International Committee of Medical Journal Editors requirements.	N/A

From: Husereau D, Drummond M, Augustovski F, et al. Consolidated Health Economic Evaluation Reporting Standards 2022 (CHEERS 2022) Explanation and Elaboration: A Report of the ISPOR CHEERS II Good Practices Task Force. Value Health 2022;25. doi:10.1016/j.jval.2021.10.008

Appendix O – Feedback on model from initial meeting with expert panel

Changes to Model Structure		
Summarised Feedback	**Decision**	**Rationale for decision**
Need a 2-way arrow between the Paediatric and Orthodontic states	Include	Valid point that needs to be included in the model as often each team will consult the other without going back to primary care dentist
Need an onward arrow from Ortho opinion to extraction	Include	Valid point that needs to be included in the model as orthodontists can ask primary care dentist to extract
Include compensating (remove of opposing health teeth) extractions	Include	How do we model an individual with 4 molars, and can we account for variation in how affected the molars are. This is a key question to answer in the model as current practice varies between practitioners. We will have some data to populate this model, however, some would need expert opinion. This is relevant for both MIH and Caries. This would mean modelling a child with 4 molars of varying degrees of disease. Depending on how complex the model becomes; we may have to assume this is either done or not.
If child going for GA, then additional events i.e. baby teeth extracted at the same GA as the cFPM	Do not include	The consequences (whatever outcome measure we use) will be small and not relatable across the life course. In addition, the costs will be very similar. Not relevant to answering the research question.
Ortho opinion should be at an earlier stage in the model	Do not include	It is not feasible that a child will be sent for an orthodontic opinion at age 6/7. Therefore, having the ortho opinion where it lies in the model is correct.
Looking at differences between England, Scotland, Wales & NI	Do not include	Model structure will be based on the English system
Does the "fillable" tooth need to be asymptomatic?	Include	Symptoms need to be included (suggestive of reversible pulpitis) to make sure the outcome is more generalisable
Include a multi-disciplinary/multi-agency opinion (safeguarding, speech and language etc.)	Do not include	This is likely to be a very rare event for the majority of patients.
Consider changing "do nothing" to "active monitoring"	Include	I would consider changing "do nothing" to "active monitoring". – Although it is purely semantics, in terms of publishing the model, it emphasises there is still a need to review these teeth.
Is a fillable tooth just for caries and not MIH? Is there likely to be different outcomes for each disease?	Further consideration	We could model for MIH and for Caries separately. The states and events will be the same, however, transition probabilities will be different up to a point in the model, although very subtly different. These will only differ from early events. This could lead to a scenario analysis. Possibly better to deal as a sub-group analysis, or include in sensitivity analysis?

Sub-group analysis considerations		
Summarised Feedback	**Decision**	**Rationale for decision**
Inclusion of children with co-morbidities, such as neurodisability, are decisions will be different compared to healthy children	Do not include	Neurodisability as a sub-group is too rare, and often clinical decision is to extract based on the need that restorations will need replaced, whereas extraction will remove the problem earlier. Also, this group is too rare to model. This model could be adapted for these sub-groups but not to be included as part of this model.
Model children who do or do not require orthodontics, based on poor OH etc.	Further consideration needed	Consideration is needed as to what orthodontic characteristics children have when they enter the model. Possibly sub-group analysis of children with different malocclusions, or different levels of OH.
The age of the child going through the pathway: extraction → space → no space, will vary in terms of outcomes of space closure, or teeth drifting	Include	This is a key question to answer, and my doing different sub-group analyses of children of varying ages, we can help answer this question. May also be included in the sensitivity analyses to highlight further areas of research? EVPI?
How do we measure, or include, the psychology of the impact of the treatment?	Further consideration needed	I am not sure psychological impacts can be built into the model, as a measure of utility given the challenges of using QALYs derived from EQ-5D in dentistry. We could consider a sub-group analyses of children who are anxious/not anxious, or possibly link to the model if anxiety (or appearance relating to psychological impact) is picked up by the DCE. We could use data from the literature relating to OHIP and include in a CEA as the cost per point score changed in OHIP values. It is likely different methodologies are needed to answer this question, but further exploration would be justified.
A group of patients who have extractions early may go onto require premolar extractions as part of ortho treatment.	Further consideration needed	Need to look at the evidence on how many children this situation will affect. Alternatively, we can consider the ideal treatment, assuming that an orthodontic opinion has been consulted as it is ok to have the molars extracted, or the extractions are delayed until later, leaving these children in the do nothing or filled teeth state.
Variable characteristics of patients • Overall stage of dentition • Motivation, support and • Malocclusion severity	Further consideration needed	These are sensible characteristics, and would merit sub-group analyses, particularly the different malocclusions and levels of OH. Need to think about OH and motivation as separate issues
Is a fillable tooth just for caries and not MIH? Is there likely to be different outcomes for each disease?	Further consideration	We may want to have different sub-groups of children with caries and those with MIH, rather than two separate models?

Consider for inclusion in the sensitivity analysis		
Summarised Feedback	Decision	Rationale for decision
Is a fillable tooth just for caries and not MIH? Is there likely to be different outcomes for each disease?	Further consideration	Could lead to a sensitivity analysis of different very subtle parameters for the different disease processes?
Changes to Model Parameters		
Summarised Feedback	Decision	Rationale for decision
Do not offer RCT to children under the age of 10.5 years old as it is unlikely to be successful	Do not include	As comments above, age will be important to consider, however, there is some evidence of success for pulp treatments for these age groups and therefore we have to model children under 10.5 going to the restorative phase including early RCT. This is more a personal clinician preference.

Appendix P– Search Strategies for parameter data for model

MIH & Caries – Treatment

*#1 (molar AND hypominerali*ation) OR (demarcated AND opacities) OR (MIH) OR (developmental AND opacit*) OR (idiopathic OR nonfluoride) OR (enamel AND opacit*) OR (enamel AND defect) OR (enamel AND (hypominerali*ation) OR (developmental AND dental AND defects) OR (developmental AND hypominerali*ation)*

#2 caries OR decay OR (caries AND defect) OR (decay AND defect)

#3 (manage) OR (treat*) OR (restor*) OR (filling) OR (extract*) OR (tooth removal) OR (resin) OR (composite) OR (orthodont*) OR (seal*) OR (crown) OR (dental crown) OR (prevent*) OR (fluorid*) OR (SDF) OR (silver diamine fluoride) OR (CPP-ACP) OR (casein phosphopeptide-amorphous calcium phosphate) OR (onlay) OR (inlay) OR (root canal) OR (pulp therapy) OR (pulpotomy) OR (pulpectomy) OR (vital pulp therap*) OR (endodontic) OR (infiltration) OR (reminerali*ation) OR (resin retained bridge) OR (dental bridge) OR (dental implant) OR (dentures)*

#1 AND #2 and #3

Total hits: 122