

Management of Children with Molar-Incisor-Hypomineralisation

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Abstract

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Introduction & Aims: Molar-incisor-hypomineralisation (MIH) is a qualitative enamel defect of permanent molar and incisor teeth which affects 14.2% of children worldwide. Its presentation is varied with the possibility of one to four molars being affected, with or without up to eight incisors. Each MIH tooth exists on a spectrum with the most mildly affected having demarcated opacities and no symptoms, and the most severely affected having post-eruptive breakdown, caries, extreme sensitivity or toothache. In the UK the majority of children have dental treatment in primary care but may see a specialist in paediatric dentistry on referral. The aim of this thesis was to investigate how children with MIH are managed in primary and secondary care in the UK.

Methods: A retrospective service evaluation within a secondary care specialist centre for paediatric dentistry was registered with the clinical governance unit. Data was collected for all 48 children diagnosed with MIH between 1st January and 31st December 2015 on consultant-led new-patient clinics, until completion of their treatment. The data collected concerned the pre-referral treatment, history and diagnoses, and the treatment completed in the hospital.

The accuracy of GDPs in diagnosis and treatment planning of MIH was assessed by an electronic survey with clinical vignettes disseminated across the UK. To assess diagnostic skill 10 clinical photographs were selected. Six cases had MIH and/or hypomineralised second primary molars (HSPM) (seven possible diagnoses). Four control cases showing caries, fluorosis, amelogenesis imperfecta and dentinogenesis imperfecta were also included. The treatment planning section presented two vignettes: a child with mild MIH who was unhappy about the appearance of his teeth

and an anxious child with severe MIH, caries and sensitivity. The survey was distributed by email and across social media platforms. Data collection occurred between February and May 2019.

Semi-structured interviews were conducted with 10 GDPs based in England to explore their experience and understanding of MIH in primary care. Semi-structured interviews with GDPs who regularly treat children were completed in May 2020. Interviews were recorded over telephone and transcribed, followed by coding and thematic analysis using an inductive approach.

Results: Service Evaluation - Out of 397 records reviewed, 48 children (12.1%) had MIH, with 81.3% and 18.8% of patients having severe and mild MIH respectively. Treatment was completed at the specialist centre for 44 (91.7%) patients. Twenty-five (52.1%) patients had an extraction of one or more first permanent molar teeth. Sixteen patients had the extractions at between eight and 10 years old and two had the extractions later as part of an orthodontic plan.

Vignette Survey – Seventy-six GDPs completed the diagnosis section. 68.4% of participants were female (n=52). 83% (n=63) graduated after the year 2000. The number of accurate diagnoses for each case were as follows – mild MIH (molars/incisors) 65.79%; mild MIH (molars only) 3.95%; HSPM & MIH (HSPM result) 0%; HSPM & MIH (MIH result) 50%; Severe MIH (post-eruptive breakdown) 63.16%; Severe MIH (caries) 31.58%; HSPM 3.95%. 58 GDPs completed the treatment planning section. Around half of participants addressed the aesthetic concerns of the child in vignette one. The majority of participants demonstrated sound treatment planning in terms of preventive care, and management of molars. More GDPs identified increased caries risk in vignette two. No significant relationship was found between the number of accurately diagnosed cases and the number of correctly treatment planned molars for vignette one (-0.054 p=0.689) or two (-0.03 p=0.808) when assessed using the Mann Whitney Test.

Telephone Interviews – themes of ‘setting the scene’, ‘fighting the tooth’, ‘working within the system’, and ‘self and interpersonal insight’ were developed after coding of transcripts. The overarching theme was of managing uncertainty. Participants had good knowledge of MIH and

treatment strategies, but barriers to optimum care existed at tooth, child, practice and systematic levels.

Conclusions: Findings from all three studies demonstrated GDPs were able to manage mild MIH effectively in primary care, whilst referral was utilised appropriately for more severe cases.

Treatment in secondary care mainly comprised management of severely affected molars by extraction under general anaesthetic. GDPs were best able to diagnose MIH when both incisors and molars were affected, and there was no caries present. Treatment planning was sound with most GDPs working at the level expected as non-specialists when faced with clinical vignettes. GDPs faced uncertainty when planning treatment for poor prognosis first permanent molars (FPM) and aesthetic treatment for incisors, and whilst administering LA and restoring FPM. In addition, the system of primary care dentistry, remuneration, and long waiting lists on referral created barriers to caring for children with MIH. GDPs managed some of these uncertainties by referring children to perceived experts in secondary care. The level at which the child became 'complex' and required referral was different for each clinician and related to the patient, their own experience, and current work environment. Clear care pathways taking into account the complexity of the case and severity of MIH are required.

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List of Abbreviations

BNF	British National Formulary
BSPD	British Society of Paediatric Dentistry
CDS	Community Dental Service
CPP - ACP	Caesin phosphopeptide- activated calcium phosphate
DCT	Dental Core Trainee
DMH	Deciduous molar hypomineralisation
EAPD	European Association of Paediatric Dentistry
EDI	Enamel Defect Indices
FPM	First permanent molars
GA	General anaesthetic
GDP	General dental practitioner
GIC	Glass ionomer cement
HSPM	Hypomineralised second primary molars
LA	Local anaesthetic
LUDH	Liverpool University Dental Hospital
mDDE	Modified Developmental Defects of Enamel
MIH	Molar-incisor-hypomineralisation
NHS	National Health Service
OPG	Orthopantomogram
PEB	Post-eruptive breakdown
PMC	Preformed metal crown
RMGIC	Resin-modified glass ionomer cement
UDA	Unit of Dental Activity

UK	United Kingdom
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Chapter 1 - Introduction

1.1 Overview of Molar-incisor-hypomineralisation

Molar-incisor-hypomineralisation (MIH) is a qualitative defect of enamel which presents as demarcated opacities on one to four first permanent molars (FPM) and often, but not always, incisor teeth (Weerheijm et al. 2001). An estimated 240 million people across the world have MIH, with around 4.8 million new cases developing every year in children (Aguirre et al. 2020). The condition has a high morbidity, with some children eventually having FPM extracted at a young age due to symptoms of pain or inability to effectively restore them, having first endured multiple unsuccessful attempts at restorative treatment. This can be stressful for both patient and dentist. Management in mild cases may be purely preventive but more complex cases may require use of adjuncts such as sedation or general anaesthetic (GA), and multidisciplinary working between paediatric dentistry and orthodontic specialists. The presence of hypomineralisation in primary molar teeth has recently been implicated as a potential risk marker for developing MIH, due to overlapping time periods of crown development (Weerheijm 2015), and the tips of permanent canines and second permanent molars may also occasionally be affected. In comparison to other enamel defects, Hubbard et al discuss its high cost to society, with a modelling analysis of the population in Australia predicting that the treatment-cost potential of MIH is similar to national expenditure on leading cancers (Hubbard et al. 2017). Despite its high prevalence, cost to families, and to society in general, in a recent survey in the United Kingdom (UK) only 57% of general dental practitioners (GDPs) felt confident in diagnosing MIH (Kalkani et al. 2016). Whilst the exact aetiology of MIH is still unknown and therefore the condition cannot be prevented, effective management is completely reliant on GDPs identifying the condition in their patients, commencing appropriate treatment or referring to a provider who can. The evidence to date from across the world indicates many GDPs do not feel confident in diagnosis and are unsure of the correct management of these patients.

1.2 Pathways for Paediatric Dental Care

Within the UK, dental care for children occurs mainly with GDPs in primary care dental practices. Where a child presents with more complex dental needs, due to medical history, moderate to severe anxiety, or complex dental disease beyond the scope of general practice, children are referred into community or hospital dental services, where they can receive specialist care, usually under the direction of specialists or consultants in paediatric dentistry. MIH is a condition that requires input from both specialists and generalists due to its variable presentation and severity. It is recommended that mild MIH can be managed wholly in primary care by GDPs, whereas severe presentations will usually require input from a specialist for treatment planning or treatment itself (British Society of Paediatric Dentistry 2020a, Almuallem and Busuttil-Naudi 2018). Since this system relies on GDPs diagnosing the condition, carrying out initial management and identifying when management would benefit from referral to a specialist, their diagnostic skill and knowledge of appropriate treatment strategies are key in the care of children with MIH. In addition, whilst the exact aetiology of MIH remains unknown, and therefore the condition cannot be prevented, early diagnosis and implementation of preventive strategies will have a large bearing on patient symptoms, and long-term prognosis of these teeth. Therefore, the management of MIH by GDPs is key to the outcomes of children presenting with this condition.

1.3 Why is this research original?

To the authors knowledge, there is no published work investigating the current practices of GDPs and potential barriers to care when managing children with MIH in UK primary care. This thesis will present a comprehensive literature review, followed by studies investigating referral practices to secondary care and the management of these children in a hospital setting, the accuracy of GDPs when diagnosing and treatment planning children with MIH, and interviews to explore the experience of GDPs regarding their thoughts, and feelings when looking after children with MIH.

Chapter 2 - Literature Review

2.1 History

The first reported cases of MIH originated as early as the 1970's (Koch et al. 1987), when a cohort of patients born in 1970 in Sweden were found to have a high prevalence of enamel hypomineralisation of FPM and incisors. The condition was reported under a number of different idioms including 'cheese molars' and 'non-fluoride hypomineralisation in permanent first molars' until it was officially defined as a unique condition at the European Academy of Paediatric Dentistry (EAPD) Congress in 2001, where several delegates presented on the phenomenon. An official name of molar-incisor-hypomineralisation was agreed in order to streamline future research activities (Weerheijm et al. 2001). Although seen as a modern developmental defect the condition has been identified in ancient remains. A recent study which analysed the teeth of ancient human remains found only 3% of cases had MIH, which is significantly less than current prevalence. This supports their hypothesis that the condition is related to modern living conditions or other health-related conditions (Kuhnisch et al. 2016). It is thought that MIH has become more widely reported in recent times due to the decrease in dental caries, which previously masked the condition.

Similar to MIH, hypomineralisation of the second primary molars has also been identified as a distinct enamel defect and its relationship to MIH investigated in a number of studies across the world. Currently there are two different terminologies being used to describe this phenomenon in primary molars, which are hypomineralised second primary molars (HSPM) (Weerheijm 2015) and deciduous molar hypomineralisation (DMH)(Costa-Silva et al. 2013).

2.2 Prevalence

The true prevalence of MIH is still disputed. Differences in prevalence may be explained by environmental and genetic factors which vary between geographical regions. Non standardised methods of data collection using different indices including the EAPD (Weerheijm et al. 2003), mDDE (modified developmental defects of enamel) and EDI (enamel defect indices) indices, have also contributed to a highly varied prevalence (Jalevik 2010), with a Brazilian study reporting that 40.2% of children examined had evidence of MIH (Soviero et al. 2009). Recent systematic reviews have estimated prevalence worldwide to be 14.2% (Zhao et al. 2018), and 13.1% (Schwendicke et al. 2018), although due to the high heterogeneity of studies included, these results should be interpreted with caution. Elfrink et al suggested that a standardised protocol for future prevalence and aetiology studies be adopted, with at least 300 children in a sample (Elfrink et al. 2015). Despite the heterogeneity of studies, it is clear that MIH is a highly prevalent condition which affects children from across the world.

The prevalence of HSPM is less well researched but probably lies between 2.7-21.8% with studies having been carried out in Australia, the Netherlands, Spain, India, Germany and Brazil (Costa-Silva et al. 2013, Kuhnisch et al. 2014, Mittal et al. 2016, Negre-Barber et al. 2016, Owen et al. 2018, Elfrink et al. 2012, Amend et al. 2020). A more recent study in Syria found a prevalence of HSPM of 41%, which is way above prevalence found elsewhere. The author commented on potential explanations for this, including the effects on health, nutrition and general wellbeing by the ongoing civil war there (Halal and Raslan 2020). In comparison to studies regarding MIH, these studies can be more accurately compared as they all follow the EAPD guidelines for diagnosis of MIH and HSPM (Ghanim et al. 2015, Elfrink et al. 2015), although there is a wide variation in the age of child examined, with some studies incorporating comparisons between prevalence of MIH and HSPM of the same cohort, whilst others looked only at children in the primary dentition.

2.3 European and UK Prevalence

MIH is well reported in Europe and the UK. A survey of members of the EAPD found that MIH was recognised in 30 countries across Europe and that members felt that it was a clinical problem (Weerheijm and Mejare 2003). In the UK several regional studies have been carried out, such as that by Balmer et al, who found a prevalence of 15.9% when 3233 12-year-old children were examined in the North East of England (Balmer et al. 2012). Unfortunately, this study did not use the EAPD index and since the children were 12-years-old, there is the chance that MIH-affected molars may already have been extracted, although the methodology did try to account for this. Currently no studies have been carried out in the UK to assess the prevalence of HSPM, however a cross-sectional study of 414 eight and nine-year-old children in Spain found a prevalence of 14.5% (Negre-Barber et al. 2016) and a cross-sectional study of 693 10-year-old children (as part of a longitudinal birth cohort) in Germany found a prevalence of 6.9% (Kuhnisch et al. 2014). In the most recent Child Dental Health Survey, it was found that 34% of 12-year-old children in England had dental decay (NHS Digital 2015) which is considered the most significant oral health issue for children across the world (Moynihan and Kelly 2014). Dental trauma (all types) affects 12% of 12-year-old children in the UK (NHS Digital 2015). This means that for every two children a GDP sees with caries, they should expect to see one child with MIH, and it is likely that there are considerably more children with MIH than those suffering dental trauma in the UK. Hubbard discussed the generalised lack of awareness of the extent of the condition and it's likely significant cost to society and families in management (Hubbard et al. 2017). MIH is a significant public health issue.

2.4 Aetiology

Multiple potential causes have been implicated with the development of MIH and HSPM, but at present the true cause remains unknown. Illness or difficulties that disrupt amelogenesis during the formation of the crown of the second primary molars, FPM and incisors have been implicated (Garot

et al. 2021). This includes a time span from 18 weeks inter-uterine life to around three years of age (Weerheijm 2015). This may include maternal illness or problems in the last trimester of pregnancy, difficult labours or caesarean births, and infantile illness until three-years-old when crown formation of the FPM and incisors is complete. In addition, it is now understood that a complex interaction of genes and environmental factors are likely to interact synergistically to cause MIH. Genetic studies using families where one child has MIH, have discovered that alterations to genes that code for amelogenesis are likely to be implicated as risk factors for MIH (Jeremias et al. 2016). In addition, a subsequent study by the same team found that immune and inflammatory cytokines in combination with altered genes for amelogenesis appeared to be related to presence of MIH (Bussaneli et al. 2019). A high quality twin study also found evidence of a genetic influence (Teixeira et al. 2018). Prolonged breast feeding was implicated at the beginning of the century, due to suspected raised levels of the toxin dioxin, however subsequent studies have found no link (Laisi et al. 2008). Fatturi et al carried out a systematic review with meta-analysis, and found maternal illness and stress, caesarean or difficult births, and respiratory disease and fever in childhood to be implicated in increased risk of MIH (Fatturi et al. 2019). More recently Garot et al completed a systematic review with meta-analysis regarding aetiology of MIH (Garot et al. 2021). In addition to discussing recent papers looking at genetic influence, they analysed potential aetiologies in the pre-natal, perinatal and postnatal periods. General maternal illness (unspecified) was identified as significant, whilst prematurity, caesarean sections and other birth complications were found to be statistically significant in the perinatal period, with hypoxia defined as the mechanism by which MIH is caused by these events. Measles, urinary tract and ear infections, gastric disorders, renal disease, respiratory infection and asthma were found to be associated with the development of MIH post-natally, in addition to fever and antibiotic use which may be a proxy of serious illness (Garot et al. 2021). Silva et al had previously found severe respiratory conditions including asthma to be associated with more severe forms of MIH where incisors were affected, in their systematic review from 2016. (Silva et al. 2016b). A criticism of many of these studies is that they are often carried out retrospectively,

and therefore rely on the memory of parents, meaning reliability is low. More high quality, prospective studies such as birth cohorts, with at least 1000 participants are required (Silva et al. 2016b, Elfrink et al. 2015, Somani et al. 2021). Whilst a clear aetiology remains elusive, steps may be taken to prevent MIH by timely identification of children with the condition and the introduction of appropriate clinical management.

The development of the second primary molars begins around the same time as the FPM, although completion of the crown occurs more quickly for the primary tooth. This shared time span means that any hypomineralisation in the primary molars indicate an increased risk for MIH, as the disruption to mineralisation may affect development of both teeth. This has been investigated through a number of studies. Elfrink et al found an odds ratio of 4.4 for the presence of MIH based on presence of primary molar hypomineralisation. A weakness of this study was that it was carried out in five and six-year-olds who may not have had all FPM teeth present, as eruption usually occurs from six years (Elfrink et al. 2012). The following year Costa-Silva et al published a study which found that although children with primary molar hypomineralisation had a higher incidence of MIH, this didn't reach significance. This study followed up children aged three to six-years-old for two years, so again there is the chance that not all FPM would have been erupted in this time span (Costa-Silva et al. 2013). In a recent study by Negre-Barber et al, 414 eight and nine-year-old children were examined by calibrated clinicians for presence of HSPM and MIH, and it was found that there was an odds ratio of 18.2 for children with HSPM also developing MIH (Negre-Barber et al. 2016). More recently, a meta-analysis investigating HSPM as a predictor for MIH found that children with HSPM were almost five times more likely to have MIH in their permanent teeth (odds ratio 4.6) in comparison to healthy children, when considering the studies above (Garot et al. 2018). Overall, it is clear that there is increased risk of children developing MIH when they have HSPM, and clinicians should be aware of this link.

2.5 Diagnosis and Clinical Presentation

MIH and HSPM affected teeth have distinct features which can distinguish it from other enamel defects (Hubbard et al. 2017). Conditions such as amelogenesis imperfecta and dentinogenesis imperfecta tend to affect all teeth equally and are also present in the primary dentition. There is usually a family history of these conditions. Fluorosis can affect multiple permanent teeth, including FPM and incisors, but tends to be characterised by diffuse opacities rather than demarcated ones. Patients with fluorosis usually do not complain of sensitivity, and there may be a history of excessive fluoride ingestion e.g. toothpaste eating habit as a child (Ghanim et al. 2017). Hypoplasia often occurs in isolated teeth, and is a defect of enamel quantity, rather than quality. This means that the tooth may have an irregular shape or pits, but the outer layer should be relatively smooth, in comparison to the rough edges of a MIH affected molar with post-eruptive breakdown (PEB) (Jalevik and Noren 2000). Considering these factors, the EAPD reported that any studies investigating MIH must consider presence of demarcated opacities, PEB, atypical restoration of FMP and incisors, and presence of extracted FPM due to MIH (Weerheijm et al. 2003). It is felt that other diagnostic tools such as the mDDE criteria were not sufficient as they didn't record PEB and the enamel defect index (EDI) didn't differentiate between diffuse and demarcated opacities (Elcock et al. 2006).

Ghanim et al have built upon this work by developing a scoring system to diagnose and grade severity of MIH (and HSPM), for use in prevalence and epidemiological studies, in order that standardised methods of data collection are used. The system combines both the EAPD criteria and the mDDE index, in order to assign a grade for the severity of MIH affected teeth, whilst also recording other enamel defects. (Ghanim et al. 2017, Ghanim et al. 2015) Ideally children should be eight-years-old at examination so that all FPM and incisors have erupted. During examination, teeth are graded on the basis of their clinical appearance, incorporating the severity based on both the amount of tooth surface affected, and the stage of visible enamel destruction. There is a short form,

suitable for screening, which only includes indexed teeth (permanent incisors, FPM and second primary molars), and a long form, where all teeth present are recorded. This grading method was found to be valid and reliable in a follow-up study in 2018 (Ghanim et al. 2019), and has been used in many epidemiological studies to date.

2.6 Structural Properties

The structure of enamel in MIH affected teeth can explain some of the associated difficulties with patient management. In normal enamel formation, ameloblasts differentiate from cells of the internal enamel epithelium, signalled to do so following the deposition of the first layer of pre-dentine by the odontoblasts. Enamel formation has distinct phases which include the pre-secretory, secretory, transition, maturation and post maturation phases (Berkovitz 2005). In the secretory phase, ameloblasts deposit the enamel matrix from the enamel-dentine junction towards the outer layer of the crown, with thin needle like hydroxyapatite crystals forming immediately. The matrix is high in water and proteins and low in mineral, and it is in the maturation phase that the mineral content of the teeth is vastly increased to around 96% by weight (Jalevik et al. 2001). Water and amelogenins are removed from the enamel, and calcium and phosphate laid down, which increases the bulk of the enamel crystals. For teeth with MIH it is thought that the disturbance of ameloblasts during the maturation phase of enamel formation leads to an increased protein, carbonate, and serum albumin content within the enamel, and lower calcium and phosphate levels, reducing the hardness and elasticity of the enamel and increasing porosity (Berkovitz 2005, Vieira and Manton 2019, Rodd et al. 2020).

Jalevik et al carried out two studies looking at the morphological and elemental properties of FPM teeth affected by hypomineralisation. The first study looked at the microscopic structure of 73

affected teeth that had been extracted due to hypomineralisation (Jalevik and Noren 2000). They discovered four distinct types of defect, with some hypomineralisation extending from the enamel-dentine junction to the outer layers of enamel, and some only found in the deeper layers, with normal enamel towards the surface. All teeth with defects had a thin layer of highly mineralised enamel on the surface, except where there had been PEB or caries progression. Defects tended to follow Hunter-Schreger lines (Jalevik and Noren 2000). It has been hypothesised that more severe hypomineralisation stems from disruption of the ameloblasts in the early maturation phase, and leads to a higher final protein content, as the ameloblasts do not recover to remove the protein matrix and the enamel crystals cannot grow. These severe lesions tend to be softer, brown or yellow in colour, and are associated with more severe symptoms for the patient, such as sensitivity when carrying out toothbrushing and PEB. The less severe lesions tend to be those where the ameloblasts could recover and so the protein content of these lesions are lower, but still above normal levels. This hypomineralisation tends to present clinically as white or creamy lesions which are less porous (Suga 1989). Neves et al found that the more severe a lesion was at baseline, the more likely it was to progress to expose dentine (Neves et al. 2019). The study was well conducted but did not consider the influence of oral hygiene, diet, and fluoride, which may alter progression for severe lesions.

In the second study Jalevik et al looked at the structure of 17 teeth using ion mass spectrometry and x-ray microanalysis (Jalevik et al. 2001). Carbon and oxygen levels were increased in hypomineralised lesions, due to increased protein content, and calcium and phosphate levels were reduced. Fluorine levels were highly variable but tended to be higher in hypomineralised enamel, apart from at the surface where there was no difference between normal and hypomineralised teeth. 'Sound' enamel in these teeth was found to have 5% less mineral content than a 'normal' control tooth (premolar extracted for orthodontic purposes) (Jalevik et al. 2001). Vieira and Manton hypothesized that the

irregular and asymmetric appearance of MIH may be down to differences in how genes are expressed between right and left, and the position of lesions on individual teeth, as a result of tissue pressures during formation on a tooth where amelogenesis had already been disrupted (Vieira and Manton 2019).

These teeth can be extremely sensitive due to constant low level pulpal inflammation. It is thought that the more porous structure and exposed dentine from PEB allows ingress of bacteria and bacterial products into the tooth structure which can irritate the pulp. Rodd et al discovered that both mildly and severely affected teeth have increased pulpal levels of transient receptor potential ion channel (TRPV1), a noxious heat receptor, which could be why these teeth are hypersensitive and difficult to anaesthetise (Rodd et al. 2007).

Hypomineralised teeth are more susceptible to caries due to several factors. Their porous physical structure lends itself to diffusion of damaging plaque acids into the enamel. Severely affected teeth can suffer from PEB and therefore the unusually shaped tooth can act as a plaque retentive factor, making plaque removal more demanding. Toothbrushing of hypersensitive teeth can be painful so children often avoid them, and the rough surfaces lend themselves to plaque accumulation.

Americano et al carried out a systematic review of 17 cross-sectional papers investigating the association between MIH and caries. 16 out of 17 papers found that dental caries was more prevalent in children with MIH (Americano et al. 2017). Although the papers were homogenous in terms of method, with all adhering to EAPD guidelines for assessment of MIH, and 16 studies using DMFT (decayed missing filled teeth) to assess for caries (one study used ICDAS-II (International Caries Detection and Assessment Scale II), none of the studies were deemed to be of high quality when assessed using the Newcastle-Ottawa scale. They went on to discuss the risk of possible overestimation of caries in MIH teeth that have been restored, since it would be impossible to know

whether this could have been from PEB alone. Americano et al closed by commenting on the confounding factors of a high risk caries population, stating that once the 'traditional' caries risk factors are controlled a clearer picture of the effect of MIH on caries susceptibility will unfold. (Americano et al. 2017). A more recent study which assessed factors contributing to caries development in the permanent dentition in a cohort of 206 children enrolled in a community dental programme in Spain, found a significant correlation between caries development and presence of MIH, alongside factors such as cariogenic diet, brushing habits and caries in the deciduous dentition. Although multivariate analysis was carried out, not all possible risk factors for caries were investigated, and therefore there is a risk of confounding variables which may have skewed results (Llena and Calabuig 2017).

Two recent studies looking at HSPM and caries risk, in Syria and Australia have found contradicting results. Amend et al, investigated for presence of HSPM and caries in four and five-year-olds in Syria, and found that children with HSPM were significantly more likely to have caries in primary molar teeth (Amend et al. 2020). In contrast, Owen et al found that there was no increased risk of caries for children with HSPM in a population of three to five-year-olds in Australia (Owen et al. 2018). Differences in the population studied including level of deprivation and caries risk may have led to these differences.

2.7 Clinical Management

EAPD differentiate presentation as mild or severe. Mild cases have occasional sensitivity, mild aesthetic concerns and well demarcated opacities without enamel breakdown (Lygidakis et al. 2010). These lesions tend to be white or cream, and have less severe symptoms due to the higher mineral content relative to brown lesions. Severely affected teeth are those which have evidence of PEB,

caries, severe hypersensitivity with spontaneous pain or pain on toothbrushing, and those children who have strong aesthetic concerns (Lygidakis et al. 2010). These are usually yellow or brown lesions, indicating lower relative mineral content, increased porosity and less resistance to fracture. Some authors use Mathu-Muju and Wright's classification of mild, moderate and severe, where mild is similar to the EAPD definition but moderate includes teeth without sensitivity that have minor PEB within enamel and without cuspal involvement, or caries (Mathu-Muju and Wright 2006). The EAPD definition will be used in this thesis.

Timely and correct management of children with MIH is paramount. Due to the structural properties of molars affected by MIH, prevention should follow national guidelines for children at high risk of caries for both mildly and severely affected teeth. This involves three-monthly applications of 2.2% fluoride varnish as soon as teeth begin to erupt which can help to increase mineral content in affected teeth (Public Health England 2017). Motivating parents and patients to carry out effective tooth brushing is extremely important (Weerheijm 2004). In addition to high fluoride toothpastes, NovaMin (NovaMin Technology, GlaxoSmithKline, Florida, USA) containing toothpastes may also help with sensitivity (Almuallem and Busuttil-Naudi 2018). Many authors also suggest the use of casein phosphopeptide - amorphous calcium phosphate (CPP-ACP) products, such as tooth mousse (GC corporation, Tokyo) in order to promote mineralisation of the hydroxyapatite, making it more acid resistant and reducing sensitivity (Lygidakis 2010, Fitzpatrick and O'Connell 2007, Onat 2013). The use of fluoride and CPP-ACP containing products have been found to have a synergistic affect in comparison to either alone (Almuallem and Busuttil-Naudi 2018). Silver-diamine-fluoride will arrest and prevent caries in the primary dentition, but is officially licensed for dentine sensitivity within the UK so could be used for this reason on MIH FPM where aesthetics aren't a concern (Seifo et al. 2020). Resin fissure sealants are recommended for teeth which are mildly affected and use of an adhesive system is recommended for increased retention (Lygidakis et al. 2009). This management,

which can improve symptoms for the patient and benefit long term prognosis, works best when the condition is identified promptly, and preventive strategies are implemented quickly.

Severely affected molar teeth may require restoration with either composite resins or preformed metal crowns (PMCs) (Willmott et al. 2008). Glass ionomer cements (GIC) may be suitable as fissure sealants and temporary restorations but are not durable and tend to need frequent replacement (Fayle 2003). Another management option is to extract poor prognosis FPMs at eight to 10 years old, in order that the second permanent molars achieve good contact and space closure with the second premolar and second permanent molar tooth (Williams and Gowans 2003). This has been shown to have good clinical outcomes with space closure and alignment in the majority of cases (Jalevik and Moller 2007). A recent cost-effectiveness analysis within German healthcare found that extraction, even with subsequent orthodontic treatment, was the most cost-effective option for severely affected MIH teeth over the lifetime of the patient. This was more cost-effective than placement of composite resin or indirect restorations, in all cases but very late extraction of FPMs when the eruption of the second permanent molar has already occurred (Elhennawy et al. 2017). Although carried out within the German healthcare setting, the findings have implications further afield. Recently, consideration of more minimally invasive approaches for severely affected FPMs have been discussed, such as coronal pulpotomies (Taylor et al. 2020) and temporising with resin-modified GIC (RMGIC) or composite before a durable cast restoration can be placed in early adolescence (Alkhalaf et al. 2020). Somani et al conducted a systematic review of treatment options but were unable to carry out meta-analysis due to the heterogeneity of relevant papers (Somani et al. 2021). Resin fissure sealants, composites, PMCs and lab-made restorations were recommended for molars, however they were unable to recommend a specific approach for incisors.

William et al proposed a clinical management approach which embodies these techniques and defines steps as 'risk identification', 'early diagnosis', 'remineralisation and desensitisation', 'prevention of caries and PEB', 'restorations or extractions' and 'maintenance' (William et al. 2006). Severity indices to help with planning have been developed by two groups. Oliver et al developed the MIH Severity Index (Oliver et al. 2014) which can be used to assess the best treatment options at tooth and mouth level, with individual characteristics such as colour of defects and presence of previous restorations as indicators for treatment type, validated using logistic regression. A criticism of the tool is that the scoring system is lengthy and is therefore unlikely to be utilised chairside by busy GPs. It also fails to consider the presence of caries as a marker of a more severe defect, which is highly relevant in high caries risk populations. The Wurzburg Group in Germany developed the MIH Treatment Need Index, which utilises a scoring system for each sextant in terms of severity and sensitivity, similar to the British Periodontal Exam (Steffen et al. 2017). Although this showed promise, no further papers demonstrating reliability and validity clinically have been published to date. Several papers concerning management discuss the need for referral and multidisciplinary care for complex cases, but don't discuss specifically what a complex case is (Weerheijm 2004, da Costa-Silva 2012, Daly and Waldron 2009). Therefore, decision making regarding referral to specialists appears to be a subjective decision.

2.8 Patient Concerns and Management Issues

Management of these patients can be extremely challenging. FPM erupt at six years of age, and many children may lack the co-operation required for restorative treatment under local anaesthetic (LA) at this age. Even when co-operation is adequate, 'simple' treatment like fissure sealants can be painful and distressing for children with hypersensitive teeth (Fayle 2003). Anaesthesia can be difficult to achieve (Rodd et al. 2007). Bonding of restorations tend to be less effective due to the lower mineral content, although Lygidakis et al found acceptable retention rates for composite

restorations at four years (Lygidakis et al. 2003). In contrast both Mejare and Jalevik found that 18 year old patients who had restorative treatment for MIH, had increased treatment need than those who had had extractions (Mejare et al. 2005), and had had treatment on affected teeth over four times as often as controls without MIH (Jalevik and Klingberg 2012). At the age of nine, in the same group of children, Jalevik and Klingberg found that those with MIH had undergone treatment on FPM 10 times as often as controls. Unsurprisingly, the same study found that these children have higher levels of dental anxiety and showed more frequent behaviour management problems than unaffected peers (Jalevik and Klingberg 2002). If GA is required to manage these patients, more invasive treatment options may be utilised in order to avoid repeat GA later. This may include teeth being extracted before the 'ideal' time, which may cause or exacerbate orthodontic problems (Cobourne et al. 2014). A guidance document created by EAPD discusses the complexity of management options and goes some way to helping clinicians decide what treatment option is best (Lygidakis et al. 2010), however it doesn't discuss what is appropriate for management in primary care. An updated guideline from EAPD is expected later this year.

Alongside issues regarding sensitivity and difficulty restoring these teeth, children often have aesthetic concerns due to opacities present on incisor teeth. Children may see an improvement in appearance with bleaching, micro-abrasion or localised composite restorations/ composite veneers (Wray et al. 2001). A modern approach involves the use of resin infiltration (ICON, DMG, Hamburg), as a microinvasive procedure that should be considered prior to macro enamel removal and restoration (Bekes 2015). This has been shown to improve the reported happiness and confidence of children with enamel defects (Rodd et al. 2011). Some of these treatments are only available when carried out by specialists in paediatric dentistry.

2.9 Delivery of Care and Outcomes

Over the last decade surveys to assess the experience of both paediatric specialists and GDPs in treating children with MIH have taken place in Europe, the Middle East, South America, South East Asia and Australasia (Crombie et al. 2008, Kalkani et al. 2016, Silva et al. 2016a, Ghanim et al. 2011, Wall and Leith 2020, Elhennawy et al. 2020). The results have found that in general only half of GDPs surveyed are confident in the diagnosis of MIH. Exceptions included the study by Gambetta-Tessini et al, where over 80% of clinicians in both Chile and Australia reported confidence in diagnosis (Gambetta-Tessini et al. 2016), and a recent study in Ireland where 71% of GDPs were confident in managing MIH (Wall and Leith 2020).

Silva et al found that a majority (64%) of 4th and 5th year dental students at King Saud University, Saudi Arabia, had not heard of MIH, which indicates a failure in the undergraduate curriculum (Silva et al. 2016a). A survey of 5th year dental students across 22 dental schools in Germany found that although 97% of students were familiar with MIH, only 34% thought they could identify it, and only 16% of those felt confident in doing so. The authors concluded that although the students had a good theoretical knowledge of MIH, perhaps their clinical exposure was not sufficient (Elhennawy et al. 2020).

The most popular materials for management varied amongst countries with clinicians in Malaysia and Norway most frequently using GIC (Hussein et al. 2014). Those in Kuwait favoured composites for moderately affected teeth and PMCs over extractions for severely affected teeth (Alanzi et al. 2018). In Ireland, composite, GIC or RMGIC were most often used, but PMCs were not favoured (Wall and Leith 2020). Differences may be explained by variations in education and general practice in different countries. A majority of dentists also reported difficulties in management due to

behavioural issues with many indicating they would prefer to refer patients with MIH onto specialist services rather than managing these patients in primary care (Gambetta-Tessini et al. 2016, Kopperud et al. 2016, Hussein et al. 2014, Wall and Leith 2020).

Most clinicians felt that further education regarding diagnosis and management would be beneficial (Silva et al. 2016a, Ghanim et al. 2011, Alanzi et al. 2018, Hussein et al. 2014), and guidelines for management and referral would aid successful patient outcomes (Kopperud et al. 2016, Gambetta-Tessini et al. 2016, Kalkani et al. 2016). A limitation of any survey is the risk that clinicians may wish to exhibit socially acceptable responses, and therefore may lead to over reporting confidence in diagnosis and management. A number of these studies targeted dentists registered with paediatric and national dental societies (Crombie et al. 2008, Silva et al. 2016a, Kalkani et al. 2016) and targeted delegates at dental conferences (Hussein et al. 2014, Alanzi et al. 2018) which means that the samples may not be representative of the wider dental population. Traditional postal surveys also tend to have low response rates which can lead to response bias, with only the most interested clinicians responding.

‘The Commissioning Standard for Dental Specialities – Paediatric Dentistry’ was published in 2018. It provides dental care providers with information on how to organise dental care for paediatric patients within NHS England (Office of the Chief Dental Officer England 2018). It describes the three levels of care that should be available to all children, based on the experience and qualifications of the clinician. Tier 1 is care which encompasses all GPs within England. Tier 2 care includes those clinicians with enhanced skills and experience, or additional facilities, which makes more complex treatment possible. Tier 3a care is specialist level care and 3b is consultant level care. The guide also sets out what type of treatment and care is expected at each level. When considering MIH, Tier 1 clinicians are expected to identify and refer developmental defects as appropriate, and Tier 2

clinicians should be able to manage 'hard-tissue defects not requiring specialist care and which are amenable to simple restoration' (Office of the Chief Dental Officer England 2018). Therefore, GDPs within the UK should be able to diagnose MIH and carry out preventive treatment such as fluoride varnish and fissure sealants. Where necessary they should refer onto Tier 2 and 3 providers, although many GDPs will also have the skills to carry out simple restorative treatment for these patients, where other modifying factors such as anxiety or cognitive maturity are not a factor.

To date there is no clear evidence of whether GDPs in the UK are skilled in identifying this important condition clinically, and whether they can construct appropriate management plans, including appropriate onward referral where the complexity is beyond their competence. If a majority of GDPs are unable to diagnose MIH, this represents a crisis in care for the one in six children with MIH in the UK. The care pathway for children referred into dental hospitals with MIH within the UK remains unreported and requires investigation in order to highlight the high treatment burden for patients within the NHS.

Chapter 3 - Aims & Objectives

Research Question

How are children with MIH managed in the UK?

Aim

1. *To understand how MIH is managed in primary and secondary care within the UK*

Objectives

1. *To assess the ability of GPs to diagnose, treatment plan and refer children with MIH*
2. *To assess how management in primary care may influence management in secondary care*
3. *To explore GPs knowledge and experience of management of MIH in primary care, including barriers to care*

Chapter 4 – Characteristics and Management of Patients Referred to Secondary Care with MIH – A Service Evaluation

4.1 Introduction

Service evaluations are part of the clinical governance activities used by health services to ensure health care is safe, adhering to standards, and consistently improving (Sally and Donaldson 1998). They can be used to assess performance and outcomes in a specific area of clinical practice or across a service. They differ from audit in that the findings are not tested against a set of pre-agreed standards or national guidelines, but can be used to identify issues within the area investigated (Twycross and Shorten 2014). In this instance a service evaluation was selected as an appropriate method to retrospectively analyse the care children with MIH receive within a hospital setting within the UK.

Although there are many published papers discussing treatment options for MIH, there are few published studies investigating the treatment children with MIH have actually received. Two studies by Jalevik and Klingberg, assessed treatment undertaken for children with MIH in the Swedish Public Dental Service, which comprises general, specialist and hospital paediatric services (Jalevik and Klingberg 2012, Jalevik and Klingberg 2002). It was not clear who carried out the treatment for this group of children, but it is likely that it comprised treatment over an extended period of time in both general and specialist dental services. No papers currently exist regarding management of children with MIH in a specialist hospital setting. Although the findings of this study relate only to this particular dental hospital in the UK, it is possible that similar patterns exist regionally and elsewhere in England.

This study was designed to investigate the characteristics of children with MIH who were referred into a hospital service, and the treatment they received for MIH prior to discharge back to primary care. In addition, information regarding previous management by the referrer, most often the GDP, would give a picture of what kind of patient, and what kind of treatment, is too complex for primary care locally in Merseyside and Cheshire.

The Commissioning Guide for Paediatric Dentistry sets out what kind of treatment is expected from GDPs (Tier 1), specialists and consultants (Tier 3). In addition, Tier 2 providers bridge the gap between general and specialist services, providing care requiring additional skills and facilities without need for specialist input. This care takes place both in primary care with GDPs and in community dental services (CDS). In England, it is expected that Tier 1 providers should manage preventive care, and Tier 2 providers should be confident to complete simple restorative treatment for children with MIH, but when the condition is more severe, referral would be appropriate (Office of the Chief Dental Officer England 2018). An additional intention of this work was to assess whether care of the appropriate level was taking place at this centre – e.g. Tier 2 and 3.

4.2 Aims & Objectives

Research Question

What is the current care pathway for patients with MIH referred to a dental teaching hospital?

Aim

1. *To evaluate the care pathway for children with MIH referred to Liverpool University Dental Hospital*

Objectives

- 1. To assess whether GDPs identified MIH in the referral and carried out treatment on affected teeth prior to referral*
- 2. To investigate the burden of care for patients with MIH in terms of treatment completed, appointment number and length of treatment plan in months*
- 3. To investigate the influence of baseline characteristics on appointment numbers and length of plan in years*

4.3 Method

This study was a retrospective service evaluation of the management of children with a diagnosis of MIH within the paediatric dentistry department of Liverpool University Dental Hospital (LUDH). Patient records for all children who attended consultant-led new-patient clinics in 2015 were requested, selected from an attendance log kept for the two consultants working at this time. Those children who failed to attend their new-patient appointment were not included. Although there was at least one additional weekly new patient clinic running at this time, no log was kept, and therefore these patients could not be assessed. Once the patient records were received, the new patient assessment proforma was checked for evidence of MIH as a diagnosis. Data was recorded only for those children with MIH. The service evaluation was registered with the local clinical governance team (Project no. 8410; RLBUHT, Liverpool Foundation Trust).

Study Population

All children with a diagnosis of MIH at their first new patient clinic appointment from 1st of January 2015 to 31st December 2015 were included. 2015 was selected as the best year to analyse as this would allow sufficient time for children to have completed a full course of treatment, as it was anticipated that a child could be referred at six years old, and not be discharged until FPM were removed at age 10 (if applicable). Demographic information collected was as follows:

- Age at referral
- Age at new patient clinic
- Dental age at extraction of FPM (early, ideal, late) (if applicable)
- Age at completion of treatment
- Sex
- First part of postcode

Study Procedures

A data collection sheet was developed and piloted within the paediatric dentistry department of LUDH. The following data was extracted from patient records:

- Specified reason for referral in letter
- Treatment on FPM/ incisors prior to referral
- Baseline anxiety (where recorded)
- Patient symptoms/ concerns e.g. sensitivity or aesthetics
- History of potential disruption to amelogenesis during pregnancy, birth or early years
- Number of teeth affected
- Severity of condition (mild or severe) based on EAPD definition (Lygidakis et al. 2010)
- Other diagnoses e.g. anomalies or caries affecting other teeth
- Treatment completed
- Whether full treatment plan complete or patient discharged before completion
- Number of appointments
- Number of cancelled or failed appointments
- Time span of treatment plan in months

An estimate of the frequency of referrals for patients with MIH out of the total referrals across one year was also made.

Data Analysis

A year was selected as a significant time period which would give a sample of several hundred children who had been referred into the hospital. This was large enough to ascertain whether any trends existed in terms of previous management by the referring practitioner. Although data collection did not include all new patient clinics, the sample still gives an idea of the frequency of referral to secondary care for patients with MIH in comparison to other referrals and the burden of care for the patient in terms of appointment number within the Merseyside and Cheshire area.

Descriptive statistics were used to explore the data. The Mann Whitney U Test for non-parametric data was used to compare the relationship between severity of MIH, and number of teeth extracted, number of appointments and length of treatment plan. All analysis was carried out using descriptive statistics in SPSS version 24 (IBM, Armonk, NY: USA). Significance was set at $p < 0.05$.

4.4 Results

Data collection took place from March to July 2019. 426 children attended new patient assessments on consultant led clinics in 2015. 29 patient records could not be retrieved by the researcher due to being lost or booked out for clinical activities. In total 397 patient records were checked for documentation of a diagnosis of MIH. 48 patients were found to have MIH – 12.1% of the sample.

Patient Characteristics

The following information relates to the 48 patients with a diagnosis of MIH. For one patient, only temporary records were found, and therefore some information regarding the initial referral and data from the new patient clinic are missing.

The mean age of patients at referral was nine years, two months (range four years eight months to 14 years four months) and nine years, seven months at new patient clinic appointment (range five years to 14 years 10 months). The mean age of patients at discharge was 10 years five months (range five years seven months to 15 years 11 months). 54.2% of patients were male (n=26).

Most children were from the Merseyside area. Liverpool postcodes accounted for 58.3% (n=28), Warrington postcodes accounted for 22.9% (n=11) and 10.4% (n=5) had a Wirral/ Cheshire postcode. The remaining children travelled long distances from Preston (n=2) Wigan (n=1), and Crewe (n=1).

Referral

The source of referral is recorded in the table 4.1

	Frequency	Percent
GDP	41	85.4
Restorative consultant	1	2.1
Orthodontist	1	2.1
CDS	4	8.3
Missing data	1	2.1
Total	48	100.0

Table 4. 1 Source of Patient Referral

Only 17.0% (n=8) of referrers stated MIH as a reason for referral. 66.0% (n=31) of referrers stated another enamel defect, most often 'hypoplasia', 36.2% (n=17) stated caries, and 10.6% (n=5) stated another reason for referral.

Treatment of MIH prior to referral, including fissure sealants, fluoride varnish, restoration, or extraction, had been attempted in 57.4% (n=27) of cases. Fluoride varnish was documented for 10 patients, and four teeth had been fissure sealed, 36 teeth had had a restoration placed (temporary or permanent), and seven teeth had been extracted.

Presenting Complaints

Most children presented with a complaint related to MIH (72.3% n=34). The most frequent complaint was of pain from MIH affected teeth (23.4% n=11). Mild sensitivity or aesthetic concerns both accounted for 19.1% of complaints (n=9). Severe sensitivity was a complaint in 10.6% (n=5) of cases followed by 'crumbling' teeth (6.4% n=3) and failing fillings (4.3% n=2). Patient anxiety was recorded for 35 patients. Of this group 57.1% (n=20) were recorded as having dental anxiety.

Details related to potential causes of disruption to amelogenesis were recorded in 42.6% of cases (n=20). These included potential factors which may have caused disruption during pregnancy (8.5% n=4), at birth (17% n=8) or as a child before the age of three years old (27.7% n=13).

Table 4.2 shows the number of affected molars and incisors. Most children had four affected molar teeth and no affected incisors. 33.3% (n=16) of patients had only molars affected, and 66.6% (n=31)

had molars and incisors affected. One patient only had a record of treatment and not diagnosis, as there had been partial loss of the patient records, so affected teeth were not recorded

		MIH incisors								Total
		0	1	2	3	4	5	6	8	
MIH molars	1	3	0	0	0	0	0	0	1	4
	2	8	2	2	1	0	0	0	0	13
	3	2	1	0	2	0	0	0	0	5
	4	3	6	9	3	1	1	1	1	25
Total		16	9	11	6	1	1	1	2	47

Table 4. 2 Number of MIH molars vs incisors

Table 4.3 shows the number of children with mild or severe MIH according to EAPD guidelines for severity (Lygidakis et al. 2010). Mild MIH includes children with mild sensitivity only, no caries, no PEB and only mild concern about appearance of teeth. Severe MIH includes children with sensitivity on brushing teeth or eating, toothache, caries, PEB and severe distress related to the appearance of their teeth.

	Frequency	Percent
Mild MIH	9	18.8
Severe MIH	39	81.3
Total	48	100.0

Table 4. 3 Severity of MIH (as per EAPD Guidelines)

Most children had an additional diagnosis alongside MIH (62.5% n=30). Caries in other teeth not affected by MIH was present in 43.8% of cases (n=21). Other dental anomalies were also present alongside MIH in 18.8% of cases (n=9).

Treatment Completed

Treatment was carried out at LUDH for 91.7% (n=44) of children. Some children had treatment completed with their GDP (8.3% n=4) and were discharged from the hospital with a treatment plan.

Table 4.4 shows the treatment completed for all FPM at tooth level

	Number	Percentage
Fissure Sealant	32	23.4
Flowable Composite	2	1.5
Composite	15	11.0
GIC	7	5.1
PMC	7	5.1
Extraction	49	35.8
Review & extraction later	25	18.2
Total	137	100.1

Table 4. 4 Treatment completed per molar

Table 4.5 shows the number of patients who had treatment with LA /behaviour management, sedation, or GA. Some patients may have had more than one treatment modality – e.g. sedation for fillings and GA for extractions.

	Number	Percentage
Behaviour management or LA	18.00	38.3
Sedation	10.00	20.8
GA	20.00	42.6

Table 4. 5 Treatment adjuncts used

Only 18.7% of patients required aesthetic treatment (n=9). Table 4.6 shows the treatment completed for incisor teeth. All results are expressed at patient level. No patients had vital bleaching, lab-made composite veneers or porcelain veneers.

	Number	Percentage
Microabrasion	5	55.6
Direct Composite Veneer	2	22.2
Localised Composite restoration	2	22.2

Table 4. 6 Aesthetic treatments per patient

For the 25 patients who had extraction of one or more FPM teeth, the mean age at time of the first extraction was 10 years 3 months (7 years 5 months to 14 years 9 months). The best time to extract FPM for optimum space closure is reported to be from eight to 10 years (Cobourne et al. 2014).

Sixteen patients had extractions in this period, two had the extractions early, seven had the extractions late and two had the extractions later than the normal 'ideal' time but as part of an orthodontic plan to avoid the loss of healthy premolar teeth.

The treatment completed and presenting complexity of the patient was assessed to categorise treatment into Tier 1, 2 or 3 care according to the Commissioning Guide for Paediatric Dentistry in England (Office of the Chief Dental Officer England 2018). The majority of patients (91.7% n=44) needed treatment at a Tier 2 or 3 level, with only 6.3% (n=3) requiring Tier 1 care. A further patient (2.1% n=1) required Tier 1 care for management of MIH but was referred for management of multiple carious primary teeth which could not be managed effectively without GA.

Outcomes of Course of Treatment

Treatment was completed in 79.2% (n=38) of cases. The remaining patients (20.8% n=11) were not brought to at least two consecutive appointments and were discharged back to their GDP for completion of the plan. One patient was referred a second time and then discharged a second time due to further missed appointments. A further 10.4% (n=5) of patients were discharged, referred again and completed their treatment at the second opportunity.

The median number of appointments necessary to complete treatment was four (range 1-16). The median treatment time from first new patient assessment to completion of treatment was 7.5 months (range 0-48). The number of missed appointments ranged from zero to six, with a median of zero. Most patients did not miss any planned appointments (61.7% n=29).

Analysis

The Mann-Whitney-U Test was used to assess the relationship between severity of MIH and the treatment length in months from first (new patient consultant appointment) to last appointment before discharge, total number of appointments and also number of extracted teeth. There was a significant relationship between number of appointments and MIH severity ($p=0.015$) but not length of plan (in years) ($p=0.92$). There was also a significant relationship between severity of MIH and number of teeth extracted ($p=0.014$). Calculation tables are shown in appendix 1.

4.5 Discussion

This study set out to investigate the current care pathway for patients with MIH referred to a hospital setting at LUDH. A service evaluation was designed to collect data regarding management prior to referral, presenting characteristics and management whilst at LUDH, for all paediatric patients attending consultant led new patient clinics with a diagnosis of MIH in 2015. No previously published studies have looked exclusively at the management of children with MIH in a hospital setting. Taylor et al. investigated the management of poor prognosis FPMs using clinical vignettes. U.K.-based GDPs and specialists in paediatric dentistry were asked to provide a treatment plan for children with poor prognosis FPMs. They found that GDPs would prefer to restore FPMs in comparison to specialists, who were more likely to extract these teeth (Taylor et al. 2019). Jalevik and Klingberg followed care for a group of children with severe MIH over 10 years and compared their treatment outcomes to controls (Jalevik and Klingberg 2002, Jalevik and Klingberg 2012). Those with severe MIH were more anxious, had higher DMFT and had treatment of FPMs 4.2 times as often as the non-MIH controls.

Most referrals were made by GDPs, followed by 8.3% from the CDS. The patients referred from the CDS may have been referred following unsuccessful treatment within the community setting. In this dental hospital additional treatment management adjuncts such as intravenous sedation and GA for comprehensive treatment are available, in addition to inhalation sedation and extraction only GA

lists which are also found in the CDS. Only 17% of clinicians referred their patient specifically to manage MIH, with most referring more generally for management of an enamel defect, most commonly 'hypoplasia'. Confusion between hypomineralisation and hypoplasia appears common, and was addressed by a recent paper by Patel et al, who discussed the differences in appearance and management approaches for these two enamel defects (Patel et al. 2019). The second study of this project will investigate this further by addressing accuracy of GDPs when diagnosing MIH.

Over half of referred patients had some treatment carried out on MIH affected teeth prior to referral. The true figure is likely to be higher, as data was taken from information recorded in the referral letter and patient assessment, which also relies on patient and parent recall of events. The data regarding previous restorations is likely to be more accurate as this is charted reliably for every patient. This information provides a limited review of previous dental history, as children may have had multiple restorations placed on the same tooth, and previous preventive care may be regular and appropriate, or sporadic. This care is also related to patient attendance which is outside the control of GDPs. Prospective research based within primary care would give a clearer view of actual patient care.

The majority of patients were symptomatic due to MIH, most frequently complaining of pain, aesthetic concerns or mild sensitivity. Just over half of the patients had dental anxiety, which is similar to normal population levels for children in the UK. (NHS Digital 2015). Jalevik and Klingberg found that children with MIH were more anxious than controls with caries (Jalevik and Klingberg 2002); however, a study of school children in Brazil found that there was no difference between children with MIH and controls (Menoncin et al. 2019). The children in the Brazilian study were mostly pre-treatment and had a range of severities of MIH, whereas the children in Jalevik and Klingberg's study had already undergone treatment for severe MIH, which may explain the difference.

The most common reason for disruption to amelogenesis was illness in the first three years of the child's life. Potential disruption to amelogenesis was recorded for less than half of patients, therefore the conclusions that can be drawn are limited. Studies investigating aetiology are best undertaken as prospective birth cohorts of at least 1000 participants, as the memory of parents cannot be robustly relied upon (Elfrink et al. 2015).

Most children had severe MIH (81.3%) according to the EAPD definition (Lygidakis et al. 2010). Most children with severe MIH required either aesthetic treatment, or restoration and extraction to manage FPM with caries, PEB, or severe sensitivity. Most children referred had four affected FPM teeth, and zero to three affected incisors, with both affected molars and incisors (66%). It has previously been reported that the greater the number of affected teeth, the more severely affected the teeth tend to be in terms of hypomineralisation (Lygidakis et al. 2008), which fits with these results. In addition, Walshaw et al found that 29% of children with MIH had a second anomaly when they reviewed the orthopantomogram (OPG) of 100 consecutive MIH patients referred to a secondary care hospital in England (Walshaw et al. 2020). In this study 19% of children had a second anomaly recorded in the notes, however, there is the possibility that some anomalies may not have been recorded.

Almost all patients had treatment completed at LUDH (91.7%), with 8.3% being discharged back to their GDP with a treatment plan. 137 molar teeth underwent treatment, with the most common treatment being extraction (35.8%) or review and extraction later (18%). The number of FPMs extracted was significantly related to severity of MIH. Although extraction of FPM in children can sometimes be straightforward, patient anxiety may necessitate referral for behaviour management, or pharmacological adjuncts. Cobourne recommends that the opinion of a specialist in orthodontics or paediatric dentistry is sought if there is a delayed approach (to address a class II or III

malocclusion), as compensating extractions or GA are often necessary (Cobourne et al. 2014). In this study many providers referred for guidance over extraction of poor prognosis FPMs. It is worth noting that 72% of children in this cohort had extraction of FPMs at the 'ideal time', indicating that most providers sought an opinion at the optimum time. The mean age at extraction was 10 years and 3 months. In contrast, a multicentre study conducted in 2007 found that the mean age of extraction of poor prognosis FPM was 11 and a half years at LUDH (Albadri et al. 2007), which is well beyond the ideal time of eight to ten years. This preceded national guidelines on planned loss of poor prognosis FPM teeth, and indicates that the guidelines have been successful in promoting referral at a better age (Cobourne et al. 2014).

Most patients had treatment with GA (42.6%), followed by LA and/or behaviour management (38.3%). A previous multicentre study looking at extraction of poor prognosis FPM teeth found that 55% of patients required GA for extractions at LUDH followed by 43% of children who had extractions with LA (Albadri et al. 2007). This study found more children had treatment with sedation, which is most likely because all treatments including restorations were included. GA for paediatric patients can only be carried out in a hospital setting after planning, ideally, by a specialist or consultant in paediatric dentistry (Davies 2008), and therefore the need for treatment with GA alone necessitates referral from primary care.

Very few patients underwent aesthetic treatment (18.7%). In 2018, Large et al carried out analysis of children who were referred to secondary care in the North of England with MIH for management of incisor opacities (Large et al. 2020). They found that 38% were unhappy with the appearance of their teeth, 24% were bullied because of their teeth and 10% felt self-conscious. The difference between studies can be explained by the fact that their sample excluded children who required treatment on FPMs. Hasmun et al discussed the anecdotal perception amongst paediatric dentists that there are

increasing numbers of children seeking corrective treatment due to distress surrounding incisor opacities (Hasmun et al. 2018). With the rise of social media and pressure on appearance, it is possible that there may be a greater number of children requesting aesthetic treatment in 2021.

Most children completed their treatment plans (89.6%) and did not miss any appointments (61.7%). The median number of appointments necessary to complete treatment was four, which reflects that many children were referred for extraction of poor prognosis FPM teeth under GA and therefore had a relatively short plan to complete treatment. Total treatment length to manage MIH may be longer, considering some of these children may have had restorative work to stabilise teeth with their own GDP prior to referral. The number of appointments ranged from one to 16. This reflects the spectrum of severity for children with MIH, with some only requiring a single appointment to create a treatment plan suitable for their GDP to complete, and with others attending multiple appointments for restorative stabilisation of FPM, aesthetic treatment and eventual extraction of FPM.

The results of this study can be compared with recommendations from the British Society of Paediatric Dentistry (BSPD) and the paediatric dentistry commissioning guide (Office of the Chief Dental Officer England 2018, British Society of Paediatric Dentistry 2020a). BSPD recommend that the majority of children with milder forms of MIH should be managed in primary care and that only those with severe MIH should be referred. The paediatric dentistry commissioning guide also recommends that moderate and severe MIH should be managed by Tier 3 providers (specialist or consultants). The use of GA or sedation as an adjunct is also considered a Tier 2 service, which is likely to occur in community and hospital settings. The results from this study show that this current guidance was being adhered to by most referring GDP in 2015, with only 6.3% of children requiring

Tier 1 care. In addition, many GDP had placed temporary or permanent restorations on FPMs prior to referral, which indicates they were working at a Tier 2 level.

The findings of this study are reassuring as it appears that appropriate referrals are being made in most cases. In addition, children have relatively short treatment plans once in secondary care, usually involving the use of GA, before being discharged back to their primary care provider.

Although a small number of patients were referred specifically for MIH, this has not disadvantaged them in general as most children had extraction at the ideal time, which is the most time critical element of an MIH plan.

Study Limitations

There are several weaknesses to the methodology of this study. A retrospective service evaluation was selected as the most time-efficient way of collecting data; however, a prospective approach may have reduced the amount of missing data, both from omitted details in the records and also from paper patient records that could not be located. The benefit of looking at data from 2015 meant that this allowed sufficient time for all patients to have finished their treatment or have been discharged. Unfortunately, due to the limited time period for completion of this doctorate programme, it would not have been possible to prospectively collect data. For example, one patient took four years to complete treatment, illustrating the timeframe necessary. Another consequence of retrospective data collection meant that not all new patient records were reviewed, as only certain clinics are logged. Although the majority of patients attend consultant led new patient clinics, other new patient clinics do exist. These mainly deal with children referred with dental caries requiring GA, but it is possible that some children on these clinics may also have MIH.

Another weakness of the methodology is the small sample size and the fact that data was only collected from one centre in the north of England. This limits the extrapolation of findings more generally. Further data collection could be carried out at other dental hospitals to explore the management of MIH more generally across the whole U.K., giving more robust results with increased external validity. This service evaluation could also be repeated in this centre, allowing for an assessment of local trends in referral and management.

These patients started their treatment six years ago, and although it was necessary to look this far back in order to ensure all children had completed their treatment plans, the picture regarding referral may have changed significantly since then. Across the North-West of England, demand for community and hospital-based paediatric dentistry has increased significantly, leading to an increase in referral-to-treatment times. The ability for GDPs to refer at the right time has become more complex, as they try to factor in potential delays the child may experience before being seen and whilst awaiting treatment. Additionally, although there was little demand for aesthetics in 2015, in the Instagram® era, it is possible that more patients are now referred for aesthetic management to improve their quality of life.

Further Research

The findings of this study pose further research questions. Due to the retrospective design, the referral practices of GDPs and the management in secondary care may have moved on since 2015. The anecdotal increases in patients requesting aesthetic treatment, and total number of referrals could be investigated. Is MIH identified prior to referral more often, and do referrals continue to be appropriate? These questions could be answered by repeating this service evaluation and comparing the two years. This study only tells half a story, in that the results only reflect those cases GDP refer, and do not demonstrate whether appropriate management has taken place in primary care. Another aspect that could be explored is the experience of managing children with MIH in primary care, and

the barriers that may exist making this challenging. The second and third studies will answer these questions.

4.6 Conclusion

The findings of this study indicate that although most referrers did not explicitly diagnose MIH prior to referral, the majority identified an enamel defect or the presence of caries. Despite this, children appeared to be referred appropriately and at the correct time, which meant those who required extraction were able to have this carried out at the best time to facilitate eruption of the second permanent molar into a good position.

The treatment carried out prior to referral, and within the dental hospital demonstrates that children were receiving care at the appropriate level in most cases, according to the Commissioning Guide for Paediatric Dentistry in England (Office of the Chief Dental Officer England 2018). In addition, most of the children treated had severe MIH which is recommended to be managed by specialists and consultants in paediatric dentistry. In general, the findings are positive and demonstrate appropriate referral by the majority of GDPs and referrers in the Merseyside area.

In terms of treatment carried out, GA was the most common modality and extraction was the most common procedure. This fits with the findings that most children had severe MIH affecting four FPMs, and therefore these poor prognosis teeth would be best lost at the optimum time to allow space closure with the second molar and premolar. Length of plan in months was highly variable due to the differences in presenting features, and treatment required. Multicentre or comparative studies within this setting could build on the findings of this small-scale study.

Chapter 5 – Diagnosis and Management of MIH by GDPs – A Vignette Survey

5.1 Introduction

Evidence to date has shown that GDPs around the world, including in the UK (Kalkani et al. 2016), do not always feel confident in managing children with MIH. This evidence comes from self-report surveys, which can be prone to reporting bias where participants wish to exhibit socially acceptable responses, including potential exaggeration of confidence. In addition, confidence may not always correlate with actual competence at a task. A vignette survey was designed to investigate GDPs skill in diagnosis and treatment planning children with MIH.

Vignette studies are a valid tool to assess clinician decision making practices, such as diagnosis and treatment planning, which are complex cognitive and behavioural processes. A well-designed vignette combines some elements of experimental research with the high external validity of traditional surveys. A criticism is that a vignette cannot faithfully recreate the true clinical situation, however, in general they are more feasible to carry out and the results more reliable than the alternatives (observation or self-report)(Evans 2015).

A systematic review of vignettes in healthcare found that most recent medical vignettes assessing diagnosis and treatment planning had between three and 130 vignette scenarios (Bachmann, 2008). Although there is no clear consensus on the number of vignettes that should be used in the literature, the author recommended that no more than 20 vignettes are used in a single study, as the reliability of the participant answers will be reduced as they become fatigued. It is also recommended that no more than 6 to 8 vignette attributes are used for the same reason. Choice

based answers lead to less bias than ranking or rating based answers, as the latter make more assumptions about human cognition (Bachmann et al. 2008).

Clinical photographs are a valid tool for use in studies that assess diagnosis of enamel defects and have been used in a number of studies to date. Elfrink et al carried out a study to assess whether HSPM and caries could be accurately diagnosed from clinical photographs (Elfrink et al. 2009). They found that the sensitivity of assessing caries using intraoral photographs was 85.5%, and the specificity 83.6%, whilst for HSPM the sensitivity was 72.3%, the specificity 92.8%. The inter-observer agreement yielded Cohen's Kappa scores of 0.76 for caries and 0.62 for HSPM. They concluded intra-oral photographs were valid for use in studies requiring diagnosis of caries or MIH. A study to assess the ability of GDP in India also used a survey format and clinical photographs to assess their ability to identify different developmental defects of enamel. They found that GDP required further training in order to differentiate between the different clinical presentations of numerous conditions (Dabiri et al. 2018). Other studies have also successfully used clinical photographs to diagnose MIH and HSPM for the purpose of estimating prevalence, such as a birth cohort study which investigated the relationship between MIH and HSPM (Elfrink et al. 2012).

The relevance of this study is that no other vignette studies had been carried out to assess the ability of GDPs to accurately diagnose MIH in the UK, or elsewhere. In addition, no studies had assessed treatment planning at patient level, rather than at tooth level, or explored referral practices for children with MIH within the general dental service.

5.2 Aims & Objectives

Research Question

How do GDPs in the UK diagnose and treatment plan children with MIH?

Aim

1. *To assess the ability of GDPs in the UK to correctly manage children with MIH (and HSPM).*

Objectives

1. *To assess the ability of GDPs in the UK to correctly diagnose MIH and HSPM based on symptoms and clinical photographs*
2. *To assess practises in treatment planning for children with MIH when presented with radiographs, photographs and clinical histories*
3. *To investigate when GDPs choose to refer patients with different presentations of MIH*
4. *To evaluate the self-reported confidence of UK based GDPs in the management of children with MIH*

5.3 Method

This study was a cross-sectional electronic vignette survey of GDPs across the UK. Ethical approval was granted from the University of Liverpool Ethics Committee (project number 4561). Participants gave consent for participation at the start of each survey.

Participants were GDPs practising in England, Wales, Scotland and Northern Ireland. GDPs who did not treat children and those who were on a specialist register were excluded. Specialist only practices (e.g. orthodontic practices) were not contacted. Practices who did not have contact details available on a public facing website were excluded.

Demographic information collected included:

- Postcode area for dental practice e.g. L for Liverpool
- Age
- Gender
- Year of graduation from undergraduate dental programme
- Location of study for undergraduate dental degree e.g UK & Ireland, EU or rest of the world
- Postgraduate qualifications
- Experience of working within a paediatric dentistry department after graduation
- Whether they currently work within a paediatric dentistry department part time
- An estimate of how often they treat patients under 16-years-old
- Case mix e.g. NHS, private or mixed

Recruitment

A sample size of 400 GDPs was selected based on a sample size calculation that approximately 50% of GDPs would accurately diagnose MIH. This was based on the assumption that those who were

confident at diagnosis would also be accurate (Kalkani et al. 2016). This would provide confidence intervals of $\pm 5\%$, whilst a response rate of 100 would give a confidence interval of $\pm 10\%$.

Strategy one –

The numbers of GDPs contacted from each nation was weighted according to the number of GDPs on the GDC register as of July 2018. A freedom of information request to the General Dental Council in July 2018 revealed that 81.5% of GDP worked in England, 10.2% worked in Scotland, 4.2% worked in Wales and 4.1% worked in Northern Ireland. This equated to 326 English GDPs, 41 Scottish GDPs, 16 GDPs from Northern Ireland and 17 from Wales to meet the sample target.

The UK has 121 postcode areas, with 97 postcode areas in England, 16 in Scotland, 1 in Northern Ireland and 7 in Wales. A random sequence of postcode areas was generated for each nation using a random number generator and an online postcode generator was utilised to randomly generate full postcodes (Bell). This postcode was inputted to the relevant national website (Care Quality Commission (England), NHS24 (Scotland), Health & Social Care (Northern Ireland) and Health in Wales) in order to find the closest dental practice to the generated postcodes. The practice website was visited, and the selected GDP was sent an invitation to participate via the publicly available practice email address. A participant was selected by allocating a number to each GDP as listed on the website, and then using a random number generator to select the number. Where only one GDP was listed, this participant was automatically selected.

Each GDP was targeted by email on two occasions, two weeks apart. An invitation email was sent to the selected GDP via the practice email address. Each email explained the nature of the study, contained a hyperlink to the survey and had participant information attached.

Strategy two –

Strategy one yielded a low response rate, and therefore a second method of recruitment was utilised. This was accepted as an amendment to the original ethics application. A link to the survey and participant information was posted and shared on social media dental groups, including Facebook and Twitter. This in turn led to snowballing of recruitment, with other members of Facebook sharing the survey.

Study Procedures

Qualtrics Survey Software (SAP, Utah) was used to build an electronic vignette survey. GDP were informed that the study was about management of dental hard tissue defects in children but weren't specifically told that it was about MIH, to avoid response bias. The study was designed with 10 vignettes in order to give a variety of presentations of MIH without the survey becoming too arduous for participants. In addition, only two cases required treatment planning to reduce response fatigue.

Diagnosis section -

Ten clinical vignettes were selected after searching the available archived clinical photography database at LUDH. Only anonymised clinical photographs of children with MIH with appropriate

consent (level 4: consent for publication) were included. A shortlist of cases was created to ensure a spectrum of MIH presentations, and that the images were of sufficient diagnostic quality. Cases included: mild MIH (molars only), mild MIH (incisors and molars), HSPM, MIH and HSPM, severe MIH with PEB of molars, and severe MIH with caries on FPM. Control cases demonstrated: caries, fluorosis, amelogenesis imperfecta, and dentinogenesis imperfecta. The final cases selected were reached by consensus opinion of consultants in restorative and paediatric dentistry, and a speciality trainee in paediatric dentistry.

Each set of clinical photographs were supplemented by a description of the patient's complaint and relevant history, to aid diagnosis. Participants were asked to give one or two 'hard tissue' diagnosis for each case, as appropriate. They did not need to specify affected teeth. Data was collected as a free-text response.

The cases were quality assured by members of CONNECT (Child Oral health National research Collaborative) to ensure diagnosis was clear for all cases and revised in order to improve the vignettes' clarity, and validity. The survey was then piloted within LUDH, and amendments made based on feedback from visiting GDP staff before finalising the version to be disseminated.

Treatment Planning Section –

Two of the vignettes from the diagnosis section of the survey were presented with more detail in order to allow treatment planning. Clinical photographs, bitewing and OPG radiographs were included, in addition to descriptive text (figures 5.1 – 5.6). Both children were considered high caries risk due to MIH, however in vignette two, the child also had active caries.



Figure 5. 1 Clinical photographs vignette one. Text read: This patient is an eight-year-old boy. He gets teased at school because of the 'dirty' patches on his front teeth and would like them to look better. He doesn't have any other complaints. He is fit and well



Figure 5. 2 Bitewing radiographs vignette one

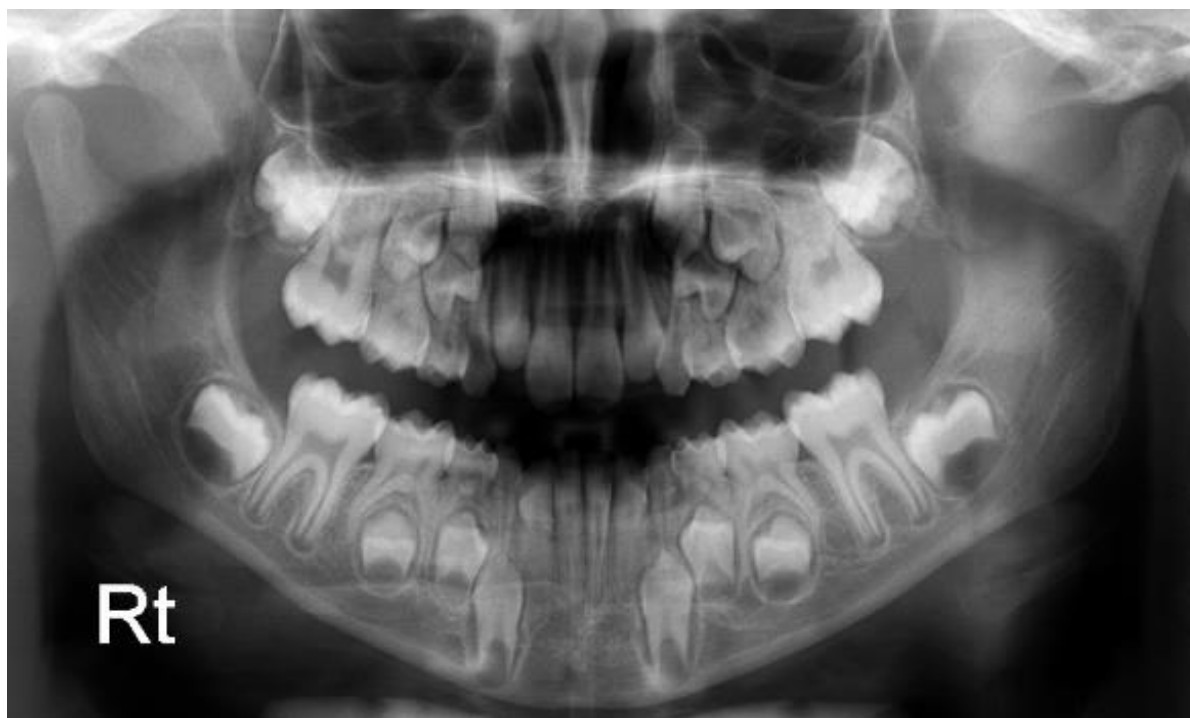


Figure 5. 3 OPG vignette two



Figure 5. 4 Clinical photographs vignette two. Text read: This is a seven-year-old girl. She tells you she doesn't like the dentist because the water and air hurt her teeth. Sometimes ice cream hurts her back teeth. Although Mum is concerned about the patches on her front teeth, the patient herself is not bothered about appearance. Fit and well with no allergies. Mildly anxious but potentially cooperative.



Figure 5. 5 Bitewing radiographs vignette two



Figure 5. 6 OPG vignette two

GDP were asked to provide treatment plans from set options for preventive care, management of FPM and management of anterior teeth. Participants were not informed that the patient had MIH, but this could be deduced from the scenarios.

Options for prevention included recall interval, fluoride varnish frequency, and diet advice/ oral hygiene instruction interval. For management of posterior teeth participants selected a treatment option for each FPM e.g. actively monitor, fissure seal GIC, fissure seal resin, restore composite, restore GIC, restore PMC, extract. For management of anterior teeth participants were asked whether they would carry out aesthetic work or not. Those who opted to treat then selected from microabrasion, bleaching, direct composite veneers, localised composite with hard tissue removal or porcelain veneers.

Following treatment planning for each case, participants were also asked:

‘What treatment would you feel competent to carry out in your own practice?’

‘If you would refer for some of the treatment, what are the important factors influencing your decision?’

‘What dental speciality would you refer the patient to?’

Self-reported Confidence Section –

The final section assessed the self-reported confidence and opinions of GDPs regarding the management of children with MIH. Questions included:

- How confident are you in diagnosis of MIH?

Very confident – Confident – Slightly confident – Not confident at all

- How confident are you in diagnosis of HSPM?

Very confident – Confident – Slightly confident – Not confident at all

- Were you aware that HSPM put children at increased risk of having MIH?

Yes / No

- Do you feel MIH should be managed in general practice or by specialists?

Always by GDP – Shared care GDP and Specialist – Always Specialist

- Do you feel confident in treatment planning for children with MIH?

Very confident – Confident – Slightly confident – Not confident at all

- Would further training in the management of MIH at undergraduate and postgraduate level be useful?

Yes/ No

Participants were not able to change their answers in previous sections once they entered the next section. This was to maintain validity by preventing them amending answers in the diagnosis section after reaching the final section where they were asked specific questions about MIH.

Data Analysis

Thematic analysis using a systematic inductive approach was used to analyse the data within the free-text response questions (Braun and Clarke 2006). A random sample of 20 responses were coded to develop a working coding framework. This was then tested against the remaining responses to

ensure the validity of the coding and to add further codes as necessary. Themes were developed from the agreed codes. Descriptive statistics were used to analyse the frequency of themes identified in the diagnosis section, the treatment planning section, and the self-reported confidence section. The Mann Whitney U Test was used to analyse the relationship between self-reported confidence and accuracy in diagnosis and treatment planning. Pearson's coefficient was used to assess correlation between accurate diagnosis and acceptable treatment planning.

5.4 Results

Data was collected electronically between 11th February and 14th May 2019 using Qualtrics survey software. Fifty-seven GDP fully completed the survey, and a further 19 GDP partially completed the survey to the end of the diagnosis section. Response rate cannot be estimated since the survey was shared publicly, and therefore the total sample size is unknown.

Demographics

Of the 76 GDPs, most were in the 20-29 age group (38.2% n=29), 35.5% were in the 30-39 age group (n=27), 18.4% were in the 40-49 age group (n=14), 6.6% were in the 50-59 age group and 1.3% were in the 70+ age group (n=1). No participants were in the 60-69 age group. 68.4% of GDPs were female (n=52). Table 5.1 shows when participants graduated

Year of Graduation	Percent	Number
2011-2018	54.0%	41
2001-2010	29.0%	22
1991-2000	9.2%	7
1981-1990	6.6%	5
1971-1980	0.0%	0
1970 or earlier	1.3%	1

Table 5. 1 Year of graduation from undergraduate dental degree

Most respondents worked in England (80.26% n=61), with 17.10% from Scotland (n=13), and 1.32% from both Wales (n=1) and Northern Ireland (n=1). In comparison to the total population of GDPs within the UK, 81% work in England, 10% work in Scotland, 4% work in Northern Ireland and 4% in Wales. In England 37 postcode areas were represented out of a potential 97. In Scotland seven postcode areas were represented out of 16, and in Ireland and Wales one postcode area was represented out of one and seven areas respectively. Appendix 2 shows a table with all represented postcode areas.

The majority of participants completed their undergraduate dental degrees in the UK (90.8% n=69), whilst 2.6% (n=2) and 6.6% (n=5) completed undergraduate degrees elsewhere in Europe, and the rest of the world. Around half of participants had postgraduate dental qualifications (54.0% n=41). Only 18.4% (n=14) of participants stated that they had experience of working within a paediatric

dentistry department after graduation and 5.3% (n=4) currently worked part time within a paediatric dentistry department.

The estimated number of children under 16 years old that each GDP saw per week ranged from one to 60. One GDP expressed the amount as a percentage and therefore the median, which was ten patients, was calculated for the 75 GDPs who expressed this result as a number. Figure 5.1 shows the case mix of each participant.

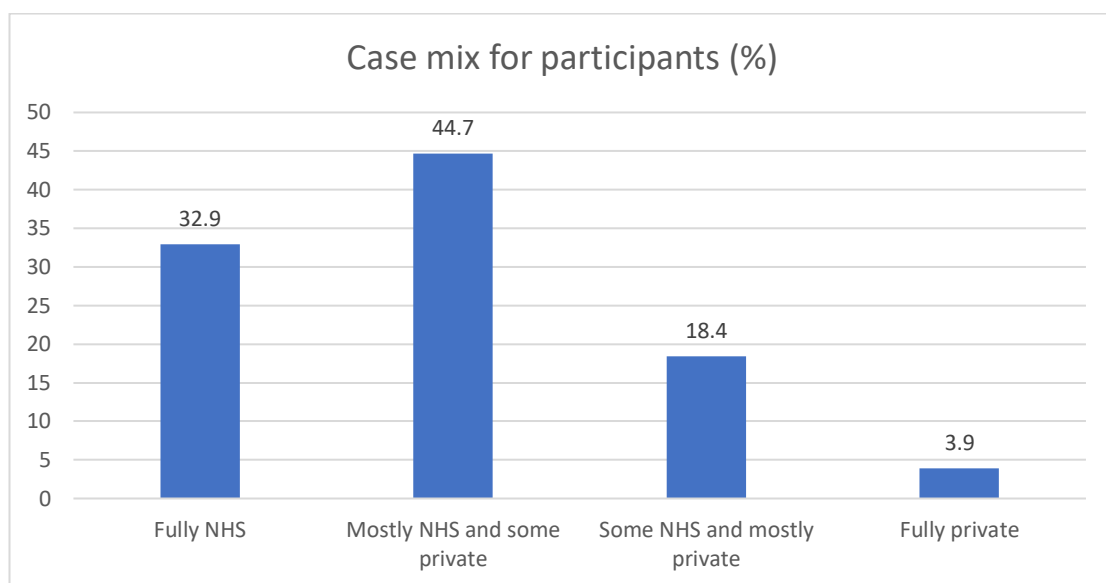


Figure 5. 7 Case mix for each GDP

Diagnosis Section

Seventy-six GDP completed this part of the survey, and this was analysed qualitatively and quantitatively. The data was first analysed using thematic analysis with an inductive approach, and then the data was analysed quantitatively using the themes selected. Only the six MIH/HSPM cases with seven possible correct diagnoses were analysed. Results from the four control cases were not explored.

Qualitative

Thematic analysis of the answers in the diagnosis section was undertaken. This was carried out by the lead researcher by combining all answers for each question into a single document, analysing the answers into codes of similar answers and then deciding on a framework and themes to describe the data. This was intended to be surface level analysis and descriptive in nature given that most answers were single words. Three main themes and five subthemes were identified, based on the type of answer given.

The hypomineralisation/hypoplasia group contained diagnoses which included the correct diagnosis of MIH or molar-incisor-hypomineralisation, but also diagnoses which could be considered partially correct such as 'hypomineralisation'. Hypoplasia was considered in this group as some GDPs gave the diagnosis 'molar-incisor-hypoplasia', and therefore it cannot be guaranteed that those using the acronym 'MIH' are using the term correctly. The second theme was 'other dental hard tissue defects' which included other enamel developmental defects such as amelogenesis imperfecta but also acquired 'defects' such as toothwear and caries. The third theme was other diagnoses, which were not dental hard tissue diagnoses, which included diagnosis of odontogenic infection and comment of orthodontic need or malocclusion. Figure 5.8 shows the themes, subthemes and codes identified.

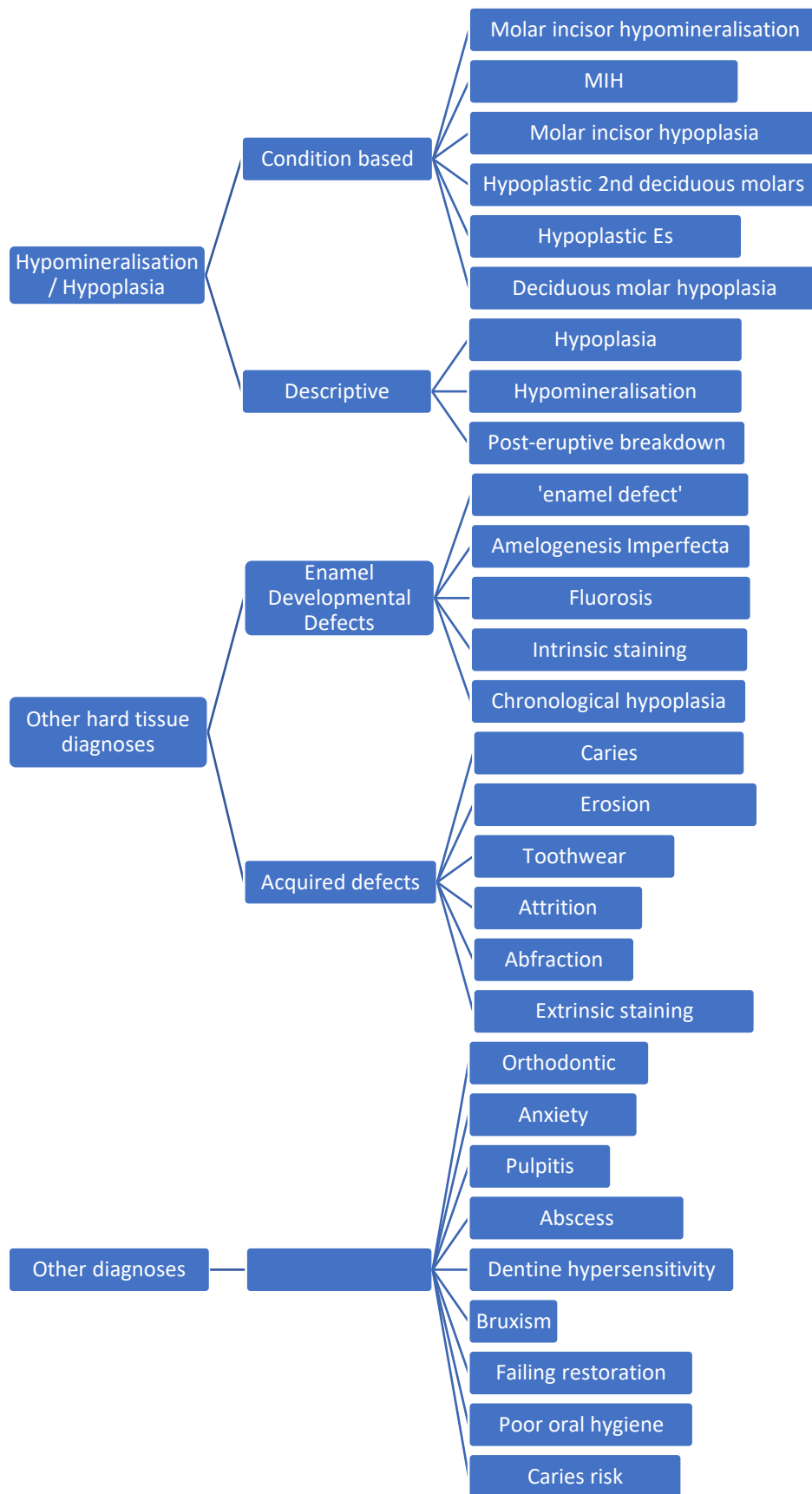


Figure 5. 8 Themes, subthemes and codes identified from diagnoses

Hypomineralisation/ Hypoplasia

Condition based

Hypoplasia was commonly confused with hypomineralisation. Where the term MIH was used, it was not always clear whether the GDP understood that 'h' was for hypomineralisation. Where hypoplasia was used it was not obvious whether this was because the GDP thought that the case showed hypoplasia or whether GDP use the term interchangeably to mean both hypomineralisation and hypoplasia.

'Hypomineralisation of 6's and hypoplastic incisors'

(Q4.8; line 44)

'Hypoplastic enamel could be MIH'

(Q4.8; line 20)

When considering HSPM affected teeth, the terminology was varied. Many GDP chose to use the term MIH, although this only applies to the condition affecting permanent teeth. Again, like for permanent teeth, hypoplasia was used frequently in place of hypomineralisation. Description of aetiologies demonstrated knowledge of potential causes of the condition.

'MIH possible febrile illness during developmental stages of Es and 6s and 1s'

(Q4.6; line 36)

Descriptive

Some GDP did not use condition type diagnoses but identified that there was either hypomineralisation or hypoplasia. Some showed increased knowledge of aetiology and disease process, for example using the term 'post-eruptive breakdown', which suggests knowledge that hypomineralised teeth can breakdown and fracture under normal occlusal forces or commenting on disruption of amelogenesis.

'Hypoplastic 6's? Systemic disease during development of the 6's'

(Q4.6; line 21)

'Molar hypomineralisation with post-eruptive breakdown'

(Q4.6; line 29)

Other Hard Tissue Defects

Enamel Developmental Defects

Some GDPs identified that the appearance was not that of caries but were not familiar with the appearance of MIH and diagnosed different enamel developmental defects. This demonstrates potential error in diagnosis, or possible lack of awareness of MIH.

'Fluorosis'

(Q4.2; line 15)

Acquired defects

Other GDPs gave hard tissue diagnoses for more commonly seen disease such as caries and toothwear. These may be participants who were not familiar with appearance of hypomineralisation or PEB, and therefore diagnosed the discolouration and defects as a more familiar oral disease.

‘Caries in deciduous teeth 55, 75, 85. Possible caries in 16 and 46. Restored 65 with possible secondary caries. Non cariogenic tooth surface loss 64’

(Q4.4; line 11)

Other diagnoses

Some GDP may have misread the question or may have found it difficult to make a hard tissue diagnosis, and therefore gave alternative, potentially correct diagnoses based on other information seen in the photos such as malocclusion or symptoms described in the text, such as sensitivity.

‘Dentine hypersensitivity, abfraction’

(Q4.10; line 4)

‘Bruxism’

(Q4.10; line 48)

Quantitative

As part of the qualitative analysis the answers given were coded into categories and quantitative analysis was undertaken based on the groups of answers given as shown in 5.2. Where a participant put more than one answer, the answer which was 'best' was recorded using a hierarchical scale with 'correct' the best and 'other diagnoses' the worst. Therefore, if a GDP correctly diagnosed there was caries but not MIH, they would be in the acquired hard tissue defect group, whereas a GDP who diagnosed MIH and caries, would be in the correct group. Likewise, a GDP who diagnosed amelogenesis imperfecta and dentine hypersensitivity would be in the other enamel developmental defects group.

Category	Examples
Correct	Molar incisor hypomineralisation (MIH), hypomineralised second primary molars (HSPM), deciduous molar hypomineralisation (DMH)
Hypomineralisation/Hypoplasia (Hypo)	Hypoplasia, hypomineralisation, molar incisor hypoplasia, hypocalcified
Other enamel developmental defects (EDD)	Amelogenesis Imperfecta, fluorosis, chronological hypoplasia, intrinsic staining
Acquired hard tissue defects (Acquired)	Caries, erosion, tooth wear, attrition, abfraction
Other diagnoses (Other)	Anxiety, dentine hypersensitivity, pulpitis, abscess, poor oral hygiene, bruxism, caries risk, orthodontic IOTN, integrity of restorative work

Table 5. 2 Categories of diagnosis

Figures 5.9. to 5.14. show each diagnosis questions involving MIH / HSPM. Tables 5.3 to 5.8 show the corresponding results for each question. Figure 5.15 demonstrates a comparative bar graph demonstrating all answers for each question.

Q4.2.



This patient is 8 years old. He gets teased at school because of the dirty patches on his front teeth.

Figure 5. 9 Mild MIH molars and incisors

	N	%
Correct	50	65.79
Hypo	19	25
EDD	7	9.21
Acquired	0	0
Other	0	0
Total	76	100

Table 5. 3 Mild MIH molars and incisors

Q4.4.



This patient is 9 years old. He is asymptomatic.

Figure 5. 10 Mild MIH molars only (caries on primary molars)

	N	%
Correct	3	3.95
Hypo	5	6.58
EDD	2	2.63
Acquired	66	86.84
Other	0	0
Total	76	100

Table 5. 4 Mild MIH molars only (caries on primary molars)

Q4.6.



This 9 year old girl has no complaints but is very anxious and jumpy in the dental chair. There is no family history of similar looking teeth.

Figure 5. 11 MIH molars and incisors, and HSPM

MIH	N	%	HSPM	N	%
Correct	38	50		0	0
Hypo	31	40.79		5	6.58
EDD	2	2.63		0	0
Acquired	4	5.26		0	0
Other	1	1.32		0	0
Total	76	100		5	6.58

Table 5. 5 MIH molars and incisors, and HSPM

Q4.8.



This is a 9 year old boy. He suffer with pain when he brushes his teeth and when eating.

Figure 5. 12 Severe MIH molars and incisors, with PEB

	N	%
Correct	48	63.16
Hypo	23	30.26
EDD	3	3.95
Acquired	2	2.63
Other	0	0
Total	76	100

Table 5. 6 Severe MIH molars and incisors, with PEB

Q4.9.



This is a 7 year old girl. She is scared of the dentist because the air and water hurts her teeth.

Figure 5. 13 Severe MIH molars and incisors, caries primary and permanent molars

	N	%
Correct	24	31.58
Hypo	17	22.37
EDD	2	2.63
Acquired	28	36.84
Other	5	6.58
Total	76	100

Table 5. 7 Severe MIH molars and incisors, caries primary and permanent teeth

Q4.10.



This patient is 5 years old and complains of sensitivity when she drinks cold water.

Figure 5. 14 HSPM (primary dentition only)

	N	%
Correct	3	3.95
'MIH'	7	9.21
Hypo	28	36.84
EDD	2	2.63
Acquired	32	42.11
Other	4	5.26
Total	76	100

Table 5. 8 HSPM (primary dentition only)

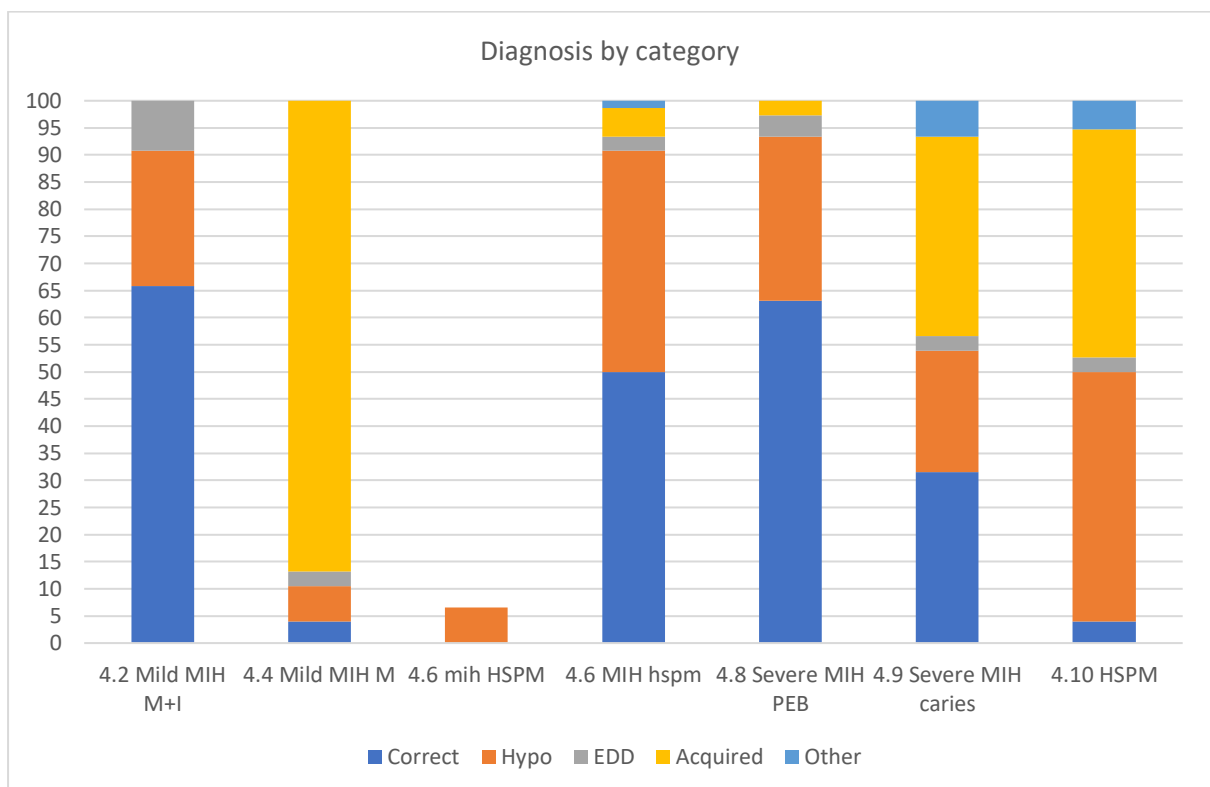


Figure 5. 15 Diagnosis by categories for each question

Management

Fifty-eight GDP completed this part of the survey. GDPs were asked to treatment plan their preventive management of each patient, management of the FPM and aesthetic management, if any, of the incisor teeth. Participants were not given the diagnosis for these cases but were advised of the patient's symptoms, complaints and given clinical photographs, OPG and bitewing radiographs for each patient.

Vignette One (Case 4.2 in diagnosis section)

- *Eight-year-old male, non-anxious*
- *Mild MIH affecting molars and incisors*
- *Patient complaint regarding aesthetics of incisors*
- *No pain or sensitivity*
- *No caries or PEB*
- *Bifurcation of lower second permanent molars not formed*

(90.79% of participants diagnosed this as MIH or hypomineralisation/hypoplasia)

Appendix 3 shows clinical photographs and radiographs included

Vignette Two (Q4.9 diagnosis section)

- *Seven-year-old girl, mild anxiety but potentially co-operative*
- *Severe MIH affecting molars and incisors*
- *Patient not concerned regarding aesthetics of incisors*
- *Patient complaint regarding sensitivity to air and water at dentist and cold foods at home*
- *Caries in three FPM and in primary molars; UL6 caries free*

- *Bifurcation of lower second permanent molars not formed*

(53.95% of GDP diagnosed this case as MIH or hypomineralisation/hypoplasia)

Appendix 4 shows radiographs and clinical photographs included

Prevention

When asked about recall interval in vignette one, 69.0% (n=40) of GDPs selected a three-month recall period, whilst 31.0% (n=18) selected a six-month recall. For vignette two 94.8% (n=55) of GDPs selected a three month recall period, whilst 5.2% (n=3) selected a six-month recall. The remaining results from the prevention section are displayed in table 5.9.

	Vignette 1 (n)	Vignette 2 (n)
Recall		
3 months	69% (40)	94.8% (55)
6 months	31% (18)	5.2% (3)
12 months	0	0
Fluoride (applications per year)		
0	1.7% (1)	1.7% (1)
1	0	0

2	29.3% (17)	6.9% (4)
3	1.7% (1)	3.4% (2)
4	67.2% (39)	87.9% (51)
OHI & diet advice		
3 months	63.8% (37)	93.1% (54)
6 months	36.2% (21)	6.9% (4)
12 months	0	0

Table 5. 9 Preventive care selected for vignette one & two

Management of Permanent Posterior Teeth

Each GDP was asked to select a treatment option including actively monitor, fissure sealant (GIC), fissure sealant (resin), restore composite, restore amalgam, restore glass ionomer cement, restore PMC, or extract. The results for each molar are shown in table 5.10.

		Vignette 1 (n)	Vignette 2 (n)
UR6	Monitor	10.3% (6)	0
	Fissure seal GIC	15.5% (9)	3.4% (2)
	Fissure seal resin	58.6% (34)	3.4% (2)
	Restore composite	6.9% (4)	55.2% (32)
	Restore amalgam	0	1.7% (1)
	Restore GIC	3.4% (2)	22.4% (13)
	Restore PMC	1.7% (1)	3.4% (2)
	Extract	3.4% (2)	10.3% (6)
UL6	Monitor	12.1% (7)	3.4% (2)
	Fissure seal GIC	13.8% (8)	6.9% (4)
	Fissure seal resin	51.7% (30)	62.1% (36)
	Restore composite	12.1% (7)	15.5% (9)
	Restore amalgam	0	0
	Restore GIC	5.2% (3)	3.4% (2)
	Restore PMC	1.7% (1)	3.4% (2)
	Extract	3.4% (2)	5.2% (3)
LL6	Monitor	12.1% (7)	1.7% (1)
	Fissure seal GIC	12.1% (7)	6.9% (4)
	Fissure seal resin	51.7% (30)	12.1% (7)
	Restore composite	12.1% (7)	46.6% (27)

	Restore amalgam	0	3.4% (2)
	Restore GIC	5.2% (3)	15.5% (9)
	Restore PMC	3.4% (2)	5.2% (3)
	Extract	3.4% (2)	8.6% (5)
LR6	Monitor	13.8% (8)	1.7% (1)
	Fissure seal GIC	15.5% (9)	1.7% (1)
	Fissure seal resin	50% (29)	12.1% (7)
	Restore composite	10.3% (6)	53.4% (31)
	Restore amalgam	0	1.7% (1)
	Restore GIC	3.4% (2)	15.5% (9)
	Restore PMC	3.4% (2)	6.9% (4)
	Extract	3.4% (2)	6.9% (4)

Table 5. 10 Management of FPM for vignette one & two

Management of Anterior Teeth

Each GDP was asked to select which incisors they would carry out aesthetic work on, including the option to select none. For vignette one, just over half of participants chose to carry out aesthetic work (58.6% n=34). For vignette two, the majority of participants did not choose to carry out aesthetic work on the incisors (94.8% n=55), however, 5.2% (n=3) opted to treat the UL1 and 3.4% (n=2) opted to treat the UR1. Treatment modalities are displayed in table 5.11.

	Vignette 1% (n)	Vignette 2% (n)
Would you carry out treatment on the anterior teeth?		
Yes	58.6% (34)	5.2% (3)
No	41.4% (24)	94.8% (55)
External bleaching	14.7% (5)	33.3% (1)
Microabrasion	79.4% (27)	66.7% (2)
Resin Infiltration	20.7% (12)	0
Composite with hard tissue removal	8.8% (3)	0
Composite veneers no hard tissue removal	20.6% (7)	0
Porcelain veneers with hard tissue removal	0	0

Table 5. 11 Aesthetic management for vignette one & two

Referral

Participants were asked a series of questions following treatment planning to explore their decision making for vignette one and two. Each GDP answered the three questions for each vignette (six questions in total) with a free text response. These questions were analysed quantitatively and qualitatively. The quantitative results are shown in table 5.12. below.

	Vignette 1 (n)	Vignette 2 (n)
Q.12 Would complete –		
Prevention	100% (58)	98.3% (57)
Restoration	34.5% (20)	79.3% (46)
Extraction	3.4% (2)	6.9% (4)
Aesthetic Rx	44.8% (26)	3.4% (2)
Q.13 Would refer for –		
Opinion	12.1% (7)	10.4% (6)
Prevention	0 (0)	1.7% (1)
Restoration	1.7% (1)	17.3% (10)
Extraction	5.2% (3)	12.1% (7)
Aesthetic Rx	25.9% (15)	3.4% (2)
None	58.6% (34)	67.2% (39)
Q.14 Referral to –		
Paediatric Dentistry	70.7% (41)	56.9% (33)
Orthodontics	8.6% (5)	12.1% (7)
Oral Surgery	0	3.4% (2)
Community	13.8% (8)	20.7% (12)
Special Care Dentistry	3.4% (2)	6.9% (4)
Restorative	6.9% (4)	1.7% (1)
Therapist	0	1.7% (1)
Sedation Service	1.7% (1)	0
None	5.2% (3)	12.1% (7)

Table 5. 12 Response to questions regarding referral

Qualitative Analysis of Referral

Qualitative analysis was undertaken for the three questions regarding referral practices, for each vignette. Thematic analysis was carried out considering all six answers from each GDP in combination. A sample of 20 responses were initially coded to create a framework. The remaining 38 responses were then reviewed, and the framework adapted where new ideas were discovered. From the framework, themes and subthemes were developed. Analysis was descriptive and at surface level due to the brief nature of responses (single words or short sentences). The overarching theme identified was of sharing care between primary and secondary care. What this meant for each GDP was dependant on their confidence when managing MIH, factors which affect complexity of the case and options for referral locally. Figure 5.16 shows the themes, subthemes and codes identified.

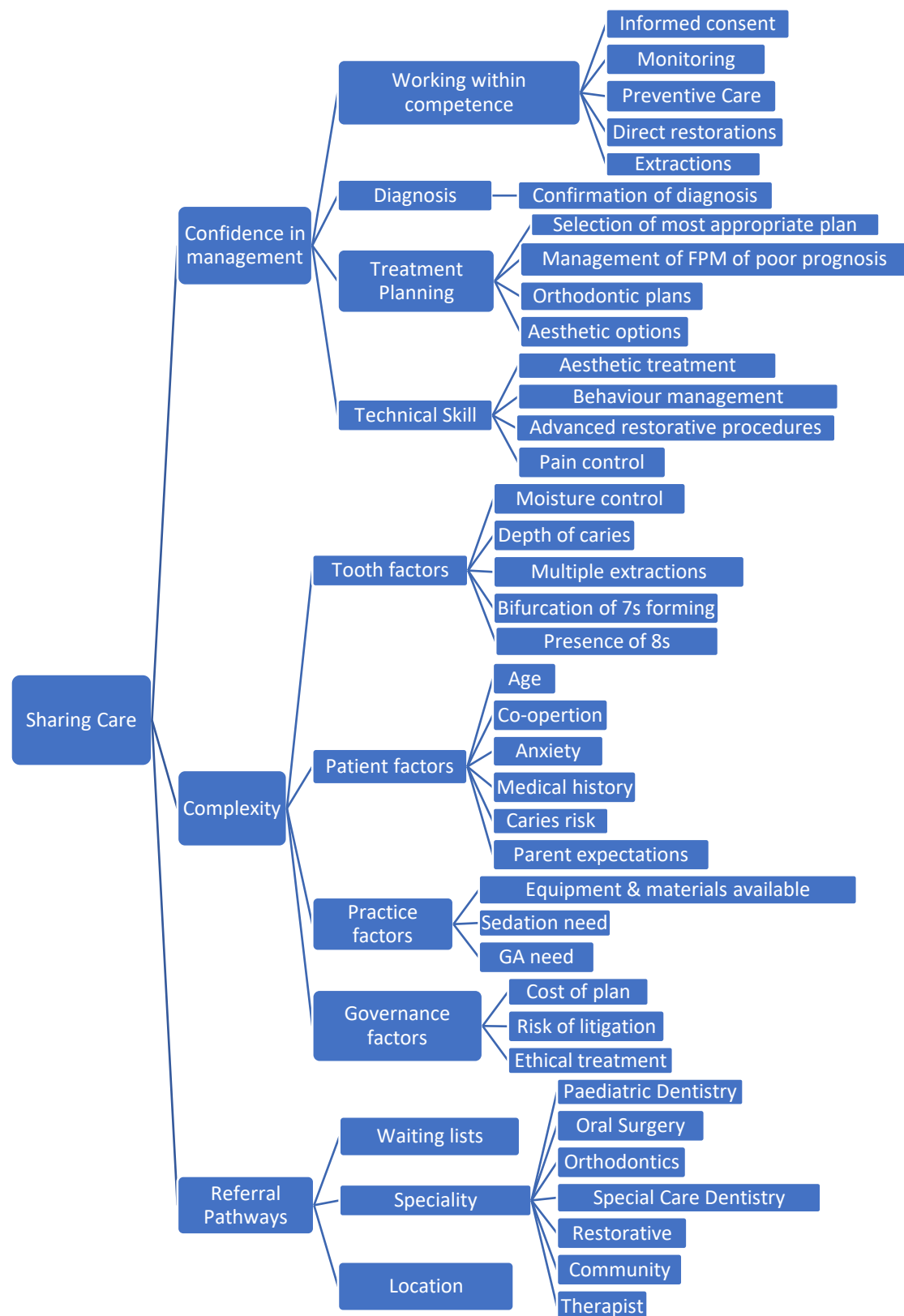


Figure 5. 16 Themes, subthemes and codes for treatment and referral decisions

Confidence in Management

Most GDPs felt confident to carry out all the treatment they had planned for the child, which was generally preventive care and direct restorations. One participant commented on the need for informed consent when planning treatment, which necessitates knowledge and experience of all potential options discussed. Some GDPs expressed they were unsure of the diagnosis and therefore would want to clarify this first before progression to treatment. Although many were happy to carry out their plan, some expressed that a second opinion regarding the best possible plan from a specialist would be helpful, prior to the treatment being completed in primary care.

‘Definitive diagnosis confirmation’

(Q6.13 line 21)

‘Possibly with specialist input for treatment plan’

(Q6.12 line 28)

Some GDPs felt part of their plan was outside their scope of practice. This included placement of PMCs on FPM teeth, microabrasion, tooth whitening and resin infiltration. Appropriate pain management was also highlighted as an area which may need specialist input or additional pharmacological agents such as sedation.

‘Preformed crowns – never done them before’

(Q7.13 line 37)

Complexity

Many GDPs commented on the need to monitor bifurcation of second permanent molars or presence of wisdom teeth when making their treatment plan. Practical factors such as ability to maintain good moisture control and depth of caries may also influence whether the GDP completed treatment or referred. The most frequently mentioned complicating factor for treating the patients in the vignettes was co-operation. This was true for both cases, despite only vignette two requiring operative dentistry and stating that the patient was dentally anxious. Age and anxiety were mentioned as well, which may influence co-operation. Caries risk and medical history were also mentioned as potential factors which may influence management. In addition, some GDPs spoke about the need to also manage the expectations of the parent.

'I'd probably refer for extractions for RA unless a very robust child!!'

(Q6.13 line 6)

GDPs discussed limitations of the dental practices they worked in, including access to materials such as PMCs, microabrasion, and resin infiltration. A need for sedation or GA due to co-operation or anxiety was also discussed, with most GDPs needing to refer for these services. Some GDPs highlighted potential ethical dilemmas in regards to carrying out aesthetic treatment on very young children. A few discussed the high cost of carrying out long treatment plans as a reason for referral, which reflects the current remuneration system in primary care within the UK.

'Availability of materials in NHS practice to carry out microabrasion'

(Q6.13 line 43)

Referral

The speciality or service the GDP would choose to refer to was varied and not always appropriate e.g. restorative or special care dentistry which are adult services. The remaining locations (paediatric dentistry, oral surgery, orthodontics, community or dental therapist) are all potentially appropriate and depend on the local services available in each area. For example, referral to oral surgery for extractions of FPMs under GA may not be appropriate in areas with a community or paediatric dentistry service, however in some locations this may not be available, or may necessitate unreasonably long distances to travel for the patient.

‘Orthodontics if required in the future, restorative opinion if required in future also’

(Q6.14 line 44)

Some GDP highlighted that access to secondary care could be difficult for some families, based either on geographical distance, and also considering the patient’s own financial circumstances, e.g. access to transport, cost of travel. In addition, long waiting times for appointments once referred and lack of specialists in rural locations may prohibit referral. The effects of long waiting lists for appointments on referral to secondary care was also discussed, as this may influence the GDPs willingness to refer the patient.

‘Nowhere. No specialists available’

(6.14 line 50)

‘Time taken to be assessed at the dental hospital / long waiting times.’

(6.13 line 21)

Confidence

Fifty-seven GDPs completed this part of the survey. Participants were asked to rate their self confidence in relation to several aspects of care for children with MIH or HSPM (figures 5.17 to 5.19). They were then asked to state whether they were aware of the link between HSPM and MIH, who should manage children with MIH, and whether further training in the management of MIH at undergraduate and postgraduate levels would be useful. Just under two-thirds of participants were aware of the link between HSPM and developing MIH (n=35; 61.4%). Most participants felt that care should be shared between primary and secondary care for children with MIH (n=53; 93.0%), with 5.3% (n=3) and 1.8% (n=1) feeling specialists or GDP should solely manage children with MIH respectively. Almost all participants felt that further training at both postgraduate and undergraduate level would have been useful for them (n=55; 96.5%).

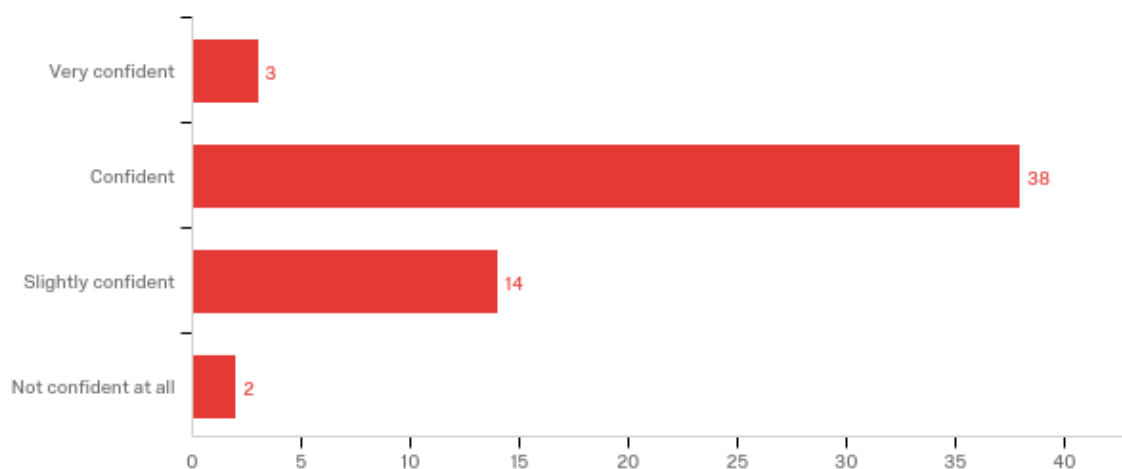


Figure 5. 17 How confident do you feel in the diagnosis of MIH?

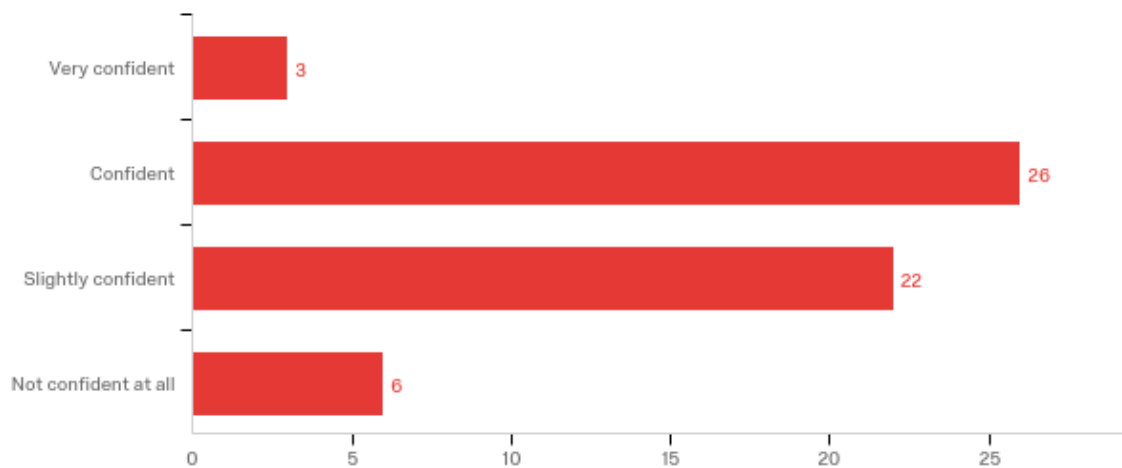


Figure 5. 18 How confident do you feel in the diagnosis of HSPM?

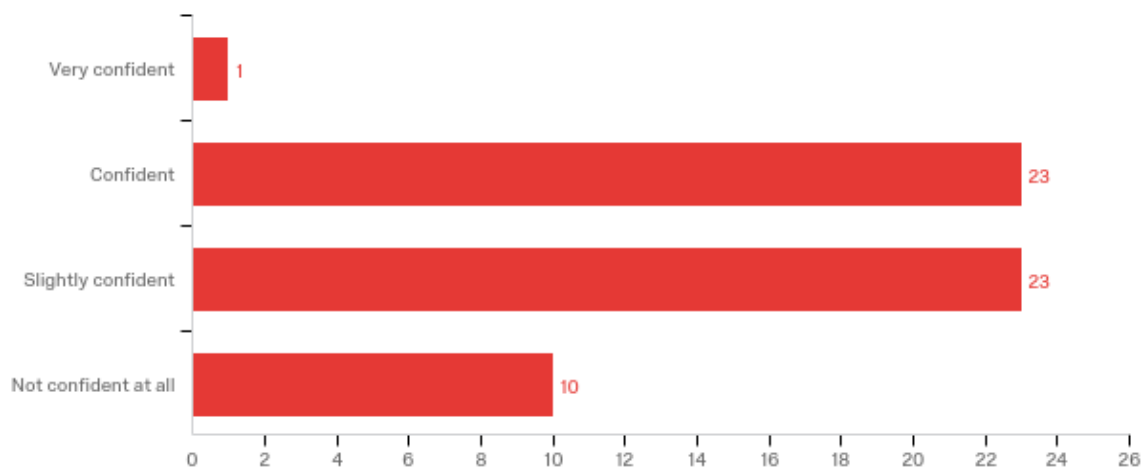


Figure 5. 19 Do you feel confident treatment planning children with MIH?

Data was then analysed using the Mann-Whitney-U test to assess associations between confidence in diagnosis and treatment planning, with accuracy of diagnosis and treatment planning. Pearson's correlation test was used to assess correlation between accurate diagnosis and accurate treatment planning. Significance was set at $p < 0.05$.

Accuracy of diagnosis was expressed in terms of the number of correct diagnoses (minimum 0; maximum 7). Accuracy of treatment planning was analysed separately for molars in vignette one and two, and incisors in vignette one and two. Each participant therefore received a value for these four outcomes (molars - minimum 0; maximum 4; incisors – incorrect 0; correct 1). Confidence in diagnosis and treatment planning were dichotomised into very confident/ confident and slightly confident/ not confident at all groups.

In vignette one any treatment which didn't involve restoration or extraction of molar teeth was acceptable, as there was no caries, PEB or symptoms. In terms of the incisor teeth, the child's main complaint was regarding appearance, so any treatment to address this was acceptable.

In vignette two treatment options were acceptable as follows:

UR6 – any restoration, no extraction as too early and fissure sealant not appropriate

UL6 – any treatment except restoration or extraction

LL6 - any restoration, no extraction as too early and fissure sealant not appropriate

LR6 - any restoration, no extraction as too early and fissure sealant not appropriate

Incisors – child not bothered by appearance, so no treatment indicated.

Diagnosis

Table 5.13. show the number accurately diagnosed cases of MIH / HSPM by participants in relation to confidence in diagnosis. No participants accurately diagnosed all seven cases. The relationship between confidence in diagnosis and accurate diagnosis was significant ($p=0.016$). No significant difference in diagnostic skill was found between those with postgraduate qualifications and those without ($p=0.703$). Appendix 5 shows the Mann Whitney U Test Calculations.

	Number of Accurate Diagnoses by Participant							Total
	0	1	2	3	4	5	6	
Not confident at all/slightly confident	8	1	4	1	1	1	0	16
Confident/very confident	9	3	6	7	10	5	1	41
Total	17	4	10	8	11	6	1	57

Table 5. 13 Number of participants accurately diagnosing cases in relation to self-reported confidence in diagnosis

Treatment Planning

The following tables demonstrate the number of accurately treatment planned molars for vignette one (table 5.14.) and two (table 5.15.), and accurately treatment planned incisors for vignette one (table 5.16.) and two (table 5.17). The only significant result was for vignette one where being unconfident in treatment planning was related to accuracy in planning for incisor ($p=0.036$).

Appendix 6 shows the Mann Whitney U Test calculations for confidence in treatment planning and accuracy of treatment planning.

Associations between accurate diagnosis and accurate treatment planning of molars was assessed for correlation. No significant relationship was found between the number of accurately diagnosed cases and the number of correctly treatment planned molars for vignette one (-0.054 $p=0.689$) or two (-0.03 $p=0.808$).

	No. Accurate Planned Molars Vignette 1					Total
	0	1	2	3	4	
Not confident at all/slightly confident	4	2	6	4	17	33
Confident/very confident	0	0	4	3	17	24
Total	4	2	10	7	34	57

Table 5. 14 Number of accurately treatment planned molars by participants in relation to self-reported confidence in treatment planning (vignette one)

	No. Accurate Planned Molars Vignette 2					Total
	0	1	2	3	4	
Not confident at all/slightly confident	3	0	2	7	21	33
Confident/very confident	1	4	3	7	9	24
Total	4	4	5	14	30	57

Table 5. 15 Number of accurately treatment planned molars by participants in relation to self-reported confidence in treatment planning (vignette two)

	No. Accurately Planned Incisors V1		Total
	0	1	
Not confident at all/slightly confident	10	23	33
Confident/very confident	14	10	24
Total	24	33	57

Table 5. 16 Number of accurate treatment plans for incisors in relation to self-reported confidence in treatment planning

	No. of Accurately Planned Incisors V2		Total
	0	1	
Not confident at all/slightly confident	1	32	33
Confident/very confident	2	22	24
Total	3	54	57

Table 5. 17 Number of accurate treatment plans for incisors in relation to self-reported confidence in treatment planning (vignette two)

5.5 Discussion

This study was designed to investigate how GDPs in the UK diagnose and treatment plan patients with MIH. The results demonstrate that diagnosis can be affected by the presenting features and severity of the condition. Most GDPs chose acceptable treatment plans and felt able to complete most or all the plan without referral. Confidence in diagnosis was associated with accuracy in diagnosis.

An electronic survey was chosen as this gave the ability for quick dissemination, without the cost (financial and environmental) of traditional paper surveys. As the survey was shared on social media, an accurate response rate could not be estimated, as it is impossible to assess how many dentists may have seen the survey and opted not to complete it. GDPR guidelines meant that restrictions existed on how participants could be contacted, reducing dissemination options. In comparison to the total population of GDPs in each nation of the UK, the spread of responses was similar to the proportions of GDPs in this study.

Clinical photographs were selected using a rigorous process, and their accurate diagnosis confirmed by the whole research team. Members of the CONNECT Research Group, comprising mainly speciality trainees in Paediatric Dentistry, reviewed the images and confirmed each diagnosis was clear before finalisation of the survey. The use of clinical photographs to diagnose hypomineralisation and other enamel defects has been found to have a good sensitivity and specificity in comparison to clinical diagnosis in previous studies (Elfrink et al. 2009, Chen et al. 2013, Dabiri et al. 2018). The participants were not informed that the survey was about MIH, in order to recreate, as closely as possible, the natural diagnostic process during examination of teeth.

This was the first study to assess how GDPs diagnose MIH when presented with vignettes, although Jalevik et al conducted a study to assess the difference between trained and untrained dentists in the detection of developmental defects of enamel (DDE). Their main findings were that the untrained staff tended to call all types of DDE hypoplasia and seemed to have limited knowledge to discern between different types of defect (Jalevik et al. 2019). Patel et al discussed the differences between hypoplasia and hypomineralisation in their paper in order to address this issue (Patel et al. 2019). Weerheijm and Mejare asked members of the EAPD if they were familiar with the appearance of a tooth with MIH, using photographs of FPM and incisor teeth, but did not ask participants to give a diagnosis for the tooth (Weerheijm and Mejare 2003). Crombie et al used the same format in a survey of the Australian and New Zealand Society of Paediatric Dentistry (Crombie et al. 2008).

Other surveys have assessed treatment options for individual teeth with MIH. Kopperud et al disseminated their vignette survey to GDPs and specialists in Norway, investigating practices in tooth tissue removal when treating molars with MIH, alongside assessment of knowledge and experience of MIH(Kopperud et al. 2016). This survey of 652 participants sampled the total population of dentists working within the public dental service of Norway, including specialists in paediatric dentistry. Alanzi et al asked 221 GDPs and specialists in Kuwait how they would manage individual teeth, alongside a traditional survey assessing knowledge, experience and confidence (Alanzi et al. 2018). This study utilised a convenience sample of delegates at a national dental conference and therefore may not be representative of the whole population of GDPs in Kuwait. A similar survey was conducted in Ireland (Wall and Leith 2020), using elements from Alanzi, Gambetta-Tessini and Kopperud's survey (Kopperud et al. 2016, Alanzi et al. 2018, Gambetta-Tessini et al. 2016). A sample of 230 dentists (mostly GDPs) were asked about knowledge and experience of MIH, in addition to selecting tooth tissue removal and restoration material for individual molars. Taylor et al investigated how 159 specialists and 74 GDPs treatment planned children with compromised FPM,

including molars with MIH (Taylor et al. 2019). None of these studies assessed ability to diagnose MIH.

Diagnosis

Correct diagnosis of HSPM was much lower than for MIH, indicating that GDPs are not as familiar with the condition affecting primary teeth. It may also be true that many GDPs are not aware of the correct terminology to describe the condition when it affects primary teeth. Where the cases demonstrated caries (Mild MIH (caries in primary teeth) and severe MIH with caries), accurate diagnosis was also lower. It may be that GDPs are more comfortable diagnosing caries than MIH, or simply that the caries was more obvious in these photographs.

The qualitative analysis of incorrect answers found a spectrum of results, with some answers more closely related to the correct diagnosis than others. For example, description-based answers such as hypomineralisation, or condition-based answers such as molar-incisor-hypoplasia, demonstrate awareness of hypomineralisation and hypoplasia (which can be confused with PEB), but are technically incorrect. Within the enamel developmental defects group, diagnoses that are often confused with MIH were recorded, such as fluorosis. The acquired defects group included common dental disease such as caries and tooth wear, which may have occurred alongside MIH but have a distinctly different appearance. Finally, the 'other diagnoses' theme included diagnoses which were not related to hard tissue at all, such as orthodontic diagnoses. Where fewer clinicians gave a diagnosis under the hypoplasia / hypomineralisation theme, this indicates that diagnosis was more challenging.

Over the last decade surveys to assess the experience of both paediatric specialists and GDPs in treating children with MIH have taken place, in Europe, Kuwait, the Middle East, Australia and South America (Crombie et al. 2008, Kalkani et al. 2016, Silva et al. 2016a, Ghanim et al. 2011). The results have found that in general only half of GDPs surveyed are confident in the diagnosis of MIH. The only exception was in a study by Gambetta-Tessini et al, where over 80% of clinicians in both Chile and Australia reported confidence in diagnosis (Gambetta-Tessini et al. 2016). Crombie et al found that 98.3% of GDPs recognised the appearance of MIH teeth, but were not asked to diagnose the condition (Crombie et al. 2008). A survey from 2016 in the UK found that 57% of the 31 GDPs attending a study day on paediatric dentistry felt confident or very confident when diagnosing MIH (Kalkani et al. 2016). Our findings are in keeping with results from across the world but show an increase in confidence in the UK, with 71.93% of GDPs feeling confident or very confident when diagnosing MIH. Accurate diagnosis was found to be significantly related to confidence within this sample, however presence of postgraduate qualifications was not related to accurate diagnosis. Education at postgraduate and undergraduate level should focus on equipping clinicians with the skills to differentiate between different severities of MIH, hypoplasia and caries.

Treatment

Two vignettes were selected to demonstrate children with common presentations of MIH but with key differences. These included differences in aesthetic concern, presence of caries, and anxiety levels. Again, the participants were not informed that the children had MIH, in order to recreate the clinical decision-making process that would take place in practise.

Four studies investigating treatment options for MIH and FPM have been carried out previously. Alanzi et al presented three tooth-level vignettes as part of a survey and asked both specialists and

GDPs in Kuwait how they would manage each tooth (Alanzi et al. 2018). This included selecting treatment options for a mildly and severely affected FPM, and aesthetic options for an MIH affected incisor. Taylor et al used a vignette survey to investigate how general dentists and specialists in paediatric dentistry would manage poor prognosis FPM at ages seven, nine and 15 – with the seven and nine-year-old having mild and severe MIH respectively (Taylor et al. 2019). Both Wall and Kopperud et al used the same tooth-level vignettes to assess management of MIH molars with mild and severe MIH, in terms of tissue removal and restoration type (Wall and Leith 2020, Kopperud et al. 2016).

The majority of participants implemented the most frequent recall and preventive regime for both vignettes, in keeping with the guidance by public health England (Public Health England 2017). More participants identified this need for the child with caries which highlights that GDP may not always associate MIH with increased caries risk. The majority of participants chose acceptable treatment options for the molars in both vignettes, but there was a spread across all possible options, which is concerning when irreversible options such as restoration or extraction were selected. Consideration of removal of poor prognosis FPMs at the ideal time is an important topic but was not investigated in this study - neither child was at the correct developmental stage (Cobourne et al. 2014).

In vignette one of this study, where the child had mildly affected FPM without caries or PEB, participants selected non-invasive options of fissure sealant with GIC or resin, or active monitoring most frequently (UR6: 84.4%, UL6: 77.6%, LL6: 75.9%, LR6: 79.3%). All participants, except one, elected to use fluoride varnish one or more times a year for both cases. Alanzi et al found that composite resin was the most popular treatment option (47.8%) followed by PMCs (20%) for a mildly affected FPM (Alanzi et al. 2018). Taylor et al also found that the majority of GDPs (95%) opted for prevention and/or fissure sealants for the child with mild MIH (Taylor et al. 2019). In Wall and Leith's

study in Ireland, they found 83% of participants would place fissure sealants with GIC or resin on a mildly affected MIH tooth, followed by 16% using fluoride varnish (Wall and Leith 2020, Kopperud et al. 2016). Kopperud et al found 54% of Norwegian participants would favour fluoride varnish, with 38% opting for a fissure sealant (Wall and Leith 2020, Kopperud et al. 2016). The participants of this study identified that it wasn't appropriate to carry out conventional restorations on a tooth without PEB or caries.

In vignette two, where the child had severely affected FPM with caries, the most common option was for restoration with composite (UR6: 55.2%; LL6: 46.6% LR6: 53.4%), and then GIC (22.4%, 15.5% 15.5% respectively). Taylor et al also found composite (44%), followed by GIC (17%) were the most popular options for restoration (Taylor et al. 2019). Similarly, for the severely affected molar, Irish dentists preferred GIC (31.4%) and composite (28.5%), and Norwegian dentists preferred GIC (58%) and composite (21%)(Wall and Leith 2020, Kopperud et al. 2016). For a FPM with PEB, Alanzi et al found that 64.3% of GDPs would use a PMC and 20% would use composite resin and fissure sealants in combination (Alanzi et al. 2018). In this study, less than 7% of participants opted to use PMCs, which was similar to the findings of Taylor et al (6.3%)(Taylor et al. 2019). Irish dentists opted to use PMCs by 16.2% of participants, and for Norwegian dentists this was 11% (Wall and Leith 2020, Kopperud et al. 2016). It appears that in Europe, and particularly in the UK, GDPs do not feel confident in using PMCs on FPM teeth.

Just over half of participants were willing to address the aesthetic concerns of the child in vignette one, with a quarter of participants opting to refer for this treatment. GDPs in the UK opted for a more minimally invasive approach, with microabrasion being the most popular treatment option for both cases. Alanzi et al, presented a nine-year-old child with MIH affecting the UR1 tooth. Most GDPs opted to remove the affected area and restore with composite (42.6%), followed by equal

numbers opting to use microabrasion or resin infiltration (25.2%) (Alanzi et al. 2018). In vignette one of this study, of those who opted to carry out treatment on the incisor teeth, 79.4% opted for microabrasion, followed by 20.7% opting for resin infiltration. Only 8.8% opted for hard tissue removal and localised composite.

Referral

When considering referral, 41.4% (vignette one) and 32.8% (vignette two) of GDPs would refer for part of the treatment plan. Although case one was a milder form of MIH, GDPs felt less comfortable managing elements of the condition. In vignette two, the child has obvious caries and did not require any aesthetic management, and therefore perhaps GDPs felt more confident in the management. The most popular destination for referral in both cases was to paediatric dentistry services. Some participants specified referral to adult services including restorative dentistry and special care dentistry, which was not appropriate but may reflect the limitations of services available locally. In a survey of Australian GDPs, 78.7% would refer to a specialist in paediatric dentistry when managing a child with MIH (Gambetta-Tessini et al. 2016). A similar study in Malaysia found that 57.1% of GDP would refer to a specialist for management of MIH (Hussein et al. 2014). These results are not directly comparable as the GDPs were not given a specific scenario to consider.

In study one, it was demonstrated that referring practitioners generally referred at a suitable time for consideration of removal of poor prognosis FPMs and waiting lists did not adversely affect the timing of treatment, although waiting lists have likely increased since 2015. Referral was most often for management of molars, and treatment most often involved GA. This relates to the factors discussed in the qualitative analysis including patient and facility factors.

The qualitative analysis of referral practices demonstrated that care of children with MIH exists on a spectrum with straightforward cases being amenable for primary care management, and the complex cases requiring management in secondary care. The overarching theme of shared care reflected this, with the main themes of confidence in management, case complexity, and local referral pathways dictating when and why each GDP may choose to refer, and what treatment they were able to do themselves. What is interesting are the many factors outside the GDP's control which dictate referral – including tooth, patient, and practice factors. Furthermore, the process of referral may be dictated by practice location and what services are available, in addition to waiting list times. Although these findings are interesting, as participants tended to answer with single words or short sentences, a deeper understanding of these factors could not be explored. Further qualitative research has been planned in study three so that the emotional impact and social experience when treating children with MIH can be explored.

Confidence

Several surveys assessing the experience of both paediatric specialists and GDPs in treating children with MIH have taken place across the world (Crombie et al. 2008, Kalkani et al. 2016, Silva et al. 2016a, Ghanim et al. 2011). Generally only half of participants felt confident when managing MIH, however Gambetta-Tessini et al, found over 80% of clinicians in both Chile and Australia felt confident when diagnosing MIH (Gambetta-Tessini et al. 2016). Crombie et al found that 98.3% of GDPs recognised the appearance of teeth with MIH, but were not asked to diagnose the condition (Crombie et al. 2008), whilst a survey from 2016 in the UK found that 57% of the 31 GDP attending a study day on paediatric dentistry felt confident or very confident when diagnosing MIH (Kalkani et al. 2016). The findings of this study are in keeping with results from across the world but show an increase in confidence in the UK, with 71.93% of GDPs feeling confident or very confident when diagnosing MIH.

Almost all participants identified that children with MIH should be managed by both GDPs and specialists. This was reflected in the treatment planning section, where only one GDP opted to refer for the whole plan in vignette two. Most participants felt further training or education regarding MIH would be useful (94.8%), which is greater than results found for GDPs in Saudi Arabia (90.5%) (Silva et al. 2016a), Iraq (69.8%)(Ghanim et al. 2011) and Kuwait (33.9%) (Alanzi et al. 2018). The results may have been higher in this study because participants had attempted diagnosis and treatment planning throughout the survey, and therefore any gaps in knowledge would have been apparent when answering this question. More teaching on the management of MIH, particularly at undergraduate level, may help increase the confidence of GDPs when managing MIH.

Accurate diagnosis was found to be significantly related to confidence in diagnosis within this sample. This was true when considering answers that were completely accurate and shows that those who use the correct terminology are more likely to identify the condition in their patients. Confidence in management of MIH was not related to acceptable treatment planning. Acceptable treatment planning was not related to accurate diagnosis. Reassuringly many GDPs who were inaccurate in the diagnosis section, still implemented a good preventive strategy and opted to restore teeth with PEB and caries.

Study Limitations

There are several limitations to the findings of this study. Firstly, as the survey was shared on social media, an accurate response rate cannot be reported. It would be impossible to track how many clinicians had seen the survey, and the demographics of this group. With any survey, only the most interested clinicians will complete it, leading to a potential response bias. The initial methodology using randomly selected GDPs was designed to try to reduce this bias, however, due to a small

response, additional recruitment methods (social media) were deemed necessary to boost numbers. Additional steps to increase the response rate included the chance to enter a prize draw to win shopping vouchers as a 'thank you' to participants for being involved, personalised emails and the sending of a second reminder to each selected dental practice as recommended by Edwards et al (Edwards et al. 2009). Despite this, the sample size was still small. Taylor et al recruited 74 GDPs to their study which is similar to the level achieved for this study. This may indicate that for many GDPs, paediatric dentistry is not a topic of interest. It may also be true that GDPs found the content of this particular survey challenging, leading them to abandon it before completion. Partial completion may also be related to the length of the survey. Two shorter surveys, addressing diagnosis and treatment planning separately, may have improved response rate.

The true results for GDPs across the UK may be different to the results found in this study. Most clinicians graduated in the last 20 years, and therefore the results cannot be extrapolated to more experienced GDPs. In addition, the majority of participants were female, whereas the workforce across the UK is split more closely 50/50 male to female. The geographical locations of participants based on postcode was varied and the proportion of participants from each nation of the UK was similar to the proportion of GDPs registered with the General Dental Council in each country.

The clearest clinical images were selected to aid diagnosis and treatment planning, but this cannot fully replace the process of clinical decision making. Other studies have also successfully used clinical photographs to diagnose MIH and HSPM, such as a birth cohort study which investigated the relationship between MIH and HSPM (Elfrink et al. 2012). The images in this study were selected using a rigorous three stage process to ensure that the diagnosis was as clear as possible. The lead researcher selected multiple suitable images showing MIH and HSPM (and control images) from the available database, before the best images for each vignette were selected by consensus opinion of

the lead researcher, and consultants in paediatric and restorative dentistry. The correct diagnosis was then positively identified for each case by members of the CONNECT Research Group, which comprises trainees in paediatric dentistry.

Further Research

The results of this study pose further questions. The low response rate calls into question whether MIH is considered an important topic in general practice in the UK. Recent advice from the BSPD recommended that the majority of MIH patients should be managed in primary care (British Society of Paediatric Dentistry 2020a). Given the prevalence of MIH and the increased caries risk for these children, GDPs should be as confident managing mild cases as they are in managing caries. Another question is whether GDPs regard MIH as uniquely challenging or whether they perceive children to be challenging in general. Many participants in this survey discussed problems associated with co-operation as a major factor influencing referral. It was unclear whether this was due to the child having MIH specifically. Almost all GDPs wanted more training regarding MIH. Feedback as to what form this education might take, in addition to exploration of the knowledge GDPs have retained from previous teaching would be useful to plan this. The third study of this thesis will address these questions.

5.6 Conclusion

GDPs are able to accurately diagnose MIH best when both incisors and molars are affected, and no caries is present, on either affected or unaffected teeth. Diagnosis of HSPM was challenging for GDPs. Hypoplasia is incorrectly used to describe hypomineralisation frequently. Confidence in diagnosis is significantly related to actual accuracy. Education surrounding diagnosis should focus on three areas - differentiating hypoplasia from hypomineralisation, the key differences in clinical

appearance of MIH and other enamel defects, and differences between MIH, caries and teeth with MIH and caries, both clinically and radiographically.

The majority of participants demonstrated sound treatment planning in terms of preventive care, and management of FPMs. More GDPs identified increased caries risk in the vignette with caries. Around half of participants were also willing to address the aesthetic concerns of the child in vignette one. Almost all participants identified that children with MIH should be managed by both GDPs and specialists. These findings demonstrate most GDPs in this study were working as effective tier one and two providers when faced with management of children with MIH. Confidence in treatment planning was not related to acceptable treatment planning. Undergraduate education dental education should have a greater focus on MIH in comparison to other rarer enamel defects, such as amelogenesis imperfecta, since GDPs are more likely to deal with these patients in primary care. UK based guidelines for management of MIH in primary care could be useful for those clinicians who have already graduated.

Chapter 6 – How do GDPs Experience Managing Children with MIH in Primary Care? - Exploratory Qualitative Interviews

6.1 Introduction

Qualitative approaches involve studying people in their ordinary settings to explore how they attach meaning to their experiences of the world. Several qualitative approaches can be used depending on the focus of the research (Green and Thorogood 2014). As this particular topic was narrow, involving the experience of a specific population and a particular dental condition, individual semi-structured interviews were selected as the most efficient way to answer the research question. This meant key elements of the objectives could be included in the questioning, whilst still allowing the participant the ability to talk freely around these areas, as appropriate according to their frame of reference (Rubin 2005).

No previous qualitative research has been done to capture the perceptions and experiences of clinicians caring for children with MIH. All information to date regarding the challenges that clinicians may face treating children with MIH have been collected using surveys. Kalkani et al conducted a survey of GDPs and specialist trainees in paediatric dentistry within the UK (Kalkani et al. 2016). They found that GDPs were significantly more likely to experience difficulty deciding prognosis in comparison to the trainees. Sensitivity of teeth and behaviour was often a challenge for both groups. Trainees were much more likely to access second opinions from specialists in paediatric dentistry or orthodontics than GDPs.

Multiple surveys of GDPs knowledge and experience of MIH have also been completed in the Middle East, South East Asia, Australia and Chile. Behaviour management was often discussed as the most

significant barrier to care for these children. Alanzi et al found that 60.9% of their sample of GDPs in Kuwait felt behavioural issues made treatment difficult (Alanzi et al. 2018). In a survey of specialists and GDPs in Saudia Arabia, 74.3% of GDPs felt behaviour management was the main barrier to care, with long treatment plans the second most common pitfall at 54% (Silva et al. 2016a). Kopperud et al found that 48.2% of dentists felt children with MIH had increased anxiety, in their survey of the public dental service in Norway (Kopperud et al. 2016). Hussein et al investigated perceived barriers to care for GDPs in Malaysia (Hussein et al. 2014). They found that the child's behaviour was the second most common reason that they struggled to manage a child with MIH, which they surmised was due to increased sensitivity of teeth and inadequate control of pain. Failure to achieve good LA was discussed as a barrier in multiple studies, but the significance of this for the GDP varied, with 23.5% to 57.6% reporting this as an issue (Silva et al. 2016a, Kopperud et al. 2016, Alanzi et al. 2018, Crombie et al. 2008). Crombie et al surveyed members of the Australian and New Zealand Society of Paediatric Dentistry, including GDPs (Crombie et al. 2008). Providing adequate and long-lasting restorations was felt to be an issue for 87.4% of GDPs when treating children with MIH. When selecting a material to use for restoration, adhesion was a key factor for all clinicians.

Qualitative interviews of GDPs have taken place within the UK regarding other topics. Marshman et al investigated the experiences of 31 dental professionals' experience of managing children with carious lesions in primary teeth (Marshman et al. 2020), as part of the FiCTION randomised control trial (Innes et al. 2019). Semi-structured interviews took place face to face or by telephone.

Negotiating LA was seen as a challenge to providing conventional restorations in primary teeth and was a source of tension for GDPs. The current infrastructure of the NHS was seen as responsible for the perceived time pressures regarding giving preventive advice and clinicians discussed the perception that the prevention alone approach was seen as 'doing nothing', both professionally and by patients. Treatment choices were dependant on the child's cooperation, with this often dictating

the plan. Trying to get this right was a source of stress, as GDPs tried to maintain a sense of professional confidence in their decision making in front of patients and parents.

Dailey and Threlfall conducted qualitative interviews with 93 GDPs based in the North West of England, concerning their experiences of treating young children in primary care. They published three papers addressing the areas of caries preventive advice, use of PMCs and the influence of time from graduation on the GDPs approach to caring for children (Threlfall et al. 2007, Threlfall et al. 2005, Dailey et al. 2007). Time pressure was a theme that influenced both prevention and use of PMCs, in addition to remuneration. The cooperation of the child was seen as a barrier to placement of PMCs, and there was a distrust of professional bodies who produced guidelines encouraging use of PMCs, which were seen as idealistic and impractical for use within the NHS primary care setting. The greatest influence on clinical practice appeared to be for the newly qualified dentists, who had to adapt what they learnt at university to the constraints of the primary care system.

The research to date has not explored how GDPs experience the management of MIH in primary care, and the complex reasons and processes behind their behaviour in this respect. The previous studies in this thesis identified that referral was an option utilised by GDPs when they reached barriers whilst caring for children, such as cooperation or complexity of decision making. The purpose of this study was to gain a richer understanding of the experience and perceptions of the GDPs who manage children with MIH in a primary care setting. In the second study, free text response answers concerning what treatment dentists would be willing to do in primary care and reasons for referral were answered briefly by participants but did not explore fully the environment and contexts which lead to these decisions. The fully qualitative nature of this third study allowed for analysis of thoughts, feelings and emotions related to caring for children with MIH, and children in general, which would not have been easily captured with quantitative methods or survey.

6.2 Aims & Objectives

Research Question

How do UK based GPs understand and experience management of MIH?

Aim

1. *To explore UK based GPs' understanding and experiences regarding MIH*

Objectives

1. *To explore how they understand MIH*
2. *To explore how initial education about the condition has been translated into clinical practice*
3. *To identify how GPs perceive their own management of children with MIH and any challenges they encounter*

6.3 Method

Semi-structured telephone interviews were designed to allow greater flexibility in terms of recruitment of GPs based on their geographic location. It has been reported that participants may feel more relaxed as they can undertake the interview at a place where they feel comfortable, and the technique also reduces the cost incurred by both the researcher and the participant related to travel (Novick 2008). In this particular circumstance, the participants may have felt a greater degree of anonymity and may have felt freer to talk about perceived weaknesses in their clinical practice (Irvine 2011). In addition, due to the timing of interviews during the COVID-19 pandemic, face to face interviews would not have been possible. Despite these advantages, there are several perceived downsides to telephone interviews in comparison to traditional face to face interviews. Interviewers do not have the benefit of rapport building prior to the start of the interview and cannot comment on non-verbal cues and body language during it. There is often an increased ratio of talk from the interviewer relative to the participant, which may cause a decreased depth of coverage of the

themes, leading to a reduced quality (Johnson 2019). Despite these potential drawbacks, it was felt that telephone interviews would overall offer more benefit and be a more pragmatic approach for interviewing busy professionals within the timeframe of the DDSc programme.

This study involved semi-structured qualitative interviews with GDPs across the UK. A semi-structured interview schedule was designed and refined several times after feedback from experienced qualitative researchers and visiting GDPs to the lead researchers place of work. The first draft and final schedule used for the interviews are included as appendices 6 and 7. Ethical approval was granted from the University of Liverpool Ethics Committee (project number 5997). Participants gave written consent prior to commencement of the interviews.

Recruitment and sampling

GDPs were recruited through GDP networks in May 2020 using two methods. The dental leads of the National Institute of Health Research (NIHR) clinical research networks in the North-West Coast and in the North-East & Cumbria areas were contacted and acted as gatekeepers for recruitment to comply with GDPR. Recruitment emails were sent to those GDPs who had previously given consent to be contacted by NIHR for research purposes, and included participant information, and contact details of the lead researcher. In addition, a post was made on social media groups for dentists in the UK explaining the purpose of the study and requesting interested parties to contact the researcher. A purposive sampling technique was utilised to ensure both male and female GDPs, and clinicians of varying experience were included. It was intended that this should not be a representative sample but would ensure that different perspectives related to the length of practice were considered. Potential participants contacted the lead researcher to show interest and the consent form was then sent by email to those who met the inclusion criteria and fitted the profile required for the

purposive sample. Ten participants were initially recruited, and no further participants were contacted following initial data analysis. Participants met the inclusion criteria if they were currently a GDP practicing within the UK, and regularly treated children. Initially multiple female GDPs qualified within the last six years showed interest and were recruited. The lead researcher then awaited more interest from males, and those practicing for over 10 years before requesting consent for participation.

Procedures

Telephone interviews were conducted by one person, the lead researcher, over loudspeaker and were recorded on a M-Audio Microtrack 24/96 audio recording device. Field notes were taken during each interview. The interviews were transcribed by the lead researcher, and the original recordings deleted after resolving any unclear parts of text in the transcript, confirmed with the participants directly. This allowed for increased familiarity with the data to be achieved prior to actual analysis. Initial data analysis was completed concurrently with data collection and transcription of the interviews.

Analysis

Thematic analysis using a systematic inductive approach was used to analyse the data within the transcripts (Braun and Clarke 2006). Coding of text with important, interesting or poignant significance were created across all data. Initial themes and subthemes were drafted from the codes, which were independently analysed and checked by a second researcher. Disagreements were discussed and resolved by the full research team. A coding framework was produced from the initial six transcripts, and this was then developed and adjusted from the analysis of the additional four

transcripts. The themes and overarching theme were developed and amended several times based on feedback from research supervisors.

The analysis was semantic in nature and follows realist paradigms. This was appropriate to explore individual motivations, experience and meaning from the situations described and language used by participants (Sandelowski 2000). Beyond purely describing the findings, analysis has involved interpretation of the codes and themes to contextualise the broader meanings and implications of the findings within the management of MIH, and primary care dentistry for children. The overarching theme was developed as a uniting concept underpinning all the themes, and a central idea throughout all interviews.

6.4 Results

Telephone interviews were conducted by the researcher during May 2020. Participants were asked to identify a convenient time to be contacted and supplied a preferred contact telephone number. Most participants were at home during the interview, which took place during the first lockdown of the Covid-19 pandemic. Interviews lasted between 18 and 37 minutes, with a mean time of 29 minutes. The final sample included four males and six females. Participant demographics are demonstrated in appendix 9. Demonstration of data saturation is challenging with only 10 interviews completed however the author felt no significantly new themes were being identified from the coding at this point.

Overarching Theme – Managing uncertainty

Participants experienced managing children with MIH as a highly variable experience which was dependent on the severity of MIH, the child and the clinician themselves. There was a great deal of

uncertainty surrounding 'doing the right thing' across the themes, and an attempt to try to control these uncertainties, with solutions frequently suggested. Situations where participants felt a lack of control were uncomfortable and stressful.

GDPs were knowledgeable regarding the potential aetiologies, presentation and treatment options for MIH. Mild cases were seen as straightforward and clinically simple, but severe MIH was described as complicated in terms of planning and execution. When planning, the main area of uncertainty was regarding decision making and many participants opted to refer these patients to secondary care where it was perceived they might receive the 'right' care from 'experts'. The unit of dental activity (UDA) system was described as restrictive and out of touch, and prevented participants from carrying out preventive care and behaviour management over several visits which would be useful for the anxious child with MIH.

When treating children with MIH, difficulty in restoring and extracting FPMs was often encountered. Doubt as to whether things would go to plan or not was expressed at several stages of the restorative / surgical process. This included uncertainty surrounding success of LA, whether they could use their preferred material (usually composite) or whether they may be forced to compromise due to clinical factors. After treatment, the uncertainty continued as many worried whether their restoration would last due to compromised bonding. Aesthetic treatment to manage incisors presented an ethical challenge as GDP tried to work within the confines of the UDA system whilst also meeting the needs of their patients. In particular, the use of bleach in under 18s was discussed, with many feeling that this was an excellent non-invasive option, but most feeling unable to carry this out for children.

Referral was utilised frequently for children with severe MIH, when participants felt uncertain regarding options for FPMs, where treatment had been attempted but was unsuccessful, or for aesthetic treatment they felt unable to provide in primary care. The process of referral was described as a 'bureaucratic nightmare' due to the need for repeat referrals after rejection, lost referrals, and patients failing to attend their appointment in secondary care. In addition, waiting list length was seen as an issue, and made referring at the best time for removal of FPMs challenging. Participants described a disconnect from colleagues in secondary care, with no opportunity to learn from the management of MIH by specialists.

The management of children with MIH depends on the relationship between child, parent and clinician functioning well. This worked best when the family attended regularly and engaged with preventive advice for home care, as the relationship dynamics became well established. Where the families did not attend regularly more uncertainty was introduced regarding how individual sessions might progress and the long-term development of rapport. Parents played different roles during appointments, usually viewed as either passive and supportive, or vocal and conflicting with the clinician's approach. Participants also reflected on their perceived role in the management of MIH, and areas for personal or systemic improvement regarding these children's care, including their opinion on issues they had experienced related to other dentists' management of MIH.

Subthemes

- Setting the scene
- Fighting the tooth
- Working within the system
- Self and interpersonal insight

Setting the Scene

Diagnosis

Participants experienced MIH as a spectrum of disease, with mild cases being perceived as ‘easy’ to manage, and severe cases presenting increasing levels of complexity for treatment planning and decision making. Participants talked about features that helped identify MIH and were able to describe a mildly affected tooth as having brown or white patches, which was often asymptomatic, whereas a severely affected tooth would have enamel that was breaking down, ‘unhealthy looking’ or with caries. The presence of MIH affecting molars *and* incisors improved diagnostic confidence. One GDP reflected that perhaps some teeth they had treated with caries, may also have had MIH, which they hadn’t realised at the time.

‘Obviously when I see it, I’m thinking about my differential diagnoses, and I’m thinking ‘oh, could it be caries?’, and those sorts of things. Um, but obviously when it’s the molars and the incisors involved, then that sort of confirms the diagnosis.’

Interview 10, qualified 2019

Most clinicians felt that they saw the condition frequently in its mild form and less frequently or not at all in its severe form, although a few participants reported that they generally saw it infrequently overall. The perceived commonness of a condition may be related to the demographics of the practice, with participants in affluent areas feeling they saw it less, and participants in areas of deprivation coming across it more frequently. In some cases, it may also relate to the experience of the clinician in diagnosing MIH. Some reported that they felt it was becoming more common, and they were seeing it in their patients more often.

'And then when you start looking for it, you know, you realise that it might be a lot more prevalent than you think. And because it manifests in such different levels of severity, and generally speaking in the UK things like that are, um, ignored. [...] But um, I've found it more and more prevalent, in more and more kids, the more and more I look for it.'

Interview 5, qualified 2004

Explaining MIH to the family

Clinicians talked about the importance of early diagnosis of MIH as an opportunity to educate the family about the condition and outline likely prognoses, give preventive advice, and empower them to make positive changes for oral health generally. In addition, early diagnosis was felt important to offer reassurance to parents who may feel guilty that they may be responsible for the condition. Participants expressed increased sympathy for the child with MIH, who was perceived to be unlucky, in comparison to a child with caries.

'And I think, you just, it's important with this to... even if there has started to be some breakdown, so you know, there's starting to be caries in the tooth, it's quite important to reassure them [the parent] that this isn't something that they've done wrong.'

Interview 8, qualified 2003

It was felt that the general public were not aware of MIH as a condition, and that creation of resources for patients with MIH would be helpful. This early information sharing gave the participants opportunity to try to alleviate some of the uncertainties of the condition which is not well known by the general public. With MIH, clinicians are unable to give many certainties about why

it has happened, the long term prognoses and treatment outcomes. To manage some of this uncertainty, participants discussed talking about the potential need for treatment in the future, or of possible loss of the FPMs, thus giving a worse-case scenario from which any less invasive options would then seem like a good outcome.

'So, I think really getting... for the parents to realise that early on... I mean, managing expectations, like, so they're not surprised if the tooth becomes carious, eh, in the future. But then also, I always find, actually, that when I say that, that there are some parents who then latch onto that, and they'll be like 'oh the teeth are weak', and if anything does happen to the tooth, then it's because the tooth is weak. Um, so it's kind of, just, a delicate line to say, yes, the teeth are weak, but that means you have to spend more time on the weak teeth.'

Interview 4, qualified 2016

Treatment Planning Mild & Severe Cases

Management of children with mild MIH was seen as straightforward with prevention and education of the family key. Often the family were unaware of the presence of MIH when mild, so explanation regarding what may have caused the condition, and reassurance that it couldn't be prevented was perceived as important. Confidence in management of mild cases may relate not only to the low technical demand of preventive dentistry, but also to the fact that children and parents who were unaware of a condition, are unlikely to have particularly high expectations related to the outcomes of treatment.

'So, in the mildest form of MIH, patients were not even aware and nor were their parents, that their children had a mild form of MIH, as it was asymptomatic.'

Interview 9, qualified 2005

Participants were able to discuss possible options for managing severe cases broadly as restoration or extraction of FPMs, or aesthetic treatment of incisors. Although knowledgeable, confidence related to making the 'right' decision for FPMs was variable. Often this uncertainty was managed by referring to specialists who were viewed as experts able to make this decision. What constituted a severely affected molar requiring specialist treatment was different for every clinician with some referring straight from diagnosis, some after failed treatment in primary care, and some only for specialist facilities such as GA, rather than specialist opinion.

'Um, but if there are any, um, cases of, especially, severe MIH, involving the 6s I would consider doing a referral then as well, especially if it looks as if they might have a poor prognosis, making sure that they're referred, at, um, the appropriate time to, to consider having them extracted. In secondary care. [...] I feel that its better, probably better in that [borderline] case, when it can go either way, to get a specialist opinion. To get an opinion in secondary care, so that I know that I've got an opinion from someone more knowledgeable, someone who's an expert in that field.'

Interview 2, qualified 2014

'And, um, I wouldn't be trying take 6s out of people, eh, of children. I think that's a job for general anaesthetic really.'

Interview 8, qualified 2003

Poor Prognosis First Permanent Molars

Discussion of FPMs of poor prognosis was generally relating to teeth with MIH although some participants also discussed carious FPMs. Some GDPs were happy to do all the treatment themselves, feeling confident in deciding whether a FPM was of poor enough quality to warrant extraction, but most felt that the decision regarding FPMs should be made in secondary care. Participants discussed factors which made them feel concerned regarding prognosis. Often it was the cases where participants felt a lack of control such as those with spontaneous PEB, as this was viewed as difficult to prevent. In contrast, managing caries in FPMs was something participants felt confident handling. There was also a fear of doing the wrong thing regarding estimating the long term prognosis of MIH teeth, and often participants described these as borderline cases, although exactly what a borderline case was, was not elaborated on. It is likely that this was different for every participant with the bar being teeth where they felt they had exhausted their own knowledge and competence in managing.

'So, if I see cases where it's just gone, you know, large areas of caries, we try to manage it as best we can. But then we're going into the realms of orthodontics options as to what we do – what do we extract, what do we leave behind?'

Interview 5, qualified 2004

Fighting the tooth

Prevention

Prevention of further oral disease was seen as an important task for the GDP, particularly when managing MIH. GDPs discussed giving oral hygiene and diet advice, and use of fluoride, as well as broader public health methods. The use of Toothmousse™ was touched on by two participants, although only one had used it with MIH patients. Fissure sealants on FPMs was highlighted as something that could be time consuming and technique sensitive especially for MIH teeth. There was a sense that some children with severe MIH may have progression of disease even with excellent at home oral hygiene and additional professional interventions, which made participants feel helpless. In comparison, participants felt more confident that they could identify what was causing progression of caries, and therefore prevent it effectively. Some GDPs recognised that they would like to be able to spend more time giving preventive advice than they currently were able to in their practice, and within the NHS dental system. Having more time to carry out prevention and being remunerated for the additional time was seen as something that would benefit the whole population and those who particularly require additional prevention, such as children with MIH.

‘Um, I would still do fissure sealants, if I think it’s appropriate, um, because I, I, think that long term that’s going to benefit that child. And actually, realistically, I don’t think it takes that long. But again, I’m in a practice where I’m not pushed for UDAs, and I’m fully supported, whereas I think that people who are in other practices are forced to be very UDA driven, um, probably wouldn’t do that.’

Interview 4, qualified 2016

'And I think there needs to be a greater drive towards prevention, and that means, you know, starting from, you know, nursery level, or whatever, where oral hygiene instruction and education is built in as a focus of education for these children and their parents.'

Interview 8, qualified 2003

Materials & Restorations

Many participants discussed the difficulties they had experienced when trying to restore a FPM with MIH. Some of these challenges related to co-operation, however a key issue was bonding and perceived compromises in the quality of these restorations, in comparison to the quality they were able to achieve on a carious tooth in an adult. This caused frustration for clinicians and one participant felt you could never guarantee a restoration would be permanent on an MIH tooth. Related to this, clinicians questioned whether they were using the best material, and there was a consensus that the current options for restoring FPMs with MIH all led to some kind of compromise in outcome. Some clinicians were aware that PMCs could be used but did not feel confident in the technique to do so. The age of the patient, small mouth size, moisture control, variable success of LA, and stress of behaviour management, all contributed to challenges when restoring the MIH FPM. Within NHS primary care dentistry restorations are guaranteed for up to a year, which means patients do not need to pay for replacing in this time period. Although this only applies to adults who pay for treatment, the model implies that dentists are at fault if restorations fail within a year, which is unhelpful for managing children with MIH where despite best practices, restorations may need to be replaced more regularly.

'But I feel with MIH, it's, like, really difficult because you're battling with, like, the condition of the teeth, and the enamel. So, um, it's always quite hard... like much harder than with a child who just has a small carious lesion on the 6.'... 'So, um, I feel like when you do restore the teeth, it's like, never a definitive restoration – it will fail at some point.'

Interview 7, qualified 2016

'I kind of think that I might actually have done GIC, which I hate doing, GIC fillings, particularly on, um, obviously adult dentition. But I think in that situation I may have because I was worried about the bond, and also, I wasn't going to...the child wasn't super, super cooperative. So, it was going to be difficult, eh, to get good moisture control etcetera.'

Interview 4, qualified 2016

Aesthetics

Aesthetic management was a complex area for the GDP to navigate due to laws and regulations in primary care. Participants were knowledgeable about potential options such as microabrasion, bleaching, resin infiltration and direct or lab made composite restorations. The participants all preferred a minimally invasive approach, with the aim to do as little irreversible aesthetic treatment whilst children were young.

Trying to persuade patients to wait until adolescence was frequently discussed. One reason as discussed previously, was to limit irreversible treatment of an incisor that was otherwise functionally sound, to avoid entering children into a potential restorative cycle at a young age. Another possible reason related to the best timing for aesthetic treatment – as the gingival margin of teeth doesn't

stabilise until adulthood, it is possible that the aesthetic result would be better the later treatment is left. Although many GDPs are skilled in aesthetic treatment in adult patients, which would usually be paid for privately, this becomes a more complex issue when children are considered. Very few children pay for private dentistry in the UK since NHS dentistry is free until the age of 16. One participant talked about the blurred lines between NHS restorative dentistry and private cosmetic dentistry in this instance. With a congenital dental condition such as MIH it is unclear whether treatment to improve appearance should be viewed as simply managing the condition to get appearance in line with social norms, or whether it is aesthetic treatment to improve appearance from what could be considered an extreme of the social norms. This is a grey area, and most GDPs in this study tried to avoid doing aesthetic treatment until patients were older teens, or by referring into secondary care settings where clinicians are allowed to provide a much greater range of treatments, including aesthetic treatments for children with enamel defects, under the NHS.

Another ambiguous area regards the legislation about use of bleach in under 18s. The General Dental Council states that bleaching products should not be used in patients under the age of 18, unless this is for management or prevention of disease. Again, this has created a state of uncertainty within the profession as some indemnity providers have said they would support clinicians to carry out bleaching in under 18s, and others would not. Several participants expressed frustration over this situation, in which more destructive and damaging aesthetic treatments are allowed in the current system, but they did not feel supported to carry out this less damaging and often, more effective option. Again, sometimes referral to secondary care was seen as a potential solution.

'It's a case of whitening, infiltrating and then restoring the tooth, in the absolute severe cases. And that's probably where we slightly cross the GDC boundary of what's restorative dentistry and what's aesthetic dentistry. And, um, you know, that's on a case-by-case basis.'

Interview 5, qualified 2004

'Now again, going back to my DCT [dental core trainee] experience. We did, in hospital, do bleaching for these patients. On the balance of it, I personally believe that's much better than doing anything restorative to these teeth, especially at such a young age. But I know that there are like, regulations, with regards to bleaching in children under 18, and whether I would feel happy doing that in general practice myself? I would probably say no. Not because I feel that it's unsafe but because I don't feel that the regulations would back you for doing it, unless you're in a paediatric department.'

Interview 4, qualified 2016

Local Anaesthetic

Administering LA was a major source of stress for both patients and GDPs. It was also an area in which the more recently qualified clinicians felt less confident, although even experienced GDPs found it stressful. This was a problem in terms of looking after all children, however the issue was heightened for children with MIH, as sometimes the LA was ineffective, or required topping up before adequate anaesthesia was achieved due to hypersensitive pulps. Finding out the LA had been ineffective usually involved starting treatment and then the patient complaining of pain, which

affected the trust the child had in their dentist. Again, this introduced uncertainty into management for the GDP, as their usually effective LA technique may not work first time – was it the tooth, or had they done something ‘wrong’? Would cooperation now be compromised? Having to give further LA also takes additional time which may not be accounted for within the appointment, creating further time pressures. For those who had attempted restoration of the severely affected molar, increased sensitivity and increased failure of LA was noted.

‘I’ve had a couple of cases where I’ve given local anaesthetic and it’s still felt... ‘oh, this is still very sensitive’, etcetera. Um, so again, that perceived fear, that eh, actually, if this child’s going to need an intervention, um and then I’m starting to give local anaesthetic, and I’m getting everything ready, and then I try to drill, and the child’s in pain, and then they’re no longer happy with the idea of having dental treatment, um, and then I do the referral, I feel like then the child is you know, a bit more compromised in that way.’

Interview 3, qualified 2015

‘I think there’s a level for children, of, um, when they can cope with, um... I think, you use topical, and you use your best injection technique, but in spite of that, having a local anaesthetic is just a little bit uncomfortable, is a little bit painful, and I think there’s an age where they’re willing to accept that little bit of pain, for the greater good, and um, I also think there’s an age when they can’t. And you’ve hurt them once, and then they just won’t let you do anything.’

Interview 8, qualified 2003

Solutions for MIH Treatment

Despite expressing uncertainties regarding treatment, most participants offered their ideas for possible solutions that would improve the ease of management for these children. It was felt Toothmousse™ (CPP-ACP) would be better utilised if it was listed in the British National Formulary (BNF), and available to prescribe on the NHS. Having a dedicated oral hygiene nurse within the practice who could focus on this area was seen as a solution for the issue of time when giving preventive advice. Having better restorative materials and adjuncts for LA would make restoring FPMs in children easier. As well as improving care for children, the solutions suggested aimed to reduce some of the uncertainties described throughout the themes, and therefore make their working life less stressful.

‘Clinically, I do admit, I’m not super confident about what material, you know, would be ideal to use in this situation. And when I have done it, it’s like, is that enough?’

Interview 4, qualified 2016

‘Maybe access to the wand, which would be quite nice, for LA. And, um, I think that’s it really. I think it’s mainly the LA for me.’

Interview 6, qualified 2016

Working within the system

Practice Demographics

Participants worked in varied locations across England, in NHS, private and mixed practices. All participants provided NHS treatment to children. Local working environments and professional expectations were dictated by the ethos of the individual practice participants worked at and the wider socio-economic status of the area they worked in. The affluence of patient cohorts varied, and this was reflected in the generalised caries risk reported by GDPs. Most participants were aware of the increased caries risk of children with MIH. Those who worked in more deprived areas linked this to 'higher need' or higher risk of developing oral disease. Those in deprived areas also seemed to come across MIH more frequently. Therefore the 'experience' of managing children with MIH was unique to each participant and with its varied presentation, also unique for every child.

'So, um, where I... I work in East London, so there's like a really high ethnic population. So especially where I work there's a lot of, like, Bengali families, um, and I think because of that, and maybe lack of, like knowledge, um, the children tend to have a really high caries risk.'

Interview 7, qualified 2016

'Um, I guess in terms of caries risk. Well, from my knowledge, it tends to be that the 6s are more affected. Or if you have kind of more, milder forms, it's the 6s that are more affected, and that's more relevant in terms of caries risk, as its more, higher, in 6s anyway, especially if the 6s have got MIH.'

Interview 2, qualified 2014

UDA system

Much of the treatment planned and executed by clinicians was influenced by the boundaries of the current system of remuneration of NHS primary care dentistry. Many participants felt that the remuneration system was no longer fit for purpose, particularly in relation to behaviour management and prevention. There was an acknowledgement that ideally part of the acclimatisation process should involve children gradually being exposed to the dental environment, which was achievable in the current system when children came regularly from a young age and this process could occur during routine examination appointments. For children presenting with issues requiring immediate treatment, often there was no capacity within busy diaries to accommodate this gradual introduction, and no payment within the system for carrying out behaviour management alone, or over multiple visits with additional enhanced prevention.

'I have had my principal tell me a few times, being like, oh you know, 'why have you booked so and so back in?' And I'm like, you know, 'just so I can make sure she's acclimatised'. Eh, but obviously, the way the national health service remunerates dentists – that is not part of it.'

Interview 3, qualified 2015

The pressure felt regarding UDA targets was variable depending on the individual circumstances of their contract, but as an associate, some felt more pressure to be efficient and productive with principals not approving of extended time spent on behaviour management. One participant discussed a culture in some practices where dentists were encouraged to spend less time on children by placing GIC restorations in permanent teeth, to balance more lengthy treatments in adults where they were likely to lose money. Others felt this pressure less and were supported by their principle

to make longer appointments for children when necessary. Geographically, those practising away from London reported less time pressures from their practice.

'I've always been able to, sort of, book however long as I've wanted. Um, so you know, I've got 15 minutes for a kids' check-up, which is probably quite a long time for an NHS kids' check-up. And if I want half an hour to do a filling, or if I want 45 minutes to do a filling, nobody says no.'

Interview 8, qualified 2003

'Um, but sometimes, you know, you get the impression that people think, ok you'll lose money on adult treatment, but you would make that money back on paediatric treatment. You know, just a quick GIC or something, which is a horrible way to think about it. But I think, I do get the impression that there are dentists that do things like that.'

Interview 4, qualified 2016

Reasons for Referral

Participants discussed broad reasons for referral including where the child required sedation or GA, for aesthetic treatment, dental anomalies and trauma. The main reasons to refer a child with MIH regarded behaviour management issues and anxiety, for planning of poor prognosis FPMs, or aesthetic treatment. In addition, practical issues such as not having an OPG machine, also contributed to referral onto secondary care. The opportunity for closer working between primary and secondary care, and access to specialist opinion was something that was felt might support GDPs to carry out more treatment for severely affected teeth. Although the individual reason varied

for each clinician and each patient, the common barrier met was uncertainty over how to achieve the best result for their patient. Participants demonstrated a feeling of professional isolation where they were unlikely to find any answers to address this uncertainty locally and understanding of the processes and what underpins decision making in secondary care was not relayed back, offering no opportunity to learn clinically.

[On referral] 'Um, relevant to this, eh, some cases of MIH, 100%. Especially, you know, post-eruptive breakdown of 6s, where its more, you know, on the moderate to severe end of the spectrum.'[...] 'Um, we also don't have access to an OPG machine. So, eh, being able to appropriately assess, you know, the ideal time, or you know, look for the bifurcation of the 7 – that's a bit more challenging.'

Interview 3, qualified 2015

'And dare I say, um, the primary care sector, on the NHS, I feel, has a huge question to answer on this, because there is, almost, very little collaboration, or little possible collaboration between orthodontists, NHS primary care, um, general practitioners and secondary care. There's almost... it's almost impossible to be able to properly approach this on, like a, multiple disciplinary level.'

Interview 5, qualified 2004

Referral – Positive and Negative Experiences

The referral system was seen as both a saviour and adversary. It was there to take over care when co-operation proved impossible, or to negate uncertainties in planning or treatment. However actually making a referral was the source of a massive amount of stress regarding bureaucracy, rejected referrals, and timing. Only one GDP talked about positive experiences of referral. The

negatives of the referral system and the frustration this caused the participants was discussed at length. Participants acknowledged long and increasing waiting times for referral and for treatment such as GA. This created difficulties regarding trying to refer at the best time for the child to have removal of FPMs, and guilt if they started treatment which they were unable to finish, essentially leaving a child suspended halfway through a treatment plan for months, before secondary care was able to finish the job. Having to send referrals for the same child more than once, either due to rejection or because that child had failed to attend their hospital appointment was another issue. They also wrestled with the uncertainty of whether they should refer potentially difficult cases from the outset to avoid getting stuck midway or making the patient more difficult to manage in a hospital setting by having done the wrong thing. Participants commented on both the perception that some colleagues didn't utilise referral enough, but also that other clinicians were over referring, causing them to deskill in the management of children. Some clinicians felt their referrals were readily accepted, some felt they were rejected for no good reason, and others thought that hospital and community settings accepted too many referrals from colleagues that should really be dealt with in primary care.

'And then if we refer to secondary care, then we start a bureaucratic nightmare, where we have to exchange 20 different referral letters because the NHS number wasn't on it, because the parents don't know the NHS number, because the hospital lost the referral, because the patient didn't turn up for their appointment, so their referral got cancelled. So, we've got to refer them again, and then we've got to wait another 8 weeks, 12 weeks, 16 weeks, and so on and so forth. It's a bureaucratic nightmare. So, I find myself, the biggest issue I have, is when I want an orthodontic or a specialist opinion on an ongoing case, which are the more severe cases. Sorry to rant.'

Interview 5, qualified 2004

'Being able to get them a GA at the time that they need it, I suppose! You know, because you sort of need to be thinking quite a long time ahead. If they're going to need a GA, you need to be thinking about it. I kind of try to get them referred and hope that they'll put them on like a... okay so they're only 9, 10 or whatever, they're going to need poor prognosis 6s extractions, but not yet. So, I kind of hope that they'll keep them on a - 'oh yeah, we'll see them once a year basis' - until they're ready. Um, because otherwise it's like, when I think it's time, I might miss the window or I might refer and there's a really long waiting list for a GA, you know. They might be in pain before they have it, do you see what I mean? I tend to try to refer quite early, because of the problems with the NHS.'

Interview 8, qualified 2003

Participants offered up possible solutions to some of the perceived systemic issues, or ways to reduce uncertainty. This included better liaison between primary and secondary care, UK based guidelines on management of MIH for primary care, and the opportunity to shadow specialists in secondary care to improve the translation of knowledge to decision making. In terms of referral, being able to receive a quick opinion from a specialist without actually referring, was one way to avoid waiting lists. Another was to allow children who had been planned in secondary care previously but had failed the treatment under LA in primary care, to be seen again without a new referral. Both methods would require excellent information sharing between primary and secondary care, and major changes to current referral pathways. In addition, one clinician felt that the letter they received back from secondary care could be more in depth and could potentially act as a learning opportunity for the primary care clinician.

'Um, possibly better access to advice, over the phone, to like secondary care, so if there were any questions or queries you would be able to pick up the phone, maybe, and ask a simple question. [...] so even if it's simply sharing an image of a tooth with somebody, or like, a consultant, being able to discuss a case. It would be quite nice to be able to do that.'

Interview 6, qualified 2016

'So nowhere in that [response to referral] letter does it say, the reason we have chosen, eh, to extract this tooth, is because of this, that and that. It will just have, eh, 'thank you for referring... the treatment plan is... extraction of this tooth under general anaesthetic'. And therefore, it limits your development to be able to make any kind of, you know, to feed into that decision-making process, or to gain from that decision-making process.'

Interview 3, qualified 2015

Self and Interpersonal insight

Relationship with the Child

The importance of developing a sound relationship with the child to allow treatment of MIH affected teeth was a key goal for participants. As a child with MIH may require invasive dental treatment using LA from the age of six, a good rapport was felt to increase the chances of success. Fear of LA, fear of the unknown and the patient's age were the most commonly encountered hurdles to cooperation. Participants expected young children to be innately anxious, and they took responsibility to reduce this anxiety prior to commencing invasive treatment. As well as acclimatising the child, regular attendances also allowed for acclimatisation of the *dentist* to the child. As the dentist becomes more familiar with the child, they become better able to accurately assess cooperation and are likely to have more success when it comes to treatment. Participants discussed

many different behaviour management techniques they utilised, including bringing patients back for follow-up reviews if treatment had not gone to plan at the last visit. This offered an opportunity to rebuild trust with the child, so that they would feel comfortable to return for routine care later. This was not a technique used with adults, so it seems that the participants placed more importance in ensuring positive experiences for children in their formative years. Referral for behaviour management reasons was usually seen as a last resort, with continuity of care locally the preferred option. Conversely, there was a perception that secondary care clinicians had increased behaviour management skills, better access to pharmacological options and importantly, fewer time pressures.

'I feel like if I can't do something first time round, we'll leave the appointment, and have, you know, like a really positive appointment, and give them OHI and give them stickers, and just, um, get to them to leave on a like a happy note. And then I'll always, like, see if we can try again, just to try to win them over, to be able to do the treatment.'

Interview 7, qualified 2016

Generally, participants reported that treating children took more of an emotional strain than adults. Participants were especially sympathetic to children with MIH as they viewed it as something that couldn't be prevented. Managing an anxious child, extracting teeth as an emergency and giving LA was reported as stressful. An element of uncertainty exists in all of these situations, particularly since the clinician is less able to predict how successful treatment is going to be at the outset, in comparison to treatment for adults which is much more likely to go to plan. Children with severe MIH who require invasive treatment are much more likely to be anxious, and LA is less likely to be successful, lending these situations to be particularly nerve-racking for the GDP. Participants also discussed their fear of being the cause of dental anxiety for the child, which may also be a factor in

deciding to refer a child. The stakes are perhaps higher for the general dentist, who have life-long relationships with many patients, in comparison to specialists who are more usually only involved in a single treatment plan.

'So, for me, having a day full of, you know, surgical teeth needing to be drilled out, or super complex procedures, is a lot less mentally draining, or cumbersome, than a day where I've had to manage a series of anxious children. [...] With a child, you feel a bit more of a sense of duty, to be like, you know, well they might not understand the consequences of their actions. So, you put a lot more into, you know, the convincing, and into, you know, bringing them along with you.'

Interview 3, qualified 2015

'My worry is that I'll traumatise the child and then they'll refuse any kind of treatment, lose any kind of cooperation in the future. [...] I think it's a life-long, you know, a journey. It's not just a one step with the general practitioner, it's a continuous management.'

Interview 1, qualified 2014

Influence of the parent

Dentistry for children is unique in that the relationship is between three people – the child, parent or carer, and the dentist. Where there is good rapport with both child and parent, the relationship is harmonious. When either child or parent is working against the dentist, this creates tension.

Participants discussed having to manage the behaviour and expectations of the parent, in addition to the child. This took several forms, including parents unhelpful voicing of their own dental anxieties in

front of the child, and becoming frustrated and chastising the child when they wouldn't co-operate. It also included parents pushing for treatments that the clinician did not feel were appropriate.

'Sometimes you can get someone [child] who's brilliant, and you know, the parents, it's just like 'just shut up!'. 'Just sit there, quietly!'. And don't say anything, and don't, you know, try and, you know... and I'll talk them through it, using appropriate language, which is hopefully going to be better than, you know saying things like 'oh it's not going to hurt' ... you know. It scares them.'

Interview 8, qualified 2003

For the more recently qualified GDPs, the pressure simply from being watched whilst working was experienced acutely. In addition, when treatment didn't go to plan and had to be abandoned due to co-operation, there was a fear that parents may equate this to incompetence. It seems that some participants put a lot of pressure on themselves to get things 'right', and in situations where there is increased uncertainty, such as when trying to restore an MIH FPM in an anxious child, that the dentist projects their own worries over getting things wrong and experiences this as perceived judgement from the parent. The more experienced dentists seemed less susceptible to this type of self-doubt and were more adept at handling disagreements with the parent.

'But the other one is just them looking over your shoulder, just having someone there, watching you, for an hour. For example, I'll sometimes have two children who come together at the same time, and both parents will just stand there, watching you for the whole hour. That in itself can just be a little bit, a little bit daunting.'

Interview 10, qualified 2019

'So in that particular scenario, eh you know, just in terms of self-perception, you know, you feel like, you know, is this parent starting to question my judgement, because I've laid out a comprehensive treatment plan, and I wouldn't have laid it out, had I thought it wasn't achievable, and yet there's direct evidence that, you know actually, the child didn't do it, so you know, does this dentist know what they're doing?'

Interview 3, qualified 2015

Many GDP also discussed positive experiences relating to the presence of the parent. Some parents were extremely helpful from a behaviour management point of view, letting the GDP take the lead on the techniques implemented, whilst supporting the child. Having the trust of the parent when treating a child was seen as highly important, with children taking the lead from the atmosphere created between GDP and parent. GDPs also discussed taking the measure of the whole family, as an indicator of likely compliance both in the dental chair, and with oral hygiene and diet. This agreeable type of parent were the regular attenders who showed interest in advice from the dentist and engaged in recommended lifestyle change. Although it is easier to build a strong relationship with a family that attends regularly, it is likely that GDPs also feel more comfortable when parents are interested and allow them to control the environment. Parents who counteract methods used by the dentist create uncertainty regarding treatment outcome, as both GDP and parent compete for attention of the child.

'We've got quite good, quite motivated parents who want to try really hard with their children's teeth. Most of the ones that I see.'

Interview 8, qualified 2003

'I think also, the other aspect that plays into it, is, you know, the parents of the child. Eh, as professionals and clinicians, a lot of what we do relies on, you know, having the trust, you know, of our patients, and you know, their family members.'

Interview 3, qualified 2015

Self-perceptions

The perception of participants regarding their role in MIH varied from clinician to clinician. Some saw themselves as the most important clinician for the child with MIH, due to the role they played in making the initial diagnosis, educating the family, and in continuity of care for the long term, with or without referral. Others saw themselves as a clinician who linked the patient to part of the wider dental team when necessary, with support sometimes needed from specialists in paediatric dentistry and orthodontics. Most GPs felt that decision making for severely affected molars presented a challenge, although one participant felt confident to make the decision in primary care. It appeared he had a clear and robust strategy for his decision making, which allowed him to reduce the uncertainty felt by other participants and avoid referral in these cases.

'I see like my role as probably the most... yeah, probably the most important dental sort of person in the patient's timeline because I'm going to be seeing them probably every 3 months. I'm going to be applying fluoride. I'm probably going to have the best relationship with them. When it comes to referral, I'm probably going to be doing that. When it comes to making the decision, or helping them make the decision about extraction, I'm probably going to be doing that. If it comes out, and then we have a space, the management is probably going to be coming from me, or from me referring to someone else. So, I think that's quite an important role.'

Interview 10, qualified 2019

'And I think when it's more severe, it's quite difficult to know what to do for the best really. And I quite often tend to refer them when I see a more severe one.'

Interview 8, qualified 2003

[Do you ever refer children with MIH to specialists in paediatric dentistry?]

'Not so much for MIH because I think the management of it is pretty straightforward, you know. And I think ultimately the treatment of a tooth with MIH will be determined by the severity of the symptoms and the presentation, and I think, you know, depending on that, you can scale your degree of intervention, and that's a decision I can make without needing a paediatric specialist opinion.'

Interview 9, qualified 2005

Participants were asked to reflect on their previous education regarding MIH in the interviews and how they had applied this knowledge. Recently qualified clinicians felt that they had had a robust education on MIH as an undergraduate, which they had built upon and adapted for clinical practice. Those qualified less recently had developed a greater understanding of MIH in general practice, and through postgraduate courses. Most clinicians felt they had a sound education, however they were also able to talk quite extensively about how treating MIH patients had been more difficult than expected, and possible resources or training which might make them more skilled in management of these patients. From these interviews it appears that participants have a good knowledge base, but that this doesn't always translate into adequate clinical experience and confidence. One participant who had spent time in a paediatric department as a postgraduate felt she was much better able to manage children in general and suggested that actual clinical exposure to patients with MIH under the guidance of specialists was the best way to learn.

[Has your experience of MIH as a GDP changed your understanding of MIH since you were first taught about it?]

'Changed my understanding? Not particularly. I'd say I was lucky to have quite an extensive, you know, undergraduate training on MIH. We spent quite a lot of time on it.'

Interview 3, qualified 2015

'So, my first experience with MIH was really at university, so in 2002 and 2003, in the Paeds Department. I think MIH at that time, and this is more my anecdotal opinion more than anything, from my memory, MIH was really very poorly understood. It still is really poorly understood, I think.'

Interview 5, qualified 2004

Other GDPs

Some participants reflected on other clinicians' management of children with MIH that they had come across which they felt could have been improved. Some commented on the perception that GDPs that had come before them may have misdiagnosed MIH. Fluorosis was a commonly cited diagnosis, which the participants had to then inform the patient and family was incorrect. This led to confusion for the patient, and participants then felt they had to work harder to regain the trust of the family, despite it being the previous clinician's error. This introduces an element of uncertainty into the relationship between dentist and patient, and also more widely to the profession. Medical professionals traditionally have a high level of trust from the public, and for some this may cast into doubt the professional standards for dentists. UK dentists are the most sued in the world and anecdotally there appears to be a growing distrust of professionals worldwide. Getting simple diagnoses wrong will not help in the public image for those who already have reservations about dental probity. In addition, it was felt this also had the potential to create missed opportunities for optimal care, such as extracting FPMs, rather than restoring. Participants tended to feel sympathetic towards the patient in these situations, having perhaps not had the most optimal treatment leading up to this point.

'It's sometimes, you know... your memory misleads you. But from my memory, um, it's been common where we've seen patients who've had a 6 taken out... let's say a lower left 6 has been extracted. [...] The other lower 6 has been root treated, not doing very well. And then you start looking at the other 6s that have survived a bit better, and then you see that they have brown patches on them. You know the typical, orangey, brownie colour that the teeth can go. And then you start to think, well this poor kid, actually, just, it's not as if they might have had the worst, kind of, OH in the world, but these teeth were really vulnerable, and

they've just not been protected properly, and the patient has not been aware from a very young age.'

Interview 5, qualified 2004

6.5 Discussion

Exploring how GDPs understand MIH

GDPs were familiar with MIH as a concept and could easily describe a mildly and severely affected tooth, even for those who didn't report they had treated any severe cases. It was generally understood to present as a spectrum of disease, with increasing complexity in treatment planning and clinical management for the severe cases. Mild cases were universally seen as easy to manage, however disagreement on whether GDPs should be treating severe cases existed. Despite the competence of GDPs in discussing aetiology, presentation and treatment options, some still used the term 'hypoplastic' interchangeably with 'hypomineralisation'. It was also perceived that other colleagues may be misdiagnosing MIH, or not diagnosing it at all. In study two, GDPs were asked to diagnose and treatment plan cases of MIH based on clinical vignettes with photographs. There was varying success at diagnosis with the more severe cases being more easily diagnosed. In addition, where there was caries, or the condition only affected molars, GDPs were less likely to diagnose MIH. These findings also correlate with study one, in which it was found around 17% of referrers referred for MIH specifically, with a further two-thirds referring for another enamel defect, most often 'hypoplasia'. The presence of MIH is known to affect anxiety and cooperation, bonding, and efficacy of LA in comparison with children who have caries alone (Jalevik and Klingberg 2002, Rodd et al. 2007, de Souza et al. 2017). A clinician who treats a child with MIH without diagnosing it, may be surprised with the additional demands required to manage that child and achieve a quality restoration, and may also have missed opportunities to implement additional preventive measures.

Although the system of referral could be frustrating to navigate, it meant clinicians were able to refer the severe cases into secondary care and therefore had to deal with less of the issues related to severe MIH itself. Therefore, although there wasn't absolute confidence in treatment planning, MIH wasn't seen as a uniquely problematic condition, since the difficult cases would usually be accepted into secondary care in most cases. Guidelines exist regarding balancing and compensating FPMs when enforced extractions are required (Cobourne et al. 2014), however no guide exists to define how to estimate prognosis and which tooth should be extracted. This is a complex decision to make as it not only takes into account the current condition of the tooth but also estimating oral health behaviours into the future. The same guidelines recommend an opinion from a specialist regarding loss of FPM teeth (Cobourne et al. 2014). Most participants appeared to follow this guidance and would refer for decision making regarding extraction. In study one, most children were referred for management of poor prognosis FPMs, and most required GA to aid removal. In contrast, a recent paper by Alkhalaf et al considered whether too many FPMs are extracted in children and suggested that development of high-viscosity reinforced GIC may enable some severely affected teeth to be restored successfully in children with limited cooperation before definitive cast restorations are provided during adolescence (Alkhalaf et al. 2020). Further research is required to precisely define a poor prognosis FPM, and when restoration may be an option instead of extraction, in the long-term.

To explore how initial education about the condition has been translated into clinical practice

Most recently qualified GDPs felt that they were well taught about MIH as undergraduates and had not learnt a great deal more since. Some who graduated almost 20 years ago remembered some teaching on hypomineralised FPMs, even if it wasn't explicitly about MIH. The more experienced GDPs had updated knowledge through clinical experience and courses. Some felt that the condition was becoming more prevalent, or simply that the reality of how frequently they came across it in

their patients was surprising to them, and they hadn't quite grasped this as an undergraduate when presented with prevalence data in a lecture setting. Similarly, some participants reflected on how some practical aspects of management had surprised them despite being taught about them, such as issues with sensitivity during treatment, and poorer bonding. A couple of participants felt that they had learnt nothing new, apart from greater understanding of aesthetic options which could be used for hypomineralised incisors generally. The greatest insight came from the participant who had worked in a paediatric department as a DCT, who reflected on how her book knowledge had developed into clinical understanding. Her insights included that despite being taught about MIH quite thoroughly at university, when she came across lots of children with caries in MIH molars she was still surprised. In addition, in her hospital position she had witnessed the improvements to appearance that could be achieved by vital bleaching, the simplicity of the procedure, and the massive effect this had on the self-confidence of patients. She felt that she 'hadn't quite grasped' that MIH was such a 'big deal' as an undergraduate. Elhennawy et al carried out a survey of final year students in Germany to assess their knowledge and confidence in management of MIH (Elhennawy et al. 2020). They found although their basic knowledge was good, that they were not confident in decision making. The authors discussed the possibility that this was down to insufficient exposure to children with MIH clinically as undergraduates. The same theory may explain why the participants in this study found themselves being surprised clinically by things they had been taught in lectures.

Although no participants felt critical about previous education, most felt that there were elements of knowledge, or skill that they might be able to improve on. In study two, 96.5% of GDP felt more information about MIH at undergraduate and postgraduate level would be useful, which correlates with the findings from this study. In surveys done with GDPs outside the UK, between 36.9% and 90.5% also wanted further training on MIH, and there was a higher demand for information

regarding treatments and treatment planning, than aetiology or diagnosis (Hussein et al. 2014, Ghanim et al. 2011, Silva et al. 2016a, Alanzi et al. 2018). This may reflect the complexity of treatment planning for severe cases and the practical aspects of treating the compromised tooth, and young child.

No previous qualitative work has investigated the general dentists' education regarding MIH, but many surveys have addressed self-reported confidence. Results from Europe, the Middle East, South East Asia and Australasia, have found that generally 50% of GDPs surveyed felt confident in the diagnosis of MIH (Crombie et al. 2008, Kalkani et al. 2016, Silva et al. 2016a, Ghanim et al. 2011). In the UK, Kalkani found that 57% of the 31 GDPs attending a study day on paediatric dentistry felt confident or very confident when diagnosing MIH (Kalkani et al. 2016). In terms of confidence when treatment planning, Hussein et al found that 74.2% of GDPs would feel comfortable managing a child with MIH, and 57.1% would refer to a specialist. Gambetta-Tessani found that 62.1% and 83% felt confident treating children with MIH in Chile and 83% Australia respectively (Gambetta-Tessini et al. 2016). In study two, 42.1% of participants were confident or very confident in treatment planning a child with MIH. Differences may exist as participants in study two had just carried out diagnosis and treatment planning for MIH within the vignettes, which may have highlighted learning needs. It appears undergraduate education prepares the GDPs well to treat mild cases, however some severe cases may not be suitable to treat in primary care due to systemic barriers and difficulty in planning and executing treatment.

To identify how GDPs perceive their own management of children with MIH and any challenges they encounter

The overarching theme of uncertainty was reflected in the management strategies that the participants discussed, and the challenges they encountered when doing so. No participants discussed finding MIH hard to diagnose, however some commented that they felt other clinicians may have misdiagnosed, or not diagnosed MIH at all. The findings from study one and two were in agreement with this. All three studies suggest that although some clinicians are skilled in diagnosis, there may be some who are not aware of the full diagnostic spectrum of presentation, leading to missed diagnosis in very mild cases.

When considering the planning and execution of treatment, participants expressed variable levels of self-doubt and uncertainty. Decision making related to restoring or extracting FPMs, and the right time to do so was something that the majority of participants referred to specialists in orthodontics or paediatric dentistry to manage. Participants discussed the concept of a poor prognosis FPMs without explaining fully what that meant to them. Taylor et al found that UK based GDPs were more likely to restore a FPM than specialists, indicating that there is not a general consensus in the UK (Taylor et al. 2019). Likewise, there was little consensus on planning poor prognosis FPM with MIH amongst specialists in the UK in a recent study which asked specialists in orthodontics and paediatric dentistry to treatment plan FPMs in comparison to expert consensus (Alkadhimi et al. 2021). Few studies have looked at long term outcomes for children who have had FPMs extracted (the preferred option in the UK) versus FPMs restored (the preferred option elsewhere) (Elhennawy et al. 2017). It appears that a poor prognosis FPM is therefore an arbitrary measure based on clinician experience, which is understandably confusing for many dentists.

Participants also expressed uncertainty regarding issues whilst treating children with MIH, related to both the tooth and the system of NHS dentistry. LA was already something that participants found stressful, but with MIH children who were more likely to be anxious and have hypersensitive pulps, the stress increased. LA has been discussed as a source of tension and challenge for cooperation for GDPs in previous qualitative work. Marshman et al carried out individual and group interviews of GDPs involved in the FICTION trial (Marshman et al. 2020). In the conventional arm of the randomised controlled trial, LA was discussed as a reason some clinicians avoided conventional restorations in children, while another discussed being reliant on support from the parent and avoidance of negative conversation related to injections prior to the procedure. Participants in this study reported similar scenarios. Patient anxiety regarding dental injections is well reported and is particularly high in young children (19% of four to six-year-olds), but gradually decreases and plateaus at 11% by age 10 (Majstorovic and Veerkamp 2004). Dower et al conducted a questionnaire of dentists in USA, and found that the biggest cause of anxiety for dentists during injections were anxious patients (67%) and children (16%)(Dower et al. 1995). It is clear that further research to improve anaesthesia specifically for children with MIH would reduce stress for both dentist and child.

Difficulties with bonding were described, which resulted in restorations that needed frequent replacement. One participant described restoring an FPM with MIH as 'a battle'. In some circumstances, participants compromised by using a less durable but less technique sensitive option (GIC). These issues have been discussed in many studies and were reported as barrier to care and reasons for referral in study two, however, the frustration and self-doubt that this caused the participants hasn't been explored before. Avoidance of uncertainty itself, may be a driver for referral regarding MIH. Research has previously been conducted to investigate retention of fissure sealants and composite resin restorations. It has been suggested that the addition of an adhesive prior to

placement of the fissure sealant may increase retention (Lygidakis et al. 2009), in addition to treating with sodium hypochlorite prior to etching to dissolve the excessive amount of protein in hypomineralised enamel (Lagarde et al. 2020). Kopperud et al discussed the importance of finishing margins within sound enamel to increase retention, which goes against the current ideologies for minimally invasive dentistry (Kopperud et al. 2016). Although these techniques will increase retention in controlled conditions, they may be more difficult to undertake in clinical practice. Jalevik reported that restorations were replaced four times as often on MIH affected teeth, which is in keeping with the experience of the GDPs in this study (Jalevik and Klingberg 2012).

Another area of ambiguity was managing aesthetic concerns in children. Working within the UDA system under the NHS, it was unclear for some participants whether they should be carrying out work to mask hypomineralisation on the incisors. In addition, most would not feel able to carry out vital bleaching in primary care due to current legislation. The General Dental Council's statement is itself ambiguous, leading to indemnity providers disagreeing over whether tooth whitening can legally be carried out in children under the age of 18, in *any* circumstance. Some participants felt quite conflicted regarding this situation, and most opted to delay any aesthetic treatment as late as possible. For some, they opted to refer children into hospital settings where more flexible rules exist regarding aesthetic treatments. Participants also felt restricted regarding time for acclimatisation, behaviour management and prevention. This meant in order to work within the system (and get paid for the work they have done), they often compromised on how they would ideally like to manage children with MIH. Participants in the FiCTION trial discussed how they felt able to give more time for patients in the 'prevention alone' arm, than they would normally have outside the trial (Marshman et al. 2020). Arheiam et al found that remuneration was also a barrier to using diet diaries in his survey of GDPs (Arheiam et al. 2016). A report into primary care contracts found that GDPs felt that the 2006 contract for dentistry had increased the feeling of pressure related to

targets, and had reduced the focus on preventive care – the opposite of the intended purpose (McDonald 2010). The current system therefore presents a lose-lose situation where GDPs are likely to feel guilty no matter the option they choose. Changes to how GDPs are paid for prevention and behaviour management will be important for the child with MIH and would also improve the working environment for the GDP.

As discussed above there were many reasons for referral, but all involved reaching a barrier where the participant was not able to proceed with any more treatment. In many cases this was a clinical barrier, such as decision making on FPM or a request for bleaching in a secondary care facility. The interviews also demonstrated that in some cases participants reached an emotional barrier. These existed when participants reached the limits of clinical uncertainty they were able to tolerate. In study two, a commonly cited reason for referral was cooperation or due to anxiety, which corroborates with the findings in this study. A survey of GDPs in Liverpool found that anxiety and need for GA or sedation were the most common reasons for referral to hospital or CDS (Harris et al. 2008). In a recent paper, Mills et al discussed the shortfall of specialist paediatric dentists in relation to other specialities, and information obtained from the General Dental Council demonstrated that 44% of postcode areas in the UK were without a specialist in paediatric dentistry (Mills 2020). The number of specialists has plateaued over the last 20 years, whilst the population of children continues to grow, leading to increased waiting lists as demand outstrips supply (British Society of Paediatric Dentistry 2020b). In a recent letter to the British Dental Journal, a DCT discussed his experience of managing the care of children in the South of England with extremely long waiting lists of up to a year (Yoong 2020). This he described as having his 'hands tied behind [his] back'. Some participants of study two advised they wouldn't, or couldn't refer because there was no specialist paediatric dentist locally and waiting lists were too long. The GDPs in study three shared a similar feeling of powerlessness when it came to referral. It is clear that the current status quo means that

children who require specialist dental care, are not being cared for in an acceptable timeframe, and the GDP shoulders the burden of this stress.

The challenges met during management of children with MIH in primary care were at tooth, child (and family), clinician, practice and systemic levels. Participants described 'battling' the composition of the FPM when giving LA, managing sensitivity, bonding to the tooth and trying to prevent PEB. When it came to managing the child the importance of the relationship between GDP, parent and child was key to enabling treatment to take place. Cooperative children with supportive parents were the easiest to work with, however anxious children with vocal parents created an uncertain environment for the GDP in terms of predicting the dynamics of the treatment session. The participants expressed differing levels of skill and knowledge related to performing treatment for MIH teeth, and level of confidence appeared to be another factor in referral. Additional to this was the influence of the practice itself. Barriers included not having an OPG machine, and therefore not being able to estimate the best time for removal of poor prognosis FPMs, perceived or real pressure from the principal to hit targets, and the general motivation and level of disease present in the local area the practice serves. The actual system of NHS dentistry itself was the final barrier. The UDA system doesn't give any explicit payment for behaviour management and only a single payment of the value of one UDA for preventive care per treatment plan, which means there is no incentive to spend additional time on these areas which are important for children with MIH, and for children generally. In addition, legislation surrounding vital bleaching in children, and the UDA contract meant that although most participants were familiar and confident with aesthetic options, they often did not feel able to carry this treatment out for children with MIH. Referral was a useful adjunct to manage many of these challenges, but the referral process itself was challenging as GDPs tried to get the timing right taking into account waiting lists. Ultimately the participants expressed

professional isolation from specialists and hospital dentists, leaving little opportunity to learn and develop from the referral process.

Strengths and weaknesses

The interview schedule went through several versions before finalisation. Through an iterative process and feedback from experienced qualitative researchers who reviewed the schedule and pilot transcripts, open and exploratory questions were developed. An opening question exploring the GDP's general experience of treating children in primary care allowed for a more relaxed and open conversation that led naturally into the topic of interest and allowed for discussion of the child with MIH in the context of children generally. It is possible that the questions selected could have been refined further to better answer the research question. In particular, the answers regarding previous education and translation into clinical practice lacked depth and could have been improved through the core topics and with follow-up and probing. The lead researcher had not carried out any previous qualitative interviewing, and therefore on analysis of the transcripts it was noted that more targeted questioning may have clarified some statements made by participants, or that some areas could have been explored in more depth, such as the relationship between previous education on MIH and current practice. Further experience in qualitative interviewing will improve the quality of work in the future.

The researcher did not meet any of the participants, and they were not known to her prior to the study. A reported downside of telephone interviews is that the information collected is not as in depth as face-to-face interviews, with the balance of conversation more equal between interviewer and participant (Irvine 2011). This was not the case for these interviews, with the interviewer speaking relatively infrequently in most interviews, however better probing may have taken place in

person, with the interviewer able to read non-verbal cues. Perhaps the dental practitioner is used to having one sided conversations with their patients, and therefore were able to speak at length about MIH without much prompting. Field notes were taken during the telephone conversation to aid analysis of the transcripts, however comment could not be made on non-verbal communication which may have imparted additional nuance to content. Only 10 interviews were completed which means the work is more exploratory in nature, and makes it difficult to prove whether data saturation was reached.

The timing of the recruitment was fortunate in that clinical practice was severely reduced due to the COVID-19 pandemic. This meant that GDPs may have been spending more time at home, and therefore had more time to be involved in research. The researcher cannot comment on the reasons that each participant wanted to be involved in the research, however as a group it gave them the opportunity to voice difficulties within NHS primary care dental services, and general dentistry for children in an anonymous way. Participants received £10 shopping e-voucher, but this is unlikely to have been a large influence in their participation. As expected, more female GDPs contacted the researcher to participate. Traditionally the speciality of paediatric dentistry and the CDS are staffed mainly by a female workforce. The purposive sampling technique aimed to minimise this bias. More recently qualified GDPs showed an interest in participating and experienced GDPs were harder to recruit. Many will have additional managerial roles, such as practice principals, or personal commitments which may make finding the time to participate in research difficult. No participants had been qualified more than 20 years which may be in part due to the reasons already stated but may also be related to their familiarity with MIH as a concept, since the term has only been in use for the last 20 years. The results are therefore biased to reflect the experience of the clinician qualified within the last 20 years. In addition, a high proportion of the participants practiced in

London, and no participants were recruited from outside of England. Therefore, the results only reflect those GPs who work in primary care in England, with a London bias.

Reflexivity

The interviews were carried out by a single person, the lead researcher. Participants were not made explicitly aware of the interviewer's position as a postgraduate student in paediatric dentistry however email correspondence to exchange consent forms was from a university email address which demonstrated that the researcher was an Academic Clinical Fellow in Paediatric Dentistry. The interviewer's knowledge of clinical dental practice allowed for relaxed conversation with the GPs without need for explanation of technical terms. Her position as an academic may have influenced how the participants answered questions, however other influences such as the age or appearance of the researcher were not factors due to the interviews taking place remotely. The analysis will have been framed within the context of the lead researchers' knowledge and experience of clinical paediatric dentistry, which would differ from those who are not clinically trained, or those who do not treat children regularly. To increase rigor, a second researcher read the transcripts and reviewed the coding, and changes to coding and themes were then agreed by the whole research team. The themes were revised several times before the final versions discussed in the results.

Further Research

Further work regarding optimising treatment for children with MIH is required. Issues of bonding and restoration longevity are frequently encountered, in addition to the difficulty of performing operative dentistry in children under the age of 10. A material that is not moisture sensitive, can be placed in single or bulk increments, has a desensitising effect on the pulp and excellent bonding properties to enamel with reduced mineral content, should be developed so that restoring an MIH

tooth becomes quick and straightforward. Pulpotomies of adult molar teeth is not a procedure on the NHS scope of practice for primary care currently but are straightforward to complete in comparison to full molar endodontic procedures. In addition, little qualitative work has looked into how children experience care during treatment of MIH. This would help to ensure treatment provided is meeting the needs of children, from their own perspective.

Reform of the UDA system in primary care is needed so that two cornerstones of paediatric dentistry – behaviour management and preventive care – are rewarded appropriately. Currently GPs do not feel able to spend sufficient time on these areas, which would have lifelong benefits for children and would likely save the NHS money on treatment of caries and expensive adjuncts like sedation or GA, down the line. In addition, communication between primary and secondary/tertiary dental care should be improved so GPs feel more supported to complete more care for children with MIH, in the knowledge they can access guidance and advice from a specialist easily. Further research would be required to investigate what clinicians in each area require, and potential methods of implementing change to address these areas within the budget of NHS dentistry.

6.6 Conclusion

Despite being knowledgeable about MIH, participants expressed varying levels of uncertainty in all aspects of management for children with MIH. Mild MIH was easy to manage, and did not challenge the participants, however severe MIH presented issues when dealing with both molars and incisors for the primary care dentist. The level at which the child became ‘complex’ was different for each clinician and related to their previous skill and experience, and current work environment. Often referral was used to manage this uncertainty, however, ultimately this did not result in the participant overcoming these same barriers in the future, due to a lack of multidisciplinary working between primary and secondary care. Beyond the participant themselves, fundamental flaws exist in

the current system of remuneration for primary care dentistry which makes managing children with MIH difficult, and participants relied on colleagues in secondary care who do not work within the UDA system to manage those children.

Chapter 7 – Conclusions & Future Research

7.1 Conclusions

The aim of this thesis was to investigate how children with MIH are managed in primary and secondary care in the UK. A service evaluation was designed to assess the management of children with MIH locally in a specialist dental hospital setting. The study also analysed pre-referral treatment, which linked into the second study which used clinical vignettes to assess the accuracy of UK based GDP in diagnosing and treatment planning children with MIH. In the third study, semi-structured telephone interviews explored the experience and understanding GDPs have regarding managing children with MIH. The detailed research findings from the three studies are discussed in chapters 4, 5 and 6.

This thesis demonstrates how children with MIH may experience dental care within the current primary care and hospital systems in the UK. Participants of the interviews discussed at length their pivotal role in the first line management of these children as GDPs, including identification of the condition and implementation of preventive strategies. However, evidence from the surveys demonstrated that GDPs accuracy in diagnosis was affected by the presence of caries on FPMs or other teeth, milder presentations and when only molars were involved. In addition, fewer clinicians were aware that children with MIH should receive more intensive prevention, including more frequent application of fluoride varnish. Difficulties in treating children with MIH are already well documented. The interviews identified that completing prevention and restorative procedures in practice was often difficult. Frustration was expressed regarding restorations that frequently failed, stress surrounding provision of LA, and behaviour management issues that were difficult to control without adjuncts. Referral was dealt with in all studies. Local data from 2015 demonstrated that almost all referrals into this hospital setting regarding MIH were of appropriate complexity to merit

specialist care. A catch-22 situation was described by participants of the interviews, where GPs wanted to do as much as possible locally to avoid long waiting lists on referral, however they were fearful of doing the wrong thing, or causing dental anxiety by trying to do too much.

Through the interviews it became clear that the current system of remuneration in primary care makes providing dentistry for children under the NHS system difficult. This includes the UDA system, where no payment for behaviour management, and only a single payment for preventive care for each patient exists. When we consider children with MIH are more likely to be anxious, should have enhanced preventive care, and may have fissure sealants or restorations which need frequent replacement, it is particularly obvious this system does not work for these children. In addition, when GPs opt to refer into secondary care, they are often met by long waiting lists. Due to professional isolation and lack of access to specialist advice, during this period the child is left in a 'no-mans-land' of dental care, without the specialist care they need and with GPs unable to bridge the gap.

7.2 Recommendations & Further Research

- Education which addresses MIH should address the diagnostic process and situations in which diagnosis may be more challenging. Case based discussion with real cases of MIH should also be utilised so practical tips for management of the child with MIH in real life, and solutions to manage uncertainty can be discussed.
- Resources should be developed to guide GDPs in decision making in primary care. Delivering Better Oral Health(Public Health England 2017) should specifically identify MIH and other developmental enamel defects at increased risk of caries. They should also be developed for families with children who have a diagnosis of MIH, and these should be made publicly available in the UK. Research to provide clarity over the definition of a poor prognosis FPM and the best treatment options for these teeth should be carried out.
- Information sharing and collaboration between GDPs and specialists / tier 2 providers should be improved. Access to specialist opinion should be available without referral in limited circumstances to reduce the number of referrals. The system of referral should be reformed so that it is less arduous for the GDP, and more fluid and patient-centred for the child.
- The remuneration system should be altered so that the additional time and skill required in completing behaviour management for children is honoured. A contract that rewards preventive dentistry should also be developed. The ability to carry out minimally invasive aesthetic treatment on the NHS in primary care should be explored perhaps using an ‘index of treatment need’ as used for orthodontic treatment.

- The law in the UK should be changed to allow bleaching in children under the age of 18 where the tooth is discoloured due to a developmental defect or as a result of dental trauma. The wording in the reformed legislation should make it explicit in what circumstances bleaching in children is legal.
- Materials should be developed to use on FPM with MIH. Ideally the material should be hydrophilic, easy to handle, quick to set, durable, tooth coloured, and biocompatible. It should reduce pulpal sensitivity and promote pulpal healing, and therefore reduce symptoms of hypersensitivity.
- CPP-ACP should be included in the dental formulary of the BNF, so that it can be prescribed on the NHS.

Further research should involve developing the resources described above and assessing how changes implemented may influence the practice of the GDP, and care outcomes for children with MIH in primary care. By investigating the care of children with MIH within the UK, the current dental system has come under scrutiny as these children are treated by both generalists and specialists. GDPs have all the skills necessary to treat children with MIH, but may lack practical knowledge and confidence regarding diagnosis, treatment planning, and the time to focus sufficiently on behaviour management and prevention within the current NHS system. It is important to remember that in the UK, the majority of children will be treated in primary care by GDPs and will never see a specialist in paediatric dentistry. Therefore, there remains a high need for pragmatic studies based in primary care, so that improvements in dental care can be implemented in a way which will benefit the majority of children, and all children with MIH.

Appendices

1. Mann Whitney U Test for difference in number of teeth extracted in terms of severity of MIH

		Ranks		
		N	Mean Rank	Sum of Ranks
Extraction	Mild MIH	9	15.50	139.50
	Severe MIH	39	26.58	1036.50
	Total	48		

Table 10. Mann Whitney U Test for difference in number of teeth extracted in terms of severity of MIH

Test Statistics ^a	
	Rx_X
Mann-Whitney U	94.500
Wilcoxon W	139.500
Z	-2.466
Asymp. Sig. (2-tailed)	.014
Exact Sig. [2*(1-tailed Sig.)]	.031 ^b

a. Grouping Variable: Severity severe

b. Not corrected for ties.

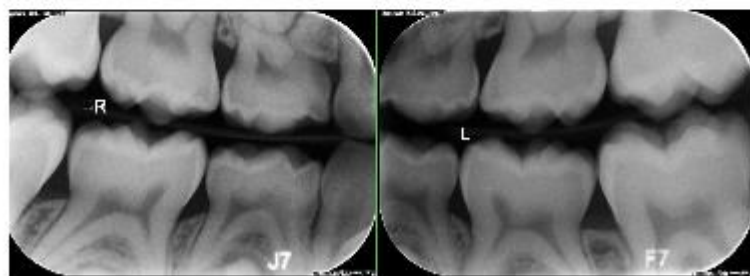
2. UK Postcode areas represented in survey

	Total No.	England	Scotland	Wales	NI
Aberdeen	1		1		
Blackburn	1	1			
Belfast (Northern Ireland)	1				1
Bristol	3	3			
Bradford	1	1			
Bath	1	1			
Birmingham	2	2			
Coventry	5	5			
Chester	1	1			
Dumfries	1		1		
Derby	1	1			
Dundee	1		1		
Edinburgh	3		3		
Exeter	1	1			
Blackpool	1	1			
Guildford	1	1			
Glasgow	4		4		
Huddersfield	1	1			
Kingston Upon Thames	2	2			
Kilmarnock	1		1		
Leicester	1	1			
Leeds	2	2			
Llandudno	1			1	
Liverpool	2	2			
Milton Keynes	1	1			
Motherwell	2		2		
Rochester	1	1			
Manchester	3	3			
Newcastle upon Tyne	5	5			
Nottingham	3	3			
North London	1	1			
Preston	1	1			
Reading	1	1			
Shrewsbury	1	1			
South West London	1	1			
Sunderland	1	1			
Stockport	2	2			
Sheffield	2	2			
Twickenham	1	1			
Cleveland	1	1			
Torquay	3	3			

Truro	1	1			
Telford	1	1			
Wigan	2	2			
Wakefield	1	1			
York	2	2			
TOTALS	76	61	13	1	1
%		80	17	1	1
Real life GDP weighting %		81	10	4	4

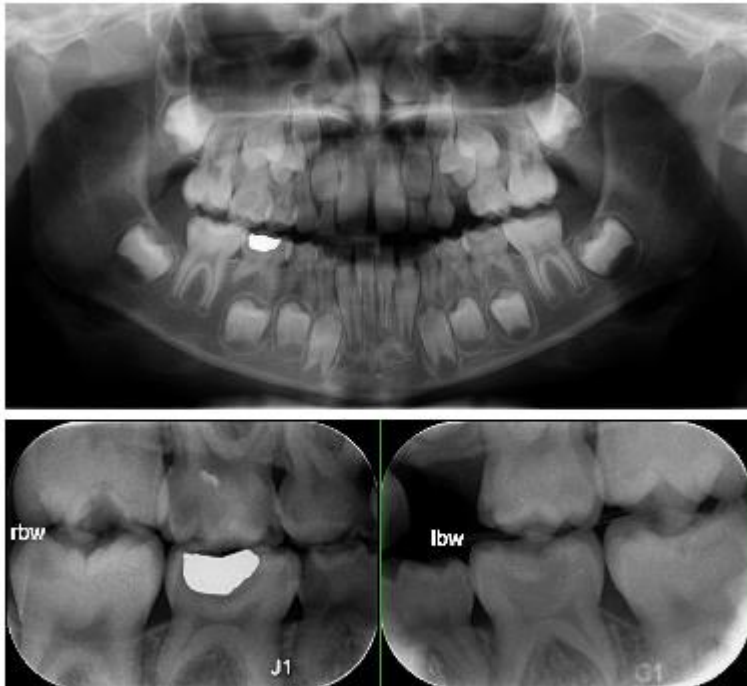
3. Orthopantogram and bitewing radiographs, and clinical photographs for Vignette 1

Q6.1.



4. Orthopantomogram and bitewing radiographs, and clinical photographs for Vignette 2

Q7.1.



5. Mann-Whitney U Test of diagnostic accuracy and confidence

Ranks

		N	Mean Rank	Sum of Ranks
Diagnosis accurate	Not confident at all/slightly confident	16	20.72	331.50
	Confident/very confident	41	32.23	1321.50
	Total	57		

Test Statistics^a

	Diagnosis correct
Mann-Whitney U	195.500
Wilcoxon W	331.500
Z	-2.406
Asymp. Sig. (2-tailed)	.016

6. Mann-Whitney U Test of treatment planning accuracy and confidence

Ranks

		N	Mean Rank	Sum of Ranks
V1 molars	Not confident at all/slightly confident	33	26.02	858.50
	Confident/very confident	24	33.10	794.50
	Total	57		
V1 incisors	Not confident at all/slightly confident	33	32.36	1068.00
	Confident/very confident	24	24.38	585.00
	Total	57		
V2 molars	Not confident at all/slightly confident	33	32.29	1065.50
	Confident/very confident	24	24.48	587.50
	Total	57		
V2 incisors	Not confident at all/slightly confident	33	29.64	978.00
	Confident/very confident	24	28.13	675.00
	Total	57		

Test Statistics^a

	V1 molars	V1 incisors	V2 molars	V2 incisors
Mann-Whitney U	297.500	285.000	287.500	375.000
Wilcoxon W	858.500	585.000	587.500	675.000
Z	-1.802	-2.098	-1.915	-.877
Asymp. Sig. (2-tailed)	.072	.036	.055	.380

7. First draft semi-structured interview schedule

Introduction

- *Introduction of interviewer and explanation of research purpose*
- *Confirmation of interview confidentiality, and anonymity in reporting the data*
- *Confirmation of interviewee's name and that they are happy for the interview to be taped*

Scene setting

1. *Can you remember any details about the first time you were taught or heard about MIH?*
2. *Thinking about children you may have seen in general practice who have MIH – do you have any thoughts about your management of those children?*

Diagnosis

3. *MIH can sometimes look similar to other dental conditions – have you found this to be an issue?*
4. *During a patient exam appointment, are there any signs or symptoms that prompt you to think of MIH as a potential diagnosis?*
5. *Does being certain in your diagnosis affect your management of the child?*

Treatment

6. *Have you encountered challenges when treatment planning or carrying out treatment for children with MIH in the past?*
7. *Do the treatment plans made for children with MIH you have referred seem logical/ make sense?*

Referral

8. *Can you think of any scenarios when you have referred a child with MIH and the reason for doing so?*
9. *In your experience, do challenges at diagnosis, when treatment planning or during treatment most often lead to referral?*
10. *Are there additional reasons which influence this decision?*
11. *When you have referred a child with MIH in the past, what contact do you maintain whilst they are seen in secondary care?*
12. *Once children are discharged from secondary care, how do you manage these children?*
13. *Do you see management of MIH as an issue in general practice?*

Closing remarks

- *The participant will be thanked for their time and asked whether they can be contacted again for clarification of any points within the transcript*

8. Final semi-structured interview schedule

Introduction

- *Introduction of interviewer and explanation of research purpose*

The purpose of this interview is to explore your experiences of molar-incisor-hypomineralisation as a GDP in primary care. I want to understand how MIH is perceived by GDP through exploring your experiences from the time you first learnt of the condition, to your most recent clinical practice. As a recap of the condition, MIH can present on a spectrum, from opaque white and yellow/brown patches on the enamel of first permanent molar and incisor teeth which are asymptomatic, to teeth which lack any normal looking enamel, which have post-eruptive breakdown, are more susceptible to caries and are sensitive or painful when eating and brushing teeth.

- *Confirmation of interview confidentiality, and anonymity in reporting the data*
- *Reminder not to disclose any identifiable patient information*
- *Advised if information is disclosed that calls into question the participants fitness to practice, confidentiality would be broken in order to follow the 'raising concerns' standard of the GDC*
- *Confirmation of interviewee's name and that they are happy for the interview to be taped*

Questions (recorded)

- *Can you tell me a little about your experiences of treating children in general practice?*
- *Can you tell me about your experience of treating children with MIH?*
- *Could you give me some examples of factors that you feel make a child with MIH easy or challenging to manage?*
- *Can you tell me about the first time you were taught or heard about MIH?*
- *Can you tell me about how your experiences of MIH in practice have shaped your understanding of MIH since you first learned about it?*
- *How do you see your role in the management of children with MIH, as a GDP?*
- *As a general practitioner what would improve your ability to manage children with MIH?*

Closing remarks

The participant will be thanked for their time and asked whether they can be contacted again for clarification of any points within the transcript

9. Participant demographics of GDPs interviewed

Participant No.	Sex	Qualified since	Location	Additional Information
1	F	2014	London	Associate. Dental core training – not paediatric dentistry
2	F	2014	London	Associate. Dental core training – not paediatric dentistry
3	M	2015	London	Associate. Dental core training – not paediatric dentistry
4	F	2016	South East	Associate. Dental core training in paediatric dentistry
5	M	2004	London	Practice principal
6	F	2016	London	Associate in affluent area
7	F	2016	London	Associate in multicultural and deprived area
8	F	2003	Yorkshire	Associate in private practice with NHS contract for children
9	M	2005	London	Practice principal & clinical tutor in dental hospital
10	M	2019	North West	Foundation dentist



Article

Management of Molar Incisor Hypomineralisation (MIH): A 1-Year Retrospective Study in a Specialist Secondary Care Centre in the UK

Judith Humphreys * and Sondos Albadri

School of Dentistry, University of Liverpool, Liverpool L3 5PS, UK; sondos.albadri@liverpool.ac.uk

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Received: 29 October 2020; Accepted: 20 November 2020; Published: 24 November 2020



Abstract: (1) Background: Molar incisor hypomineralisation (MIH) is an enamel defect that affects an estimated 14.2% of children worldwide. Care takes place in primary and secondary care facilities. (2) Aim: To investigate how children with MIH are managed within a specialist centre in the north of England. (3) Method: A retrospective service evaluation within the paediatric dentistry department was registered with the clinical governance unit. Children who attended consultant-led new-patient clinics between 1 January and 31 December 2015 with a diagnosis of MIH were included. The data collected concerned the pre-referral treatment, the history and diagnoses and the treatments completed. (4) Results: Out of 397 records reviewed, 48 (12.1%) had MIH, where 81.3% and 18.8% of patients had severe and mild MIH, respectively. The majority of patients ($n = 44$ (91.7%)) were referred appropriately. Treatment was completed at the specialist centre for 44 (91.7%) patients. Twenty-five (52.1%) patients had an extraction of one or more first permanent molar teeth. Sixteen patients had the extractions at between 8 and 10 years old and 2 had the extractions later as part of an orthodontic plan. (5) Conclusion: Most children had severe MIH and were referred at an appropriate time to facilitate the consideration of loss of poor prognosis of first permanent molars (FPMs). Most children required specialist management of their MIH.

Keywords: molar incisor hypomineralisation; paediatric dentistry; poor prognosis first permanent molars

1. Introduction

Molar incisor hypomineralisation (MIH) is a developmental defect of enamel causing cream, yellow or brown opacities on the first permanent molars (FPMs) and sometimes also the incisor teeth [1]. It is a common defect, with the worldwide prevalence estimated to be approximately 14.2% [2], and a local prevalence of 15.9% in the northeast of England [3]. The presentation can vary, with some children presenting with mildly affected asymptomatic teeth and others presenting with severely affected teeth that can be painful or carious, in addition to having aesthetic concerns regarding the incisors [4]. Treatment options are varied and sometimes complicated by the young age of the presenting patients, who may struggle to cooperate with restorative and surgical options.

Treatment planning for MIH is dependent on multiple factors, including severity. Prevention should follow national guidelines for children at high risk of caries for both mild and severe cases [5,6], in addition to using desensitising treatments, such as casein phosphopeptide–amorphous calcium phosphate (CPP–ACP) [7–9] and silver diamine fluoride [10]. In Europe, a shared care approach between generalists and specialists is recommended [11,12], with mild cases being managed by general dental practitioners (GDPs) in primary care and the more complex cases being managed by specialists in paediatric dentistry [13] or in orthodontics, where FPMs are to be removed [14].

11. Accepted abstract for poster study one (EAPD 2020 - No poster as virtual event due to pandemic)

'CAN YOU FIX MY CRUMBLY BROWN TEETH?'
– A SERVICE EVALUATION OF CHILDREN REFERRED
WITH MOLAR-INCISOR-HYPOMINERALISATION (MIH)

J Humphreys; F Jarad; S Albadri

AIM

To evaluate the care pathway for children with MIH referred to a dental hospital in the U.K.

METHOD

Patient records for all children (n=426) who attended their first Paediatric Dentistry consultant clinic in 2015 were reviewed.

Data collected for children with a diagnosis of MIH included:

- *Demographic information*
- *Reason for referral*
- *Previous treatment for MIH prior to referral*
- *Patient symptoms/ concerns*
- *Number of teeth affected*
- *Severity of condition (mild or severe) based on presence of caries/ post-eruptive breakdown/ severe sensitivity / severe aesthetic concerns*
- *Treatment planned and completed*
- *Number of appointments*

RESULTS

- *48 children (11%) had a diagnosis of MIH*
- *Ages ranged from five to 14 years (mean nine)*
- *22 were female (46%)*
- *MIH was the reason for referral in 16.7% of cases (n=8)*
- *62.5% (n=30) of children had had some treatment attempted prior to referral*
- *Most children (70.8% n=34) had a complaint related to MIH - most commonly toothache (22.9% n=11)*
- *Children most frequently had four affected molars (52.1% n=25) and no affected incisors (33.3% n=16)*
- *The majority of children (81.3% n=39) had severe MIH*
- *41.7% (n=20) completed treatment with general anaesthetic, however only 25% (n=12) were planned for this*
- *56.3% (n=27) of patients had FPM extracted, with 66.7% (n=18) being removed at the 'ideal time'*
- *18.8% of patients were planned for aesthetic treatment to their incisors (n=9) but only 12.5% (n=6) completed this*
- *79.2% (n=38) completed treatment, with median of four appointments being required (range one to 16).*

CONCLUSIONS

Most children were not referred regarding MIH explicitly. Most had severe MIH which required treatment under general anaesthetic or specialist management, and therefore referral was appropriate. The presentation and treatment required varied significantly, reflecting the spectrum of disease severity.

Management of molar-incisor hypomineralisation by general dental practitioners – part one: diagnosis

Judith Humphreys,^{*1} Fadi Jarad² and Sondos Albadri³

Key points

General dentists are not familiar with the terminology for hypomineralisation of second primary molars in addition to using the term hypomineralisation and hypoplasia interchangeably.

Dentists should be aware that the presence of caries may make it more difficult to diagnose molar-incisor hypomineralisation (MIH).

Postgraduate and undergraduate education should ensure that dentists are skilled in avoiding these pitfalls when diagnosing MIH.

Abstract

Introduction Molar-incisor hypomineralisation (MIH) affects one in six children in the UK. For the majority of patients who have mild MIH, this should be managed in primary care.

Aims To assess UK-based general dental practitioners' (GDPs) ability to diagnose MIH when presented with multiple clinical vignettes.

Design An electronic vignette survey was designed with use of clinical photographs – six cases had MIH and/or hypomineralised second primary molars (HSPMs) (seven possible diagnoses). Four control cases showing caries, fluorosis, amelogenesis imperfecta and dentinogenesis imperfecta were also included. Participants were UK-based GDPs. The survey was distributed by email and across social media platforms. Data collection occurred between February and May 2019.

Results Seventy-six GDPs completed the survey; 68.4% (n = 52) of participants were female and 83% (n = 63) of participants graduated after the year 2000. The number of accurate diagnoses for each case were as follows – mild MIH (molars/incisors) 65.79%; mild MIH (molars only) 3.95%; HSPM and MIH (HSPM result) 0%; HSPM and MIH (MIH result) 50%; severe MIH (post-eruptive breakdown) 63.16%; severe MIH (caries) 31.58%; HSPM 3.95%.

Conclusion GDPs are able to accurately diagnose MIH best when both incisors and molars are affected and caries is not present.

Introduction

Molar-incisor hypomineralisation (MIH) is a qualitative defect of enamel which presents as demarcated opacities on first permanent molars (FPMs) and incisor teeth.¹ It has a worldwide prevalence of 14.2%,² with a prevalence of 15.9% reported in the North East of England.³ Recent papers in the *British Dental Journal* and by the British Society of Paediatric Dentistry have indicated that MIH should primarily be managed by general dental

practitioners (GDPs), with only severe cases requiring specialist care.^{4,5,6}

Several papers have been published in the UK discussing management options for MIH;^{4,7,8} however, this is complicated by difficulty in diagnosis.⁹ MIH can present with demarcated cream, yellow or brown patches, can be asymptomatic or acutely sensitive, and severely affected children may have teeth with a lack of any normal-looking enamel, which is prone to post-eruptive breakdown (PEB).⁹ Patel *et al.* recently discussed the key difference between hypomineralisation and hypoplasia.³ Hypoplasia is a defect of enamel quantity, which means the shape of the tooth is altered, often presenting with pits or striations. The enamel itself will appear shiny and hard, unlike an MIH tooth with PEB where the enamel is rough and porous. Other conditions such as fluorosis, amelogenesis imperfecta and dentinogenesis imperfecta can

be distinguished from MIH with a good family and fluoride history, and from the appearance across both dentitions.^{10,11}

MIH teeth are more prone to caries due to the lower mineral content and can be acutely sensitive.^{12,13,14} Early diagnosis of MIH by GDPs is key in order to implement prevention strategies for children of increased concern, for symptomatic relief,^{15,16,17} timely restorative intervention or referral to specialist providers when necessary.

Despite its high prevalence, in a recent survey in the UK, only 57% of GDPs felt confident in diagnosing MIH.¹⁸ Other surveys around the world have found similar levels of confidence in diagnosis, ranging from 51.1% in Iraq¹⁹ to 92.9% in Australia.²⁰ The evidence to date from across the world indicates many GDPs do not feel confident in diagnosis and are unsure of the correct management of these patients.^{19 21,22,23} No previous studies

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Refereed Paper.

Accepted 9 July 2020

<https://doi.org/10.1038/s41415-021-2735-3>

Management of molar-incisor hypomineralisation by general dental practitioners – part two: treatment

Judith Humphreys,^{*1} Fadi Jarad² and Sondos Albadri³

Key points

Most dentists felt able to provide the majority of care for the children with molar-incisor hypomineralisation (MIH) in the presented vignettes.

Dentists should be aware that all children with MIH present an increased caries risk.

A shared-care approach for management of children with MIH between primary and secondary care was almost unanimously agreed upon by general dental practitioners

Abstract

Introduction Molar-incisor hypomineralisation (MIH) is a common occurrence in primary and secondary care settings. While severe cases may need specialist care, mild cases should be managed in primary care.

Aims To assess how UK-based general dental practitioners (GDPs) plan treatment for children with MIH using two clinical vignettes

Design An electronic vignette survey was designed using clinical photographs and radiographs. Vignette one presented a child with mild MIH who was unhappy about the appearance of his teeth. Vignette two presented an anxious child with severe MIH, caries and sensitivity. Further questions relating to confidence in management of MIH and referral were included. Participants were UK-based GDPs who regularly treat children. The survey was distributed by email and across social media platforms. Data collection occurred between February and May 2019.

Results Fifty-eight GDPs completed the survey. Around half of participants addressed the aesthetic concerns of the child in vignette one. The majority of participants demonstrated sound treatment planning in terms of preventive care and management of molars. More GDPs identified increased caries risk in vignette two.

Conclusion These findings demonstrate most GDPs in this study were working as effective tier one and two providers when faced with management of children with MIH.

Introduction

Molar-incisor hypomineralisation (MIH) is a common condition affecting 14.2% of children worldwide.¹ In the UK, where 15.9% of children have MIH,² it is more common than dental trauma (12% of 12-year-olds) and around half as common as dental caries (34% of 12-year-olds).³ This means that, for every two children with caries, you could expect to see one child with MIH.

Treatment planning for MIH is dependent on multiple factors, including severity. Prevention should follow national guidelines for children at high risk of caries for both mild and severe cases.^{4,5} Many authors also suggest the use of casein phosphopeptide-amorphous calcium phosphate (CPP-ACP) products to promote mineralisation of the hydroxyapatite.^{6,7,8} This management, which can improve symptoms and benefit long-term prognosis, works best when implemented quickly.

Severe MIH includes affected teeth which have evidence of post-eruptive breakdown (PEB), caries, severe hypersensitivity or pain and children who have strong aesthetic concerns,⁹ which necessitates more invasive treatment. First permanent molars (FPMs) may require restoration with either composite resins or preformed metal crowns (PMCs).¹⁰ Glass-ionomer cements (GICs) may be suitable as fissure sealants and temporary restorations

but are not durable in the long term.¹¹ Another management option is to extract poor prognosis FPMs when the patient is 8–10 years old, in order that the second permanent molars achieve good contact and space closure with the adjacent teeth on eruption.¹²

Children with MIH often have aesthetic concerns when incisors are affected. They may see an improvement in appearance with microabrasion, bleaching, resin infiltration or localised composite restorations/composite veneers.¹³ This has been shown to improve the reported happiness and confidence of children with enamel defects.¹⁴ Some of these treatments are not widely available in NHS primary care.

Even with a clear plan in mind, execution can be challenging. FPMs erupt at six years of age and many children may lack the cooperation required for restorative treatment under local anaesthetic. 'Simple' treatment like

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Refereed Paper.

Accepted 9 July 2020

<https://doi.org/10.1038/s41415-021-2842-1>

14. Abstract for poster study two (International Association of Paediatric Dentistry 2019)

Hypoplastic or Hypomineralised?

- A vignette survey to assess diagnosis of Molar-Incisor-Hypomineralisation

S.J Humphreys; S. Albadri; R. Harris; F. Jarad

Background

Molar-incisor-hypomineralisation (MIH) is a common enamel defect which affects 14% of children world-wide (1). Its management and diagnosis can be challenging (2). Research in several countries has found that general dental practitioners (GDPs) do not always feel confident when diagnosing MIH. Prompt initial management and preventive interventions are important as affected teeth are at risk of caries and post-eruptive breakdown.

Aim

To assess the ability of GDPs in the United Kingdom to diagnose MIH.

Design

Ethical approval was granted from University of Liverpool. An electronic vignette-based survey was designed, piloted and disseminated electronically to UK based GDPs. Six sets of clinical photographs were selected through rigorous processes to show a variety of phenotypes of MIH and hypomineralised second primary molars (HSPM) with or without caries and post-eruptive breakdown. Four control conditions were also selected. Descriptive statistics and a systematic inductive approach were used to analyse the data.

Results

Quantitative

Data was collected over six weeks (February and March 2019). 52 GDPs completed the survey. 29% were male, 69% were female and 2% preferred not to say. There were seven possible diagnoses of MIH or HSPM, in six case vignettes. Two (4%) GDPs identified five cases, 23% (n=12) identified four, 23% (n=12) identified three, 17% (n=9) identified two, 6% (n=3) identified one, and 27% (n=14) did not identify any cases.

Only one dentist gave the correct diagnosis for the case with HSPM alone and none gave the diagnosis of HSPM in the case of MIH and HSPM. MIH cases where patients also had caries were more difficult to diagnose (4% n=2; 19% n=14) compared with cases without caries (65% n=34; 67% n=35).

Qualitative

GDP often confused the term hypoplastic and hypomineralised or used descriptive terms instead of the recognised diagnosis term, such as 'hypomineralisation' instead of MIH or HSPM.

Conclusions

The majority of GDPs in this sample could identify MIH from clinical vignettes but were unable to spot the condition when there was a second diagnosis of caries. GDPs were unable to diagnose HSPM with or without MIH. GDPs frequently confuse hypomineralisation with hypoplasia.

References

- (1) ZHAO, D., DONG, B., YU, D., REN, Q. & SUN, Y. 2018. The prevalence of molar incisor hypomineralization: evidence from 70 studies. *Int J Paediatr Dent*, 28, 170-179.
- (2) WEERHEIJM, K. L. E., M.E.C; KILPATRICK, N 2015. *Molar Incisor Hypomineralization and Hypomineralized Second Primary Molars: Diagnosis, Prevalence and Etiology - Planning and Care for Children and Adolescents with Dental Enamel Defects: Etiology, Research and Contemporary Management*, Springer-Verlag Berlin Heidelberg.

15. Poster study two (International Association of Paediatric Dentistry 2019)

16. Abstract for oral presentation study three (Alliance of Molar Incisor Hypomineralisation Investigation and Treatment 2021)

Molar-Incisor-Hypomineralisation in Primary Care – An Exploratory Study Investigating

How General Dentists Fit in the Puzzle

J. Humphreys; R. Harris; S. Clayton; S. Albadri; F. Jarad

Background

Molar-incisor-hypomineralisation (MIH) is a common enamel defect affecting 14.2% of children worldwide. In the UK, most children have dental care with their general dental practitioner (GDP), and only see specialists by referral. Therefore, the experience of GDPs form an important part of the child's overall dental journey.

Aim

To investigate the experience of GDPs when managing children with MIH in England.

Method

Ethical approval was granted by University of Liverpool. A sample of four male and six female GDP with one to 15 years' experience was achieved through purposively sampling interested parties following advertisement via professional groups. Ten semi-structured telephone interviews were undertaken in May 2020. Recorded interviews were transcribed verbatim and coded. Thematic analysis using an inductive approach was semantic in nature and followed realist paradigms which was appropriate to explore individual motivations, experience and meanings described.

Results

The overarching theme was of managing uncertainty with four subthemes -setting the scene, fighting the tooth, working within the system, and self and interpersonal insight.

Despite being knowledgeable about MIH, participants expressed varying levels of confidence in all aspects of management for children with MIH. Severe MIH presented issues when planning and executing treatment for both molars and incisors. Families who attended irregularly missed opportunities to develop a sound relationship with the GDP, making appointments less predictable. There was a great deal of uncertainty surrounding 'doing the right thing' across the themes, with referral often used to manage these uncertainties. Situations where participants felt a lack of control led to reduced confidence regarding outcomes.

Conclusion

Participants experienced managing children with MIH as a highly variable experience which was dependent on the severity of MIH, the child, the clinician and system of primary care dentistry. These factors dictated the level at which the child became 'complex' to manage for general practitioners.

17. Ethical approval study two



Health and Life Sciences Research Ethics Committee (Psychology, Health and Society)

9 January 2019

Dear Prof Jarad

I am pleased to inform you that your application for research ethics approval has been approved. Application details and conditions of approval can be found below. Appendix A contains a list of documents approved by the Committee.

Application Details

Reference:	4561
Project Title:	Management of Children with Molar-Incisor-Hypomineralisation
Principal Investigator/Supervisor:	Prof Fadi Jarad
Co-Investigator(s):	Miss Sarah Humphreys
Lead Student Investigator:	-
Department:	Restorative Dentistry
Approval Date:	09/01/2019
Approval Expiry Date:	Five years from the approval date listed above

The application was **APPROVED** subject to the following conditions:

Conditions of approval

- All serious adverse events must be reported to the Committee (ethics@liverpool.ac.uk) in accordance with the procedure for reporting adverse events.
- If you wish to extend the duration of the study beyond the research ethics approval expiry date listed above, a new application should be submitted.
- If you wish to make an amendment to the study, please create and submit an amendment form using the research ethics system.
- If the named Principal Investigator or Supervisor leaves the employment of the University during the course of this approval, the approval will lapse. Therefore it will be necessary to create and submit an amendment form within the research ethics system.
- It is the responsibility of the Principal Investigator/Supervisor to inform all the investigators of the terms of the approval.

Kind regards,

Health and Life Sciences Research Ethics Committee (Psychology, Health and Society)

iphsrec@liverpool.ac.uk

0151 795 5420

Research ethics amendment approved - Prof Fadi Jarad: 4561 - Management of Children with Molar-Incisor-Hypomineralisation



donotreply@infonetica.net

Fri 3/8/2019 7:03 PM



To: Jarad, Fadi; S.J.Humphreys@liverpool.ac.uk

Cc: IPHS Ethics; sysadmin@infonetica.net; S.J.Humphreys@liverpool.ac.uk

The answers within the form indicate this is a minor amendment, therefore we are pleased to inform you that the amendment to your study has been approved.

Conditions of the approval

- All serious adverse events must be reported to the Committee within 24 hours of their occurrence, via the Research Integrity and Ethics Officer (ethics@liverpool.ac.uk).
- If the named Principal Investigator or Supervisor leaves the employment of the University during the course of this approval, the approval will lapse. Therefore please contact the Committee (details below) in order to notify them of a change in Principal Investigator or Supervisor.

Kind regards,

Health and Life Sciences Research Ethics Committee (Psychology, Health and Society)

Need help? Download a user guide: [User Guide \(Staff\)](#) | [User Guide \(Student/Supervisor\)](#)

19. Ethical approval study three



Health and Life Sciences Research Ethics Committee (Human participants, tissues and databases)

20 April 2020

Dear Prof Jarad

I am pleased to inform you that your application for research ethics approval has been approved. Application details and conditions of approval can be found below. Appendix A contains a list of documents approved by the Committee.

Application Details

Reference: 5997
Project Title: How do UK based GDPs understand and experience molar incisor hypomineralisation (MIH)?
Principal Investigator/Supervisor: Prof Fadi Jarad
Co-Investigator(s): Miss Sarah Humphreys, Prof Rebecca Harris, Prof Sondas Albadri
Lead Student Investigator: -
Department: School of Dentistry
Approval Date: 20/04/2020
Approval Expiry Date: Five years from the approval date listed above

The application was **APPROVED** subject to the following conditions:

Conditions of approval

Please note: this approval is subject to the restrictions laid out in the [Policy on research involving human participants in response to COVID-19](#). Therefore all face-to-face contact with human participants for the purpose of research should be halted until further notice; unless the study qualifies as one of the exceptions specified in the Policy and has been discussed with Research Ethics and Integrity team.

- All serious adverse events must be reported to the Committee (ethics@liverpool.ac.uk) in accordance with the procedure for reporting adverse events.
- If you wish to extend the duration of the study beyond the research ethics approval expiry date listed above, a new application should be submitted.
- If you wish to make an amendment to the study, please create and submit an amendment form using the research ethics system.
- If the named Principal Investigator or Supervisor changes, or leaves the employment of the University during the course of this approval, the approval will lapse. Therefore it will be necessary to create and submit an amendment form within the research ethics system.
- It is the responsibility of the Principal Investigator/Supervisor to inform all the investigators of the terms of the approval.

Kind regards,

D Prescott

Health and Life Sciences Research Ethics Committee (Human participants, tissues and databases)

edreseth@liverpool.ac.uk

20. Qualtrics screenshot showing study two survey blocks

DDSc GDP Survey

▸ Inclusion/ Exclusion | 4 Questions

▸ Consent | 8 Questions

▸ Demographics | 10 Questions

▸ Diagnosis | 23 Questions

▸ Treatment Planning | 2 Questions

▸ Treatment Planning Case 1 | 17 Questions

▸ Treatment Planning Case 2 | 17 Questions

▸ Confidence | 6 Questions

▸ Prize draw | 1 Question

Add Block

End of Survey

We thank you for your time spent taking this survey.

Your response has been recorded.

21. Consent imbedded into study two survey

Q2.1. Please consult the study participant information attached before continuing.

[Participant information v1](#)

Q2.2.

I confirm that I have read and have understood the information sheet dated October 2018 for the above study, or it has been read to me. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.

Yes

Q2.3.

I understand that taking part in the study involves completing this questionnaire.

Yes

Q2.4. I understand that my participation is voluntary and that I am free to stop taking part and can withdraw from the study at any time without giving any reason and without my rights being affected. In addition, I understand that I am free to decline to answer any particular question or questions.

Yes

Q2.5. I understand that following submission of the questionnaire I will no longer be able to request withdrawal of the information I provide.

Yes

Q2.6. I understand that the information I provide will be held securely and in line with data protection requirements at the University of Liverpool.

Yes

Q2.7.

I understand that original questionnaires will be retained in the Qualtrics cloud, Utah, USA, until 6 months after the end of the research project. Only the lead investigator will have access to this data.

Yes

Q2.8.

I agree to take part in the above study.

Yes

22. Consent for study three

Version 1; November 2019

Participant consent form

Research ethics approval number: 5997

How do UK based GDPs understand and experience management of molar incisor hypomineralisation (MIH)?

Sarah Humphreys

Please initial box

1. I confirm that I have read and have understood the information sheet dated February 2020 for the above study, or it has been read to me. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.
2. I understand that taking part in the study involves an audio recorded interview over telephone.
3. I understand that my participation is voluntary and that I am free to stop taking part and can withdraw from the study at any time without giving any reason and without my rights being affected. In addition, I understand that I am free to decline to answer any particular question or questions.
4. I understand that I can ask for access to the information I provide and I can request the destruction of that information if I wish at any time prior to anonymization. I understand that following anonymization I will no longer be able to request access to or withdrawal of the information I provide.
5. I understand that the information I provide will be held securely and in line with data protection requirements at the University of Liverpool.
6. I understand that signed consent forms and original audio will be retained by the researcher on a password protected computer until publication and reparation of further grants. This is estimated to be around 5 years.
7. I understand and agree that my participation will be audio recorded and I am aware of and consent to your use of these recordings for the following purposes: transcription into text and qualitative analysis.
8. I understand that the confidentiality of the information I provide will be safeguarded and won't be released without my consent unless required by law. I understand that if I disclose information which raises considerations over the safety of myself or the public, the researcher may be legally required to disclose my confidential information to the relevant authorities.
9. I agree to being contacted at a later date and invited to take part in future studies. I understand that I am only agreeing to receive information and I am under no obligation to take part in any future studies. If you decide not to consent to being contacted in the future it will not have any influence on your involvement in this particular research study.
10. I agree to take part in the above study.

Participant name

Date

Signature

SJ Humphreys

04/05/20



Name of person taking consent

Date

Signature

Principal Investigator

Student Investigator

Fadi Jarad

Sarah Humphreys

Liverpool University Dental Hospital
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