# A follow-up study of children referred for management of Molar Incisor Hypomineralisation: an analysis of treatments received, changes in self-reported oral health-related quality of life, dental fear and anxiety, and the impact of the condition and treatment on the child's family.

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The candidate confirms that the work submitted is his own. The candidate confirms that appropriate credit has been given within the thesis where reference has been made to the work of others.

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## Abstract

**Introduction:** There is currently only limited data about the longer-term impact of Molar Incisor Hypomineralisation (MIH) on affected children and their families.

**Aims:** This follow-up study aimed to investigate the impacts that MIH, and its management, have on affected children and their families.

**Methods:** 18 children, originally referred to a specialist centre for the management of MIH, were re-recruited for this follow-up study. Pre-treatment OHRQoL and DFA were captured post-treatment and compared with pre-treatment OHRQoL and DFA scores previously captured using the same tools. Details of original diagnosis, treatment and patient management were captured from clinic records. The Family Impact Scale questionnaire was used to captured family impact.

**Results:** Follow-up OHRQoL and DFA scores showed little average change from pre-treatment baseline. There was a small decrease in average OHRQoL and a small increase in average DFA, but the majority reported decreased DFA and increased OHRQoL (especially Functional well-being). Only two subjects had permanent molar extractions of under LA, but reported the largest increases in DFA. All but one of those receiving treatment under general anaesthesia had MIH classified as severe at initial diagnosis. Family impact was generally low, being greatest on parental/family activities. Financial impact was scored as zero by all, but longer-duration of the journey to the specialist centre was associated with greater impact. Children and their families praised the dental team regarding explanation of procedures and communication, but reported problems with waiting times and appointment booking.

**Conclusions:** Whilst the small numbers of subjects recruited for this follow-up study precludes drawing firm conclusions, some interesting trends worthy of further investigation were identified. With the possible exception of permanent molar extractions under LA, MIH and its treatment seemed to

have little effect on OHRQoL and DFA. Problems highlighted included, waiting times and appointment booking.

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# List of Abbreviations

| | |
|---|---|
| AI | Amelogenesis Imperfecta |
| AT | Atypical restoration |
| CDFP | Child Dental Fear Picture Test |
| CFSS-DS | Children's Fear Survey Schedule – Dental Subscale |
| COHIP | Child Oral Health Impact Profile |
| COHIP-SF19 | Child Oral Health Impact Profile- short form 19 |
| CPP-ACP | Casein Phosphopeptide-Amorphous Calcium Phosphate |
| DA5 | Dental Anxiety Scale for 5-year-old children |
| DAS | (Corah) Dental Anxiety Scale |
| DCLG | Department for Communities and Local Government |
| DDE | Developmental defects of enamel |
| DDQ | Dental Discomfort Questionnaire |
| DHC | Dental Health Component (of IOTN) |
| DMFT | Decayed Missing Filled Teeth (permanent dentition) |
| dmft | decayed missing filled teeth (primary dentition) |
| DO | Demarcated Opacity |
| EAPD | European Academy of Paediatric Dentistry |
| ECC | Early Childhood Caries |
| ECOHIS | Early Childhood Oral Health Impact Scale |
| EPR | Electronic patient record system |
| FIS | Family Impact Scale |
| FPM | First permanent molar(s) |
| GA | General Anaesthesia |
| HPM | Hypomineralised primary molars |
| ICON | Index of Complexity and Orthodontic Treatment Need |
| IOTN | Index of Orthodontic Treatment Need |
| IHS | Inhalation Sedation |
| LA | Local Anaesthesia |
| LDI | Leeds Dental Institute |
| LTHT R&I | Leeds Teaching Hospitals NHS Trust Research & Innovation |
| MDAS | Modified Dental Anxiety Scale |
| MCDAS | Modified Child Dental Anxiety Scale |
| MCDASf | Modified Child Dental Anxiety Scale – faces version |
| mDDE index | Modified Developmental Defects of Enamel index |
| MIH | Molar Incisor Hypomineralisation |
| MIH-SSS | MIH severity scoring system |
| NPEU | National Perinatal Epidemiology Unit, University of Oxford |
| OHRQoL | Oral health-related quality of life |
| OPT | Orthopantomagram radiograph |
| PEB | Post-eruptive Enamel Breakdown |
| POQL | Pediatric Oral Health-Related Quality of Life |
| QoL | Quality of Life |
| REC | Research Ethics Committee |
| SFP | Smiley face programme |
| VAS | Visual analogue scale |
| VPT | Venham picture test |

# Chapter 1: Literature Review

## 1.1 Introduction

Developmental defects in the enamel of first permanent molars were first recognised to be a relatively prevalent phenomenon in the late 1980s and early 1990s (Koch et al., 1987). Various names have been given to this condition, including 'hypoplastic first permanent molars', 'idiopathic enamel hypomineralisation in permanent first molars' and 'cheese molars' (Weerheijm et al.,2001a). In 2001, the term 'Molar Incisor Hypomineralisation' (MIH) was suggested to describe the clinical picture of a hypomineralisation of a systemic origin of one or more of the four permanent first molars, as well as any associated and affected incisors (Weerheijm et al., 2001b). Molar Incisor Hypomineralisation (MIH) has since been widely used to describe this condition.

## 1.2 Prevalence of MIH

MIH has been observed in many populations in all parts of the world. The prevalence of this condition was first described in the Swedish population by Koch in 1987. It was noticed that children born in the late 1960s/early 1970s had idiopathic hypomineralisation related to the first permanent molars and incisors. An epidemiological study was conducted to investigate the prevalence of such phenomena. Some 2252 children born between 1966-1974 were divided into groups according to the year they were born. The examinations of the children took place in 1979, 1980 and 1983. The prevalence ranged from the lowest at 4.4% for children born in 1972 to the highest at 15.4% for those born in 1970 (Koch et al., 1987).

Elsewhere In Europe, in a study conducted amongst 3233 children of 12 years of age in Northern England (Balmer et al., 2012), the prevalence of MIH was 15.9%. Lygidakis et al. examined a sample of 3518 children aged from 5.5 to 12 years old in Greece and reported a prevalence of 10.2% (Lygidakis et al., 2008a). A sample of 550 Spanish children aged between 6 and 14 years showed the occurrence of MIH at 17.8% (Martinez Gomez et

al., 2012). Out of 227 children at 7.3-8.3 years, 13.7% were affected in Italy (Calderara et al., 2005). Four regions in Germany showed a mean MIH prevalence of 10.1% after examining 23957 children of 10 years old (Petrou et al., 2014). Norway reported 28.2% from examining 3013 8-9 year old Children (Haque Afzal et al., 2023). Over 30,000 children aged 4 to 12 years took part in an annual dental screening in Switzerland, and the result showed a 6.6% prevalence of MIH (Abdelaziz et al., 2022).

The Swedish study conducted by Koch and Jalevick (Koch et al., 1987) was the first to demonstrate that the prevalence of MIH in a population might vary significantly in children born in different years. Likewise, among 2408 children who were born in Dresden, Germany, a higher prevalence MIH rate was found in children born between 1989 and 1991 when they were compared to those who were born before and after this period. Nevertheless, no specific explanation could be given for such a higher prevalence (Dietrich et al., 2003).

Outside of Europe, MIH was found to be present in 6.3% of 1792 children between the ages of 6 and 9 years in Northern India (Mittal, 2016). Across Africa and Asia, eleven Middle Eastern countries averaged 15% in a recent meta-analysis involving 32,636 participants from 29 studies (Bukhari et al., 2023). New Zealand had an 18.8% prevalence of MIH amongst 235 children with a mean age of 8.2 years (Mahoney and Morrison, 2011). The lowest reported MIH prevalence was found to be 2.8% in a 2635 sample of children in Hong Kong with a mean age of 12 years (Cho et al., 2008). The highest was reported when 100 cases were diagnosed with MIH in Brazilian children, or 40.2% out of 249 participants aged 7 to 13 years in Rio De Janeiro (Soviero et al., 2009).

Globally, the average MIH prevalence has been determined to be 14.2% according to a meta-analysis consisting of 70 studies that included 89,520 people and 10,823 MIH cases (Dave and Taylor, 2018). Another meta-analysis reviewed 99 studies from 43 countries and 113,144 individuals, resulting in a calculated 13.1% prevalence rate of MIH (Schwendicke et al., 2018). Recently, an additional meta-analysis of 116 studies noted the prevalence at 13.5% (Lopes et al., 2021). Even though there appears to be

some variation in reported and estimated prevalence, MIH is certainly a relatively widespread condition in child populations around the world.

## 1.3 Aetiology of MIH

In 2001, Swedish parents of MIH-affected children answered questionnaires regarding their own health, medications taken during pregnancy, birth complications, and the health of the child during their first three years (Jälevik et al., 2001). After comparing the findings with a control group, the study concluded that MIH is associated with infancy health problems especially those related to respiratory diseases. A similar study was conducted with a group of UK children and their mothers who took part in a study to investigate the cause of MIH by asking the mothers to complete medical history interviews (Whatling and Fearne 2008). Although the cause of MIH remained uncertain, there was a significant relationship between the presence of MIH and mothers who experienced pregnancy problems, children who had chickenpox between 3 and 4 years of age and children for whom the only antibiotic prescribed to them for infections was amoxicillin.

Alaluusua et al. (1996), found that there was an association between breastfeeding for more than eight months and the presence of mineralisation defects. The total duration of breastfeeding also had an association with demarcated hypomineralisation in Finland (Hölttä et al., 2001). MIH in the first molars had a significant relation to breastfeeding when this was continued for more than six months (Fagrell, 2011). It is worth noting that the previous study was not exclusive to MIH, It studied mineralisation defects in general. Conversely, no such association could be established in a later retrospective study where Finnish mothers of MIH-affected children were surveyed about the total duration of breastfeeding (Wuollet et al., 2014).

Through a combination of face-to-face interviews with parents of 267 MIH-affected children in Saudia Arabia who filled out questionnaires, Allazzam et al. (2014) showed that MIH was associated with illnesses such as asthma, adenoid infections, and tonsillitis during the first 4 years of the child's life. Interestingly, in that study, birth complications and breastfeeding duration showed no association with MIH. Recently, in a study conducted where

exposure to aetiological factors were surveyed in mothers of 613 children from Poland, MIH was significantly associated with otitis media in early childhood, atopic dermatitis and premature childbirth (Ilczuk-Rypuła et al., 2022).

In recent years, evidence has accrued supporting a potential genetic component in the aetiology of MIH. In a study of DNA samples from 163 cases, Jeremias et al. (2013) suggested that genetic variations in ENAM and AMELX genes that are responsible for amelogenesis imperfecta may result in a lesser degree of enamel formation disturbance in MIH. The authors also suggested that environmental factors interacting with such genetic factors may also play a role in the aetiology. Similarly, a study conducted on 176 pairs of twins, 94 of whom were monozygotic and 73 dizygotic, came to the conclusion that the greater consistency of MIH diagnosis among the monozygotic twins suggested a genetic influence in addition to shared environmental factors (Teixeira et al., 2018). Medical records of 360 children in Greece with MIH showed more than one medical problem during the perinatal and postnatal periods, such as birth complications, respiratory conditions, and neonatal illnesses, compared to a control group. (Lygidakis et al., 2008b).

In summary, conflicting results from numerous studies indicate that the aetiology of MIH is not yet clearly determined, and the broad consensus is that it is believed to be multifactorial. Jeremias et al. (2013) suggested that genetic variations may interact with environmental factors as several genes that are involved in enamel formation are associated with MIH. A recent meta-analysis found no association between aetiological factors such as prolonged breastfeeding or a mother's habits during pregnancy, including smoking and alcohol consumption (Garot et al., 2022).
MIH is now globally recognised as a potential public health issue. Current thinking is that the defect is not simply of systemic origin but a condition with a complex aetiology that may be the result of genetic and environmental interactions (Bussaneli et al., 2022).

## 1.4 Classification of MIH

The European Academy (EAPD) classified MIH into mild or severe based primarily on the presence or absence of enamel breakdown at presentation, induction of sensitivity on brushing or not and the degree of the aesthetic concern (Lygidakis et al,. 2022). Earlier, Leppaniemi et al. (2001) proposed that MIH be classified into three categories according to their severity, ranging from mild, moderate and severe. Da Costa-Silva et al. clearly described the Leppaniemi et al. classification, stating that mild MIH is where the demarcated opacities are without structural loss. Moderate MIH is characterised by opacities in the enamel associated with PEB limited to the enamel. MIH is considered to be a severe type when the structural loss is affecting the dentin and enamel (Da Costa-Silva et al., 2011)

In 2020, Cabral et al. introduced a new scoring system called MIH-SSS that focuses on the severity of the condition. This Severity Scoring for the FPMs system ranges from score = 0, where there is no enamel opacity, to score = 9, where extraction due to MIH has occurred.
A different approach to classification has been proposed by Mittal (2016). The scheme consisted of 5 variations of the presentation of enamel hypomineralisation based on which teeth are affected. Maxillary teeth and buccal surfaces were found to be more commonly affected than mandibular and lingual surfaces (Mittal, 2016).

## 1.5 Consequences of MIH

### 1.5.1 Post-Eruptive Breakdown

Mahoney and Farah (2015) reviewed the available evidence relating to the physical, mechanical, mineral and histopathological characteristics of enamel affected by MIH. The darker the enamel, the greater the severity of the defect, i.e., yellow-brown enamel is more porous, has more protein, and has lower mineral density than white-chalky enamel, suggesting that the risk of fracture under occlusal load increases in darker enamel opacities (Mahoney and Farah, 2015).
Post-eruptive breakdown (PEB) is more likely to occur in MIH-affected teeth with yellow or brown opacities rather than those with white opacities, as

darker lesions reflect a greater deficit in mineral content and a higher protein. The findings came after conducting two clinical assessments over an 18-month period of 6-12-year-old children. (Da Costa-Silva et al., 2011). The different appearances of the enamel in MIH basically reflect hypomineralised defects of varying severity. The effect on the cuspal areas is usually mild, while the cervical enamel is always sound (Farah et al., 2010). A 3D X-ray microscopic study revealed that the pattern of 20% of the mineral concentration reduction suggests a disturbance in the enamel maturation stage, thus explaining the asymmetry of the defect (Fearne et al., 2004). Furthermore, MIH post-eruptive breakdown and structural loss are different from those caused by dental caries, as pre-existing opacities surrounding the borders of lesions are often associated with this non-carious breakdown (Da Costa-Silva et al., 2011).

### 1.5.2 Hypersensitivity

Hypersensitivity is another symptom frequently associated with MIH. Rodd et al. attributed the increased sensitivity in MIH teeth to the weakness of the enamel seen in some MIH patients, resulting in rapid post-eruptive structure loss. Immunocytochemical findings on 19 MIH-affected first permanent molars showed pulpal changes in innervation, vascularity and immune cell accumulation, indicating an inflammatory response. The innervation showed an increased density, and within the pulp, immune cells were the most abundant, and the vascularity was the same in the hypomineralised teeth (Rodd et al., 2007).

In a study of MIH-affected children, daily activities such as tooth brushing and eating were reported to be associated with discomfort in 31% of subjects (Petrou et al., 2015). Raposo et al. (2019) demonstrated that over half of subjects with Mild and Moderate MIH reported hypersensitivity, mostly of low intensity when compared to non-affected molars (Raposo et al., 2019). A similar association of sensitivity in severe MIH has also been reported, but this was considered by the authors of that study to be unreliable, as there was a high frequency of carious lesions that involved dentine in affected teeth (Raposo et al., 2019).

Younger MIH patients who are less than eight years of age have been reported to suffer from more severe hypersensitivity in their first permanent molars immediately after eruption. However, the hypersensitivity tends to decrease as the patient's age increases (Linner et al., 2021).

### 1.5.3 Aesthetic concerns

Marshman et al. (2009), conducted qualitative semi-structured interviews with 21 young people aged 10-15 years who had varying severities of developmental defects of enamel (DDE) affecting their incisor teeth. All young people recruited were already aware they had DDE, and they were interviewed in their own homes. Open-ended questions were used to permit each young person to describe their experiences in their own words. The results showed considerable variation in how much the dental defects of enamel (DDE) marks 'bothered' the participants. Those whose 'appearance' was a defining characteristic of their 'sense of self' were impacted more than those whose 'sense of self' was more characterised by their 'personality'. Interestingly, in this study, gender, age and severity of DDE did not seem to be associated with severity of impact (Marshman et al., 2009).

Tooth discolouration has been reported as a cause of concern for both mothers and 131 MIH-affected children between 7 and 13 years of age when compared to a control group of 131 children of the same age and gender from the same school (Leal et al., 2017). In that study, children in the MIH group reported avoiding smiling because of the appearance of their teeth. MIH did not affect the aesthetic satisfaction of 8-12-year-old children with mild and severe lesions in 467 Brazilian students who completed surveys and had clinical examinations (Fragelli et al., 2021). In another study conducted by Da Silva et al. in 2019, 56 children and their parents were divided into equal groups of MIH, and a control group of patients without MIH was surveyed. The group of severe MIH showed that staining and discolouration significantly impacted the aesthetic perception scores. Parents even exhibited higher dissatisfaction compared to the children (Da Silva et al., 2019).

A cross-sectional study of 16-year-old children conducted by Sujak et al. (2004) combined oral examination and a questionnaire survey for 1024 subjects with enamel defects. Two-thirds of participants had enamel defects, but only 12.5% reported dissatisfaction with their teeth condition. However, the enamel defects were not specifically MIH-related (Sujak et al., 2004).

### 1.5.4 Dental anxiety and fear

Childhood dental fear and anxiety have been measured by several methods over many years. For example, Paryab and Hosseinbor 2013 investigated the demographic and dental factors affecting dental anxiety among Iranian schoolchildren by selecting a total of 150 children between the ages of 6 and 12 years old. After local anaesthesia administration, mothers were asked to fill out the CDAS, and children filled out MCDAS questionnaires. Results showed that nearly 30% of the children experienced severe dental anxiety, with age and regularity of dental visits being significant predictive factors. Additionally, around 29% exhibited behavioural problems, significantly influenced by past negative dental experiences. The study concluded that severe dental anxiety is fairly common in early school years, and general family factors seem to have less impact on children's behaviour during dental visits.

Conversely, Soares et al. (2017) examined the prevalence and factors associated with high dental anxiety in 416 Brazilian children aged 5-7 years over a two-year period. Interviews with parents and clinical evaluations were conducted. High dental anxiety was found in 16.2% of children at the start and increased to 19.8% at the two-year mark. Notably, 38% of those initially anxious remained so, while 62% improved. Chronic medication use, parental dental anxiety, and poor dental health were identified as significant risk factors for high dental anxiety in children (Soares et al., 2017). Furthermore, Brazilian pre-school children showed an association between the number of siblings and dental anxiety. Children with three or more siblings had higher dental anxiety (Abanto et al., 2017).

## 1.6 Tools and Scales developed to measure dental fear and anxiety.

A number of different tools have been proposed to measure dentally-related anxiety and fear in children, and a number of different principles have been employed to attempt to make this assessment. Some tools involve observation and rating of child behaviour by the dental team providing the dental care or by a trained observer. Examples include The Frankl Scale (Frankl, Shiere, & Fogels, 1962) and The Houpt Scale (Houpt et al., 1985). This type of approach has the advantage that it does not rely on comprehension of verbal or written questions and so is most often used to assess dental anxiety in young children whose understanding of such questions might be limited. Disadvantages include the need to involve trained observers and the logistics of either live or recorded ratings of episodes of dental care.

For older children (and adults), responses to questions and/or self-reported information captured from the child patients themselves are commonly used, and a variety of methods have been used to achieve this.
One of the earliest of these was the Fear Survey Schedule-Dental Subscale (FSS-DS). This question-and-answer-based approach was developed in Sweden in 1968 for children between the ages of 4 and 14 years (Scherer et al., 1968) and was based on the Fear Survey Schedule originally proposed by Wolpe and Lang in 1964, which itself had been developed as a method of determining stimuli associated with neurotic anxiety. The FSS-DS comprised 80 items, each of which was scored by the participant on a Likert scale. Cuthbert and Melamed (1982) later developed this into the Dental Fear Survey Schedule for Children (CFSS-DS), reducing the original 80 items to 15, again answered on a 5-point scale ranging from “not afraid at all” to “very much afraid”. Scores thus range from 15-75, with scores of 38 or above usually considered to be indicative of significant clinical dental fear (Milgrom et al., 1995). Visual Analogue Scales (VAS) have also been used to capture patient responses to dental questions about dental anxiety (Luyk et al., 1988) but are more suited to older children and adults.

Beena (Beena J.P. 2013) employed the CFSS-DS to assess the level of fear associated with dental procedures among school children aged 6-12 years to correlate the prevalence of dental caries with their dental fear scores. The study involved 444 school children, consisting of 224 girls and 220 boys. The children were asked to complete the Children's Fear Survey Schedule - Dental Subscale (CFSS-DS) questionnaire. Those with a score of ≥ 38 were categorised in the "dental fear" group, while those scoring <38 were placed in the "without dental fear" group. The results revealed that the highest fear scores were associated with "Injections", "choking", and "dentist drilling".

The study found that there was no statistically significant correlation between the level of dental fear and the prevalence of dental caries in children.
In order to make a question-based approach more suitable for even younger children, some researchers adopted picture-based scales as either an adjunct or replacement for a Likert or Visual analogue scale.
The Venham Picture Test (VPT) was created in the UK in 1979 and is suitable for 3-18 years of age (Venham, 1979) and uses stylised cartoon figures of children representing various emotional states.
The Child Dental Fear Picture Test (CDFP), developed by Klingberg and Hwang in 1994, used ‘dental setting’ pictures of animals in stress-evoking dental-care scenarios, ‘pointing pictures’ of children in five dental situations, and a sentence completion task to test for dental anxiety 5-12-year-old Swedish children.

The use of picture analogue scales using simple facial images representing different emotions has been used widely in paediatric care for many years (e.g. the Varni-Thompson Paediatric Pain Questionnaire - Varni et al. 1987). In 2002, the development of five-point Facial Image Scales to capture dental anxiety in young children was described both by Buchanan and Niven (Buchanan and Niven, 2002) and separately by Humphris et al. Buchanan and Niven’s Facial Image Scale (FIS) was deemed to be suitable for 3 to 18 years of age (Buchanan and Niven, 2002), and the Dental Anxiety Scale for 5-year-old children (DA5) was aimed specifically at 5-year-olds (Humphris et al., 2002) both were developed in the UK in 2002. Buchanan later developed

a computerised facial image scale for 4-11-year-old children based on FIS called The Smiley Face Program (SFP) (Buchanan, 2005).
The Corah Dental Anxiety Scale (DAS), created in 1969 by Dr. Norman L. Corah (Corah, 1969), is a four-question survey designed to gauge a patient's dental anxiety by capturing the respondent's rating of anxiety to dental scenarios (the prospect of making a dental visit the next day; sitting in the dentist's waiting room; having a scale and polish; receiving a filling) using a 5-point scale. Depending on the total score, it categorises anxiety levels as moderate (9-12), high (13-14), or severe (15-20), with the latter possibly indicating a phobia.

Later, in 1995, the Modified Dental Anxiety Scale (MDAS) was introduced by Professor Gerry Humphris at the University of St. Andrews, Scotland (Humphris et al., 1995). This enhanced the Corah DAS scale by using simplified, concise language and adding a secondary assessment to identify specific dental procedures potentially causing anxiety, such as ‘injection in the gum’, ‘being put to sleep for treatment’ and ‘having a tooth taken out’. While both scales aim to identify and address dental anxieties, the MDAS is quicker for patients due to its streamlined questions. In 1998, Wong, Humphris and Lee further developed MDAS, producing the Modified Child Dental Anxiety Score (MCDAS), an eight-question tool designed and validated to be suitable for children 8-15 years of age (Wong et al., 1998)
In order to make the MCDAS more suitable for younger children, Karen E. Howard and Ruth Freeman (2007) introduced a new version that incorporated a faces rating scale, based on the DA5 (Humphris et al., 2002) and the SFP (Buchanan 2005), alongside the original numeric form. In their study, Howard and Freeman aimed to describe the psychometric properties of this face version, termed MCDAS(f), and to provide normative data for dental anxiety in children using this scale. To assess the scale's reliability and validity, various tests were conducted:

- Test-retest reliability was determined using 287 schoolchildren aged 8-10 years who completed the MCDAS(f) twice, 17 weeks apart.
- Criterion validity was assessed with 207 schoolchildren aged 10-12 years who completed both the MCDAS(f) and the CFSS-DS.

- Construct validity was evaluated using a cohort of 206 consecutive child dental patients and their parents.

The findings indicated that the MCDAS(f) exhibited good test-retest reliability and internal consistency. It also showed a significant correlation with the CFSS-DS. The study concluded that the MCDAS(f) is a reliable and valid tool for measuring dental anxiety in children aged 8-12 years (Howard and Freeman, 2007). Nonetheless, MCDAS(f) is limited due to not being able to assess unhelpful thoughts and behaviour or physical reactions, which may be associated with children's dental anxiety and fear. Additionally, the sedation and the anaesthesia questions can be unfamiliar to children who have not experienced those procedures (Porritt et al., 2012).

### 1.6.1 Dental fear and anxiety in children with MIH

Jälevik and Klingberg conducted a 9-year follow-up of 32 patients with severe MIH. Those who had reported increased management problems and anxiety at the baseline still had management problems after nine years. However, their anxiety and fear had decreased (Jälevik and Klingberg, 2012). In a similar study of 498 children in Greece, those with mild or severe forms of MIH showed no significant relationships with dental fear (Kosma et al., 2012).

Özükoç et al. (2019) used the Child Fear Survey-Dental Subscale (CFSS-DS) to measure dental fear in 58 children with mild, moderate and severe MIH. A statistically significant difference in the severe MIH group showed they were more afraid of dentists (Özükoç et al., 2019).

A systematic review of 14 studies suggested children and adolescents diagnosed with MIH did not suffer significantly increased dental fear and anxiety; however, their OHRQoL was impaired (Jälevik et al., 2022).

In the original study to this follow-up study, Al-Bahar, (2017), the MIH group of children had higher mean dental anxiety scores than the caries or amelogenesis imperfecta groups. However, the differences were not statistically significant (Al-Bahar, 2017).

## 1.7 Tools developed to investigate the quality of Life in relation to oral health

The impact of various oral and dental conditions on oral health-related quality of life has been documented in a number of studies using a variety of measurement tools, which have been reviewed by Culler et al. (2021). These tools include Michigan-OHRQoL, which is a tool developed to assess the oral health-related quality of life in children. Developed at the University of Michigan, it has the advantage of allowing children as young as three years old to self-report (Filstrup et al.,.2003).

The Dental Discomfort Questionnaire (DDQ) by Versloot is aimed at identifying dental discomfort or pain in children aged 2-5 years, which can be a significant factor affecting their oral health-related quality of life. However, the questionnaire is only pain-specific and does not explore the OHRQoL issues (Versloot et al., 2006).

In 2007, Pahel et al. collaborated to develop the Early Childhood Oral Health Impact Scale (ECOHIS), a tool that measures the impact of oral health problems on the well-being of preschool children at 3-5 years of age. One of its many strengths is that it also assesses the impact of oral health on the children's families as well. (Pahel et al., 2007).

Introduced in 2011 at Boston University, the Pediatric Oral Health-Related Quality of Life (POQL) is a tool designed to measure the oral health-related quality of life in preschool paediatric populations, capturing various dimensions of their experiences. It can examine the changes before and after the treatment of ECC. It is limited in that it has only been validated to be used in caries (Huntington et al., 2011).

The Scale of Oral Health Outcomes for 5-year-old children, as the name suggests, is specifically designed for 5-year-old children to assess the outcomes of their oral health and its impact on their quality of life (Tsakos et al., 2012)

The Child Oral Health Impact Profile (COHIP) was initially developed by Broder et al. (2007) using a multi-staged scheme based on the process

originally used ed by Guyatt et al. (1996) when designing a system to measure QoL in children with asthma. Guyatt's scheme used several phases for the development of their questionnaire. It includes an initial pool of items, face validation of those items, impact evaluation of the initial item pool, development of positive items, impact evaluation of the revised questionnaire and finally, factor analysis and final revision of the questionnaire. COHIP was designed to measure self-reported oral health-related quality of life (OHRQoL) of children aged between 8 and 15 years. The questionnaire consisted of five domains: oral health, functional well-being, social-emotional well-being, school environment, and self-image. The resulting final COHIP questionnaire was made up of 34 questions.
Broder and Wilson-Genderson first tested the COHIP tool later in 2007 in a study designed to assess its reliability and convergent and discriminant validity (Broder and Wilson-Genderson 2007). The study involved children from paediatric, orthodontic, and craniofacial clinical settings in the USA and Canada, as well as a comparison group of children not seeking dental treatment from two US elementary schools. The results showed that the COHIP had excellent scale reliability and test-retest reliability. Discriminant validity was supported by significant differences among the clinical groups, with the craniofacial group reporting the lowest OHRQoL scores. Within specific clinical groups, associations were found between COHIP scores and clinical indices. Convergent validity was established through significant associations between COHIP scores and Global Health Ratings. The study concluded that the COHIP showed excellent reliability and both discriminant and convergent validity, making it a valuable tool for assessing OHRQoL in children).

Later, the same group of researchers developed a shorter version of the COHIP questionnaire derived from the validated 34-item COHIP with a reduction to 19 items, which became COHIP-SF19 (Broder et al., 2012). They assessed the reliability and validity of the COHIP-SF19 involving participants from paediatric, orthodontic, and craniofacial anomalies (CFAs) groups. For COHIP-SF19, the 19 items were categorised into three domains: Oral Health (five items), Functional Well-Being (four items), and a combined subscale named Socio-Emotional Well-Being (10

items). The study found that the COHIP-SF19 had an internal reliability of ≥ 0.82 across the three samples. The tool was able to discriminate within and across treatment groups based on the extent of the defect (EOD) and within a community-based paediatric sample. The results suggest that the COHIP-SF 19 is a psychometrically sound instrument to measure oral health-related quality of life across school-aged paediatric populations. The COHIP-SF19 is a shorter, more efficient version of COHIP, consists of 19 items and retains excellent psychometric properties appropriate for clinical research and epidemiological studies (Broder et al., 2012).

It was for these reasons (especially its high validity and reproducibility) that Albahar selected COHIP SF19 to assess OHRQoL in the original baseline study, from which this current study is a follow-up (Al Bahar, 2017).

### 1.7.1 Impact of MIH on Oral Health-Related Quality of Life

A number of studies have investigated the impact that MIH may have on children's OHRQoL.

The Child Perceptions Questionnaire (CPQ8-10) was employed in a study conducted in Mexico to assess the impact of MIH on schoolchildren. The study involved 116 children with moderate and severe MIH-affected teeth who were aged 8 to 10 years; 63.2% reported a negative impact on their quality of life (Gutiérrez et al., 2019).

The same tool (CPQ8-10) was used in children of the same age in another study in Germany to investigate the impact of MIH on OHRQoL among 94 children (Joshi et al., 2022). The results showed an increased impairment of OHRQoL with the increase in MIH severity.

A study from Brazil with 594 participants aged 11-14 years of age investigated the impact of MIH on both children and their parents. The researchers utilised the Parental-Caregivers' Perceptions Questionnaire (P-CPQ) and the Child Perception Questionnaire (CPQ11–14 ISF:16). The authors concluded that severe MIH has a greater negative impact on functional and oral symptoms than those without MIH (Dantas-Neta et al., 2016).

Bekes et al. (2021) assessed changes in OHRQoL before and after treatment for hypersensitivity in MIH children aged 6 to 10 years in Germany and Austria. After sealing the MIH-affected molars with glass ionomer cement or composite, a significant improvement in OHRQoL showed throughout the 12 weeks of follow-up. The assessment was made using the CPQ8-10 Questionnaire before treatment and after 1,4,8 and 12 weeks (Bekes et al., 2021).

Dias et al. (2012) employed two versions of the Child Perceptions Questionnaire designed for children of different age ranges (CPQ8-10 and CPQ11–14, respectively) and the Parental–Caregiver's Perceptions Questionnaire (P-CPQ) for the parents in a cross-sectional study conducted on a group of 253 children between 6 and 12 years of age in Germany. The study demonstrated that MIH had a significant negative impact on the OHRQoL of children with MIH, especially in both the 'functional limitation' and 'emotional well-being' domains. Their parents were also significantly impacted in the emotional well-being domain (Dias et al., 2021).

## 1.8 Impact of children's medical and/or dental conditions on their families

### 1.8.1 General medical conditions

It is well recognised that when a child has a health-related condition, the condition itself and/or the treatment of that condition may have a significant impact on both the child and their family/carers. There are a number of different ways in which children and their families might be impacted, and many of these impacts may be potentially negative.

At West Virginia University, data from a sample of 18,136 special needs children from the ages of 3 to 17 years diagnosed with autism spectrum syndrome (ASD), mental health conditions (MHC) and developmental disabilities (DD) was investigated in a cross-sectional study. Compared to children with DD or MHC, caregivers of children with ASD reported issues with insurance coverage, using services, having an adverse family impact, and lack of shared decision-making and source of care (Vohra et al., 2014). In the same year, Towe-Goodman et al. conducted a study that aimed to

understand the impact of anxiety disorders in preschool-aged children on their families was conducted in North Carolina, USA. It involved 110 four-year-old children diagnosed with anxiety disorders, with 63 healthy children acting as a control group. Structured diagnostic interviews and maternal questionnaires were employed to gauge child anxiety symptoms and their impact on family functions. It concluded that anxiety disorders in preschool-aged children can have a significant impact on family activities and the emotional well-being of parents (Towe-Goodman et al., 2014).

Yantzi et al. (2001) examined the effect of the distance to the hospital and its impact on 113 families with children with chronic medical conditions in Ontario, Canada, using data from Burke et al. (1994-1996), which looked at repeatedly hospitalised children. It showed that distance to the hospital is a significant factor affecting the well-being of families. Those living further away from the hospital (more than 80 km) experienced more stress, financial burden, and disruption to family life. The study discovered that families living closer to the hospital had better access to services and support, which helped alleviate some of the challenges faced by those living further away (Yantzi et al., 2001). Cleaton et al. (2020) found that families of UK children aged 6-18 years with developmental coordination disorder (DCD) encountered significant direct and indirect costs along with difficulties accessing services. Also, in the UK, cochlear implantation, especially for children at the age of three years, was shown to cause an increased financial burden on the children's families (Barton et al., 2006), even though the cost was at most equal to 3% of the overall cost of the implantation, the latter being covered by the health sector.

A meta-analysis of 15 studies of children with chronic medical conditions, such as asthma and diabetes, revealed that parents experience stress, time constraints, and challenges in balancing work and family, which leads to an impact on their quality of life (Kish et al., 2018). In a large-scale retrospective study, Dullet et al. (2017) came to the conclusion that modern emerging solutions in the form of telemedicine, where it can be utilised, may have a positive effect on the reduction of cost, time and burden on families, in addition to the benefit of long-term environmental sustainability.

### 1.8.2 Impact of oral and dental conditions and treatments

In 2004, a UK study came to the conclusion that indirect costs of traumatic injuries to incisors in children and adolescents constituted 39% of the total cost (Wong and Kolokotsa 2004). They suggested in the UK setting that there was a need for more specialists in paediatric dentistry to enhance more localised access to care and to reduce the indirect costs associated with travelling for care when specialists were required.

In an analysis of data from 3859 children from England, Wales, and Northern Ireland who had been included in the 2013 UK Children's Dental Health Survey (CDHS), Abed et al. (2019) used the Family Impact Scale (FIS) to measure the effects on families. Only seven items of the full 14-item FIS questionnaire were used in this survey due to space and time. Children with severe dental caries (identified as having at least one tooth with a condition such as pulpal involvement, ulceration, fistula, or abscess – 'PUFA') were compared to those without. Results showed that severe dental caries in children significantly affected family life. Specifically, parents of affected children were more likely to take time off work, feel their child needed more attention, experience guilt, feel stressed, have their regular activities disrupted and have their sleep disrupted. However, severe dental caries was not shown to be linked to financial difficulties in the family. These effects were consistent regardless of the child's age, gender, UK country of residence, or the family's sociodemographic characteristics (Abed et al., 2019).

After reviewing 25 papers, Das et al. (2022) noted that traumatic dental injuries significantly impacted the oral health-related quality of life of children and their caregivers, while treatment of such traumatic dental injuries improves aesthetics in addition to enhancing the OHRQoL as children enjoy food, smiled, showed their teeth without impressment and socialised.

### 1.8.3 Impact of MIH on families of affected children.

Elhennawy et al. (2017), evaluated the cost-effectiveness of treatment options for molars with severe molar-incisor hypomineralisation (MIH) within the German healthcare system. Three treatment approaches were

compared: tooth extraction followed by orthodontic alignment, restoration using resin composite, and restoration with an indirect metal crown after using a preformed metal crown. Results showed that extracting the affected molar at the optimal age and following it with orthodontic alignment if needed was the most cost-effective management in the German environment. However, patient-specific factors such as compliance and aesthetic concerns should be taken into consideration during treatment planning. (Elhennawy et al., 2017).

### 1.8.4 Tools for measuring the impact of a child's medical/dental condition on their families.

Some tools have been developed to measure and assess the burden on families and caregivers of children with chronic diseases. The Impact on Family Scale, developed in 1980 by Stein and Riessman, is a 27-item set of questions that can be used in an interview form or a questionnaire form. It consists of four domains: economic, social, familial and personal strain. It has a four-point scale ranging from strongly agree to strongly disagree (Stein and Riessman, 1980). In 2003, Stein revised the scale and found it to be short, inexpensive and valid. It requires minimal training and can be administrated by phone (Stein and Jessop, 2003). The PedQL Family Impact Module was developed by James W Varni in 2004. It is a comprehensive questionnaire that contains Physical Functioning (6 items), Emotional Functioning (5 items), Social Functioning (4 items), Cognitive Functioning (5 items), Communication (3 items), Worry (5 items), and two scales measuring parent-reported family functioning: Daily Activities (3 items) and Family Relationships (5 items). It was found to be a reliable and valid method for measurement (Varni et al., 2004).

### 1.8.5 Impact on the family – the Family Impact Scale (FIS)

The Family Impact Scale (FIS) was crafted by Locker in 2002 in order to assess the impact on families, both socially and economically, using an initial set of 21 items created from existing health questionnaires, interviews with 41 parents of children with dental and oro-facial conditions, and expert discussions. From these, 14 key items were chosen based on frequency and importance from a study involving 93 parents (Locker et al., 2002). The FIS

consists of 14 items that are divided into four main categories: parental/family activity, parental emotion, family conflict and family finance. The total scores range from 0 to 56, with each question scoring: never = 0, once or twice = 1, sometimes = 2, often = 3, everyday = 4. The FIS scores are computed by summing all of the item scores.
In a study using a translation of Locker's FIS tool, Barbosa and Gaviao (2009) investigated how MIH in Brazilian children had impacted their families. Firstly, they found that the Brazilian Portuguese version of the FIS was both valid and reliable. Their study also further highlighted that children's oral conditions can have a negative impact on the family. The findings of this study were based on a cross-sectional study and convenience samples (Barbosa and Gavião, 2009).

In a study to assess the impact of malocclusion on families of adolescent school children aged 12-15 years in India, Vinayagamoorthy et al. (2020) used and aimed to validate the Kannada (a language spoken in Southwestern India) translation of the Family Impact Scale (FIS) questionnaire. The sample comprised 768 schoolchildren who were chosen randomly from private and public schools. The FIS questionnaire and informed consent forms were distributed via the schools to parents through their children. Data was collected on socio-demographic details, past dental visits, and school type. Clinical examinations for malocclusion were conducted using the Dental Aesthetic Index by a trained and calibrated examiner. The study found a malocclusion prevalence of 59.9%. The FIS demonstrated high internal consistency. Parents of children with malocclusion had significantly higher FIS scores. After adjusting for variables like age, gender, and past dental visits, parents of children with malocclusion had 1.86 times higher FIS scores than those without children with malocclusion. The study concluded that the Kannada version of the FIS is a reliable tool for assessing the impact of a child's oral condition on their family. Furthermore, malocclusion in children significantly negatively influenced family impact scores (Vinayagamoorthy et al., 2020).

## 1.9 Management of MIH

### 1.9.1 Minimally Invasive Approaches

#### 1.9.1.1 Microabrasion

Enamel microabrasion has been advocated as a technique for improving the appearance of MIH-related enamel opacities on upper incisors. A variety of techniques have been advocated. A simple technique of removing a maximum of 0.1mm of enamel thickness by applying 18% hydrochloric acid or 35% phosphoric acid with pumice on the discoloured area of the MIH-affected incisors was described by Wray & Welbury (2001). Other authors have commented that when the opaque enamel defects extend through the full thickness of the enamel down to the amelodentinal junction ADJ, this technique might not produce much improvement when used on its own in teeth with yellowish/brown discolouration (Fayle, 2003).

Pliska et al. (2012) examined the effects of using casein phosphopeptide amorphous calcium phosphate (CPP-ACP); it was found that it had no significant effect when used alone. Microabrasion, on the other hand, was demonstrated to improve the fluorescence of the white spot lesions when used with or without CPP-ACP. In a systematic review of 11 qualified studies, the method was found to be reliable and effective for the management of enamel discolouration of permanent teeth, especially in fluorosis (Blanchet., 2023).

#### 1.9.1.2 Resin Infiltration

Resin infiltration treatment has been proposed recently as an alternative micro-invasive option for enamel defects in an attempt to inhibit caries progression, enamel breakdown and improve aesthetics. Currently, only one enamel infiltration resin material, (Icon® system, DMG, Hamburg, Germany) has been evaluated in incisor enamel opacities associated with MIH. An evaluation of resin infiltration treatment of 116 incisors in 37 patients has recently been published, documenting effective and stable masking of non-cavitated MIH enamel lesions over a 6-month follow-up period (Altan and Yilmaz, 2023).

A recent systematic review of enamel resin infiltration in MIH-affected teeth reported that whilst there was some encouraging evidence that the ICON system can be used to improve the appearance of MIH-affected teeth, all cited authors commented on variability in their results and possible failure. The authors concluded that the lack of standardised protocols for resin infiltration treatment in hypomineralised teeth meant that clinicians currently rarely use resin infiltration and that there is still a need for further research into the use of the ICON system in MIH.

### 1.9.2 Restorative intervention for affected permanent anterior teeth

Due to the high prevalence of MIH, it is logical that diagnosing and managing such cases should be done in primary care whenever possible (Almuallem and Busuttil-Naudi, 2018). Restoration of MIH-affected teeth should be done while considering the long-term prognosis, along with managing sensitivity or pain if present. MIH-affected anterior teeth should be managed conservatively to preserve tooth structure whilst improving aesthetics. This can be achieved with direct composite veneers with or without minimal enamel reduction. If enamel reduction leads to an increase in discolouration of the defect appearance, using a dark grey opaquer can help to mask it (Fayle, 2003). In a series of cases describing five female patients who complained of MIH-affected anterior teeth, treatment involved microabrasion, resin infiltration, bleaching and/or composite restoration. Resin infiltration showed the best results. However, the selection of the appropriate approach depends on several factors, including the patient's expectations, dental age, presence of sensitivity, the severity of the condition and ongoing orthodontic treatment needs (Natera-Guarap et al., 2023).

### 1.9.3 Management of permanent molars.

Restorative intervention for affected first permanent molars presents a number of different challenges for both the child and the dentist. The post-eruptive enamel breakdown (PEB) commonly associated with MIH does not usually follow classical caries-related patterns, which results in many restorations following 'atypical' outlines. PEB is more significantly associated with yellow-brown opacities as they have been shown to be five times more

likely to experience atypical restorations, caries and extraction after one year of applying glass ionomer sealant (Schraverus et al., 2021). These atypical outlines can present challenges with historic non-adhesive restorative materials, such as amalgam, and hence, full coverage or adhesive restorative solutions have become the most popular approaches.

Following a study conducted by Lygidakis et al. (2003), the authors concluded that posterior permanent teeth with enamel hypomineralisation exhibit good long-term prognosis when restored with resin composite restorations. They restored 52 teeth in 46 children aged 8-10 years. All teeth were sensitive-free after the 48 months of the study period (Lygidakis et al., 2003). In a later study, 48 MIH-affected first permanent molars were restored with GIC and then evaluated at the baseline, at six and at 12 months. It was found that 78% of GIC restorations maintained their protective function (Fragelli et al., 2015). Linner, in 2020, conducted a randomised clinical trial and concluded that traditional composite and CAD/CAM ceramic fillings showed moderate to high durability when used on teeth affected by MIH following preparation of the teeth and any cavitation. Conversely, in that study, simpler treatments, like non-invasive composite and particularly glass ionomer cement (GIC), were used for less cooperative children. Although these simpler treatments had lower longevity, they served as a temporary measure to protect the compromised hard tissue of the teeth. These treatments also helped to increase the child's willingness to cooperate until they become mature enough for more permanent and invasive treatment options (Linner., 2020).

#### 1.9.3.1 Extraction of FPM of Poor Prognosis

In the UK, the attitude towards the treatment of the FPM with poor prognosis (cFPM) varies as there appears to be a slight preference towards extraction among specialists in paediatric dentistry, whereas general dental practitioners tend to restore FPM. This conclusion was derived from 51 paediatric dental specialists and 138 GDP who completed online questionnaires which related to three clinical vignettes of compromised FPM (Taylor et al., 2019).

Teo et al. (2016) attempted to identify better radiographic predictors for spontaneous space closure of lower second permanent molars (SPMs) following the extraction of first permanent molars (FPMs) of poor prognosis. Traditionally, FPMs with poor prognosis are planned for extraction at an 'ideal time' to allow SPMs to erupt favourably in their place. However, for lower FPM extractions, timing alone has not been an accurate predictor of successful space closure. The research re-analysed data from a previous study involving 127 lower SPMs from 66 patients, incorporating additional radiographic factors. These factors included the calcification stage of the SPM's bifurcation, the position of the second premolar, the mesial angulation of the SPM in relation to the FPM, and the presence of the third permanent molar. The results showed that only 58% of FPMs extracted at the 'ideal time' had complete space closure. The best outcomes were observed when combining several factors: SPMs not at a specific development stage, the presence of mesial angulation of the SPM, and the presence of the third permanent molar. In such cases, 85% achieved complete space closure. The study concluded that, in addition to the extraction timing of the FPM, other factors like the presence of the third permanent molar and the angulation of the SPM should be considered to ensure reliable, spontaneous space closure of the lower SPM. In the UK, some orthodontic treatment is funded, but this may not cover managing space closure when there are no other orthodontic needs.

Before the decision is made to extract the first molars, (Almuallem, and Busuttil-Naudi, (2018) recommend a full dental assessment of the presence, position and normal formation of the developing permanent dentition. They argue that with orthodontic support, extraction can be a valid treatment as it limits repeated restoration sessions, which may cause less child anxiety. However, orthodontic support is not always available. A recent study concluded that extraction might prove more cost-effective than restorative approaches over the longer term, especially in some cases of severe MIH (Elhussein and Jamal 2020).

Generally, it has been recommended that to give the second permanent molar a better opportunity to drift into the FPM position, extraction should be

considered at a dental age of 8-10.5 years has been shown to give better spontaneous outcomes compared to extractions done at other ages (Eichenberger et al., 2015). The latest guidelines for FPM management in the UK (Noar.J. et al., 2023) state: “For effective management of compromised FPMs with uncertain outcomes, a comprehensive set of variables needs to be taken into account, extending beyond just occlusion-related issues for the individual. These key considerations include:

- Immediate clinical signs associated with the problematic FPM tooth or teeth;
- A precise evaluation of the affected tooth or teeth's prognosis, along with an assessment of potential future symptoms.
- The patient's ability to undergo complex dental procedures, such as surgery, restorative work, and orthodontic treatments.
- Accessibility to dental services.
- The current state of the patient's occlusion or malocclusion and
- The immediate dental care priorities and long-term care responsibilities as perceived by both the child and their parent.

## 1.10 Further assessment of the impact of MIH on children and their families

### 1.10.1 Content analysis

Content analysis is a research method that is used across several academic disciplines. It involves the systematic examination and interpretation of textual, visual, or audio content to extract meaningful insights, identify patterns, and uncover underlying themes. It can be defined as “ A research technique for the objective, systematic and quantitative description of the manifest content of communication” (Berelson, 1952). Another definition that emerged later states, “Any technique for making inferences by systematically and objectively identifying special characteristics of messages” (Holsti, 1968). content analysis enables researchers to explore both quantitative and qualitative aspects of the information, making it an indispensable tool in social sciences, communication studies, psychology and marketing.

Historically, content analysis can be traced back to the early 20th century when it was primarily applied to study communication and media content (Berelson, 1952). Since that time, content analysis has evolved and expanded its scope, embracing various data sources, including traditional media, online content, interviews, surveys, and archival documents. Content analysis is commonly employed in text contents, where researchers examine written or spoken words to detect and further investigate certain patterns and themes. Its data can be in a variety of documents, transcripts, social media posts, and advertisement forms. Researchers also use content analysis for visual content that involves examining images, photographs, or video footage to derive insights from visual cues and symbols (Neuendorf, 2017). Content analysis is an important research method in various disciplines. However, it can be time-consuming and challenging to automate. It is particularly well-suited for longitudinal studies, allowing researchers to track changes in content over time (Krippendorff, 2018).

## 1.11 Rationale for conducting this study.

The impact of MIH on children and their families is multifaceted. Clinically, affected teeth can be more susceptible to rapid caries progression and post-eruptive enamel breakdown, often leading to pain and increased sensitivity. Additionally, the aesthetic concerns related to MIH can affect a child's self-esteem and social interactions with their peers. For families, the condition can lead to increased numbers of dental visits, higher treatment costs in some countries, and the emotional strain of seeing their child in discomfort.

There is a growing recognition of the impact of MIH. None the less, there is still a need for further research in the fields of understanding the aetiological factors, improving diagnostic criteria, and developing effective preventive and therapeutic strategies are areas that require more in-depth exploration. Moreover, the psychosocial implications of MIH on children and the emotional, functional and financial impact on their families are relatively under-researched areas. The psychosocial and economic aspects, in particular, were recommended as a field for future research in the most recent Clinical Practice Guidance for Children with MIH produced by the

European Academy of Paediatric Dentistry (EAPD) (Lygidakis et al., 2022). Long-term studies into the effect of MIH and its treatment on patients and their families are currently extremely lacking in the literature. The current study was planned and undertaken with these recommendations in mind.

# Chapter 2: Materials and Methods

## 2.1 Study Design

This study followed up and investigated a cohort of children with a diagnosis of MIH who had been involved in a previous descriptive observational study that aimed to report the initial clinical features, baseline dental anxiety and OHQoL of children presenting for specialist consultation at Leeds Dental Institute in relation to defective first permanent molars (FPMs) (Al Bahar, 2017). This prospective follow-up study was designed to evaluate each subject's current dental anxiety and OHQoL to investigate if either had changed since their original visit some 7-8 years previously. In addition, the impact that the presenting condition and/or associated specialist treatment had had on each subject's family was also investigated.

The previous study (AlBahar 2017) was a descriptive observational study of a cohort of children recruited from those presented in 2015 for specialist consultation at Leeds Dental Institute in relation to defective first permanent molars (FPMs). This previous study aimed to evaluate a range of baseline clinical variables, including baseline dental anxiety and oral health-related QOL (OHRQoL), (Al Bahar, 2017). These children were originally recruited at ages 6-12 years. The present follow-up study aimed to investigate children from this group with a specific diagnosis of MIH (n=82) to determine what hospital-based specialist care they had subsequently required and received, as well as what hospital-based specialist care was still planned. It also aimed to reassess the current self-reported OHRQoL and dental anxiety and compare these with the findings recorded at their original specialist paediatric dentistry consultation at the Leeds Dental Institute. The current OHRQoL and the dental anxiety were assessed by asking the adolescent participants to complete the same two questionnaire tools they had completed in the previous study at the start of their treatment (the Quality of Life Questionnaire: Child Oral Health Impact Profile – Short Form 19 [COHIP SF 19] and the Faces version of the Modified Child Dental Anxiety Scale [MCDASf]). The current study also aimed to investigate what impact the dental condition, and any associated treatment had had on the adolescents'

families using the Family Impact Scale questionnaire (FIS). Additional open questions were added to capture what participants and parents thought about the treatment they had received at LDI and if they had any further comments about their experience while coming to LDI.

### 2.1.1 Original Plan Prior to COVID-19

The onset of the global pandemic brought about a shift in our strategy and the method of execution. Our original plan was to invite the participants to clinical visits to LDI for clinical examination and to fill out the questionnaires in addition to conducting face-to-face interviews. However, as the pandemic's impacts became evident, this resulted in the halt of postgraduate clinical duties for an indefinite period. It was also thought likely that many families may have not wished to attend non-essential hospital appointments for some time in the future. These unexpected factors, therefore, necessitated an alteration of our approach to conducting the study. Clinical examination was not feasible, nor were the participants asked for face-to-face interviews. In response, we had to adopt a remote working approach and shift to telephone and mail/email correspondence instead. The revised plan, although different in its method, aimed to capture the same core objectives of the study.

## 2.2 Ethical approval

Ethical approval for this study was sought and received from:

- The NHS HRA - Research Ethics Committee Yorkshire and The Humber 21/12/2021 (REC 289650) (REC: 21/PR/1069). (Appendix 1)
- The Leeds Teaching Hospitals NIHS Trust Research & Innovation Committee: LTHT R&I number DT22/148475 REC 21/PR/1069, 17/08/2022.

## 2.3 Data Protection and Confidentiality

Data protection regulations, as required by the University of Leeds and The Leeds Teaching Hospitals NHS Trust, and contemporary GCP guidelines were followed. All patient information was kept securely at the University of Leeds in a locked cupboard in the principal investigator's office, only

accessible by the lead investigator and the supervisors. No identifiable patient data other than a uniquely generated identification number which could only be decoded by the principal investigator or lead investigator. They were used for reporting and statistical analysis. No identifying participant information has been or will be included in any reports of this research.

## 2.4 Participant selection

Following ethical approval, attempts were made to recruit all the adolescents with MIH and their parents/carers who had participated in a previous study conducted from 2015 to 2017 following referral at that time to the Leeds Dental Institute (LDI) for the management of defective first permanent molars.

Some 105 participants were recruited into the original study, 82 of whom had a diagnosis of MIH. The recruited children were referred to the LDI from several sources, which included general dental practitioners and specialist paediatric dentists.

### 2.4.1 Inclusion criteria

- Participants with a diagnosis of MIH who had been recruited to the previous study carried out in 2015-17.
- Participants and their parents/guardians who consented to take part in this follow-up study.

### 2.4.2 Exclusion criteria

- Adolescents and/or their parents/guardians who did not wish to take part.
- Adolescents/family who could not be contacted.

## 2.5 Family Contact

### 2.5.1 Initial invitation letter

Each potential participant and their family were contacted by sending an invitation and information letter by post (Appendix 2). This letter served both as an introduction to the study and why they had been contacted. It avoided the possibility of a phone call being perceived as an unexpected ‘cold call’.

The addresses of the potential participants were sourced from the old and new Salud system (the clinical recording system used in the Leeds Dental Hospital).

The principal investigator and lead clinical supervisor's email addresses and a contact telephone number were included in the letter, although no positive action was required from the participants or their guardians at this stage. The letter gave a brief outline of the study and its purpose, reminded the adolescents and parents of their participation in the previous study and informed them that the lead investigator would be making a telephone call to explain the current study. It was explained that participation was entirely voluntary and that a telephone call would be made in the next few days to ask if the family was willing to participate. In addition to the invitation letter, an age-appropriate participant information sheet for participants (i.e., 'below 16 years of age', or '16 years and above'), and a parent/carer information sheet were included in this first postal package.

### 2.5.2 Telephone call

Following 7-10 working days after posting the invitation letter, the first telephone call was made by the lead investigator (MH). The period between postage and the first telephone call was adjusted accordingly during periods of postal disruption caused by national postal strikes, which also impacted the running of the study. The telephone numbers of the potential participants were sourced from the old and new Electronic Patient Record (EPR) system (Salud®). The landline telephone numbers were called first if available on the system.

During the first telephone call, the lead investigator asked if the invitation letter had been received. If the letter had not been received, the home address was checked for any changes. In either case, the present study was briefly explained, and the lead investigator asked if the parent/caregiver and the adolescent would be interested in taking part in this updated study. The family was advised that their participation would be entirely voluntary, and if they did not wish to take part, they were reassured that this would not affect any of their child's clinical treatment or follow-up. The participants who

wished to take part were also asked to choose to fill out the questionnaires and answer the questions in a paper version by post or electronically by email. For those who gave a verbal agreement and preferred a postal method for participation, a package of documents was subsequently sent by post. Others who requested the electronic route received the package by email.

#### 2.5.2.1 Non-Responding Telephone Numbers

For potential participants who could not be contacted initially due to unavailable or answered phone numbers using the Dental EPR system, the Paediatric Department Secretary's office was asked to search for any additional contact phone numbers using their Trust PAS system. A pre-dialling code was dialled before calling to ensure that the landline number appears to the call receiver instead of "Unknown" or "Withwheld". Telephone calls for non-responding numbers were made during five different months on different days of the week and at different times of the day to try to reach potential participants. The non-responding numbers of the potential participants were excluded at the end of the recruitment period.

### 2.5.3 The main study package

Following the first phone contact, the main package included documents as follows: The study package was sent to the confirmed, agreed address by Royal Mail tracked delivery mail or by email according to each participant's preference.

- A further copy of the invitation letter (Appendix 2)
- Age-appropriate information sheet for the participant (Appendix 3, 4)
- Participant consent form for participants above 16 years of age or a participant assent form for under 16 years of age (Appendix 5, 6)
- Participant questionnaires included dental anxiety and QOL questions in the MCDASf & COHIP-SF19 questionnaires, as well as instructions about how to complete the questionnaires. There were also four open-ended questions inviting feedback from the participants about their experiences at their visits to LDI (Appendix 7)
- Information sheet for the parent/carer (Appendix 8)

- Consent form to be completed by the parent carer (Appendix 9)
- Parent/carer questionnaire comprising of the Family Impact Scale (FIS) and instructions about how to complete the questionnaire. There were further questions designed to ascertain details of how they travelled to LDI for the treatment, and three open questions inviting feedback about the participants' and parents' experiences during their visits to LDI (Appendix 10)

A postage-paid pre-addressed return envelope and instructions about exactly which documents to return were also included to facilitate the return of the completed forms and questionnaires. It was addressed to the project lead clinical supervisor with 'MIH Study' written on the envelope.

When the completed forms were returned, the lead investigator checked to ensure that all the required forms and questionnaires had been completed and enclosed. In case of incomplete forms, an additional phone call was planned to obtain the missing part by phone if possible. If no contact could be established, then the only option was to exclude from the study. Consent forms were then checked and countersigned by the lead investigator. A copy of the completed and countersigned consent forms was then sent back to the family for their records with a thank you letter containing a £10.00 voucher as a token of appreciation for the time taken to complete the questionnaires. (Appendix 11)

### 2.5.4 Online Preference

To facilitate the process of receiving, filling and sending back the questionnaires, online receiving and return of questionnaires by email was another choice for participants. In the first phone call, the participants were asked if they preferred to be contacted by post or email. In case email was chosen, participants were asked for their email addresses so the same documents could be sent as electronic documents by email. These included fillable forms (PDF) of the consent forms and questionnaires to be emailed back to the lead investigator. When the completed electronic questionnaires were received, the returned documents were printed out, and then the same process was followed, with the printed questionnaires being kept with all other data.

A thank you letter was sent by post together with the countersigned consent form and the £10.00 shopping voucher. (Appendix 11)

## 2.6 The Child Participant Questionnaire

### 2.6.1 Quality of life: COHIP-SF19

Each participant's self-reported current oral health-related quality of life was assessed using The Child Oral Health Impact Profile - Short Form 19 (COHIP-SF19) (Appendix 7). This is a self-reported questionnaire consisting of 19 items, of which five are under the 'Oral Health subscale, four under the 'Functional Well-being' subscale, and 10 under the combined 'Social-Emotional Well-being' subscale. Answers to the questions, were assessed on a five-point Likert scale from «never» (4 points) to «almost constant» (0 points) and on the reverse scale for two questions with positive wording. The last two questions' scores were reversed as they are positively worded questions. The total score was calculated by summing the scores on all responses, the maximum possible score was 76 points which corresponded to the highest quality of life OHRQoL (Kriachkova et al., 2022). The results of this questionnaire were compared with results captured when the same tool was administered at the beginning of the previous study. The lead investigator recalculated the baseline scores for the 18 participants who were recruited for this study. The reason for the recalculation is to have a unified method for calculating the COHIP-SF19 baseline scores from the previous study and the current scores from this study.

### 2.6.2 Dental Anxiety: MCDASf

The participants' self-reported current dental anxiety was assessed using the Modified Child Dental Anxiety Scale (faces) Modified Child Dental Anxiety Scale (faces) (Appendix 7). The (MCDASf) is a simple scale with five faces. The scores range from 8 to 40 with "not worried" = 1, "very slightly worried" = 2, "fairly worried" = 3, "worried a lot" = 4 and "very worried" = 5 (Barbosa et al., 2022). The results of this questionnaire were compared with results captured when the same tool was administered at the beginning of the previous study.

### 2.6.3 Open questions inviting experiential feedback from participants

Four additional questions, based on questions used in previous Department of Paediatric Dentistry surveys of patient experience, were included in the package to invite participants to comment on their experiences at LDI. Each question left space for free-text answers.

The additional questions were:

*When you had your teeth treated at Leeds Dental Institute:*

*- Was there anything you remember as being particularly difficult?*

*- Was there anything that was good or easier than you thought it would be?*

*- Was there anything we could have done better?*

*- Is there anything else you would like to tell us about your teeth or visits to LDI?*

## 2.7 The Parent/Carer questionnaire

### 2.7.1 Impact of the Condition and Treatment on the Family (Family Impact Scale)

In order to assess the social and economic impact on families, the Family Impact Scale (Appendix 10) was used. It consists of 14 items that are divided into three main categories. Parental/family activity, parental emotion, family conflict and financial burden.

The questionnaire instructions were modified slightly to capture a better picture of the impact on the family as they were asked, “We want you to think about how your child's dental enamel problems, and the treatment provided, have affected the family overall since your child’s treatment at LDI started”. “Related to your child’s dental enamel condition and treatment. The FIS scores are computed by summing all of the item scores. Since there were 14 questions, the final score could vary from 0 to 56, for which a higher score translates to a greater degree of impact of a child's oral condition on the functioning of parents/caregivers and the family (Barbosa and Gavião, 2009).

### 2.7.2 Questions to capture information relating to appointment-associated travel to and from the LDI

Four additional questions were developed to ask about transport details for clinical visits to the LDI from the time of the participants' initial diagnosis. The questions aimed to capture the mode of transport used, the approximate time of travel and the approximate cost of travel if known:

*-When you attended appointments at Leeds Dental Institute, what mode(s) of transport did you use for your journey?*

*Car, Bus, Train, Taxi, Walking, Other (please specify)*

*-Approximately what was the cost of transportation for each journey (in one direction)?*

*Nil £1-5 £5-10 £10-20 £20-30 £30-40 £40-50 more than £50*

*-What was the approximate total time for your journey in one direction, door-to-door, from your home to the hospital?*

*-Overall, how would you describe your journeys to and from LDI?*

*Very Easy Easy Moderate Difficult Very Difficult*

### 2.7.3 Open questions inviting experiential feedback from parents/carers

Three additional questions based on those used in Departmental surveys of patient experience were included to invite participants to comment on their experiences at LDI.

*What did we do well?*

*What could we have done better?*

*Is there anything else you would like to tell us about your child's dental enamel condition or visits to LDI?*

## 2.8 Clinical record data

To determine the clinical history for each participant, information about each visit to the LDI from the first contact at the beginning of the previous study was collected. The data collected included: date, department, who saw the patient, reason for visit (consultation, treatment, type of treatment, who saw the participant, and relevant comments from the clinician. This included a review of LDI dental clinical records for all clinical entries from the first date of inclusion in the preceding original study up to the 23rd of May, 2023. For each

participant, the information was collected by reviewing records within the LDI electronic patient clinic record systems (SALUD Enterprise) and (New SALUD) in addition to written records that were kept in a box that was secured in the principal investigator's office from the previous study (Figure 1).

**Table2. 1** Data Extracted from the Clinical Records

| Data extracted from the records | |
|---|---|
| **Gender** | |
| **Age** | |
| **Ethnicity** | |
| **Date of first visit** | |
| **Relevant medical history** | |
| **Postal code** | The first section (3/4 characters) of the postcode and first digit of the second section |
| **Original compliant** | |
| **Oral Hygiene status** | |
| **State of FPM** | Number of FPM affected and the severity* of each one. |
| **State of incisors** | Incisors demarcation involvement, yes or no. |
| **Orthodontic factors** | Skeletal pattern, malocclusion presence, yes or no |
| **Mode of treatment** | No intervention, L.A., IHS, G.A, or combination of the previous modes |
| **Planned treatment** | Initial consultant opinion and their initial treatment plan |
| **Received treatment** | Actual performed treatment and if it is different from the planned treatment |
| **Discharge date** | |
| **Previous COHIP-SF19 and MCDASf scores** | Recalculated the previous scores in the same method of calculating the current ones. Recalculation of the scores from the previous study to ensure uniformity of the method of calculations to ensure accuracy. |

* In the baseline study, MIH severity was recorded as mild, moderate, or severe, as per Da Costa-Silva et al., 2011

## 2.9 Piloting the Questionnaires

Seven University students (three post-graduate dental students, three undergraduate dental students and one medical student) provided feedback on drafts of the study questionnaires, which have already been detailed in section 2.9. The feedback from the pilot was incorporated into the final study. Based on the feedback, file formats were edited to ensure compatibility with smartphones' different operating systems. Additionally, the

questionnaire instructions were made to be more appropriate to the age group of study participants. No other significant issues were identified.

## 2.10 Data Storage and Security

All the physical records (consents/assents, filled questionnaires, additional notes written by the participants) were stored in a file box and placed in a locked cupboard in a secure office in the School of Dentistry. Access was only available to the principal investigator and the lead investigator.

## 2.11 Participant Confidentiality

Each participant's data set was allocated a unique reference number accessible only by the principal investigator and the lead investigator, and no identifiable patient data was extracted from these original physical records. Each participant was assigned the same unique identification number that was used in the previous study for continuity and uniformity purposes. The scores calculated from the questionnaire and all other answers were extracted and recorded using only a unique, sequentially generated identification number for each individual participant. All data was stored on a flash USB drive in an Excel file format and analysed without any identifying participant information being included. Consequently, no patient-identifying data is used in any reports of this research.

## 2.12 Negative Feedback

Any reports of unsatisfactory treatment or concerns experienced by the participants or their parents/caregivers during treatment were identified and reported to the principal investigator and the care team. The care team discussed the area of concern and further investigated the issue. Where appropriate, the participant or the participant's parent/caregiver was contacted by the clinical team for further details and to agree on any further actions.

## 2.13 Safeguarding

It was recognised that answering questionnaires that have a social-emotional well-being aspect, such as the COHIP-SF19 and/or MCDASf, could potentially bring back unpleasant memories and could cause distress for the participants or serve to bring up questions or concerns about current or future care. In the event that such a situation were to arise, the participant information sheet invited the participants and/or parents/carers to either alert the principal investigator or to contact the lead clinical supervisor directly for help and support (contact details were listed).

## 2.14 Data analysis

Originally, data were to be analysed by using SPSS software. Later, due to the low response rate and the subsequent small participants number, it was decided that data would be analysed by Excel datasheet, comparison tables and graphs. Nvivo software version 1.7.1 (QSR International) was used for the coding process for the free text content.

**Study outline**

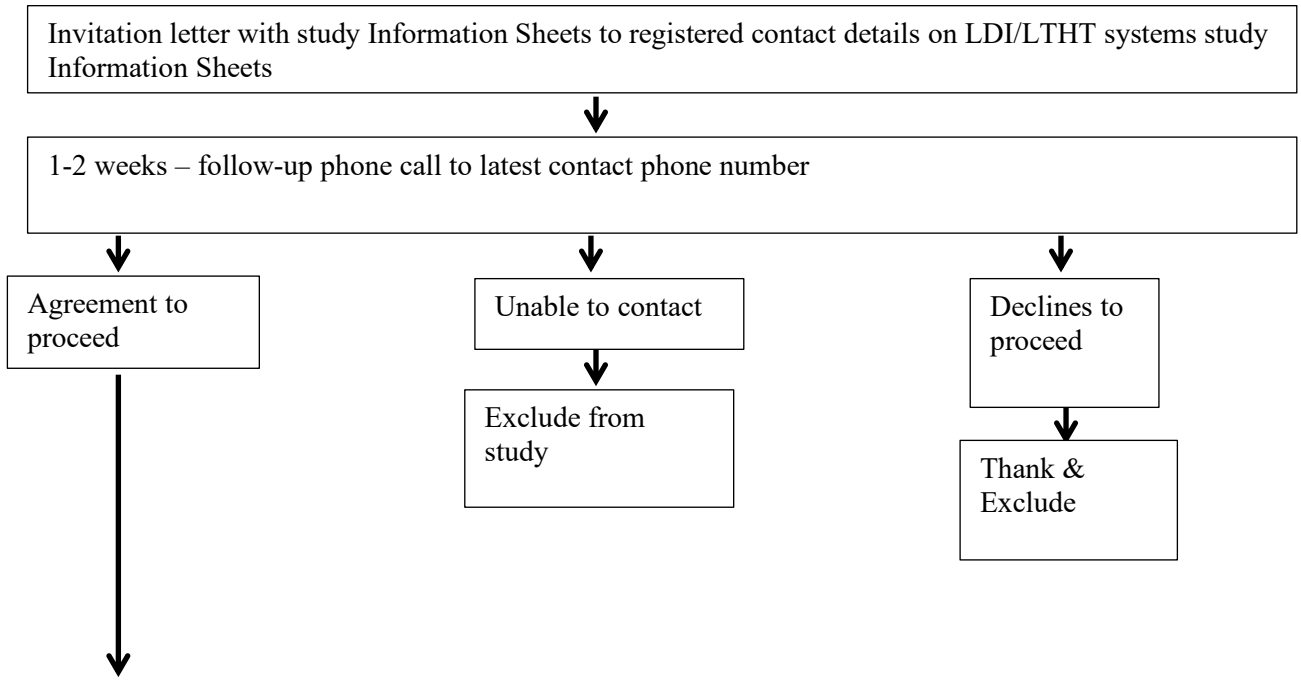
**Phase 1**

- Pilot studies to test the design of questionnaires

**Phase 2**

Prospective study to investigate current level of self-reported dentistry-related anxiety (MCDAS) and Oral Health Impact Profile (COHIP) of study participants and self-reported impact of their dental condition on their family/carers (FIS)

Communications with parents/carers and study participants



Verbal explanation of study and what would be involved. Explain voluntary and remote completion of questionnaires and return via post. Ask the participants if they would like to fill the questionnaires and the answer the questions online (by email) or by post.

- Explain package will be sent out and contents
- Explain if participants want to fill the forms online or by post
- Explain information sheets, consent forms and questionnaires
- Explain second call after package/email received to answer any questions
- If they decide to take part parent/carer and child participant can complete consent forms and questionnaires and returned in the pre-paid envelope/send the email.
- If they have any questions, are uncertain about anything or simply if they would prefer to, they can delay completing the consent forms or any parts of the questionnaires until the investigator's second phone call.

↓

Send out instruction letter, consent forms and questionnaires

↓

Second phone call.

- Check package has arrived
- Answer any questions.
- Confirm consent and request return/email of completed consent forms and questionnaires.

↓

Receive return package.

- Data extracted from questionnaires into Excel spreadsheet
- Consent forms and paper /printed questionnaires stored in a flash memory.
- If not received, one further phone call attempt to remind – if this fails, excluded. Still OK to decline at this stage – if declines: exclude

**Phase 3**

Participant-specific data capture from clinical records regarding clinical appointments and treatment delivered at each appointment

Signed consent, thank you letter, and the voucher sent

↓

Individual participant's LDI patient clinical records were reviewed, and information was recorded as detailed in Table 2.1

**Phase 4**
**Data analysis and reporting**

| |
|---|
| Statistical analysis of variation between original (pre-treatment) and new (post-treatment) MCDAS(f) and COHIP scores.<br>Reporting of findings from the Family Impact Scale and additional satisfaction/feedback questions<br>Reporting of data captured from clinical records and analysis in anonymised and collated form. |

Figure2. 1: Flow Chart of the study phases

# Chapter 3: Results

## 3.1 Piloting questionnaires

The updated questionnaire that was developed following the occurrence of COVID-19, was pre-tested with seven university students who completed the questionnaires on paper and electronically with a focus on identifying any challenges they faced or errors they identified during the process.

After completing the process of filling out the questionnaires, their verbal feedback was documented by contacting them by telephone.

They were asked to provide their insight on:

- The clarity of the questions
- The clarity of the instructions
- The length of time to complete the task
- Compatibility of the electronic files on their devices (PCs, mobile phones, tablets)

The respondents gave the following feedback which was used to improve the design:

- No participants found any difficulty in understanding the questions

-Three participants found the instructions confusing

- The average length of time to complete all questionnaires and answer the questions was 15 minutes.

- Three participants could not manage to open the electronic file due to compatibility issues with their devices.

Changes were mainly made to the electronic files compatibility with the several electronic devices taking the feedback into consideration before sending the packages to the participants.

## 3.2 Participant selection and re-recruitment

### 3.2.1 Initial screening of records

Some 105 participants were recruited into the previously completed original study, 82 of whom had a diagnosis of MIH. The records of these 82 were reviewed in the SALUD patient record system, and no reasons for not re-contacting any participants were identified.

### 3.2.2 Contact information

A most recently recorded postal address was available for all 82 participants, as was a most recently recorded contact telephone number.

Introductory letters were generated and posted to all 82 participants. These were generated and posted in two separate tranches to avoid disruption by UK postal strikes and the Christmas holiday period.

The outcome of each stage of the recruiting process - contacting and subsequently securing consent and the return of Main Study Packages - is summarised in Figure 2-1

### 3.2.3 Telephone contact

Telephone contact was attempted following the agreed methodology. Forty-nine potential participants could not be contacted by telephone after the maximum attempts and therefore were excluded. Thirty-three families were successfully contacted by telephone, and of these 12 indicated they did not wish to take part in the study, so were also excluded. A total of 21 families agreed to proceed with the study at this first stage and were subsequently sent the Main Study Package.

### 3.2.4 Postal package

Main Study Packages were sent by post to the 21 families who had given verbal agreement. Only 18 families subsequently returned completed packages, leading to three further families being excluded. All returned packages included appropriately completed consents from both the participant and a parent /guardian and all questionnaires were found to have been adequately completed. Only one participant chose to receive the main study package electronically by email, while 17 participants preferred the package by post.

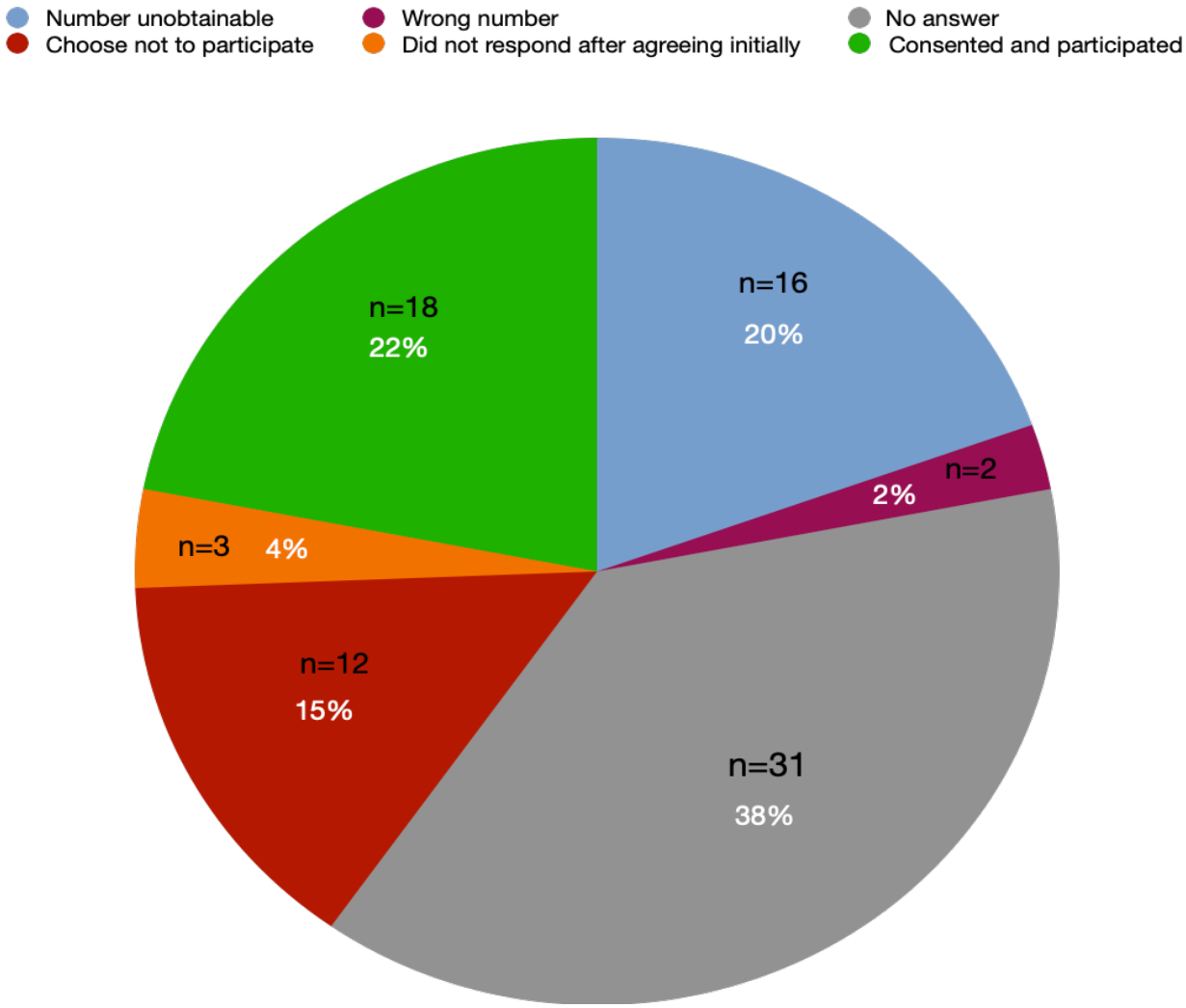



**Figure3. 1:** Pie chart of the outcome of potential participants' contact and recruitment process.

## 3.3 Participant demographics

### 3.3.1 Gender

Of the 18 participants returning completed packages, 13 were male and five were female.

### 3.3.2 Ethnicity

Of the final 18 participants, 12 participants were of White-British ethnicity; five participants were of White-non-specified ethnicity and one participant was of mixed White-British and Arab ethnicity.

### 3.3.3 Age

**Figure 3.2** shows the age distribution of the subjects agreeing to participate in this follow-up study at the time of receiving their response questionnaires. The mean age of participants in this follow-up study was 15.6 years. For the same cohort of participants, the mean age was 8.05 years at the time of recruitment to the original baseline study.

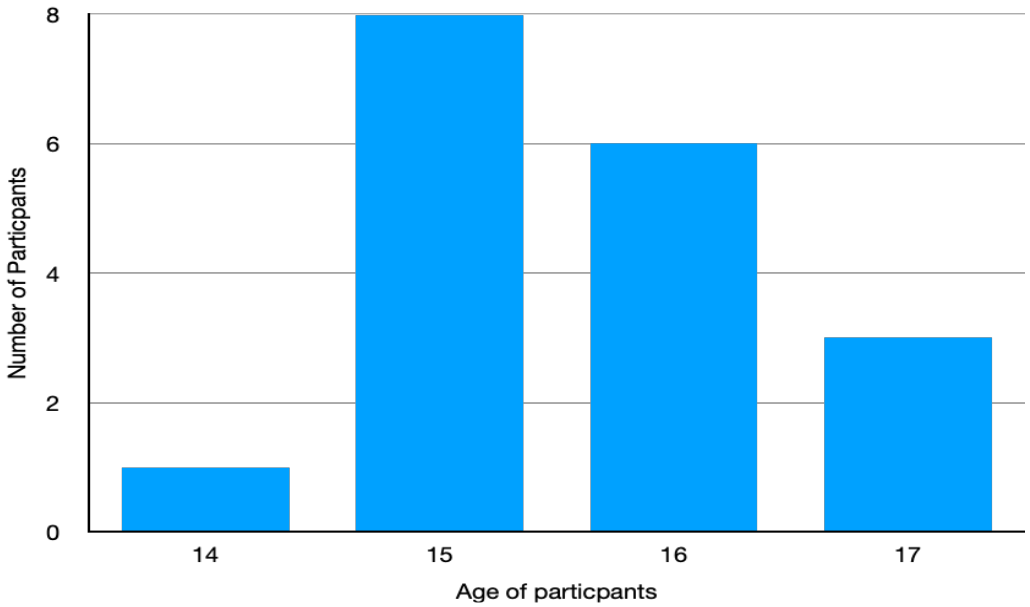



Figure3. 2: Age distribution of the participants at the time of recent follow-up study questionnaire completion.

## 3.4 MIH severity

The participants were scored for MIH severity at the initial presentation during the baseline study (Da Costa-Silva et al., 2011). Three participants were recorded as having an MIH severity score of mild, four were moderate and 11 were severe.

**Table3. 1** MIH Severity for each participant recorded pre-treatment in the baseline study

| Participant ID | Severity |
|---|---|
| 2 | Mild |
| 11 | Severe |
| 12 | Moderate |
| 28 | Severe |
| 30 | Mild |

| | |
|---|---|
| 43 | Severe |
| 46 | Moderate |
| 49 | Severe |
| 51 | Mild |
| 60 | Moderate |
| 63 | Severe |
| 65 | Severe |
| 66 | Severe |
| 97 | Severe |
| 99 | Severe |
| 100 | Severe |
| 107 | Severe |
| 108 | Moderate |

## 3.5 Mode of pain/patient management for treatment

The modality(ies) of pain/patient management for delivery of treatment as determined by retrospective review of Dental Hospital records is summarised in Table 3.2, where the subjects have also been ranked by MIH severity. Local anaesthesia was used for nine participants. Treatment under general anaesthesia was used for 8 participants. Inhalation sedation was used for two participants in combination with other modes of management. Only one participant had a combination of all three modes of management during the treatment. Four participants were managed without administrating L.A., G.A. or IHS.

Seven out of the total eight of G.A. admissions were for participants with severe MIH. None of the three participants with mild MIH had G.A, L.A. or IHS interventions, and only one of those with moderate MIH had G.A.

Seven of the 11 subjects with severe MIH had GA.

**Table3. 2** Mode(s) of patient management and MIH severity ranked by increasing baseline MIH severity

| Participant ID | Severity | G.A. | L.A. | IHS |
|---|---|---|---|---|
| 2 | Mild | ✗ | ✗ | ✗ |
| 30 | Mild | ✗ | ✗ | ✗ |
| 51 | Mild | ✗ | ✗ | ✗ |
| 12 | Moderate | ✗ | ✓ | ✗ |

| | | | | |
|---|---|---|---|---|
| 46 | Moderate | ✗ | ✓ | ✗ |
| 60 | Moderate | ✗ | ✗ | ✗ |
| 108 | Moderate | ✓ | ✗ | ✗ |
| 11 | Severe | ✗ | ✓ | ✓ |
| 28 | Severe | ✗ | ✓ | ✗ |
| 43 | Severe | ✓ | ✗ | ✗ |
| 49 | Severe ★ | ✓ | ✓ | ✓ |
| 63 | Severe | ✓ | ✓ | ✗ |
| 65 | Severe ★ | ✓ | ✓ | ✗ |
| 66 | Severe | ✗ | ✓ | ✗ |
| 97 | Severe | ✓ | ✗ | ✗ |
| 99 | Severe | ✓ | ✗ | ✗ |
| 100 | Severe | ✓ | ✗ | ✗ |
| 107 | Severe | ✗ | ✓ | ✗ |

✓ - indicates this modality was used for this participant
★ - subjects subsequently had GA following failed treatment attempts under LA

## 3.6 Dental Treatment received

A range of dental procedures and treatments were performed for the participants from their first visit to LDI until the time of conducting this current study (table 3.3). Extraction was the most performed procedure, performed on 11 participants, of which nine had severe MIH. Seven participants had restorations, and three participants had extractions in addition to restorations. Fissure sealants were provided for three participants. Tooth Mousse was given to three participants and two participants had temporisation. Six had some form of active orthodontic intervention.

**Table3. 3** Dental Treatment received and MIH severity grouped and ranked by increasing baseline MIH severity.

| Participant ID | Severity | Restoration | Extraction | F/S | Tooth Mousse | Temporisation | Orthodontic Intervention |
|---|---|---|---|---|---|---|---|
| 2 | Mild | ✗ | ✗ | ✓ | ✓ | ✗ | ✓ |
| 30 | Mild | ✗ | ✗ | ✗ | ✓ | ✓ | ✗ |
| 51 | Mild | ✗ | ✗ | ✗ | ✗ | ✗ | ✓ |
| 12 | Moderate | ✓ | ✗ | ✗ | ✗ | ✗ | ✗ |
| 46 | Moderate | ✗ | ✓ | ✗ | ✗ | ✗ | ✓ |
| 60 | Moderate | ✓ | ✗ | ✓ | ✗ | ✗ | ✗ |
| 108 | Moderate | ✗ | ✓ | ✗ | ✗ | ✗ | ✗ |
| 11 | Severe | ✓ | ✓ | ✗ | ✓ | ✓ | ✗ |
| 28 | Severe | ✓ | ✗ | ✓ | ✗ | ✗ | ✓ |
| 43 | Severe | ✗ | ✓ | ✗ | ✗ | ✗ | ✗ |
| 49 | Severe | ✓ | ✓ | ✗ | ✗ | ✗ | ✗ |
| 63 | Severe | ✓ | ✓ | ✗ | ✗ | ✗ | ✓ |
| 65 | Severe | ✗ | ✓ | ✗ | ✗ | ✗ | ✗ |
| 66 | Severe | ✓ | ✗ | ✗ | ✗ | ✗ | ✗ |
| 97 | Severe | ✗ | ✓ | ✗ | ✗ | ✗ | ✗ |
| 99 | Severe | ✗ | ✓ | ✗ | ✗ | ✗ | ✗ |
| 100 | Severe | ✗ | ✓ | ✗ | ✗ | ✗ | ✗ |
| 107 | Severe | ✗ | ✓ | ✗ | ✗ | ✗ | ✓ |

✓ - indicates this modality was used for this participant

## 3.7 Number of clinical visits

The total number of clinical visits per participant was calculated and is shown in Table 3.4 and Figure 3.3. The lowest number of visits was two, while the highest number of visits for a participant reached 42 visits in the combined Paediatric and Orthodontic departments. The average number of

visits specifically to the Paediatric Department only (including G.A. visits) is 4.94 visits per patient with a range of 2 to a maximum of 10 visits. For the 4 participants visiting the orthodontic department, total visits to that Department ranged from 23 to 40.

**Table3. 4** Number of clinic visits and final treatment pathway outcome for each participant at the time of the current study.

| Participant ID | Number of Paediatric Visits | Number of Orthodontic visits | Total Number of Visits for those also visiting Orthodontics | Outcome at time of follow-up study |
|---|---|---|---|---|
| **2** | 5 | 27 | 33 | Not yet discharged from Orthodontic Department. |
| **11** | 10 | | | Discharged |
| **12** | 7 | | | Discharged |
| **28** | 10 | | | Discharged |
| **30** | 5 | | | Discharged |
| **43** | 3 | | | Discharged |
| **46** | 2 | 40 | 42 | Discharged |
| **49** | 7 | | | Not yet discharged from the Paediatric Dept. Pt returned after being discharged. |
| **51** | 4 | | | DNA |
| **60** | 2 | | | Discharged |
| **63** | 7 | 25 | 32 | Not yet discharged from Orthodontic Department. |
| **65** | 5 | | | Discharged |
| **66** | 5 | | | DNA |
| **97** | 4 | | | Discharged |
| **99** | 3 | | | Discharged |
| **100** | 3 | | | Discharged |
| **107** | 3 | 23 | 26 | Not yet discharged from Orthodontic Department. |
| **108** | 4 | | | Discharged |

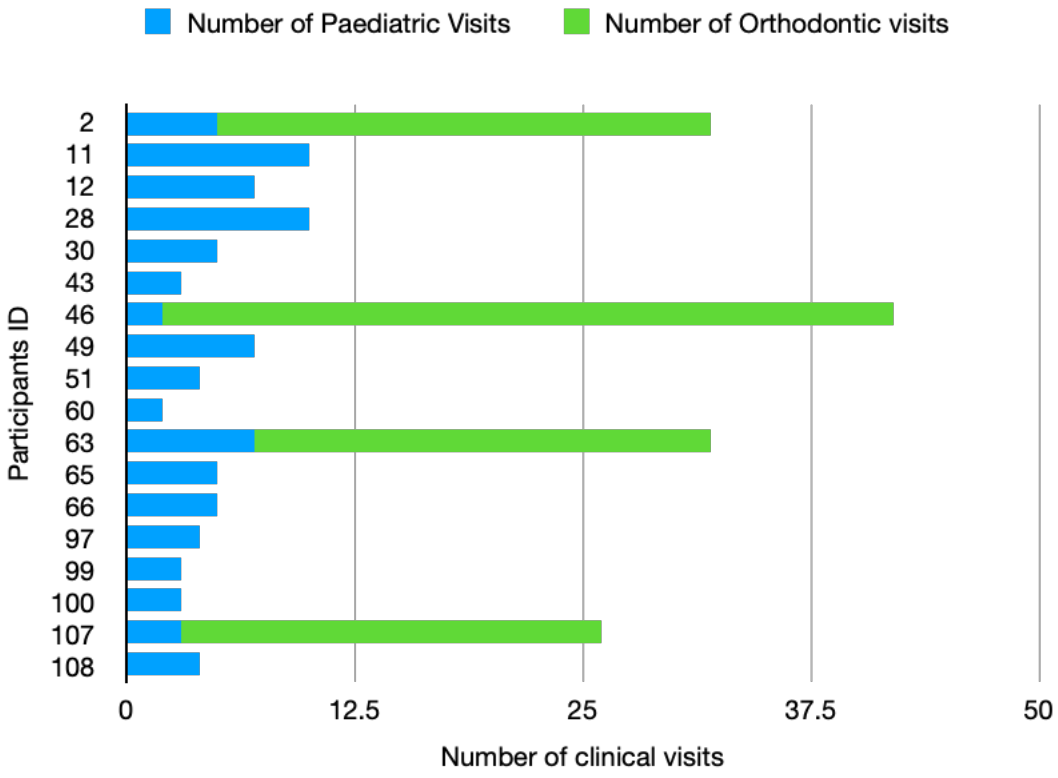



**Figure3. 3** Number of clinical visits by department

## 3.8 Discharged/Continuing treatment

At the time of completing the follow-up questionnaires, 12 participants had been discharged from the LDI after treatment completion. Two further participants had failed to attend for follow-up. Four participants were still being seen at LDI for continuing treatment. In three of these cases, they are for orthodontic treatment only. One case is still being seen in the Paediatric Department for caries treatment after being referred back again in 2023.

## 3.9 Active orthodontic treatment

Among the 18 participants in this follow-up study, six participants had orthodontic intervention. Two participants had relatively short courses of interceptive orthodontic treatment at the Paediatric Department without additional visits to the Orthodontic Department. The four having active treatment in the Orthodontic Department had visit numbers to orthodontics ranging from 23 to 42, and 3 of these were still under active orthodontic care/follow-up at the time of this follow-up (Current) study.

**Table3. 5** Active Orthodontic treatment

| Participant ID | Orthodontic Factors | Orthodontic Intervention | Number of Orthodontic Visits |
|---|---|---|---|
| **2** | Severe crowding<br>Posterior Cross-bite | Wears Elastic<br>Unlikely to be fully recovered | 27 |
| **28** | Thumb sucking | Habit breaker constructed and given | Treated at Paediatric Department |
| **51** | Thumb sucking | Tongue crib device | Treated at Paediatric Department |
| **46** | UL1 Impacted & Ectopically Erupted | Removable Partial Denture<br>Denture broke several times | 42 |
| **63** | Missing UR5<br>Anterior Cross-bite<br>Spacing in both arches | Fitted Cervical Headgear | 25 |
| **107** | Overjet = 9 mm | Twin Block | 23 |

## 3.10 COHIP-SF19 scores

**Table 3.5. 1** shows a summary of COHIP SF19 scores giving the mean and median for baseline, current and change in the total scores and the scores for each individual domain.

| Domain | | Mean | Median | Range |
|---|---|---|---|---|
| OH | Baseline | 13.6 | 13.5 | 6-20 |
| | Current | 13.1 | 14 | 6-20 |
| | Change | -0.4 | 0 | -8 to 5 |
| FN | Baseline | 12.6 | 13 | 7-16 |
| | Current | 13.6 | 14 | 6-16 |
| | Change | 1 | 2 | -6 to 7 |
| SC | Baseline | 30.1 | 31 | 18-39 |
| | Current | 27 | 29.5 | 8-38 |
| | Change | -3.1 | -1 | -21 to 8 |
| TOTAL | Baseline | 56.3 | 58 | |
| | Current | 53.6 | 58 | |
| | Change | -2.6 | 2 | |

### 3.10.1 Changes in individual COHIP-SF19 domain scores

#### 3.10.1.1 COHIP-SF-19 Domain 1: Oral health well-being

The first domain of the COHIP-SF-19 questionnaire comprised of five questions designed to explore Oral Health and Well-being. Only seven participants showed improvement in this domain, with, eight participants showing a decline, and three participants showing no change in their score (Table 3.6).

**Table3. 6** Baseline and follow-up COHIP-SF19 Domain 1: Oral health well-being domain (OH) scores, and their changes between baseline and follow-up studies, ranked according to the difference between baseline and current scores.

| Participant ID | Baseline Oral Health well-being score | Current Oral Health well-being score | Difference in Oral Health well-being scores |
|---|---|---|---|
| 46 | 15 | 20 | 5 |
| 12 | 14 | 17 | 3 |
| 28 | 10 | 13 | 3 |
| 43 | 15 | 18 | 3 |
| 108 | 11 | 14 | 3 |
| 63 | 13 | 14 | 1 |
| 65 | 13 | 14 | 1 |
| 51 | 6 | 6 | 0 |
| 66 | 14 | 14 | 0 |
| 100 | 10 | 10 | 0 |
| 11 | 13 | 12 | -1 |
| 97 | 18 | 16 | -2 |
| 107 | 12 | 10 | -2 |
| 49 | 18 | 15 | -3 |
| 99 | 13 | 10 | -3 |
| 30 | 16 | 12 | -4 |
| 60 | 20 | 14 | -6 |
| 2 | 14 | 6 | -8 |

Note
Green indicates an increase in the COHIP-SF19 score (an improvement in QoL).
Red indicates a decrease in COHIP-SF19 score (i.e. a decline in OH QoL)
The maximum possible score for **Oral Health well-being** domain = 20

#### 3.10.1.2 Functional well-being

12 participants showed improvement when ranked according to their score difference in the functional well-being domain of the COHIP-SF19, which

was the greatest number of improving scores in any of the three domains. Six participants had a decline in their scores.

**Table3. 7** Baseline and follow-up COHIP-SF19 Domain 2: Functional well-being scores, and their changes between baseline and follow-up studies, ranked according to difference in Functional Domain (FN) score from Baseline to Current studies.

| Participant ID | Baseline Functional well-being score | Current Functional well-being score | Difference in Functional well-being scores |
|---|---|---|---|
| 108 | 7 | 14 | 7 |
| 11 | 12 | 16 | 4 |
| 51 | 11 | 15 | 4 |
| 63 | 13 | 16 | 3 |
| 97 | 12 | 15 | 3 |
| 28 | 10 | 12 | 2 |
| 43 | 14 | 16 | 2 |
| 65 | 14 | 16 | 2 |
| 99 | 11 | 13 | 2 |
| 100 | 9 | 11 | 2 |
| 12 | 13 | 14 | 1 |
| 66 | 15 | 16 | 1 |
| 46 | 16 | 15 | -1 |
| 60 | 15 | 14 | -1 |
| 107 | 15 | 14 | -1 |
| 49 | 14 | 12 | -2 |
| 30 | 14 | 10 | -4 |
| 2 | 12 | 6 | -6 |

The maximum possible score for **Functional well-being** domain = 16

### 3.10.1.3 Social-Emotional well-being

Eight participants showed improvement in their scores, but 10 participants showed a decline in their social-emotional scores, the greatest number of declining scores of all three domains. This domain also accounted for the largest component of the decline in the total scores, partly due to the relatively high possible score for this domain. However, improvements in the SC domain scores were relatively smaller, and declines were relatively higher than those seen in the other two domains.

**Table3. 8** Baseline and follow-up COHIP-SF19 Domain 3: Social-Emotional well-being domain scores, and their changes between baseline and follow-up studies, ranked according to difference.

| Participants ID | Baseline Social-Emotional well-being score | Current Social-Emotional well-being score | Difference in Social-Emotional well-being scores |
|---|---|---|---|
| 11 | 25 | 33 | 8 |
| 108 | 23 | 31 | 8 |
| 30 | 31 | 35 | 4 |
| 65 | 27 | 30 | 3 |
| 66 | 32 | 35 | 3 |
| 46 | 34 | 36 | 2 |
| 28 | 18 | 19 | 1 |
| 97 | 31 | 32 | 1 |
| 43 | 30 | 29 | -1 |
| 63 | 39 | 38 | -1 |
| 12 | 33 | 31 | -2 |
| 49 | 31 | 25 | -6 |
| 107 | 29 | 23 | -6 |
| 2 | 31 | 22 | -9 |
| 99 | 30 | 20 | -10 |
| 51 | 33 | 20 | -13 |
| 60 | 35 | 19 | -16 |
| 100 | 29 | 8 | -21 |

The maximum possible score for **Social-Emotional well-being** domain = 40

### 3.10.2 Changes in COHIP-SF19 total scores

In the COHIP-SF19 questionnaire, higher scores correspond to higher reported oral health-related quality of life for the participants. Baseline total COHIP-SF19 scores ranged from 38 to 70 with a mean of 56.3, and Current total COHIP-SF19 scores ranged from 29 to 68 with a mean of 53.7. However, changes between the two census points for individuals varied considerably with the range of change being +18 to -23.

10 participants showed higher total scores in their current COHIP-SF19 questionnaire compared to their baseline scores in the original study (before they received any treatment for the MIH). This suggests some improvement in the oral health-related QoL for these 10 participants. The other eight participants scored lower scores in the COHIP-SF19 questionnaire compared to their baseline scores (before they received any hospital treatment) indicating a decline in their oral health-related QoL.

**Table3. 9** COHIP-SF19 baseline and current total scores

| Participant ID | Baseline COHIP-SF19 Total score | Current COHIP-SF19 Total score | Change in total COHIP SF-19 score |
|---|---|---|---|
| 2 | 57 | 34 | -23 |
| 11 | 50 | 61 | 11 |
| 12 | 60 | 62 | 2 |
| 28 | 38 | 44 | 6 |
| 30 | 61 | 57 | -4 |
| 43 | 59 | 63 | 4 |
| 46 | 65 | 71 | 6 |
| 49 | 63 | 52 | -11 |
| 51 | 50 | 41 | -9 |
| 60 | 70 | 47 | -23 |
| 63 | 65 | 68 | 3 |
| 65 | 54 | 60 | 6 |
| 66 | 61 | 65 | 4 |
| 97 | 61 | 63 | 2 |
| 99 | 54 | 43 | -11 |
| 100 | 48 | 29 | -19 |
| 107 | 56 | 47 | -9 |
| 108 | 41 | 59 | 18 |
| | 1013 | 966 | |

Figures 3.4 and 3.5 compare changes in total COHIP-SF19 scores with COHIP-SF19 total scores at Baseline and Current studies. There appears to be an inverse relationship between the Baseline total COHIP-SF19 score and subsequent change and, conversely, a positive relationship between the Current total COHIP-SF19 score and the change in total score between studies.

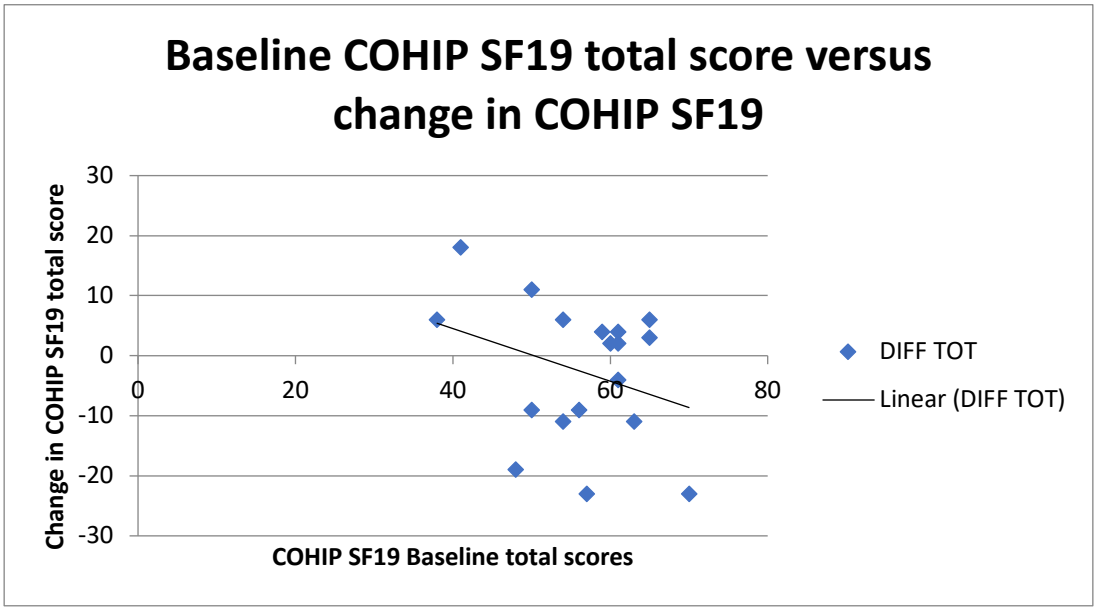



**Figure3. 4** Baseline COHIP-SF19 total score versus change in COHIP-SF19

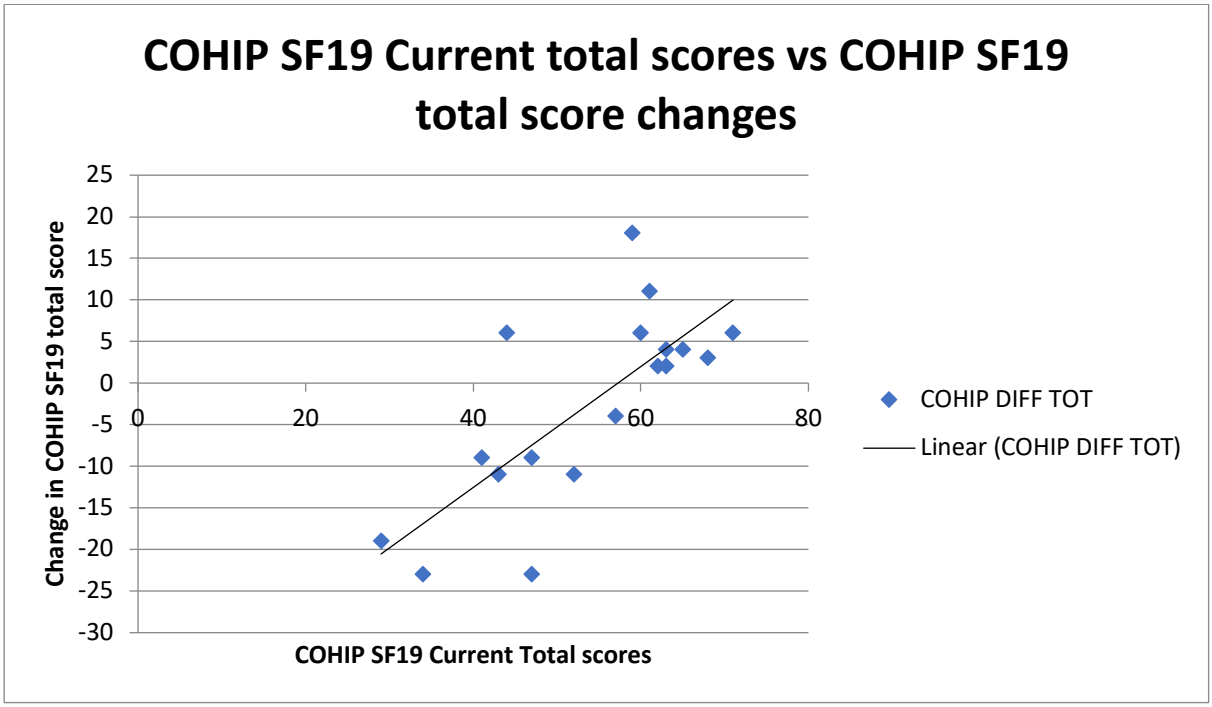



**Figure3. 5** Current COHIP-SF19 total score versus change in COHIP-SF19

### 3.10.3 Change in Total COHIP-SF19 score compared to change in individual domain scores

The change in total COHIP SF19 score and the changes in the individual domain scores are presented in Table 3.10. The socio-emotional (SC) domain score had the most influence on the change in overall score (Table 3.12 and Figure 3.4), but this would be expected, as this domain contributes more than half of the overall score. It is, however, noteworthy that some of

the largest changes in total-score-changes were in a negative direction, and in many of those individuals, the majority of total-score-change was in the SC domain.

**Table3. 10** COHIP SF19 domain and sum-total score differences sorted by ascending sum-total COHIP SF19 score difference

| Participant ID | OH domain score change | FN domain score change | SC domain score change | Change in total COHIP SF19 score |
|---|---|---|---|---|
| 108 | 3 | 7 | 8 | 18 |
| 11 | -1 | 4 | 8 | 11 |
| 28 | 3 | 2 | 1 | 6 |
| 46 | 5 | -1 | 2 | 6 |
| 65 | 1 | 2 | 3 | 6 |
| 43 | 3 | 2 | -1 | 4 |
| 66 | 0 | 1 | 3 | 4 |
| 63 | 1 | 3 | -1 | 3 |
| 12 | 3 | 1 | -2 | 2 |
| 97 | -2 | 3 | 1 | 2 |
| 30 | -4 | -4 | 4 | -4 |
| 51 | 0 | 4 | -13 | -9 |
| 107 | -2 | -1 | -6 | -9 |
| 49 | -3 | -2 | -6 | -11 |
| 99 | -3 | 2 | -10 | -11 |
| 100 | 0 | 2 | -21 | -19 |
| 2 | -8 | -6 | -9 | -23 |
| 60 | -6 | -1 | -16 | -23 |

| | |
|---|---|
| | - indicates a negative change from baseline |
| | - indicates a positive change from baseline |
| | - indicates no change from baseline |

OH – Oral Health domain (max possible score 20)
FN – Functional domain (max possible score 16)
SC – Socio-emotional domain (max possible score 40)
Maximum possible summated score = 76

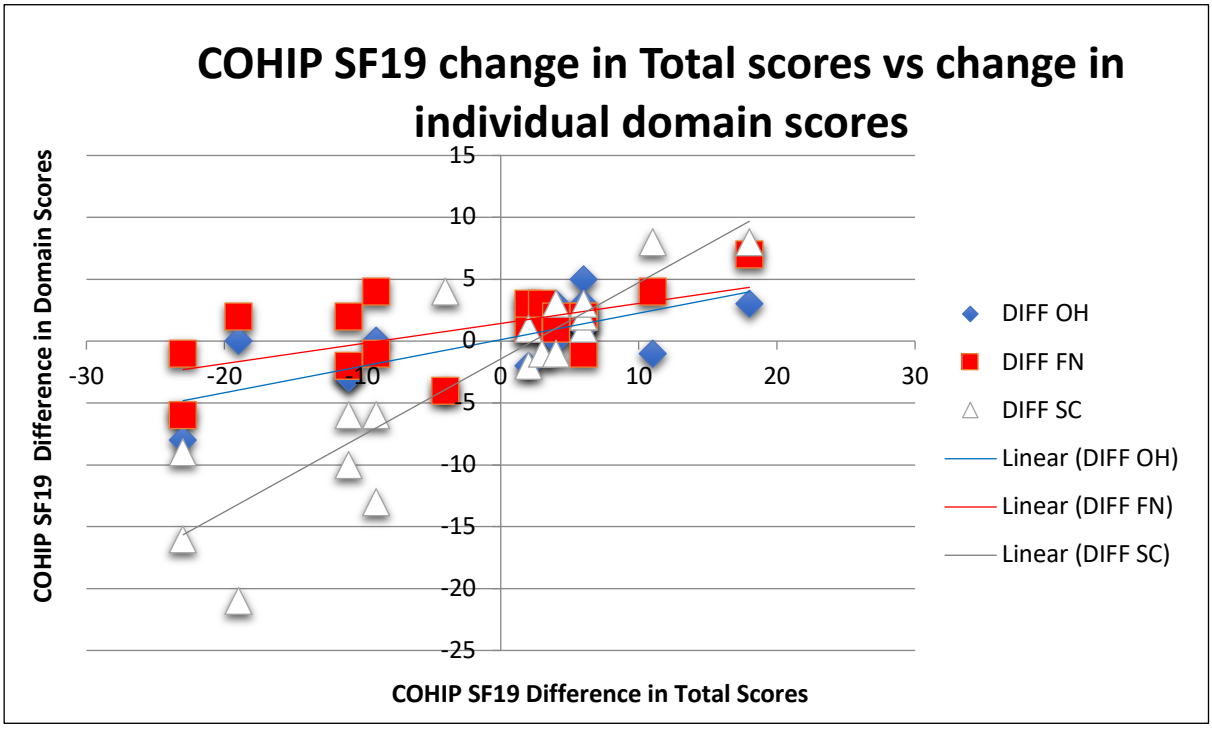



**Figure3. 6** COHIP SF19 change in Total scores vs change in individual domain scores.

### 3.10.4 changes in COHIP-SF19 scores compared to MIH severity

**Table3.10. 1** Baseline and changes in COHIP-SF19 scores ranked by increasing baseline MIH severity

| | | Pre-treatment COHIP-SF19 | | | | Change in COHIP-SF19 | | | |
|---|---|---|---|---|---|---|---|---|---|
| Pt ID | MIH Severity | Oral Health | Functional | Social-Emotional | Total | Oral Health | Functional | Social-Emotional | Total |
| 2 | Mild | 14 | 12 | 31 | 57 | -8 | -6 | -9 | -23 |
| 30 | Mild | 16 | 14 | 31 | 61 | -4 | -4 | 4 | -4 |
| 51 | Mild | 6 | 11 | 33 | 50 | 0 | 4 | -13 | -9 |
| 12 | Moderate | 14 | 13 | 33 | 60 | 3 | 1 | -2 | 2 |
| 46 | Moderate | 15 | 16 | 34 | 65 | 5 | -1 | 2 | 6 |
| 60 | Moderate | 20 | 15 | 35 | 70 | -6 | -1 | -16 | -23 |
| 108 | Moderate | 11 | 7 | 23 | 41 | 3 | 7 | 8 | 18 |
| 11 | Severe | 13 | 12 | 25 | 50 | -1 | 4 | 8 | 11 |
| 28 | Severe | 10 | 10 | 18 | 38 | 3 | 2 | 1 | 6 |

| | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|
| 43 | Severe | 15 | 14 | 30 | 59 | 3 | 2 | -1 | 4 |
| 49 | Severe | 18 | 14 | 31 | 63 | -3 | -2 | -6 | -11 |
| 63 | Severe | 13 | 13 | 39 | 65 | 1 | 3 | -1 | 3 |
| 65 | Severe | 13 | 14 | 27 | 54 | 1 | 2 | 3 | 6 |
| 66 | Severe | 14 | 15 | 32 | 61 | 0 | 1 | 3 | 4 |
| 97 | Severe | 18 | 12 | 31 | 61 | -2 | 3 | 1 | 2 |
| 99 | Severe | 13 | 11 | 30 | 54 | -3 | 2 | -10 | -11 |
| 100 | Severe | 10 | 9 | 29 | 48 | 0 | 2 | -21 | -19 |
| 107 | Severe | 12 | 15 | 29 | 56 | -2 | -1 | -6 | -9 |

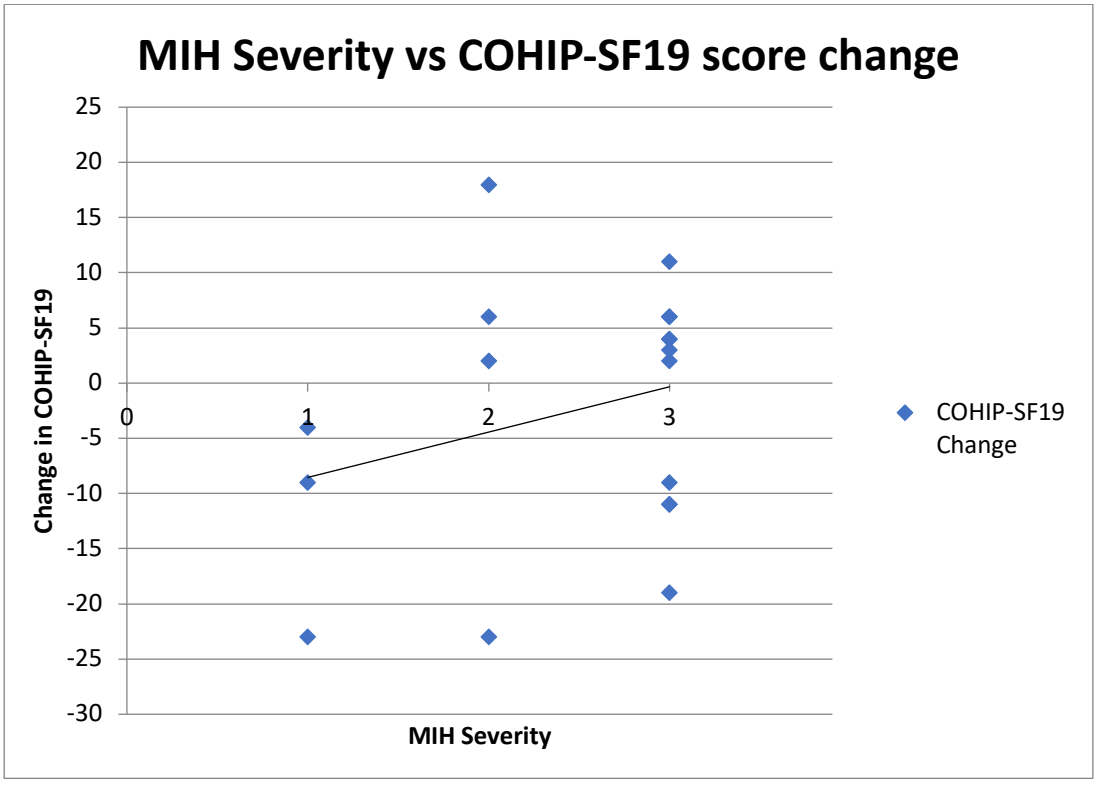



**Figure 3.6. 1** COHIP SF19 change in Total scores vs change in individual domain scores.

## 3.11 MCDASf scores

In the MCDASf questionnaire, higher scores correspond to a higher level of dental anxiety. In the current questionnaire, eight of the participants showed higher scores indicating more anxiety and ten participants had lower scores

indicating less anxiety compared to their original baseline scores (Table 3.11).

**Table3. 11** MCDASf baseline and current scores and change from baseline to current ranked by ascending change.

| Participant ID | Baseline MCDASf Score | Current MCDASf Score | Change in MCDASf score |
|---|---|---|---|
| 97 | 20 | 8 | -12 |
| 108 | 32 | 22 | -10 |
| 63 | 23 | 15 | -8 |
| 51 | 24 | 19 | -5 |
| 28 | 26 | 22 | -4 |
| 11 | 36 | 33 | -3 |
| 12 | 18 | 15 | -3 |
| 66 | 12 | 9 | -3 |
| 30 | 20 | 18 | -2 |
| 43 | 23 | 21 | -2 |
| 60 | 19 | 22 | 3 |
| 65 | 15 | 18 | 3 |
| 99 | 16 | 19 | 3 |
| 2 | 21 | 25 | 4 |
| 49 | 13 | 18 | 5 |
| 100 | 11 | 19 | 8 |
| 107 | 21 | 31 | 10 |
| 46 | 11 | 35 | 24 |

Green indicates a decrease in MCDASf score (less anxiety)

Red indicates a decrease in MCDASf score (more anxiety)

MCDASf score range 5-40, where lower score indicate less anxiety

The mean, median and range of MCDASf scores are shown in Table 3.11.1.

**Table3.11. 1** Mean, Median and Range of MCDASf scores

| | Mean | Median | Range |
|---|---|---|---|
| Baseline | 20.1 | 20 | 11-36 |
| Current | 20.5 | 19 | 8-35 |
| Change | 0.5 | -2 | -12 to 24 |

Plotting change in MCDASf score for each individual against their Baseline and Current scores shows some interesting trends (Figures 3.5 and 3.6). Those children with lower MCSADf at baseline (i.e., lower dental anxiety) tended to show increases in MCDASf from baseline to current and vice versa. Current MCDASf shows the opposite trend, ie those with higher MCDASf currently (post-treatment) tended to be those individuals where MCDASf had increased from baseline to current (ie the individual had become more dentally anxious) and vice versa.

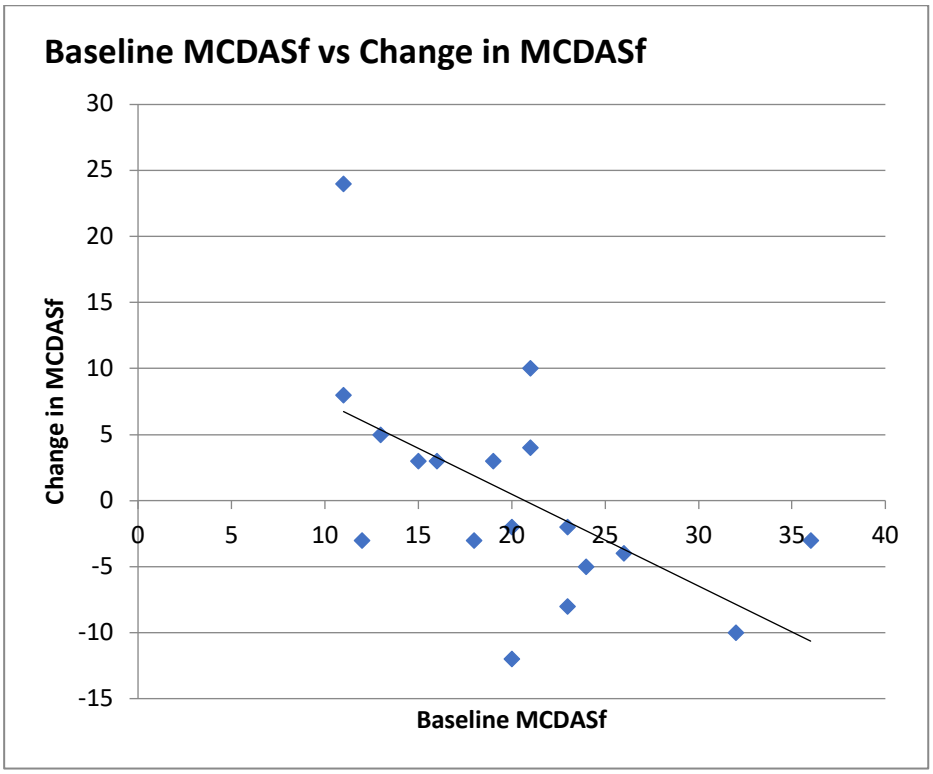



**Figure3. 7** Baseline MCDASf versus Change in MCDASf

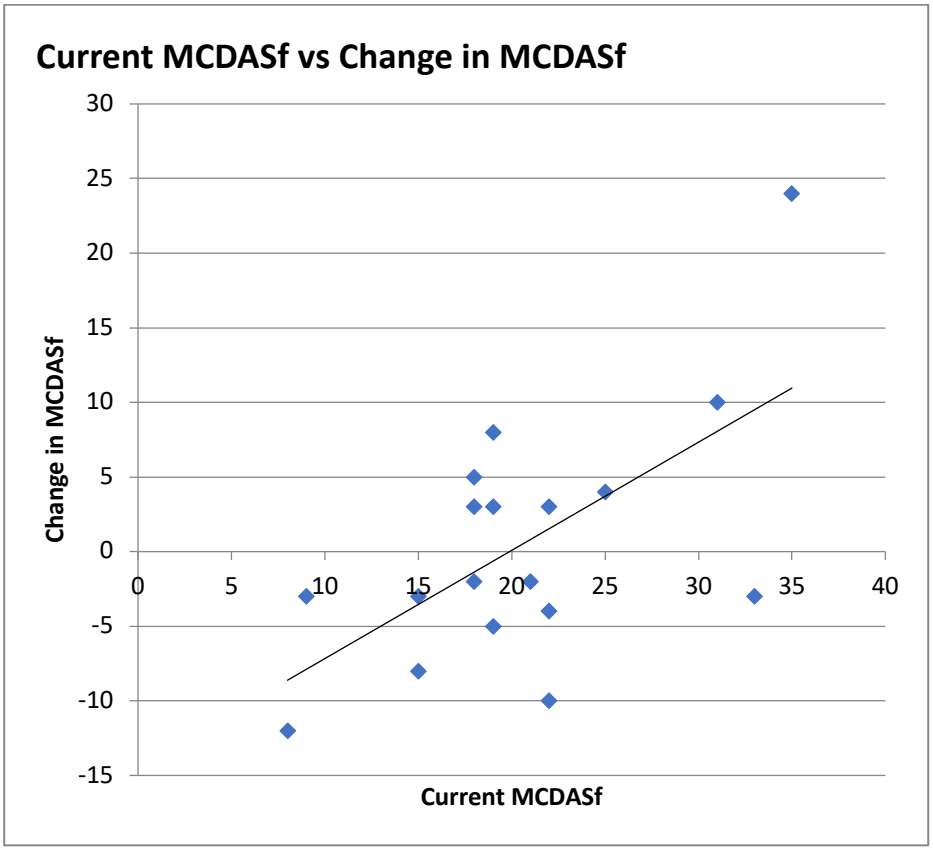



**Figure3. 8** Current MCDASf versus Change in MCDASf.

### 3.11.1 changes in MCDASf scores compared to MIH severity

**Table3.11 1** Baseline and changes in MCDASf scores ranked by increasing baseline MIH severity

| Participant ID | MIH Severity | MCDASf | |
|---|---|---|---|
| | | Pre Rx | Change |
| 2 | Mild | 21 | 4 |
| 30 | Mild | 20 | -2 |
| 51 | Mild | 24 | -5 |
| 12 | Moderate | 18 | -3 |
| 46 | Moderate | 11 | 24 |
| 60 | Moderate | 19 | 3 |
| 108 | Moderate | 32 | -10 |
| 11 | Severe | 36 | -3 |
| 28 | Severe | 26 | -4 |

| | | | |
|---|---|---|---|
| 43 | Severe | 23 | -2 |
| 49 | Severe | 13 | 5 |
| 63 | Severe | 23 | -8 |
| 65 | Severe | 15 | 3 |
| 66 | Severe | 12 | -3 |
| 97 | Severe | 20 | -12 |
| 99 | Severe | 16 | 3 |
| 100 | Severe | 11 | 8 |
| 107 | Severe | 21 | 10 |

## 3.12 Relationship between COHIP-SF19 and MCDASf score differences

For the majority of subjects (14) the direction of change in COHIP-SF19 total score (OH-related QoL was accompanied by a commensurate change in dental anxiety score (MCDASf) i.e. increases in COHIP SF19 (improved OH QoL) were accompanied by decreases in MCDASf (Dental anxiety) or vice versa. For only four subjects did dental anxiety increased when OH-related QoL improved or vice versa (Table 3.12 and Figure 3.5). In spite of the small numbers in this study, this suggests an association between these two variables (Figure 3.5).

Eight subjects had an improvement in OH-related QoL score (COHIP-SF19) accompanied by a decrease in dental anxiety score (MCDASf). A further eight subjects had a decline in their OH-related QoL score (COHIP-SF19) accompanied by an increase in dental anxiety score (MCDASf).

**Table3. 12** Differences in COHIP-SF19 total scores and in MCDASf total scores, and their changes between baseline and follow-up studies, ranked according to the difference in total COHIP-SF19.

| **Participant ID** | **Difference in COHIP-SF19 total score** | **Difference in MCDASf total score** |
|---|---|---|
| 108 | 18 | -10 |

| | | |
|---|---|---|
| 11 | 11 | -3 |
| 28 | 6 | -4 |
| 46 | 6 | 24 |
| 65 | 6 | 3 |
| 43 | 4 | -2 |
| 66 | 4 | -3 |
| 63 | 3 | -8 |
| 12 | 2 | -3 |
| 97 | 2 | -12 |
| 30 | -4 | -2 |
| 51 | -9 | -5 |
| 107 | -9 | 10 |
| 49 | -11 | 5 |
| 99 | -11 | 3 |

| | | |
|---|---|---|
| 100 | -19 | 8 |
| 2 | -23 | 4 |
| 60 | -23 | 3 |

Note
- A positive change (increase) in total COHIP SF-19 is indicative of an improvement in OH QoL (indicated in green). A decline in COHIP-SF19 score is indicative of a decrease in OH QoL (Red).
- A positive change (increase in MCDASf score indicates an increase in dental anxiety (indicated in Red). A decrease in MCDASf score is indicative of a decrease in dental anxiety (Green).

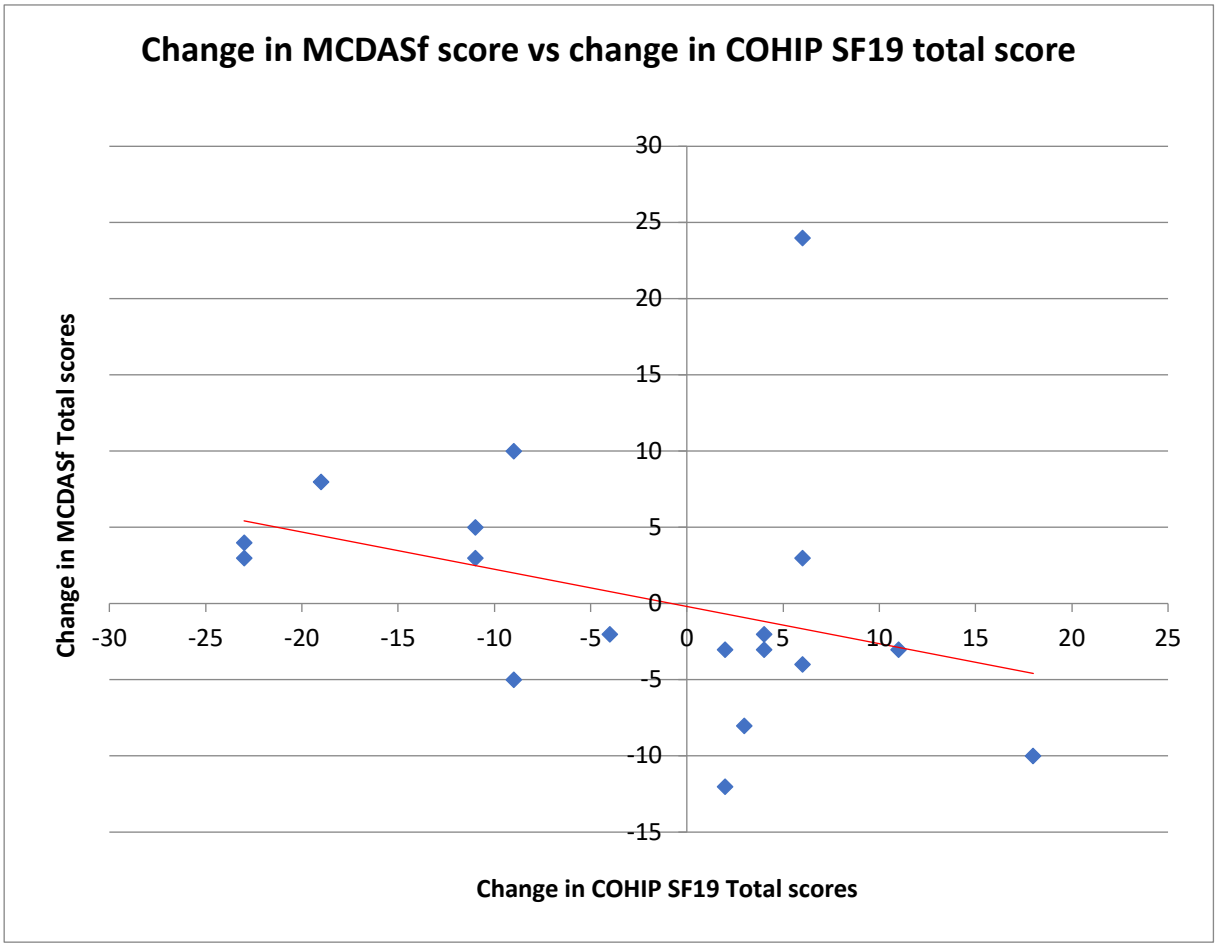



**Figure3. 9** Change in MCDASf score and change COHIP-SF19 total score.

## 3.13 FIS scores

In the FIS questionnaire, higher scores correspond to a greater impact on the family. The total score can range from 0 to a maximum of 56, where a higher score equates with greater impact. One participant's family scored 0

which indicates they felt there was no impact in all the areas of the FIS questionnaire. The highest score was 25 and the average score was 6, which suggests most families felt that impact on the family resulting from their child's MIH was relatively low.

### 3.13.1 Individual FIS domain scores

A full breakdown of the FIS individual domain and total scores for each participant's family is given in Table 3.13. The participants' scores are ranked in order of increasing total FIS score (i.e. increasing impact).

**Table3. 13** Family Impact Scale (FIS) domain and total scores ranked in order of increasing FIS total score

| **Participate ID** | FIS Parental Activities | FIS Parental Emotion | FIS Family Conflict | FIS Financial Burden | FIS Total score |
|---|---|---|---|---|---|
| 28 | 0 | 0 | 0 | 0 | 0 |
| 2 | 1 | 0 | 0 | 0 | 1 |
| 49 | 1 | 0 | 0 | 0 | 1 |
| 60 | 1 | 0 | 0 | 0 | 1 |
| 30 | 0 | 0 | 2 | 0 | 2 |
| 65 | 2 | 0 | 0 | 0 | 2 |
| 12 | 0 | 3 | 0 | 0 | 3 |
| 51 | 3 | 0 | 0 | 0 | 3 |
| 63 | 1 | 0 | 2 | 0 | 3 |
| 107 | 2 | 1 | 0 | 0 | 3 |
| 66 | 3 | 1 | 0 | 0 | 4 |
| 43 | 4 | 1 | 0 | 0 | 5 |
| 97 | 3 | 4 | 0 | 0 | 7 |
| 108 | 5 | 2 | 0 | 0 | 7 |
| 46 | 5 | 4 | 0 | 0 | 9 |
| 99 | 6 | 5 | 5 | 0 | 16 |
| 100 | 3 | 6 | 7 | 0 | 16 |
| 11 | 10 | 10 | 5 | 0 | 25 |

PA = Parental/Family Activities domain (score range 0-20)
PE = Parental Emotions domain (score range 0-16)
FC = Family Conflict domain(score range 0-16)
FB = Financial Burden domain (score range 0-4)
Total score range 0-56

#### 3.13.1.1 FIS Domain 1: Parental activates

Three participants scored 0 indicating no reported impact on parental/family activities. The highest score was 10 and the average score is 2.77 (maximum possible score 20)

#### 3.13.1.2 FIS Domain 2: Parental emotions

Eight participants' parents/carers reported no impact at all, scoring 0. The highest score was 10 and the average score was 2 (maximum possible score 16)

#### 3.13.1.3 FIS Domain 3: Family conflict

Family conflict was not impacted for thirteen participants who scored 0. The highest score was 7 while the average score was 1.16 (maximum possible score 16)

#### 3.13.1.4 FIS Domain 4: Financial burden

All participants scored 0 "never" in their response to the last question concerning the impact on family finances (maximum possible score 4), reporting no financial impact relating to their child's MIH.

## 3.14 Change in COHIP-SF19 total score and FIS total score

Both Table 3.14 and Figure 3.10 compare the change baseline in COHIP SF19 total score from baseline to current studies and the total FIS score. No obvious relationship is apparent between the change in COHIP-SF19 total scores from baseline to current when compared to the current FIS total scores.

**Table3. 14** Change in COHIP-SF19 total score compared to FIS total score ranked from most positive to most negative change in COHIP-SF19 total score.

| participant ID | COHIP-SF19 difference in total score | FIS Total score |
|---|---|---|
| 108 | 18 | 7 |
| 11 | 11 | 25 |
| 28 | 6 | 0 |
| 46 | 6 | 9 |
| 65 | 6 | 2 |

| | | |
|---|---|---|
| 43 | 4 | 5 |
| 66 | 4 | 4 |
| 63 | 3 | 3 |
| 12 | 2 | 3 |
| 97 | 2 | 7 |
| 30 | -4 | 2 |
| 51 | -9 | 3 |
| 107 | -9 | 3 |
| 49 | -11 | 1 |
| 99 | -11 | 16 |
| 100 | -19 | 16 |
| 2 | -23 | 1 |
| 60 | -23 | 1 |

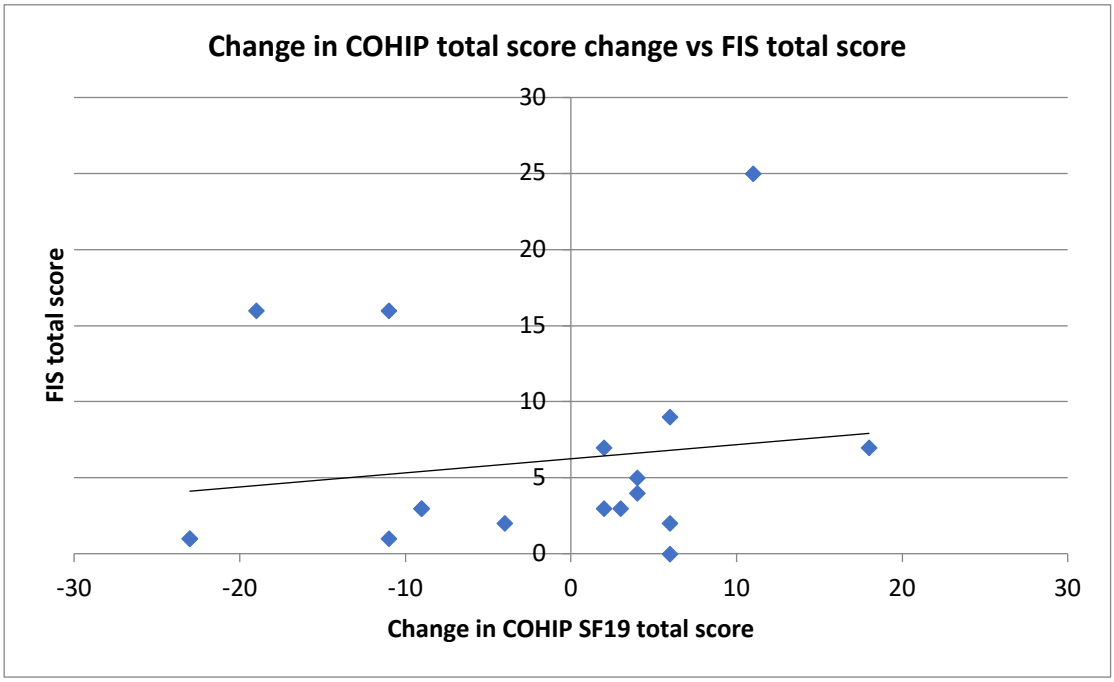



**Figure3. 10** Change in COHIP-SF19 total score vs FIS total score.

## 3.15 Change in MCDASf score and FIS total score

There was no apparent relationship between the change in MCDASf scores from the Baseline study to the Current study when compared to the current FIS total scores (Table 3.15 and Figure 3.11).

**Table3. 15** Change in MCDASf score versus FIS total score, ranked by ascending FIS total score.

| Participant ID | FIS Total score | Baseline MCDASf Total Score | Current MCDASf Total Score | Change in MCDASf |
|---|---|---|---|---|
| 28 | 0 | 26 | 22 | -4 |
| 60 | 1 | 19 | 22 | 3 |
| 2 | 1 | 21 | 25 | 4 |
| 49 | 1 | 13 | 18 | 5 |
| 30 | 2 | 20 | 18 | -2 |
| 65 | 2 | 15 | 18 | 3 |
| 63 | 3 | 23 | 15 | -8 |
| 51 | 3 | 24 | 19 | -5 |
| 12 | 3 | 18 | 15 | -3 |
| 107 | 3 | 21 | 31 | 10 |
| 66 | 4 | 12 | 9 | -3 |
| 43 | 5 | 23 | 21 | -2 |
| 97 | 7 | 20 | 8 | -12 |
| 108 | 7 | 32 | 22 | -10 |
| 46 | 9 | 11 | 35 | 24 |
| 99 | 16 | 16 | 19 | 3 |
| 100 | 16 | 11 | 19 | 8 |
| 11 | 25 | 36 | 33 | -3 |

Note: an increase in MCDASf score (shown in Red) indicates an increase in dental anxiety.

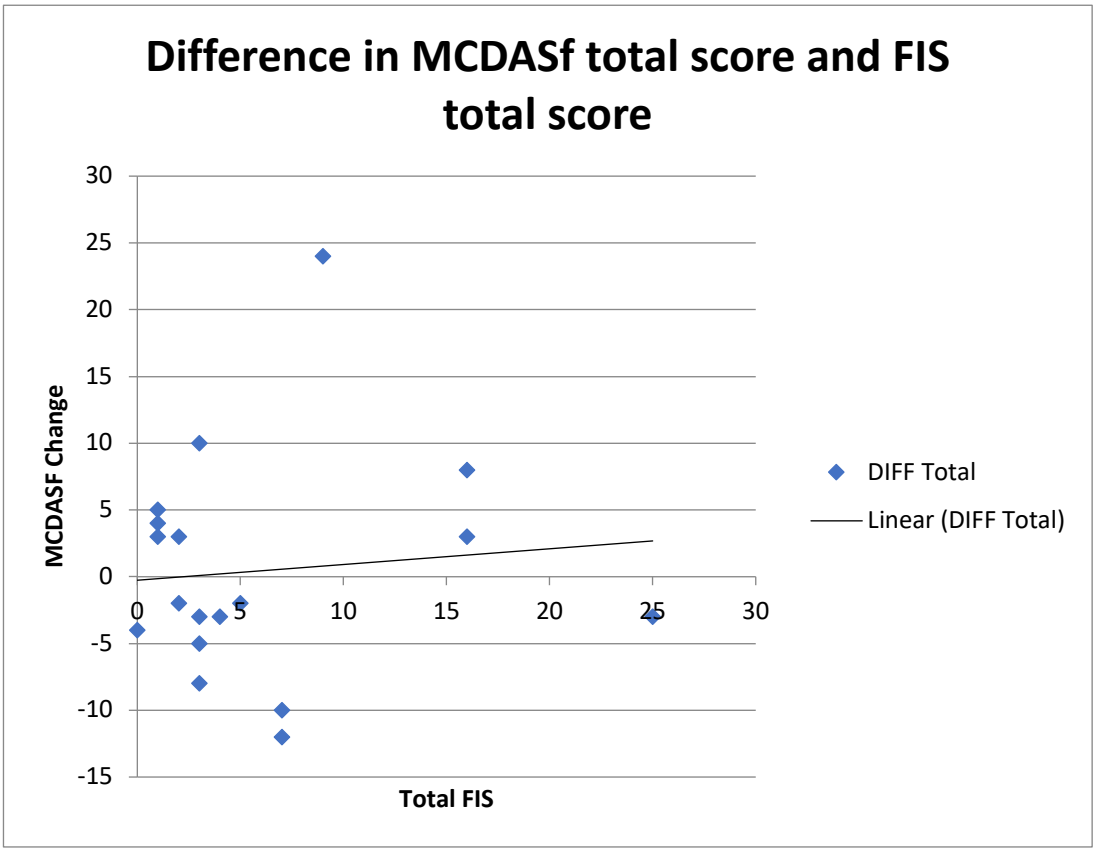



**Figure3. 11** Difference in MCDASf total score and FIS total score.

## 3.16 The relationship between Pain/Anxiety management employed and COHIP SF19, MCDASf and FIS

Patients were sorted according to the most invasive pain/anxiety management used for their care using the following scheme:
*No specific management* < *Local Analgesia* < *Inhalation Sedation* < *General Anaesthesia*
Change in total COHIP SF19, change in MCDASf and FIS score were then compared graphically to identify if any possible trends were apparent. In addition, baseline total COHIP-SF19 and MCDASf were investigated for any association between these baseline scores and the treatment modalities subsequently employed.

### 3.16.1 Change in COHIP SF19

The most invasive pain/anxiety management modality employed was compared to the change in COHIP SF19 scores and is represented graphically in Figure 3.12. Overall, there was little evidence that different

management modalities were consistently associated with changes in total COHIP SF19 scores, either positive or negative. However, of some note is that 5 of 8 subjects who received GA exhibited an increase in COHIP SF19 scores (i.e. suggesting an increase in OH-related QoL), and the subject with the highest overall increase was in the GA cohort. Interestingly, the largest negative changes occurred in three subjects who received treatment under GA (subject 100), LA (subject 60) and no specific management respectively (subject 2).

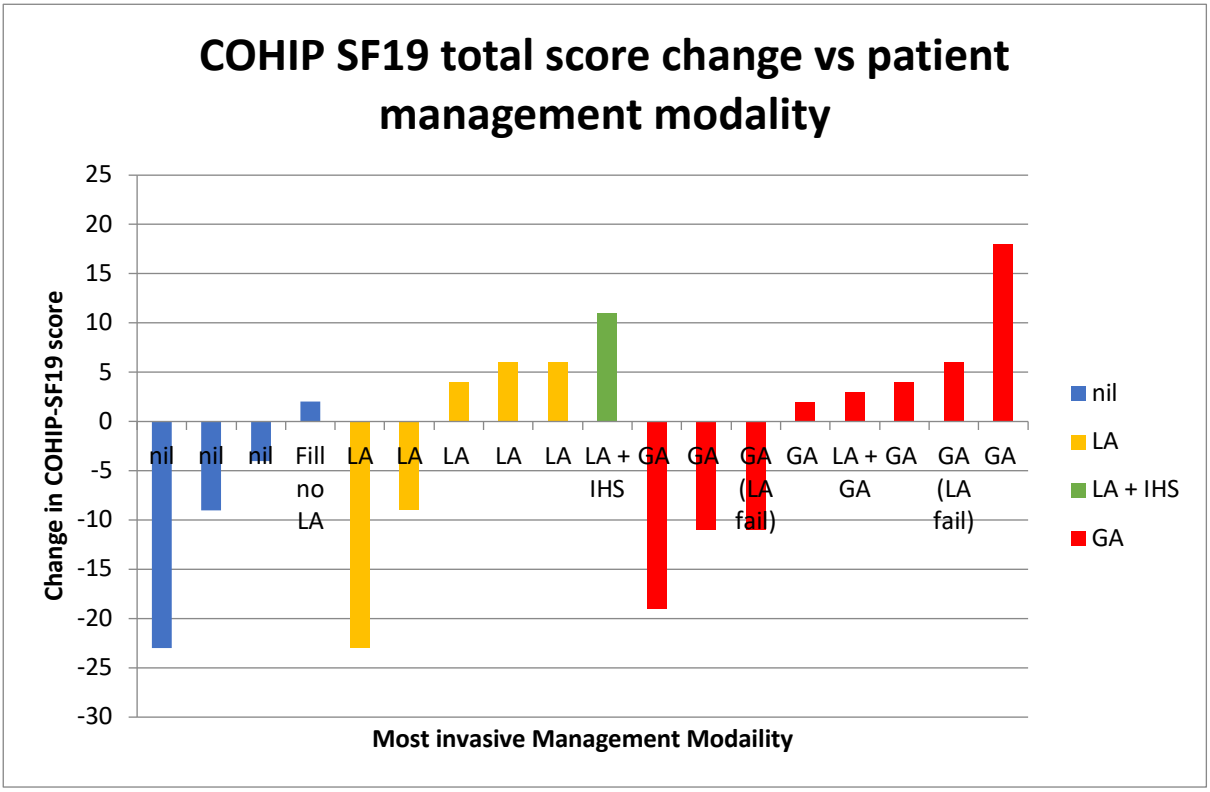



**Figure3. 12** COHIP SF19 total score change vs patient management modality.

### 3.16.2 Baseline COHIP SF19 scores and most invasive patient management modality subsequently employed

Baseline COHIP SF19 scores versus the most invasive patient management modality subsequently employed are displayed in Figure 3.13. Overall, no trend suggesting that baseline COHIP SF19 might be related to the management modality employed was apparent.

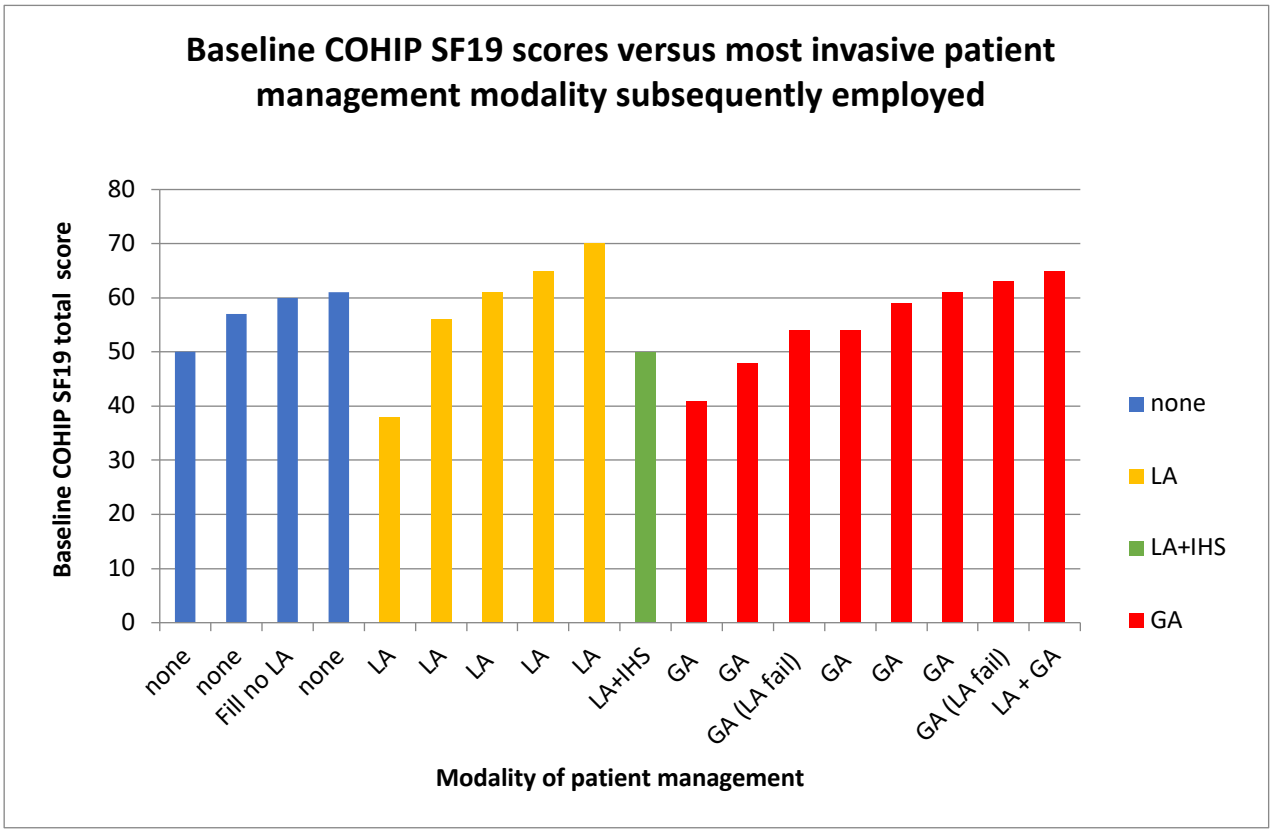



**Figure3. 13** Baseline COHIP SF19 scores versus most invasive patient management modality subsequently employed..

### 3.16.3 Change in MCDASf

The most invasive management modality was compared to changes in MCDASF score and this is shown graphically in Figure 3.14. The greatest increases in MCDASf from Baseline to Current studies (i.e. increases in Dental Anxiety) were seen in two subjects who received treatment under LA (subjects 46 and 107). Overall, three patients receiving only LA were more dentally anxious in the follow-up study, with two showing slight decreases in dental anxiety. For general anaesthesia (GA), four subjects had increased, and four had decreases in dental anxiety. The average decreases in dental anxiety following GA were greater than those of subjects in the other three modality categories. Both subjects experiencing failed treatment under LA and then progressing to GA showed small increases in dental anxiety.

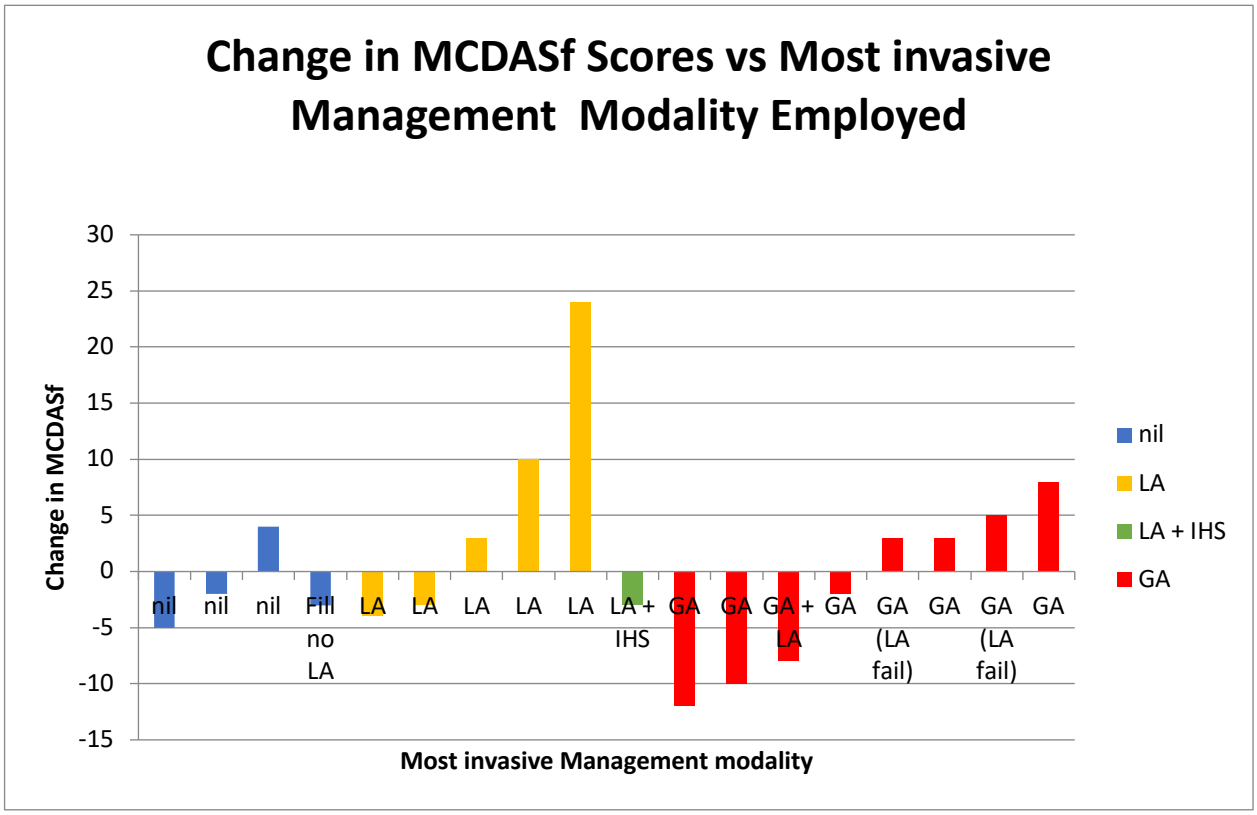



**Figure3. 14** Change in MCDASf Scores vs Most Invasive Management Modality Employed.

### 3.16.4 Baseline MCDASf scores and most invasive patient management modality subsequently employed

The relationship between Baseline MCDASf and the most invasive pain/anxiety management modality employed is shown graphically in Figure 3.15. No trend suggesting that baseline MCDASf might be related to the management modality employed was apparent. It is worthy of note that the subjects who received LA plus IHS had the highest baseline MCDASf score.

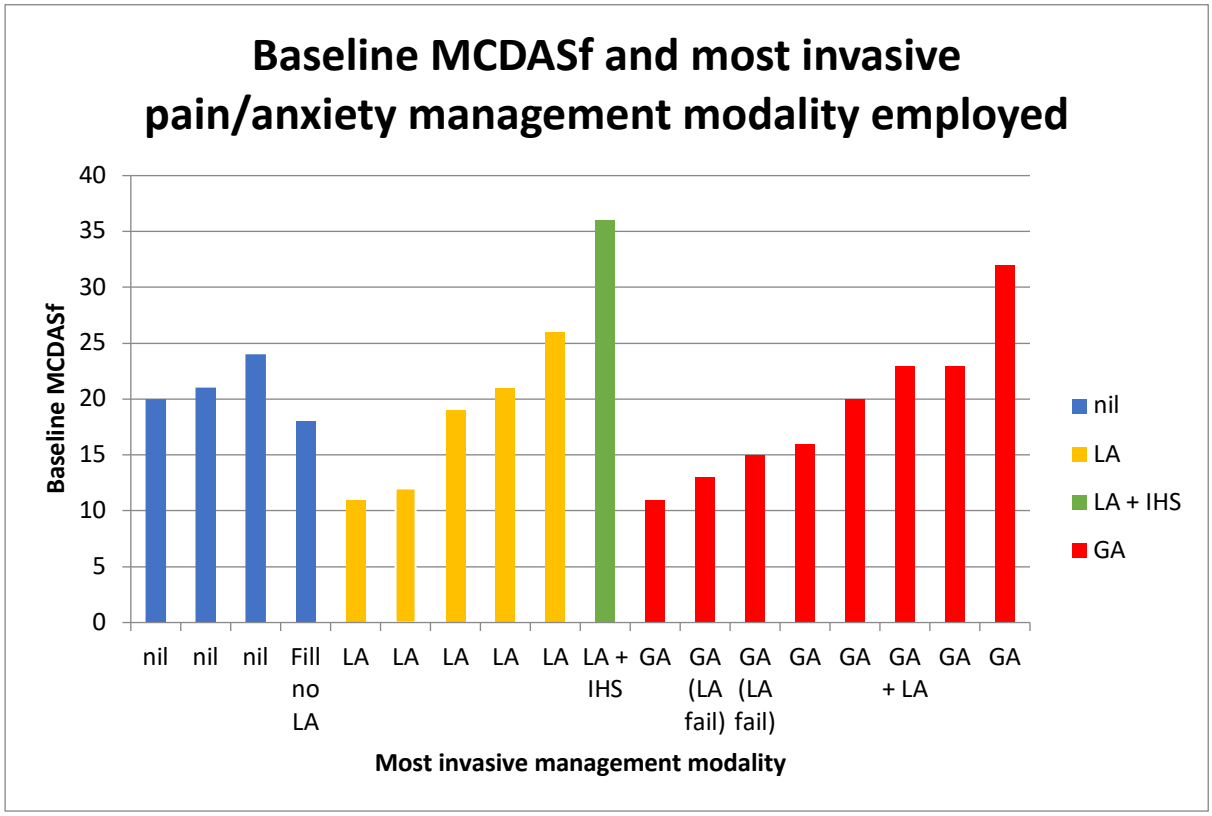



**Figure3. 15** Baseline MCDASf and most invasive pain/anxiety management modality employed..

## 3.17 The relationship between the type of dental treatment delivered and COHIP SF19, MCDASf and FIS

Patients were sorted according to subgroups based on the most invasive dental treatment delivered during their care at the hospital.
Changes in total COHIP SF19 and changes in MCDASf and FIS scores were then compared graphically to identify if any possible trends were apparent. In addition, the baseline total COHIP-SF19 and MCDASf were investigated for any association between these baseline scores and the treatment modalities subsequently employed.

### 3.17.1 Change in COHIP-SF19 total score and most invasive dental treatment provided

The change in COHIP-SF19 total score versus the most invasive dental treatment provided is shown in Figure 3.16. Subject 2 (prevention only) had a 23-point drop in COHIP total, with the drop spread fairly evenly across all

three domains. Subject 60 (fill LA) had a 23-point decrease in COHIP total with a 16-point drop in the SC domain.

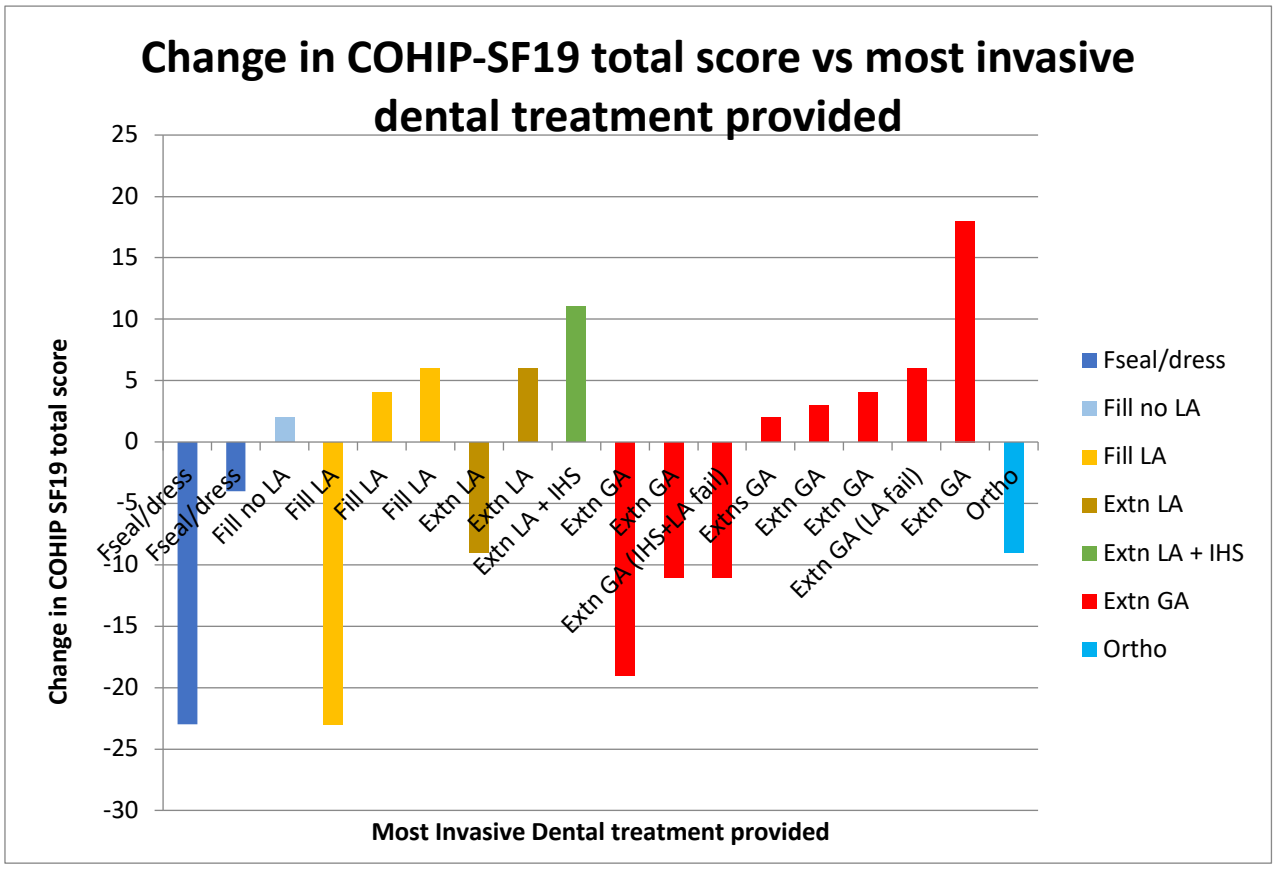



**Figure3. 16** Difference in COHIP-SF19 total score and most invasive dental treatment provided.

### 3.17.2 Change in MCDASf total score and most invasive dental treatment provided

Changes in MCDASf scores related to various dental treatments are shown in Figure 3.17. Extraction under L.A. showed the largest increases in MCDASf scores. Conversely, extractions under GA were associated with the three largest declines in MCDASf score (i.e. reduction in dentally-related anxiety). Filling under L.A. showed much smaller changes, with 2 out of 3 decreasing (i.e. less dentally anxious). IHS and LA patients had the highest baseline but did show a slight decrease in the Current study.

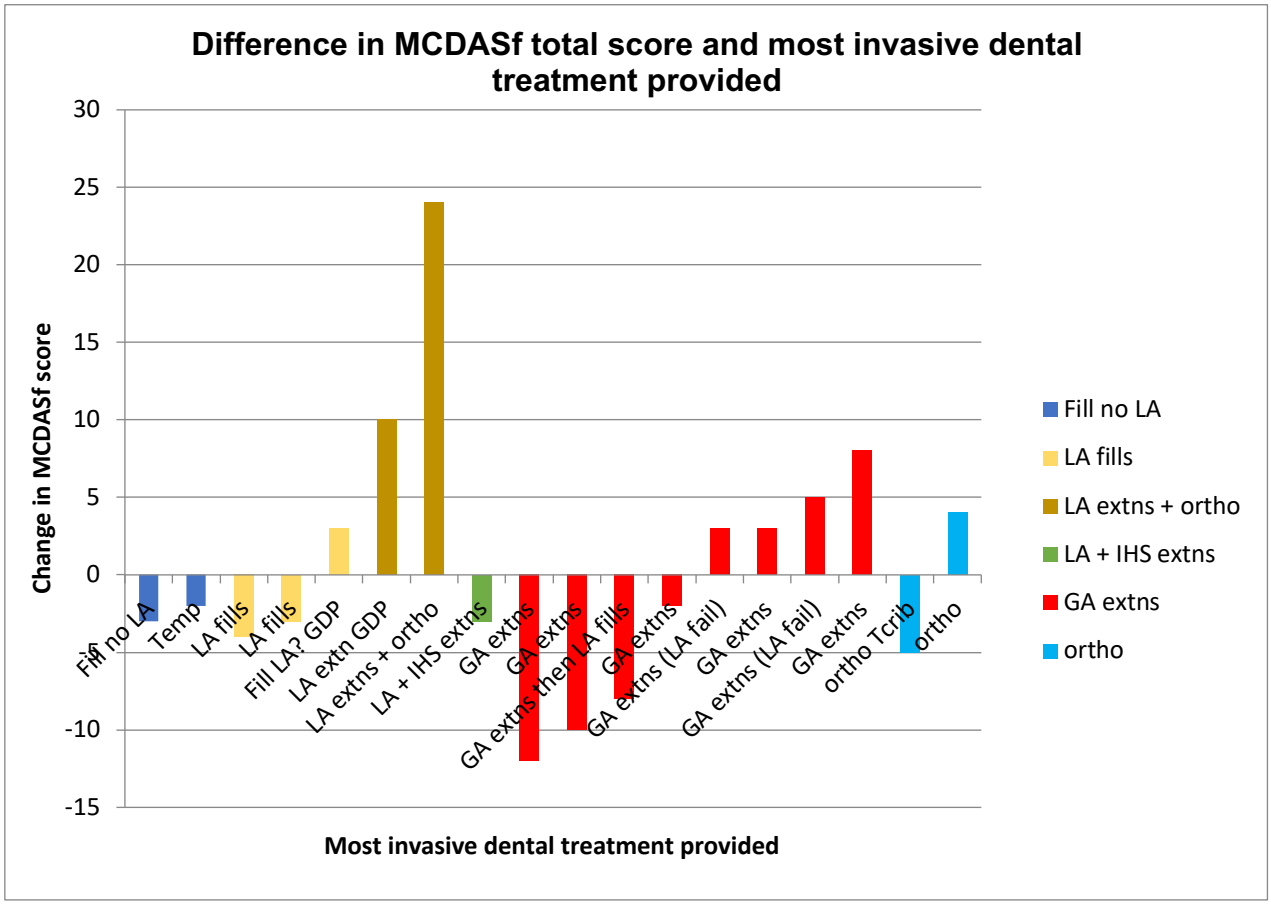



**Figure3. 17** Difference in MCDASf score and most invasive dental treatment provided.

## 3.18 Association between the total number of clinical visits and the changes in COHIP-SF19 total and MCDASf scores

The total number of clinical visits versus changes in COHIP SF19 and MCDAS scores is shown in Figure 3.20. No obvious relationship between changes in COHIP SF19 and the total number of clinical visits is apparent. However, Figure 3.19 suggests there might be a weak tendency for increased dental anxiety as visit numbers increase, although the trend is skewed by two relative outliers.

**Table3. 16** Difference in COHIP-SF19 total scores and the total number of clinical visits ranked by ascending number of visits.

| Participant ID | Total number of clinical visits | Change in COHIP-SF19 total scores | Change in MCDASf scores |
|---|---|---|---|
| 60 | 2 | -23 | 3 |
| 43 | 3 | 4 | -2 |
| 99 | 3 | -11 | 3 |
| 100 | 3 | -19 | 8 |
| 51 | 4 | -9 | -5 |
| 97 | 4 | 2 | -12 |
| 108 | 4 | 18 | -10 |
| 30 | 5 | -4 | -2 |
| 65 | 5 | 6 | 3 |
| 66 | 5 | 4 | -3 |
| 12 | 7 | 2 | -3 |
| 49 | 7 | -11 | 5 |
| 11 | 10 | 11 | -3 |
| 28 | 10 | 6 | -4 |
| 107 | 26 | -9 | 10 |
| 2 | 32 | -23 | 4 |
| 63 | 32 | 3 | -8 |
| 46 | 42 | 6 | 24 |

Note: An increase in COHIP SF19 indicates an improvement in OHRQoL
An increase in MCDASf indicates an increase in DFA.

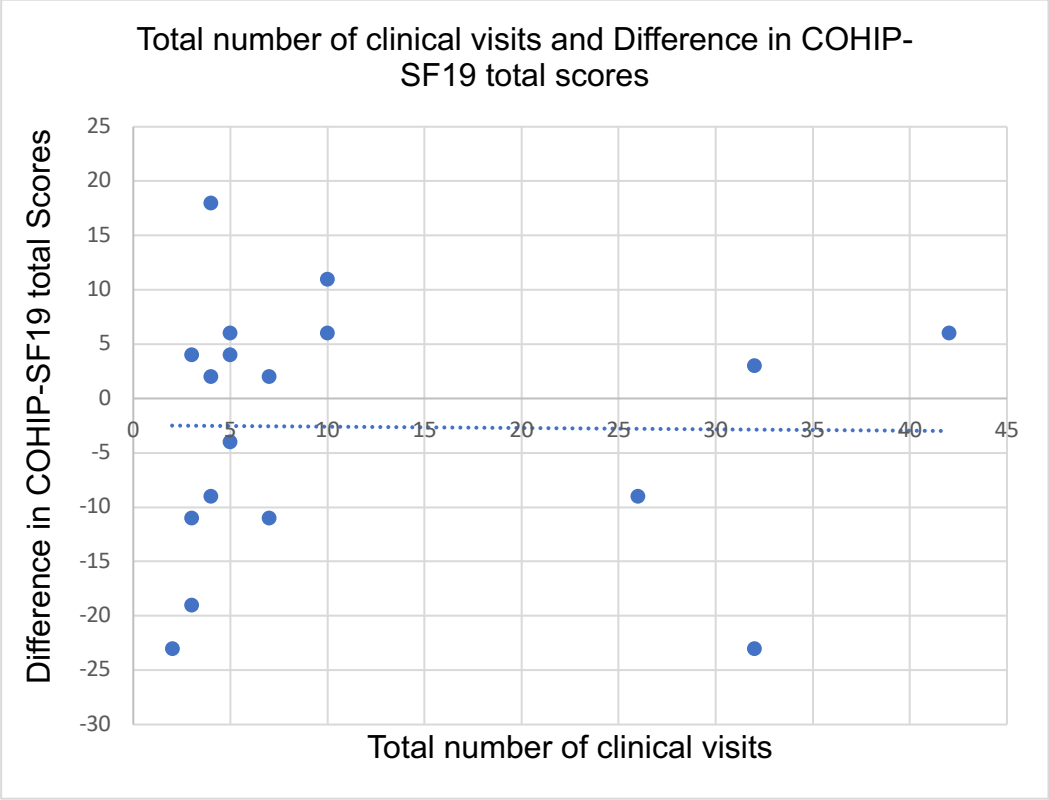



**Figure3. 18** Change in COHIP-SF19 total scores and the total number of clinical visits.

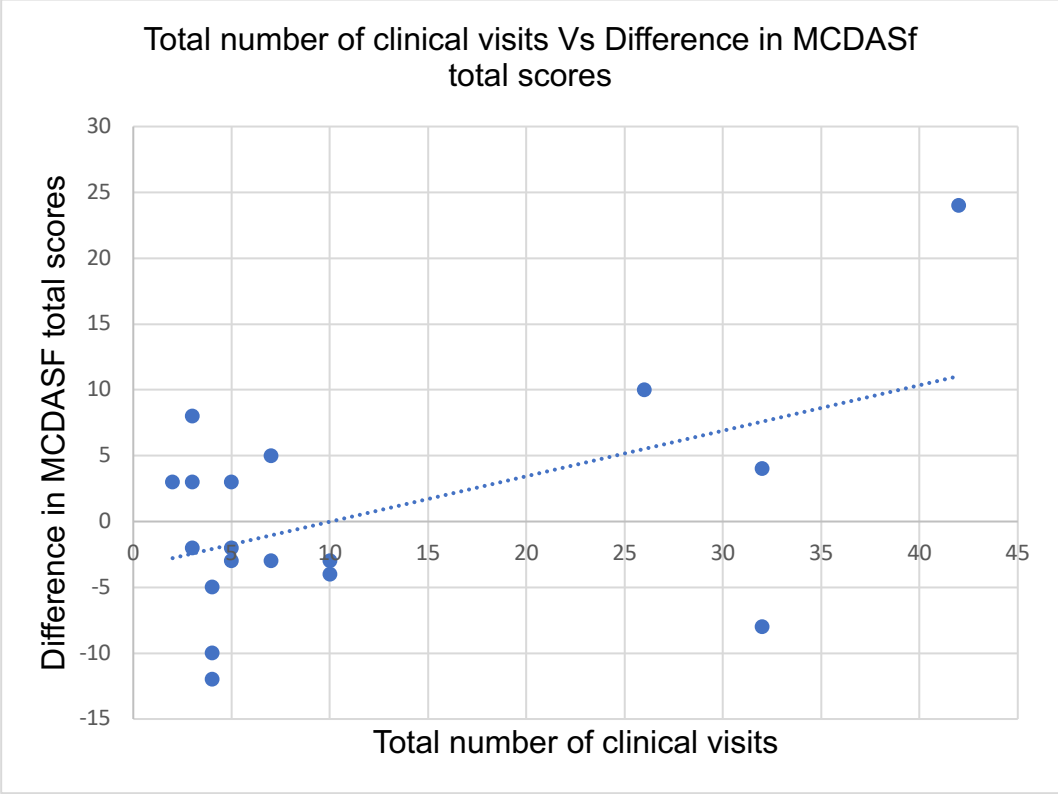



**Figure3. 19** Change in MCDASf total scores and the total number of clinical visits.

## 3.19 Relationship between FIS total score and the total number of clinical visits

When the FIS total score was compared to the total number of clinical visits, no clear relationship was apparent.

**Table3. 17** Difference in FIS total scores and the total number of clinical visits.

| Participant ID | Total number of clinical visits | FIS total scores |
|---|---|---|
| 60 | 2 | 1 |
| 43 | 3 | 5 |
| 99 | 3 | 16 |
| 100 | 3 | 16 |
| 51 | 4 | 3 |
| 97 | 4 | 7 |
| 108 | 4 | 7 |
| 30 | 5 | 2 |
| 65 | 5 | 2 |
| 66 | 5 | 4 |
| 12 | 7 | 3 |
| 49 | 7 | 1 |
| 11 | 10 | 25 |
| 28 | 10 | 0 |
| 107 | 26 | 3 |
| 2 | 32 | 1 |
| 63 | 32 | 3 |
| 46 | 42 | 9 |

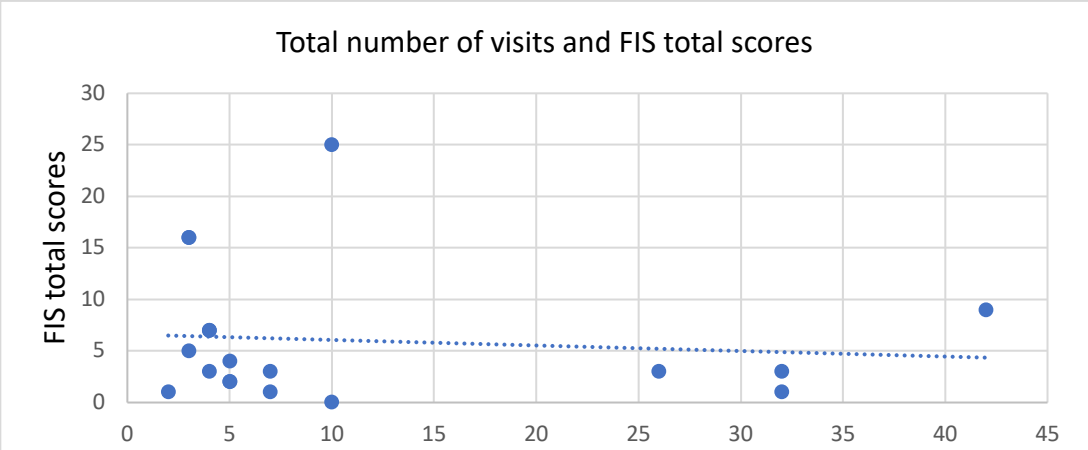



**Figure3. 20** Difference FIS total scores and the total number of clinical visits.

## 3.20 Duration of journeys for families to access the LDI

The shortest reported journey time amongst the participants was 18 minutes, while the longest reported travel time was 90 minutes (Table 3.18). The average reported travel time was 42.66 minutes. The travel time captured was for a one-way journey.

### 3.20.1 Relationship between the difference in COHIP-SF19 total score and the duration of the journey to LDI

When the time of the journey to LDI ranked from the least time to the most in minutes and compared to the difference in the COHIP-SF19 total scores, no direct relationship was apparent (Table 3.18).

**Table3. 18** Difference in COHIP-SF19 total scores and the journey time to LDI ranked from the least time to the most in minutes.

| Participant ID | Total time for the Journey to LDI | Difference in COHIP-SF19 total score |
|---|---|---|
| 28 | 18 | 6 |
| 51 | 20 | -9 |
| 46 | 20 | 6 |
| 30 | 20 | -4 |
| 43 | 25 | 4 |
| 2 | 25 | -23 |
| 66 | 30 | 4 |
| 60 | 30 | -23 |
| 108 | 35 | 18 |
| 107 | 40 | -9 |
| 49 | 45 | -11 |
| 12 | 45 | 2 |
| 100 | 60 | -19 |
| 99 | 60 | -11 |
| 97 | 60 | 2 |
| 65 | 60 | 6 |
| 63 | 60 | 3 |
| 11 | 90 | 11 |

Note: The travel time captured was for a one-way journey.

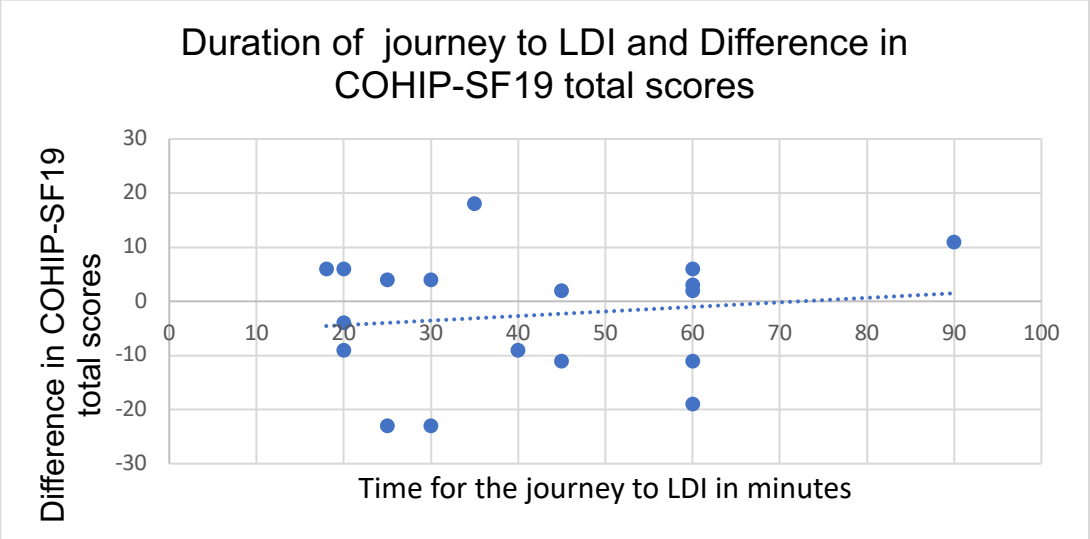



**Figure3. 21** Difference in COHIP-SF19 total scores and the journey time to LDI.

### 3.20.2 Relationship between the difference in MCDASf total score and the duration of the journey to LDI

When the time of the journey to LDI ranked from the least time to the most in minutes and compared to the difference in the MCDASf total scores, no direct relationship was apparent.

**Table3. 19** Difference in MCDASf total scores and the journey time to LDI ranked from the least journey time to the most in minutes.

| Participant ID | Total time for the Journey to LDI | Difference in MCDASf score |
|---|---|---|
| 28 | 18 | -4 |
| 51 | 20 | -5 |
| 46 | 20 | 24 |
| 30 | 20 | -2 |
| 43 | 25 | -2 |
| 2 | 25 | 4 |
| 66 | 30 | -3 |
| 60 | 30 | 3 |
| 108 | 35 | -10 |
| 107 | 40 | 10 |
| 49 | 45 | 5 |
| 12 | 45 | -3 |
| 100 | 60 | 8 |
| 99 | 60 | 3 |

| | | |
|---|---|---|
| 97 | 60 | -12 |
| 65 | 60 | 3 |
| 63 | 60 | -8 |
| 11 | 90 | -3 |

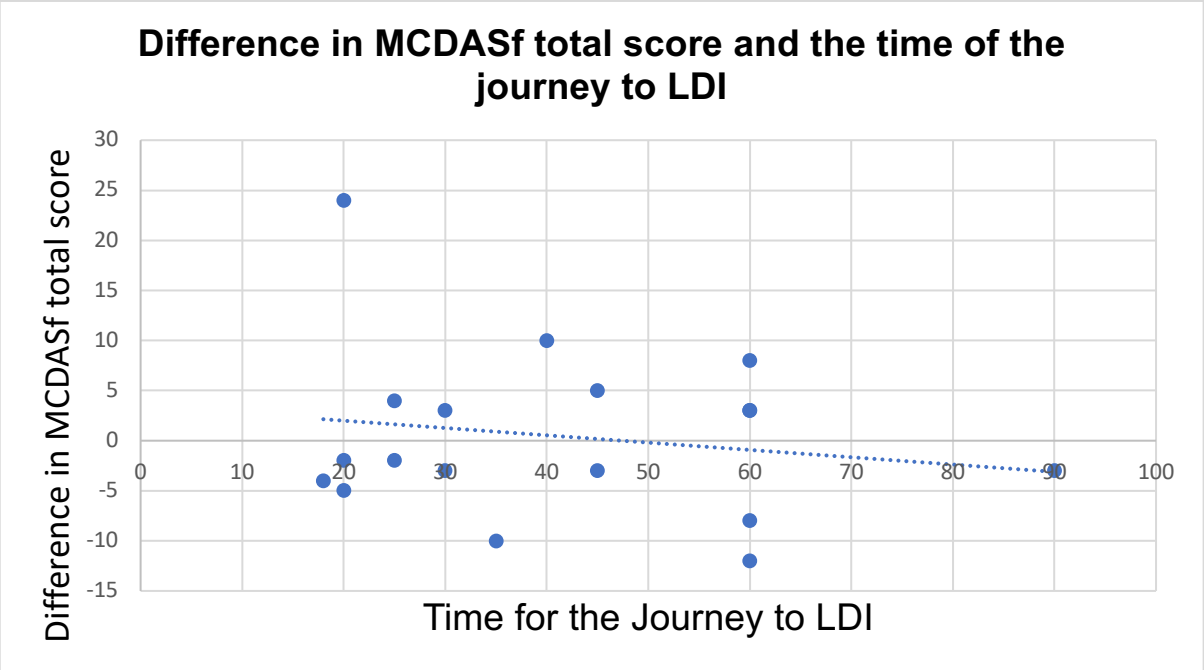



**Figure3. 22** Difference in MCDASf total scores and the time of the journey to LDI.

### 3.20.3 Relationship between the difference in FIS total score and the duration of the journey to LDI

Journey times to the hospital clinic and Family Impact Scale total and individual domain scores are shown in Table 3.26. When the time of the journey to LDI ranked from the least time to the most in minutes and compared to the difference in the FIS total scores, a possible weak relationship emerged, with a tendency for longer journey times to be associated with higher total FIS scores (Figure 3.23). The three subjects with the highest FIS scores also had the longest journey times. A similar association is apparent for the parental activities domain (PA), parental emotions domain (PE) and the family conflicts domain (FC) of the FIS (Figures 3.24, 3.25 and 3.26), showing that all 3 of these domains made a contribution to the trend.

**Table3. 20** The difference in FIS total and individual domain scores and the journey time to LDI ranked from the least journey time to the most in minutes

| Participant ID | Total journey time (mins) | FIS total score | FIS Parental Activities | FIS Parental Emotions | FIS Family Conflict | FIS Financial Burden |
|---|---|---|---|---|---|---|
| 28 | 18 | 0 | 0 | 0 | 0 | 0 |
| 51 | 20 | 3 | 3 | 0 | 0 | 0 |
| 46 | 20 | 9 | 5 | 4 | 0 | 0 |
| 30 | 20 | 2 | 0 | 0 | 2 | 0 |
| 43 | 25 | 5 | 4 | 1 | 0 | 0 |
| 2 | 25 | 1 | 1 | 0 | 0 | 1 |
| 66 | 30 | 4 | 3 | 1 | 0 | 0 |
| 60 | 30 | 1 | 1 | 0 | 0 | 0 |
| 108 | 35 | 7 | 5 | 2 | 0 | 0 |
| 107 | 40 | 3 | 2 | 1 | 0 | 0 |
| 49 | 45 | 1 | 1 | 0 | 0 | 0 |
| 12 | 45 | 3 | 0 | 3 | 0 | 0 |
| 100 | 60 | 16 | 3 | 6 | 7 | 0 |
| 99 | 60 | 16 | 6 | 5 | 5 | 0 |
| 97 | 60 | 7 | 3 | 4 | 0 | 0 |
| 65 | 60 | 2 | 2 | 0 | 0 | 0 |
| 63 | 60 | 3 | 1 | 0 | 2 | 0 |
| 11 | 90 | 25 | 10 | 10 | 5 | 0 |

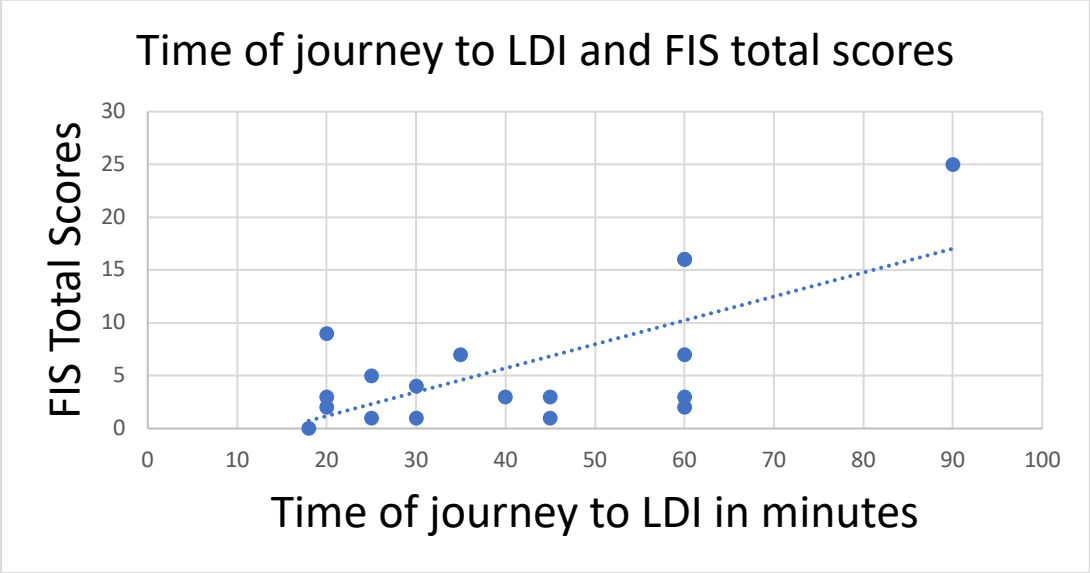



**Figure3. 23** Difference in FIS total scores and the time of the journey to LDI.

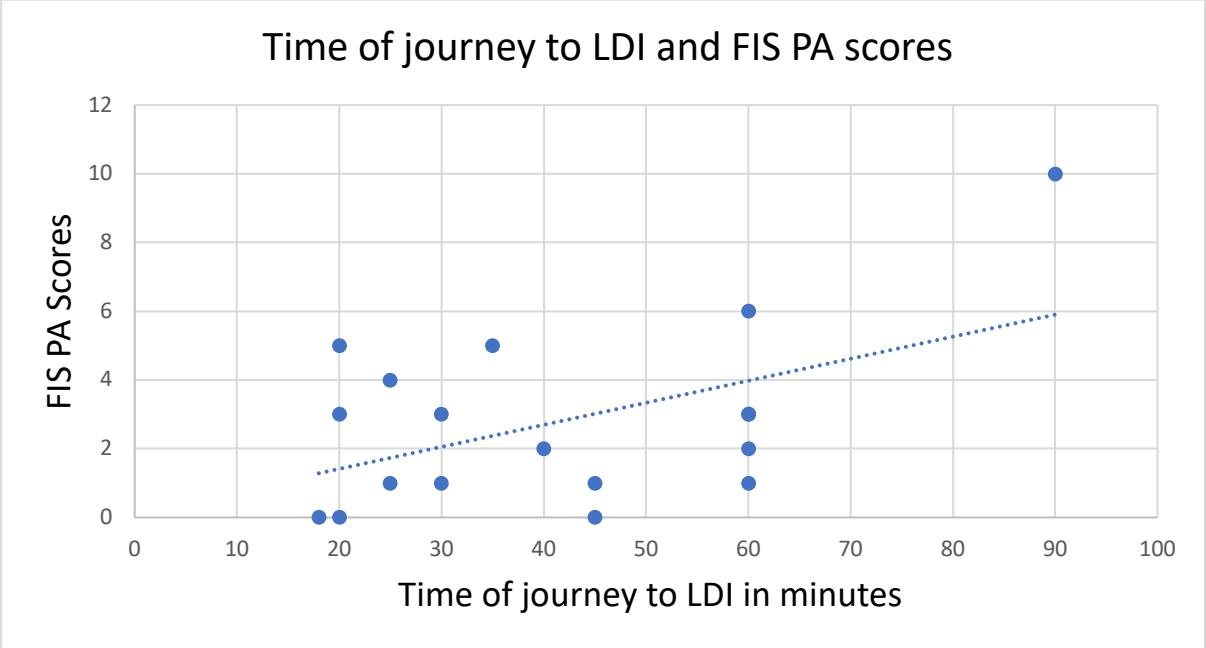



**Figure3. 24** Difference in FIS: Parental Activities domain scores and the time of the journey to LDI.

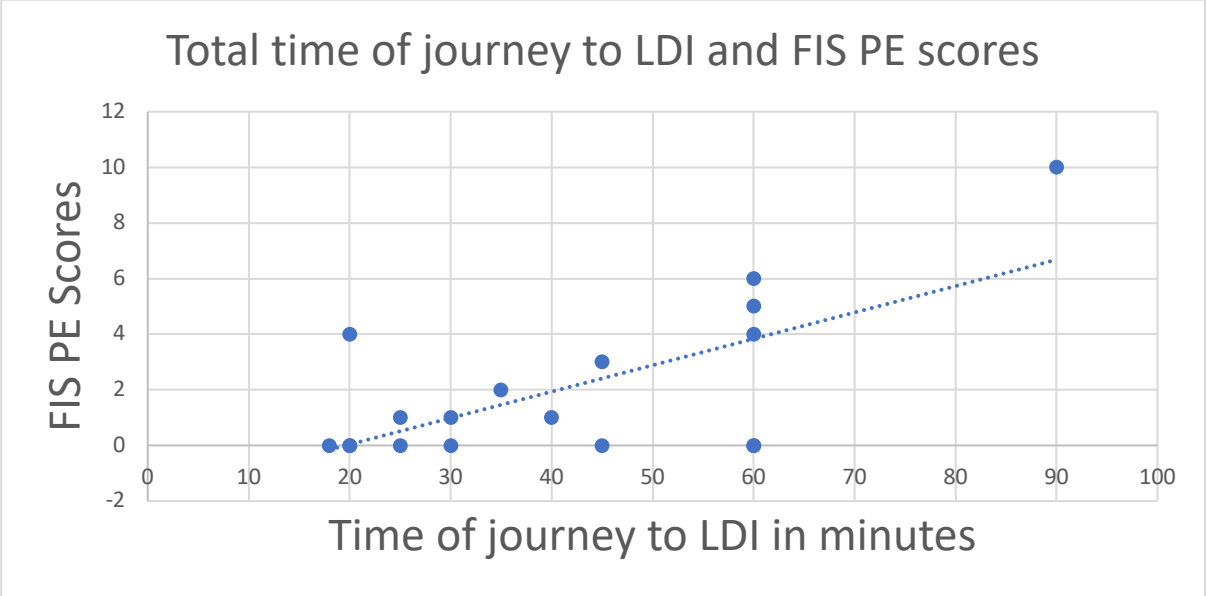



**Figure3. 25** Difference in FIS: Parental Emotion domain scores and the time of the journey to LDI .

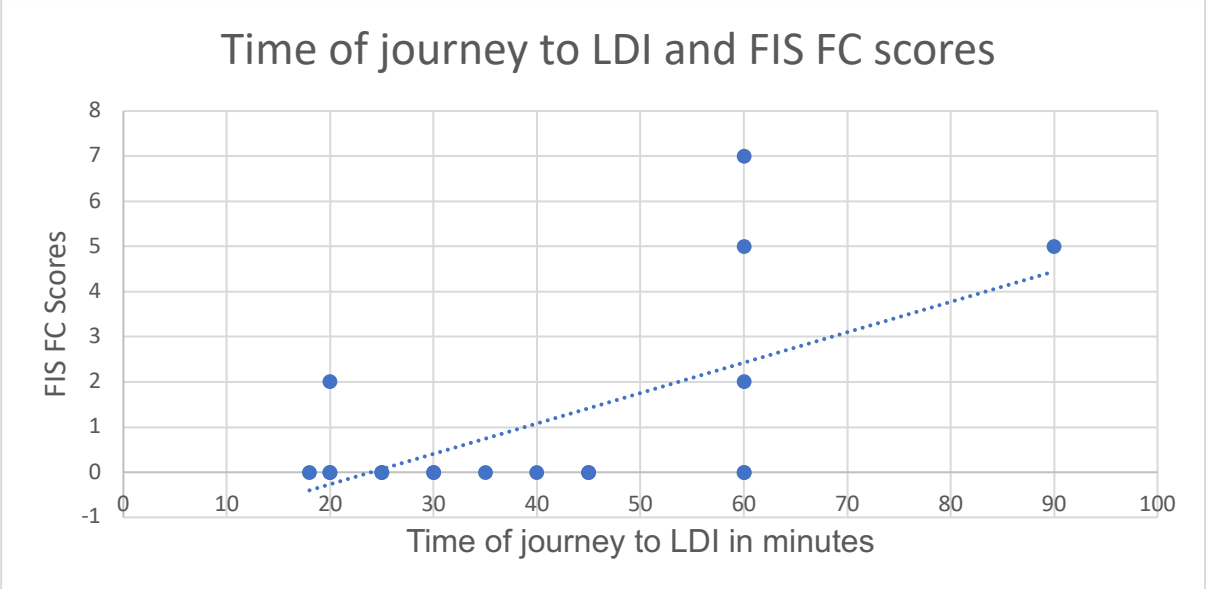



**Figure3. 26** Difference in FIS: Family Conflict domain scores and the time of the journey to LDI.

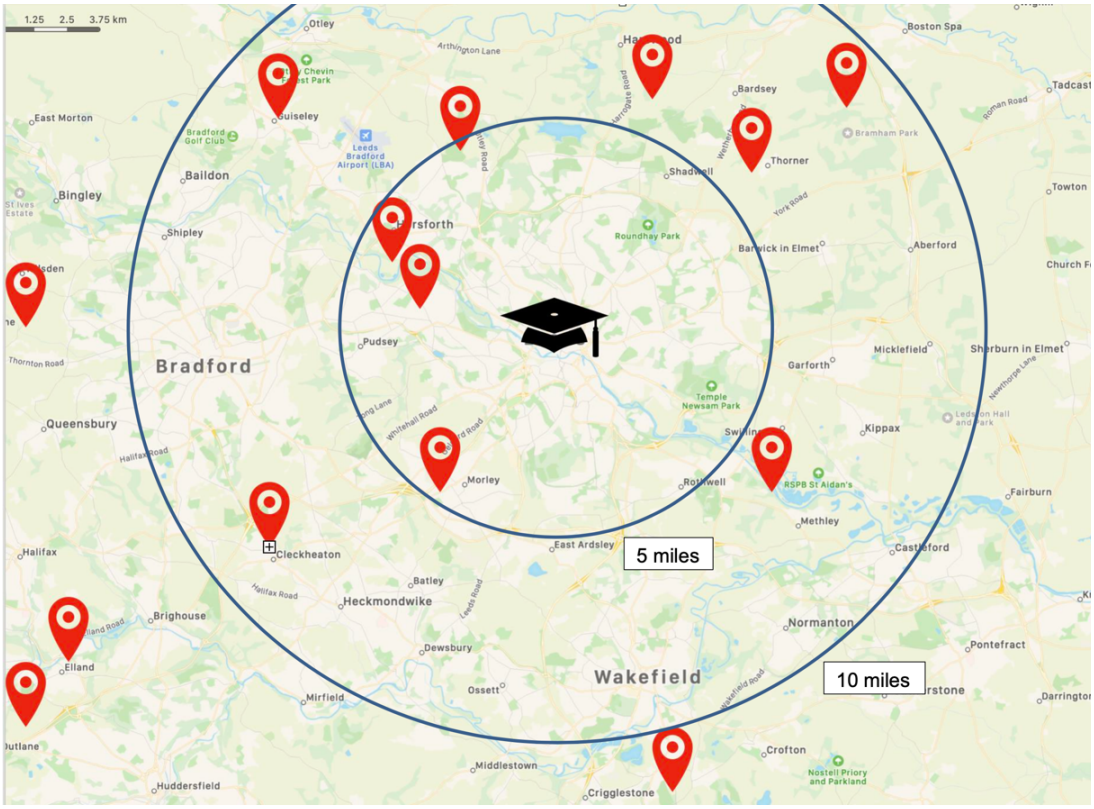



**Figure3. 27** Map illustrating the approximate home location of the participants in relation to the Dental Hospital.

## 3.21 Cost of transportation to LDI

The reported cost of travel in one direction ranged from £0 for one participant to £10-£20 (3 participants). The £5-10 option was the most common estimate, chosen by 10 participants. No participants chose the £20-30, £30-40, £40-50, or more than £50 options. Interestingly, the only family indicating a Financial Burden in the FIS questionnaire was the family that reported zero transportation costs. Figure 3.28 shows FIS total scores plotted against the cost of the journey. No obvious trend is apparent.

**Table3. 21** Cost of transportation to LDI

| Participant ID | Cost of transportation |
|---|---|
| 2 | 0 |
| 11 | £5-10 |
| 12 | £5-10 |
| 28 | £10-20 |

| | |
|---|---|
| 30 | £1-5 |
| 43 | £1-5 |
| 46 | £5-10 |
| 49 | £5-10 |
| 51 | £1-5 |
| 60 | £10-20 |
| 63 | £1-5 |
| 65 | £5-10 |
| 66 | £5-10 |
| 97 | £5-10 |
| 99 | £5-10 |
| 100 | £5-10 |
| 107 | £5-10 |

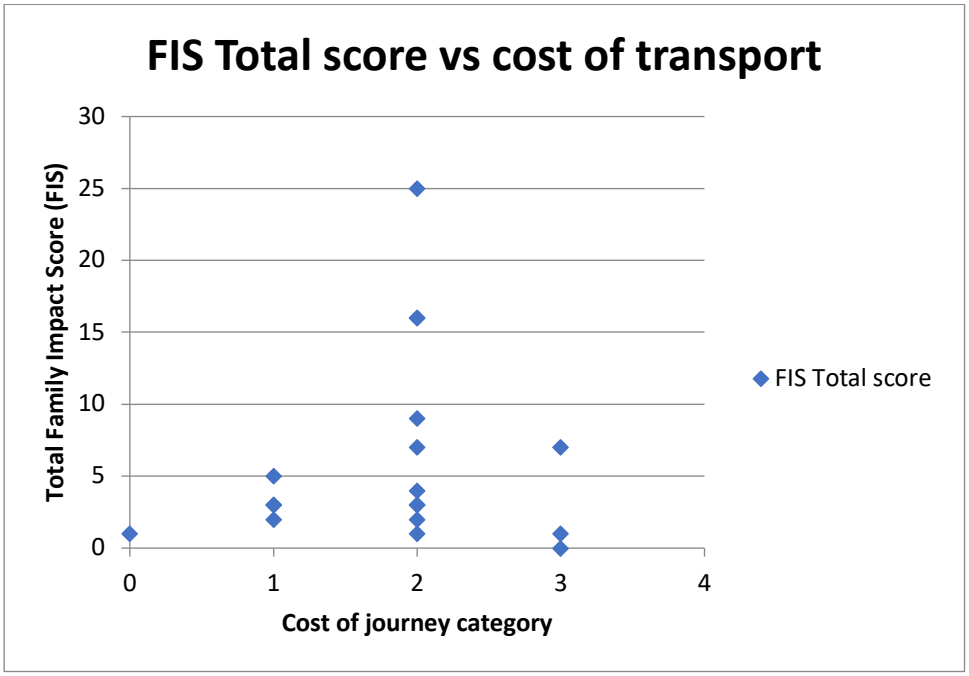



**Figure3. 28** Family Impact total score vs cost of transport.

## 3.22 Mode of transportation to LDI

When participants' families were asked to choose from a mode of transportation, the majority of families (14) travelled by car. Of those using public transport only two used buses and two travelled by train.

**Table3. 22** Mode of transportation

| Participant ID | Mode of transportation |
|---|---|
| 2 | Car |
| 11 | walking/train |
| 12 | Car |
| 28 | Bus + Taxi |
| 30 | Car |
| 43 | car |
| 46 | Car |
| 49 | Bus |
| 51 | Car |
| 60 | Car |
| 63 | Car |
| 65 | Car |
| 66 | Car |
| 97 | Car |
| 99 | Car |
| 100 | Car |
| 107 | Train |
| 108 | Car |

## 3.23 Description of the journey

The journey was found to be moderate when asked to be described by the families of 10 participants. Four families described the journey as very easy, and four described it as easy (Table 3.23). No one described the journey as being difficult or very difficult when they were asked to choose from the multiple answers. As might be expected, families with longer journey times tended to rate their journeys as more difficult (Figure 3.29)

**Table3. 23** Journey description

| Participant ID | Describe the journey |
|---|---|
| 2 | Very easy |
| 11 | Moderate |
| 12 | Easy |
| 28 | Easy |

| | |
|---|---|
| 30 | Very easy |
| 43 | Easy |
| 46 | Moderate |
| 49 | Moderate |
| 51 | Very easy |
| 60 | Moderate |
| 63 | Moderate |
| 65 | Moderate |
| 66 | Moderate |
| 97 | Moderate |
| 99 | Moderate |
| 100 | Moderate |
| 107 | Easy |
| 108 | Very easy |

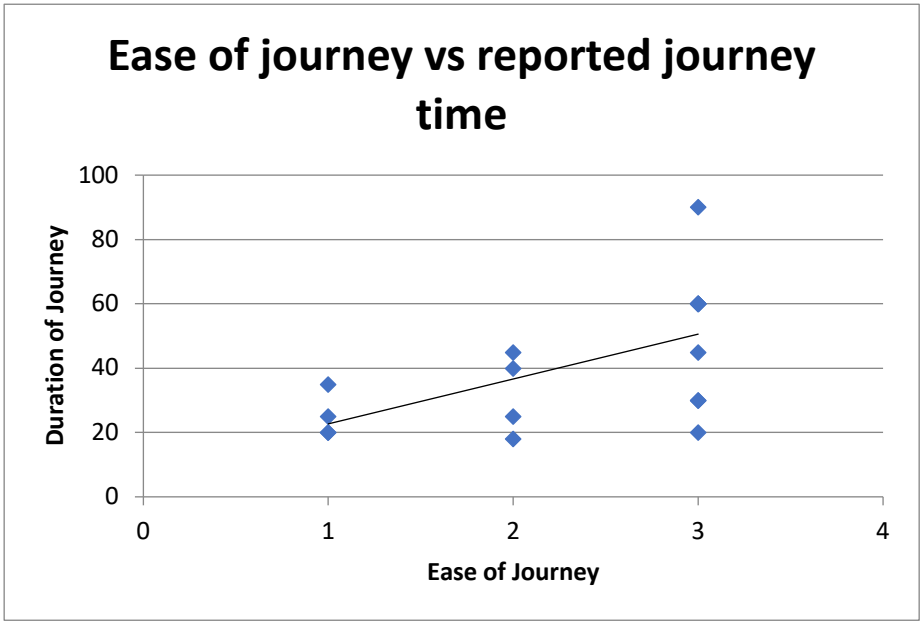



**Figure3. 29** Reported ease of journey versus reported journey time.

1 = 'very easy'
2 = 'easy'
3 = 'moderately difficult'
No families reported 'difficult' or 'very difficult'

## 3.24 Experience of care at LDI

Nine parents of participants reported that they were mostly satisfied with their experience at the LDI and eight reported that they were very satisfied. Only one reported that they were neither satisfied nor dissatisfied with the experience of visiting LDI and none reported being ‘dissatisfied’/’very dissatisfied’

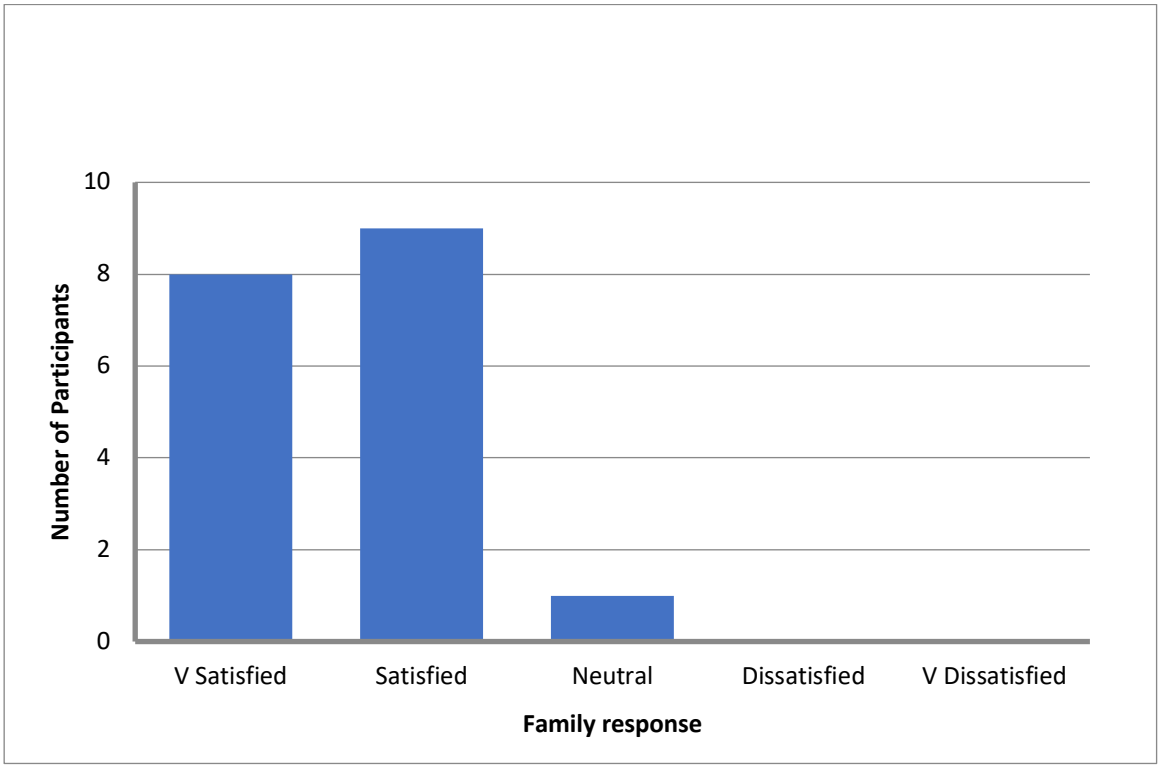



Figure3. 30 Reported satisfaction of families with care provided at LDI.

## 3.25 Content Analysis

Seven codes were derived from the questionnaire answers and assigned within NVivo for analysis. Codes were created by the chief investigator and an experienced supervisor researcher (Kate Kenny) by looking for keywords repeated in the answers of the participants and the participants’ parents. They were:

- Explanation
- Information
- Friendly
- Nice
- Appointments
- Waiting times

- Treatment

There were two main themes under which the answers for most of the participants and their parents' responses fell:

- Communication
- Challenges

The codes that were assigned were related to one of the two main themes:

| **Communication** | **Challenges** |
|---|---|
| Explanation | Waiting times |
| Information | Appointments |
| Friendly | |
| Nice | |
| Treatment | |

The following verbatim quotes were extracted from the participants and the participants' parents answers to the provided questions. The quotes indicate the comments that allowed the codes to be assigned.

**Explanation**

*"Communicating with … and explaining procedure"*
Participant's parent 012

*"Everything was explained in detail to myself and my child"*
Participant's parent 028

*"Great explanation of what was going on. Made ….. feel really comfortable"*
Participant's parent 063

*"Explained procedures in an appropriate way"*
Participant's parent 065

*"Explaining the condition and treatment"*
Participant's parent 100

**Information**

*"Provided detailed information"*
Participant 002 parent

*"Being informed about the treatments"*
Participant 100

**Friendly**

*"I got a goody bag at the end and the staff were friendly"*
Participant 060

*"Treated child & parent with respect in a friendly manner"*
Participant 065 Parent

*"Staff were friendly"*
Participant 108

*"Friendly staff"*
Participant and participant parent 093

**Nice**

*"My dentist was really nice"*
Participant 028

*"Very Nice nurses + doctors"*

Participant 053

*"Staff were nice and made me feel slightly less nervous"*

Participant 100

**Waiting times**

*"Reduce waiting time as this increases anxiety"*
Participant's Parent 108

*"Waiting times could have been better, especially waiting for xrays"*
Participant's Parent 028

*" Waiting times have improved significantly since before covid"*
Participant 002

**Appointments**

*" I had to trace me follow up appointments. I contacted me the secretaries each time who sorted out appt for me"*
Participant's 012 Parent

*"Waited long time for appointments. Follow-up slow. Had to ring to chase appointments"*
Participant's Parent 030

*"The availability of appointments is quite limiting. I feel like treatment has taken longer due to this"*
Participant's Parent 063

*"I would like to know when the follow-up appointments are going to be as it is a long time since ….. Was at LDI"*

Participant's Parent 100

**Treatment**

*"There was part of the treatment that I didn't want so they have found a solution"*

Participant 063

*“Treated appropriately”*
Participant’s parent 066

*“Seen on time, staff polite, treatment good”*
Participant’s parent 107

# Chapter 4: Discussion

## 4.1 Re-recruitment Rate

The re-recruitment rate for this current study was 22%. There is no specific threshold for the response rate in questionnaire studies (Chapple, 2003). However, the response rate is well below the median response rate of 73.5% for Europe reported in a systematic review of 133 dental field articles in 2022 (Al Khalaf et al., 2022).

To improve the re-recruitment rate, two methods of sending and collecting the questionnaires were employed before conducting the follow-up study. A physical postal mail and an electronic email option were offered to the participants (Funkhouser et al., 2014). Nevertheless, the mixed mode approach in the delivery did not seem to improve the response rate as the postal mail was used for 17 participants out of the total 18, and only one participant used the email method for the questionnaire delivery only while filling them physically.

A face-to-face interview and self-completed questionnaires filled in at the clinic visit was the initial method planned for collecting the data from the participants, as this was how the baseline questionnaires were administered in the previous Baseline study (Al Bahar, 2017). This is a longstanding and effective method that dates back to 1912 (De Leeuw, D., 2005). Later, De Leeuw (2005) used face-to-face interviews and questionnaires combined with mail surveys to form the first mixed mode of collecting data in scientific research. Unfortunately, the global pandemic necessitated a modification of the research methodology into a remote approach. Funkhouser in 2014 suggested that response rate is not the only indicator of the survey quality as factors including data quality, timelines and cost are other indicators to be considered (Funkhouser et al., 2014). The process of sending the invitation letters and the questionnaire packages to the potential participants was done in batches from December 2022 to May 2023 to monitor the response rate and adjust accordingly.

Coincidently, the period of postal questionnaire distribution and return had multiple postal service interruptions due to Royal Mail workers' strike action. The strikes started in May 2022 and ended in July 2023 after the union members reached an agreement with the Royal Mail (Wikipedia, 2023). These interruptions required posts to be sent at times most likely to result in successful timely delivery and return, which added to the increased period of conducting the research.

In addition, families might have been less likely to respond positively to the postal questionnaire compared to direct hospital appointments for review, which may have also contributed to the low re-recruitment rate. The long period of time since patients have been discharged, combined with the children/adolescents not having any further problems following the treatment, may have also led to parents being less keen on responding to further contact with LDI. The years that had elapsed since the discharge of the patients may also have resulted in many changing their addresses and mobile telephone numbers, which might also have had an impact.

Hurwitz et al. emphasised the role of a participant-centred approach in re-recruitment, including collaboration with schools, direct outreach and maintaining rapport through regular updates (Hurwitz et al., 2017).

## 4.2 Participant demographics

### 4.2.1 Gender

13 male and five female subjects agreed to take part in this follow-up study. The previous baseline study group included 42 female and 40 male MIH participants (Al Bahar, 2017), so female participants were relatively underrepresented in the present study sub-group. Gender has not been demonstrated to significantly influence the risk of MIH (M. Koruyucu et al., 2018). However, females are 1.5 times more likely to take part in a survey than males (Smith, W.G. 2008.). Whilst the number of male participants agreeing to take part in the current study is more than double the number of female participants, which seems to be unexpected, one possible factor could have been that many of the actual primary responders were the

mothers of the participants who took part, which may have negated any gender-bias.

### 4.2.2 Ethnicity

The most common ethnic group participants identified as White-British ethnicity (12 participants), followed by five white non-specified ethnicities and one participant of mixed White-British and Arab ethnicity. The previous baseline study group (Al Bahar, 2017) was also predominately White-British or another White background (82.9%), so it is not surprising that most of those agreeing to re-participate in this study were of White background. Studies have reported conflicting results on the role of ethnicity in the prevalence of MIH. Balmer et al. suggested MIH may have a variation in different ethnic groups, while Zagdwon et al. found that the difference between White-Caucasian and Asian-Caucasian was not significantly different (Zagdwon et al., 2002).

### 4.2.3 Age

The age of the participants at the time of completing the questionnaires in this follow-up study ranged from 14 to 17 years, with a mean age of 15.6 years. For the same cohort of participants, the mean age was 8.05 years at the time of recruitment to the original baseline study (when the baseline questionnaires were completed). The relatively narrow age range in this follow-up study reflected the narrow age range in the original study, which had Children's ages ranging from 6.5 to 12.8.

The 15 years-of-age group had the largest number of participants, with 8 participants. Half the participants were over 16 years old when they received their questionnaire packages, which allowed them to sign their own consents, while the other half signed assents as they were below the age of 16 years. The MCDASf is a reliable and valid measurement tool for measuring dental anxiety for children, primarily aged 8-12 (Howard and Freeman, 2007). The MCDASf was an ideal tool for the original study due to its suitability for the age of the participants. However, the age of the participants in this follow-up study is beyond that age range, which is a

possible criticism. However, MCDASf was developed from tools with a greater upper age limit, and it was decided to use the same tools to measure DFA and OHRQoL as previously so results from both studies could be directly compared. In contrast, COHIP-SF19 covers a wider age group than MCDASf, as it has been validated to be used on children aged 7-17 years (Broder et al., 2012).

## 4.3 MIH Severity

Table 3.1 shows that the majority of participants in this follow-up study had severe MIH at the time of diagnosis (11 participants), followed by those with moderate severity (4 participants) and 3 participants having mild severity. The re-recruited subgroup, therefore, included a reasonable number of subjects with all three of the different severities of MIH from the previous study, albeit a somewhat skewed distribution. In the previous study, out of the 82 children, 72 (87.8%) had severe MIH, 5 (6.0%) had moderate, and 5 (6.0%) had mild MIH (Al Bahar, 2017).

## 4.4 Mode of pain/patient management for treatment and dental treatment received

Table 3.2 and Table 3.3 show the modalities of pain/patient management and treatment received, respectively. L.A. was used for nine participants, followed by G.A. for eight, and IHS for two participants combined with other modes. Two participants had failed attempts at extraction with L.A. before performing the needed first permanent molar extraction under G.A. Treatment received according to the retrieved dental records for the 18 re-recruited participants had extractions performed on 11 participants, restorations followed on seven, three had fissure sealing, and two had temporary restorations.

Jälevik and Klingberg found that children with MIH require comprehensive treatment approaches, including early intervention and effective pain management. Their study concluded that children with MIH had almost 10 times more dental treatments for their FPMs compared to children attending for problems other than MIH. It is worth noting that, at the time of their initial referral, their sample of children with MIH frequently had a history of

treatment with inadequate pain management, as most of the treatment was done without L.A (Jälevik, and Klingberg, 2012).

In the current study, all the dental treatments received by the participants at the specialist centre were within the recommendations for MIH management that have been reported in numerous scientific research articles (William et al., 2006; Elhennawy et al., 2016; Somani et al., 2022). The severity of the MIH condition was a determining factor in the type of dental treatment received by the participants, with FPM extractions being frequent in those with severe MIH and relatively few in moderate. This is in accordance with the updated European Academy of Paediatric Dentistry policy document of best clinical practice guidance for clinicians dealing with children presenting with molar-incisor-hypomineralisation (Lygidakis et al., 2021). The dental treatment received at LDI for this follow-up study further underlines that MIH severity is an important factor in planning and performing the treatment.

## 4.5 Number of clinical visits

This follow-up study focused on and captured specialist-level care. In this current study, the average number of visits to the Paediatric Department at LDI (including treatment under G.A. in ODU) for managing MIH-affected children was 4.94 visits. (Table 3.4) with the minimum being two visits and the maximum 10 visits. Previous studies have reported an increased number of clinical visits for MIH patients when compared to non-MIH patients. Kotsanos et al. reported that the frequency of restorative intervention was higher in patients with MIH, and, in addition, their need for retreatment was greater (Kotsanos et al., 2005). Folyan et al. came to similar conclusions in regard to the increased time spent to diagnose and provide treatment for children with MIH (Folyan et al., 2018).

In this current follow-up study, the number of clinical visits inflated considerably when the participants also had an active orthodontic intervention. The total number of orthodontic/paediatric clinical visits for this sub-group ranged from 23 to 40 visits with a mean of 28.75. In 2016, Tsichlaki et al. reported a mean of 17.81 in their systematic review derived

from 5 studies of non-MIH orthodontic treatment with fixed appliances (Tsichlaki et al., 2016). In another study conducted at LDI in 2013, traumatic dental injuries (TDI) resulted in a median total number of clinical visits per tooth reaching six visits with a range of 1-22 visits. The involvement of severe hard tissue injury, complicated periodontal injury and non-vital tooth contributed to the increase in the number of clinical visits (Keasberry et al., 2013).

## 4.6 Oral Health-Related Quality of Life (OHRQoL)

There is a lack of consensus about the impact of MIH on OHRQoL in affected children. Some studies have concluded that MIH may have a significant effect, whilst others have failed to demonstrate any definite impact. For example, in a cross-sectional study conducted by Dantas-Neta et al. (2016), who classified the MIH similar to this follow-up study into severe, moderate and mild, found a higher negative impact on OHRQoL in schoolchildren with severe MIH than those without MIH In contrast, in a similar cross-sectional study, Dias et al. (2021) concluded no significant negative impacts on OHQoL for MIH-affected children.

The original baseline study (Hussa 2017) did suggest that, at the time of initial attendance at the specialist clinic, children with MIH had significantly lower OHRQoL than children referred for management of caries. The baseline COHIP-SF19 scores at initial diagnosis for the 18 participants in this follow-up study showed considerable variation but with no obvious trends between those with mild, moderate or severe MIH *(3.10.1, Fig 3.6.1)*. This may reflect the lack of previous treatment for MIH in the baseline study group.

When baseline (pre-treatment) and follow-up (post-treatment) COHIP SF19 total scores were compared, ten participants had an increase in total COHIP SF19 score when compared to their baseline total score (Tables 3.10 and 3.12), indicating an improvement in their OHRQoL whilst eight participants recorded decreased total scores (decline in their OHRQoL). However, some

interesting trends emerge when changes (from pre-treatment to post-treatment) in COHIP-SF19 scores are considered in more detail.

Firstly, all three subjects with mild MIH had a reduction in their total COHIP-SF19 scores, suggesting a decrease in OHRQoL. This is in marked contrast to subjects with moderate/severe disease, where more than half the subjects in each subgroup (3/4 with moderate MIH and 7/11 with severe MIH) had higher COHIP-SF19 scores after treatment, indicating improved OHRQoL *(3.10.1, Fig 3.6.1)*. Although numbers are small, so caution in interpreting results is needed, this does suggest that those subjects with moderate and severe MIH may have gleaned the most benefit from treatment, which in turn suggests their baseline condition may have been having a greater impact on them.

Furthermore, when changes in individual domain scores are reviewed, some other interesting variation is apparent. When considering the individual domains (3.10, Fig 3.6) this trend in difference between the severity groups is most marked in the OH and functional domains.

The ‘Functional well-being’ domain showed the greatest number of participants experiencing an increase in their scores, suggesting improvements in this aspect of the subjects’ OHRQoL. Table 3.8 shows that 12 participants had a higher COHIP-SF19 score, suggesting an improvement in OHRQoL, with 6 participants returning lower scores. All but two subjects with severe MIH showed an improvement in their COHIP-SF19 functional domain. This tends to suggest that OHRQoL may impacted negatively in those children with severe MIH and that this impact may be most significantly related to functional impairment. It also suggests that for the majority of those with severe or moderate MIH, treatments are effective in improving this aspect of OHRQoL.

These findings concur with those of a previous cross-sectional study of 594 children of a mean age of 12.45 years attending an integrated children’s dental clinic in a Brazilian city (Dias et al., 2021). Children with MIH were

compared with a control group of children without MIH. It was concluded that the greatest negative impact on OHRQoL for those children with severe MIH was due to oral symptoms and functional limitation. According to the parents'/caregivers' perception, the functional limitation of severe MIH patients reached 42% higher than those without MIH.

In the current study, apart from one exception with mild MIH, for all those subjects whose COHIP-SF19 scores decreased, the 'social-emotional' domain scored the highest relative decline, suggesting possibly that aesthetic/appearance factors may be playing a significant role in the decline of OHRQoL over time.

The Social-Emotional well-being domain also showed the greatest overall number of participants experiencing a decrease in their OHRQoL scores. Ten participants scored lower in this domain at follow-up, suggesting a decline in OHRQoL, with 8 participants returning a higher score (Table 3.9). Interestingly, only 2 of the participants with a decline in their social-emotional score expressed their opinions in their free-comments section of the questionnaires. Participant ID 66 mentioned "having the moulds - they were uncomfortable", and participant ID 100 recalled when asked if anything could have been done better that "Being informed about the treatments" and added, "I think I should be seen at LDI since last time because it's now been around 6 years the last time I was seen properly."

Elhenway et al., in a cross-sectional study, examined the data from 317 children in Germany aged between 7 and 14 years to explore the relationship between MIH and OHQoL. Their results showed that MIH had a substantial negative impact on affected children that increases with MIH severity (Elhennawy et al., 2022). These workers recommended that future MIH research should be designed to consider a longitudinal approach and to look into the effects and outcomes of prospective treatment. Hence, the current follow-up study aimed to fulfil such a recommendation. The results of our original baseline study (i.e. that children with MIH had lower reported OHRQoL than those with dental caries) and do support Elhennawy's findings

of a negative impact. However, it is also notable that in our follow-up study, the vast majority of those with more severe disease returned scores indicating that their OHRQoL had increased post-treatment.

## 4.7 Dental fear and anxiety (DFA)

For the subjects agreeing to participate in this follow-up study, there was a considerable variation in their previous baseline MCDASf scores. There was no evidence to suggest that the severity of MIH at first presentation had any significant impact on DFA. If anything, children with moderate or severe disease tended to report slightly lower DFA than those with mild MIH, although the small numbers of subjects made it impossible to have any certainty about this *(Table 3.11.1)*.

When comparing MCDASf scores from the baseline study to the scores in the current follow-up study, 10 participants showed a decrease in their scores, indicating a decrease in anxiety. Eight participants showed an increase in their scores corresponding to a higher state of anxiety. Again, changes in MCDASf scores between the different severity groups showed no obvious differences.

When considering the current repeat MCDASf scores for each individual against their baseline score, some interesting trends emerged (Figures 3.5 and 3.6). Those children with lower MCDASf scores at baseline (ie lower dental anxiety) tended to show greater increases in MCDASf (increased DFA) from baseline to the current time and those with higher baseline MCDASf scores (ie higher DFA) tended to return larger declines (decrease in DFA). Current (ie post-treatment followup) MCDASf scores show the opposite trend, with those with higher post-treatment scores tending to be those individuals where MCDASf had increased from baseline to current (ie the individual had become more dentally anxious) and vice versa.

## 4.8 Relationship between changes in COHIP-SF19 and changes in MCDASf scores

In the current study, a pattern emerged when comparing changes in COHIP-SF19 scores and changes in MCDASf scores. For 14 out of the total 18 subjects, an improvement in the OHRQoL score (COHIP-SF19) was accompanied by a decrease in the DFA score (MCDASf) or vice versa. In only four subjects, dental anxiety increased when OH-related QoL improved or vice versa (Table 3.14 and Figure 3.5). This pattern suggests a relationship between oral health/well-being and dental anxiety. Improvements in OHQoL, possibly due to successful treatment or intervention leading to the perception of improved comfort, function or appearance, could potentially lead to a reduction in DFA, whereas a worsening perception of oral health may be associated with an increase in DFA.

Similar associations have been reported previously, but mostly in studies of adult dental patients. In 2003, in a study of 3000 UK residents aged 16 years of age and over, who had been selected using a random probability sampling methodology, high dental anxiety scores were shown to be associated with the poorest oral health-related quality of life (McGrath and Bedi 2004). In a later study conducted in Hamburg, Germany, the Dental Anxiety Scale (DAS), the Dental Fear Survey (DFS) and the Oral Health Impact Profile (OHIP) were utilised in a study of 173 adult patients with severe dental anxiety (Mehrstedt et al., 2007). These workers found a low to moderate positive correlation between DFA and OHRQoL. More severe dental anxiety correlated with worse OHRQoL. Vermaire et al. (2008) conducted a study in the Netherlands to determine the association between dental anxiety and OHRQoL and to test if treating 35 very anxious adult patients would have a significant effect on OHRQoL. Their study found that higher dental anxiety was significantly associated with lower OHRQoL. Following often extensive dental treatment, including for most subjects restorations, extractions and root canal treatments, reduced dental anxiety,

rather than improved oral health, was found to most significantly predict enhanced OHRQoL.

Whilst the above-mentioned studies relate to adult patients, one Finnish study did focus on children aged 11-14 years attending a university hospital clinic for management of cleft lip and/or palate (Luoto et al., 2009). DFA (using the modified Children's Fear Survey Schedule-Dental Subscale) and OHRQoL (using CPQ[11-14]) were measured in 133 children (53 with cleft lip and/or palate and a control group of 82 unaffected schoolchildren). This study demonstrated a clear association between DFA and OHRQoL, with the authors concluding that DFA negatively affects children's OHRQoL, especially social and emotional well-being.

The results of our current follow-up study concur with these previous studies, suggesting that higher levels of DFA are related to poorer OHRQoL and vice-versa for child patients with MIH.

## 4.9 The relationship of pain/anxiety management and dental treatment with changes in Oral health-related quality of life (OHRQoL) and Dental Fear and Anxiety (DFA)

In this current study, the average OHRQoL was slightly lower at follow-up when compared with the pre-treatment baseline (Table 3.11.1). With such small numbers of subjects, it is important not to draw firm conclusions, but this does seem to concur with the findings of the follow-up study conducted by Jalevik (2012), who reported ongoing poorer oral health post-treatment in MIH-affected children.

All the various active pain/anxiety management modalities (LA, LA+IHS and GA) seemed to have relatively similar effects on changes in reported OHRQoL. Overall, however, local analgesia (LA) was associated with the two largest increases in reported dental anxiety, both these cases being patients who had permanent molar extractions under LA. Whilst numbers are very small, this might suggest that permanent molar extractions under LA may result in a significant increase in dental anxiety in some children.

Interestingly, both of these two subjects showed relatively small changes in OHRQoL, one actually returning increased OHRQoL (by 6 points), whilst the other returned a decrease (by 9 points).

In the only previous study to investigate DFA before and after treatment, Jälevik et al. (2002 and 2012) found that when MIH-diagnosed children and adolescents were compared to unaffected controls, they did not generally suffer increased anxiety following appropriate treatment. As mentioned above, the MIH-affected children in this study did, however, continue to have poorer dental health, which was also associated with higher ongoing treatment needs. It is not known how many of the children in Jalevik's study had extractions under LA only and there is generally a paucity of scientific evidence concerning the impact of extraction of first permanent molars under LA only in children, but this is certainly an important question, very worthy of further study.

The current study found that equal numbers of participants receiving GA reported an increase or decrease in dental anxiety, with the largest decreases in dental anxiety scores overall being reported by these individuals. Although the numbers are small, these results might suggest that treatment under GA (including having permanent molar extractions) might equally be associated with either an increase or decrease in dental anxiety in children with MIH requiring permanent molar extractions.

According to the updated European Academy of Paediatric Dentistry policy document of best clinical practice guidance for clinicians dealing with children presenting with molar-incisor-hypomineralisation (Lygidakis et al., 2021), the severity of the MIH condition is a determining factor in the type of dental treatment received by children with MIH. The current study demonstrated a similar association.
All the participants in this follow-up study whose OHQoL improved were satisfied with their experience at LDI except for participant ID 66, who had a neutral experience.

## 4.10 Family Impact

The maximum score that can be recorded in the FIS questionnaire is 56. In the current study, the highest score was 25, while the lowest score was 0 (the minimum possible score). The mean total FIS score was six (Table 3.13). The 'financial burden' domain scored zero amongst all the participants' parents, suggesting that none of the families were significantly impacted financially. The 'Parental Activity' domain was the most affected domain, with a mean score of 2.7, followed by the 'Parental Emotion' domain at 2 and the 'Family Conflict' score at 1.1.

The FIS questionnaire has been utilised in dentistry, showing a negative impact on families of children affected by malocclusion. Abreu et al. (2015) surveyed 123 parents/caregivers of children having orthodontic treatment and found negative impacts affecting mostly the Parental Emotion, Family Conflict and Financial Burden domains. Unlike the UK, orthodontic care is not subsidised in Brazil (Abreu et al., 2015).

In a study of 1000 children between the ages of two and six in South Bangalore, India, early Childhood Caries was found to have a greater negative impact on the families of affected children compared to families with children who did not have ECC (Siddaiha and Vijaya, 2024).

A Turkish version of FIS was utilised in a study that included 110 parents of 7-15-year-old children who had experienced Traumatic Dental Injury (TDI) (Bani et al., 2017). The greatest impact was seen in the PE (Parental Emotions) domain, whilst the FC (Family Conflict) was the least affected.

## 4.11 Association between the total number of clinical visits and the changes in oral health-related quality of life and anxiety

This current study found no evidence that prolonged treatment (i.e. a total number of visits) had any consistent impact on the OHRQoL, but there was a weak suggestion that prolonged treatment may be associated with an

increase in dental anxiety. There was also no evidence to suggest any impact on the families of the study children.

## 4.12 Relationship between the change in FIS total score and the duration of the journey to LDI

In the Current study, there appeared to be a directly proportional relationship between journey time and FIS score, with the total FIS score tending to increase as the journey time increased (Fig 3.23). This suggests families with longer travelling times felt a greater impact when bringing their child for dental care. This trend was seen in three of the four FIS domains (Fig 3.24, Fig 3.25, Fig 3.26). Having said this, total FIS scores were relatively low, suggesting only limited family impact across the whole group.

Yantzi et al (2001) examined the effect of travelling distance to the hospital and its impact on 113 families with children with chronic medical conditions in Ontario, Canada. The study evaluated what appears to be previously unpublished data captured by the same team from repeatedly hospitalised children and their families in 1994-1996*. Burke et al using the results of their surveys and questionnaires, they concluded that distance to the hospital was a significant factor affecting the well-being of families. Those living further away from the hospital (more than 80 km) experienced more stress, financial burden, and disruption to family life. They found that families living closer to the hospital had better access to services and support, which helped alleviate some of the challenges faced by those living further away (Yantzi et al., 2001).

## 4.13 Mode of transportation to LDI

The majority of families (14) travelled by car. Of those using public transport, two used buses, and two travelled by train. This suggests that public transport is only used by the minority of families visiting this particular centre.

* This data is referred to as ‘Burke et al (1994-1996)’ in the text, but with no specific citation given.

Leeds is widely recognised as the largest city in Western Europe not to have a mass transport system (Wikipedia, 2024). This may explain the low use of public transport by this cohort of patients. A proposed project for a mass transit system for Leeds valued at £2 billion is currently pending approval (Wikipedia, 2024). Improved access to hospital-based specialist dental and medical care may be a key benefit of such a scheme.

## 4.14 Experience of care at LDI

All but one of the families in this current study (17/18) rated their experience at LDI as either ‘very satisfactory’ or ‘satisfactory’. Only one participant was neutral to the experience, reporting neither satisfaction nor dissatisfaction. Very similar findings were reported in a Nigerian study. Adult patients were surveyed using a modification of the Dental Satisfaction Questionnaire (DSQ), and they showed a high degree of satisfaction with the treatment provided. However, the waiting time was the least liked aspect of the service provided to them (Adeniyi, 2013)

Another study explored a similar aspect of assessing the satisfaction/dissatisfaction at Jordan University Hospital, School of Dentistry. They found out that adult patients who received periodontal and fixed prosthetic treatments were satisfied with the dental setting and the treatment quality. However, their experience was negative towards the old, crowded clinical building (Ismail et al., 2024). In this current follow-up study, no negative comments were directed toward the dental clinic settings or environment.

## 4.15 Global pandemic effect

The Covid-19 global pandemic had its effect on almost every aspect of life. This study has gone through a design change from face-to-face interviews of the participants and their families to a remote approach. Communications were achieved through telephone calls and by post. Consequently, the researcher and the supervisors relied mainly on online meetings. A narrative review conducted by Goriuc in 2022 pointed out the impact of the global pandemic on dental research in the limitation and the disturbance of clinical

trials and research progress in addition to the psychological impact on the students, causing stress and anxiety (Goriuc et al., 2022).

## 4.16 Content analysis

When participants and their parents were asked to answer the free questions in the questionnaires (appendix 7, 10), ease of communication and challenges faced by the participants and their parents emerged as the two main themes in their responses.

Explaining the procedure by the staff and dentists was mentioned and praised by five participants:
*"Communicating with … and explaining procedure"*
*"Everything was explained in detail to myself and my child"*
*"Great explanation of what was going on. Made ….. feel really comfortable"*
*"Explained procedures in an appropriate way"*
*"Explaining the condition and treatment"*

Being informed about treatment is another positive response reported by two participants:
*"Provided detailed information"*

*"Being informed about the treatments"*

The feedback from the participants and their parents suggested that practices in the clinic were broadly in accordance with the GDC principles (General Dental Council, 2024).

Furthermore, statements describing the dentists and staff as being nice and friendly, were quoted seven times and there were no negative comments:
*"I got a goody bag at the end and the staff were friendly"*
*"Treated child & parent with respect in a friendly manner"*
*"Staff were friendly"*
*"Friendly staff"*
*"My dentist was really nice"*
*"Very Nice nurses + doctors"*

*"Staff were nice and made me feel slightly less nervous"*
These responses support the findings that good communication accompanied by trust between the dentist and the patient leads to patient satisfaction and locality (Szabó et al., 2023).

Challenges experienced by subjects and their parents were mainly due to the long waiting time to be seen for review appointments, and also the waiting times experienced whilst at the hospital clinic.

*"Reduce waiting time as this increases anxiety"*

*"Waiting times could have been better, especially waiting for xrays"*

*" Waiting times have improved significantly since before covid"*

Previous authors have highlighted that patients waiting for longer times in the waiting room after arriving at the clinic may have higher anxiety levels than those who waited for shorter periods of time (Coffey et al., 1983).

An additional challenge faced by the participants and their families was the shortage of appointment availability.

*" I had to trace me follow up appointments. I contacted me the secretaries each time who sorted out appt for me"*
*"Waited long time for appointments. Follow-up slow. Had to ring to chase appointments"*
*“The availability of appointments is quite limiting. I feel like treatment has taken longer due to this"*

Hence, as well as the positive feedback from subjects and their families, there are two key areas where improvements need to be considered by the team, namely waiting room waiting times (including possibly looking at ways to improve the waiting room environment/experience) and trying to minimise waiting times for appointments. Long waiting times for appointments are unfortunately not unusual in the UK National Health Services currently, often reflecting limitations in overall capacity, but this study suggests that efforts should be made to improve waiting times and the ability of families to access

information about their current waiting status and estimated further waiting times.

## Conclusions

- The re-recruited subgroup included a reasonable representation of the three different severities of MIH and a reasonably representative range of paint/anxiety management and dental treatment modalities.

- Participants had relatively small numbers of visits to the paediatric dentistry department (range 2-10, mean 4.94). Those going on to having active orthodontic treatment required a significantly higher total number of visits (range 26-42)

- A slight majority of participants (10 out of 18) showed an overall improvement in total oral health-related quality of life (OHR QoL) scores in the current study with the greatest number of participants showing improvement in the Functional Wellbeing domain. Where a decline in OHRQoL occurred (8 subjects) the largest relative declines were seen in the Socio-Emotional Wellbeing domain.

- A slight majority of participants (10 out of 18) showed an overall decline in dentally related anxiety scores.

- For the majority of participants (14/18), improvements in OHRQoL were inversely associated with changes in dental anxiety, with the majority of those with increased OHQoL scores reporting lower dental anxiety scores and vice-versa.

- All the various active pain/anxiety management modalities (LA, LA+IHS and GA) seemed to have relatively similar effects on changes in reported OHRQol, but local analgesia (LA) was associated with the two largest increases in dental anxiety, both these cases being patients who had permanent molar extractions under LA. The results of this study suggest that permanent molar extractions under LA can result in a significant increase in dental anxiety in some children.

- Equal numbers of participants receiving GA reported an increase and decrease respectively in dental anxiety, with the largest decreases in dental anxiety scores overall being reported by these individuals. These results suggest that treatment under GA (including having permanent molar extractions) whilst being potentially associated with increases in dental anxiety, might equally be associated with a significant decrease in dental anxiety in children with MIH requiring permanent molar extractions.

- MIH and its treatment appear to have a relatively low overall impact on the family, with the majority of impact reported on Parental/Family Activities and Parental Emotions. The financial impact was reported as 0 for all the participants.

- There was no evidence that prolonged treatment (ie total number of visits) had any consistent impact on OHRQoL but there was a weak suggestion that prolonged treatment might be associated with an increase in dental anxiety.

- Longer journey time to the hospital was associated with increased Family Impact.

- The majority of families gave positive responses to patient experience questions. However, some expressed dissatisfaction with long waiting times, and the limited availability of, appointments.

- Study design changes and delays largely associated with the 2020 global pandemic resulted in just under one-quarter of participants from the original study group being re-recruited. This relatively low level of re-recruitment requires that caution be exercised when interpreting the study results.

## Strengths and Limitations

This follow-up study has significant limitations. The re-recruitment rate is low at 22% which translated to a smaller sample size. The interruption of the study due to COVID-19 and the change of the original plan led to a longer period between the treatment and the questionnaire administration that had originally been envisaged. Additionally, the questionnaires were administered in a home setting, which may have been influenced by parents, or the parent's questionnaires may have been influenced by the participants.

On the other hand, this is one of the few prospective studies to date of children being treated for MIH comparing oral health-related quality of life and dental anxiety pre and post-treatment. The extended period also allowed the study to be a valid long-term follow-up to assess the effectiveness of MIH management from the perspective of the patients and their families.

## Future Research Recommendations

Researchers planning prospective follow-up studies should ensure prospective ethical approval and consenting arrangements are in place to ensure the capture of robust, comprehensive contact information, and to enable relocation of participants of long-term studies using digital tools that are unlikely to change such as email addresses and social-media platforms.

Further investigation of the use of reliable and confidential web-related e-resources for collecting data from participants, such as university webpages with direct access to the questionnaires, should be conducted.

More research is needed on the impact that MIH has on families of affected children.

Research investigating impact from the perspective of MIH-affected children/adolescents is important for a better understanding of the long-term effect of the condition.

Short-term MIH-related follow-up immediately pre and post-treatment studies are also needed as they may give more accurate feedback.

Future studies may also include assessing the dental outcomes to consider what impact the condition of MIH and its treatment has on permanent dentition in mid to late adolescence.

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# Appendix 1

Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

NHS
Health Research Authority

Mr. Mohammad Hussein
Flat 80 Axis Tower
9 Whitworth Street West
LS2 9LU

Email: approvals@hra.nhs.uk

21 December 2021

Dear Mr Hussein

**HRA and Health and Care Research Wales (HCRW) Approval Letter**

| | |
|---|---|
| **Study title:** | **A follow-up study of children referred for management of Molar Incisor Hypomineralisation: an analysis of treatments received, changes in self-reported oral health-related quality of life, dental anxiety, and the impact of the condition and treatment on the child's family.** |
| **IRAS project ID:** | **289650** |
| **REC reference:** | **21/PR/1069** |
| **Sponsor** | **University of Leeds, School of Dentistry** |

I am pleased to confirm that **HRA and Health and Care Research Wales (HCRW) Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the "Information to support study set up" section towards the end of this letter.

**How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?**
HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) have been sent to the coordinating centre of each participating nation. The relevant natior

Please see IRAS Help for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

**How should I work with participating non-NHS organisations?**
HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

**What are my notification responsibilities during the study?**

The standard conditions document "After Ethical Review – guidance for sponsors and investigators", issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Progress reports of the study

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

**Who should I contact for further information?**
Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **289650**. Please quote this on all correspondence.

Yours sincerely,
Juliana Araujo

Approvals Specialist

Email: approvals@hra.nhs.uk

*Copy to: Ms Jean Uriacke*

## Appendix 2

The Leeds Teaching Hospitals NHS Trust

UNIVERSITY OF LEEDS

University of Leeds School of Dentistry
Leeds Dental Hospital

Clarendon Way, Leeds LS2 LU
Tel. Switchboard 0113-2440111

[10/00/2021, Version 7]

IRAS Project ID is: 289650

# Invitation to take part in a research project

Dear Parent/caregiver

About four years ago, you may remember that you and your child took part in a research study at Leeds Dental Institute. The research was about enamel problems on adult molars and your contribution was very valuable to us. We are now conducting a follow-up study of all the children interviewed in that study.

We would like to invite you and your child to join this follow-up research study ***"A follow-up study of children referred for management of Molar Incisor Hypomineralisation (MIH): an analysis of treatments received and changes in self-reported oral health-related quality of life, dental anxiety and the impact of the condition and treatment on the child's family."***

This study is a postgraduate student (Mr. Hussein) project contributing to the Doctor of Paediatric Dentistry Research Degree.

We are interested to find out how this adult-molar enamel problem and its treatment have affected you and your child since we first met you. This will help us to understand what effects the condition has on children and their families. It may also help us to improve the treatment for children with the same condition in the future.

The study does not involve returning to the LDI, but we will ask volunteers to fill out similar questionnaires to the ones they filled out in the previous study and then return these to the University of Leeds so we can see if things have changed since the first time. The questionnaires will be sent to you and returned to us through the post or if you prefer, electronically by email. Mr Hussein will also make two phone calls:

1) A first short phone call before we send anything else out to you. This is to explain a bit more about the study and ask if you and your child are willing to volunteer again for this research. Participation is entirely voluntary and if you do not want to take part, it will not affect any future treatment at LDI. You do not need to do anything else right now.
He will therefore call within the next 1-2 weeks to ask if you and your child are willing to take part. If you are happy to take part, He will explain more about the study when I call or agree a convenient time to call back to complete the explanation.
2) He will make a second call once you have received your questionnaire package. This will be just to check if you have been able to fill in the questionnaires and answer any further questions. We will also make sure this is done at a time convenient to you.

Your and your child's participation in this research is entirely voluntary and you can decide not to take part if you wish without any effect on any future treatment for either your son/daughter or yourself at Leeds Dental Institute.

If your telephone number or address has changed since you were last seen at Leeds Dental Institute, but you still want to take part, please email an updated telephone number and address to my email below.

Yours sincerely,

Richard Balmer
Clinical Lecturer and Consultant in Paediatric Dentistry University of Leeds, School of Dentistry
Clarendon Way
LS2 9LU
Email:
r.c.balmer@leeds.ac.uk
0113 3436228

Mohammad Hussein
Postgraduate in Paediatric Dentistry
Leeds Dental Institute
Clarendon Way
LS2 9LU
Email:
dnmakh@leeds.ac.uk
mohammad.hussein@nhs.net

## Appendix 3

UNIVERSITY OF LEEDS

University of Leeds School of Dentistry
Leeds Dental Hospital

Clarendon Way, Leeds LS2 LU
Tel. Switchboard 0113-2440111

# Participant Information Sheet

**(under 16 years)**

**Title of study**

**A follow-up study of children referred for management of Molar Incisor Hypomineralisation (MIH): an analysis of treatments received and changes in self-reported oral health-related quality of life, dental anxiety and the impact of the condition and treatment on the child's family.** (IRAS Project ID is: 289650)

**Why are we giving you this leaflet?**

Four years ago, you helped us to understand how you felt about your teeth and going to the dentist. We want your help again to tell us what you think about your teeth and going to the dentist now, and the treatment you had since then.

**What Is this study?**

Your teeth have a special but very common condition called MIH. We want to know more about how you feel about it and the treatment you received. We will compare your new answers to your original ones to see if things have changed over the last few years.

**If I join this Study, What Will Happen?**

If you decide to join this study:

- If you agree to take part, the details of the treatment you have had at LDI will be collected by the research team from your clinical records and we will not need you to come back for the study.
- We will send to you and your parent/caregiver two sets of questions about how you feel about your teeth and about what it was like when you came for treatment to Leeds Dental Institute.
- We will speak to you and your parent/caregiver on the telephone to check you and your parent/caregiver are happy to take part before sending the questions
- You can do this at home, you do not need to come back to the hospital

☞ We may also contact you again in the future and ask if you can come back to the Dental Hospital as part of further research to see how your adult teeth are and how you are getting on. When that time comes it will be OK if you don't want to come back to the Dental Hospital.

**Do I have to join this study?**

•You decide whether you are happy to take part in this study

•You don't have to join this study if you don't want to

•Even if you join this study, you can change your mind at any time and that's fine

**Thank you for taking the time to read this information sheet!**

## Appendix 4

UNIVERSITY OF LEEDS

University of Leeds School of Dentistry
Leeds Dental Hospital

Clarendon Way, Leeds LS2 LU
Tel. Switchboard 0113-2440111

[1/12/2021, Version 7]

# Participant Information Sheet (16 years)

## Title of study

**A follow-up study of children referred for management of Molar Incisor Hypomineralisation: an analysis of treatments received and changes in self-reported oral health-related quality of life, dental anxiety ,and the impact of the condition and treatment on the child's family.** (IRAS Project ID is: 289650)

I would like to invite you to take part in the research study named above. The University of Leeds is the sponsor for this study.

### 1- Why have I been given this leaflet?

About 5 years ago you took part in a similar study carried out when you attended Leeds Dental Institute (LDI). That study was called "The Dental and Orthodontic features, baseline Anxiety and Quality of life of Children referred to a specialist centre for management of First Permanent Molars with Molar Incisor Hypomineralisation (MIH) or caries" At that time you may remember we said we might contact you again to find out what has happened about your dental health and treatment since then.

### 2-What is the purpose of this study?

- To find out what dentalcare you have received since we did the first study.
- To assess how the tooth-enamel problem you have (called 'MIH') had affected you since then.
- To understand what effect the MIH and its treatment has had on you and your family
- To help us to improve treatment of this condition.

### 3- Who is doing the study?

Due to the current pandemic, the study has been designed to be carried out remotely. If you agree to take part, we will not need you to come back to LDI. Details of any treatment carried out at LDI will be taken by a research team from your clinical records held at LDI. Other information will be captured using questionnaires which we will send to you.

The study will be conducted remotely by a postgraduate in Paediatric Dentistry (Mohammad Hussein) under the supervision of a clinical Professor in Paediatric Dentistry (Prof.Bernadette Drummond), a Consultant in Paediatric Dentistry (Dr.Richard Balmer), and a Honorary Senior Lecturer involved in the previous study (Mr.Stephen Fayle) and a consultant in Orthodontics (Mr.James Spencer).

## 4- Do I have to take part?

If you do not want to take part that is OK and will not affect in any way future treatment which might provided at Leeds Dental Institute.

## 5-Risks and benefits

There are no recognised risks to you taking part in this study. The benefits are that we will be able to understand the treatment progress for you and others who have Molar Incisor Hypomineralistion (MIH).

## 6- What will be involved if I take part in this study?

If you agree to take part in the study you will be information sheets, consent forms for you and your parent/caregiver, and two questionnaires. You will be able to fill these in at home and you will not need to come back to the clinic. The questionnaire that you would be asked to complete is largely the same as the ones you completed in the first study (a dental anxiety questionnaire and a dental quality of life questionnaire) with a couple of extra questions. We will also include a new questionnaire for your parent/caregiver to fill out called the 'Family Impact Scale questionnaire'. It should take no longer than 15-20 minutes to fill in the questionnaires. You will be given a choice if you prefer the questionnaires to be sent by post or emailed to you.
There will also be a follow-up phone call where Mr.Hussein can answer any further questions and make sure you have been able to fill in the questionnaires. We will send participants a small shopping voucher to thank them for their participation.

We also ask for your agreement to possibly be contacted again in the future for further research into how your teeth are. Again taking part in any future research will be entirely voluntary.

## 7- Can I withdraw from the study at any time?

Yes, you are free to withdraw from this study at any time without giving any reasons but the research team will keep the research data about you that they already have. All research data will be included in reports but will not include any identifying information. If you or your parent/caregiver decide to withdraw that is OK and will not affect in any way future treatment we might provide for you or your parent/caregiver at Leeds Dental Institute.

## 8- Will the information I give be confidential?

Yes. We will use information from the questionnaires filled in by you and your parent/caregiver and from your dental records. We will only use the information that we need for the research study. All the data collected from the dental records and questionnaires will be coded so that you can not be identified in any reports. It will be kept secure on password-protected University of Leeds computers within a secure research facility at the Dental Institute. The access codes will be held by a senior supervisor. People who do not need to know who you are will not be able to see your name or contact details. No names or personal data will be published and data will remain anonymous. You can read the University's privacy notice here: https://dataprotection.leeds.ac.uk/wpcontent/uploads/sites/48/2019/02/Research-Privacy-Notice.pdf

Health research authority can be found here:
https://s3.eu-west
2.amazonaws.com/www.hra.nhs.uk/media/documents/My_data_and_research_qbmsVYc.pdf
Should you need to, the University Data Protection Officer can be contacted by email: dpo@leeds.ac.uk

## 9-What will happen to the results of this study?

The results of this study will be discussed with other colleagues in the department and will be published in Mr Hussein's thesis. They will be presented at scientific conferences and in scientific journals. No identifiable information will be used in any presentation or publications, so no one except the researchers carrying out this study will know your identity. This information will not be used for any other purpose. We will send you a summary of the result of this study.

## 10- What if I have any other questions or a problem?

If you have any questions or concerns about any aspect of this study, you should ask to speak with the researchers who will do their best to answer your questions. You can do this by :

- Sending an email.
- Phoning us.
- Asking Mr.Hussein when he calls you.

If something goes wrong or you are harmed during the research study, there are no special compensation arrangements. If you are injured and this is due to someone's negligence, then you have grounds for legal action for compensation, but you may have to pay your legal costs. The standard NHS complaints mechanisms will still be available to you.
The University, when acting as Sponsor, has insurance cover in force, which meets claims against it and where those claims arise from the Universities own negligence and its role and activities relating to the study (and which is subject to the terms, conditions, and exceptions of the relevant policy). Clinical negligence indemnification will rest with the participating NHS Trust under standard NHS arrangements.

If you are still unhappy and wish to complain formally, you can do this by contacting Patient Advice and Liaison Services (PALS). Telephone: 0113 2066261, email: patientexperience.leedsth@nhs.net

## 11- Who has reviewed this study?

This research has been internally reviewed by senior academics within the University of Leeds, and reviewed by the NHS Ethics committee and the Health Research Authority. Research Ethics (IRAS) project ID: 289650.

## Thank you for taking the time to read this information sheet

If you would like more information or have any questions or concerns about this study or would like to request a summary of the results of the study, please contact any member of the study team:

Mr. Mohammad Hussein
Postgraduate in
Paediatric Dentistry
Leeds Dental Institute
Clarendon Way
LS2 9LU

Email:
dnmakh@leeds.ac.uk

Mr. Mohammad Hussein
Postgraduate in

Dr.Richard Balmer
Clinical Lecturer and
Consultant in
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# Appendix 5

A2 16

NHS
The Leeds Teaching Hospitals
NHS Trust

UNIVERSITY OF LEEDS

[14/10/2021, Version 1]

University of Leeds School of Dentistry
Leeds Dental Hospital

Clarendon Way, Leeds LS2 LU
Tel. Switchboard 0113-2440111

IRAS Project ID: 289650

Participant Identification Number: ☐

## Participant Consent Form

**Title of study**

**A follow-up study of children referred for management of Molar Incisor Hypomineralisation (MIH): an analysis of treatments received and changes in self-reported oral health-related quality of life, dental anxiety and the impact of the condition and treatment on the child's family.**

Name of the researcher: **Mohammad Hussein**

***Please write your initials in each box to indicate your agreement***

1. I confirm I have read and understood the participant information sheet dated May 2021 for the above study. I have had the opportunity to consider the information ☐
2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without it affecting my medical care or legal rights. ☐
3. I understand that relevant sections of my medical notes and data collected during the study, maybe looked at by relevant individual/staff at the Leeds School of Dentistry or from regulatory authorities, or Leeds Teaching Hospitals NHS Trust, where it is relevant to us taking part in this research. I give permission for these individuals to have access to my records. ☐
4. I agree for data collected to be kept for a future research. ☐
5. I agree to be contacted again in the future for follow-up research into my dental condition ☐
6. I agree to take part in the above study. ☐

……
Name of person giving consent

…… ……
Signature Date

……
Name of researcher

…… ……
Signature Date

# Appendix 6

IRAS ID: 289650
[10/10/2021, Version 5]
Participant Identification Number:

## Participant Assent Form (under 16 years)

**Title of study**
A follow-up study of children referred for management of Molar Incisor Hypomineralisation: an analysis of treatments received and changes in self-reported oral health-related quality of life, dental anxiety and the impact of the condition and treatment on the child's family.

Name of the researcher: **Mohammad Hussein**
***Please read the 'Patient Information Sheet' and then read each question below. Please write your initials in all the boxes to show you agree with each question***

1. I have read the information sheet about this project ☐
2. I understand what this project is about ☐
3. I have been able to ask questions about this research ☐
4. I understand the answers to any questions I have asked ☐
5. I understand it's ok to stop taking part in this research at any time ☐
6. I understand I might be contacted again in the future about more research into the problem with my tooth enamel ☐
7. I am happy to take part in this research study ☐

| | | |
|---|---|---|
| ……. | …….. | …….. |
| Name of patient | Signature | Date |
| ……. | …… | ……... |
| Name of parent/carer | Signature | Date |
| ……. | …… | ……... |
| Name of researcher | Signature | Date |

## Appendix 7

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SITY OF LEEDS

UNIVERSITY OF LEEDS

[10/10/2021, Version 9]
IRAS Project ID: 289650
Participant ID ----------

# Participants Questions

**Please answer the three sets of questions below**

### PART 1: Modified Child Dental Anxiety Scale (MCDASf)

For the first set of questions below I would like you to show me how relaxed or worried you get about the dentist and what happens at the dentist. To show me how relaxed or worried you feel, please use the simple scale below. The scale is like a ruler going from 1, which would show that you are relaxed, to 5, which would show that you are very worried.

Please circle the appropriate number on the scale

**1** would mean: relaxed/not worried
**2** would mean: very slightly worried
**3** would mean: fairly worried
**4** would mean: worried a lot
**5** would mean: very worried

| How do you feel about... | | | | | |
|---|---|---|---|---|---|
| .. going to the dentist generally? | 1 | 2 | 3 | 4 | 5 |
| .. having your teeth looked at? | 1 | 2 | 3 | 4 | 5 |
| .. having your teeth scraped and polished? | 1 | 2 | 3 | 4 | 5 |
| .. having an injection in the gum? | 1 | 2 | 3 | 4 | 5 |
| .. having a filing? | 1 | 2 | 3 | 4 | 5 |
| .. having a tooth taken out? | 1 | 2 | 3 | 4 | 5 |
| .. being put to sleep for treatment? | 1 | 2 | 3 | 4 | 5 |
| .. having a mixture of gas and air which will help you to feel comfortable for treatment but cannot put you to sleep? | 1 | 2 | 3 | 4 | 5 |

### PART 2: Child Oral Health Impact Profile-Short Form 19 (COHIP-SF19)

For this last set of questions, please read each statement carefully and choose the answer that best describes you in the past 3 months regarding your teeth, mouth or face.
We want to know how you really feel. Please put a tick in the box under the best answer.

**In the last 3 months have you ….**

| Domains | Scoring | | | | |
|---|---|---|---|---|---|
| Oral Health – Well-Being | 0= Almost all the time | 1= Fairly often | 2= Sometimes | 3= Almost never | 4= Never |
| Q1. Had pain in your tooth/teeth? | | | | | |
| Q2. Had discoloured teeth or spots on your teeth? | | | | | |
| Q3. Had crooked teeth or spaces between your teeth? | | | | | |
| Q4. Had bad breath? | | | | | |
| Q5. Had bleeding gums | | | | | |
| Functional Well-Being | 0= Almost all the time | 1= Fairly often | 2= Sometimes | 3= Almost never | 4= Never |
| Q6. Had difficulty eating food you would like to eat | | | | | |
| Q7. Had trouble sleeping | | | | | |
| Q8. Had difficulty saying certain words | | | | | |
| Q9. Had difficulty keeping your teeth clean | | | | | |
| Social-Emotional Well-Being | 0= Almost all the time | 1= Fairly often | 2= Sometimes | 3= Almost never | 4= Never |
| Q10. Been unhappy or sad | | | | | |
| Q11. Felt worried or anxious | | | | | |
| Q12. Avoided smiling or laughing | | | | | |
| Q13. Felt that you looked different | | | | | |
| Q14. Been worried about what the people think about your teeth, mouth or face | | | | | |
| Q15. Been teased, bullied, or called names by other children | | | | | |
| Q16. Missed school for any reason | | | | | |
| Q17. Not wanted to speak/read out loud in class | | | | | |
| | 0= Never | 1= Almost never | 2= Sometimes | 3= Fairly often | 4= Almost all the time |
| Q18. Been confident | | | | | |
| Q19. Felt that you were attractive (good looking) | | | | | |

**PART 3: About your experience at Leeds Dental Institute**
(please write your answers in the spaces below each question)

1. **When you had your teeth treated at Leeds Dental Institute ….**
   was there anything you remember as being particularly difficult?

   was there anything that was good or easier than you thought it would be?

   was there anything we could have done better?

2. **Is there anything else you would like to tell us about your teeth or visits to LDI?**

## Thank you for taking the time to answer these questions!

**If you have not managed to fill in any parts of this questionnaire or have any questions, don't worry. Either call or email us to let us know and we will respond back, or just keep hold of all the forms and we will call you in a few days.**

**Mohammad Hussein**
dnmakh@leeds.ac.uk
mohammad.hussein@nhs.net

## Appendix 8

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[13/10/2021, Version 11]

# Parent Information Sheet

## Title of study

**A follow-up study of children referred for management of Molar Incisor Hypomineralisation: an analysis of treatments received and changes in self-reported oral health-related quality of life, dental anxiety ,and the impact of the condition and treatment on the child's family.** (IRAS Project ID is: 289650)

I would like to invite you and your child to take part in a research study, which is named above. The University of Leeds is the sponsor for this study.

This information sheet is to help you understand what the study is about and what you would need to do. Please read it before deciding. After reading this, if you or your child are not sure, do not worry, as I will telephone-call in a week or so to answer any questions and check if you and you child agree to take part. This call should take no longer then 5-10 minutes. This research is entirely voluntary, so you do not need to take part if you do not want to.

## 1- Why have I been given this leaflet?

About 5 years ago, your child took part in a study at Leeds Dental Institute which aimed to find out more about how Molar Incisor Hypomineralisation (MIH) affects children with the condition, and what their teeth were like at the time they first came along to Leeds Dental Institute. We now want to carry out a follow-up study of the same children to find out what has happened since. The previous study was called "The Dental and Orthodontic features, baseline Anxiety and Quality of life of Children referred to a specialist centre for management of First Permanent Molars with Molar Incisor Hypomineralisation (MIH) or caries" and at that time you may remember we said we might contact you again to find out what has happened about your child's dental condition and treatment since then.

## 2-What is the purpose of this study?

At the moment, very little is known about how MIH and its treatment affects children over the longer term. We want to find out how the condition affects children and their families. We also want to find out how children with MIH and their families feel about their dental health after they have had treatment. This will help us to improve how we treat children with this condition.

To do this we want to do the following things:

- To review children in the original study, who were diagnosed with MIH around 5 years ago and determine what care they have received. We would do this by reviewing the records of your child's treatment at Leeds Dental Institute
- To assess how the condition has affected your child and your family since then and to understand what effect the condition and treatment have had on your child and your family. We would do this by asking your child to re-answer the same questions they were asked in the first study. We would also like to ask you to answer some questions to help us understand how your child's MIH condition has affected you and the family.

If you agree to take part, your child's treatment details at LDI will be collected by the research team from their clinical records held here. The questionnaires can be completed at home. We will not need you to come back to Leeds Dental Institute.

## 3- Who is doing the study?

Due to the current global pandemic situation, the study will be conducted remotely by a postgraduate in Paediatric Dentistry (Mohammad Hussein) under the supervision of a clinical Professor in Paediatric Dentistry (Prof.Bernadette Drummond), a Consultant in Paediatric Dentistry (Dr.Richard Balmer), and a Honorary Senior Lecturer involved in the previous study (Mr.Stephen Fayle) and a consultant in Orthodontics (Mr.James Spencer).

## 4- Does my child have to take part?

No, it is entirely voluntary. If you or your child do not want to take part that will not affect in any way future treatment provided at Leeds Dental Institute for you or your child.

## 5-Risks and benefits

There are no perceived risks to you or your child taking part in this study. The benefits are that we will hope to improve our understanding of how MIH affects children and families. This would help us to improve the care we provide for your child and other children who have Molar Incisor Hypomineralistion..

## 6- What will be involved if I and my child take part in this study?

If you agree for your child to participate in the study you will be sent a package that includes consent forms for you and your child, and two questionnaires (one for you, one for your child). You will be asked to sign a consent form. If your child is under 16, they will sign an assent form. If they are over 16 they will sign their own consent form. You will be given a choice if you prefer the questionnaires to be sent by post or emailed to you.

You will be able to complete all forms and questionnaires at home. You will not need to come back to the clinic. The child questionnaire is the same as your child completed in the first study (questions about a dental anxiety and dental-related quality of life) with a couple of extra questions added at the end about their experiences. We will also include a new questionnaire for you to fill out called the Family Impact Scale questionnaire, which helps us to understand how medical conditions and their treatment affects a child's family. It should take no longer than 15-20 minutes to fill in both questionnaires.

We will also have a follow-up phone call where Mr Hussein can answer any further questions, and to check you have been able to fill in the questionnaires OK. This second phone call will take about 5-10 mins.

We will also ask for your agreement to be contacted again in the future for further research into how your childs's teeth are. This, again, will be entirely voluntary.

We will send participants a small shopping voucher to thank them for their participation.

## 7- Can I withdraw from the study at any time?

Yes, you are free to withdraw from this study at any time without giving any reasons. The research team would only keep the research data about you that they already collected. If you or your child decide to withdraw at any stage, that is OK and will not affect in any way future treatment we might provide for you or your child at Leeds Dental Institute.

## 8- Will the information I give be confidential?

We will use information from the questionnaires filled in by you and your child, and from your child's dental records. We will only use information that we need for the research study. All the data collected from the dental records and questionnaires will be coded so that you or your child can not be identified in any reports. It will be kept secure and confidential on password-protected University of Leeds computers and in a lockable secure office in the Worsley building on level 6. The code will be held by a senior supervisor. People who do not need to know who you are will not be able to see your name or contact details. No names or personal data will be published and data will remain anonymous. If you agree, the data will be kept for up to 5 years with your contact details so that you may be contacted for any future study on your child's condition. After that time it will be destroyed. You can read the University's privacy notice here:
https://dataprotection.leeds.ac.uk/wpcontent/uploads/sites/48/2019/02/Research-Privacy-Notice.pdf.
Health research authority can be found here:
https://s3.eu-west-2.amazonaws.com/www.hra.nhs.uk/media/documents/My_data_and_research_qbmsVYc.pdf
Should you need to, the University Data Protection Officer can be contacted by email: dpo@leeds.ac.uk.

## 9-What will happen to the results of this study?

The results of this study will be discussed with other colleagues in the department and will be published in Mr Hussein's thesis. They will be presented at scientific conferences and in scientific journals. No identifiable information will be used in any presentation or publications, so no-one except the researchers carrying out this study will know your identity. This information will not be used for any other purpose. We will send you a summary of the result of this study.

## 10- What if I have any other questions or a problem?

If you have any questions or concerns about any aspect of this study, you should ask to speak with the researchers who will do their best to answer your questions. You can do this by :

- Asking Mr.Hussein when he calls you.
- Sending an email.
- Phoning us.

If you believe or your child are adversely affected by any issues raised by this research project at any stage, you can contact the Senior Investigator, Dr Balmer directly who will be able to discuss these issues and arrange any further support or advice as necessary. Telephone: 01133436228, email: r.c.balmer@leeds.ac.uk

If you are still unhappy and wish to complain formally, you can do this by contacting Patient Advice and Liaison Services (PALS). Telephone: 0113 2066261, email: patientexperience.leedsth@nhs.net

If something goes wrong or you are harmed during the research study, there are no special compensation arrangements. If you are injured and this is due to someone's negligence, then you have grounds for legal action for compensation, but you may have to pay your legal costs. The standard NHS complaints mechanisms will still be available to you.
The University, when acting as Sponsor, has insurance cover in force, which meets claims against it and where those claims arise from the Universities own negligence and its role and activities relating to the study (and which is subject to the terms, conditions, and exceptions of the relevant policy). Clinical negligence indemnification will rest with the participating NHS Trust under standard NHS arrangements.

## 11- Who has reviewed this study?

This research has been internally reviewed by senior academics within the University of Leeds, and reviewed by the NHS Ethics committee and the Health Research Authority. Research Ethics (IRAS) project ID: 289650.

## Thank you for taking the time to read this information sheet

If you would like more information or have any questions or concerns about this study or would

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Mr. Mohammad Hussein
Postgraduate in Paediatric Dentistry
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LS2 9LU

Dr.Richard Balmer

Mr.James Spencer
Consultant in Orthodontics University of Leeds, School of

# Appendix 9

**A1**

IRAS Project ID: 289650
[10/10/2021, Version 8]

NHS
The Leeds Teaching Hospitals
NHS Trust

UNIVERSITY OF LEEDS

University of Leeds School of Dentistry
Leeds Dental Hospital

Clarendon Way, Leeds LS2 LU
Tel. Switchboard 0113-2440111

Participant Identification Number:

**Parent Consent Form**

**Title of study**

**A follow-up study of children referred for management of Molar Incisor Hypomineralisation (MIH) : an analysis of treatments received and changes in self-reported oral health-related quality of life, dental anxiety and the impact of the condition and treatment on the child's family.**

Name of the researcher: **Mohammad Hussein**

***Please write your initials in each box to indicate your agreement***

1. I confirm I have read and understood the parent information sheet dated May 2021 for the above study. I have had the opportunity to consider the information ☐
2. I understand that my child's and my participation is voluntary and that we are both free to withdraw at any time without giving any reason, without it affecting my or my child's medical care or legal rights. ☐
3. I understand that relevant sections of my child's medical notes and data collected during the study, maybe looked at by relevant individual/staff at the Leeds School of Dentistry or from regulatory authorities, or Leeds Teaching Hospitals NHS Trust, Where it is relevant to us taking part in this research. I give permission for these individuals to have access to my child's records. ☐
4. I agree for data collected from my child to be kept for a future research. ☐
5. I agree to be contacted again in the future for follow-up research into the dental condition my child has. ☐
6. I agree for me and my child to take part in the above study. (if my child is 16 or over, I understand they will sign their own consent form) ☐

| | |
|---|---|
| Name of parent/carer giving consent | Name of your child |
| Signature | Date |
| Name of researcher | |
| Signature | Date |

When completed; 1 copy researcher site file, 1 copy to participant, 1 copy scanned SALUD electronic dental file.

## Appendix 10

[13/10/2021, version 2]

IRAS Project ID: 289650

Participant ID ----------

**Family Impact Scale Questionnaire**

***This questionnaire is to be filled by the parent/caregiver***

We want you to think about how your child's dental enamel problems, and the treatment provided, have affected the family overall since your child's treatment at LDI started.

**Related to your child's dental enamel condition and treatment, how often…**

1) **Have you or has the other parent taken time off work?**

Never Once or twice Sometimes Often Daily Don't Know

2) **Has your child required more attention from you or the other parent?**

Never Once or twice Sometimes Often Daily Don't Know

3) **Have you or has the other parent had less time for yourselves or other family members?**

Never Once or twice Sometimes Often Daily Don't Know

4) **Has your sleep or that of the other parent been disturbed?**

Never Once or twice Sometimes Often Daily Don't Know

5) **Have the family activities been interrupted?**

Never Once or twice Sometimes Often Daily Don't Know

6) **Have you or has the other parent been upset?**

Never Once or twice Sometimes Often Daily Don't Know

7) **Have you or has the other parent felt guilty?**

Never Once or twice Sometimes Often Daily Don't Know

8) **Have you or has the other parent worried that your child will have fewer life opportunities?**

Never Once or twice Sometimes Often Daily Don't Know

**9) Have you felt uncomfortable in public places?**

Never Once or twice Sometimes Often Daily Don't Know

**10) Has your child argued with you or the other parent?**

Never Once or twice Sometimes Often Daily Don't Know

**11) Has your child been jealous of you of other family member?**

Never Once or twice Sometimes Often Daily Don't Know

**12) Has your child's condition caused disagreement or conflict in the family?**

Never Once or twice Sometimes Often Daily Don't Know

**13) Has your child blamed you or the other parent?**

Never Once or twice Sometimes Often Daily Don't Know

**14) Has your child's condition caused financial difficulties for your family?**

Never Once or twice Sometimes Often Daily Don't Know

Please also answer the following questions about your visits to Leeds Dental Institute

**15) When you attended appointments at Leeds Dental Institute, what mode(s) of transport did you use for your journey?**

Car, Bus, Train, Taxi, Walking, Other (please specify) ……..

**16) Approximately what was the cost of transportation for each journey (in one direction)?**

Nil £1-5 £5-10 £10-20 £20-30 £30-40 £40-50 more than £50

**17) What was the approximate total time for your journey *in one direction*, door-to-door from your home to the hospital?**

**…….**

**18) Overall, how would you describe your journeys to and from LDI?**

**Very Easy Easy Moderate Difficult Very Difficult**

**19) Overall, how satisfied are you with you and your child's experience at the LDI during the past 4 years?**

**Very Satisfied Mostly Satisfied Neutral Dissatisfied Very Dissatisfied**

**What did we do well?**

**What could we have done better?**

**20) Is there anything else you would like to tell us about your child's dental enamel condition or visits to LDI?**

**Thank you for taking the time to answer these questions.**

**Please return this together with the other blue forms in the postage-paid envelope provided.**

**If you have not managed to fill in any parts of this questionnaire or have any questions, don't worry. Either call us to let us know and we will call back, or just keep hold of all the forms and we will call you in few days.**

**Mohammad Hussein**
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## Appendix 11

IRAS Project ID: 289650

**Title of study**

**A follow-up study of children referred for management of Molar Incisor Hypomineralisation (MIH): an analysis of treatments received and changes in self-reported oral health-related quality of life, dental anxiety and the impact of the condition and treatment on the child's family.**

Dear Family,

Thank you for taking part in our study. We really appreciate the time you took. Your participation will help us improve our knowledge about the treatment for MIH and this improve our care in the future.

Please accept the attached £10 voucher as a token of our appreciation.

Yours Sincerely,
Mohammad Hussein