

**Parental Perception of Children Affected
by Amelogenesis Imperfecta (AI) and
Dentinogenesis Imperfecta (DI):
A Qualitative Study.**

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Doctorate in Dental Surgery.

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Declaration

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Summary

Aims: This study aimed to investigate and explore parental attitudes, beliefs, values and expectations regarding aesthetics and treatment need of children in primary dentition affected by AI and DI.

Methods: A descriptive qualitative study was conducted involving a purposive sample of thirteen parents of young children affected by AI and DI. Parents were divided into two separate focus groups; mothers (n=7) and fathers (n=6). A topic guide was formulated and included open-ended questions. Eleven standardised photographs of a smile showing primary teeth affected by varying severity of AI/DI and photographs of different aesthetic treatments were utilised to stimulate the discussion. Data were audio-recorded and transcribed verbatim. A cross sectional thematic analysis approach was followed in data analysis using MAXQDA software.

Results: Analysis identified six main themes; the impact on affected children, the impact on parents, the life course of the disease, coping mechanisms, treatment need, and experience of treatment. Parents highlighted the emotional and psychosocial challenges experienced by families from the time of teeth eruption and believed that young children would be aware of their affected teeth. A feeling of guilt was evident among fathers affected by the same condition. Most parents sought dental treatment when child was approaching school age due to worries of bullying at school. Dental treatment was viewed as a way of achieving normality, protecting children from feeling different and/ or experiencing negative social reactions and as a way of maintaining structure and function. Whether affected by same condition or not, parents appeared to rely solely on the professional advice of the paediatric dentist in making all treatment related decisions.

Conclusions: The personal experience of parents affected by AI/DI plays a pivotal role in parent's judgements of their children's teeth and perceived need for dental treatment. The paediatric dentist has a powerful influence on parental decisions and must acknowledge the parents' perspectives when discussing options for treatment.

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1 Background

1.1 Introduction

Amelogenesis Imperfecta (AI) is a hereditary developmental disorder which affects the structure and clinical appearance of enamel of all or nearly all the teeth in a more or less equal manner (Crawford et al., 2007). The prevalence rate has been reported to range from approximately 1.4:1000 to 1:16 000 depending on the population studied (Seow, 2014). It is a genetic defect and can be inherited as autosomal dominant, recessive, x-linked or sporadic.

Depending on the stage of enamel formation that is affected, AI affected teeth can present as hypoplastic (thin enamel, surface pitting or vertical grooving), hypomineralised/hypocalcified (soft enamel), or hypomature (Mottled/ opaque /altered enamel translucency). Both hypomineralisation and hypoplastic defects can coexist in the same patient and even the same tooth (Crawford et al., 2007). Diagnosis is based on a combination of family history, pedigree plotting and clinical findings. Genetic tools can allow for more precise diagnosis, however are currently limited for research use (Crawford et al., 2007). The most commonly used classification was developed by Witkop and classifies AI into four types based on clinical appearance; hypoplastic, hypomaturation, hypocalcified and hypomaturation-hypoplastic with taurodontism (Witkop, 1988). AI affected teeth are often discoloured, creating aesthetic problems in addition to sensitivity, tooth wear, loss of vertical dimension of occlusion, increased calculus formation and the need for lifelong dental care.

Dentinogenesis imperfecta (DI) is the most common type of hereditary developmental disorders of dentine with an estimated incidence ranging between 1:6000 and 1:8000 (Seow, 2014). It can be inherited as autosomal dominant or autosomal recessive. DI affected teeth vary in colour and can exhibit grey, blue, yellow, brown and opalescent discolouration. Tooth discolouration tends to be more marked in the primary dentition due to thinner primary teeth enamel. Also, DI affected teeth may become altered in shape due to wear and attrition.

Parents present to the dentist with concerns about the appearance of their child's affected teeth. The paediatric dentist plays a substantial role in early diagnosis, preventative dental care and management early in the life course of both developmental anomalies. The aims of management are to improve the aesthetics, function, reduce sensitivity of the affected dentition and protect against tooth wear.

The paediatric dentist is also responsible to promote the child's positive dental attitude and prepare the young patient for life long dental treatment.

Several restorative options are considered when addressing aesthetic and functional issues with primary teeth affected by AI or DI including polishing, microabrasion, composite strip crowns, zirconia crowns, Stainless Steel Crowns (SSCs) and overdentures. There is a lack of evidence of superiority of one treatment option over another due to a lack of good quality evidence assessing the success rates of restorative treatments for children and adolescents with AI in terms of patient satisfaction and function (Dashash et al., 2008). Operator's preferences and aesthetic demands by parents are among the most important factors that affect the decision on which restorative option is chosen (Waggoner, 2015).

Parents of young children are involved in the clinical decision-making process and are responsible for facilitating the child's access to dental treatment. Due to the complexity of treatment, children's young age and anxiety in dental settings, restorative treatment for young children may incorporate inhalation sedation or general anaesthesia. A better understanding of both mothers' and fathers' attitudes, beliefs, values, and expectations regarding aesthetics and treatment need is required to facilitate increased communication between parents and the dental provider.

1.2 Literature Review

1.2.1 Facial Appearance and Social Perception

Facial appearance has been of interest throughout the centuries. There has been a worldwide increase in social concerns about aesthetics with a media driven emphasis on appearance. Facial aesthetics plays a substantial role in physical attractiveness and has been described as ‘the most consistent and compelling determinant of self and social perceptions and attributions’ (Albino et al., 1990).

The concept of an association between physical and facial attractiveness and attribution of positive characteristics was first introduced in the psychology literature by Dion in the 1970s when he proposed the physical attractiveness stereotype, ‘what is beautiful is good’, (Dion et al., 1972). This concept has been supported by several studies and confirmed by several reviews and meta- analyses (Baldwin, 1980; Eagly et al., 1991; Feingold, 1992; Langlois et al., 2000). This effect of facial attractiveness has been demonstrated to extend beyond the initial impressions to actual interactions with known people, with attractive children and adults regardless of gender being judged and treated more positively than unattractive people and thus exhibit more positive behaviours and traits than unattractive people (Langlois et al., 2000; Baldwin, 1980).

A significant association between social judgements of popularity and sociability and facial attractiveness is evident in the literature, however, the extent to which attractiveness affects the intellectual/academic judgements of children and occupational judgements of adults’ remains controversial (Baldwin, 1980; Langlois et al., 2000; Feingold, 1992).

1.2.2 Social and Psychosocial Impact of Facial Disfigurement

Visible facial disfigurement, whether congenital or acquired, can impact significantly on the psychosocial wellbeing of children and adolescents (Rumsey and Harcourt, 2007; Baldwin, 1980). The most commonly reported difficulties are related to negative self-

perceptions, development of self-concept and negative social reactions (Rumsey and Harcourt, 2007; Marik and Hoag, 2012).

Rumsey (2007) has reported four key developmental challenges for patients with facial disfigurement in early childhood including: developing a secure attachment in early parent-child interactions, development of self-perceptions that starts as early as two or three years of age and social relationships and responses from others especially when changing social groups (Rumsey and Harcourt, 2007).

The developmental changes in late childhood and adolescence have been demonstrated to further increase the focus on appearance. Around this age, social comparison (how we feel we compare with others) becomes a significant factor in the development of children's self-image/ concept and that creates considerable challenges the self-perception of unfavourable appearance (Baldwin, 1980; Rumsey and Harcourt, 2007; Marik and Hoag, 2012).

Most of the existing research on the effect of facial differences on the self-concept of children and adolescents has focused on patients with craniofacial differences, especially cleft lip and palate (CL(P)) (Marik and Hoag, 2012; Rumsey and Harcourt, 2007). A systematic review of the literature on the psychosocial impact of cleft lip and palate has revealed a lack of consensus, with many studies providing contradictory evidence on the psychosocial consequences of CL(P) (Hunt et al., 2005). Some studies reported significant behavioural, emotional, and interpersonal implications associated with facial impairment (Albino et al., 1990; Hunt et al., 2006) while other studies reported an average to above average self-concept (Slifer et al., 2003; Krueckeberg et al., 1993) and no significant difference in self-esteem when compared to unaffected children (Hunt et al., 2005; Walters, 1997). Hunt concluded that the majority of children and adults with CL(P) do not appear to experience major psychosocial problems, though some individuals might experience dissatisfaction with appearance, depression, anxiety and some behavioural problems (Hunt et al., 2005).

Facial differences are quickly perceived in social discourse, therefore children with facial differences are at high risk for experiencing stigmatising behaviours, such as staring, startled reactions, teasing and expressions of pity (Masnari et al., 2012; Strauss et al.,

2007). In a cross-sectional multisite study in the US, over 35% of adolescents with facial differences experienced stigmatising behaviour (Strauss et al., 2007).

A history of teasing was found to be the most significant predictor of psychosocial problems for children when rated by both children and parents. Children who had been teased have been reported to experience increased anxiety, less satisfaction with appearance and greater behavioural problems (Hunt et al., 2006). Despite these challenges, some affected children develop resilience and positive psychological and social adaptation to their altered appearance influenced by the degree of their emotional functioning (degree of depression and/or anxiety), satisfaction with appearance and the child's subjective report and evaluation of other people's reactions (teasing, stating, and questioning), regardless of the visibility/ severity of deformity. Resilient children place less importance on other people's evaluation of their appearance, facilitating better adjustment. Other factors like temperament, social skills, social acceptance and support were also suggested to promote positive psychosocial adjustment (Feragen, 2009; Juneja et al., 2016; Strauss, 2001; Strauss and Fenson, 2005).

The perception and experience of parents of children with CL (P) have been investigated mainly in cross-sectional surveys that have focused on the mothers' views in the early stages of child's life. Both cross-sectional surveys and some qualitative studies have documented parent's feelings of anger and worry. In-depth qualitative studies have revealed parental concerns and worries in relation to the social reactions to altered facial appearance. Feelings of parental guilt, self-blame and associated anxiety were also reported (Nelson et al., 2012a). Parents appeared to manage their emotional 'strain' by cognitively reframing their circumstances (change the way they look at them) and/or pursuing cleft treatment (Nelson et al., 2012a; Nelson et al., 2012b). A perceived parental 'moral' obligation to be 'good' parents by pursuing the elective 'normalising' treatments, particularly surgeries for their children was also reported. Such treatments were viewed by parents as a way of facilitating their child's social inclusion (Nelson et al., 2012c).

1.2.3 Social and Psychosocial Impact of Dental Appearance

Dental attractiveness plays a substantial role in determining facial attractiveness, particularly visible teeth in a smile. The mouth is central to social interactions as shown by studies of eye movement in a face-face situation which revealed that the person's eyes and mouth area are primarily scanned, with little time spent in observation of other features (Miller, 1970). Teeth shape, size, alignment, proportion, number and colour contribute to dentofacial appearance. Alterations in any one of these factors may affect facial attractiveness and influence how people are perceived by society and how they perceive themselves.

The individual's age, race, gender, personality, preferences and established guidelines are usually taken into consideration when restoring the anterior teeth. For an ideal aesthetic outcome, clinical guidance advises the central incisor to be the dominant element in the anterior dental composition, with a mesial axial inclination, a width/length ratio between 0.75-0.8, 4mm overbite, 2mm overjet, and maxillary incisal edges parallel to the curvature of lower lip and in contact with its mucosal surface (Ahmad, 2005; Holyoak, 2013). These values were mostly based on studies involving white subjects and found to vary among different ethnic groups (Tsukiyama et al., 2012).

Several theories were postulated to provide criteria for evaluating the tooth-tooth proportions for an aesthetic smile. The traditional concept of golden proportion (1:1.618) was introduced to restorative dentistry by Levin in 1970s when he proposed a theoretical correlation between the successive widths of maxillary anterior teeth and dental aesthetics (Levin, 1978). Later, the "recurring aesthetic dental proportion" concept was introduced, stating that continuous proportion -not necessarily limited to the golden proportion, as long as it remains consistent, proceeding distally in the arch must be used by clinicians when designing an aesthetic smile (Ward, 2001). In parallel, other authors advocated applying harmony (that relies on similar repeated proportions) and symmetry principles irrespective of actual teeth size or ratio (Ahmad, 2005).

In recent years, several studies as well as systematic reviews were carried out to assess laypeople's perceptions and preferences regarding frontal dentofacial aesthetics of what is socially perceived as an attractive smile in an attempt to improve the outcomes

of prosthodontic, orthodontic and periodontal treatment. These studies have identified several dental aesthetic parameters and defined threshold values of public acceptance for these parameters using adult observers (Del Monte et al., 2017; Witt and Flores-Mir, 2011; Parrini et al., 2016) as detailed in **Table 1-1**. Among the dental parameters are the smile arc, maxillary central incisor width/height ratio and shape, symmetry, buccal corridors, tooth angulations and tooth colour. Maxillary central incisors appeared to be the most important teeth in defining smile aesthetics.

A recent systematic review on children's and adolescents' perceptions of smile aesthetics concluded that unaltered smiles (with well aligned teeth) were always associated with significantly better children's evaluation when compared with altered smiles (crowding, diastema or proclined incisors). No studies have qualitatively or quantitatively analysed thresholds values for aesthetic smile characteristics from children's perspective (Rossini et al., 2016).




Dental Variable	Threshold/Ideal Value (Range of Acceptability)	
Diastema	Ideal value: 0 Cut-off value: 1.5 (0-2) mm	
Tooth size and shape	Crown-length discrepancy range: 2-4 mm.	
	Maxillary central incisor width: Length: 80% (75%-85%)	
	Tapered incisors: the most attractive shape, square shape more preferred for females with ovoid facial contours.	
	Laypeople prefer unworn dentition. Laypeople prefer small teeth for females and large teeth for males. Laypeople prefer square-round incisors to square incisors and flat canines (when paired with round incisors)	
Maxillary incisor vertical position	Lateral incisors' edge position (maxillary to the central incisor plane) Ideal value: 1.2 (1, 1-2) mm	
	Vertical discrepancy between central incisors: 0 (0- 0.6)mm	
	Vertical discrepancy between lateral incisors: 0 (0-1)mm	
Maxillary central- lateral width proportion	(50% -74%)	
 (Del Monte et al., 2017)	 (Witt and Flores-Mir, 2011)	 (Parrini et al., 2016)

Table 1-1 Dental variables and laypeople acceptance of an aesthetic smile.




Dental Variable	Threshold/Ideal Value (Range of Acceptability)	
Maxillary central incisors mesiodistal angulation	Ideal value: 0 Acceptable range: (0- 8.6) degrees.	
Maxillary central incisors labiolingual inclination	Ideal angle between the Incisor inclination and the horizontal: 93 degrees.	
Maxillary anterior contact area	Ideal: 50%	
Midline discrepancy	Ideal value: 0 Cut-off value: 2.38 (1.83-2.92) mm	
Buccal corridors	Ideal value: 11.5 (5-16 mm/17% total smile)	
Occlusal plane cant	Ideal value: 0-4.	
Smile arc	Ideally the lower lip profile.	
Overbite	2-5 mm	
		
(Del Monte et al., 2017)	(Witt and Flores-Mir, 2011)	(Parrini et al., 2016)

Table 1-1 Continued

The impact of dentofacial attractiveness on social judgements of personal characteristics, social interactions and thus the psychosocial well-being of children (Baldwin, 1980; Shaw, 1981; Shaw et al., 1980; Klages and Zentner, 2007) and adults (Feng et al., 2001; Klages and Zentner, 2007; Baldwin, 1980) has been well documented.

Shaw conducted a cross sectional study in which 42 children and 42 adults were shown standardised photographs of a boy's and a girl's face exhibiting variable dental arrangements (normal incisors, prominent incisors, missing lateral incisors, and unilateral cleft lip). He found that children (11-13 years) with normal dentofacial appearance are judged by other children as more attractive, more desirable as friends, more intelligent, and less inclined to aggression than those with impaired dentofacial

appearance. From the adult perspective, the results were similar with regard to physical attractiveness and desirability as a friend, but not for aggressiveness (Shaw, 1981).

The concept of facial aesthetics has been of primary research interest and concern in orthodontics. The physical, social, and psychological consequences of malocclusion and its influence on the oral health related quality of life (OHRQoL) of affected individuals have been frequently investigated, though focused on the permanent dentition (Ukra et al., 2013; Scapini et al., 2012; Sardenberg et al., 2012; Peres et al., 2009).

Several cross-sectional observational studies reported a negative impact of anterior malocclusion on OHRQOL in children and adolescents, predominantly in the dimensions of emotional and social wellbeing and irrespective of OHRQOL measures used (Dimberg et al., 2014). **Table 1-2** illustrates some of these studies. The quality of evidence supporting this negative impact was judged until recently to be "albeit modest" due to its predominantly cross sectional component (Liu et al., 2009). More recent cross sectional studies of higher quality were conducted and were included in a systematic review published five years later (Dimberg et al., 2014). According to this review, there is a growing higher quality evidence to support a negative impact of anterior malocclusion on OHRQL in children and adolescents (Dimberg et al., 2014).

Kragt and co-workers performed a meta-analysis (using all six studies included in the Dimberg review in addition to 34 cross sectional studies). All the included studies had control groups with good dental features or lesser malocclusions and subgrouped the participants based on their orthodontic treatment need. He concluded that children (age 8-18 years) with malocclusion were 1.74 times more likely to have a detrimental impact on OHRQOL when compared to children without malocclusion (Kragt et al., 2016).

Another systematic review reported facial attractiveness as the main motivational factor for parents and children seeking orthodontic treatment. Parents wanted their children to look nice and worried that they may be accused of neglecting parental duties if treatment was not pursued (Samsonyanová and Broukal, 2014). Early orthodontic treatment is perceived to yield a great psychosocial benefit. As a consequence, the request for orthodontic treatment has been increasing.

Author/ year	Country	Participants	Important findings
(Ukra et al., 2013)	New Zealand	783 schoolchildren (411 boys and 372 girls) Age: 12-13 years	Malocclusion appeared to have a negative impact on OHRQOL, subscales of emotional and social wellbeing.
(Scapini et al., 2012)	Brazil	632 schoolchildren (270 boys and 362 girls) Age: 11-14 years	Increased severity of malocclusions was associated with higher impact on OHRQOL, subscales of emotional and social wellbeing.
(Sardenberg et al., 2012)	Brazil	1204 schoolchildren Age: 8-10 years	Schoolchildren with malocclusion were 1.3 times more likely to experience a negative impact on OHRQOL than those without malocclusion.
(Peres et al., 2009)	Brazil	339 children nested in a birth cohort (182 boys, 157 girls) Age: 12 years	Incisal crowding was related to worse OHRQOL.

Table 1-2 Studies on negative impact of anterior malocclusion on OHRQOL in children and adolescents.

From a different perspective and irrespective of demographic characteristics, tooth colour was reported to be the most important variable in predicting dental attractiveness of adults (Dunn et al., 1996). Adults are more likely to make negative judgments of social and intellectual competence based on dental appearance. Individuals with whitened teeth are judged more positively in terms of social competence, intellectual ability, psychological adjustment, and relationship satisfaction (Kershaw et al., 2008).

Individuals with visible dental caries are perceived as less socially competent, less intellectually competent and less psychologically adjusted than individuals with visible healthy anterior teeth (Feng et al., 2001; Newton et al., 2003). In the view of that, tooth colour plays a pivotal role in motivating individuals to seek dental care and this can be noted with the increased public requests for teeth whitening.

In young children, even though teeth discoloration may pass unnoticed when the focus is on the whole facial appearance (Newton and Minhas, 2005; Soares et al., 2015), when focus is drawn to the mouth, discoloration was significantly associated with negative social perceptions by preschool and young children (6-10 years) even more than tooth loss (Soares et al., 2015; Vlok et al., 2011).

Dental trauma is one of the main causes of altered anterior aesthetics which can range from discoloration and crown fracture to complete tooth loss. These alterations are noticed by other children, can impact on social interactions and can have a negative psychosocial impact on affected children, however, most of the studies in this area have focused on traumatised permanent teeth (Vlok et al., 2011; Soares et al., 2015; Rodd et al., 2010).

In one study, children (age 11-12, 14-15 year olds) were shown standardised photos of other children with normal and visible permanent incisor trauma. Younger children judged those with visible incisor trauma more negatively while older children judged them more positively suggesting that children with visible trauma can experience more social problems at younger age. Self-monitoring behaviour at older age was suggested to explain the difference in social judgments, where children will modify their responses to what they consider more socially acceptable. A further gender implication was noted as females of both age groups gave more favourable overall rating (Rodd et al, 2010).

A different perceived aesthetic impact of various dental injuries among different age groups was noted in another study with tooth discoloration due to trauma, complicated crown fracture and lateral luxation being less acceptable to younger age group (6-10 year old) when compared to adolescents (11-17 year old) and adults (18-24 year old), with avulsion and uncomplicated crown fracture being more acceptable. This is probably

due to that younger children are more familiar with missing and partially erupted teeth in the phase of early mixed dentition (Vlok et al., 2011).

The only study to question preschool children was a recent cross sectional study that investigated the social perception of Brazilian preschool children of altered aesthetic appearance of traumatised primary anterior teeth and found a significant association between altered aesthetics and negative social and self-perception in children (Soares et al., 2015).

The available evidence on dentofacial attractiveness demonstrates the importance of dental aesthetics in social context. Negative social judgments can be made based on dental aesthetics and a stereotype of a social handicap can be attributed to individuals with impaired dental aesthetics (Klages and Zentner, 2007).

Dentofacial aesthetics contributes to psychosocial wellbeing of both children and adults, however, the evidence related to social perception and attribution of aesthetically impaired primary teeth is limited to three studies. **Table 1-3** illustrates studies that investigated the perception of aesthetically impaired primary teeth.

Author/ year	Country	Participants	Causes of aesthetic defects	Methods	Important Findings
(Woo et al., 2005).	Washington	103 dentists. 67 Paediatric dentists. 97 parents (low income). 112 parents (high income).	- Caries. - Caries with visible sinus tract. - Dark incisor.	- Dentist/ Parent specific questionnaire presenting photos and periapical x-rays of asymptomatic three-year-old child with aesthetic defects of upper primary incisors.	- All groups recognised carious teeth with and without sinus tract as unhealthy and unattractive. - For the dark incisor, 83% of dentists favoured no treatment, as compared to 71% of parents who thought treatment was needed (P<.001).
(Holan et al., 2009).	Israel	362 parents of 294 children aged 1-6 years: 211 with aesthetic defect (A). 83 without aesthetic defect (NA).	- Trauma (discolouration, fracture, infraoccluded, missing, ectopic alignment). - Caries. - Malocclusion (crowding, open bite, spaced teeth).	- Clinical exam of the child's primary incisor. - Questionnaire for parents concerning their own perception of their children teeth.	- 73% of parents of (A) recognised an aesthetic problem versus 17% of parents of (NA). - 87% (A): 97% (NA) advocated dental treatment even if chances of success were only 50%.
(Soares et al., 2015).	Brazil	431 four-five year old children.	- Discolouration due to trauma. - Crown fracture. - Missing tooth due to trauma.	- Questionnaire presenting photos of children with altered aesthetic defects and photos of mouths with altered Aesthetic defect. - Face to face Interviews with children.	A significant association between both negative social and self-perceptions and altered dental aesthetics.

Table 1-3 Studies on the perception of aesthetically impaired primary teeth.

1.2.4 Social and Psychosocial Impact of Developmental Defects of Enamel (DDE)

Developmental enamel defects result from altered enamel formation due to a variety of hereditary and environmental factors that can affect both primary and permanent dentition. It can present as enamel hypomineralisation (brown/creamy/yellow/white opacities), hypomaturation (altered enamel translucency) or enamel hypoplasia (pits/grooves/thin or missing enamel). These visible differences to normal dental appearance can create aesthetic problems in addition to problems of wear and sensitivity.

Several studies with variable methodologies have investigated the aesthetic perception and psychosocial impact of developmental enamel defects affecting the anterior permanent teeth with a focus on adolescents and young adults (Sujak et al., 2004; Marshman et al., 2009; Chankanka et al., 2010; Craig et al., 2015). A qualitative study involving adolescents with permanent incisors affected by a variable degree of developmental enamel defects in UK has revealed that adolescents can make judgements based on teeth appearance and DDE can be a subject of teasing. The greatest impact was on individuals whose sense of self was more contingent on appearance and who depended on perceived approval from their peers about their appearance (Marshman et al., 2009).

A recent study utilising a mixed qualitative quantitative methodology was conducted in the UK involving 547 adolescents (11-15 years old) from areas of different socioeconomic status. Findings from two focus groups with a total of 12 children were used to develop a social attribute questionnaire. Using photographs, the total attribute score was significantly lower for subjects with enamel defects suggesting that adolescents can make negative psychosocial judgements on the basis of altered enamel appearance. Subjects with enamel defects were perceived as being lazy and not caring about appearance (Craig et al., 2015). This can result in personal dissatisfaction with appearance and can have a significant impact on affected individuals. In another study, children (7-16 years) with DDE reported being worried and embarrassed about their

dental appearance and aesthetic management of affected permanent incisors made them happier and more confident (Rodd et al., 2011).

The impact of DDE on affected people seems to be variable among populations. The psychosocial impact on Malaysian sixteen year old school children was found to be generally low. In that population, affected females, adolescents dissatisfied with their overall health, and those who are subjected to teasing were more likely to express dissatisfaction with altered teeth colour due to DDE (Sujak et al., 2004).

Parental aesthetic perception of younger children (8 years old) affected by fluorosis was assessed in several areas of the world. Parents' dissatisfaction with the aesthetic appearance of their children's teeth was greater with increased severity of fluorosis in terms of both colour and enamel pitting (Chankanka et al., 2010; Sigurjóns et al., 2004).

Parental and childrens' perception of MIH (Molar Incisor Hypomineralisation) was a major area of interest for a recent case-control study (Leal et al., 2017). In this study, 262 Brazilian school children (age 7-13 years) and their parents were interviewed using the Child and Parents Questionnaires on dental appearance. Both mothers and children were concerned about enamel opacities in terms of appearance and discoloration regardless of the severity and even when the incisors were not affected.

Studies which have investigated the aesthetic perception and psychological impact on preschool children with DDE are limited. A recent population based cross sectional study have investigated the oral health related quality of life of preschool children from both children and parent perspective and did not find a significant association between DDE and negative impact on the quality of life of young Brazilian children (Corrêa-Faria et al., 2016).

1.2.5 Social and Psychosocial Impact of AI and DI

Several studies have examined the impact of Amelogenesis Imperfecta on the patients' quality of life as shown in **Table 1-4** (Poulsen et al., 2008 Coffield et al., 2005; Hashem et al., 2013; Lundgren et al., 2015; Parekh et al., 2014; Sneller et al., 2014).

A cross sectional controlled study by Coffield and co-workers in 2005 demonstrated a significant psychosocial impact of AI on affected adults in terms of social interaction, anxiety, self-image, self-esteem and self-perceived quality of life. Affected individuals were found to be unhappier with their dental appearance with 93% reporting being teased about their dental appearance. Moreover, affected individuals had higher levels of social avoidance and distress compared to other unaffected family members who served as controls (Coffield et al., 2005). Similarly, a negative psychosocial impact on AI affected adults and adolescents was reported in Ireland and Sweden, however, the self-esteem was not significantly affected (Hashem et al., 2013; Lundgren et al., 2015).

A recent study with mixed qualitative and quantitative research methodology have investigated the impact of AI on children and adolescents (10-16 years) in the UK. In depth interviews of seven AI patients were utilised to develop a questionnaire that was completed by 40 AI patients (10-16 years). This study revealed a huge aesthetic concern in affected children. The majority of children started to notice the altered dental appearance at very young age (around 6 years old) and 50% reported being teased. Furthermore, the most important reason for undergoing treatment was to improve teeth colour (90%). The study concluded that there was a detrimental aesthetic, social and psychosocial impact of AI on children and adolescents (Parekh et al., 2014).

In contrast, in another exploratory qualitative study in the same region, adolescents (11-16 years) denied any negative psychological impact of AI, however only four adolescents attended this focus group discussion. The authors suggested three explanations to this contradictory finding; either that is a true feeling, that they are used to living with AI, or that these responses were just a function of the study context. The adolescents' parents were involved in different focus group and expressed a different opinion. Parents described how their children used to cover their mouth in certain social situations. In addition, this study provided preliminary evidence of potential impact of

having an AI affected child on parents. Parents reported that having a child with AI 'gets them down'. Parents expressed a need for support groups held by professionals. Parents needed information-based support as well as emotional and practical support to help them in caring of their affected children. However, all parents were not affected (Sneller et al., 2014).

Collectively, studies to date have indicated variable psychosocial impact of AI on affected adolescents and adults, however, studies utilising qualitative approaches to examine perceptions in-depth have been relatively rare. Surprisingly, the psychosocial impact of Dentinogenesis Imperfecta (DI) on affected people and their perceptions of the condition have never been investigated. There is a paucity of both quantitative and qualitative research which has explored parental perception of young children affected by AI or DI. No study has assessed the perception of parents whose children have primary teeth affected by AI/DI, particularly where many of these parents may be affected themselves or have a family history of these dental anomalies. Parents affected by either condition may experience distress and challenges, and may have different perceptions than parents who are unaffected. These issues have not been explored in the research literature.

Author / year	Country	Participants	Methods	Important findings
(Coffield et al., 2005)	North Carolina	30 AI: 29 without AI Adults (14 years and above)	A questionnaire and a number of psychometric scales.	- Subjects with AI (n = 30) had higher levels of social avoidance and distress, higher levels of dysfunction, discomfort and disability attributable to their oral condition compared with subjects without AI (n = 29).
(Hashem et al., 2013)	Ireland	27 AI: 27 without AI Age 18-45 year.	Oral health impact profile and Rosenberg Self Esteem Scale.	-AI has marked negative impacts on the oral health related quality of life of affected patients under 4 of 7 domains (psychological discomfort, physical disability, psychological disability, social disability). -Self-esteem was not significantly affected. -AI has a more negative impact than hypodontia.
(Lundgren et al., 2015)	Sweden	69 patients with AI, aged 6–25 year: 33 males and 36 females (mean age 14.5 ± 4.3); Healthy controls (n = 80).	Three questionnaires measuring OHRQoL (OHIP-14), dental fear (CFSS-DS), and dental beliefs (DBS-R) before and after treatment.	-Adolescents and young adults with AI reported significantly lower oral health related quality of life than healthy controls (mainly orofacial appearance and orofacial pain) -Two years after crown therapy, OHRQoL was improved significantly (psychosocial impact and orofacial impact). -Therapy did not increase dental fear or negative attitudes toward dental treatment.

Table 1-4 The impact of AI on the patient's quality of life studies.

Author / year	Country	Participants	Methods	Important findings
(Parekh et al., 2014)	UK	7 AI patients interviewed. 40 AI patients completed questionnaire. Age: 13-16 years.	In depth interviewing to develop an AI specific questionnaire.	Children and adolescents exhibited concerns about aesthetics, comments by others and self-consciousness associated with this.
(Sneller et al., 2014)	UK	1 focus group: 4 AI adolescents (11-16 years) 1 focus group: their parents(4)	Focus groups	-The adolescents reported that they are 'not bothered' by having AI. -Parents acknowledged that having a child affected by AI sometimes 'gets them down'. -Parents believed that 'deep down it bothers the child too'. -Parents need further informational, emotional and practical support.

Table 1-4 Continued

1.3 Aims and Objectives

1.3.1 Study Aims

This study aimed to investigate and explore parental attitudes, beliefs, values and expectations regarding aesthetics and treatment need of children in primary dentition affected by AI and DI.

1.3.2 Study Objectives

- Qualitatively report the aesthetic, social and emotional perception of primary teeth affected with AI or DI by parents of affected children.
- Elucidate the issues that influence parental perceptions and treatment decisions.

2 Methods

2.1 Introduction

This study was a qualitative study that took a descriptive approach to investigate and explore parental attitudes, beliefs, values, and expectations of both mothers and fathers regarding aesthetics and treatment need of children in the primary dentition affected by AI and DI.

Qualitative research is a valuable approach that provides in-depth insight into people's personal perspectives, beliefs, attitudes and experiences. It is commonly used to explore, interpret and gain deeper understanding of the social world of research participants especially for aspects where little is known or understood (Stewart et al., 2008; Ritchie et al., 2013).

Focus groups were used for data collection utilising smile photographs of primary teeth affected by AI and DI. Focus groups are a commonly used research tool in qualitative research. It is defined as a carefully planned group discussion that is guided, monitored and recorded by a moderator, with the aim of generating information on collective views and the influences behind these views (Stewart et al., 2008; Smithson, 2000). The group participants interact with each other and share their views and experiences about a certain topic and data are generated through their interaction. Focus groups offer insights into a wide variety of participants' opinions and perceptions, and in particular provides different forms of interactions between participants including argumentative behaviours and spontaneous responses. These interactions result in a depth of dialogue of multiple participants simultaneously and highlights arguments and conflicts in a quicker and more convenient way when compared to individual interviews. However, some participants may find a focus group intimidating and feel under pressure to agree with dominant or socially acceptable views which makes the data generated from focus groups contingent on the skills of the moderator to deal with dominant voices that might override the discussion as well as the irrelevant discussions which may distract from the main focus (Smithson, 2000; Onwuegbuzie et al., 2009).

The photographs were standardised so that only the smile with visible teeth were shown and magnified to a similar size intending to clearly show the area of interest and limit the influence of other extraoral confounding factors such as facial form or hair colour

on parents' perceptions. Parents' visualisation of these dental anomalies is assisted by the use of the photographs, stimulating discussion, and eliciting parents' opinions and perceptions. However, it is known that other factors such as facial characteristics and social interaction with individuals influences individual perceptions and judgments in real life, which makes the photographs not entirely realistic.

2.2 Ethical Approval

Ethical approval was granted for the study by the Ethics Committee of the Faculty of Health Sciences in Trinity College, Dublin (Reference number: 160603) (**Appendix 1 and 2**).

Initially the research aim was to assess parents' perception of primary teeth affected by AI, however the number of potential participants was insufficient to proceed. This generated an amendment to include parents of children affected by DI since both are genetic dental anomalies that affect both primary and permanent dentition and can influence aesthetics. In addition, affected individuals with either conditions may benefit from early dental intervention and are likely to require lifelong dental care.

2.3 Sampling and Setting

This study was conducted in the Dublin Dental University Hospital (DDUH), which is a national referral centre for children with developmental dental anomalies. All patients diagnosed with AI or DI are recorded on a specific database since 2002 (Special Dental Need Database (SPNS)). Dental treatment for patients on this database is provided at no cost to the patient. Various dental interventions are provided for children and include the use of both pharmacological and non-pharmacological behavioural management techniques.

Eligible parents were approached through the gatekeeper (AB) and those who agreed to participate were divided into two focus groups; one including mothers and one including fathers as this was assumed to facilitate disclosure and help in shaping effective group dynamics. Purposive sampling is a sampling approach that choose research participants based on their characteristics which would enable detailed

exploration and understanding of the research topic (Ritchie et al., 2013). Purposive sampling was applied to include parents of boys and girls affected by either AI or DI with a wide range of age (above 35 and below 35 years of age), and where different levels of dental intervention has been provided for affected children. In addition, purposive sampling was utilised to ensure that each focus group included parents affected by AI/DI.

Including participants with different characteristics and experiences permitted covering the entire range of experiences and aids in understanding the impact of the characteristics (Morse, 2000; Coyne, 1997; Ritchie et al., 2013).

In qualitative research it is often difficult to know exactly how many participants are required in advance, however, the focus groups were planned to comprise of 6-12 participants as is recommended to provide appropriate group dynamics (Fusch and Ness, 2015). The total number of focus groups was determined by theoretical saturation. Theoretical saturation is the point in data collection that is achieved when no more new themes emerge from the latest collected data (Mack et al., 2005; Fusch and Ness, 2015). That was applied using the following criteria:

- Agreement among the research team on the overall quality of data obtained from each focus group and the quality of data obtained from each subgroup of participants in light of the research aims and objectives.
- Comparison between the themes emerged from focus groups until no more new themes were identified from the latest focus group data.

2.4 Inclusion and Exclusion Criteria for The Focus Group

Inclusion Criteria

- Parents of children 10 years old or under referred to the DDUH.
- Agreed diagnosis of AI /DI affecting their primary dentition.
- Able to speak English.
- Able to provide consent.

Exclusion Criteria

- Parents who could not speak English.
- Parents of children with full permanent dentitions.
- Parents of children with altered anterior teeth aesthetics due to other conditions (e.g. caries, trauma, hypodontia).

2.5 Training

The main researcher (AA) attended a training course in qualitative research methodology by the Social Research Association in Edinburgh – Scotland (April 2016). The course was intended to teach researchers who are involved in qualitative research how to design a qualitative study, undertake focus groups, and how to analyse and interpret qualitative data. The course gave the researcher (AA) the opportunity of practicing interviews by interviewing other course participants.

2.6 Identification, Approaching and Recruitment of Participants

The electronic patient management system (SALUD) and the existing Special Dental Needs database (SPNS) held in the DDUH were used to identify participants who fulfilled the inclusion criteria. Potential participants were approached by the gatekeeper (AB) who is a member of the administration team in the Division of Public and Child Dental Health. The invitation letter, participant information leaflet, information sheet and consent form were prepared and sent by the gatekeeper (AB) to potential participants in one envelope by mail (**Appendices 3, 4, 5 and 6**).

Invitees were given seven days from receiving the study information to respond. Failure to respond within that time period generated a reminder phone-call by the gatekeeper. Once the invitee expressed interest in participation, they were asked to return the information sheet (**Appendix 6**) to the gatekeeper. In cases where invitee had questions or requested further clarifications, the researcher called the invitee and answered any questions.

Focus group sessions were flexible depending on the needs of the participants; mornings, afternoons, evenings and were arranged at a mutually agreeable time and date. The consent forms were signed on the basis of parents having understood the information sheets and asked subsequent questions. Relevant demographic data including the parent's age, number and gender of affected children, type of dental anomaly, child's age at first dental visit, reasons behind first dental visit, child's age when receiving their first dental treatment, whether the parent is affected or not, and whether parents possess a Medical Card or not were all obtained before the focus group meeting so the impact of these various characteristics can be explored.

Parents were divided into two focus groups by gender, a group of fathers and another group for mothers intending to provide more comfortable group dynamics and aid the in depth exploration of the topic.

2.7 Topic Guide

A topic guide was formulated to elaborate on the aspects the researchers were interested in. The guide consisted of open ended questions that explored five key areas including:

- 1. Aesthetics and attractiveness.**
- 2. Impact on the child.**
- 3. Impact on parents.**
- 4. Treatment need.**
- 5. Influence of dental interventions.**

The questions were informed by the literature regarding AI and DI as well as the literature regarding parental perception of dentofacial anomalies and clinical experience. The topic guide was formulated by the researcher (AA) and then was revised by the supervisor (AOC) and an independent experienced qualitative researcher (AD).

The questions were used as prompts during the focus group meeting but were not strictly adhered to. The topic guide allowed great flexibility for discussions to flow and the moderator allowed for spontaneous emergence of new issues where relevant. The topic guide was revised by the researcher (AA), the supervisor (AOC) and the experienced qualitative researcher (AD) after the first focus group in conjunction with

the researcher's field notes, with attention to some emergent ideas that were relevant to the topic but were not included in the topic guide. The information obtained from the first focus group were then used to update the topic guide and made it easier to explore with greater depth in the second focus group. **Appendix 7** outline the first focus group guide while **Appendix 8** outlines the second focus group guide.

Eleven standardised photographs of a smile showing teeth affected by varying severity of AI/DI and photographs of different aesthetic treatments (composite strip crowns, zirconia crowns, a mix of anterior composite/zirconia crowns and posterior stainless steel crowns) were utilised to stimulate the discussion and parents were encouraged to express their attitudes and feelings toward these photographs (**Appendix 9**).

2.8 Moderating The Focus Group

The focus groups were guided by an independent moderator (AD), who had extensive experience of in depth qualitative interviews and focus groups moderation. The main researcher (AA) provided the moderator with the topic guide, attended the focus group session and was the note keeper. The researcher indicated to the moderator when certain issues needed further probing. Neither the moderator nor the note keeper had any relationships with the participants. The approach used was inductive and nondirective to provide a greater opportunity for individual views to emerge through group interactions. Interjection was mainly for clarification that they had understood, to raise another topic for discussion, to enable another participant to speak or to steer the discussion back to the topic when tangential discussions were judged irrelevant.

2.9 Participation

To ensure that all participants of the group were allowed to give their opinion, the moderator encouraged the quieter speakers to give their opinions where appropriate and/or allowed them to agree or disagree and/or share their experiences on certain issues.

The moderator and note keeper were attentive for nonverbal cues of facial expressions or gestures to include other participants. Field notes were prepared before and after the focus group meeting and were used to record observations, non-verbal gestures and other significant events (**Appendix 7**). The moderator probed deeper to ensure issues were covered in depth and abstract themes were clarified.

2.10 Reflexivity

Reflexivity is a concept used in qualitative research as a mean of reducing bias, limiting the influence of the researcher's opinion and prior assumptions and the research process in shaping the collected data (Mays and Pope, 2007).

Efforts were made to imply social research policy and involve a non-dentist moderator, however, that was not feasible due to limited resources. Both the moderator and the researcher were dentists. The moderator was a specialist in Special Care Dentistry who treats adults only and had extensive experience of in depth qualitative interviews and focus group moderation. The researcher was a member of the paediatric dentistry team at the DDUH.

In order to limit the influence of the researcher and research process on the collected data, the experienced and independent moderator raised focus group questions based on the prepared topic guide in a neutral non leading way with the aim of being responsive to participants' views and exploring emerging ideas as they appeared.

Furthermore, to allow the participants to feel more at ease with their conversation, the moderator and the researcher were unknown to the participants, the moderator introduced herself as a person with no experience with AI and DI, dental jargon was avoided and the participants were assured there was no right or wrong answers. In addition, the location for focus group meeting was a quiet meeting room in the Dublin Dental University Hospital away from the dental working area to ensure privacy and provide a less stressful non-clinical environment.

The involvement of another independent qualitative researcher (BD) after data collection during thematic analysis was made to ensure analysis was based on parents' voices and to preclude paediatric dentists' imposed views constraint on the emergent data. This independent experienced qualitative researcher (BD) (who is another

specialist in Special Care Dentistry who treats adults only with extensive experience in qualitative research) and the supervisor (AOC) reviewed the data independently before the main themes were agreed to ensure both consistency through validity and comprehensiveness in coding and data analysis and to limit any researcher bias.

2.11 Arrangements for The Focus Group

The focus groups were held in a quiet meeting room in the DDUH, where the participants were provided with water, tea, coffee, sandwiches and biscuits. This introduced an air of informality and made participants feel welcome.

Participants were encouraged to take breaks through the session and replenish their drink and food. This proved to be an icebreaker and increased the informal and social nature of the group.

2.12 Recording

Data were audio-recorded on two separate recording systems to guarantee no loss of data. A trial of recording was conducted prior to participants' arrival. There was no videotaping of participants as it was not considered necessary. Once no more ideas emerged, the moderator and researcher thanked the participants and the recording was terminated. The researcher listened to the participants' feedback after the meeting and while guiding them to the DDUH main entrance. All feedback was very positive.

The fathers' focus group lasted for one hour and fifty minutes and the mothers' focus group lasted for one hour and twenty-four minutes.

2.13 Transcription

The audiotapes were transcribed verbatim through a recognised professional transcription services provider. A confidentiality agreement was signed in relation to storage of digital and text information. The participants were given different names in the transcribed data to ensure full confidentiality. All data were anonymised at source and stored on an encrypted and password protected computer. Once digital recordings

had been transcribed and the text documents received by the researcher all recorded data were destroyed by the transcriber.

2.14 Data Analysis

2.14.1 Use of MAXQDA Qualitative Data Analysis Software

After each focus group, a transcript of the study was formatted. These transcripts were uploaded as text documents to a qualitative data analysis software (MAXQDA). MAXQDA is the successor of winMAX and was developed and distributed by VERBI Software based in Berlin, Germany. The software was used by the researcher for thematic analysis by systematically evaluating and interpreting textual data.

In comparison with manual methods, computer assisted qualitative data analysis software aid the process of data analysis and facilitates organisation and display of data in more systematic and accessible way to all research team members (Ritchie et al., 2013). **Figure 2-1** shows the software interface.

The screenshot displays the MAXQDA software interface, which is used for qualitative data analysis. It is divided into several main sections:

- Document System (Top Left):** Labeled with a '1' in a box, it shows a list of documents. The selected document is 'ladies full focus group' with 478 segments. Other documents include 'fathers focus group part 2' (103 segments) and 'fathers focus group part 1' (190 segments). A 'Sets' folder is also visible with 0 segments.
- Code System (Bottom Left):** Labeled with a '2' in a box, it shows a list of codes. The selected code is 'feeling of guilt' with 12 segments. Other codes include 'timing of treatment' (26 segments), 'influence of child personality' (1 segment), 'willingness to pay' (15 segments), 'child protection' (53 segments), 'parents own experience' (33 segments), 'coping mechanisms' (40 segments), 'self consciousness' (23 segments), 'school, teasing and confidence' (27 segments), and 'Impact of early intervention' (11 segments). A search bar at the bottom of this panel shows 'Aesthetics and attractiveness'.
- Main Document View (Center):** Titled 'Document Browser: ladies full focus group', it displays the text of the selected document. A pink horizontal line highlights a segment, with a '3' in a box next to it. The highlighted text is: '1624 I: Absolutely, nothing, no. 1625 D: My husband would feel guilty about passing it on. 1626 I: Your husband would, would he? 1627 D: Very much so, and any time they've been to the dentist, like when (name) was crying the last time and I, he always rings me and says how was it? And I don't let him go to school, he kind of gets the day off 1628 I: Yeah. 1629 D: and we make a fuss of it, to try and make it 1630 Multiple speakers: Yeah. 1631 D: not be the ordeal 1632 I: Yeah. 1633 D: it is for him. 1634 I: Okay.' Below the text, a vertical axis shows the distribution of codes. The 'feeling of guilt' code is highlighted in pink, corresponding to the highlighted text segment. Other codes visible include '..coping mechanism', 'childs dental anxiety', and 'impact on parents'.
- Right Panel (Bottom Right):** Contains a search bar with the text 'Simple Coding Query (OR combination of codes)'.

Figure 2-1 Example of MAXQDA Interface

There are three main display windows that provide the following features:

- 1- A Document System Window offers options to import data (text documents, table documents, PDF files, video and audio files and images). Imported data files are accessed through this window.
- 2- A code System Window. This allows assignment of a code to a particular part of the document. Codes then are ordered into a hierarchical structure, a main code/ theme having several sub codes.
- 3- A Document Browser Window. This allows visualisation of the selected document, so codes can be assigned to text segments.

2.14.2 Thematic Analysis

A cross sectional thematic analysis approach was followed in analysing the data of all focus groups. It is a flexible qualitative research analysis approach for identifying and reporting patterns of meaning across the whole data set thus providing a rich, detailed interpretive, yet complex account of data (Braun and Clarke, 2006).

Analysis was an ongoing, iterative process that began in the early stages of data collection and continued throughout the study. It involved multiple steps as illustrated in **Figure 2-2**.

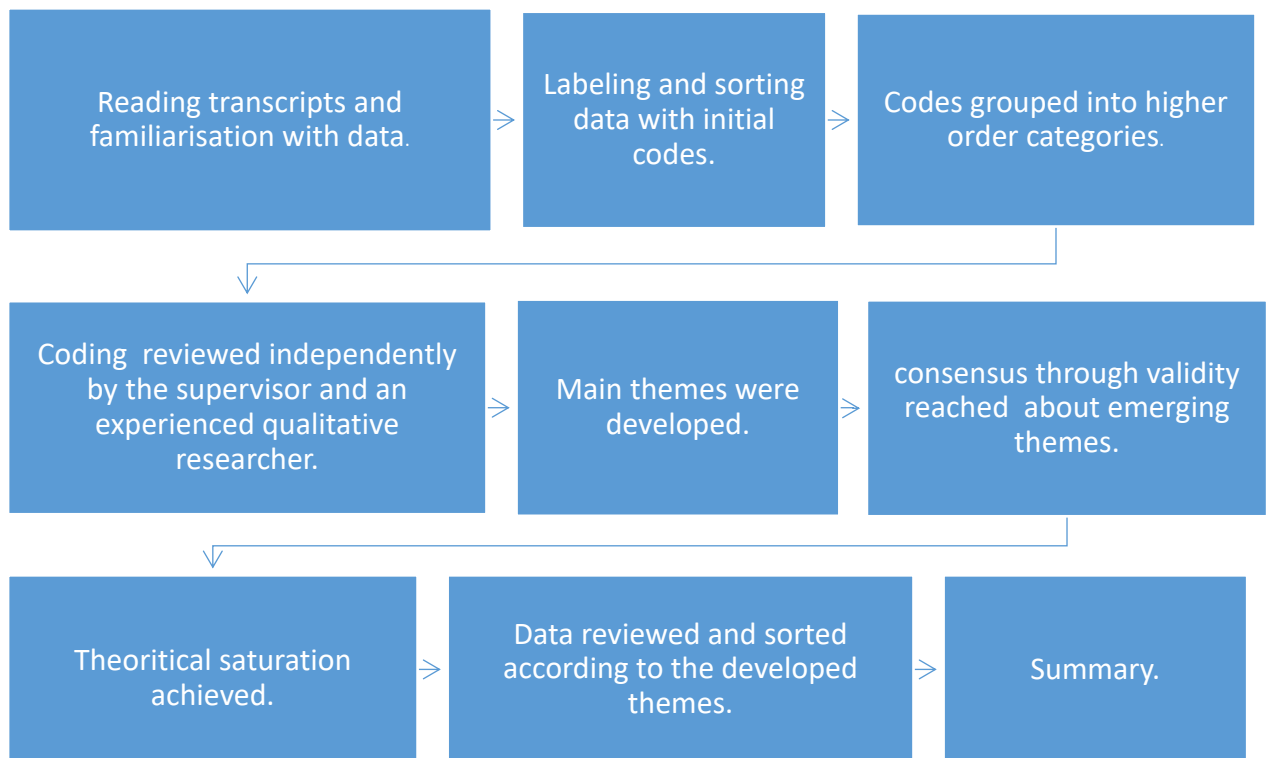


Figure 2-2 Steps for Thematic Analysis

2.14.2.1 Familiarisation with Data

The researcher read through the entire transcript of each focus group to become familiar with the depth and breadth of data prior to coding. Attendance of the researcher at the focus group meetings and writing the field notes helped in developing an initial knowledge and general impression of data prior to reading transcripts, which in turn facilitated further immersion in the data.

2.14.2.2 Initial Coding

The researcher reread the entire transcript of each focus group, line by line, and coded ideas as they emerged. Codes were assigned to relevant selected segments of the text using the MAXQDA program in a systematic fashion. Codes included both 'In vivo' codes which utilises the language and terms used by parents and 'Emergent' codes that were

devised and captured by the researcher through reading the transcripts. Text segments were assigned to multiple codes at this stage.

2.14.2.3 Coding Grouped into Higher Order Coding Categories and Themes

The researcher read along the list of the different codes that were identified across the data set of both focus groups systematically to identify topics and ideas that were integrated and related. These integrated codes were allocated into higher order codes. Codes and sub codes were ordered into a hierarchical structure using the MAXQDA functions. Each category and its sub codes were then revised by the researcher to ensure coherence. Using MAXQDA, the coded segments under each code were overviewed in one window, through which the researcher could comment on each coded segment and assign a weight score of relevance, as shown in **Figure 2-3**. Towards the end of this stage, the researcher captured and listed the emerged themes, sub themes (higher order categories) and sub codes.

Coded segments

Code: child protection\feeling of guilt

12 coded segments from 3 documents and 0 document groups

D: My husband would feel guilty about passing it on.

I: Your husband would, would he?

D: Very much so, and any time they've been to the dentist, like when (name) was crying the last time and I, he always rings me and says how was it? And I don't let him go to school, he kind of gets the day off

I: Yeah.

Comment	Document name	Code	Begin	End	Weight score	Preview	Area	Coverage %	Author	Creation date
○ guilt of passing it	fathers focus group...	child protection\feeling of guilt	1053	1068	5	I: Can I say some people or the only bits of things that, is th	648	0.95	areej	26/03/2017 18:12
○ guilt of passing it	fathers focus group...	child protection\feeling of guilt	1071	1099	5	Cl: You're still going to beat yourself up a little bit <because	1039	1.53	areej	26/03/2017 18:14
○ no guilt	fathers focus group...	child protection\feeling of guilt	1362	1367	0	Do, do you think the guilt thing was there with your wife as we	263	0.39	areej	29/03/2017 11:07
○ guilt of passing it	fathers focus group...	child protection\feeling of guilt	1616	1636	5	MC: and yes, there is guilt, I: Yeah. JMC: I mean I would, be	917	1.35	areej	29/03/2017 11:21
○ grandparents guilt	fathers focus group...	child protection\feeling of guilt	219	229	5	CC: Yeah. Like I'd be honest with you, her grandparents feel gu	770	1.74	areej	29/03/2017 15:10
○ no guilt	fathers focus group...	child protection\feeling of guilt	706	716	0	JMC: it's em, it's a case where it's there, it's inevitable, it	500	1.13	areej	29/03/2017 16:00
○ guilt not to treat	fathers focus group...	child protection\feeling of guilt	842	868	5	JMN: I, I don't have the guilt thing, I: Good, yeah. JMN: be	939	2.13	areej	29/03/2017 16:10
○ grandparents	ladies full focus group	child protection\feeling of guilt	1562	1576	0	F: My, my father feels really guilty, I: Your father feels F	760	0.48	areej	16/04/2017 21:46
○ guilt of passing it	ladies full focus group	child protection\feeling of guilt	1625	1637	5	D: My husband would feel guilty about passing it on, I: Your h	711	0.45	areej	24/04/2017 15:25
○ guilt of giving giv...	ladies full focus group	child protection\feeling of guilt	1591	1601	0	D: up till (name) had it or was diagnosed with it I: Yeah. D:	382	0.24	areej	16/04/2017 21:54
○ no guilt	ladies full focus group	child protection\feeling of guilt	1643	1676	0	E: Can I ask you to ask him a question? I: Yeah. D: Yes. E:	1130	0.71	areej	16/04/2017 21:56
○ guilt of treatment...	ladies full focus group	child protection\feeling of guilt	2317	2342	5	H: my little fella then eh was, I felt eh he got one strep thro	908	0.57	areej	17/04/2017 10:17

Figure 2-3 Example of MAXQDA coded segment window

2.14.2.4 Coding and Thematic Review

This was carried through two phases. The first phase involved the researcher review of the coding at the level of coded segments and in relation to the entire data set.

At the second phase, the supervisor (AOC) and an independent experienced qualitative researcher (BD) reviewed the data independently before the main themes were agreed to ensure both consistency and comprehensiveness in coding and data analysis and to limit any researcher bias.

The transcripts were sent along with the coding hierarchy and list of themes to the supervisor (AOC) and the experienced qualitative researcher (BD) separately. Both reviewed the coding independently and read through the transcripts and searched for themes. A meeting was arranged subsequently between the researcher and the reviewers to discuss the coding, quality of data obtained and the abstract themes. New themes identified and some themes were collapsed into each other. Controversies were resolved by discussion and consensus reached about six main emergent themes. The data were rich and experiential and were judged to be sufficient to reach thematic saturation based on the following criteria:

- Agreement among the research team on the overall quality of data obtained from each focus group and the quality of data obtained from each subgroup of participants in light of the research aims and objectives.

- Comparison between the themes emerged from focus groups until no more new themes were identified from the latest focus group data.

2.14.2.5 Interpretive Analysis

The researcher performed ongoing analysis by reviewing the collated data extracts for each theme in an attempt to account for patterns that had been found, the diversity and range of perception, views, experiences and beliefs under each theme, their recurrence and their broader meaning and implication. Attention was given to areas of disagreement, affirmation, conflict and connection between characteristics and experiences of the participants and their attitudes. Coding was an ongoing process,

additional data within the themes that had been missed in earlier coding stages were coded and the coding was refined during the process of data analysis.

2.14.2.6 Writing Results

The researcher wrote the results in light of the above detailed descriptive and interpretive analysis.

2.15 Rigour and Trustworthiness

Rigour and trustworthiness were established by adherence to quality guidelines for the qualitative research conduct (O'Brien et al., 2014), revisiting issues and requesting clarification during the course of the focus groups and by triangulating interpretations among the research team. For example, the repetitive referral of affected parents to their childhood experience with bullying and the only father who didn't seek referral for dental treatment of his child until the late mixed dentition stage report of his own traumatic dental experience with extractions at young age were both agreed by the research team to suggest that the personal experience of parents affected by AI/DI plays an important role in parent's judgements of their children's teeth and perceived need for dental treatment. The supervisor (AOC) and the experienced qualitative researcher (BD) reviewed the data independently before the main themes were agreed to ensure both consistency through validity and comprehensiveness in coding and data analysis. The strength of qualitative research lies in its validity which is the extent to which the findings accurately represents the social phenomena studied (Mays and Pope, 2007). All data were evaluated using the checklist of standards for reporting qualitative research (SRQR), developed by O'Brien et al, 2014. **Appendix 10** shows the SRQR with each item referenced to the appropriate thesis section.

3 Results

3.1 Introduction

A total of fifty-six eligible parents were contacted through the appointed gatekeeper, eighteen parents agreed to participate (eight fathers and ten mothers).

Two focus groups were conducted involving a total of thirteen parents of affected children (three mothers and two fathers failed to attend on agreed dates). Seven mothers participated in one focus group held on 2/3/2017 and six fathers participated in the other focus group held on 16/2/2017. Both meetings were arranged in the afternoon (at 5:30 P.M) based on participants' preference. Data review and analysis were completed in conjunction with data collection and the quality of data obtained from two focus groups was rich and experiential and was judged to be sufficient to reach thematic saturation.

3.2 Demographic Characteristics of Participants

3.2.1 Fathers Focus Group

Six participants attended this meeting, five of whom were aged 35 or above. None of the fathers had a Medical Card. Families are considered eligible for Medical Cards if the combined accessible net weekly income is less than the qualifying financial threshold calculated as per the Irish Health Service Executive (HSE) (HSE, 2014) . All participants were Irish and no parents from ethnic minorities volunteered to participate.

Two fathers had children affected by DI and four fathers were parents to children affected by AI. Three of the six fathers were affected by the same condition.

Within the time frame of the study, we identified only one parent (a father of children who never had any dental intervention in the primary dentition stage). The demographic characteristics of participants are shown in **Table 3.1**.

Participant	Age	Medical Card	Affected by the same condition	Child affected by AI/ DI	Number gender and age of affected children	Dental Intervention in primary dentition stage
1	≥35	No	No	AI	1 Male (Age 6)	Yes
2	≥35	No	No	DI	1 Female (Age 10)	Yes
3	≥35	No	No	DI	2 Female (Age 2) Male (Age6)	Yes
4	≥35	No	Yes	AI	1 Male (Age 9)	No
5	<35	No	Yes	AI	2 Male (Age 6) Female (Age 2)	Yes
6	≥35	No	Yes	AI	1 Male (Age9)	Yes

Table 3-1 Demographic characteristics of fathers focus group

3.2.2 Mothers Focus Group

Seven mothers attended this meeting, six of whom were aged 35 years or above and none qualified for a Medical Card. All participants were Irish and no parents of ethnic minorities volunteered to participate.

Three mothers had children affected by DI and four mothers were mothers of children affected by AI. Two of seven mothers were affected by same condition. Within the time

frame of the study, one mother of a child who never had any dental intervention participated and she was the partner of the father who attended the fathers focus group. **Table 3.2** outlines the demographic characteristics of participants.

Participant	Age	Medical Card	Affected by the same condition	Child affected by AI/DI	Number, gender and age of affected children	Dental intervention in primary dentition stage
1	≥35	No	No	DI	3 Males (Age 16, 13 and 10)	Yes
2	≥35	No	No	AI	1 Male (Age 9)	No
3	≥35	No	Yes	AI	1 Male (Age 6)	Yes
4	≥35	No	No	DI	1 Female (Age 7)	Yes
5	<35	No	No	AI	2 Male (Age 6) Female (Age 2)	Yes
6	≥35	No	No	AI	2 Male (Age 7) Female (Age 10)	Yes but in mixed dentition stage.
7	≥35	Yes	Yes	DI	2 Male (Age 6) Female (Age 2)	Yes

Table 3-2 Demographic characteristics of mothers focus group

3.3 Focus Group Duration

The fathers' focus group lasted for one hour and fifty minutes and the mothers' focus group lasted for one hour and twenty-four minutes. The number of transcripts pages analysed were a hundred and twenty-nine and a hundred and seventy-eight for the fathers and mothers focus groups respectively.

3.4 Main Themes

Cross-sectional analysis of the whole data set of the focus group discussions was carried out with coded, transcribed data. Six themes were identified as represented in **Figure 3-1**.

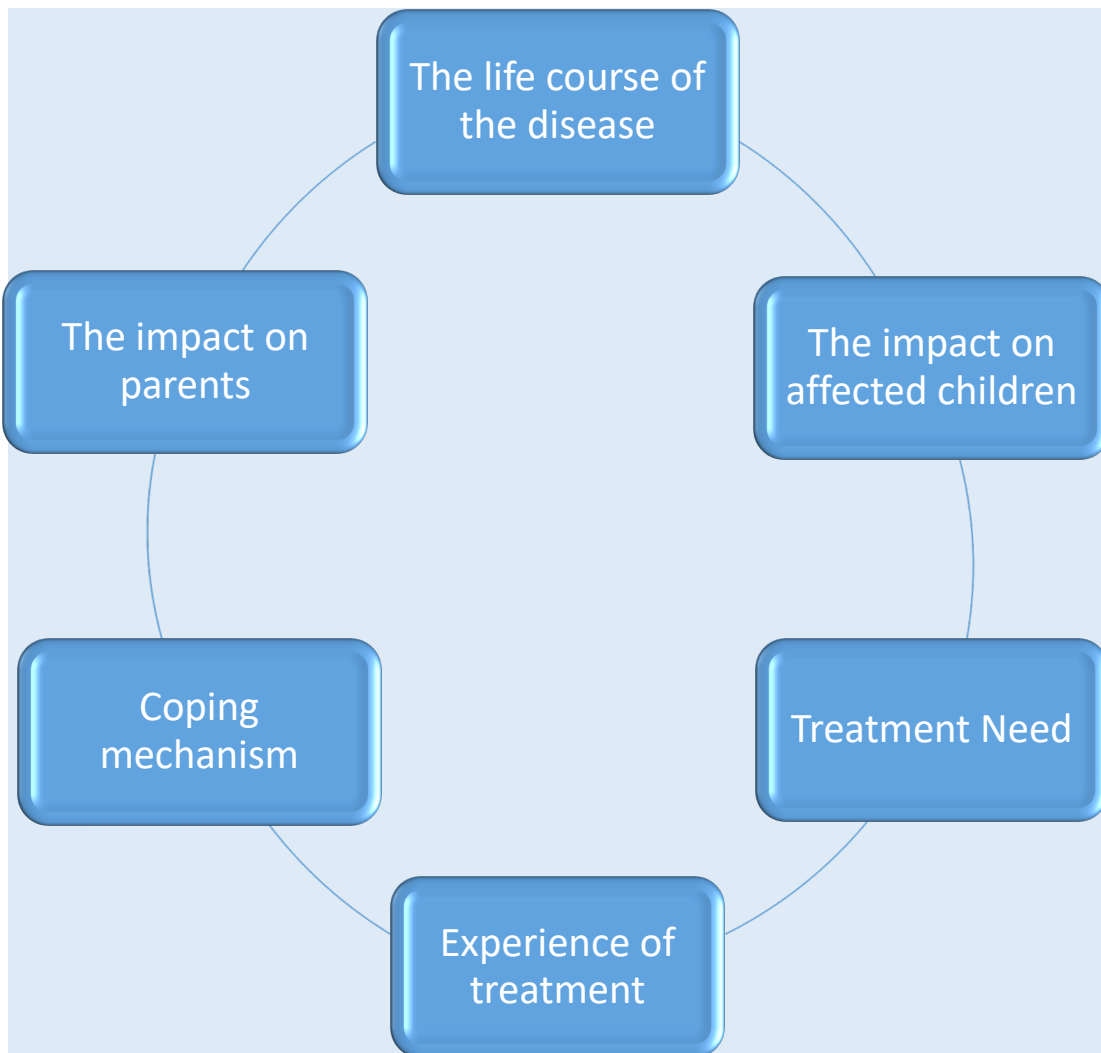


Figure 3-1 Six main themes identified from cross sectional thematic analysis

Each theme is presented below and and illustrated with quotes from the focus groups.

3.5 The life Course of The Disease

3.5.1 Disclosure and Diagnosis

Most parents started to notice an altered appearance of their children's teeth as soon as the first primary teeth were erupting.

CC: *" Like you'd see, the minute you'd see it come through the skin, you can see it. You know straight away. "* (Father of a child affected by DI)

CS: *"I suppose as soon as the teeth had come through we could see that, so we knew it wasn't something that the teeth had been white and then they'd actually degraded quite quickly."* (Father of a child affected by AI.)

Affected parents and their partners appeared to be more conscious about the condition especially when there is a family history of a developmental dental defect.

JMC: *" I would have been conscious of it from an early stage."* (Father of a child affected by DI-Father is affected)

Concerns and apprehension of getting affected teeth were expressed by several affected parents. The following example demonstrates the affected parent's apprehension, worries and upset as they were awaiting eruption of their child's teeth.

B: *"And you know the feeling, your heart sinks in your mouth, you're just waiting and waiting for them to come through and then you see that they're brown, oh it's horrific. I cried and cried. Oh I hated it,,,,, the waiting for them to come through and you're just waiting, and everyone keeps saying oh, their teeth will be through and you're just dreading. You're dying to find out but then you're dreading it, Oh, it's awful. Because you know what they're going to go through, like."* (Mother of a child affected by DI- mother is affected)

From a different perspective, a sense of shock and lack of understanding in association with tooth eruption was expressed by the unaffected parents and those who were not aware of any positive family history :

C: *"I didn't know what it was, like. <I just knew her teeth> wasn't right when they were cutting through her gums, and I said God, there must be skin or something that's growing over her teeth, that's what I thought,, So I was scraping her tooth with my nail thinking there was something on over her teeth."* (Mother of a child affected by DI)

Several reasons were proposed for the altered teeth appearance and texture including decay, pregnancy related problems and antibiotics:

A: *"Actually I thought it was something to do with me, even though there was no history of it in the family, I actually thought it was something to do with the pregnancy. And I was asked about antibiotics when they were babies, and things like that, "*

B: *"<just thought they were decayed, like.>"*

F: *"I was asked that question."*

(Dialogue between mothers.)

Interestingly, one parent reported not getting his wife's own diagnosis until her children were diagnosed with the condition.

CS: *"My wife had it quite extensively as a child, but she'd always been told that she got it because she had pneumonia as a child and it was the antibiotics <when she was a baby.> And then we saw it with my boy, and we started thinking well maybe there's a hereditary aspect here, and you know that's I suppose kind of confirmed I suppose with the dentists, and eh....., the dentist diagnose it , you know, the boy, he wasn't bottle-fed hugely anyway."* (Father of child affected by AI- wife is affected- .)

Regardless of the family history of developmental dental defects, parents were keen to get dental advice as soon as they started to notice the defect in their children teeth. Some were referred to the DDUH through their own dentists, while others used Google search to get to the DDUH and arranged for their children's dental assessment by themselves. Affected parents who were themselves patients of the DDUH found their

way to the paediatric service in DDUH at an early timepoint . The age of the child when parents sought referral varied but most referrals occurred between one to three years of age. Those who sought referral later had been attending their family dentist for regular check ups and teeth debridement.

In general, parents were reluctant to accept the myth “that baby teeth are not important and will fall out anyway” and exhibited a persistent attitude to get expert assessment , advice and treatment.

C: *“ I said there’s something wrong, I’m bringing her to the baby nurse for that, and he was looking at me going, you know, don’t be silly or whatever, and he says don’t worry about it, it’s only her baby teeth. I said she has some type of a deficiency, I says I want to get to the bottom of it and the dentist that was with the nurse says you’re wasting your time, it’s only the baby teeth, you’ll be on a waiting list for about five to six years and then teeth will have fallen out again, so you’re worrying about nothing, basically. So I said, I wasn’t happy with the answer he gave me, went to my own dentist, he checked it out,,, and I ended up here in the Lincoln in few months.”*

D: *“But it is that old-fashioned thing of, ah, the baby teeth, don’t worry about them.”*

C: *“They have the baby teeth till they’re seven or eight years of age. Of course they’re important, you know.” (Dialogue between mothers.)*

3.5.2 Transition to Mixed Dentition

Opinions differed as to whether primary teeth were more or less affected when compared to permanent teeth. A number of parents postulated that their children’s teeth worsened over time. In contrast, other parents believed that the permanent teeth are usually less affected. A minority of participants recalled that the diagnosis for their children was not made until their early mixed dentition. The examples below displays these contradictory views.

Permanent teeth are more affected:

G: *"I have one out of four, <my youngest boy> so his baby teeth, they weren't too bad, they were always a bit yellow looking but I didn't really pass any heed. So I knew then when the next ones were coming, they were just rough, yellowish and discoloured."* (Mother of a child affected by AI)

A: *" Well, their baby teeth were slightly, not as white as my nephew's baby teeth and that kind of thing, and they were quite small,,,,, and also they wore down like as if she was grinding a lot more. But no cavities, no problems, and didn't, we didn't discover Amelogenesis Imperfecta until the two new front teeth had come down and were very thin,, My kids in their new teeth it is more. "* (Mother of a child affected by AI.)

Permanent teeth are less affected:

F: *"But they always say, That the next teeth aren't usually as bad as the baby teeth."*

D: *" My three now, the permanent teeth are much better than the baby teeth."*

B: *" Yeah, <mine were the same, yeah.> "* (Dialogue between mothers of children affected by DI and a mother of a child affected by AI.)

The focus group discussions provided a fascinating insight into the children's experience with transition into early mixed dentition after having had a partial/full mouth restorative treatment. Children were upset with the new affected permanent teeth and for some of them this was the first time to notice that their teeth were different.

CC: *"My son had crowns put in when he was three and I think it's probably only now when he's in, coming to the end of senior infants, that anyone is even noticing, including kind of him,,,,,so he's got his crowns and now some of his teeth are falling out and he can see the ones coming through and he's like I want to keep the ones <That he has.> he's probably now for the first time getting so aware he's realising that there is a problem. The new teeth coming they're this colour (grey) and they're very weak, but the ones around them are shiny white."* (Father of a child affected by DI, Child had full mouth restorative treatment in the primary dentition.)

B: *"He was really upset. So eventually and he said and then they're sore, the ones that are coming up are really sore, so he was very upset with the change, <and for good reason.> Yeah, there's a pain in it, and they feel different, like the rest are smooth like."*(Mother of a child affected by DI, Child had restorative treatment in primary dentition.)

Parents appeared well informed about limitations in restoring the permanent teeth when partially erupted. Some parents believed that awaiting the exfoliation of all primary teeth is necessary before restoring permanent teeth.

I: *"And I suppose you can't have the work done straight away on the new ones coming through, can you?"*

CC: *" No."*

JMA: *"No, no. <You have to wait.> well I didn't have this problem but I would imagine you would have to wait till the teeth developed"*

CL: *" Yeah."*

CS:*"I think you have to wait till they all fall out. And you have to get it done again. "(Dialogue between fathers.)*

3.5.3 Lifelong Dental Treatment Need

A recurrent theme in the focus groups was a sense of acknowledgement of the lifelong dental treatment need of affected teeth, mixed with uncertainty about future treatment needs.

CL: *"My sister has it and she has a boy the same age as my son and his teeth, thank god are one hundred per cent perfect, which is great because at least he doesn't have to go through the twenty years of treatments,,,,, It is lifelong." (Father of a child affected by AI.)*

A: *"So I think there is no hard and fast rule, it's a long road ahead and it's gonna basically just see what happens every six months, there's no point in thinking too far ahead because just things are gonna be changing all the time anyway, it really depends on how the mouth develops and everything." (Mother of a child affected by AI)*

The parent's own experience and information provided by the dentist attributed to shaping this future attentiveness. A variety of perspectives were expressed in relation to that including;

- Focusing on present:

***B:** "As I say to AOC, I go appointment by appointment, and I do because any time you try and think too far ahead it changes or something, so I can only go from literally each visit, that's kind of how I did it." (Mother of a child affected by AI.)*

- Preparing the child to anticipate progressive and continuous dental treatment:

***JMC:** "We're teaching, letting him know slowly but surely kind of what's ahead of him, and it is a bit of a long road to get to it, but at some point in time the intention is he'll have his teeth like the others <afterwards.> When he's eighteen he can get it sorted." (Father of a child affected by AI.)*

- Investing family savings:

***CC:** "Because it is a lot of years, it's not just one. The parent has to be prepared like it's not going to be a short-term fix, like it's a lot of years and travelling, money and operations, like they have to be prepared for everything then." (Father of a child affected by DI.)*

Sixteen, eighteen and twenty years of age were suggested as an age for permanent or semi-permanent solutions. Parents expressed an interest for traveling abroad if needed, to seek these permanent solutions. One father shared his experience of travelling abroad for treatment. Concerns about financial burden of treatment were widespread among parents.

3.6 The impact on Affected Children

3.6.1 Appearance and Associated Negative Social Reactions

Parents, on the whole, demonstrated a belief of an increased social expectation of white well-aligned teeth and a social perception of white teeth as an indication of personal hygiene. It was suggested that young children were influenced by this social phenomenon and so they were expecting normal primary teeth to be pearly white and well-shaped teeth. In fact parents described their children's teeth as 'not as white as other children teeth', 'discoloured', 'yellow looking', 'smaller', 'thinner', 'not solid looking', 'jagged and pointed', 'spiky', 'rough', 'not healthy looking', and 'see-through – if you shone a light you could see nearly through the tooth'.

JMN: *"There is an expectation now to be personal hygiene, you know, like we're not going out the back garden to the toilet so yeah. So there's an expectation." (Father of a child affected by AI.)*

JMC: *"And it can kind of hit you a little bit more when your son at three years of age turns round and says "Dad, why are your teeth green?" because he's expecting white." (Father of a child affected by AI.)*

A recurrent theme was a sense among parents that social pressures are increasing over time, driven by media and based on social norms from a cultural perspective. Parents provided examples from their own experiences confirming their above mentioned beliefs.

B: *"The other day and they were saying oh, the person has lovely teeth and I was like [laughing] it's not something that's not big here."*

A: *"Yes, people judge people by their teeth, people look at your teeth when you're speaking."*

D: *"I mean I remember back in, I was seventeen, so what, twenty, thirty years ago, thirty-two years ago, getting braces on,,,and the only reason, I had a gap, my teeth were always yellow. I had a gap between my two front teeth, my aunt in the States came home regularly, and I mean they got braces on over there just because, to be part of the*

fashion, it literally was. And she was on to my mother, you know, you need to do something with her teeth and whatever.”(A mother affected by DI and her own experience with different cultural perspectives.)

Parents considered the Irish culture more relaxed when compared to other cultures like the American culture, however, they thought that it is still influenced by the worldwide move toward aesthetics and perfection.

JMN: “I guess it’s based on the norm, isn’t it?”

I: “Yeah, on the norm, yeah. Do you think it’s, do you think it’s an Irish thing? That teeth should be straight and white? Do you think that’s a cultural thing?”

JMC: “<I don’t think> so, no. We’re more relaxed <than lots> of other cultures. ”

I: “Like who do you think wouldn’t be relaxed? From a culture point of view.”

JMN: “<US.>”

CC: “<Americans> <The US, I would have thought.> Now I don’t think your teeth to be perfect, they have to be all precisely spaced or white ,, there’s a bandwidth there I think that.” *(Dialogue between fathers about social norms from cultural perspective.)*

One mother referred to the national caries rate as an important factor on developing cultural attention to affected teeth. Countries with high caries rate were believed to be less concerned about discoloured teeth.

D: “ In Ireland generally speaking people’s teeth are quite good because there’s fluoride in the water, now my husband’s from Scotland so you’d be more inclined to see decayed teeth over there because they don’t have the fluoride in the water. So it’s not as, you know, instantly obvious, but over there, I would spot like that people their teeth wouldn’t be as kind of pearly white as here.” *(Mother of a child affected by DI-mother is affected-)*

Two divergent and conflicting views emerged in relation to social reactions to affected teeth, some parents believed that there is an increased general acceptance of diversity by the society including children while other parents felt that society and children nowadays are more likely to make negative judgments based on appearance.

One mother, for example, talked about increased social acceptance of diversity among children at school:

A: "I think in school now, like I think it's amazing. My little girl's class has a girl who's diabetic, there's loads of people who are different, can't have gluten, people who are on EpiPens, there's people with Coeliac, there's loads of different things. So actually that's nearly a good thing because she just thinks there's something, she was saying to me one day about special needs in school and stuff like that, and she goes but I'm special needs,,, because she's under special dental needs in here, ,,I think it's good in some ways because they're so aware of that, this person is diabetic and this person is this, and they get told everything in school ,they don't mind being a little bit different in some ways." (Mother expressing her beliefs of social acceptance of diversity.)

While other mothers talked about people increasingly becoming more conscious about appearance:

D: "I think probably was easier before, I think now everything is just perfect, you know, the perfect figure and the hair and the look, the fake, you know, the teeth. I do think we're far more aware. I feel they're so self-conscious now, and conscious of what everyone else looks like as well."

H: "I think it will be going forward for life."

Parents who believed in an association between the appearance of their children's teeth and negative social reactions referred to their own experience when they were children and their children's experience to support their beliefs. These accounts focused on being seen and/ or treated differently, predominantly because of the visual differences caused by teeth appearance. Negative social reactions from parents perspective varied and included :

- Staring:

JMN: "You might kind of catch somebody having a look, they wouldn't necessarily say anything." (Father of a child affected by AI)

- Questioning teeth appearance:

C: *"Maybe the neighbour's kids, could be about seven and he used to go oh what's wrong with your teeth." (Mother of a child affected by DI)*

A: *"My daughter as well in primary school. And kids in school, they used to say why are your teeth so small or, and it's not even that, it's not very obvious." (Mother of a child affected by DI)*

- Questioning teeth brushing:

CL: *"A lot of my friends and stuff, like when I was in junior, senior infants would say things like that and well why don't you brush your teeth?" (Father of a child affected by AI- Father is affected- .)*

- Assuming over-consumption of sugary food:

E: *"When I was a child everyone thought that I was eating too many sweets, because you know, obviously what do you tell your children? If you eat too many <sweets they'll rot your teeth.> ,,there's an assumption that you're eating the wrong food." (Mother affected by AI describing her own childhood experience)*

- Bullying : Several comments on teeth texture, size and colour were encountered by affected children.

A: *"kids in school, they used to say why are your teeth so small or, yellow, And her friend said no matter how long she brushes them, they'll always be yellow, or something." (Mother of a child affected by AI)*

A mother affected by DI described her own affected sister's experience with bullying at school:

B: *"Like my sister was called Sellotape teeth. <One of her, one of her teeth came down white,> and this girl stood up in front of the class and said I just want to make an announcement, (name) has a white tooth."*

There was a general agreement among parents that young children at preschool and primary school age could notice affected teeth and might show innate unfavorable responses.

CS: *"I don't have it, so like we never really kind of mentioned it to my son, and he's in senior infants now and I suppose it's been mentioned. He is come back from school a couple of times and said oh, some of the boys and girls were saying my teeth are a bit yellow."*

I: *"<So even at a young,> so at a young age, the children will pick that up, the other children."*

JMA: *"<Oh, definitely, yeah, children more so than adults.>"*

Multiple speakers: *<Yeah. Yes.>*

JMA: *"Like children have no filter"*

On the other hand, other unaffected children within the family, family members and friends demonstrated assimilation and understanding and are less likely to show undesirable responses. This suggests that the perceived negative responses are socially contextualised.

I: *"Do the other children say anything? In your own family?"*

JMC: *"No, in my case, because I think they know what he's going to go through, knowing what I've gone through in the last while, at some point in time the intention is he'll have his teeth like the others <afterwards.> Grandparents would know about it and <family friends, for example> but nobody would ever say it." (Father of a child affected by AI)*

F: *"My oldest daughter now, she's seven, she doesn't have it. <She doesn't realise it,> she didn't, they've never mentioned anything about her younger sister's teeth." (Mother of a child affected by AI)*

3.6.2 Appearance and Psychosocial Impact on Children

A number of issues were identified in affected children including self-consciousness, feeling different, shyness, social avoidance and decreased self-confidence. Parents referred to their own children's experiences and affected parents explicitly referred to their own experiences to support the believed impact on child's confidence and self-esteem.

A mother of a girl affected by DI described her child's self-consciousness and associated shyness at age three:

C: *" My child would shy away like that was what I noticed when my daughter got it, when you were talking to her, she'd always sort of hide her head, she got the crowns in when she was three, three and a half. And she was shying away at that age,,,, She even asked me like Mammy why my teeth like this is and why have you not got it and blah, blah like." (Mother of a girl affected by DI.)*

This view was echoed by another father affected by AI based on his own experience:

JMC: *"The minute somebody's mentioned a dentist visit or whatever, I'd close my mouth. Em that was the difference. So I can see that going to affect my son. "(Father affected by AI describing his own experience.)*

Another mother followed this up by providing an anecdote from her affected young brother's experience to support her believed association of decreased self-confidence of affected children especially at school age:

B: *" My younger brother, my two brothers have it and my younger brother is so quiet and everything, and I really believe that it's because of the teeth. I really think he had no confidence growing up. And I, I honestly do believe it's because of the teeth. Yeah, my mam does too. " (Mother affected by DI describing her affected brother experience.)*

The age at which children started to notice their teeth being different were varied. Some parents reported that their children became aware as soon as the primary teeth were erupting and even before reaching three years of age.

I: *"So what age did you think it started to matter? Really? "*

C: *"From the moment like she could maybe talk and her teeth started showing, maybe."*

I: *"How old is that?"*

H: *"One? When her teeth were up."*

Interestingly, a two year old child affected by AI used to think that she 'had cheese' on her teeth.

CL: *" I'm actually telling, my two-year-old already knows her teeth are different to her mam's teeth and she'll say it like,,, <It's a visual thing.> And she'll say things like oh, why do I have cheese on my teeth, is usually what she's saying. She barely would know what's in the world but she can already distinguish that." (Father of a child affected by AI)*

Looking different and perceiving oneself to be different at toddler age was linked to children's attitudes that took the form of questioning teeth appearance as evident in the above example and an increased attention to the appearance of other people's teeth as illustrated in the example below.

H: *"She's two and four months, she says to my mam. My mam has gorgeous white teeth and they're perfect, she would say regularly to my mam, "Granny show me your teeth", all the time, and we don't really kind of make a big deal about teeth when we talk, but if you want to talk about something, talk about it, and she is always on about teeth." (Mother of a child affected by AI)*

Other parents believed that children won't notice any difference until starting school, around the age of four, where they become more aware of difference and people responses.

CS: *"To be honest I'd say he wouldn't have realised it himself until probably starting school."*

I: *" And how old would he have been then?"*

CS: *"He was very young starting, youngest in the class so he was kind of four, four and a half when he started school. " (Father of a child affected by AI)*

The child's own character was suggested as an important factor to determine the extent of impact on social life and psychological well-being of affected children. A parent of a

child with “ introspective character” expressed great concerns about the negative psychosocial impact on his child’s future:

JMN: “ But what worries me is he’s quite an introspective kid anyway. That’s my concern. I don’t think it’s because of it. I think he’s just he’s a thinker, and it’s not a good combination.” (Father of a boy affected by AI, Father is affected.)

In contrast, parents of children with strong personalities believed their children are less affected.

A: “she’s actually quite a strong girl, she’s confident, but one day we were up at gymnastics and a girl kept saying to her what is wrong with your teeth? Why are your teeth so yellow? and I was like going (muttering). But she just said oh, it’s just there’s something wrong with them, and she just brushed it off. Kids in school, they don’t ask her anymore because they know her, she’s actually, very good, it doesn’t upset her. “ (Mother of a girl affected by AI.)

Regardless of the physical appearance of their teeth, children were able to build friendships, yet some uncertainty was expressed about future relationships.

JMN: “ He’s got a good circle of friends, he’s in a good school as well, and I think they’ve got the kids understanding people are different in different ways. But look, there’s always gonna be, as I say, those kids that’ll say it to be mean, even, not out of ignorance. And, of course, as he gets older, subject of girls and all <that type of thing, that’s>, you know, it all comes into focus.” (Father of a boy affected by AI.)

One mother referred to her own childhood experience to illustrate on the questionable impact on child’s confidence and self-esteem.

E: “Believe it or not I never lacked confidence, and I still don’t lack confidence ,,,I don’t think I can recall being conscious, till about maybe first or second class. I’d say one or two people who would have picked on somebody for something or other. I don’t think it ever affected my smile. <For years, until I got,> until I got the white teeth.” (Mother affected by AI talking about her childhood experience.)

Opinions of parents differed as to whether the child’s gender will influence the extent of psychosocial impact. Some thought that girls were more affected, others argued that

boys were becoming more concerned about their own appearance and so might be more or equally affected.

H: "I think it's more, that hits girls a bit more than boys"

Multiple speakers: <Yeah.>

H: "<I do think boys,> that will hit, but I think girls can be a bit more."

B: "<A little bit more?> <I think boys in their> teenage years."

D: "I think the lads are better to deal with it, but I do agree with you, I think boys are becoming very body-conscious way more so than they ever were, you know, in what they wear, how they look, so like there's definitely none of them like it,,, Boys like to look at themselves too."

The factors that determine the psychosocial impact on affected children and the aspects of psychosocial impact from parents' perspectives are summarised in **Figures 3-2 and 3-3**.

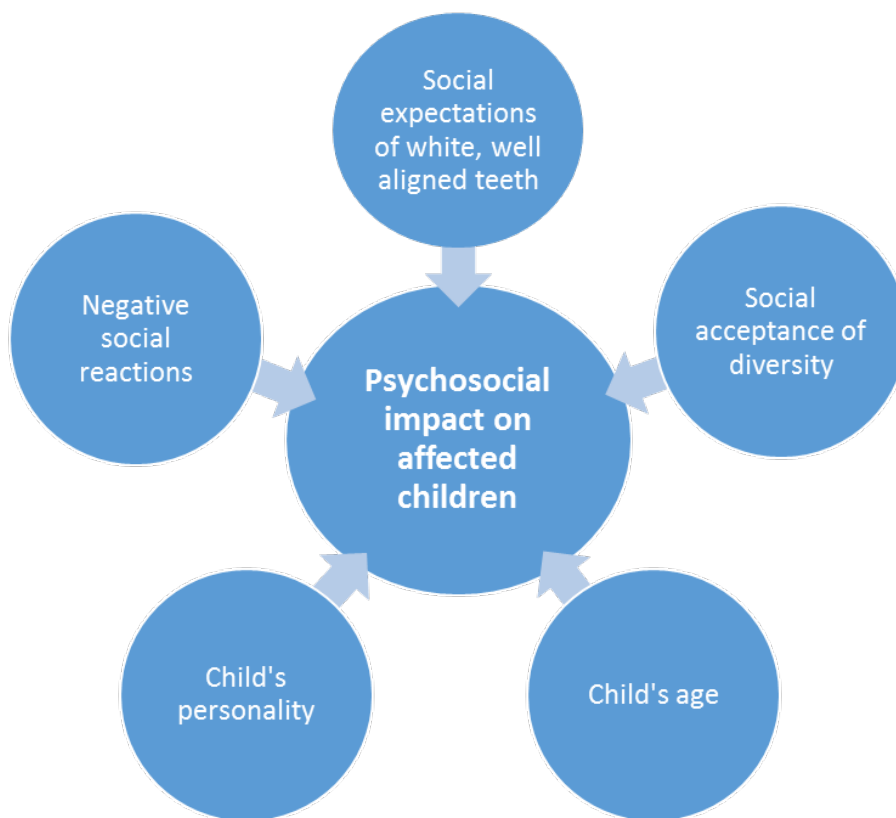


Figure 3-2 Factors that determines the psychosocial impact on children from parents' perspective.

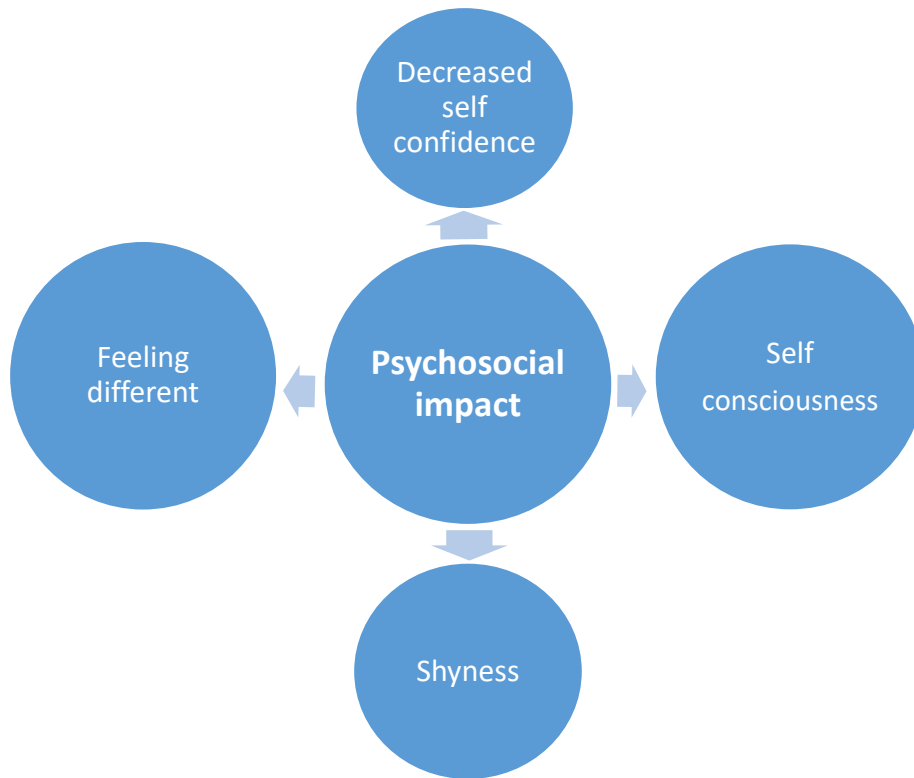


Figure 3-3 The psychosocial impact on affected children.

3.6.3 Impact on Children Health and Function

The majority of parents expressed their concerns in relation to the affected teeth being friable and vulnerable to break down and wear.

A: *"The front ones are chipping on forks, when she bites too hard on a fork. Also, they wore down like as if she was grinding a lot more. But no cavities, no problems."*

G: *"I mean when you see, see them from the side it's just so thin, you know."*

B: *"<It's the wear and tear.>"*

G: *"<I'd be afraid to sort of give him an apple, it's...>"*

F: *"They chip easily."*

G: *"My ones just suck it more away, from like chewing and stuff."*

(Dialogue between mothers of children affected by DI and AI.)

Most of affected children experienced sensitivity, pain, tooth wear and higher rates of calculus build up.

CL: *"For my kids anyway, because they don't have enamel,, when they have cold things they're sore and they're quite sensitive, like my two-year-old has it and she struggles quite a bit with sensitivity and pain and things like that."* (Father of a child affected by AI)

JMN: *"We're brushing, we're flossing, we're doing all sorts, but if there's a big gap between appointments it will build up, plaque will build up."* (Father of a child affected by AI)

Interestingly, affected parents relied solely on their own experiences to assume that children's teeth were sensitive and friable even when they were not.

E: *"For a long time I wouldn't let him eat ice cream because I couldn't touch ice cream because of the cold, I was very sensitive to the cold,,, and then he was in the crèche and they had this birthday party thing and they gave out ice cream and I had put on the thing don't give him ice cream, so he comes back and he says Mammy, the ice cream, it's lovely,,, no he didn't have the sensitivity that I had."* (Mother of a child affected by AI, Mother is affected.)

Parents avoided offering their children certain foods and drinks (ice cream, fizzy drinks, toffies, jellies, hard food) due to sensitivity and breakdown concerns before having dental treatment and due to fears of 'pulling off' crowns after having had dental restorations.

CC: *"Our lads haven't even ever, they got one lollipop on holidays this year for the first time ever, but we probably won't give them another one again, just in case they bite and break one or something like this, like we're so conscious of that. Lollipops, anything, any hard sweets, anything like that, no-go."*

I: *"What about carrots and things like that, would you be worried about crunchy foods?"*

CC: *"Soft, even when we're cooking the dinner, we have to have it soft for them."* (Father of a child affected by DI.)

JMN: *“The only thing I probably would have been fairly stickler on was toffees that type of thing, and especially now because toffees will pull the temporary crowns off.” (Father of a child affected by AI.)*

Those parents who did not modify their children’s diet attributed that to lack of symptoms, and kids’ preference for such food. The discussion in relation to children’s diet highlighted some misunderstanding among parents about cariogenic food:

A: *“If they’re having treats, I only let them have chocolate and ice cream,,, Because chocolate isn’t as bad, because there’s fat in chocolate, there’s fat in ice cream and seemingly the sugar clings to the fat and that’s why it’s not as bad for your teeth.” (Mother of a child affected by AI)*

There was a general belief among parents of affected children that they are more conscious about their children’s food intake when compared to parents of unaffected children and their diet is healthier.

However, this might not be socially acceptable in some circumstances, e.g. when on holiday. This diet consciousness umbrella covers both affected and unaffected children of the same family.

I: *“What I’m hearing is that you’ve become very aware of teeth, do you think all parents are the same?”*

Multiple speakers: *“No.”*

E: *“Definitely not.”*

I: *“Do you think you’re more conscious?”*

A: *“<Way more.>” (Dialogue between mothers.)*

One mother talked about social implication of dietary restriction while on holidays:

E: *“I remember we were on holidays two years ago and we were in Lanzarote and you know the way they have the kiddies disco, they were playing musical statues, when you get out you get a lollipop, and my little boy said through the microphone I’m not allowed have lollipops,,, And the whole table looked at me. So there me the bad mother.” (Mother of a child affected by AI)*

A minority of parents related AI to other personal health issues like narrow maxilla, mouth breathing, digestion, and possible reduced hearing capacity.

JMN: "uh, digestion also suffers. In my situation the implications of AI impacted on my breathing,,, Mouth breathing, <more than through my nose, because it narrowed, it narrowed my palate > I had a, quite a long face, so I actually had jaw surgery as well." (Father affected by AI).

H: "I was told it can affect the ears, that your hearing can be impaired. All my family has selective hearing in my house, three lads on my father's side." (Mother of a child affected by AI)

Figure 3-4 summarises the impact on affected children health and function.

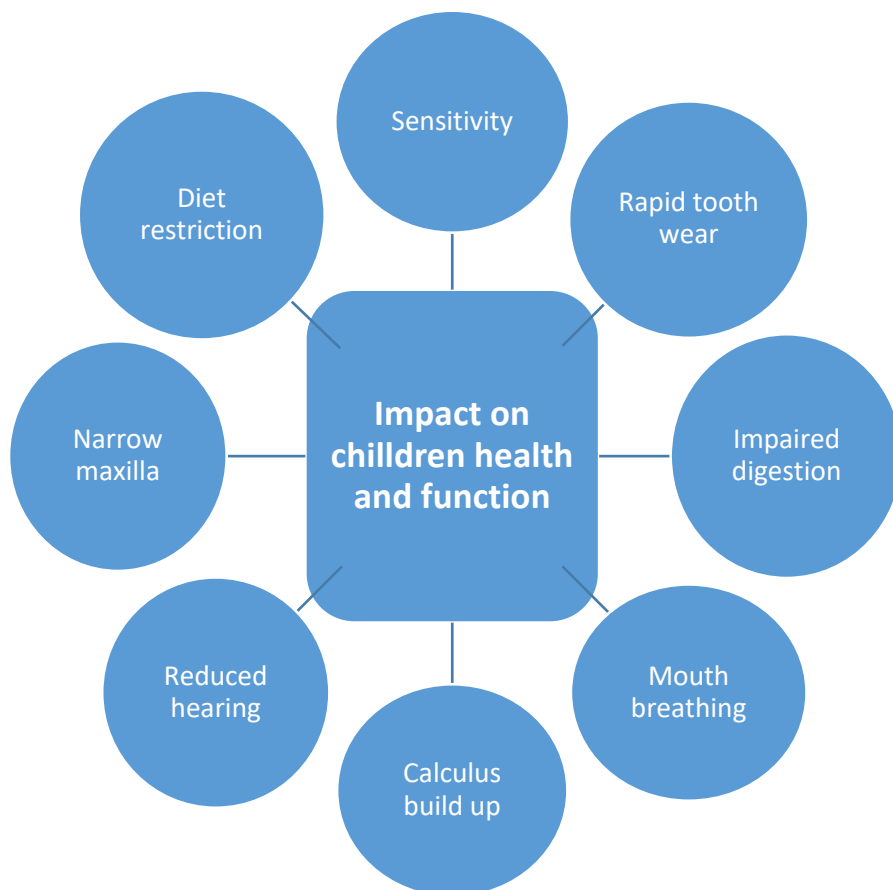


Figure 3-4 The parental perceived impact on children's health and function.

3.7 The Impact on Parents

Four key emotions of parents about having an affected child arose during the discussions; sadness, fear of bullying and negative social reactions, worry about associated impact on the child and the lifelong treatment needs and a feeling of guilt. In addition to these feelings, the apprehension associated with teeth eruption was discussed previously in the life course of the disease section. Parents expressed and discussed these feelings repetitively and in both focus group discussions. Parents believed that within their experiences, their consciousness about their children's dental appearance as well as other children appearance has been considerably increased after having affected children.

E: "I didn't feel guilty, like I think I felt very sad,,, like you feel terrible for the child."

B: "Oh it's horrific. I cried and cried. Oh I hated it. They're just brown and, do you know?"

I: "what do you hate most about them?"

F: "I hate the look of them."

C: "I haven't ever heard of it in my life, I was roaring and crying then going to her, she's gonna be teased."

H: "Oh, my heart breaks, because I'm looking at them, because it's something different like, I think anything different, you don't want your kid to be bullied."

(Dialogue between mothers.)

There was two main ways in which the feeling of guilt manifested within the experiences of affected parents. Firstly, the inheritance of a developmental dental defect is a reason to feel responsible for all the difficulties the child is expected to go through.

CL: "Yeah, it's just a strange thing because you know it's genetics, well in my case it's genetics and there's very little you can do about it, but you still feel a little bit responsible because I guess it's your genes and not your partner's and then some of it's also just a bit of cursing your luck as well." (Father of a child affected by AI)

JMC: *"I do feel the guilt that he inherited it <from Me.>,,,, he's nine now and like he's now going to know what pain is like and whatever it is that" (Father of a child affected by AI)*

The second manifestation relates more specifically to seeking dental treatment and the difficulties and complications that might occur in association with early dental treatment.

JMC: *"There is guilt to do with, and the more the worry then, he is nine now and so, and you're going to keep trying to drag him up from Roscommon so, <Donegal,> a long distance up and down up and down, it's not going to be easy." (Father of a child affected by AI)*

H: *"My little fella then he got one strep throat after another, and of course I put it all back to getting the teeth done, in my own mind,,,, I did feel guilty because he got brutal strep throats to the point where I brought him to the consultant to see should I get the tonsils out." (Mother of a child affected by AI)*

This feeling of guilt appeared more prominent in the father's focus group. Conflicting views, however, were expressed about the degree of this guilt, with some expressing a huge feeling of guilt that extended to affected grandparents, and others denying any feeling of guilt based on the personal lack of control on genetic inheritance and their own positive experience with dental treatment.

JMN: *"I don't have the guilt thing, because he's brilliant in every other way, I'd feel guilty if I wasn't taking steps to help him, that's guilt,,,, It just happens to be especially visible, <because it's, it's there> front and centre,,, like it does get resolved,,, I know the road I had to leaving dentist's chair with my teeth and being able to go out and smile without being conscious, it's a huge thing. And all of our kids will get to that stage." (Father of a child affected by AI- father is affected-)*

This example demonstrated denial of the feeling of guilt of inheritance, however, expresses a feeling of guilt if not seeking child's dental treatment. In addition, it highlighted the expectation that dental treatment over time will lead to a reduction in the degree of visible difference, and thus an improvement both aesthetically and psychologically.

Interestingly, parents found the generous financial support for seeking dental interventions by affected grandparents was a result of their internal feeling of guilt and was driven by worries about their grandchildren’s experience. One parent thought that they want their grandchildren to ‘get what grandparents couldn’t have’.

CC: “ I’d be honest with you, her grandparents feel guilty because they came up with that,, Like the amount of offers, every time we come to Dublin, do you want money,,, And the kids are like, go on, stuff like that. I believe she’s wanting what she couldn’t have.” (Father of a child affected by DI)

F: “My father feels really guilty. He does, and so does my mam, and like when (name) got born with it, Mammy was like right, and we’re going to set up a little Credit Union account and she said I’m going to give a few pound every week.” (Mother of a child affected by AI)

Figure 3-5 illustrates the impact of AI/DI on parents and variable guilt manifestations.

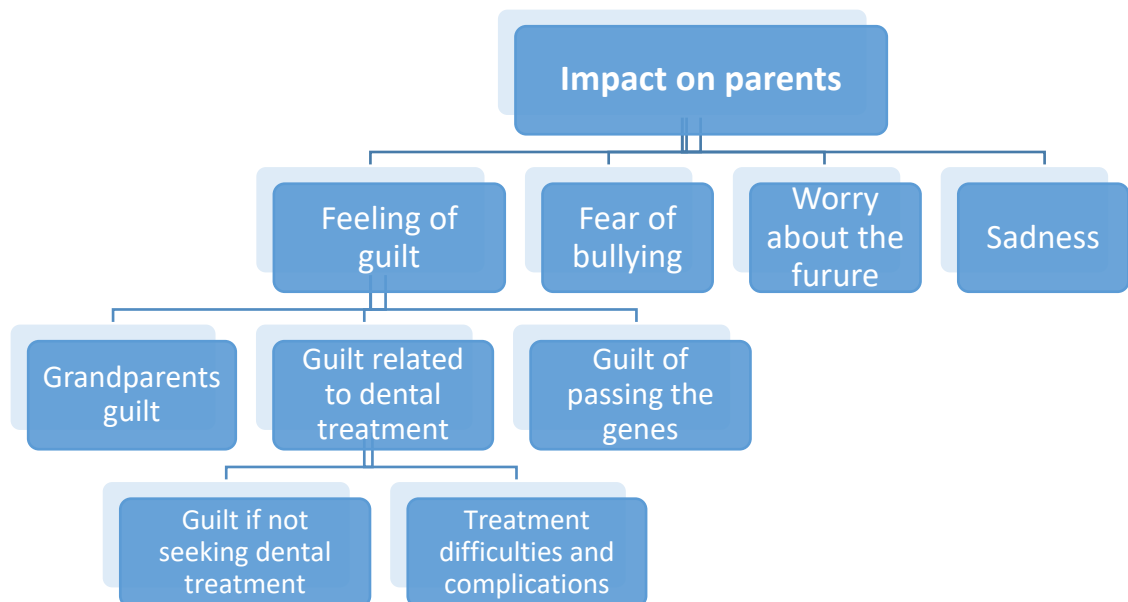


Figure 3-5 The Impact on parents.

3.8 Coping

A number of strategies were employed by parents to cope with having children affected by AI/DI. Within this theme, the following subthemes were evident from the data:

- Whitening toothpaste and home remedies: There was a general knowledge among parents that 'whitening toothpaste' and 'home remedies' offered no help to improve the affected teeth appearance.

J: " Did you try anything else yourself first, some people say there's sometimes whitening toothpastes and all this "

JMA: "<No, no, no.>"

JMN: "If you know what you're dealing with, you're not wasting your time."

Multiple speakers: "<Yeah.>"

(Dialogue between fathers including a father of a child affected by DI and a father affected by AI.)

- Acknowledgment and explanation: 'Acknowledgment' and explanation to affected children and family friends was suggested by several parents. Many parents used the term 'special teeth' to describe the condition to their children.

JMN: "We've taken a different approach on it,,,,, We've just talked about it from day one,,, like me and him are in this together, if that makes sense,,, because I know it's important, little pep talks and stuff like that, so he knows he has special teeth." (Father of a child affected by AI- Father is affected-)

One mother had her own way of describing AI to her child at preschool age by saying: 'the front white part of tooth is not there', though another mother found it difficult to explain AI to her two year old daughter at this very young age. Moreover, parents persuaded their children that seeking dental treatment is a normal part of everybody's life and they clearly denied treating the affected children differently.

D: *"I have a control group, I have twins. [Laughing] Perfect science experiment. So girl boy twins. Girl has pearly whites, boy has, this picture very slight discolouration. I wouldn't treat them all, they all have to have their teeth washed the same way, I have two three-year-olds who come in saying wash the teeth you want to keep and all of this, dental hygiene,,, it's something that we do and that's it. It's going to the dentist, we go to the dentist, everyone goes to the dentist, it's something that's done, <it's like a normal.>" (Mother of a child affected by DI)*

- Positive parents' comments on appearance: Regardless of teeth appearance, parents were careful about their comments to their affected children and were assiduous in giving positive comments.

G: *" He'll brush his teeth, when he was younger and he was, oh look, they're so shiny, but they were terrible, I used to say oh yeah, they are great, great." (Mother of a child affected by DI.)*

- Promise of future definitive dental treatment: Promise of future definitive dental treatment was adopted by a number of parents. Parents who had positive dental experiences referred to their own experiences to reassure their children and they postulated that this definitive treatment is not achievable before the age of eighteen.

JMN: *"And when he's eighteen he can get it sorted, so he knows the story, you know. What I try to say to him is look, this isn't forever, you know Just need to get this sorted and one day you won't even think about it,,, At some point in time the intention is he'll have his teeth like the others <afterwards.>,,, I know the road I had to leaving dentist's chair with my teeth and being able to go out and smile without being conscious." (Father of a child affected by AI- father is affected-)*

- Photoshop of family photos: One parent explained his remarkable coping strategy with his child's dental appearance before having the dental treatment

by whitening his child's teeth in family portraits so that his son will not be embarrassed because of his dental appearance as he grew up.

JMN: " I'll tell you something, you know these family portraits, and we went and did one of those, I got the person who finishes the photo, I got them to whiten his teeth,,,, because I wanted him to have that picture and to be able to pull it out and not be embarrassed. That's the only reason,,,, I said, don't Hollywood, I said I just want you to bring up the shade a little bit." (Father of a child affected by AI-father is affected-.)

- Seeking dental intervention: Mostly, parents were keen to get dental intervention as early as possible and were motivated by several factors that will be discussed under the treatment need section. One mother who didn't seek dental intervention in the primary dentition referred to her unique way to overcome her child's sensation of teeth roughness by letting him " put his tongue on her –smooth- teeth".

G: "So I let him put his tongue on my teeth just to, let him believe the smoothness of my teeth because they're rough." (Mother of a child affected by AI)

- Diet modification: Diet modification was explained in further details under the section of impact on children's dental health and function.
- Feeling grateful not having other major systemic diseases: This positive way of thinking was expressed by several parents and parents perceived the availability of dental treatment as a blessing when compared to other life-threatening or chronic systemic diseases that affect children attending the two main paediatric tertiary care hospitals in Dublin; Our Lady's Children's Hospital, Crumlin and Children's University Hospital, Temple Street.

F: "That's what I always tell myself when I'm driving up here and my heart would be in my mouth and you know, just wait,, I'm not going to Crumlin, I'm not going

to Temple Street.,,, you look at people who have sick children, and you look at that thing and you think, no,,, it's not the worst thing that you could. Its shit, but it's not the worst."

E: "<That's what my> mam keeps telling me, if you were going to Temple Street, <rather than the Dental Hospital.>"

A: "<I do say that a lot, >,,, It's a pain in the neck, of course it is, but you'll get by. You know. " (Dialogue between mothers)

3.9 Treatment Need

Most parents sought dental treatment of their children around the age of two and three years and were concerned about getting dental treatment before their children started primary school.

Several reasons for seeking dental treatment were advocated by parents including mainly psychological reasons attributed to appearance as well as other functional reasons. The main reasons proposed by parents were child protection, achieving normality, and maintaining structure and function.

3.9.1 Child Protection

There was a general expectation that early dental treatment will protect children from 'feeling different' at young age and thus protect their self-esteem and confidence as they grow up. Starting primary school was referred to as a critical period within the life course of the disease as most parents believed that the children's awareness of difference as well as other children's innate negative responses to the appearance increase significantly around this age. Concerns and fears about bullying at school as a main motivating factor for treatment were repetitively referred to by parents in both focus groups.

JMA: " It's not the fact of what the teeth look like, you're trying to protect the child, you know, you don't want them to be slagged, and I think that's why we went for the option when she was so young to get it done, because before she starts school then, that's even what the dentist said, he says you know it's a good time to get it done because she'll know no different." (Father of a child affected by DI.)

C: *"I think it's the fear of bullying."*

B: *"<It's the fear of bullying.>"*

Multiple speakers: *"<Yeah. The bullying.>" (Dialogue between mothers)*

Interestingly, parents believed that young children lacked the capability to respond to other people's comments at this age thus the dental treatment can provide them a shield of protection until they get older and be more capable to respond to people's comments. Parents wanted to 'make life easier on the child' (in the words of participants).

CS: *"I think it's <more about self-confidence and getting the caps done kind of protects them, at least until they get old enough to be confident enough to deal with it and be able to tell people"*

CL: *"It's jeering at three, though, is it?"*

CS: *"It's more you're different, but then that comes later <from five, six> onwards they'll start."*

JMN: *"And kids, kids can be ruthless."*

CS: *"Like you would blame yourself if you knew your kid was getting jeered at school for something that could have fixed, not fixed, that could help the problem. I just want to make life as easy as possible on her" (Dialogue between fathers)*

Apparently, affected parents' concerns of bullying were attributed to their own experience of bullying during childhood and the childhood experience of other family members.

CC: *"Because it's on my wife's side, as she said, she was kind of jeered in school and she didn't want him end up in school and be jeered and not want to go to school, so we got it done before he even went to school, we got it done <when he was three.>,,, I think that was our main thing." (Father of a child affected by DI)*

B: *" Like I don't know I was quite mild with it. But my sister, and she told me to tell you today, when she was a child, she used to think because it was so rare that she was going to be on The Late Late Show, and she said the thought used to get her through because she was really badly bullied over it.*

I: *She was bullied, was she?*

B: *Oh, completely, yeah, she was told she had Sellotape, she was really severe, <Her teeth were really brown,> they were really brown like and at the time they couldn't do much. So she was bad, till she got them crowned in secondary school.*

E: *That must have been hard for her.*

B: *<Oh it was, yeah, I think that's why you feel so much for the children." (Mother of a child affected with DI)*

However, the affected parents' experiences with dental treatment significantly influenced their motivation for early dental treatment of their children. Those who had positive dental experiences showed enthusiastic attitude toward dental treatment.

One father, for example, referred to his affected wife's positive experience with dental treatment:

CS: *" I imagine my wife would have been here as a girl, so she would have had a lot of treatment here as a child, so she was more I suppose up to speed on the issue and wanted the same level of treatment as well." (Father of a child affected by AI)*

And his wife confirmed that and went further to deny any 'fears of dentists':

E: *"All this is because I went through it and I have no fear of dentists, I'm quite happy."*

On the other hand, the affected father who did not seek dental interventions of his child's teeth until he was eight year old explicitly referred to his own traumatic negative dental experience, supported by advice from his family dentist to wait until permanent teeth had erupted.

JMC: *" In my case all I remember is sitting in a dentist's chair coming out and a mouthful of blood and all top teeth were gone in one go. And like I had dentures in, he fit them three weeks later or something like that, and I've had that ever since.,,,, We did bring him to Noel (family dentist) and Noel said look, there's no big rush but there is going to*

be a case where you really have to get them along to the eighteen years of age type of thing.” (Father affected by AI)

Another interesting proposed reason for seeking dental treatment early was the belief expressed by one mother and echoed by another, that children do not recall experience memories before the age of five and so will not recall having had dental treatment. Therefore, parents perceived that early dental treatment could guard children against feeling different when they grow up.

F: “I was assuming I’d get them done at two as well, before they’re old enough to kind of know what’s going on, that’s what I was thinking. Before they’re old enough to talk to you and to be conscious of it, to remember all of it, because he doesn’t remember and that’s a big thing. They forget,,, I asked the last time I was bringing (name) up, I was like to (name) do you remember being in hospital? And he has absolutely no recollection of it.”

E: “Generally speaking, kids forget things that happened before the age of five, they have a learned memory, now the positive or the negatives, sensation, that does impact obviously, but the actual memories, they tend to disappear rapidly.”

3.9.2 Achieving Normality

Achieving ‘normality’ (in the words of participants) or close to it was suggested as a considerable reason for seeking dental treatment.

JMA: “ I just wanted what was best for the child. Just as long as her teeth were happy and they’re, not happy but healthy and looked half-normal.” (Father of a child affected by AI.)

3.9.3 Keeping Structure and Function

“Teeth protection” (in participant’s words) was suggested as another reason for seeking dental treatment. Parents were concerned about keeping the teeth longer in order to preserve the bite and balance in the mouth. Managing infraocclusion of teeth was another functional purpose of treatment though not directly related to AI and DI.

JMA: *“Alluded to there, you know, just thinking of the kids, but also protecting the teeth so the back teeth are very important, that you retain the molars, because the integrity of the molar is very important. So (name) would have had steel crowns put on at around that age (three) to protect the back teeth” (Father of a child affected by DI.)*

D: *“,,, <that was purely to> keep them in place for longer, and then the older guy then had the front few whatever, he was done down here and that again was just purely for functionality, for the bite and for all that.” (Mother of a child affected by DI)*

Issues related to sensitivity and dietary problems as motivating factors for treatment were not particularly prominent in the focus group data.

3.9.4 Willingness to Pay for Primary Teeth Treatment

All children received dental treatment at the DDUH at no cost. However, when parents were asked about their willingness to pay for dental treatment, most parents showed a great willingness to pay for treatment for pure cosmetic reasons.

Moreover, the impact of dental appearance on the child, the disease severity, and expert advice were suggested to influence their willingness to pay. The impact of dental appearance on the child and the child’s self-image were repeatedly referred to as perhaps the most important factor to influence the parents’ willingness to pay.

B: *“If I thought it was having a negative impact on the child,,, If I thought that for the child, it meant the difference between being happy and not being happy.”*

C: *“You’d do anything for your children.”*

B: *“You do. Again, it’s always what the cost-benefits is? Like. Doesn’t matter what I think they look like, it’s what the child thinks they look like and how they feel about their teeth.”*

Again, parents referred to their own childhood experience as a motivating factor for treatment at any cost, as long as they can afford it.

F: *“, I was so self-conscious growing up, if I didn’t have it, you can’t go in and rob a bank and get it, but if there was any way possible that I could make it happen, it would happen.” (Mother of a child affected by AI - Mother is unaffected-).*

Some parents expressed their willingness to pay only if the child needed treatment for keeping structure and function and were not concerned to pay for cosmetic treatment of primary teeth solely as they will fall out anyway and the child will still need treatment in the permanent dentition.

H: "No, not on baby teeth."

F: "<Unless he> needed it like."

H: "Yeah, unless it was a case of you had to do so-, I'd pay. Like the silver panels, you need to get these done because they're going to protect the tooth that's there for whatever number of years, if you don't, the tooth's gonna fall out and it will affect the permanent teeth coming in. But not for aesthetics, I wouldn't." Dialogue between mothers.

3.10 Experience of Treatment

3.10.1 Treatment Choices

Several treatment choices were considered when the child was to receive dental interventions at young age including the timing of treatment, choices between adjunctive pharmacological behavioural management techniques, and choices between various dental interventions.

Most children had received stainless steel crowns to restore and preserve posterior primary teeth, though anterior dental treatment varied between no treatment, composite strip crowns and zirconia crowns. A minority of children have received aesthetic restorative intervention of anterior and posterior primary teeth with zirconia crowns and one child did not receive any dental intervention until the early mixed dentition.

All children of attending parents who received dental treatment at the age of two and three received dental treatment under general anaesthesia while for older children two children were treated under nitrous/ oxygen inhalation sedation, one child's dental treatment was performed without any adjunctive pharmacological behavioural management techniques and others were treated under general anaesthesia.

The affected parents pointed to the general improvement in the understanding of both dental anomalies and the number of treatment options available in comparison to what was available for older generations.

I: " Do you think your own child has the same experience as say your father? "

JMN: "Em, well he's got much better care, there's more options available now,,,"

That's just medical science having developed and an understanding of it." (Father of a child affected by AI- Father is affected-.)

In general, treatment related decisions were based on mutual agreement between parents of affected children with no conflict, however, affected partners and their affected family members were generally more likely to encourage starting dental treatment early.

H: ", because I remember saying oh God, even though we'd decided to do it, I'd say oh God, I'm putting him under, you know, and his whole, all of them that have had it, and even the sibling that didn't have it growing up with him said get it done, it's the best thing to do for him." (Mother of a child affected by DI)

Several factors influenced the parents' treatment choices, including expert clinician opinion, parents' desire to get an aesthetic smile and the impact of the dental anomaly on the child. These factors were explored further under the following subheadings.

3.10.1.1 Expert Opinion

Expert opinion and advice were identified as a particularly critical factor in treatment decisions. This view was held by almost all parents. Most parents relied on the paediatric dentist who was the expert from parents' perspective as the main source of information and the decision maker on most of treatment related decisions, particularly when choosing between different pharmacological behavioural interventions.

H: "I did feel, and I thought long and hard about it as well, anaesthetic, <I did absolutely.>"

I: "What motivated you in the end to go for it? "

H: *“Well Dr AOC advice, I mean obviously I’m always going to take the professional’s advice, she’s not gonna push you into a theatre if it’s not necessary.” (Mother of a child affected by DI)*

JMA: *“Well I think in our case Doctor (AOC) put it to us that this was the best option, and it was the best time to have it done so we knew nothing about it so we trusted her.”*

However, parents’ preferences competed with the expert preferences sometimes in terms of choices between aesthetic and apparently less aesthetic treatment options. Parents’ decisions on these cases will be explained further in the ‘aesthetic smile’ section.

Parents who were unaffected by the condition showed variable attitudes toward electronic and paper searches to get more information about the dental anomaly and treatment options. Parents who had an experience of reading about it from different sources were frustrated with the type of information they obtained in terms of limited amount and ‘depressing’ quality. The most frequent finding that the parents recalled from this searching experience was the depressing photos of severe cases that did not necessarily represent the appearance of their child’s teeth.

A:’ *“I wish there was more information on it, <it’s very hard to get,>, I remember trying to get as much information as possible and I got my own dentist to give me a document from a dental manual and I got another dentist to give me a document, just so I could read about it. But there’s so many different types of cases.”*

C: *“No, I’m not doing it again, because the images, I was tearful when I seen the images like. I was putting myself through things I shouldn’t have been putting myself through, you know through going on the internet.”*

Interestingly, some parents did not care about reading more about it and were solely dependent on the expert input due to variability in presentation and management of dental anomalies.

D: *“I sound like I’m probably disinterested in my children’s teeth; I haven’t Googled anything. I never Googled it. I just do everything Dr (AOC) says I go with and if she advises*

to do X, Y or Z,,, as you said earlier on, they're all different stages, as in all our kids are obviously affected to a different degree."

H:" <I prefer not to know,> as in I don't really like thinking about it, you know, I just want to deal with it appointment by appointment."

3.10.1.2 'Aesthetic Smile'

Another important factor in treatment decisions was the "aesthetic smile". Not surprising, all parents wanted beautiful aesthetic smiles of their children and were completely against getting silver (metal) teeth.

*E: "My preference would have been for a white one, crowns, Aesthetic, because it doesn't stand out, and then it kind of brings him closer to where you ultimately want him to be."
(Mother of a child affected by AI)*

However when the expert advice contradicted their preferences, they were comfortable with the dentist advice as long as the silver crowns did not appear when the child's smile. Parents, in general, accepted the silver stainless steel crowns (SSCs) to restore the posterior teeth based on the expert advice of strength and durability.

A: "<You always go with whatever's gonna be the strongest.> "

JMN: " I felt well, if that's the most comfortable thing for him, I wasn't too worried about aesthetics of the back teeth. Like day-to-day, even with the bottom teeth, you know, some people don't show their bottom teeth that much."

Interestingly, one couple of parents who initially insisted on getting white restorations, on every tooth, changed their position when the dentist advised the difficulty in getting the lower anterior teeth crowned for technical reasons. They accepted no treatment of these teeth, as it did not show in the child's smile but insisted on having the rest of the teeth crowned with white zirconia crowns.

H:" My son had the bottom four, they were so tiny and his baby teeth, they couldn't crown them, so they would have been polished up. It did lighten them a little bit, but you

can very definitely see, but it wouldn't, and when he's smiling you'd only really ever see the top."

Parents' acceptance of different anterior teeth restorative options (zirconia crowns, composite strip crowns, polishing, and no treatment) varied when they were shown photos of these different interventions. Not all parents were familiar with the restorative options and only had one of these interventions provided by the dentist without understanding the choice, but trusted the opinion of the dentist.

B: *"But there was no option for cosmetic reasons other than capping, wasn't there? I didn't think there was. Well it was either to get it done or not to get it done."*

Interestingly, some mothers perceived the composite strip crowns restored teeth as a mild variation of AI that did not receive dental intervention and the same mothers were not against using composite strip crowns to improve the appearance of severely affected teeth.

F: *"Well compared to what I have, I'd probably be delighted yeah, but you know, if I had a choice."*

The cost- benefit of these different restorative options was suggested to determine the parents' decisions, with the cost- benefits being viewed as whether the restorative option will need anaesthesia or not and whether that is local or general anaesthesia.

H: *"I'd go for it, like if there was no anaesthetic or anything like that, if it was a simple, wham, bam, thank you ma'am, I'd go for that one. But if it was going to involve being more medical type intervention."*

C: *"There's negatives with every treatment and responses to that, so what is the balance and what and how does it balance out and, for the child? Not for me."*

One mother, for example, chose composite strip crowns and stainless steel crowns (Hall technique) for her four year old child based on the ability to provide treatment without the use of local or general anaesthesia. This child had spacing between his upper hypoplastic anterior teeth that allowed treatment of anterior teeth without anaesthesia and Hall technique was utilised to restore his posterior teeth with SSCs.

E: " I was like, no, not doing it because it was general anaesthetic, I'm still going like there's, is it worth, the cost-benefit of general anaesthetic versus aesthetic,, the veneers,, That was in the chair downstairs, that's safe,, and my little fella got the silver ones recently, no anaesthetic hardcore, he's five, so we were able to separate the teeth, and over a week, and they just slot them" (Mother of a child affected by AI)

3.10.1.3 Impact on The Child

Parents referred to the impact of the dental anomaly on the child to influence their treatment related decisions. This impact was related to the severity of the dental anomalies and associated psychosocial implications as well as the child's own preferences.

CS:" There's no right or wrong answer, I mean I think it's just giving parents the choice and children the choice because it'll depend partly on the child if they're having issues at school or anything like that if it's, what's happening with the child, what's best for the child, and it might be the, the full veneers or it might just be some of the teeth."

Generally, affected children liked the stainless steel crowns.

D: "He was delighted. He's getting more silver done,, Doesn't bother him. He was very popular for a while after he got it done, they were all like my God, Mammy, silver teeth."

I: "Do you think there's a difference between girls and boys on that one?"

D: "No, not at all."

However, in the child's account of the events surrounding the procedure of getting the stainless steel crowns the child might dislike to have them again:

E: "He's only five, and he'd never had any problem with the coloured teeth, but the silver teeth, and I think it may have been to do more with the process than the aesthetic of it, he had fallen, knocked loose four baby teeth, and very shortly, we had been into the dentist's on the Friday,, so in his mind this is why he had the silver teeth, was because he fell."

3.10.2 Obstacles Related to The Experience of Dental Treatment

3.10.2.1 Treatment Under General Anaesthesia

Parents talked about their experiences with their children of dental treatment under general anaesthesia (G.A.). They highlighting the difficulties they have faced at several stages including making the decision for treatment, the time of induction, during and after the operation. These difficulties were particularly prominent in the mothers' focus group.

Making the decision for treatment was not easy for parents and they expressed different perspectives in relation to the "cost effectiveness" of general anaesthesia for dental treatment at young age.

CL: *" Because it's really, it's not an easy choice to even elect to have them done, because you could not do it then they won't be protected and the aesthetics won't look as nice, but you can decide not to because giving a three-year-old a general anaesthetic is not something <you actually do lightly>"*

Irrespective of the parents being affected or not, some parents believed that the aesthetics and dental protective outcome of dental treatment is worth it while others preferred to postpone the dental treatment for aesthetic reasons until the age of five due to health related concerns of general anaesthesia at young age.

F: *"I shouldn't say this because I'm putting her through a general anaesthetic for cosmetic reasons but I will get her teeth crowned as soon as I can.,,, what makes me do it as well is because I can do it, whereas we didn't have that opportunity when we were children."*

E: *"I was like, no, not doing it because it was general anaesthetic for this.,,, I'll wait till he's five. So he had done playschool and junior infants, but as long as that, I'm still going like is it worth, the cost-benefit of General anaesthetic versus aesthetic. And that's, if I can prepare for it to have the answers, he doesn't need to have the general anaesthetic."*

The data revealed that most of children had no experience with general anaesthesia before receiving this dental treatment and this added to the difficulty of making the

treatment decision. In addition, parents referred to being blamed by the general society for putting the child through the risks of general anaesthesia only for dental treatment of primary teeth that will be lost eventually.

D: *"Oh, do you know, it was awful because none of my kids till then had ever been in hospital or anything."*

CC: *"So nervous like, because we'd never experienced anything like this in our lives."*

F: *"And people were saying why are you putting the child through that? They thought I was a really bad mother, why are you putting the child through that when it's only baby teeth and he's going to lose them?"*

Parents expressed concerns such as leaving the child alone, the lengthy general anaesthesia treatment sessions, the fear of the child not waking up, overnight stay and immediate post general anaesthesia complications. These concerns conspired to foster parents' nervousness and worry in the context of dental treatment under general anaesthesia.

C: *"While she goes asleep, I remember sitting on the bench like on the bed with her and she's looking at me with like the eyes on her, <Mammy, what's happening? >,,, I was walking out of the room, and I can't believe I'm walking and leaving my daughter in there like, it was just horrible."*

H: *“, And then the minute he goes asleep, I just burst in tears,,, So he was under for about three hours, two and a half, and I kept thinking oh my god, there's something after going wrong or what if he's not waking back up."*

Dental treatment under general anaesthesia lasted for two to five hours according to parents. Some children had to stay overnight and immediate post general anaesthesia complications were mostly vomiting and throat ulceration. However, parents whose children had dental treatment under general anaesthesia a second time believed that treatment duration and postoperative complications were considerably less compared to the first dental treatment episode. These parents were keen to have the same dental treatment for their other affected children.

F: *“ He did get quite sick after the first one because they took so many x-rays and everything, and he had a tube down and then he got ulcers in his throat and vomiting and I feel really bad that I put him through it again, whereas the next one they have all that information and they’re kind of just finishing off. But I’m gonna do the same with my daughter because he has beautiful teeth, now.”*

The data revealed interesting parental beliefs of an association between dental treatment under general anaesthesia and other general health issues. One father believed that greater forces are used when seating stainless steel crowns under general anaesthesia and correlated that with an intermittent neuropathic facial pain his child developed few months after the operation.

JMN: *“,,, Now our little guy started experiencing pain in his jaw up here, not in the tooth,,, So he has had this issue of intermittent pain every four hours, and we were feeding him the Calpol, Nurofen. I think because the kids are under general anaesthetic, there’s greater force being used.”*

Another mother related her son’s experience with recurrent strep throat infections to the intraoral general anaesthetic tube and eventually she felt guilty for putting her child through general anaesthesia to treat his primary teeth.

H: *“ My little fella, He got one strep throat after another, and of course I put it all back to getting the teeth done, in my own mind,, but I actually did feel very guilty for a while,,, because he got brutal strep throats to the point where I brought him to the consultant to see should I get the tonsils out.”*

3.10.2.2 Treatment Under Nitrous Sedation

Treatment under nitrous sedation for older children was generally an acceptable method for parents. One couple referred to their child immediate postoperative complications of dizziness and nausea, however that did not impact on their willingness to continue dental treatment under nitrous sedation.

JMA: *“The only thing was, just a little bit when he left here, bit upsetting the tummy, that type of thing,,, If he was asked again would he go back in with the happy gas I think*

he would, he was okay, I mean he knew himself that he wasn't quite right afterwards but I mean an hour or two down the road he was fine."

3.10.2.3 Child's Coping with Dental Treatment

The experience of dental visits by affected children varied. Some affected children 'love visiting the dentist' while others show dental anxiety and limited cooperation.

JMA: *"She loves that because she gets a day out in Dublin you know, we go and do all the good things afterwards, so I think she loves coming to the dentist."*

C: *"She'll come in and she'll jump up on the chair and she'll turn on the lights and the whole lot."*

Several anxiety provoking sensory stimuli at the dental clinic were suggested by parents of anxious children. Specifically, parents referred to their children's fear of needles, sensitivity to scaling, and unpleasant experiences with intra-oral photography using occlusal mirror, or taking x-rays of their teeth.

A: *"She's very afraid of needles and everything."*

JMN: *"Plaque will build up at the back of the teeth and stuff So that has to be cleaned out and he's quite touchy about that, so he was building it up a bit in his head."*

F: *"(Name) hates getting the mirror, you know the way they take so many pictures,,, He also hates the x-ray. There's a certain one at the back, it's the things being stuck into their mouths. Any objects."*

Parents placed considerable value on rewarding their children's behaviour at the dentist, with the belief that children can forget the negative dental experience easily once rewarded.

CS: *"Maybe not enjoying it too much when they're in the chair but it's a day out in town and getting some stickers and being on the bus home and that's what they remember, I think."*

Most parents will let their children take the day off school, some will take them on a trip or visit the toys store and 'sugar coat the day':

JMN: *"He screams his lungs out downstairs. But we always kind of sugar coat the day then. Trip to the park or something."*

D: *"And I don't let him go to school, he kind of gets the day off and we make a fuss of it, to try and make it not be the ordeal."*

E: *"When we get to go to Dublin, we go to the Disney Store."*

However, other parents 'do not make anything of it at all'.

G: *"We don't go to the Disney Store. [laughing] We go straight home and, I just don't make anything of it at all."*

Dental treatment for children with limited cooperation was carried under general anaesthesia. Nevertheless, general anaesthesia was not a pleasant experience for some children. Parents referred to the mask induction and others referred to the intravenous line access as being threatening and unforgettable by their children.

A: *"The anaesthetist said to her I'm gonna just put a gas, you're gonna feel a little bit wobbly and she went don't want to feel wobbly,,, I had <to hold her hands> while they put the mask over her, because she was fighting,,, she said afterwards, she says I hope I never have to get those crowns done, she still brings it up."*

H: *" , that when I got the Lego in my hands, the needle, that's what he remembers, and I said if you had to get it again well could I get something else other than the Lego? I just <don't want the Lego.>"*

3.10.2.4 Time Off School and Work

Affected children need continuous dental review and therefore parents alluded to the time implications of attending ongoing appointments, both for affected children and their parents. This time implication was complicated by parents' commitment to reward their children following the dental visits as well as the need to travel a long distance for families living outside Dublin.

C: *"It's my day off. A half day. "*

JMC: *"He is nine now and you're going to keep trying to drag him up from Roscommon so, <Donegal,> a long distance, you know, and it is, as we do, and make a day out of it then try and cheer it up in some way."*

Parents, however, did not mention any negative effect on schooling or employment. One father followed that up by suggesting that the number of appointments as well as the gap between appointments decreased by moving from the treatment phase to the review phase until the eruption of permanent teeth.

B: *"She had more visits like earlier when she got the crowns in and then say one or two had fallen out and I had to come back and she had to get them redone and stuff like that, but like now there's like three-month, four-month gap in between appointments. So it's settling down, she's not here as often as she was."*

3.10.3 Satisfaction with Early Dental Intervention

The participants demonstrated general satisfaction with early dental intervention of their children's primary teeth. Apparently, this satisfaction was threefold, the positive psychosocial impact on the child, the improved aesthetic appearance and the feeling of smoothness of the teeth.

Talking about the psychosocial impact on the child, one parent who sought dental treatment of his child when he was two years old believed that this early dental intervention has achieved his aims of protecting his child, precluding any feeling of difference by the child and allowing him 'to grow up to be confident'.

CL: *"I can see the difference in my son, he's not self-conscious, he is an extremely confident little man and, I think by the time his front main ones come down, I think if someone comes round to him and says something, I think he'll turn back around and say well, do you know what, I don't care what you think, <he'll have the resilience built up.>*

JMC: *"<Exactly.>"*

CL: *"he's kind of had that protection and been allowed to grow up to be confident."*

This view was echoed by another parent who said:

F: *"He was so young, it was before he was two he got crowns, so he doesn't remember that. So he doesn't know anything different."*

Parents were delighted with the improved aesthetic appearance and felt that people did not note any difference in their children's teeth because of the early dental treatment.

F: *" I'm gonna do the same with my daughter. Because he has beautiful teeth, now."*

I: *" Do people say anything to you about your child's teeth?"*

CC: *"No. Never. Because no-one has ever seen them really because he was so small, we got it them done straight away."*

Parents of children with hypoplastic AI placed a considerable value on the feeling of smoothness experienced by their children after treatment.

B: *" They'd be very sharp and I know when my son got his crowned, what took, the biggest adjustment was the feeling of the smooth surfaces."*

JMA: *"The first thing that (name) said, at nine years of age, was the smoothness when he was rubbing his tongue, that was the very first thing."*

In all cases, apart from one mother, parents denied any current feeling of regret in relation to treatment decisions for their children. This mother regretted that she delayed dental intervention for her older affected child until he was 10 years of age.

H: *" I'm sorry that I didn't start the ball rolling sooner for the older two, I don't know would they be any better off now if I had done it. Well I can only judge it on the younger, she was about three, three and a half, and so the older guy would have been ten. So maybe to get him in sooner."*

F: *"now I'm so delighted I did it, and at least it gave me a break for those two years."*

G:*"I wouldn't do anything differently."*

4 Discussion

4.1 Discussion of Main Findings

This study has explored the experiences and perspectives of both mothers and fathers of young children affected by AI and DI. Both mothers and fathers nowadays share parental duties in providing physical and emotional care to their children, though to a variable extent in different families. Nevertheless, fathers were seldom included in parental perception studies and such studies were dominated by mothers (Levy et al., 2005; Woo et al., 2005; Naidu et al., 2012; Pani et al., 2013; Nelson et al., 2012a). There is a growing body of research to suggest that father's involvement in raising their children is increasingly expanding (Cabrera et al., 2000; Yeung et al., 2001), and a considerable increase in children's time spent with fathers has been reported (Yeung et al., 2001). Although these findings emerged from US investigations, they reflect the social perspectives in most western countries, including Ireland (Ralph, 2016) and several Asian countries (Pease and Pringle, 2001).

Mothers' and fathers' views on their children's oral health related quality of life and treatment need can vary. While fathers and mothers tend to agree on their perception of the oral health related quality of life of their children at group level, they are reported to disagree at family level (Zhang et al., 2007). Mothers may show more accurate knowledge of their young children (2-6 years) when compared to fathers (Pani et al., 2012), but this does not lessen the value of assessing the perceptions of both parents. In fact, there is evidence from behavioural research to suggest that the maternal judgement of preschool children behavioural problems tend to be influenced by the mother's own psychological functioning which might result in overestimation of child's problems (Hay et al., 1999). In addition, a gender variation in the perception of the social and psychosocial impact of their own oral health has been previously reported in men and women (Mc Grath and Bedi, 2000).

The decision for treatment can be led by mothers, fathers or both. In addition, in most cases, males with X-linked Amelogenesis Imperfecta (including some affected fathers) experienced more severe dental presentation than females in their families (Crawford

et al., 2007). Exploring the fathers' perspectives alongside mothers' perspectives is paramount to reveal the parents' views from all aspects.

Parents were divided into two focus groups by gender, a group of fathers and another group for mothers intending to provide more comfortable group dynamics and aid the in depth exploration of the topic. Evidence from social research suggests that men and women interact differently in group situations and that they interact differently in mixed gender groups as opposed to same gender groups. In comparison to same gender groups, concerns about interpersonal relations in mixed gender groups are greater and might limit the diversity of opinions expressed. Women in mixed gender groups tend to be less dominant than in all female groups. Men might speak less about themselves in same gender groups due to increased concerns about status and competition, however, they are generally less likely to conform to group pressure (Stewart et al., 1990).

In this study, mothers and fathers highlighted the emotional and psychosocial challenges experienced by families from the time of tooth eruption. Both mothers and fathers expressed concerns about their children feeling different and shared similar concerns in relation to the impact of these dental anomalies on their affected children.

This study agrees with the contemporary literature demonstrating that dentofacial aesthetics is considered important during childhood starting from a very early age and without any gender difference (Di Blasio et al., 2009; Rumsey and Harcourt, 2007; Klages and Zentner, 2007). This study revealed parents' beliefs of increased worldwide media driven social expectation of white well aligned teeth that extends to affect young children which correlates with previous reports in adolescents and adults (Baldwin, 1980; Mattick et al., 2004; Newton and Minhas, 2005; Kershaw et al., 2008). This society pressure is significantly increasing over time. Two major contributing factors to the influential effect of social pressure emerged from the parents' narratives; the social norms from cultural perspective and the level of caries nationally. However, their belief of an appearance –associated negative social reactions varied based on their own variable childhood experience as well as their children's experiences.

In this study, parents reported that their children's self-perception of "being different" started to develop in conjunction with the eruption of primary teeth and before or approaching three years of age. This finding supports the growing evidence from children's developmental psychology that self-perceptions starts to develop as early as two to three years of age (Rumsey and Harcourt, 2007; Marik and Hoag, 2012; Tremblay et al., 2011; Tatangelo et al., 2016).

It must be acknowledged, however, that the findings reported here are based on parents' perspectives which might be distinct from the child's own reports especially in terms of emotional and social wellbeing of affected children (Barbosa and Gavião, 2008b; Eiser and Morse, 2001; McGrath et al., 2004). In a qualitative study involving AI affected older children (10-16 years), the majority of children self-reported that they started to notice the altered dental appearance around 6 years of age (Parekh et al., 2014). Another qualitative study of children (10-15 years) reported that the transition to secondary school was the age at which developmental enamel defects became a concern (Marshman et al., 2009). Children involved in those studies were older than affected children in our study. Younger children may lack the linguistic and cognitive skills to communicate their thoughts and perceptions, which might explain the lack of child's self-reported studies for younger age groups. In addition, child's perceptions are known to change over time as part of the social, emotional, cognitive and language development (Barbosa and Gavião, 2008a).

The way the child views himself (self- concept) is hypothesised to develop over time as a function of social interactions and interpretations of the evaluative reactions by others (Marshman et al., 2009; Argyle, 2017). The number and consistency of social reactions, qualifications of others from the child's perspective, and the extent of child's interest to attain positive attributes can all contribute to the extent of social reaction effect on self-rating (Argyle, 2017). This may explain differences in age at which child's self-consciousness starts as well as the different impact on different individuals.

The exposure to the concept of " ideal beauty" through media from a very young age might increase the vulnerability for children's dissatisfaction with appearance, though its effect remains unclear (Tatangelo et al., 2016). A systematic review on body image and body dissatisfaction have identified three sociocultural factors to influence the

development of body dissatisfaction of preschool children (age 3-6 years old); verbal messages by parents, verbal messages by peers and media. According to this review, parental influence remains to be the most important factor of development of body dissatisfaction at preschool age with peers being more influential around school age (Tatangelo et al., 2016). Nevertheless, the interaction between these social factors and individual factors will determine the impact of altered dental appearance on the way the child views himself.

Both mothers and fathers in our study believed there is an increased emphasis on appearance as children approach school age due to increased social interactions and comparison with peers. It was apparent from focus group transcripts that young children at preschool and primary school age could notice affected teeth and judge and react negatively to affected individuals. (Soares et al., 2015). Parents in this study also supported these concepts with various examples from their childrens' experiences as well their own personal experiences in childhood. Many of them reported being teased as a child. Almost all parents believed that changing social groups like starting school, or encountering new people on holidays could be particularly challenging. These observations are in agreement with what was previously reported for individuals with visible body disfigurement (Rumsey and Harcourt, 2007).

A major finding of this study is the parental perception that starting primary school was a major cornerstone in the child's development of self-concept, which correlates with previous reports for patients with cleft lip and palate (Abualfaraj et al., 2017). A repetitive theme of fear of bullying was dominant among almost all parents. Self-concept can impact on the quality of peer interactions, coping styles, academic achievement, self-esteem and overall behavioural and psychosocial development (Marik and Hoag, 2012). Although standardised protocols are developed for surgical repair of CL(P), no such protocols are currently available for management of AI and DI. Our study indicated that, from parents' perspective, the aesthetic management of dental appearance at preschool age can yield a significant positive psychosocial impact on affected children.

A recurring theme when discussing the impact of AI and DI on children and parents was the range of individual and collective coping strategies adopted by parents to help them

deal with the condition both in the short term and long term. These could be categorised into 'emotion focused coping' and 'problem focused coping'. Both coping categories were frequently identified to be employed by parents of children with cleft lip and palate as well as parents of long-term conditions (Nelson et al., 2012b).

'Emotion focused coping' employs cognitive reframing of the condition. In this study, this was seen in the parent's focus on the present rather than on the lifelong dental treatment need and future uncertainties, comparison with others felt to be less fortunate than their children due to major systemic diseases, and reframing the condition when describing it to their children using the term 'special teeth' and when describing dental treatment as a normal part of everyone's life.

'Problem focused coping' employs taking actions and that was seen primarily in seeking early dental assessment and intervention. In addition, measures were taken by parents in relation to future adult definitive dental treatment of their children including preparing the child to anticipate progressive and continuous dental treatment and investing family savings.

Another example of problem focused coping was parental control on their children's dental condition by altering their children's diet. Parents increased emphasis on dental issues both for their affected and unaffected children but the discussion in relation to children's diet revealed some parents' misconception about cariogenicity of food e.g. 'Fat-containing foods such as chocolate and ice cream are not cariogenic'. It is important that the dentist ensures that parents and children understand basic preventive oral health education and diet advice provided, in addition to specific advice relating to their dental anomaly. The dentist needs to be aware of the variable coping strategies adopted by parents and support families in coping with these dental conditions.

Worries about bullying and its impact on the child's wellbeing from tooth eruption led parents to seek early dental care in an attempt to help their children. Such treatment was viewed as a way of achieving normality and 'protecting children' from feeling different and/ or experiencing negative social reactions, thus facilitating child's social inclusion. No parent regretted their decision to obtain dental treatment for their child. These findings mirror those in the cleft lip and palate literature for parents of children

from infancy to early adulthood (Nelson et al., 2012b; Nelson et al., 2012c), however, has not been previously reported for parents of young children with dental anomalies.

All parents believed that 'baby teeth are important' and, regardless of family history, almost all parents exhibited a joint persistent attitude to get expert assessment, advice and treatment as soon as the condition was detected even if the teeth were asymptomatic. One couple did not seek referral for early dental intervention of their child's asymptomatic primary teeth, however, this couple monitored their child's teeth through their own family dentist. The discussion with this couple exposed a previous negative dental experience of the father, himself affected by AI, that obstructed him from seeking referral for dental intervention of his child's primary dentition.

The results of this study indicated that the personal experience of parents affected by AI/DI plays a pivotal role in parent's judgements of their children's teeth and perceived need for dental treatment. Fear of bullying as well as the childhood experience with bullying of affected parents motivated them to seek early dental interventions. In addition, affected parents who had positive dental experience showed enthusiastic attitude toward dental treatment while negative dental treatment experience of one affected parent hindered him seeking early dental interventions for his child. This is a novel finding that has not been reported previously in the AI and DI literature.

Previous negative dental experience of mothers and grandmothers was reported to adversely affect their attitude toward their children's and grandchildren's oral health (3 months- 20 years) in a qualitative study conducted in the island of Saipan (Riedy et al., 2001). This contrasts with another study that assessed parent's attitudes toward dental treatment of their children (< 6 years) under G.A. due to caries in British Columbia, Canada where previous negative experiences of parents appeared to be less relevant (Amin et al., 2006). The findings from our study supported the evidence on the impact of parental negative dental experience, yet revealed new insights on the impact of the childhood experience of the parent with bullying on parents' attitude to the dental care of their young children.

Almost all fathers and mothers exhibited a desire to achieve an aesthetic smile for their child which supported reports of high parental demand to improve the aesthetic

appearance of discoloured anterior primary teeth in multiple countries (Holan et al., 2009; Woo et al., 2005). When the aesthetic appearance of teeth is altered due to caries or trauma, 87.3% of parents advocated dental treatment to save primary anterior teeth even if the chances for success were only 50% (Holan et al., 2009). When considering parental preferences for care of primary teeth, social and cultural norms must be noted, although there are no previous reports from the Irish culture on parental attitude to care of the primary dentition.

Dental treatment at the DDUH is provided at no cost, however, most parents expressed a willingness to pay for aesthetic management of primary dentition. This willingness to pay appeared to be mainly driven by the psychosocial impact of altered dental appearance on their children. They also reported taking steps to plan for possible future expenditure related to dental treatment of AI or DI.

Although the main themes that emerged from both focus groups were similar, certain concepts were more prominent in one group over another. While, the feeling of guilt was more prominent in the fathers' focus group, the emotional stress and guilt associated with treatment under general anaesthesia were more prominent in the mothers' focus group. This might be attributed to a gender difference in parents' feelings and/ or their ability to interpret their emotional stress.

To the best of our knowledge, this is the first study to report parental feeling of guilt due to inherited dental anomalies which appears more likely to affect fathers of young children. Parental guilt in association with other oral health problems and/ or treatment needs have been reported to be 22.8%, 35.8%, and 24% respectively (Gomes et al., 2014; Carvalho et al., 2012; Carvalho et al., 2018).

The likelihood of feeling guilty was reported to increase with the severity of dental caries (Gomes et al., 2014; Carvalho et al., 2012; Carvalho et al., 2018) and toothache of preschool children (Gomes et al., 2014; Carvalho et al., 2012). Parents' perception of poor child's oral health, severe dental traumatic injuries (avulsion and luxation) and discolouration were identified as contributing factors for the parental feeling of guilt in a population-based cross sectional study (Gomes et al., 2014). Nevertheless, no significant association between parental feelings of guilt and dental trauma or

malocclusion was found in another cross sectional study (Carvalho et al., 2012). Increased parental guilt with increased severity of dental caries and/or severity of dental trauma could be attributed to multiple issues including the associated pain the child might experience, the increased treatment need, loss of school days, and to the fact that it would be more recognisable (Gomes et al., 2014).

An important distinction must be highlighted here as the feeling of guilt of parents of young children affected by the inherited dental anomalies (AI/DI) appears more complex and distinct from feelings of guilt due to dental caries. Parents feel responsible for their child's health and acknowledge that caries and its associated consequences can be avoided and prevented which contribute primarily to their feeling of guilt and fear of being blamed for their children's oral health problems (Gomes et al., 2014; Amin et al., 2006; Arora et al., 2012). A very recent population based study involving 1313 parent-child pairs found a significant association between parental feelings of guilt and the presence of caries and parents belief that their child has an oral problem or that this problem could have been avoided (Carvalho et al., 2018). In contrast, inherited dental anomalies are inherited and not related to oral health behaviour of parents.

Our study provided a fascinating insight into the diverse aspects for this parental feeling of guilt in conjunction with dental anomalies. Parents attributed their feeling of guilt to passing the genetic defect to the child and the consequences the child might experience as well as the difficulties associated with dental treatment. In addition, a feeling of guilt was anticipated if dental treatment was not pursued.

In this study, the extent of guilt was variable though more common among fathers. Some affected fathers attributed it in part to passing the genetic defect to the child and the fact that it is not preventable and others denied any feeling of guilt due to lack of control on genetics. This can be best explained by different experiences of affected parents. Furthermore, and similar to previous reports, paternal dental anxiety might contribute to an increased feeling of guilt in relation to the child's dental problems and associated treatment need (Goettems et al., 2011). This parental feeling of guilt might be established based on parental negative impressions about dental treatment and the anticipated difficulties that the child will experience.

The parental feeling of guilt of not pursuing dental treatment of their children indicates the perception that seeking dental treatment is consequently seen an indicator of parental competency. This finding is replicated in the cleft lip and palate literature. A perceived parental moral obligation to be 'good parents' by pursuing early surgical treatment of children's cleft lip and palate was previously reported (Nelson et al., 2012c). Similarly, in the orthodontic literature, 64% of 674 parents seeking orthodontic treatment of their children's (age 7-18 years) reported that they were motivated by future worries of being accused by their children for neglecting parental duties (Wędrychowska-Szulc and Syryńska, 2009).

Unlike paternal guilt evident in fathers' focus group discussion, feelings of 'fear', 'worry', 'concerns', and 'guilt' in relation to dental treatment under G.A. were more dominant in the mothers' focus group. Amin and co-workers previously reported a similar result in a qualitative study in which mothers showed higher levels of anxiety compared to fathers in relation to dental treatment under G.A. (Amin et al., 2006). In our study, mothers expressed higher levels of G.A. associated emotional stress and anxiety at all stages including the stage of making the decision for treatment, the time of induction, during and after the operation. That is possibly due to that women are more sensitive, more anxious and better able to interpret emotions than men (Stewart et al., 1990).

Having mothers in the same group seemed to provide a comfortable environment for detailed disclosure of all associated feelings that appeared relevant to almost all participated mothers. Maternal anxiety seemed to increase immediately prior to G.A. This increase in paternal anxiety (no details on parents gender) is mostly influenced by baseline parental anxiety and child's distress at assessment visits (Balmer et al., 2004).

Two important factors seemed to complicate the parents' decision for treatment under G.A., the first being experience of the child with general anaesthesia and the second social blame for putting the child through the risks of general anaesthesia for restoring primary teeth. The dentist needs to acknowledge all these factors contributing to the emotional burden of dental treatment under general anaesthesia and support parents at all stages of treatment.

Interestingly, the feeling of guilt associated with dental treatment under G.A. appeared as temporary and more related to the context of putting the child to sleep and the risk of associated comorbidities as developing a sore throat and immediate postoperative complications (nausea and vomiting). Almost all parents acknowledged the hazardous risks of G.A., however, they did not regret dental treatment under G.A. and were keen to have the same dental treatment for other affected children. This decision to repeat G.A. if necessary has been reported in two previous studies (El Batawi, 2014; Vinckier et al., 2001). It must be noted that experiencing a previous G.A. with other siblings did not seem to reduce associated parents' emotional distress of a current G.A. (Amin et al., 2006).

After dental treatment, parents reported an overall improvement in the child's wellbeing similar to studies reported previously following rehabilitation under GA for dental caries (Cantekin et al., 2014; Knapp et al., 2017; Yawary et al., 2016; Jankauskiene and Narbutaite, 2010; Baghdadi, 2015).

The psychosocial and aesthetic impacts were more frequently described in our study compared to the functional impacts. Parents believed that early dental interventions precluded child's 'feeling different' or being seen or treated differently. An important and previously undescribed outcome for those who had early partial/ full mouth restorative treatment was that they did not notice that their teeth were different until getting their permanent teeth. Another novel positive outcome after dental treatment of children's primary teeth affected by hypoplastic AI, was the satisfaction with smoothness. This outcome was highly valuable for these children.

Some parents in this study reported that G.A. dental treatment was a difficult experience to their children. Some parents referred to the mask induction and others referred to the intravenous line access as being threatening and unforgettable for their children. This threatening impact of intravenous line access and associated discomfort was interestingly only once reported in another qualitative study on the children (6-11 years old) experiences with dental extraction under general anaesthesia. In that study for some children, the discomfort associated with IV cannula was greater than that associated with extractions (Rodd et al., 2014). Parents in our study did not mention children avoidance of dental visits as a consequence of unpleasant experience with

treatment under G.A., only that some children did not want to have the IV cannula specifically.

Whether affected by same condition or not, parents appeared to rely solely on the professional paediatric dentist in making all treatment related decisions. This echoes the work of Nelson(2012) with parents of children with cleft lip and palate (Nelson et al., 2012b; Nelson et al., 2012c). Placing trust in the specialist can be viewed as a way to resolving their feelings of anxiety in relation to their children's treatment by constructing a sense of 'being in the right hands'. Parents relied on the paediatric dentist to get information and advice on the short term and long term needs of their children. Professional opinion on decisions between different types of treatment and how it should be provided was essential. Hastings and co-workers (1994) previously proposed that dentists have the greatest influence on parents' decision to use general anaesthesia.

Not surprisingly, the findings from this study indicated that parents evaluated the dental competence of the dentist and this influenced their subsequent decisions. Parents were reluctant to accept recommendations by general dentists that no treatment of primary teeth be provided. Many sought referrals to DDUH for specialist treatment. Parents strictly followed the paediatric dental specialist advice even in cases where the recommendations were for a less aesthetic option from parents' perspective as in the case of posterior SSCs. Trust in the dentist, dental competence and good communication can influence parents' treatment related decisions (Woo et al., 2005). This places the responsibility on the paediatric dentist to understand the parents' own motivations for treatment, social considerations and the affected parents' experiences on an individual basis, provide adequate information (based on the best available evidence) to enable parents to provide truly informed consent for the dental care of their child, and most importantly act in the best interest of both child and family.

Through the use of photographs within the focus groups, it was apparent that all parents were familiar with stainless steel crowns for posterior teeth. However not all parents were familiar with the restorative options for anterior teeth and only had one of these interventions provided by the paediatric dentist without understanding the choice. This finding was significant because it indicated that the paediatric dentist's clinical

experience and preferences, intentionally or not, determined the choice between different restorative interventions. When parents were asked about their preferences, interestingly, they indicated the risk- benefit (being viewed as whether the restorative option will need anaesthesia or not and whether local or general anaesthesia could be used) to determine their decision which shows the detrimental influence of anaesthesia on parent's choices. Dental anxiety is variable between children and is influenced by internal and external factors as well as the child temperament and thus explains the variable ways children cope with dental visits and treatment (Klingberg and Broberg, 2007).

The dental appointments had an additional time implication on families, resulting from the parents' commitment to reward their children following the dental visits as well as the need to travel along distance for families living outside Dublin. Remarkably, parents did not mention any negative effect on schooling or employment.

An important outcome of this study was that focus group meetings gave parents participating in the study the opportunity to meet parents of children affected by same condition for the first time in their life. Parents were happy to share their emotions and experiences with other parents and felt belonging to a community, however, they espoused the need for informational and emotional support. Parents reported lack of electronic information relevant to their child and written in layman terms.

Parents suggested the development of a website or electronic support groups led by professionals as a source for both emotional and informational support especially for newly diagnosed parents when there is no family history. The foundation of a website can provide them with information in layman terms and give an overview of options for treatment prior to attending the paediatric dental clinic.

Social peer groups for both children and parents were also suggested to get emotional and social support for children and their parents. This need was previously identified in a qualitative study of adolescents affected by AI where their parents expressed a desire for support groups held by professionals (Sneller et al., 2014). Peer support provides parents a sense of normality, practical advice, experiential learning and allows them to

see into the future particularly when dealing with parents of older age. Such groups could yield a family support benefit. (Kerr and McIntosh, 2000).

4.2 Strengths, Limitations and Future Research

This study was a qualitative study that took a descriptive approach in order to collect detailed and rich data on parental attitudes regarding aesthetics and treatment need of young children affected by AI and DI. Focus groups were adopted to collect the research data involving a purposive sample of parents of young children. This was very useful for a variety of reasons, it allowed for in depth group discussion of unique, diverse and shared experiences and perceptions. Also, focus group interactions allowed for the emergence of new data, for example, the complex interaction between the previous experiences of affected parents' and their own perception of their children's teeth.

Both mothers and fathers hold the responsibility for their children's emotional and physical care thus facilitating the child's access to dental treatment. Parents' views can be transmitted to their children (Rumsey and Harcourt, 2007). This study was the first study to include parents of young children affected with AI and DI and it explored father's views alongside mothers' views. It was apparent that both mothers and fathers shared similar concerns in relation to affected children. However, one of the novel findings of this study (the feeling of guilt) emerged from the fathers' focus group thus the inclusion of fathers was essential. Therefore, this study was valuable as it increased the paediatric dentists' knowledge and understanding of fathers' as well as mothers' perceptions. However, perception of children might disagree with their parents' views and so future research exploring the young children's attitude to the aesthetics of their affected teeth would add to the completeness of the picture. Such research involving children is subjected to multiple ethical and methodological issues inherent to the children's limited and variable linguistic and cognitive ability to communicate their thoughts and perceptions and the complexities related to getting an informed consent as well as achieving complete confidentiality (Barbosa and Gavião, 2008a; Kirk, 2007).

The separation of participants by gender allowed for including couples in different groups which may perhaps be difficult to achieve in mixed gender groups. This separation by gender provided comfortable group dynamics for both mothers and fathers and shed light on subtle differences in their perceptions and worries. Yet, the moderator skills might have contributed to that and perhaps the same comfortable group dynamics might still be achieved with mixed gender groups.

No agreement exists about sample size in qualitative studies. For the purposes of this study, the two focus groups with a total of 13 participants provided a detailed, coherent and rich description that resulted in data saturation. As with all qualitative research, the sample sizes and selection procedures used do not permit statistical estimates to be made with any calculable degree of accuracy. This research has provided a unique insight into parent's perspectives and its findings can inform future longitudinal quantitative research to ascertain the views of more parents of children with AI and DI, both before and after dental interventions.

An acknowledged limitation of this study was the potential for recall bias on behalf of parents as some children had comprehensive dental treatment under G.A. before approaching four years of age, however all children were under ongoing dental review and/or treatment. Parents were also identified from existing patients with dental anomalies that were referred to the DDUH for dental assessment and treatment and so might have additional concerns that not necessarily reflect those with parents who did not seek referral for their AI and DI affected children. Parents did not need to pay for treatment and their willingness to pay was based on hypothetical assumption of a need to pay and so might not fully reflect their real attitudes. Another potential limitation is that all participants were Irish, all were married, and none were qualified for a Medical Card, therefore these results must be taken with caution as parents' perspectives might vary for other ethnic groups, different socioeconomic classes and/or single parents. It would be interesting to utilise the same research methodology to elicit parents' views in different cultures and different dental settings where parents needed to pay for treatment.

Rigour and trustworthiness in this study were established by adherence to quality guidelines for the qualitative research conduct (O'Brien et al., 2014) (see **Appendix 10**)

at all stages of research conduct and writing. The researcher received training in qualitative research conduct, an independent experienced qualitative researcher moderated the focus groups and another experienced qualitative researcher and the supervisor reviewed the data independently before the main themes were agreed to limit the researcher bias. However, it must be noted that all members of the research team were dentists which might impose clinician perspectives on the emergent data.

5 Conclusions

This qualitative study provided valuable insights on mothers and fathers attitudes regarding aesthetics and treatment needs of young children affected by AI/DI. The findings of this study suggested that children can notice their altered dental appearance before approaching three years of age. Most parents sought dental treatment before approaching school age. Dental interventions of primary teeth before approaching school age was perceived by parents as a way of achieving normality, protecting children from feeling different and/ or experiencing negative social reactions at school and a way of keeping dental structure and function. Seeking early dental intervention was one way of coping with the feeling of guilt of fathers affected by the same condition. In contrast, mothers' feeling of guilt was more related to dental treatment under general anaesthesia. Dental treatment of young children under general anaesthesia places a considerable emotional stress on parents, especially mothers.

The personal experience of parents affected by AI/DI in terms of childhood experience with bullying and own dental experience played a pivotal role in parent's judgements of their children's teeth and perceived need for dental treatment.

The paediatric dentist has a powerful influence on all parental treatment related decisions and must acknowledge parents' perspectives, parents' feelings, the impact of the condition on the child and the affected parents' experiences on an individual basis when discussing options for treatment. Furthermore, parents appeared to rely solely on the paediatric dentist as main source of information. This research finding points to the obligation of the paediatric dentist to ensure spending enough time with families in providing adequate information on both the short term and long term issues to enable parents to give truly informed consent for the dental care of their children and support both parents and children in coping with these dental anomalies.

6 References

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7 Appendices

7.1 Appendix 1: Ethical Approval



Coláiste na Tríonóide, Baile Átha Cliath
Trinity College Dublin

Ollscoil Átha Cliath | The University of Dublin

Dr. Areej Alqadi
Dublin Dental University Hospital,
Lincoln Place,
Dublin 2.

Ref: 160603

Title of Study: Investigation of Parental Perception of Children Affected by Amelogenesis Imperfecta (AI) Before and After Dental Intervention.

Dear Dr. Alqadi,

Further to a meeting of the Faculty of Health Sciences Ethics Committee held in June 2016, we are pleased to inform you that the above project has been approved without further audit.

Yours sincerely,

A handwritten signature in black ink that reads "Prof. Brian O'Connell".

Prof. Brian O'Connell
Chairperson
Faculty Research Ethics Committee

Dámh na nEolaíochtaí Sláinte
Foirgneamh na Ceimice,
Coláiste na Tríonóide,
Ollscoil Átha Cliath,
Baile Átha Cliath 2, Éire.

Faculty of Health Sciences
Chemistry Building,
Trinity College Dublin,
The University of Dublin,
Dublin 2, Ireland.

www.healthsciences.tcd.ie

7.2 Appendix 2: Ethical Approval After Amendment



Coláiste na Tríonóide, Baile Átha Cliath
Trinity College Dublin

Ollscoil Átha Cliath | The University of Dublin

Dr. Areej Alqadi,
Dublin Dental University Hospital,
Lincoln Place,
Dublin 2

Ref: 160603

Title of Study: Investigation of Parental Perception of Children Affected By Amelogenesis Imperfecta (AI) Before and After Dental Intervention. And Dentinogenesis Imperfecta (DI) Before and After Dental Intervention.

Dear Dr. Alqadi,

Further to a meeting of the Faculty of Health Sciences Ethics Committee held in November 2016, we are pleased to inform you that the above project (as amended with the following changes) has been approved without further audit.

The number of participants following contact for the study is insufficient to proceed. We intend to expand our inclusion criteria (section 2-6) to include:

- 1. Parents of children with another genetic anomaly affecting dental aesthetics; Dentinogenesis Imperfecta (DI).*
- 2. Parents of older children (age 11-15)*

It is anticipated that expanding the population will provide adequate number of participants. Participants will be identified and contacted in the exact same manner as previously approved. (section 1-3)

- Appendix 1: title of invitation letter changed.*
- Appendix 2: change throughout leaflet to include Dentinogenesis Imperfecta.*
- Appendix 3: title changed in consent form.*

Yours sincerely,

Prof. Brian O'Connell
Chairperson
Faculty Research Ethics Committee

Dámh na nEolaíochtaí Sláinte
Foirgneamh na Ceimice,
Coláiste na Tríonóide,
Ollscoil Átha Cliath,
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Chemistry Building,
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7.3 Appendix 3: Invitation Letter

Invitation Letter

University of Dublin

Trinity College

NAME OF PARENT

RE: Investigation of Parental Perception of Children Affected by Amelogenesis Imperfecta (AI) and Dentinogenesis Imperfecta (DI)

Dear Parent,

I am writing with regards to a proposed research study which I am undertaking as part of my clinical doctorate degree in paediatric dentistry. You have been contacted because your child attends the Dublin Dental Hospital for treatment. This study is asking parents to discuss their experiences and opinions regarding the appearance of children's teeth and of dental treatment for children. In this way we hope to better understand parents' concerns about the appearance of children's teeth and the right time for treatment.

If you are willing to participate in this project it means you would agree to attend one group discussion to discuss the issues. Attached you will find a consent form, information sheet and participant information leaflet that give you more details about the study. Please return the Information Sheet if you are interested in taking part as soon as possible. We will then contact you to arrange a suitable time.

If you have any further questions or envisage any issues related to this study please feel free to contact me by email: areej.alqadi@dental.tcd.ie

Many Thanks,

Yours Sincerely

Dr Areej Alqadi

7.4 Appendix 4: Participant Information Leaflet

Patient Information Leaflet

Title of study: Investigation of Parental Perception of Children Affected by Amelogenesis Imperfecta (AI) and Dentinogenesis Imperfecta (DI)

Introduction: Children can be born with unusual teeth. Amelogenesis imperfecta (AI) and Dentonogenesis Imperfecta are a conditions which can affect both baby and adult teeth and have unusual appearance because the enamel is defective.

This study aims to increase our understanding of how parents perceive affected and unaffected teeth of their children or other children before and after any dental intervention.

Understanding your thoughts on your child's baby teeth and how these might affect your decisions are important.

This research will help you and the dentist decide on the best time and treatment option in dealing with affected teeth.

Dentists' and parents' opinions of optimum treatment can vary; we aim to understand parents' opinions and perceptions that would be crucial in communication and satisfaction of all parties.

Procedures: you are invited to join this study because:

1. You are a parent of a child who was referred to the Dublin Dental Hospital, and diagnosed with AI or DI affecting his/ her teeth.
OR
2. You are a parent of a child who presented to the Dublin Dental Hospital without any front teeth dental issue.

We would like to invite you to participate in a focus group discussion to express your feelings and attitudes related to a set of photos of children with a range of appearance and a range of ways of dealing with affected teeth.

- Your child will not need to attend.
- Participants will be divided into groups of 4-6 people. Discussion will be held in a quiet room in the Dublin Dental University Hospital, audiotape recorded, and led by two dentists of which one will be moderator and the other will be note taker.
- We would like to involve both mothers and fathers in separate sessions.
- These group discussions will be held in the DDUH in a mutually agreed time (either during the day, after work hours or during the weekend) and won't take more than two hours.

Benefits: By participation in this study you will help us understand your own perspectives in viewing children's teeth. This is expected to improve the communication between parents of affected children and the dentist.

Risks: this study will not put you or your child under any risk.

Exclusion from participation: *you cannot participate in this study if you:*

- Cannot speak English,
- Your children had a set of adult teeth when first presented to the DDUH.
- Your child has an altered front teeth appearance due to other conditions (e.g. caries, trauma, missing teeth).
- Are not willing to provide consent.

Confidentiality:

The results of the study will be used for publication in the dental literature and scientific conferences. Your identity will remain confidential. Your name will not be published and will not be disclosed to anyone outside the study group.

You can access the transcript of the focus group you attended if you wish.

Voluntary Participation: You have volunteered to participate in this study. You may quit at any time. If you decide not to participate, or if you quit, that will not affect your child's management in the Dental Hospital.

Stopping the study: You understand that the investigators may withdraw your participation in the study at any time without your consent.

Permission: This trial has Faculty of Health Science Research Ethics Committee approval.

Further information: If you are interested in participating in this study, or have any queries please contact: **Areej Alqadi** or **Anne Marie Boon** on this number **(01-6127303)** and mention "parents study" and whether the mother, father or both would like to participate.

Thank you for your consideration.

Consent Form

Title of research study: Investigation of Parental Perception of Children Affected by Amelogenesis Imperfecta (AI) And Dentinogenesis Imperfecta (DI)

RESEARCHER: Areej Alqadi, Postgraduate student in Paediatric Dentistry, Dublin Dental University Hospital.

SUPERVISOR: Anne O'Connell, Associate Professor and Consultant in Paediatric Dentistry, Dublin Dental University Hospital.

BACKGROUND:

Participants will attend a group discussion with other parents on one occasion to express their feelings and attitudes related to the appearance of children's teeth and of dental treatment for children.

Participants will be divided into groups of 4-6 people. Discussion will be held in a quiet room in the Dublin Dental University Hospital, audiotape recorded, and led by two dentists of which one will be moderator and the other will be note taker.

Mothers and fathers will be involved in separate sessions.

These group discussions will be held in the DDUH in a mutually agreed time (either during the day, after work hours or during the weekend) and won't take more than two hours.

You may request a copy of the transcript of the focus group you attended.

DECLARATION:

I have read, or had read to me, the information leaflet for this project and I understand the contents. I have had the opportunity to ask questions and all my questions have been answered to my satisfaction. I freely and voluntarily agree to be part of this research study, though without prejudice to my legal and ethical rights. I understand that I may withdraw from the study at any time and I have received a copy of this agreement.

PARTICIPANT'S NAME: (mother, father)

Your child's name:

PARTICIPANT'S SIGNATURE:

Date:

Date on which the participant was first furnished with this form:

Statement of investigator's responsibility: I have explained the nature, purpose, procedures, benefits, risks of, or alternatives to, this research study. I have offered to answer any questions and fully answered such questions. I believe that the participant understands my explanation and has freely given informed consent.

Doctor's/Dentist's signature:

Date:

Information Sheet

Dear Parent,

Thank you for considering the participation on this study.

We would like each individual who would like to participate kindly to:

- Fill this form.
- Send it back to

Ms. Anne Marie Boon,
Dublin Dental University Hospital,
Lincoln Place, Dublin 2, D2.

Participant's name:

Mothers:

- Please indicate 3 possible time slots that suit you the most for group discussion:

	Monday	Tuesday	Wednesday	Thursday	Friday	Saturday
morning						
afternoon						
evening						

- Do you have a medical card : YES NO
- Are you > 35 year old : YES NO
- < 35 year old: YES NO

Fathers: (discussions at a separate time to mothers)

- Please indicate 3 possible time slots that suit you the most for group discussion:
- Do you have a medical card : YES NO
- Are you > 35 year old : YES NO
- < 35 year old: YES NO

	Monday	Tuesday	Wednesday	Thursday	Friday	Saturday
Morning						
Afternoon						
Evening						

7.7 Appendix 7: First focus Group Guide

Focus Group Guide 1

Site:

Number of participants:

Moderator:

Note taker:

Date:

Start time:

End time:

Seating chart:

Introduction:

- Study topic
- Explanation of study aims and objectives
- Explanation of length of discussion.
- Mobiles turned off/silent.
- Ask participants not to discuss the content of the discussion afterwards.
- Respect each other.
- Speak once at a time.
- Moderator/participant should state the participant number before speaking.
- Check if they have any questions.
- Check that they are happy to continue.

Background:

- name
- Age.
- Number of children.
- Child's age on first dental visit.
- Reason for visiting dentist.

Photos discussion:

Aesthetics and attractiveness

- How do you describe ideal baby teeth? How do you describe normal teeth?
- Colour, shape, size
- Who decides what perfect teeth are?
- Photos to be utilised. How would you describe them? Describe your child's teeth?
- Is there anything you like about these teeth? Is there anything you don't like?

Impact of having this problem on the child's life (What is it like for your child when they have teeth like this?)

- Feeling different, self-consciousness
- Friendship and social interactions.
- School.

- Confidence.

Impact of having this problem on the parent's life. (How does that make you feel?)

- Society perception (how does society judge little children with these teeth?)
- Whether it gets them down.

Treatment need

- Timing. At what age do you think it starts to matter?
- Who decided to have treatment?
- Motivational factor. (Function/ appearance, both?)
- Gender effect. (boys versus girls)
- Was there anything that was holding you back for seeking dental treatment? Barriers.
- Worth to pay for it if they had to pay? How much are you prepared to pay for dental treatment?
- Whether parent's assessment of their children's anxiety influences their preferences for the dental care of their children?
- Anything parents can do? e.g.: whitening toothpaste.

Influence of different interventions (photos to be utilised)

- Satisfaction and agreement. (Looking back would you have done anything differently?)
- Aesthetics and attractiveness.
- Impact on child and parents.
- Looking back to timing, any regrets?
- Worth G.A.? Usually 2 stages when 6 and when 8.

Conclusion:

- Thank participants for their time.
- Reassure participants that data will remain confidential.
- Reassure participants that they will be fully anonymised and the ideas discussed will be used for research purposes.

Aesthetics and Attractiveness:

Participant number	Response	Observations
1		
2		
3		
4		
5		
6		

Impact on child's life:

Participant number	Response	Observations
1		
2		
3		
4		
5		
6		

Impact on parents:

Participant number	Response	Observations
1		
2		
3		
4		
5		
6		

Treatment need:

Participant number	Response	Observations
1		
2		
3		
4		
5		
6		

Influence of different interventions:

Participant number	Response	Observations
1		
2		
3		
4		
5		
6		

7.8 Appendix 8: Second Focus Group Guide

Focus Group Guide 2

Site:

Number of participants:

Moderator:

Note taker:

Date:

Start time:

End time:

Seating chart:

Introduction:

- Study topic
- Explanation of study aims and objectives
- Explanation of length of discussion.
- Mobiles turned off/silent.
- Ask participants not to discuss the content of the discussion afterwards.
- Respect each other.
- Speak once at a time.
- Moderator/participant should state the participant name before speaking.
- Check if they have any questions.
- Check that they are happy to continue.

Background:

- name
- age.
- Number of children.
- Child's age on first dental visit.
- Reason for visiting dentist.

Photos discussion:

Aesthetics and attractiveness

- How do you describe ideal baby teeth? How do you describe normal teeth?
- Colour, shape, size
- Who decides what perfect teeth are?
- Photos to be utilised. How would you describe them? Describe your child's teeth?
- Is there anything you like about these teeth? Is there anything you don't like?

Impact of having this problem on the child's life. (What is it like for your child when they have teeth like this?)

- Feeling different, self-consciousness
- Friendship and social interactions.

Red text denotes additions following modifications after focus group 1.

- School.
- Confidence.

Impact of having this problem on the parent's life. (How does that make you feel?)

- Society perception. (How does society judge little children when their teeth are affected?)
- Whether it gets them down.
- **Any feel of guilt?**

Treatment need

- **Timing. (Optimal age to start treatment?)**
- **Experience of diagnosis and referral?**
- Motivational factor. (Function/ appearance, both?)
- **Effect of AI/DI on general health and diet?**
- **Sources of information?**
- Who made the decision for treatment?
- **Effect of child personality on seeking dental treatment?**
- Gender effect. (boys versus girls)
- Was there anything that was holding you back for seeking dental treatment? Barriers
- Worth to pay for it if they had to pay? How much are you prepared to pay for dental treatment?
- Whether parent's assessment of their children's anxiety influences their preferences for the dental care of their children?
- Anything parents can do? e.g.: whitening toothpaste.
- **Coping mechanisms?**

Influence of different interventions

- Satisfaction and agreement. (Looking back, would you have done anything differently?)
- Aesthetics and attractiveness.
- Impact on child and parents.
- **Experience with transition into mixed dentition?**
- Looking back to timing, any regrets?
- Worth G.A.? Usually 2 stages when 6 and when 8.

Conclusion:

- Thank participants for their time.
- Reassure participants that data will remain confidential.
- Reassure participants that they will be fully anonymised and the ideas discussed will be used for research purposes.

Aesthetics and Attractiveness:

Participant number	Response	Observations
1		
2		
3		
4		
5		
6		

Impact on child's life:

Participant number	Response	Observations
1		
2		
3		
4		
5		
6		

Impact on parents:

Participant number	Response	Observations
1		
2		
3		
4		
5		
6		

Treatment need:

Participant number	Response	Observations
1		
2		
3		
4		
5		
6		

Influence of different interventions:

Participant number	Response	Observations
1		
2		
3		
4		
5		
6		

7.9 Appendix 9: Photos













7.10 Appendix 10: SRQR Checklist

Standards for Reporting Qualitative Research (SRQR)* http://www.equator-network.org/reporting-guidelines/srqr/	
Title and abstract	Section number/ Comment:
Title- Concise description of the nature and topic of the study Identifying the study as qualitative or indicating the approach (e.g., ethnography, grounded theory) or data collection methods (e.g., interview, focus group) is recommended	"Parental Perception of Children Affected by Amelogenesis Imperfecta (AI) and Dentinogenesis Imperfecta (DI); A Qualitative Study." The title describes the topic concisely and indicates clearly that it is a qualitative study.
Abstract - Summary of key elements of the study using the abstract format of the intended publication; typically includes background, purpose, methods, results, and conclusions	Summary including aims, methods, results and conclusion is available page 2.
Introduction	
Problem formulation - Description and significance of the problem/phenomenon studied; review of relevant theory and empirical work; problem statement	Section 1 includes a background of the topic and discuss the lack of existing literature on the research topic. (pg. 8-25)
Purpose or research question - Purpose of the study and specific objectives or questions	Section 1.3 states the aims and objectives of the study.(pg. 26)
Methods	
Qualitative approach and research paradigm - Qualitative approach (e.g., ethnography, grounded theory, case study, phenomenology, narrative research) and guiding theory if appropriate; identifying the research paradigm (e.g., postpositivist, constructivist/ interpretivist) is also recommended; rationale**	Section 2.1 states that it is an exploratory study and discuss the rationale for this study. (pg.27) Section 2.14.2 describe the thematic analysis approach followed in this study. (pg. 36-39)
Researcher characteristics and reflexivity - Researchers' characteristics that may influence the research, including personal attributes, qualifications/experience, relationship with participants, assumptions, and/or presuppositions; potential or actual interaction between researchers' characteristics and the research questions, approach, methods, results, and/or transferability Context - Setting/site and salient contextual factors; rationale**	Section 2.10 illustrates the characteristics of the researcher and focus group moderator and considerations taken to limit bias and add to the research reflexivity. (pg.33) Section 2.3 elucidates the research setting.(pg.28)

<p>Sampling strategy - How and why research participants, documents, or events were selected; criteria for deciding when no further sampling was necessary (e.g., sampling saturation); rationale**</p>	<p>Section 2.3 refers to the purposive sampling strategy utilised, provides a clear definition and explanation of this sampling strategy and points out the participant’s characteristics that accounted for the criteria of selection. Section 2.3: indicates the theoretical saturation approach applied to determine the total number of focus groups. (pg.28) Section 2.4 states the inclusion and exclusion criteria. (pg. 29) Section 2.6 details the identification and recruitment procedure. (pg.30)</p>
<p>Ethical issues pertaining to human subjects - Documentation of approval by an appropriate ethics review board and participant consent, or explanation for lack thereof; other confidentiality and data security issues</p>	<p>Section 2.2 documents the ethical approval obtained for this study. (pg.28) Appendixes 1 and 2 include the ethical approval letters.</p>
<p>Data collection methods - Types of data collected; details of data collection procedures including (as appropriate) start and stop dates of data collection and analysis, iterative process, triangulation of sources/methods, and modification of procedures in response to evolving study findings; rationale**</p>	<p>Section 2.1 discuss the justification for using focus groups and not individual interviews. (pg.27) Sections 2. 7- 2.13 describes the data collection procedure in details. (pg.31-34) Section 3.1 states the dates of data collection. (pg.40) Section 2.14 describes the data analysis and clarifies that data analysis was a continuous procedure started with data collection and ended with the final report. (pg. 35-39)</p>
<p>Data collection instruments and technologies - Description of instruments (e.g., interview guides, questionnaires) and devices (e.g., audio recorders) used for data collection; if/how the instrument(s) changed over the course of the study</p>	<p>Sections 2.7-2.13 describes the data collection procedure in details. (pg. 31-34) Appendix 7 outlines the focus group guide.</p>

<p>Units of study - Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)</p>	<p>Sections 3.2.1 and 3.2.2 details the numbers and demographic characteristics of participants. (pg. 40-42)</p>
<p>Data processing - Methods for processing data prior to and during analysis, including transcription, data entry, data management and security, verification of data integrity, data coding, and anonymization/de-identification of excerpts</p>	<p>Section 2.13 describes data transcription. (pg. 34) Section 2.14 provides a detailed data management and analysis. (pg. 35-39)</p>
<p>Data analysis - Process by which inferences, themes, etc., were identified and developed, including the researchers involved in data analysis; usually references a specific paradigm or approach; rationale**</p>	<p>Section 2.14 and its subsections details the data analysis approach with reference to the thematic analysis approach applied. (pg. 35-39)</p>
<p>Techniques to enhance trustworthiness - Techniques to enhance trustworthiness and credibility of data analysis (e.g., member checking, audit trail, triangulation); rationale**</p>	<p>Section 2.15 refer to member checking- detailed on section 1.4- and refers to adherence to these quality guidelines on writing up qualitative data. (pg. 39) Section 2.5 outlines the researcher training in qualitative research conduct. (pg. 30)</p>

Results/findings

<p>Synthesis and interpretation - Main findings (e.g., interpretations, inferences, and themes); might include development of a theory or model, or integration with prior research or theory</p>	<p>Parents of children affected by AI and DI believe that “ baby teeth are important” and value dental treatment of their children primary teeth before starting school. The personal experience of parents affected by AI/DI plays a pivotal role in parent’s judgements of their children’s teeth and perceived need for dental treatment. The paediatric dentist has a powerful influence on parental decisions and must acknowledge the parents’ perspectives when discussing options for treatment. Fathers and mothers agree on most aspects but the feeling of guilt was more prominent among fathers while worry related to dental treatment under general anaesthesia was more pronounced among mothers.</p>
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Links to empirical data - Evidence (e.g., quotes, field notes, text excerpts, photographs) to substantiate analytic findings	Evidence from parents' quotes included to support analytic findings.
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Discussion

Integration with prior work, implications, transferability, and contribution(s) to the field - Short summary of main findings; explanation of how findings and conclusions connect to, support, elaborate on, or challenge conclusions of earlier scholarship; discussion of scope of application/generalisability; identification of unique contribution(s) to scholarship in a discipline or field	All were ensured while writing the discussion. The generalisability of the findings was discussed under section 4.2. (pg. 101-103)
Limitations - Trustworthiness and limitations of findings	Discussed in section 4.2. (pg.101-103)

Other

Conflicts of interest - Potential sources of influence or perceived influence on study conduct and conclusions; how these were managed	No potential sources of conflict.
Funding - Sources of funding and other support; role of funders in data collection, interpretation, and reporting	No influence of DDUH funding the study on the study process.

*The authors created the SRQR by searching the literature to identify guidelines, reporting standards, and critical appraisal criteria for qualitative research; reviewing the reference lists of retrieved sources; and contacting experts to gain feedback. The SRQR aims to improve the transparency of all aspects of qualitative research by providing clear standards for reporting qualitative research.

**The rationale should briefly discuss the justification for choosing that theory, approach, method, or technique rather than other options available, the assumptions and limitations implicit in those choices, and how those choices influence study conclusions and transferability. As appropriate, the rationale for several items might be discussed together.

O'Brien BC, Harris IB, Beckman TJ, Reed DA, Cook DA. [Standards for reporting qualitative research: a synthesis of recommendations. *Academic Medicine*, Vol. 89, No. 9 / Sept 2014 DOI: 10.1097/ACM.0000000000000388](#)