Patient and Parental Perception of Alveolar Bone Grafting Surgery in Children with Unilateral Cleft Lip and Palate: A Qualitative Study

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Declaration

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Summary

Aims: This study aimed to evaluate and understand patients' and parents' experience of alveolar bone grafting surgery and to gather patient and parental feedback on existing information resources related to alveolar bone grafting surgery.

Methods: A descriptive, qualitative study was conducted involving a purposive sample of children affected by unilateral cleft lip and palate and parents of affected children, who had undergone alveolar bone grafting surgery. Two topic guides were developed and in-depth, semi-structured interviews were carried out over the telephone. The interviews were audio-recorded with a dictaphone and transcribed verbatim. A thematic analysis approach was carried out for data analysis using MAXQDA software.

Results and Discussion: Following data analysis, six main themes were identified: the life course of the condition, the perception of treatment, the experience of treatment, communication and information seeking, the impact of treatment and coping mechanisms. There was a large amount of positive feedback and high satisfaction with the care that patients had received and their experience of treatment. The importance of the patient-clinician relationship was evident throughout. Parents relied heavily on the information they received verbally at the multidisciplinary clinics and tended to avoid doing their own research online, for

fear of what they may read having unreliable content. Negative feedback related to the busy nature of the clinics and the long waits to be seen, as well as some children feeling overwhelmed by the number of clinicians in the room at the multidisciplinary clinics. However, parents did acknowledge, and were appreciative of, the considerable expertise in the room at these clinics. For many parents and children, the significant discomfort and mobility issues from the donor site post-surgery was unanticipated, and a consequence for which they felt unprepared. The impact of cleft on parents was most significant psychologically at diagnosis and during the initial surgeries, and their ability to cope and rationalize their fears for their child seemed to improve drastically over time. ABG and pre-surgical orthodontics caused considerable stress and anxiety in patients. Familiarity with, and trust in the cleft care team were significant in reducing their anxiety. Acknowledging other strengths of the child, putting the condition in perspective and support of family and the cleft community, emerged as means of coping with the condition.

Conclusions: Parents were better able to cope with their child's treatments and surgeries at the alveolar bone grafting stage of treatment but children find the MDT clinic daunting and were anxious prior to their surgery. Trust in and familiarity with the clinicians and care team were important factors in reducing their anxiety prior to surgery. Parents felt unprepared for the discomfort and mobility issues of their child's hip following surgery and expressed a desire to be better prepared for this element of the surgical experience.

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List of Acronyms

CLP	Cleft Lip and Palate
ABG	Alveolar Bone Graft
MDT	Multi-Disciplinary Team
OLCHC	Our Lady's Children's Hospital Crumlin
TSCUH	Temple Street Children's University Hospital
SRA	Social Research Authority
SRQR	Standards of Reporting Qualitative Research
CLAPAI	Cleft Lip and Palate Association of Ireland
DCC	Dublin Cleft Centre

1.0 Literature Review

1.1 Orofacial Clefts

A cleft is an abnormal opening or fissure in an anatomical structure that is normally closed. Orofacial clefts can affect the lip and/or palate and may occur in isolation or in combination with a syndrome (Kummer, 2014).

1.2 Aetiology of Clefts

Clefts of the lip and palate (CLP) are congenital conditions and are the most common congenital anomaly to affect the craniofacial region in humans (Fraser, 1970). Clefts occur due to a disruption in embryological development, affecting the normal fusion of the palatal shelves and lip. Embryological development of the lip and alveolus begins at 6 to 7 weeks, while palatal development begins later at approximately 8-9 weeks of gestation (Kummer, 2014). Prior to palate formation the tongue sits in a high position with the two palatal shelves vertically on either side. The cleft can result from failure of either growth, elevation or fusion of the palatal shelves.

Ultimately the aetiology of clefts is poorly understood but believed to be multifactorial, with a combination of genetic and environmental factors implicated in its development. A number of different causative genes and teratogens have been described (**Table 1**). The sibling risk for cleft lip and palate is approximately 30 times higher than that of the normal population. The concordance rate in monozygotic twins is approximately 25-45% and 3-6% for dizygotic twins

(Mitchell and Risch, 1992). This lack of complete concordance highlights the significance of environmental factors in the aetiology.

Table 1: Factors Contributing to Cleft Development

Genes (Dixon et al., 2011)	Teratogens	Other
IRF6	Cigarette smoke (Reiter et al., 2012)	Increased parental age (Martelli et al., 2010)
PVRL1	Phenytoin (Edwards et al., 2003)	Maternal obesity (Moore et al., 2000)
MSX1	Diazepam (Edwards et al., 2003)	Maternal vitamin B6 deficiency (Munger et al., 2004)
MID1	Corticosteroids (Edwards et al., 2003)	Mechanical interferences in utero (Kummer, 2014)
TREACLE	Lead pollution (Vinceti et al., 2001)	Pre-pregnancy diabetes mellitus (Watkins et al., 2014)
COL2AI	Rubella (Metneki et al., 2005)	Maternal malnutrition
IRF6	Influenza (Metneki et al., 2005)	
VAX1	Alcohol (Watkins et al., 2014)	
8q24	Retinoic acid (Watkins et al., 2014)	
FGFR2		
FOXE1		
17q22		

1.3 Syndromes associated with Clefts

A syndrome is a pattern of multiple anomalies that are pathogenically related. A sequence, however, is a series of anomalies that result from a single initiating factor, for example Pierre Robin Sequence is the most well-known example of a sequence that is associated with orofacial clefting. The initiating

factor in Pierre Robin Sequence is the micrognathia and the secondary consequence is glossoptosis (posterior position of the tongue), which often causes a wide cleft of the palate, as the tongue prevents fusion of the palatal shelves. Pierre Robin Sequence may occur in isolation or in combination with a syndrome, such as Treacher Collins Syndrome, Stickler Syndrome or Foetal Alcohol Syndrome (Kummer, 2014).

Thirty per cent of cases of cleft lip and palate are associated with a syndrome, and half of all cases of cleft palate occur in association with a syndrome (Cobourne, 2016). It has been estimated that there are over 400 different syndromes associated with facial clefts (Gorlin, 2001). Below are some of the syndromes most commonly associated with cleft lip and/or palate and cleft palate alone (**Table 2**) (Kummer, 2014).

Table 2: Syndromes associated with CLP

Syndromes associated with CL+/-P	Syndromes associated with CP
Amniotic bands	CHARGE syndrome
CHARGE syndrome	Foetal alcohol syndrome
Diabetic embryopathy	Foetal hydantoin syndrome
Foetal alcohol syndrome	Hemifacial microsomia
Hemifacial microsomia	Kabuki syndrome
Opitz syndrome	Stickler syndrome
Orofaciodigital syndrome type 1	Van der Woude syndrome
Popliteal pterygium syndrome	Velocardiofacial syndrome
Trisomy 13	
Van der Woude syndrome	
Wolf-Hirschhorn syndrome	

1.4 Classification of Clefts

Various classifications of orofacial clefts have been described, including systems by Veau and Borel (1931), Kernahan and Stark (1958), Kernahan (1971) and Kriens (1989) (McBride et al., 2016). There was debate about the various classifications, none of which were universally accepted. The clinical presentation of clefts is so variable that it is often more useful to specifically describe each individual case. One particular controversy is the need to differentiate between cleft of the lip and palate and cleft lip alone. Historically, cleft lip alone and cleft lip with cleft palate have been considered as a single phenotype of varying severity (Marazita, 2012). Orofacial clefting can range from simple notching or submucous clefts to complete unilateral or bilateral clefts of the lip, alveolus and hard and soft palate.

Kriens' palindrome for classifying clefts (LAHSHAL) is probably the most applicable and useful (**Figure 1**) (Kriens, 1989). The letters represent Lip, Alveolus, Hard Palate and Soft Palate. An advantage of this classification is that the laterality of each anatomical area (except the soft palate) can be recorded. The completeness of the cleft may also be recorded: incomplete clefts are represented with lowercase letters, while complete clefts are represented with uppercase letters. The Royal College of Surgeons later simplified the classification by removing the second H in the palindrome.

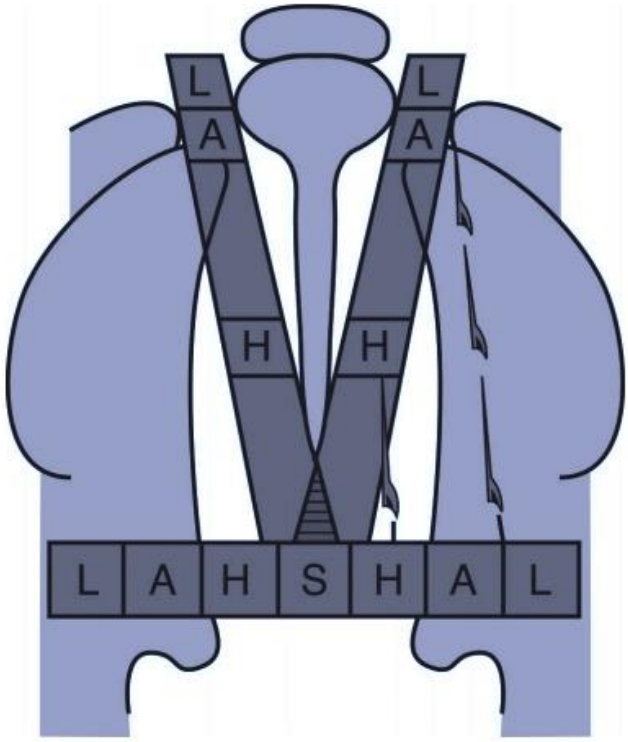

Figure 1: LAHSAL Classification

1.5 Epidemiology of Clefts

The incidence of cleft lip with or without cleft palate was reported to be 1 per 700 live births (Mossey, 2001). A more recent update on the epidemiology of orofacial clefts reported the worldwide incidence to be slightly lower at 7.94 per 10,000 live births (Tanaka et al., 2012). Considerable racial variation was found in the incidence however, with Asian populations most commonly affected (1.7 per 1000), followed by Caucasians (0.6 - 1.7 per 1000) and least common in African populations (0.4 per 1000) (Habib, 1978).

In another population based study of approximately 8 million births, the incidence of cleft lip and/or palate was 9.9 per 10,000 births. This data was

collected from 54 birth defect registries in 30 countries between 2000 and 2005. The incidence in Western Europe (12.1 per 10,000) was similar to that of the United States. However in Japan, the incidence nearly doubled (20 per 10,000) (IPDTOC, 2011). Cleft palate alone occurs less frequently, approximately 1 in 2,000 births. Incidence varies according to race, from 1.2 cases per 10,000 births in areas of sub-Saharan Africa to six cases per 10,000 births in Europe and Central Asia (Watkins et al., 2014).

Seventy per cent of cases of cleft lip and/or palate occur in isolation and the remaining 30% are associated with a syndrome, whereas half of isolated cleft palate cases occurs in association with a syndrome (Cobourne, 2016). Unilateral cleft lip and/or palate account for the vast majority of all orofacial clefts (approximately 80%), with the left side more commonly affected (Cobourne, 2016).

1.6 Gender Predilection

Males are more commonly affected by cleft lip and/or palate (approximately 3:2), whereas females are more commonly affected by isolated cleft palate (approximately 3:2) (Wyszynski et al., 1996).

1.7 Features of Cleft

1.7.1 Feeding

Feeding is a significant concern for parents of babies born with clefts. Young found that for 95% of parents, this was their primary concern (Young et al., 2001). The baby can struggle to suckle, as it is difficult to generate and maintain

sufficient intraoral pressure. Milk can also be lost through the nose before being swallowed, which can ultimately result in the baby struggling to get the necessary nutrients, gain weight and thus fail to thrive. This can be overcome with the use of specialised bottles with adapted teats, which the parents are instructed how to use by a feeding specialist within the first 24 hours of life.

1.7.2 Speech

Difficulty with speech in patients with cleft palate is caused by velopharyngeal insufficiency (inadequate functioning of the soft palate). The main problem is the lack of mobility in the soft palate, due to anomalous muscle insertions relating to the cleft as well as scar formation from the palate repair. The soft palate is unable to lift and produce an effective seal with the posterior pharyngeal wall, which results in hypernasality of the speech and nasal escape, particularly during pronunciation of certain consonants. The speech and language therapist has a role of critical importance for these patients. Spriestersbach et al. reported that 50% of children with repaired cleft palate developed normal speech spontaneously while 25% required speech and language therapy and 25% required further palatal surgery (Spriestersbach et al., 1973).

1.7.3 Hearing

Cleft palate is commonly associated with middle ear dysfunction. The Eustachian tube connects the middle ear to the nasopharynx. It is closed at rest and opens during swallowing and yawning when the tensor veli palatini muscle contracts. As the Eustachian tube opens, it provides ventilation for the middle ear,

which is important for normal function. Opening of the Eustachian tube also results in pressure equalization and allows fluid to drain. Dysfunction is often caused by failure of ventilatory function of the Eustachian tube. When the Eustachian tube fails to function normally, fluid collects in the middle ear due to negative pressure (middle ear effusion). If the fluid does not drain, bacteria can grow in the effusion and cause an infection, known as otitis media with effusion. This can be managed by the insertion of tympanostomy tubes by an ear-nose-throat surgeon under general anaesthesia. This procedure may need to be repeated.

1.7.4 Facial Features

Maxillary retrusion or midface deficiency is commonly found in patients with a repaired cleft lip and palate. This is due, in part, to an inherent deficiency in the maxilla (maxillary hypoplasia) and also due to contraction of scar tissue following surgery and a consequent restriction of maxillary growth (Kawakami et al., 2002). This can result in an anterior crossbite and a class III malocclusion, whereby the maxilla is retrusive relative to the mandible. The maxilla is also commonly deficient in the transverse and vertical dimensions. Other common facial features are a short upper lip, asymmetry of the lip, flattening of the Cupid's bow and a deviated nasal septum. The deviation of the nasal septum is caused by a lack of support from the floor of the nose and stenosis of one or both nares following repair.

1.7.5 Oral and Dental Features

Children with cleft lip and palate have an increased incidence of dental anomalies. Hypodontia of the lateral incisor on the side of the cleft is the most common dental anomaly, affecting approximately 50% of children with a cleft lip and palate (Akram et al., 2015). Often, there is hypodontia of both the lateral incisor and canine on the side of the cleft. If these teeth are present however, they tend to be irregular in size or shape. Hypodontia of other teeth outside the zone of the cleft is also common, as the MSX-1 gene appears to be responsible for the development of both clefting and hypodontia (van den Boogaard et al., 2000). Olin (1964) reported that 24% had hypodontia outside the area of the cleft (Olin, 1964). Supernumerary teeth in the region of the cleft is the second most commonly reported dental anomaly, occurring in 22.2% of children with a unilateral cleft lip and palate (Akram et al., 2015).

Delayed exfoliation of primary teeth, rotated teeth and ectopic eruption are all more common in patients with cleft lip and palate. Ectopic eruption of the maxillary first permanent molar was reported in 15.4% of children with clefts (Bjerklin et al., 1993) and a significantly higher rate of impaction of maxillary canines on the side of the cleft was also demonstrated (Akcam et al., 2008). Higher incidences of enamel hypoplasia, discoloration and dental caries were also reported in children affected by orofacial clefts.

The report of an increased prevalence of dental caries in patients with cleft lip and palate is likely multifactorial, associated with both diet and oral hygiene. Poorer oral hygiene may be due to the malalignment of the maxillary dentition, reluctance on the part of both the parent and the child to brush around the site of the cleft, as well as reduced access to the oral cavity following surgical repair of the

lip due to scarring. There may also be longer oral clearance times following eating. The diet may contain more simple carbohydrates and more frequent snacking may occur, in an attempt to encourage weight gain. However, there is evidence that both supports and refutes an increased caries experience in children with clefts. A systematic review by Hasslof and Twetman (2007) reported no good quality evidence to support an increased prevalence of dental caries in children with clefts (Hasslof and Twetman, 2007). This review had strict inclusion criteria however, which limited the number of studies included. A more recent systematic review and meta-analysis, which included twenty-four studies, concluded that individuals with cleft lip and/or palate have higher caries prevalence, both in the deciduous and the permanent dentitions (Worth et al., 2017). The authors did acknowledge that the quality of the included papers in this review were poor.

1.8 Psychosocial Impact of Cleft

1.8.1 Impact on parents

Parents of babies born with cleft can often experience a multitude of emotions initially, from shock and sadness, to disappointment, resentment and guilt, which are in conflict with other strong feelings of love and protectiveness (Despars et al., 2011). Parents of children with clefts can experience considerable stress, as they are faced with a number of additional challenges, from difficulty feeding their baby to obtaining adequate information about necessary surgeries and interventions. They also have to cope with the staring of other children and adults in public settings, which can be distressing as it compounds and highlights the fears of social rejection.

It has been reported that parents' stress levels correlate with the child's adjustment in later life. Pope *et al* (2005) examined the parents of 47 children with clefts and found higher levels of maladjustment in toddlers whose parents showed increased levels of stress at both assessments in the study period (Pope et al., 2005). This highlights the importance of providing adequate counselling and support for the parents of children with clefts, to prevent adjustment problems for the child in later life.

Family support has been shown to be a significant factor in the adjustment of having a child with a cleft. A lower level of social support is linked with depression in mothers of children with clefts (Baker et al., 2009). While having a child with a cleft is stressful, there is no evidence that it causes an increased incidence of psychiatric problems in parents (Grollemund et al., 2010). Nelson *et al* (2012a) interviewed parents of children with clefts in Manchester, United Kingdom and found that some of the positive aspects of caring for a child with a cleft was recognising personal strength, appreciating diversity and reflecting on a child's uniqueness (Nelson et al., 2012a). However, she also found that parents felt stress and guilt putting their child through invasive surgical procedures, but did so as they deemed it necessary to avoid social stigmatisation of the child in later life. As such, the importance of access to emotional support for parents is again apparent (Nelson et al., 2012b).

1.8.2 Impact on patients

For children born with clefts, there is evidence that impaired facial growth and dental anomalies are associated with adverse psychosocial outcomes (Hunt et

al., 2006). Much of this qualitative research has been carried out in North America. Children with clefts were shown to suffer from increased behavioral and emotional difficulties as well as depression and anxiety (Thompson and Kent, 2001). Chapados (2000) carried out interviews with teenagers affected by cleft and found that girls struggled with their appearance more than boys and that children often felt responsible for conflict between their parents (Chapados, 2000). Brunner *et al* (2004) found that the majority of young people with clefts had experienced bullying and had concerns about social acceptance (Brunner et al., 2004).

Alansari *et al* (2014) carried out interviews with adults who had been treated for cleft lip and palate (Alansari et al., 2014). They reported that the multiple medical visits and interventions were tiring and frustrating. Peer stigmatization was a major issue during childhood and contributed to feeling defective, unworthy and abnormal. They felt at times that the treatment process, which was supposed to make them feel more normal, paradoxically exacerbated their sense of defectiveness.

Hunt *et al* (2006) found significantly more depressive symptoms in patients with clefts on the Child Depression Inventory when compared with a control group (Hunt et al., 2006). However, Klassen *et al* (2012) carried out a systematic review of the literature on quality of life in children with clefts and found that in most of the studies, cleft patients had either equivalent or increased levels of self-concept and self-esteem when compared with non-cleft patients (Klassen et al., 2012). She also found most studies regarding peer relations showed no difference between cleft and non-cleft patients.

1.9 Management of Cleft

1.9.1 Introduction

As highlighted above, the presentation of orofacial clefts is complex and varied and often have lifelong implications for those affected. The objectives of treatment are to provide satisfactory facial appearance, good orofacial function for speaking, eating and swallowing, a stable and functional occlusion and satisfactory hearing. A significant range of healthcare professionals are involved in achieving these treatment objectives. While much of the interventions are surgically driven, a core rationale for these interventions is to achieve psychological and social well-being for the affected individual (Broder et al., 2017).

1.9.2 Multidisciplinary team approach

A multidisciplinary team approach is of paramount importance to achieve a successful outcome for patients with clefts. A number of healthcare professionals are involved in their care from birth to adulthood (**Table 3**).

Table 3: Members of the Cleft Team

Members of the Cleft team
Feeding Nurse
Cleft Co-ordinator
Speech and Language Therapist
Paediatric Dentist
Paediatrician
Orthodontist
Plastic Surgeon
Ear Nose and Throat Surgeon
Oral and Maxillofacial Surgeon
Prosthodontist
Geneticist
Audiologist

Patient outcomes are highest when their care is undertaken in a centralised unit by a multidisciplinary team who treat high volumes of cleft patients. Cleft care was centralised in the 1990s in the United Kingdom based on recommendations from a Clinical Standards Advisory Group (CSAG), in order to improve the clinical outcomes in cleft care (Sandy et al., 1998, CSAG, 1998). Cleft care in Ireland was also centralised on the basis of these findings in 2000, with the establishment of the Dublin Cleft Centre (HSE, 2007). There are 11 multidisciplinary cleft centres across the United Kingdom and Ireland, which provide a 'hub and spoke' service,

as recommended in the CSAG report. The 'hubs' are in main population centres and are where multidisciplinary clinics and surgeries are carried out. The 'spokes' are the treatment centres, situated closer to patients' homes and provide treatments such as paediatric dentistry, orthodontics and speech and language therapy (HSE, 2007).

1.9.3 Early Surgical Intervention

Surgical repair of the lip is usually carried out between 3 and 6 months, or earlier as determined by the cleft surgeon. Advantages of early repair of the lip are improved parent-infant bonding and to reduce impact of an unrepaired cleft lip on development. The primary goals are to reconstruct normal lip anatomy and restore lip function. The secondary goals are closure of the nasal floor and correction of nasal tip asymmetry (Ness and Sykes, 1993)

A Millard rotation advancement flap (Millard, 1957) is the most common method used for repair of the lip, as it hides the scar in the base of the nostril, but a disadvantage is that it can result in reduced lip length. The triangular flap of Tennison (Tennison, 1952) is easier to carry out, creates better lip lengthening and preserves the Cupid's bow but leaves a potentially unsightly horizontal scar in the lip.

Palatal repair is carried out between 9 and 12 months and involves a palatoplasty to move the tissues towards the midline, usually using a vomerine flap (Kumar, 1985). The timing of the palate repair is a balance between the benefits of early closure on feeding and speech and the consequence of inhibited maxillary growth as a result of surgical scarring.

1.9.4 Alveolar Bone Grafting

1.9.4.1 Introduction

Alveolar Bone Grafting (ABG) is a surgical procedure, carried out to repair the residual bony defect in the anterior maxilla, estimated to be present in 75% of patients with cleft lip and palate (Cho-Lee et al., 2013). The procedure was initially described in 1986 (Bergland et al., 1986). It is usually completed between 8 - 11 years and is an important part of the reconstructive process for cleft patients. It is considered by most cleft teams as the final 'compulsory' surgical procedure (McIntyre, 2014). This is because after this stage of treatment, any further surgery, such as lip or nose revision for example, is carried out on an elective basis.

The clinical consequences of a maxillary alveolar cleft are numerous, and may include a persistent palatal or nasolabial fistula with nasal discharge and regurgitation, poor speech and articulation, lack of support for the alar base of the nose, lack of bone for eruption of the permanent dentition and anterior cross-bite (Cho-Lee et al., 2013).

1.9.4.2 Donor Site

The surgery involves the placement of an autogenous bone graft directly into the maxillary alveolar bone defect. Bone is most commonly harvested from the iliac crest due to its accessibility and large quantity of cancellous bone (Murthy and Lehman, 2005) but may also be harvested from the mandible or the cranium. Cancellous bone is quick to incorporate and vascularise and does not interfere with the development of the teeth adjacent to the cleft (Bergland et al., 1986).

Cohen *et al* (1991) compared the results of alveolar bone grafting with bone harvested from the iliac crest with from the cranium and concluded that the source of the graft did not appear to influence the success of the outcome. They surmised that the main factors contributing to success of the graft were the simultaneous closure of oro-nasal and palatal fistulae, the use of only cancellous bone particles and coverage of the graft with a well-vascularized flap (Cohen et al., 1991). A Cochrane Review in 2011 also attempted to determine whether newer grafting materials were superior to the conventional grafting method but the two studies included were of lower quality and as such no conclusion could be reached (Guo, 2011).

1.9.4.3 Rationale for ABG

Alveolar bone grafting has made a significant impact on the oral rehabilitation of children with cleft lip and palate. Ultimately alveolar bone grafting facilitates the eruption of the permanent maxillary canine but there are several benefits, which are summarized in the table below (**Table 4**) (Bergland et al., 1986).

Table 4: Rationale for Alveolar Bone Grafting

Rationale for alveolar bone grafting
Facilitates eruption of maxillary canine
Facilitates alignment and orthodontic space closure in the maxillary arch (particularly if lateral incisor agenesis)
Helps to stabilise the pre-maxillary segment
Establishes good alveolar contour
Preserves vulnerable teeth adjacent to the cleft site
Increases upper facial height
Closes fistula in the anterior palate
Enhances alar base support
Provide bone for future endosteal implant placement

1.9.4.4 Timing of ABG

The timing of the bone graft is important; it is best carried out prior to the age at which the canine usually erupts, and when the root of the canine is approximately two-thirds formed (around 8-10 years). There is, however, some variation in the timing at which the ABG is completed. If the ABG is carried out after lip repair, but before palatal repair, it is referred to as primary ABG. Secondary grafting is divided into early secondary, carried out prior to the age of 12 years, and late secondary grafting, carried out after the age of 13 years. Numerous studies have demonstrated greater success rates when grafting is completed prior to canine eruption. Earlier grafting is advocated if the developing central or lateral incisor may erupt into the cleft, thereby jeopardising the health of these teeth. Kaura *et al* (2018) in their systematic review concluded that

secondary alveolar bone grafting performed during the stage of mixed dentition provides optimal outcomes for these patients (Kaura et al., 2018).

1.9.4.5 Surgical Procedure

The surgery involves two stages, preparation of the intra-oral recipient site, then harvesting and transfer of the graft. The recipient site is prepared by raising a full thickness mucoperiosteal flap intra-orally on the buccal aspect, from the sulcus of the anterior teeth to the first permanent molar, with a relieving incision to allow reflection. If a residual palatal fistula exists, then a full thickness mucoperiosteal flap will be raised palatally also. Any remaining primary teeth will usually be extracted at the time of surgery, or a few weeks prior to the surgery. The nasal floor is closed with sutures, prior to the placement of the graft. The graft harvested from the iliac crest is carried out via a 2-cm incision approximately 1cm lateral to the iliac crest. An osteotome and curettes are used to harvest the desired amount of cancellous bone, which is transferred to the recipient site. Keratinized gingival mucosa is then advanced from the posterior segments to fully cover the graft intra-orally, the papilla are approximated and the area is sutured.

1.9.4.6 Adverse Complications

Potential adverse consequences of the ABG include graft failure, infection, wound dehiscence, root resorption of adjacent teeth, as well as donor site morbidity such as possible effects on growth, gait disturbance, haematoma formation and scarring. In a systematic review by Kaura et al in 2018, the most commonly reported adverse consequences were graft recipient site infection (1-

7%) and wound dehiscence (1-24%) (Kaura et al., 2018). However, the complication rates in most of the studies included in this review were low.

1.9.4.7 Criteria for Success

Ideally the grafted bone integrates with the alveolar bone and the canine will successfully erupt. In some cases, the canine will not erupt spontaneously and a further surgery will be required to expose the canine, with orthodontic traction applied to facilitate its eruption. One study assessed the clinical outcome of 50 patients with alveolar bone grafting over three years and found that in 94% of the sample (47 patients), the permanent canines showed movement toward the oral cavity. In 72% of those 47 patients (36 patients), the permanent canines spontaneously erupted through the grafted area. Six percent of the permanent canines underwent exposure and orthodontic traction while 12% remained under observation at the time of publication of the paper (da Silva Filho et al., 2000).

The success of ABG surgery is often described using the Bergland Criteria (Bergland et al., 1986), whereby success is measured as height of interdental septum remaining following alveolar cleft bone graft. Types I and II are commonly considered successful, while Types III and IV are unsuccessful. Diagrammatic representation of the Bergland criteria are demonstrated in **Figure 2**. Toscano *et al* (2012) looked at success of alveolar bone grafting one year post-operatively. He included 49 patients with varying severity of clefts and found success rate to be 91.84% and found the severity of the cleft pre-operatively had no statistical significance on the success of the graft (Toscano et al., 2012).

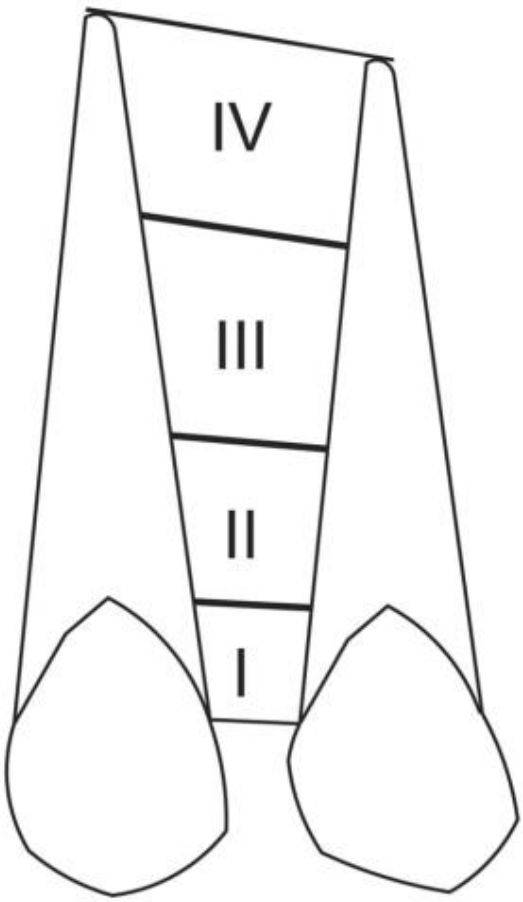

Figure 2: Schematic Representation of the Bergland Criteria

1.9.5 Pre-surgical Orthodontics

Orthodontic arch expansion is often required prior to alveolar bone grafting. This may be necessary to widen the maxillary arch, create surgical access and maximise the amount of bone placed. This is usually achieved with the use of a quad- or tri-helix device, which is a fixed expansion appliance cemented via bands to the first permanent molars. It usually achieves the desired expansion within six months. Patients will be seen on a 4-8 weekly basis to have the appliance

reactivated. The appliance is thought to achieve mostly dento-alveolar expansion with a small degree of skeletal expansion, with a ratio of 6:1 in favour of dento-alveolar expansion. Usually, slight over-expansion is achieved in order to allow for some relapse. Some initial alignment of the maxillary labial segment dentition may also be carried out prior to the bone graft. Many authors agree that pre-surgical orthodontic treatment, with or without maxillary expansion, allows for optimal preparation of the cleft site (Kaura et al., 2018). Should alignment be undertaken, it is important to be cautious in the movement of the teeth adjacent to the cleft, in order to prevent the movement of these roots inadvertently into the site of the defect.

1.9.6 Overview of Treatment of Cleft Lip and Palate

An overview of the key stages of the treatment journey is outlined in **Figure 3**.

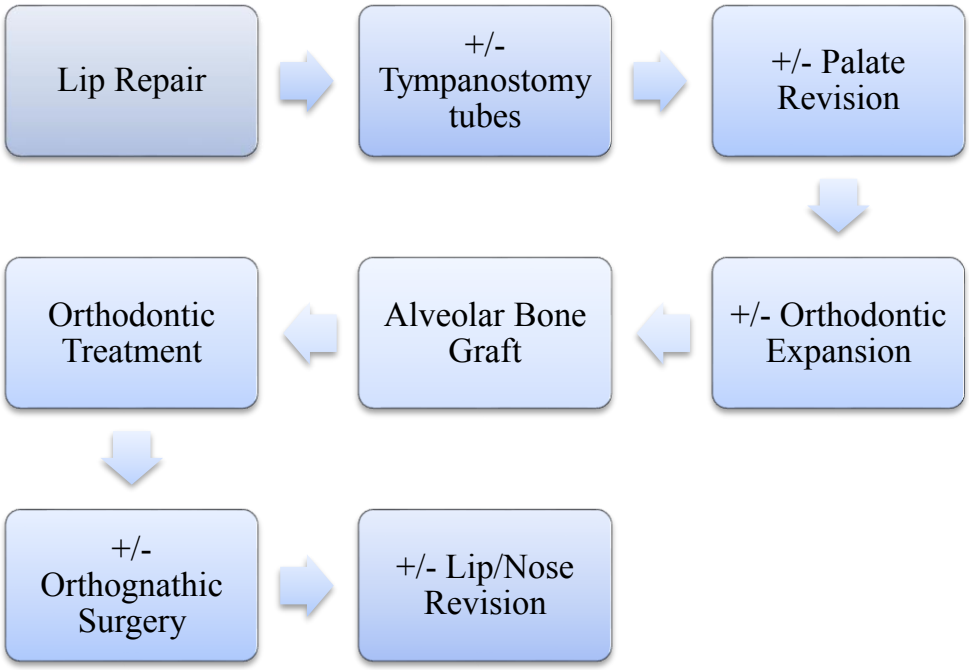

Figure 3: Overview of Treatment Journey

1.10 Provision of Services in Ireland

1.10.1 Introduction

The Dublin Cleft Centre (DCC) was established in 2000 to centralise cleft care in the Republic of Ireland. It is a multidisciplinary service for the management of cleft lip and palate as well as non-cleft related velophayngeal dysfunction. The DCC is affiliated with the Craniofacial Society of Great Britain and Ireland and submits audited data annually. Audit records are taken pre and post-operatively, as well as at 5, 10, 15 and 18 years of age. The entire multidisciplinary team meets four times a year, while multi-disciplinary clinics occur regularly and follow the 'hub and spoke' model of care, as outlined previously. The 'hub' centre is based in Dublin, with 'spoke' centres in Galway and Sligo. There are two clinics provided by the cleft surgeon and cleft co-ordinator from the DCC annually in Sligo and six

clinics annually in Galway, carried out in conjunction with the local cleft teams in the region. Care is also provided for patients with clefts by a separate team in Cork University Hospital.

1.10.2 Alveolar Bone Grafting in Ireland

Multi-disciplinary clinics are held between St James' Hospital, Our Lady's Children's Hospital Crumlin (OLCHC) and Temple Street Children's University Hospital (TSCUH). There are two Oral and Maxillofacial surgeons who carry out alveolar bone grafting surgery in Dublin, in OLCHC and TSCUH. There are approximately 25-30 alveolar bone grafts carried out in Ireland per year, most of which are carried out between these two hospitals. Approximately 3-4 of the bone grafts carried out in Ireland each year are in Cork University Hospital. Of the 25-30 ABGs done each year, the cleft care team estimate that 25% of these cases have pre-surgical orthodontics prior to this.

As the surgery is carried out in two different children's hospitals in Dublin, there are slight differences in the protocols followed and the instructions given to the patients. The post-operative protocol in OLCHC involves Elastoplast on the upper lip for a week, as well as ice packs to the site for 48 hours to reduce swelling. The advice sheet from TSCUH recommends a soft diet for two weeks whereas the leaflet from OLCHC recommends a soft diet for just one week. The recommendation for the use of mouthwash varies also, with the leaflet from TSCUH recommending mouthwash after meals and brushing for two weeks and OLCHC recommend mouthwash 8-10 times per day until reviewed back in the clinic in approximately one week. There is evidence to suggest that standardizing

peri-operative information following alveolar cleft repair significantly improves patient adherence to pre-and post-operative instructions (Chang et al., 2017).

1.11 Qualitative Research

Qualitative research can provide an in-depth insight into people's personal perspectives, beliefs, attitudes and experiences. It is commonly used to explore and gain deeper understanding of the participants' life, especially for aspects where little is known or understood (Stewart et al., 2008b). Qualitative research may involve questionnaires, surveys, interviews, focus groups, field observations or a combination of methods, which can obtain much more detailed information on the subject area. This type of research does not seek to quantify answers to research questions. Quantitative research has traditionally dominated much of healthcare research, particularly dentistry. However, qualitative approaches, which are common within social sciences, are now recognised as equally important, especially in healthcare. They are both of value, as they aim to answer different research questions.

While there is an abundance of research involving children as research subjects in the literature, nearly all of this is quantitative in nature. There is limited qualitative research involving children. This is mostly due to both pragmatic and ethical issues involved in interviewing children. However, there is research to show that conducting qualitative interviews with children can yield rich, deep, valid accounts and lead to revelations of knowledge not commonly known by adults (Gill et al., 2008). This type of research is particularly useful in acquiring

detailed information from children or those with literacy problems, as it can be acquired verbally.

1.12 Perception of services

1.12.1 Parents' perception of services

Most research shows that parents have generally positive views of cleft services, however much of this research is carried out quantitatively, with a lack of qualitative research that examines parents' perspectives in depth (Nelson, 2009). Nelson *et al* (2012a) also suggested that while the experiences of parents are mostly positive, they may have unmet needs (Nelson et al., 2012a). Some of parents' concerns related to delayed referrals, poor co-ordination of appointments and communication between clinicians as well as a lack of a single contact (CLAPA, 2007). Parents of children with clefts have a significant need for information and most of the available information is aimed at diagnosis, birth and the early years. There is significantly less information available for parents aimed at the childhood and adolescent years. The experiences of parents during these years is also under-researched (Nelson and Kirk, 2013). There is a paucity of research specifically examining experiences of cleft surgery but results of quality of life studies suggest that parents find their child's cleft surgeries to be challenging (Nelson et al., 2012b). In relation to information provision, parents show a preference for information that is individualised (CLAPA, 2007) and provided orally with written information as a supplement (Knapke et al., 2010).

Nelson and Kirk (2013) carried out interviews with parents of children with clefts to explore their perspectives of the delivery and organisation of cleft services

(Nelson and Kirk, 2013). They carried out interviews with 35 parents. In accordance with previous literature, they found that parents wanted more individualised care plans and better coordination of services with a key person with whom they could communicate. They also found that parents wanted more detailed information on the process of surgery. These needs were common across all ages and stages of treatment and regardless of the family's prior treatment experience.

1.12.2 Patients' perception of services

The vast majority of the research on patients' perceptions of the treatment journey and cleft services are carried out with patients once they have reached adulthood (Eckstein et al., 2011). More recent research aims to target the patients as children and as they undergo active intervention. Alansari *et al* (2014) carried out in-depth semi-structured interviews with 11 adults with non-syndromal cleft lip and palate in Canada (Alansari et al., 2014). There was a strong recollection of the treatment process being difficult and burdensome throughout childhood and early adolescence, and out of proportion to its potential benefits. One respondent recalled how clinicians spoke only to their parents and never addressed them directly and as such resulted in them feeling that they lacked importance or control in the treatment process. The vast majority of participants, however, had positive interactions with clinicians and remembered when they addressed them directly and showed interest in their lives, outside of their cleft.

Munz *et al* (2011) carried out a survey based study in Michigan to ascertain whether the oral health related quality of life of adolescents with clefts related to

their own and their parents' satisfaction with treatment and treatment outcomes (Munz et al., 2011). Young patients who participated in the study had a high treatment satisfaction score. The Post-Surgical Patient Satisfaction Questionnaire was used and found that both parents and patients were very satisfied with treatment (Kiyak et al., 1984). Both patients and their parents reported that they would be very likely to recommend the treatment to others. As the surveys were posted however, there may likely have been some conferring between the parent and child about their answers, which may account for the similarity in responses.

One of the aspects of the treatment journey analysed specifically in a recent publication, was the patient and parent experience of the craniofacial multidisciplinary team clinics (MDT). The findings indicated that the MDT was experienced as emotionally challenging, particularly if participants were not prepared for the number of health professionals present and their function. Myhre *et al* (2019) carried out interviews with patients and their parents about their experiences of craniofacial MDT clinics in Norway and also found that being the centre of attention and feeling 'on display' was challenging for patients and reduced their ability to process the information they received and to ask questions (Myhre et al., 2019).

1.13 Outcomes

1.13.1 Patient reported outcomes

Patient-reported outcomes are "reports that come directly from patients about how they function or feel in relation to a health condition and its therapy, without interpretation by a physician or anyone else" (Valderas et al., 2008).

Patients' perceptions of outcomes were identified as a valuable source of information, which can be used to improve services and keep treatment goals patient-centered. Historically, measuring outcomes of a treatment intervention was clinician led, using objective measurements. The appraisal of cleft lip and palate outcomes specifically, have typically focused on objective measurements as determined by the clinician (Long et al., 2011). There is now greater emphasis placed on patient-completed subjective assessments. It is becoming increasingly common to focus on patient-reported outcomes and patient satisfaction with treatment in all aspects of healthcare, as opposed to clinician opinion. There may be differences in what a clinician and a patient prioritize as important and it is vital that treatment meets the patients' perceived needs and expectations where possible.

1.13.2 Measuring outcomes in Cleft

Klassen *et al* (2012) carried out a systematic review of quality of life of children treated for CLP (Klassen et al., 2012). She reported that while health concepts were measured using various questionnaires, no specific patient-reported outcome instrument existed to measure quality of life for cleft patients. She acknowledged that the formation of a comprehensive patient-reported outcome instrument for cleft patients would be inherently difficult to create, due to the varied presentations of the condition and the large age range of individuals involved. She concluded that interviews would be necessary to identify the areas of most significance to cleft patients and that such a tool would be of benefit to both patient care and clinical research into quality of life of cleft patients.

Eckstein *et al* (2011) also highlighted the lack of a patient-reported outcome instrument for cleft patients (Eckstein et al., 2011). She identified a number of tools in the literature, few of which were validated, and thus concluded that further research was necessary to develop a valid questionnaire for measuring patient reported outcomes in cleft patients. Generic instruments such as the Paediatric Quality of Life Inventory (Varni et al., 2001) and "KIDSCREEN" (KIDSCREEN, 2006) have been used to measure patient-reported outcomes in cleft treatment but are not specifically validated for cleft patients and as such, may make it appear that interventions do not improve outcomes, as they may not be asking the right questions.

1.13.2.1 CLEFT-Q

Wong Riff *et al* (2017) carried out qualitative interviews with 136 children affected by clefts in 6 countries, in order to develop a conceptual framework for a patient-reported outcome instrument for individuals with cleft (CLEFT-Q) (Wong Riff et al., 2018). In the interviews, patients described concepts of interest in 3 main domains: appearance, health-related quality of life and facial function. These in-depth interviews have been used to form the basis of the CLEFT-Q; a questionnaire designed for cleft patients aged 8-29 years. It aims to measure the outcomes that are of most importance to patients and consists of 12 scales that assess appearance, health-related quality of life and facial function. The mean Flesch-Kincaid readability statistic for the questionnaire was found to be 1.4 (0 to 5.2) (Tsangaris et al., 2017). The questionnaire is believed to be applicable internationally, as the data from interviews was multi-centered, from various

countries and patients interviewed were from different socio-economic backgrounds (Wong Riff et al., 2017).

The results of a study utilising the CLEFT-Q questionnaire were published in 2018 (Klassen et al., 2018). Data was collected from 2434 children and young adults with cleft across 12 countries and analysed to provide evidence of validity. One hundred of these children were from Ireland and 339 from the United Kingdom. Lower scores on the CLEFT-Q scales were associated with having a speech problem, being unhappy with facial appearance and needing cleft-related surgery in the future. For most appearance scales, the results were lower in older patients. When analysed based on cleft type, the difference in mean scores were significant for 11 out of 12 scales. On all scales, patients affected by cleft lip and palate had the lowest mean scores. Patients with syndromes were under-represented in the sample and patients under 8 years of age were not included, as it was suggested that 8 years of age was the lower limit for the collection of valid and reliable patient reported outcome data. They concluded that the CLEFT-Q was a rigorous tool for measuring the impact of having a cleft on quality of life and of the impact of treatment on patients. The integration of the CLEFT-Q into electronic health records could transform health services and outcomes for cleft patients, by incorporating the patient perspective in the evaluation of treatment success and routinely evaluating and considering the effect that treatment has on this vulnerable cohort of patients.

2.0 Aims and Objectives

Aims

The aim of the study was to gain an understanding of Irish parents' and patients' experience with pre-surgical orthodontics and alveolar bone grafting as part of the management of unilateral cleft lip and palate.

Objectives

The objectives of the study were to use qualitative methods to gather information relating to:

- patients' and parents' experience of pre-surgical orthodontics and alveolar bone grafting surgery
- existing patient information resources related to alveolar bone grafting surgery

3.0 Subjects and Methods

3.1 Study Design

This was a prospective qualitative study that involved a cohort of parents and patients who had undergone alveolar bone grafting surgery in the previous twelve months. Some of the patients had also undergone pre-surgical orthodontic therapy in that time. Semi-structured interviews were conducted with the patients and their parents to assess their experience with regard to their orthodontic treatment, alveolar bone grafting surgery, and their information seeking habits prior to surgery. The interviews recorded were transcribed and imported into a qualitative software tool (MAXQDA), which allows for visualisation and organisation of the data, as well as the assignment of qualitative codes. The data was then analysed using a thematic analysis approach, where themes and subthemes were identified.

3.2 Ethical Approval

Ethical approval for the study was granted by the Joint Research and Ethics Committee of St James's and Tallaght University Hospitals (Reference number: 2017-11 **[Appendix 1]**). The approval was granted on the grounds that the parent would be present at all times when the child was being interviewed.

3.3 Participants

Participants were selected for interview using purposive sampling, based on when they underwent alveolar bone grafting surgery and pre-surgical

orthodontics. It was decided that those patients who had their surgery within 12 months would be selected, because their recall of the experience would be more detailed than that of someone who had the surgery several years previously. A shorter time frame was considered but this was not possible because the number of patients undergoing this procedure is relatively small, and limiting the time frame any further would result in a very low sample size.

Alveolar bone grafting surgery in Dublin is carried out in two children's hospitals. The cleft co-ordinator for each of these hospitals was contacted for a list of patients who had undergone the surgery within the previous 12 months. A letter was sent by post by a gatekeeper (a member of the administrative team in the Division of Child and Public Dental Health), with a Patient Information Leaflet about the study, to invite these patients and their parents to participate **(Appendices 2 and 3).** The contact details of the gatekeeper were also included, and invitees could make contact if they wished to be included in the study. If any of the invitees had any further questions about the study, the gatekeeper called them to answer these questions.

Invitees were given two weeks to consider the information. The gatekeeper followed those who had not responded up with a phone call. During the telephone call the study was explained and any further questions clarified. Those who were interested in participating were sent a consent form and a pre-paid envelope, to return the signed consent form. For those participants under 18 years of age, written and verbal consent was given by a parent or legal guardian and verbal assent was obtained from the participant **(Appendix 4)**. A suitable time was then arranged to interview either the parent alone, or the child also once the consent form had been received. The parent or guardian was requested to be in the room

with the phone on loudspeaker while the child was being interviewed. In all cases the parent was interviewed first, without the child present. The child was interviewed afterwards or at a separate time, with the parent or guardian present.

Participants were recruited until theoretical saturation was achieved. Theoretical saturation is the point in data collection that is achieved when no more new themes emerge from the latest collected data (Mack, 2005). The point of theoretical saturation was agreed upon amongst the research team on the overall quality of data obtained from each interview in light of the research aims and objectives and when the transcribed data revealed no new themes.

3.4 Inclusion and exclusion Criteria

The inclusion and exclusion criteria for the study are shown below **(Table 5)**.

Table 5: Inclusion and Exclusion Criteria

Inclusion Criteria	9 years and older
	Patients with unilateral cleft lip and palate
	Parents of patients with unilateral cleft lip and palate
	Alveolar bone grafting surgery within previous 12 months
Exclusion Criteria	Less than 9 years of age
	Clefts associated with craniofacial syndrome
	Patients who require the use of an interpreter
	Patients who were in any other research study in the past six months

3.5 Formal Qualitative Training

Formal training was undertaken by the lead investigator with the Social Research Authority in the United Kingdom (October 2017). The Social Research Authority is the professional membership body for social researchers. The courses involved lectures, as well as practical exercises in interviewing skills and analysing qualitative data. Practice interviews and focus groups were carried out as part of the course. The courses undertaken are listed below **(Table 6)**. Various analysis methods and software programs were discussed.

Table 6: Courses Undertaken with the Social Research Authority

Introduction to qualitative research
Conducting interviews and focus groups
Analysing qualitative data
Writing up qualitative data

3.6 Topic Guides and Interviews

3.6.1 Topic Guide

Prior to undertaking the interviews, two topic guides were developed, one for the patients and one for the parents/guardians **(Appendices 5 and 6)**. The topic guides were formulated based on the literature and on the supervisor's prior experience with qualitative research. A Consultant Cleft Orthodontist was asked to review the topic guides, based on his clinical experience of the service and the

surgery for feedback. The lead investigator also attended several cleft clinics, to gain an insight into the multi-disciplinary environment and the advice and information provided to the children and their parents by both the orthodontist and the oral and maxillofacial surgeon. The topic guides were then amended accordingly.

The topic guides were intended to act as a prompt during interviews, but were not to be strictly adhered to. The topic guides were divided into sections, with the main areas included listed in Table 7. The interviews were semi-structured in nature so the main items in the topic guide served only as a reminder of the areas to be discussed. The semi-structured format of the interviews allows participants to discuss areas that are not included in the original topic guides. The topic guides were then modified after each interview to include new and emerging themes that were not initially included. This modification of the topic guides continued until no new themes emerge.

Table 7: Main Areas discussed in Topic Guides

Parent/Guardian Topic Guide	**Child Topic Guide**
Introduction	Introduction
Participant background information	Participant background information
Child background information	Experience with fixed appliances
Pre-operative information and communication	Experience of surgery
Experience of surgery	Advice for other children
Experience of orthodontic treatment	Any other issues
Advice for other parents	

3.6.2 Reflexivity

Reflexivity is a concept used in qualitative research as a means of reducing bias and limiting the influence of the prior assumptions and opinions of the researcher in shaping the collected data (Mays, 2007). Ideally the interviewer should not be a clinician with detailed knowledge of the treatment process, as this could influence the responses of the participants in the interviews. This was not feasible, however, due to limited resources, and the main researcher in this study was also the interviewer. Efforts were made however to limit the influence of the researcher on the collected data, by following the pre-prepared topic guide in a neutral and non-leading manner. The interviewer carried out practice interviews in order to prepare and improve on the interviewing technique. The interviewer was also unknown to the participants and avoided dental jargon.

3.6.3 Practice Interviews

Prior to undertaking interviews with participants, practice interviews were undertaken in the Dublin Dental University Hospital. The lead researcher interviewed one of the research supervisors and other colleagues, who acted out scenarios. These interviews were recorded and assessed, with constructive feedback given and the interview technique modified accordingly. The topic guide was amended following the practice interviews also. These practice interviews prepared for deviation from the topic guide and for the management of sensitive issues, were they to arise, as well as avoiding leading questions which may influence the data, as discussed above.

3.6.4 In-depth Interviews

In-depth, semi-structured interviews were carried out in a private room within the orthodontic department in the Dublin Dental University Hospital. The interviews were carried out over the phone and recorded using both transcription software and a digital dictaphone. Participants were advised that the interviews would be recorded and their verbal consent was obtained prior to the interview beginning. They were also informed that they could withdraw from the interview at any point, and that they were not obliged to answer a question if it made them feel uncomfortable. Interviews were carried out with no set time limit. The interviews continued until the participant had no further information to give and recruitment and interviews continued until no new themes emerged and theoretical saturation had been achieved.

The interviews were transcribed verbatim by a transcriptionist in Trinity College Dublin using Microsoft Word©**.** All interviews and transcribed data was anonymised and uploaded through a secure file sharing method, only accessible by the lead investigator and the transcriptionist. All the interviews were then permanently deleted by the transcriptionist and from the dictaphone. The transcribed data was stored on a secure drive, only accessible by password by the lead investigator and supervisors.

3.6.5 Analysis of Data

A qualitative software programme, MAXQDA© (VERBI, Berlin, Germany) was used to aid in data management and analysis. It is a software package for qualitative and mixed methods research, which is distributed by VERBI Software (Berlin, Germany). In comparison with manual methods, computer assisted

qualitative data analysis software aided the process of data analysis and facilitated organisation and display of data in a more systematic and accessible way to research team members (Ritchie, 2013). The software interface for MAXQDA© is illustrated in **Figure 4**.

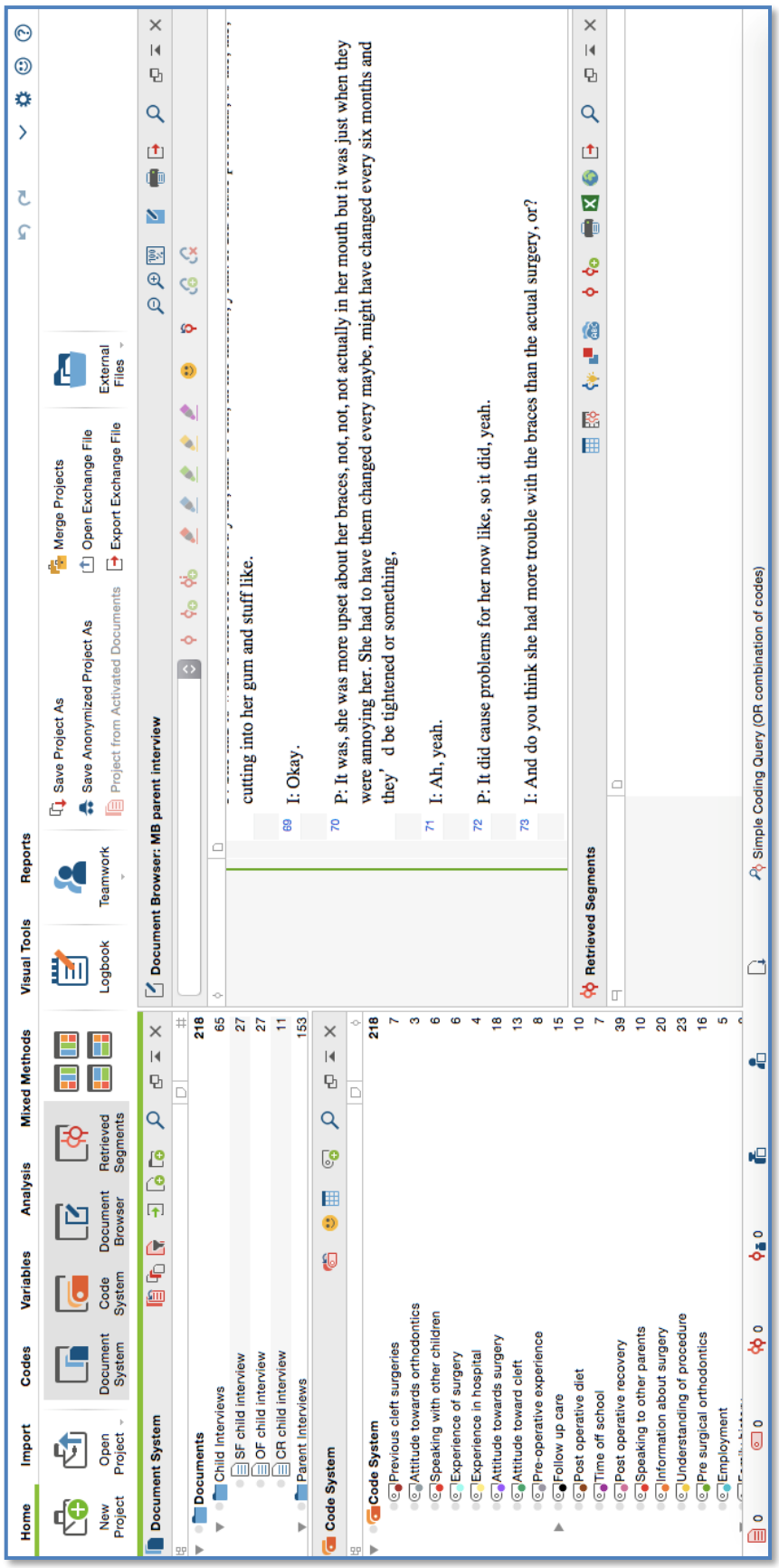

Figure 4: MAXQDA© Interface

Four main display windows provided the following features:

I. A Document System Window, where data was imported in the form of text documents, video and audio files or images. Imported data files could also be grouped through this window.

II. A Code System Window, which allowed the creation and assignment of codes to sentences or paragraphs within the documents. Codes were ordered into a hierarchical structure; a main code/theme which could have several sub-themes.

III. A Document Browser Window, which allowed visualisation of the selected document, so codes could be assigned to text segments.

IV. A Retrieved Segments Window, which allowed retrieval and visualisation of all sections from the various documents, which were assigned the same code or theme.

The data from the transcribed interviews was uploaded into MAXQDA© and analysed using a thematic analysis approach, which allowed for systematic analysis of the data. The data analysis was an iterative process that began in the early stages of data collection and continued throughout the study. It involved multiple steps as illustrated in **Figure 5** and described in further detail below.

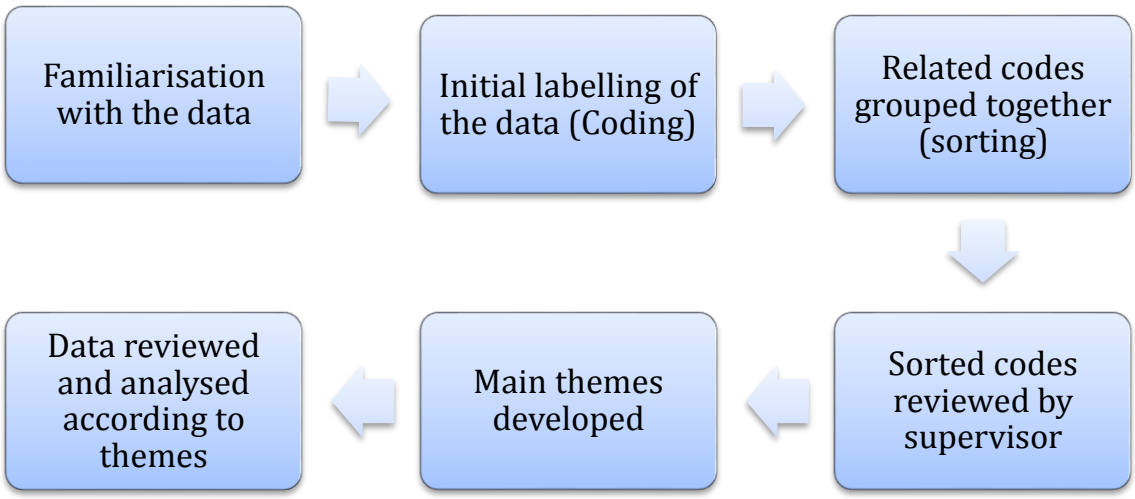

Figure 5: Process of Data Analysis

The first step was familiarisation with the raw data, by reading through the transcripts several times and gaining a thorough insight into the content of each transcript. Through conducting the interviews, the lead researcher had an impression of the data and an initial knowledge of its content, prior to reading the transcripts. The key sentences and phrases were then labelled in MAXQDA© (coding). This involved reading each transcript and highlighting segments and assigning them a colour and an initial label/code. Each segment of the transcripts may be assigned several codes, should the segment be relevant to more than one topic.

Once coding was complete, the researcher reviewed the codes that had been generated from all the data and grouped together the codes that were related (sorting) (**Figure 6**). The researcher was then able to extract initial themes and sub-themes from the data, which were reviewed by the research supervisors. The

transcripts were sent to the research supervisor and to an experienced qualitative researcher, who also helped identify the themes. This aided in the consistency and comprehensiveness of the analysis and limited researcher bias. Agreement was reached on the emergent themes and the data was deemed sufficient to reach thematic saturation.

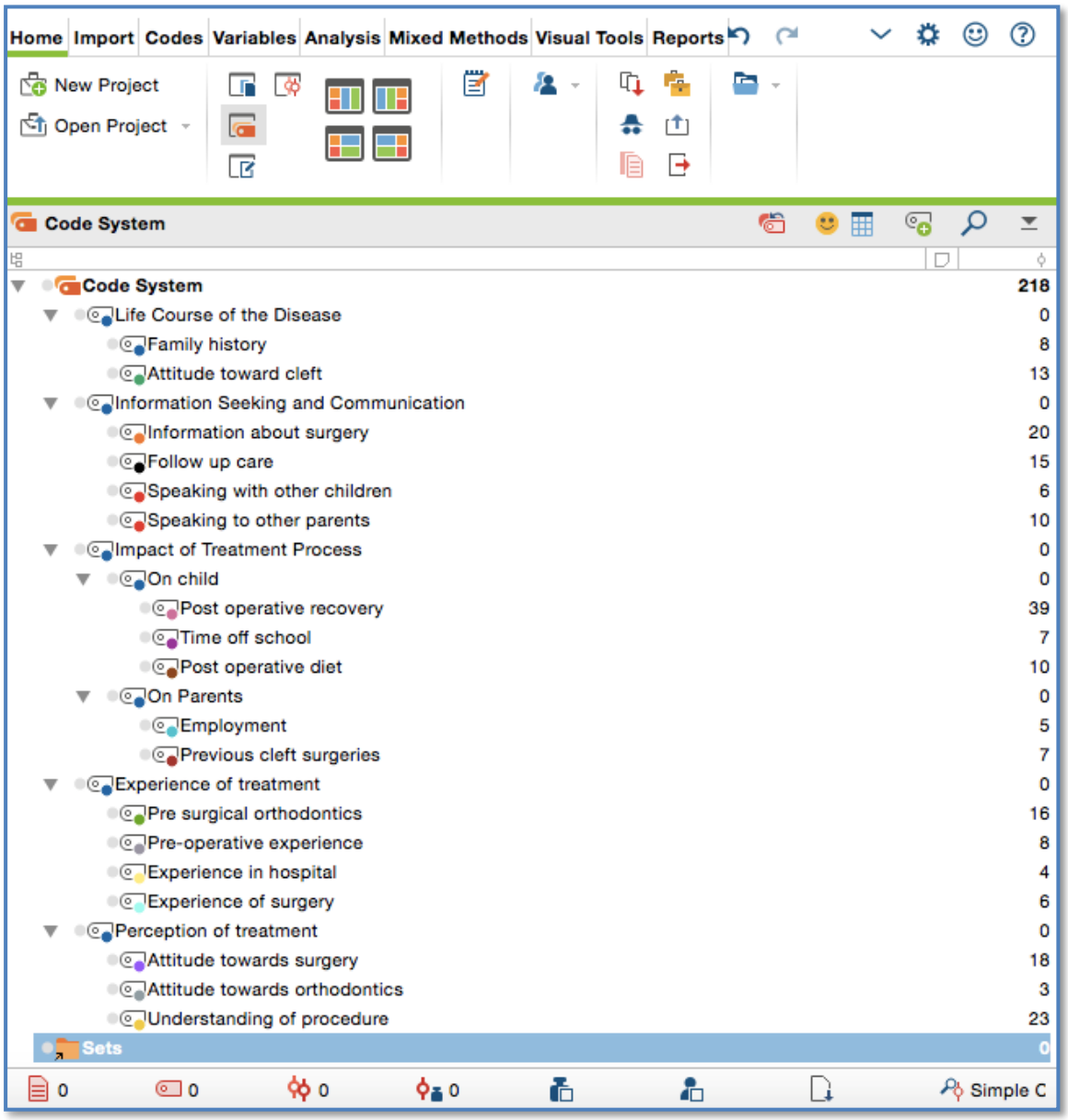

Figure 6: Grouped codes in MAXQDA

3.6.6 Rigour and Trustworthiness

Rigour and trustworthiness were established in adherence with guidelines for the conduct of qualitative research (O'Brien et al., 2014). Any ambiguity in the data were clarified with the research supervisor and all transcripts were reviewed by the supervisor as well as an experienced qualitative researcher to ensure theoretical saturation had been achieved and to ensure comprehensive coding and data analysis. Mays and Pope (2007) state that the strength of qualitative research lies in its validity, which is the extent to which the findings accurately represent the social phenomena studied (Mays, 2007). The checklist of standards for reporting qualitative research (SRQR), containing 21 items and developed by O'Brien et al 2014, was referenced and adhered to throughout (O'Brien et al., 2014) **(Appendix 7).**

3.6.7 Summary of the Method

The methodology employed is summarized below in **Figure 7**.

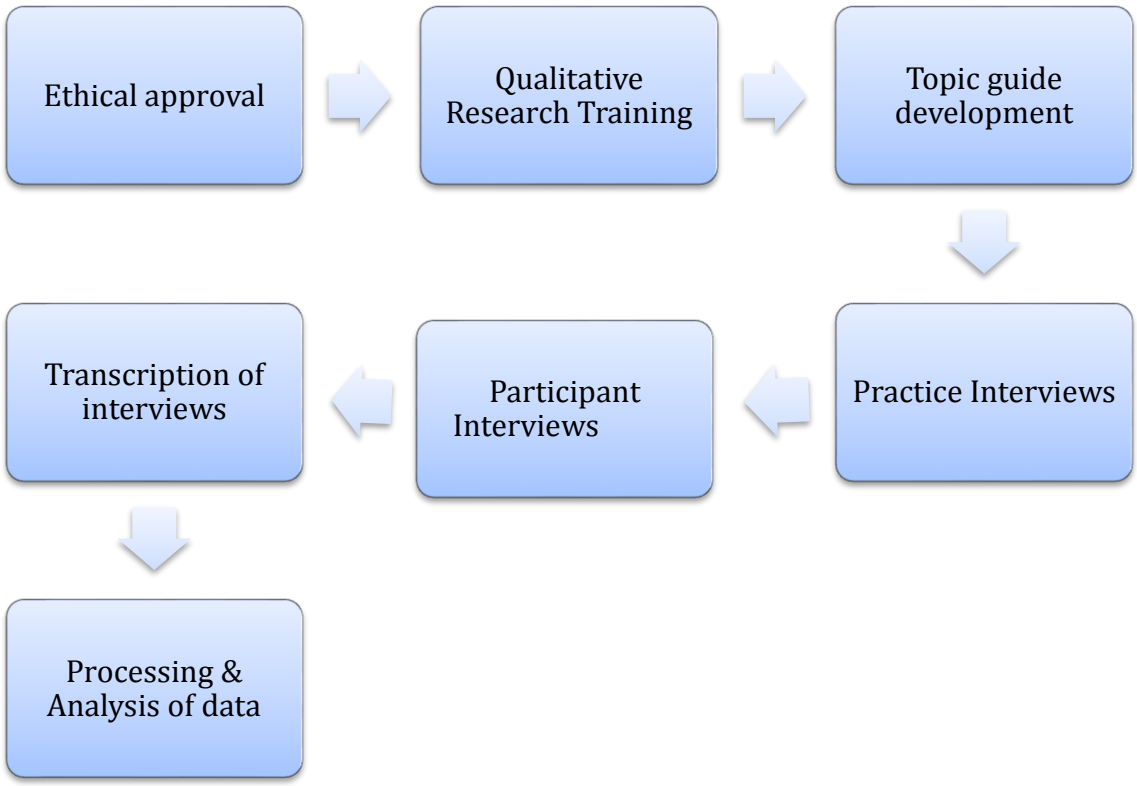

Figure 7: Summary of the Method

4.0 Results

A total of 22 participants were contacted, 11 parents/guardians and 11 children/patients. As these patients and their parents attend multiple appointments, a voucher was offered to thank them for their time and contribution to the study. Three of the 11 parents responded to the initial invitation letter expressing interest in participating for themselves and their child (27% response rate of the parents to the invitation letters). All of the others were followed up with a phone call and a further three parents agreed to be interviewed. Three of the children declined to be interviewed because they were too shy. Three of the parents and children could not be contacted on the contact number provided and two parents declined on behalf of themselves and their child, as they did not wish to participate. Nine invitees in total agreed to participate (6 parents and 3 children). The overall uptake was 41%. All of the participants had their ABG surgery carried out in the one centre (TSCUH).

4.1 Participant demographics

Table 8 shows a summary of the parent/guardian demographics and **Table 9** shows the child demographics. None of the parents/guardians interviewed had a cleft, nor was there a significant family history of clefts reported. In each case it was the family's first experience of a cleft. The parent contacted by phone was the parent listed as the contact details on the child's hospital chart. All of the parents/guardians interviewed were the primary caregivers of the child and had attended all appointments and surgeries with the child. The interviews were

carried out over the phone at a time that was convenient for the parent/child. Where children were interviewed this was usually in the afternoon, when they were home from school.

Table 8: Parent/Guardian Demographics

Patient Identifier	Relationship to child	Gender	Employment	Family history
P1	Mother	F	Catering	No
P2	Mother	F	Catering	No
P3	Grandfather	M	Retired	No
P4	Mother	F	Homemaker	No
P5	Mother	F	Accountant	No
P6	Mother	F	Waitress	No

Table 9: Child Demographics

Child Identifier	Age	Gender	Family history
C1	10 y	F	No
C2	11y	F	No
C3	11y	M	No

4.2 Interview duration

The interviews had no time limit and, as they were semi-structured, they varied in length. The parent interviews varied in length from 13 to 27 minutes with a mean time of 20.5 minutes and the child interviews ranged from 7 to 13 minutes with a mean time of 10.6 minutes. In total there were 132 minutes of parent interviews and 31 minutes of child interviews, which equated to 95 and 21 transcribed pages, respectively.

4.3 Main Themes

Following analysis of the entire data set, six main themes were identified with associated sub-themes **(Table 10)**. Each theme, and its sub-themes, was discussed in turn. Quotes were used to support the issues raised, for which participant numbers are included (e.g. P1 indicates Parent 1, C1 indicates Child 1). Quotes were chosen which the researcher and the supervisors felt best reflected the themes and subthemes. Explanatory comments were added where appropriate. There was significantly more data from the parent interviews and as such some of the themes relate to the parents only, namely Diagnosis and Coping Mechanisms. For the other themes, which are common to both parents and children, the data is discussed separately (Perception of Treatment, Experience of Treatment, Impact of Treatment and Information Seeking). This is apparent in **Table 10** below.

Table 10: Main Themes and Subthemes

Theme	Subthemes
Life Course of the Condition*	Diagnosis Experience Prior to Initial Surgical Repair Previous Cleft Surgeries
Perception of Treatment	Parents Perception of Treatment ➢ Understanding of Orthodontics ➢ Understanding of ABG ➢ Understanding of Risks Child's Perception of Treatment ➢ Understanding of Orthodontics ➢ Understanding of ABG
Experience of Treatment	Parents Experience of Treatment ➢ Experience of Orthodontics ➢ Experience of ABG ➢ Satisfaction with Treatment Childs Experience of Treatment ➢ Experience of Orthodontics ➢ Experience of ABG
Impact of Treatment	Impact of Treatment on Parents ➢ Psychological Impact ➢ Logistical Implications ➢ Post-operative Recovery Impact of Treatment on Children ➢ Psychological Impact ➢ Time off School ➢ Post-operative Recovery
Information Seeking	Information Seeking Communication with Care Team Communication with Other Parents Communication with Other Children Advice for Others Undergoing Treatment
Coping Mechanisms*	

* relates to parents only

4.3.1 Life Course of the Condition

4.3.1.1 Diagnosis

The diagnosis of the cleft during pregnancy was naturally an emotive topic for most parents. Most described their shock and upset, as well as their lack of prior knowledge or experience with clefts. One mother reported how she had never heard of a cleft lip or palate until she was advised at her scan.

> ***P2:*** *"No, I wouldn't have known until eh, like I didn't even know what a cleft lip and palate was. At nineteen weeks pregnant, I knew I was having a baby with a cleft palate, or cleft lip"*

This mother explained how unaware she was at diagnosis of the impact the cleft would have and of the long treatment journey ahead.

> ***P2****: "And so I didn't even know what that meant. I didn't realise there was so much to do with his speech and about his eating and that. I just thought it was just like a hare's lip and you just got it sewn together and that was it."*

One mother described her reaction to the initial diagnosis and her limited experience of clefts prior to the diagnosis.

> ***P1:*** *"Em. At the beginning yes, when I, I found out in my pregnancy, I found out my em, my scan, and yeah, I was when I was told about it, I was a bit shocked.*
> *I didn't know exactly what it was, I just knew, an old man years ago that he had one, like, so I thought it was going to be like that."*

The feeling of shock and sadness at the diagnosis was common to all the parents interviewed.

> ***P6**: "It's, it's very, very, very big shock. I don't know how other parents, but I'm shocked and my husband as well, shocked, you know."*

> ***P6**: "first shock to see your children is cleft lip palate and mine not perfect and I'm shocked to know and I was crying and crying and crying and, I'm, oh Jesus, and you know don't, don't, don't know what really to do"*

This mother also described wondering if she had done something wrong and the tests she underwent after the diagnosis in order to try and understand how the cleft had occurred.

> ***P6:** "..and you know you're thinking what you do wrong or what, why that's happened, you know. It's, it's hard."*

> ***P6**: "I went to genetic tests, and doctors say it could be, can be from, because I was having, you know, first three months, I was sick. I had high temperature for three, and, eh, for three months and I'm having antibiotics, antibiotics, every, you know, every week for three months, it could be maybe that some, can affect something."*

4.3.1.2 Experience Prior to Initial Surgical Repair

Some of the mothers discussed the difficulties they had with feeding prior to the initial lip or palate repairs, as well as their worries and concerns about the babies' weight loss.

P2: "*When he was born, it was hard now to feed him. He was trying to suck all the time, and it was an awful lot of milk coming down his nose. And there was em, he was actually losing weight, but I think most cleft babies do. He lost a lot of weight and I was kind of a bit concerned because instead of him getting heavier he was getting lighter"*

P6: *"And I need clean up, you know, the deep inside, that, that palate.*
Yeah, I clean, clean up all the time, you know, try be clean and oh, it's, it's very difficult…I, when you remember it all, I want to cry"

4.3.1.3 Previous Cleft Surgeries

This mother described the number of surgeries her eleven-year old son had undergone prior to the alveolar bone graft, which highlighted the extent of treatment, intervention and surgery this child underwent.

P2*: "..he got his lip done in, around eleven weeks, then he got his first em palate operation and they found that it went okay, but it wasn't good enough. So eh before he went to school, he got eh his palate done again. And then he got it done again at em, about two years ago. So he actually had three palate operations done. And he got two sets of grommets in between"*

She also described the improvement her son had after the initial surgeries.

> ***P2**: Yeah, yeah, well he still goes to speech therapy weekly, but it was far better, his speech* [after the third palatal surgery], *he was a bit, a bit nasally before that.*

4.3.2 Perception of Treatment

The perception of the treatment related to the understanding the participant had of the treatment they or the patient underwent, namely the orthodontic treatment prior to alveolar bone grafting, and the grafting surgery itself. This was discussed separately for the parents and the children. Parents were asked more specifically about the risks of treatment also.

4.3.2.1 Parents' Perception of Treatment

The parents' level of understanding of their child's treatment varied. Some had quite a detailed understanding of the procedures, risks and associated benefits, while others did not have a detailed understanding. Most parents, however, displayed a sufficient understanding of the treatments.

4.3.2.1.1 Parents' Understanding of Orthodontics

Three of the parents interviewed had children who had undergone orthodontic alignment or expansion prior to the completion of the graft. One of the parents (P4) appreciated this was in order to widen the palate and increase the area available for grafting, while another was not sure of the exact reasoning, but was

aware that it was to prepare for the surgery (P1). The grandfather of one child explained how it was to straighten his teeth (P3).

> ***P4:*** *"em, just to make more room, you know, around where the graft was going in, to widen that, move the teeth a little bit and widen her -her palate, as such, or whatever, yeah."*

> ***P1:*** *"I'm not really sure exactly, like they explained to me and to get her gum all ready, ready for the operation like."*

> **P3***: "He, yeah, he wore eh a wire going across his teeth to try and straighten them, at one stage."*

4.3.2.1.2 Parents' Understanding of Alveolar Bone Grafting

Parents were asked to explain their understanding of the alveolar bone graft, and the need for it. Most of the parents understood that there was gum missing in the palate, and some had an understanding that the ABG was to help the adult teeth erupt.

> ***P1****: "Is it, because she had no, like she had no kind of em, gum there or, so they had to take a bit of, piece of em the hip up into the gum, so the teeth coming, so they could have room to come, they have to come, come down like."*

P2: "*Yeah, completely, completely different thing, isn't it? It's more to do with the, em, it's more to do with the teeth coming down"*

***P2**:" Em, so then his second teeth, em, that's coming down will come through better. That's what I, I believe. And that he'd have a nicer set of teeth at the end of it"*

One mother had a clear understanding of the procedure and the treatment timeline and summarised it quite succinctly.

P5: *"I suppose what I would call the gum area, eh, wasn't formed properly, and I suppose she had the initial lip repair to repair the lip and the, the palate repair to do the soft and hard palate, and I suppose the remainder bit was this gum area, I suppose, as I would call it, em, and that the, the repair of that was more time-sensitive in the sense that it was to do with the growth of the adult teeth, and when they were at the, you know, most opportune time for the bone grafting to, to take place."*

This mother also discussed how her daughter had a residual fistula after the primary palate repair and that she was aware this would be repaired at the time of the alveolar bone graft.

P5: *"They said to us at that stage, look, we did as much of a repair as we could, there is a small fistula left, which would be repaired at the time of the bone grafting."*

4.3.2.1.3 Parents' Understanding of Risks

Parents described the concept of graft failure and tooth eruption failure as potential risks of the surgery, but there was no distinction made between the two.

P4: *"I suppose they would have said that in certain cases the bone just may not take fully, and indeed you may not know for like a twelve-month period, really I suppose the test would be does the tooth ultimately come through that new bone?"*

When prompted about the option of exposing the canine, should the tooth fail to erupt, she advised that she was just told that there were other ways to manage it.

P4: *"We hadn't gone into the detail of that, but they did say that there were other ways of dealing with it as well, if there, if there was a problem down the road."*

4.3.2.2 Children's Perception of Treatment

The children's perception and understanding of treatment would be anticipated to be more simplistic than that of the parent or guardian, as there may have been an element of trying to protect the child from some of the details, which they may have found distressing. The children interviewed, however, showed quite a

perceptive understanding of their treatment, which they were able to articulate coherently.

4.3.2.2.1 Children's Understanding of Orthodontics

One of the children interviewed had undergone orthodontic treatment prior to the alveolar bone graft. Her understanding did not seem to relate the orthodontic treatment to the bone graft but more so for alignment of her teeth.

> **C1: "***Em, because my tooth was em lopsided, kind of, so it was, it wasn't, it was facing the wrong way. It was facing sideways instead of em showing the, the front."*

4.3.2.2.2 Children's Understanding of Alveolar Bone Grafting

Two of the children gave a clear and succinct interpretation of the indications for their alveolar bone graft, demonstrating an understanding of the removal of bone from the hip to support the gum and to facilitate the eruption of the teeth.

> **C1***: "Em, yeah, because when I was born, I only had, on my top lip em there was a bit missing I think? So em, so when I was born, they had to just do a little surgery and then em they, they just had to, to put bone from my hip into my mouth, because I didn't have enough in there."*

C2: *"Em, I was born with a cleft lip and palate, so they had to put bone into my gums to help support it, I think. And they got that bone from my hip… Just to help my teeth grow."*

One of the girls explained further about the removal of two supernumerary ('extra') teeth during the surgery.

C2: *"Em, they took one out because it was wobbly and then they took two extra teeth that I had in my gums."*

4.3.3 Experience of Treatment

4.3.3.1 Parents' Experience of Treatment

Parent's experience of this stage of treatment appeared to be affected by their prior experiences of cleft surgeries and their ability to cope with their child undergoing surgery seems to increase as they progress further along the treatment journey. This was evident in some of the quotes below.

4.3.3.1.1 Parents' Experience of Orthodontic Treatment

The experience of orthodontic treatment was described by one of the parents as somewhat problematic and uncomfortable. One mother implied it to be worse than the alveolar bone grafting surgery itself.

P1: *"She had to wear a brace for about a year, kind of em, in her mouth, yeah. It did cause problems, cutting into her gum and stuff so it was quite uncomfortable like.."*

"She was more upset about her braces, not, not, not actually in her mouth but it was just when they were annoying her"

"That's when we had trouble. That was the worst part. She was very stressed herself, you know, we were a bit stressed with that as well like."

While another of the patients, who was slightly older undergoing this stage of treatment, had no problem with the braces, according to his grandfather.

P3: *"That was fine on him.* [the brace]. *He actually forgot that he had it on, at one stage"*

The difficulties associated with the oral hygiene and maintenance of the orthodontic appliance, as well as gingival issues, was mentioned by one of the mothers. Otherwise, the orthodontic appliance seemed to have been well tolerated.

P4: *"She was fine. There was a while when her gums started to kind of I suppose recede a bit or they were, you know, bleeding and that, so you know, we had to get on top of that and just the cleaning and stuff."*

"No, I don't think they ever had any issues with any of them breaking or anything."

4.3.3.1.2 Parents' Experience of Alveolar Bone Grafting

The impression of the parent experience of alveolar bone grafting surgery was that it was another routine surgery, and of its relative insignificance in comparison to the experience of the primary lip and palatal surgeries, which were more distressing for them.

P5: *"…and look, I mean it's the same, like she's had quite a few surgeries along the way and you know what you, you know what to expect"*

P4: *"Well I was just, I was fine about it, we just knew it had to, had to be done and we, you know, I mean at the time, with, with the length of time we were waiting for it, we just wanted to get it over and done with"*

P4*: "I mean, I suppose to me it wasn't a big, you know, it wasn't a big procedure, I don't, you know, it's, I wasn't worried about it"*

However, one of the mothers seemed to feel a little unprepared leaving the hospital and also felt an extra night in hospital would have been beneficial. Although she was given a post-operative instruction leaflet, she expressed surprise that no one went through the leaflet with her.

P2: *"I just think, you know, just to do with another night, a third nights stay in, just to get him walking and just for the eating and the pain relief. But you know, just to wind it down a bit more. I just thought it was very fast, from Tuesday to Thursday."*

***P2**: "I thought there was a woman would come round to me, when I was in the hospital, to go over stuff, more stuff on it, you know, it wasn't as much explanation as I thought I was going to get."*

4.3.3.1.3 Parents' Satisfaction with Treatment and Service

Most of the parents expressed significant satisfaction with their child's treatment, the outcomes and the care they received.

P1: *"I have to say they're all very good in Temple Street, yeah, and even the surgeon who did the operation, he was excellent so he was."*

P6: *"Thank God at having so good doctors and do a good job."*

One of the mothers expressed some disappointment that one of her son's supernumerary teeth was not noticed prior to the surgery, and was picked up on afterwards at a check-up appointment.

P2: *"They said that he needed three teeth removed. So they done that during the bone graft, and then we were up for his check-up and we were a wee bit disappointed that they missed a tooth and he actually had four teeth to come out."*

In spite of this, it was evident that there was an effort made, on the part of the mother, to justify and understand how it had occurred, as well as putting their faith in the clinicians, demonstrating the attributes of a trusting doctor-patient relationship.

P2*: "I mean if, it could be just a thing they couldn't see it in that eh, big x-ray, you know like, for his tooth…*

We just put it in their hands and they said lookit, they'll look after that when he's getting his, his further operations on his teeth."

4.3.3.2 Children's Experience of Treatment

4.3.3.2.1 Children's Experience of Orthodontic Treatment

One of the children interviewed (C1) had undergone palatal expansion, as well as labial fixed appliances. She reported finding the labial appliances more uncomfortable than the tri-helix, as they were harder to clean and hard to close her mouth around the appliance.

C1: *"Em, it was a bit em uncomfortable and it was a bit awkward to eat. And em it took a lot of cleaning. In between them and if food got stuck in it too, it'd be kind of hard to get it out."*

"Just to close my mouth, it was em a bit annoying. It was em, sometimes it'd be hard to close my mouth sometimes, to put my lip over it."

When asked which experience was more difficult for her, between the orthodontic treatment and the bone grafting surgery, she felt that they were difficult in different ways.

***C1**: "I thought the, the braces were annoying, but em, the hip was like sore. So it's kind of hard to decide."*

4.3.3.2.2 Children's Experience of Alveolar Bone Grafting

Most children remembered staying two or three nights in the hospital post-operatively. For one of the children, the part she remembered as most difficult was the induction of the general anaesthesia.

C1: *"I remember getting the gas mask, em, and lying on the bed, and em I remember I was really em, I, I was really nervous, so I put the gas mask on and then he told me to breathe slowly and then I got a bit frightened so I started like, I kind of started breathing heavily and then I got too frightened, and then he told, and then he kept telling me to breathe slowly, and then finally I, I, I br-breathed slowly."*

One of the other children reported thinking it would be worse than it was and was relieved when it was all over.

C3: *"I thought it'd be like a little bit sore or like take longer but it was actually good when it was like done and over with."*

The feeling of trust had an impact on the experience and on the emotional response of the children. The importance of familiarity with the team members was apparent; as it helped the children feel more at ease during the surgical experience.

C1: *"Em, I was a little bit nervous. I was very nervous at the start but then I kind, I was kind of more relaxed when I, I knew I had like em, I, you know, I could trust them and all."*

"Like I, I could, like I remembered the doctors I met before"

4.3.4 Impact of Treatment

The impact of treatment is discussed separately for parents and children and according to relevant subthemes, such as psychological and logistical implications.

4.3.4.1 Impact of Treatment on Parents

4.3.4.1.1 Psychological Impact on Parents

The feeling of guilt experienced by some mothers at the diagnosis stage was apparent, which seemed to be exacerbated by the comparison to other 'normal' children.

> **P6***: "I never have in my life know, don't know what is it exactly. I never see and, you know, and bring children and all the children beside it's fine and I'm alone in there and I'm crying, I don't know how many days, and not can stop it."*

This feeling of guilt seemed to follow on with a desire to achieve normality for their child, as well as a worry that they would experience bullying. This seemed to serve as a motivating factor for them and made it easier to cope with and justify the multiple appointments and surgeries their child underwent, knowing it would ultimately result in their child appearing normal and not feel different to other children.

> **P6:** *"I want him be beautiful boy, not, you know, not different for other children, you know, so no one bullying him at school you know, it's not nice."*

The worry of having more children and whether or not they would also be affected impacted on the parent also and was a concern for them.

P6: *"What you do wrong and you know, especially could be, if that's your first children, you know and what if you have second children"*

The condition and treatment process naturally had an emotional impact on parents, but the impression was that parents were content once they felt their child was happy.

P3: *But once he was happy, that was alright with us.*

4.3.4.1.2 Logistical Implications for Parents

Although the parents of these children appear to have built up a certain tolerance to the multiple procedures and surgeries their children undergo, the experience of the surgery and multiple appointments was unpleasant nonetheless, particularly the logistical issues of spending considerable time in a busy children's hospital.

P5: *"I mean, I suppose as a parent, it's never the most pleasant experience, you know, when your child's having something done, but you know what to expect, you're sleeping on the floor, you know like. It's not ideal."*

For some parents, who were not from Dublin, the frequent travelling up and down to appointments was tiring, time consuming and costly. One mother (P1)

commented on the multiple trips from Sligo, in the West of Ireland, approximately 200 kilometers from Dublin; a three-hour drive each way.

P1: *"We were up and down to Dublin like, I think once a week, was it, for about six weeks or something"*

4.3.4.1.3 Post-operative Recovery

The post-operative recovery period was difficult on the parents as well as the patient. Firstly the child was incapacitated to a certain extent and their mobility was impaired from the surgery at the donor site. Secondly their diet was quite restricted, which was time consuming for the parent, due to the need for a blended, liquid diet for a few weeks. One of the mothers was surprised at the extent of the mobility issues associated with her daughter's hip, and felt she had been unprepared for this.

P5: *"And it, that was a, that was a surprise to us, because like even getting her into the car when we were leaving Temple Street, like we, we had the, it took two of us to lift her into the car."*

This mother felt the focus had been on how to deal with the mouth post-operatively, and not as much emphasis placed on the hip.

P5: *"The one area that I felt I wasn't prepared for at all was the issue with her hip. I suppose the focus had always been where was the bone going to and you know, there'd be swelling in the face and that the feeding would be an issue, you know, on a soft food diet for a while."*

"*But we actually found her, it was her hip was much longer to recover, and much more difficult, just very practical sort of getting her out of bed the first day."*

"There was probably a four-week period where even getting up and down the stairs was, was very difficult for her."

"I suppose it was a, it was really just a practical thing, like I mean she couldn't get up to the bathroom in the middle of the night and she would have been waking those first couple of nights for pain relief, but you know, she couldn't get out of the bed to tell us she needed pain relief."

Some other parents also felt the hip was worse than the mouth, in terms of pain experience.

P6: *"Hip is more, more painful than mouth, yes him, him never ever say I have pain in my mouth, he said all the time hip, I'm very bad pain hip, around maybe two weeks him, him say I've pain, pain, pain, you know."*

However, this was not the experience of all the participants, which highlighted the individual experience of pain, and different coping abilities of both parents and children. Some parents reported very little post-operative difficulties.

P1: *"No, she recovered very quickly. I'd say like by the, you know, she got it done on the Tuesday, by the Friday she was great like."*

P4: *"Sure I thought she would have been, you know, I thought she would have been hobbling around a lot more like, you know, and, than she was, you know, she, she definitely, you know, she recovered very quickly from it."*

The swelling and bruising on the face was discussed, but parents seemed to anticipate this and have been prepared for those particular post-operative complications.

P5: *"There was a lot of swelling, yeah, on, eh, her, her cleft is on the left side, so yeah, a lot of swelling on the left, you know, right up to the eye level and a lot of bruising as well. I would have anticipated that she would have been, yeah, like that."*

4.3.4.2 Impact of Treatment on Child

4.3.4.2.1 Psychological Impact on Child

The impact on the children of the alveolar bone graft appeared to be more significant than it was on the parents. For the child, this surgery was at an age where they were old enough to have an understanding of what was going on, and to experience emotions of fear and anxiety. One of the quotes from a mother demonstrated the impact it had on her son, when he was told he would have to have surgery, and of his fear of the pain associated.

P6: *"You know, afraid the pain and everything, you know. Him afraid, yeah. Him crying, lots and lots"*

One of the children interviewed volunteered how the children in school were interested in what had happened to her after the alveolar bone graft, which highlighted the extra attention that she received from her peers. When C1 was asked how it made her feel, she responded that it didn't bother her.

C1: *"And then at school everyone was kind of asking me, like what happened and how was it and was it sore and whatever, like that"*

"I didn't, I didn't really mind telling them about it. I, they, they were just wondering"

4.3.4.2.2 Time off School

Most of the children reported being off school for 2 or 3 weeks. For some, this conveniently fell over holiday periods so they didn't miss too much school. One of the mother reported the concern her son had about missing school.

> **P6***: "and you know, he say why I can't go to school, why I can't eat normal foods and just after, you know"*

Some of the parents had concerns about missing out on schoolwork, for those whose surgery didn't fall over a holiday period.

> **P4:** *"I spoke to the teacher before and they knew it was coming up and I had said, you know, to send home, send home whatever homework"*

One of the children was happy to go back to school, because she had nothing to do at home.

> **C2***: "It was a bit boring being at home."*

The greater concern for many of the parents and the children was the length of time that they would be unable to play sport. For one boy and his mother, their biggest concern was not being able to play football, and he was happy when he could play sooner than anticipated.

C3: *"I thought I wouldn't be able to play football for eight weeks, but actually four or five."*

P2*: "How am I going to keep him out of sports for the summer and all? But no, he bounced back and he was playing his Gaelic and, and you know, in the summer, and no problem to him."*

4.3.4.2.3 Post-operative Recovery following ABG

The children found the hip more difficult than they anticipated, and felt this affected them more so than their mouth.

C1: *"Em, yeah, espe-, it was kind of a bit awkward to walk because I was kind of like limping on one foot."*

"My hip was the sorest because em, well I just, because I was walking on it"

While they found eating uncomfortable, this did not seem to bother them as much, as they had anticipated it in advance.

C2*: "Like I kind of knew it would be hard to eat, em, like hard food"*

C1: *"It was a bit awkward to eat, so I only, I was trying to eat on one side"*

The restrictive diet was unpleasant for some of the children. One mother describes her son's reaction to the diet post-operatively, while another boy found he was hungry on the soft diet.

P6: *"…need blended all food and him not want it, see, him not understand why it's, him not can eat, you know, normal food after that bone graft, that he need everything blended and not nice."*

C3: *"Yeah, like soup or beans. It was. It was okay, but you'd be starving after a while."*

One of the children mentioned a cream she had to put in her nose and a mouthwash she had to use after the surgery. She didn't like the sensation of the cream in her nose, nor the fact that she did not know she would have to use these beforehand.

C2: *"And just the, the nose cream that I had to use and the mouthwashes, I didn't really know about them. I didn't, I didn't know I had to take them. Like in my nostrils. It was kind of, it felt really weird when it was in there."*

4.3.5 Information Seeking & Communication

An objective of the current study was to ascertain where parents sourced information about their child's surgery and which sources they found most reliable, as well as their communication with other parents and children, who had

prior experience of the surgery. An emerging theme was also the communication with the care team and what advice the parents and children could pass on to others regarding their experience.

4.3.5.1 Information Seeking

Parents were asked to discuss where they had sourced information about the alveolar bone graft surgery and which sources they found reliable, as well as whether they felt they had received sufficient information and for which format they had a preference i.e. written/ verbal. Many participants found the verbal information they received at the clinic the most valuable, both from the Oral and Maxillofacial Surgeon and the Orthodontist.

> ***P1:*** *"I suppose the verbal is nice, you can ask questions back like and stuff, you know?"*

> **P3**: "*So then we were, we were up in eh, eh, the hospital, and we were all there talking to the surgeon, and he told us exactly what he was going to do… like the way he explained it to us, it was absolutely spot-on, you know.."*

> **P4***: "I would say probably the orthodontist would have explained a lot of it, lot of it to us initially because you know, he would explain why the braces were going on or what, what was going to be done"*

Some parents felt they had sufficient information from the verbal advice at the clinics and didn't feel the need to do any further research, for most this was out of fear of what they may read on the Internet. The dangers of 'knowing too much' seemed to be a recurring theme with parents.

P5: *"I would have done some online searching, but I'm sure you know yourself, you know, you can em, you can read so many things that maybe aren't too reliable."*

P4: *"I felt I knew enough of what was going on without having to, to be researching it anymore. Sometimes you can know too much as well, not a good thing either…"*

P1: *"You know, the more you, the more you look at that stuff, like the more you like, you know, it can scare you, like."*

P2*: "a lot of people say that's the worst scenarios, so not to be really always looking at the Internet."*

Whereas another mother reported how she was advised to source online information by the cleft team, specifically being directed towards videos, which she found very helpful.

P6: *"It's the lady, she coming and she say 'you can see some videos as well, if you're looking in the Internet, you can find everything, you know.'"*

"I found, you know, I found videos Temple Street exactly how to do that surgery, bone graft in Temple Street"

As regards written information about the surgeries, not all could remember receiving a written leaflet prior to surgery, but all recollected getting post-operative leaflets. Those who didn't receive the former still seem satisfied that they had received adequate information.

P5: *"Em, I think the only written information was the little leaflet that's actually prepared by the Cleft Lip and Palate Association. It's just like a two-page summary of what happens."*

One of the children described a book that she read before the surgery, which was sourced from the Cleft Lip and Palate Association, through her mother who was actively involved in the association.

C2: *"And then I had a book on it, called Making Faces. Yeah, so I read that before, and then just listened to what the doctors were saying."*

4.3.5.2 Communication with Care Team

The vast majority of participants spoke very highly of their experience with the Cleft Care Team, how they felt very well prepared by them, and that any interaction with them was pleasant. One mother mentioned the relationship that was built up over the years with the team. There was a sense in the interviews of the parents having a lot of trust in the team and the surgeon.

> **P1:** *"When you trust in the surgeons like, you know, you know, you don't, not really* [need to look online]."

> ***P5:*** *"She's eleven now, so, you know, a lot of them have been on the team since Orla was born, so you know, you have built up a relationship with these people, so it's em, you know, so you do, really do, get all the information that you need."*

In relation to the multidisciplinary clinics, some parents found this very reassuring, to have so many specialists in one room during the consultations, who are involved in the decision making process.

> **C1:** *"I was kind of more relaxed when I, I knew I, you know, I could trust them and all."*

> **P6:** *"No, we having appointment, which eh, around maybe five or six specialists, you know, doctors and now everything"*

P5: *"As a team, they're great because, you know, you'd have quite a few people in the room at the time when they're making decisions to go ahead with the bone grafting, there would be a cleft nurse, a cleft coordinator, the orthodontist, the surgeon, actually the plastic surgeon popped in as well, you know, it's very much a team eh environment in there"*

While one of the children and her mother said that on one occasion they found the number of people daunting, as she did not know whom a number of them were.

P5: *"…there was loads of them, and there was obviously quite a busy clinic on, and they sort of came in one by one. I knew who they were, but she didn't, and the way she described it when we came out, was she said 'Mum, people just kept on coming in the door, putting on gloves and putting their fingers in my mouth', and she said 'I didn't know who they were or what they were doing.'"*

One mother described how busy the clinics were and the difficulty for patients travelling a distance on public transport.

P2: *"…and then the appointments could be running late and you have people anxious, they're trying to get buses or trains back and I find that very hard. And I was very disa-disappointed before Christmas, there was this man and he said I have to go and catch a bus, and his, his child, his son didn't get seen that day because they were waiting three or four hours, you know, to be seen. And eh, he just went off, he said I have to catch a train, he was from down the country. And I felt sorry for the wee boy because, this is a little boy that's going to miss out."*

4.3.5.3 Communication with other Parents

Some parents found it helpful and reassuring to speak with other parents of children with clefts. There seemed to be a sense of support from shared experiences, as well as the potential to learn from other parents.

> **P1:** *"It's nice to talk to parents, that, you know, have it, they understand like, they can tell you like what's the next step and what will happen like, you know, as well like."*
>
> *"From my pregnancy I would have talked to a woman, I think she had three kids with cleft lip and palate, like, of her own like, so she was a good help to me like… she came to me the first day I had her, when I, in the hospital, like, you know, helping, showing me how to feed her and stuff like.'*

Another mother had a different experience of offering to talk to another woman who had recently had a baby with cleft lip and palate.

> **P2**: *"There's a girl in the town here and her granny em asked me to talk to her em, and I went to talk to the mother and the mother says 'I'll deal with it myself, I don't want to talk about it'"*

The outings organised by the Cleft Lip and Palate Association were mentioned as a nice way to meet other families.

P2: *"But it's good to meet families, and you seen the same families there all the time and they could be expanding, and there could be two or three kids with the cleft, you know, in the one family."*

One of the mothers involved in the association (CLAPAI) felt that the interaction with other parents was extremely helpful, but felt she wouldn't have that contact, if she was not involved with the organisation.

P5: *"Oh, a-absolutely yeah, no, it is, because it's, it's, it's back to those, the practical things that maybe mightn't be picked up in a hospital, like the, the walking or the diet, you know, just the small day-to-day things that mightn't have been picked up"*

"if I wasn't involved in that [CLAPAI], I probably wouldn't have had any exposure to other parents with children with clefts."

4.3.5.4 Communication with other Children

None of the patients had spoken with other children prior to their surgery about their experience. Some of the patients felt that they would have found it helpful to talk to another child of their age who had had the procedure done before.

C1: *"It probably would have helped, because I didn't really know what was going to happen."*

C2: *"Em, I think it might have helped. But I wasn't too nervous anyway."*

5.3.5.5 Advice for others undergoing treatment

The patients interviewed were asked what advice they would give to other patients undergoing alveolar bone graft surgery. One of the patients had advice in relation to the day of the surgery, while others were more practical for the recovery period.

C1: "*Em. I'd probably say like don't be really nervous, because it's actually not too bad when you get it done, and like if you just relax and em, espe-especially with the gas mask, because it's just going to take forever if you don't breathe slowly"*

C2: "*I would probably say that just beforehand, make sure that like in your house you have like soft foods and comfy chairs. And a few books to read. And, and just that you do get quite swo-swollen and your hip is quite hard to walk on for the first couple of days, and that stairs are really hard."*

The parents were also asked what advice they would pass on to other families undergoing the same treatment journey. Some of the mothers who were involved with the CLAPAI recommended this as a way of meeting other families and sourcing helpful information from other parents. Some of the parents' advice

regarded the mobility issues and difficulty with the hip, such as getting up the stairs or into bed.

P5: *"Yeah, nothing really. Em. No, as I said the, the real surprise for us was that the, the mobility issues when she came out initially."*

"...but just in terms of getting a, a more comfortable chair for her, like we sort of brought her home and put her on the couch and [she said] *'I can't get into it, I can't get out of it.'"*

4.3.6 Coping Mechanisms

The theme of coping, with the condition and the treatment journey, emerged through the data analysis, although participants were not specifically asked about this. Several different mechanisms for coping arose from the data. One Mother talks about how her own mother had been a great source of support for them.

P2: "*Yeah, and she was very positive, you know, she was always praying for him and making sure everything, you know, she'd always worry for him, lighting candles for him and everything."*

The idea of putting the condition in perspective certainly seemed to help some parents cope. While most parents expressed shock and sadness at the diagnosis, on reflection were grateful the diagnosis had not been worse, and were able to put it in perspective.

P2: *There's a bit to it, but yeah, there's a lot worse. Could be a lot worse, there's cancer. I was dealing with that for the last three months, and that's a disaster.*

One mother's attitude towards her daughter's cleft was put in perspective by her son's congenital heart condition, which highlights also the effect of individual's previous life experiences on their outlook.

P4: *"I had another son who went through three heart surgeries so in, in comparison, do you know, it was minor, so I suppose other parents might see it as quite a traumatic… And I don't want to make little of it, but I suppose it- it's all relative to what my experience was"*

Meeting other families affected by clefts and attending outings organised by the Cleft Lip and Palate Association of Ireland also served as a coping mechanism for some families.

P2: *"But it's good to meet families, and you seen the same families there all the time and they could be expanding, and there could be two or three kids with the cleft, you know, in the one family…We were very lucky that the other three* [children] *had not* [been born with a cleft]"

However, other participants did not seem to find this helpful, and hence the individual variation in the different methods of coping became apparent also.

P4: *"I didn't feel that I needed to talk to anyone else about it, you know, that it wouldn't give me any more insight, you know because I wasn't nervous. I suppose if I was nervous and anxious about it, yeah, I probably would have liked* [to talk to other parents]"

The idea of 'getting on with it' appeared as a coping mechanism for some parents, and the concept of taking it in one's stride.

P3: *"We just, and he even took it, took it in his stride and that was the end of it, you know"*

P5: *"You get on with everything else and eh, you know, you just try and get them back out again as soon as you can."*

Recognising and acknowledging the strengths of their child, such as their determined ability, was another coping mechanism that emerged.

P1: *"Not, no, she was grand. She was up, no, she was up and about, no, she was flying it like. She'd go past you too fast sometimes like, you'd need to slow her down."*

"You would be always, you know, worried like, you know, but for her like, and upset, upsetting for her like, but em, no, she's a tough wee cookie so she is."

P2: *"Well you know what, he surprised us all the time. He had a great pain threshold. He made an unbelievable recovery… He was super, absolutely."*

P3: *"He's that type of a child, he just bounces back into, to his usual routine."*

P4: *"Now she would be fierce independent and, and she would, you know, she would have a, she would have a high threshold for pain, like I mean the orthodontist would always say to her that, you know, he's her best patient, because she would, she'd sit up in the chair and no matter what he done, she'd just lie there, you know."*

5.0 Discussion

5.1 Discussion of Methodology

This was a qualitative study, using semi-structured interviews and thematic analysis to understand the experiences of children with cleft lip and palate, and that of their parents, at certain key stages in their treatment journey, namely the alveolar bone grafting surgery and pre-surgical orthodontics.

5.1.1 Topic Guide Development

Throughout the interviewing process new topics, which had not been initially included in the topic guide, emerged. The topic guide was amended after each interview to include these new topics, as is common practice in qualitative research. The idea was to capture new and emergent themes as they arose, so as to include them in subsequent interviews. As the interviews are semi-structured in nature and not intended to be standardized, this modification to the topic guide allowed for flexibility in the discussion and for these areas to be further probed as necessary with subsequent participants, until no further new themes emerged and theoretical saturation had been achieved.

5.1.2 Participant Interviews

The initial protocol for the study involved a mixed methods approach for data collection, including focus groups with parents and face-to-face interviews with parents and children. Focus groups are a commonly used research tool in qualitative research and are defined as carefully planned group discussions that

are 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., 2008a, Smithson, 2000). The group participants interact with each other in a focus group and share their views and experiences about a certain topic and these interactions can result in a greater depth of dialogue and data. However, some participants may find a focus group intimidating and feel under pressure to agree with dominant personalities or socially acceptable views, which can result in generic data. It was decided that because this patient cohort was located throughout the country, organising a time and location for a focus group would not be feasible. Therefore the format for data collection was individual semi-structured interviews.

As this patient cohort had a significant number of appointments and interventions, and spent a considerable amount of time at, and travelling to, appointments, it was felt that telephone interviews would be most convenient for the patients and their parents, and would result in a greater participation in the study. This was also based on the recommendation of an experienced qualitative researcher. As such the study protocol was amended to include individual semi-structured interviews, which would be carried out via telephone calls and audio-recorded.

There are some advantages to carrying out interviews over the phone. It is more convenient and less time consuming for both the participants and the interviewer, as it requires no travelling on either account. It can be easier to schedule at a more convenient time for participants, and is also less expensive, as there are no travel costs to be accounted for in the research budget. Participants were interviewed while in their own homes, as opposed to a clinical environment,

which can put people at their ease. This resulted in a more informal atmosphere for the interviews and may result in the data being more reliable, as participants are more relaxed and comfortable. There was also an increased element of anonymity. In this case, the researcher had not treated or met any of the patients or their parents, so they were more likely to speak freely and disclose sensitive information with less embarrassment than might occur if they were to attend for a face-to-face interview. These advantages of telephone interviews have been discussed in the literature (Vogl, 2013).

The use of the telephone for interviews, however, has previously been discouraged by traditional qualitative researchers, who view it as an inferior method of data collection and not suitable for qualitative research (Gillham, 2005). However, there is no empirical evidence to support this. The absence of visual cues via telephone is thought to result in loss of contextual and nonverbal data and to compromise rapport, probing, and interpretation of responses (Novick, 2008). The face-to-face encounter is often thought to be necessary for the interviewer to build and maintain rapport with participants and thus to enable the gathering of rich in-depth information. During face-to-face interviews, body language and cues from the interviewees can also supplement a researchers' understanding (Gillham, 2005).

Vogl (2013) compared face-to-face interviews with telephone interviews in children and found that there was no significant difference in the duration of the conversation, the number of words spoken, the proportion of words spoken by interviewees, or the need for clarification between the two interviewing modes. The study also concluded that there was no difference in the level of rapport

achieved. The differences in the response to sensitive questions were attributed to the personality of the interviewee (Vogl, 2013).

The interviews were carried out once adequate training had taken place. While the interviewer had carried out practice interviews with colleagues, no practice interviews had been carried out with children. The interviews with the children were more challenging and the data gained was not as rich as perhaps may have been achieved by a researcher with experience interviewing children. A researcher with more experience may have had been able to employ different interviewing methods with the children to gain further information, as it is acknowledged that children are more difficult to interview. This was accepted as a limitation of the study. Other limitations of the interview process were that the parent was present in the room while the child was being interviewed. This was necessary as it was a stipulation from the ethics committee. The parents' presence resulted in some prompting of the child by the parent, and may have had an impact on the child's responses and willingness to speak freely. The interviews with the children had to be carried out on loudspeaker, which sometimes resulted in difficulty being heard, and impacted on the continuity of the conversation.

Although all participants were interviewed within a year of the alveolar bone graft, there were times when they were unable to remember details about the experience. This recall bias was considered when analysing the results. For example, where a parent reported not to have received any information leaflets about surgery, it was acknowledged that there is a possibility that they may have received some leaflets but have simply forgotten. When both parent and child from the one family agreed to be interviewed, it was requested that either the child be interviewed first, or that the child not be present while the parent was being

interviewed. This was to avoid the child overhearing what the parent had said and merely repeating the answers. There was no way of ensuring that this did not happen in some instances, and again, was accepted as a limitation.

5.1.3 Purposive Sampling

The sampling of the population was carried out purposively, based on when the alveolar bone graft had been carried out, to reduce recall bias. However, the numbers of patients treated with alveolar bone grafts in Ireland each year is small (approximately 25 – 30 patients), and so this limited the number of suitable patients. It was intended to include more participants in the study, but the response rate was low (41%), despite an incentive offered for participation. Many of the children declined to be interviewed, either personally by their parents, who felt they would not want to be interviewed. The gender distribution ideally would have had more male participants, but it was the mothers who volunteered to be interviewed in nearly all cases. Following review of the transcripts however, it was deemed by an experienced qualitative researcher, that the data was sufficiently rich and that theoretical saturation had been achieved. As such, no further recruitment of participants was attempted.

5.1.4 Analysis of Data

A thematic analysis approach was planned to manage and analyse the data within MAXQDA© software. 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 account of the data (Braun, 2006). Due to the

familiarity of the researcher with cleft lip and palate literature and the treatment journey, some themes were anticipated, which shaped the research aim and objectives, interview questions, and interpretation of data, as is often the case in qualitative research. Thorough content analysis of the transcribed data also took place in MAXQDA, where segments of text were coded line by line through constant comparison to look for themes with an inductive approach, ultimately resulting in the development of the themes and subthemes. During the coding and analysis of the data and development of the themes, some data was relevant to more than one theme. As such, there was some crossover of content between the themes.

5.2 Discussion of Results

5.2.1 Life Course of the Condition

The subthemes that arose regarding the life course of the condition were as follows;

- disclosure and diagnosis
- experience prior to initial cleft repair
- previous cleft surgeries

This research project aimed to look at particular stages of the treatment journey experienced by patients with unilateral cleft lip and palate, namely alveolar bone grafting and pre-surgical orthodontics. Therefore, the diagnosis of the cleft lip and palate, or previous cleft surgeries, had not been included in the original topic guide for parents, as it was deemed outside the scope of this research project. However, as the interviews were semi-structured in nature, this allowed for the conversation

to diverge from the topic guide to explore other emergent topics. Most of the parents freely reported the diagnosis and the first year of their child's life, including their initial lip and palate repair surgeries.

The discussion of the diagnosis and early years without prompting by the researcher, demonstrated the significant impact the original diagnosis and early surgeries had on the parents, and highlighted the importance of emotional support for parents during this time. Many of the parents had no prior knowledge or experience of cleft lip and palate, and their sense of shock is evident. The concept of fear of the future and of not knowing what was in store for their child with the management of the cleft are apparent, as well as other emotions of guilt, worry, self-blame and upset. Mothers felt guilty and wondered if they were responsible for the cleft, worrying whether they had done something wrong in the course of their pregnancy. Although no fathers were interviewed in this study, this multitude of emotions was in keeping with the literature on the impact of cleft lip and palate on parents and particularly on mothers (Despars et al., 2011). This experience is often referred to as 'emotional strain' on mothers, many of whom seek support and professional counselling during this time. With hindsight, the mothers acknowledged that they had no knowledge of the treatment required for their child. The first few months of life, prior to the initial surgical repair, were described as difficult and stressful for the parents. Feeding difficulties and the constant worry of their child not gaining weight and failing to thrive prevailed.

The life course of the condition with a cleft lip and palate, compared to other chronic conditions however, had an upward trajectory, where patients continually improved with ongoing treatment, and ultimately led a normal life. It was evident from speaking with mothers, how their outlook on the condition

changed; how they gained perspective into the condition and became more optimistic, acknowledged the excellent care, treatment and outcomes their children had received and showed appreciation that their child did not have a more serious illness. Mothers acknowledged the special attributes and strengths of their child, which was in keeping with literature on parental experience of caring for children with clefts (Nelson et al., 2012a).

5.2.2 Perception of Treatment

This theme was discussed in relation to both parents' and patient's understanding of their treatment with regard to both alveolar bone grafting `and pre-surgical orthodontics, as well as the risks of treatment according to the parents. The parents all showed an understanding of their child's treatment, to a greater or lesser extent. In all instances, it was the parent's first experience of a cleft in the family, so this was not a factor in some having a greater understanding than others. The variation in understanding could be due to a number of factors. Firstly there could be an element of recall bias, as the surgery had been carried out several months prior. As such, they may have had a better understanding at the time of surgery but had subsequently forgotten some of the details by the time of the interview. Secondly, the information they received at the clinic may have varied, which could be due to a differences in the information provided to the parents by the clinicians and/or differences in the information seeking habits of the parents. Some parents may have asked more questions at the clinic to gain further insight into the procedure and its risks, as well as carrying out their own research online.

Some parents had access to more information resources, through their involvement with the Cleft Lip and Palate Association of Ireland (CLAPAI). Those who were involved seemed to have a better understanding of the rationale for treatment, as well as the risks involved. This may be to do with increased access to information resources through their involvement with CLAPAI, as well as frequently discussing the treatment journey with other parents. The level of employment status of the parents seemed to have an impact on the extent of their understanding of treatment, with those with professional employment having a more detailed understanding of the procedure and its associated risks. Generally there was little acknowledgement of the risks of the procedure in this study. There is research to show that patients want to be informed of major risks which would have long term complications as well as the consequences of not undergoing treatment (El-Wakeel H, 2006). The only significant factor between the groups questioned in the study by El-Wakeel *et al* was the level of education. While the parents in this study did not recollect risks of surgery, it cannot be assumed that they were not informed of the major risks at the time of the ABG and were just unable to recall them at the time of the interview.

The patient's understanding of their treatment, particularly considering the ages of those interviewed, was relatively comprehensive. The level of understanding the child had of their treatment could be correlated with that of their parents. The extent to which the children were prompted during, or prepared for, the interview by the parent is uncertain. The researcher, while carrying out the interviews, noticed one or two episodes of prompting, but it did not seem to occur to a significant extent. Similar to the parents, some of the children had access to more information resources than others. One of the girls interviewed had a specific

children's book on alveolar bone grafting, as her mother was involved with CLAPAI, so her knowledge of treatment was more extensive.

5.2.3 Experience of Treatment

This theme related only to the experience of alveolar bone graft (ABG) and the pre-surgical orthodontics, and not to previous cleft surgeries. However the parent experience of the ABG appeared to have been influenced by their experience of the initial cleft surgeries, which occurred at a much more stressful time, and when the child was only a few months old, which was understandably more distressing. This primary lip and palate repair was the parents' first experience of surgery and was far more emotive for them. As such, the parents' experience of ABG was moderated to an extent by their prior experiences, as well as by the complexity and number of previous surgeries.

Many of the parents referred to the ABG as 'another routine surgery' and there was an attitude of just wanting to be completed. It was not suggested that the parents were unconcerned about their child's surgery, but it emphasized the number of procedures and surgeries some of these children had undergone and that it had become somewhat routine for their parents. This may have been influenced by the established relationship built up with the cleft care team over the years and trust in the service and the clinicians, which put parents at ease. While most felt that the surgery was routine, one of the mothers felt underprepared leaving the hospital afterwards and commented that no one had discussed post-operative instructions with them. This demonstrated the desire for an individualised care plan, which had been found previously in the cleft literature

(Nelson and Kirk, 2013). It was possible that literacy may be an issue for some parents, although this was not recorded in this study. Approximately one in six adults in Ireland have significant literacy difficulties (Central Statistics Office, 2012), and simply providing a written instruction leaflet is obviously inadequate for these parents.

In relation to the orthodontic treatment prior to the ABG, some parents found this aspect of treatment more challenging, in relation to discomfort, frequency of appointments and difficulty in maintaining adequate oral hygiene. Most of these children were 8 or 9 years old undergoing this orthodontic treatment, so their tolerance and ability to maintain good oral hygiene may be less than that of an adolescent patient. Those whose children had been slightly older undergoing the treatment did not report any untoward experience with the fixed appliances. One of the mothers felt the orthodontic treatment was more difficult for her daughter than the surgery. This may be useful for orthodontists to be aware of the impact this treatment process can have on younger patients, when making treatment decisions about whether pre-surgical orthodontic treatment is beneficial in cases which may be deemed borderline in requiring expansion.

A subtheme that emerged was the satisfaction of parents with their child's treatment. All of the parents expressed satisfaction with the care their child had received and with the care team, which is in keeping with cleft research on parental satisfaction from other countries (Nelson et al., 2012a, Nelson and Kirk, 2013). While satisfaction with treatment was generally high in the literature, the reports were vague, as did not define how satisfaction is assessed. One of the parents expressed disappointment that a supernumerary tooth had been missed during the ABG surgery. There was potential for recall bias in the parent's account

of proceedings. However her attitude, towards what she interpreted as an error, demonstrated a level of understanding towards the surgeon and trust that it will be dealt with in the future.

One of the patient's experience of orthodontic treatment was that it was uncomfortable and annoying, particularly with food impaction, difficulty cleaning and discomfort closing her mouth over the labial fixed appliances. The difficulty for young patients managing both labial and palatal appliances should be considered and orthodontic treatment simplified for these patients where possible, as it is has been debated whether orthodontic treatment prior to grafting influences outcomes (Kindelan and Roberts-Harry, 1999). The children's experience of surgery was very individual, and their memory quite specific to certain aspects of the experience, with one girl focusing on the induction of the general anaesthesia as the most difficult part, while the other two children focused more on the post-operative experience.

5.2.4 Impact of Treatment

The treatment of cleft lip and palate is evidently a long road for both the affected children and their parents. There are psychological implications for parents as well as the patients, as well as logistical issues such as time off work and school, attending multiple appointments, caring for other children and the difficulties associated with the immediate post-operative recovery following surgery. The psychological impact of the treatment on the mothers began with the initial diagnosis of the condition, with the feeling of guilt, as well as a desire to achieve normality for their child. This seemed to serve as a motivating factor for

them throughout their child's multiple treatments and surgeries, as well as serving to justify the interventions, as ultimately they had a strong desire for their child not to feel different from their peers or be bullied. This was a recurrent theme in the qualitative literature on the experience of parents. Klein et al (2006) reported that parents worried about social issues, including concerns about a child's acceptance by their peers, experiences of teasing, and finding employment. Children experiencing bullying at school, or when entering new and unfamiliar settings, was particularly distressing to parents (Klein et al., 2006).

A concept found in previous literature of cleft patients and their parents/guardians was that parents' happiness was correlated with that of their children. The results of this study would support these findings. Sischi *et al* (2017) found that parents' well-being was influenced by their children's health and reactions to their children's cleft and cleft treatment (Sischo et al., 2017). The concept that the treatments intended to make cleft patients feel normal paradoxically exacerbating their sense of defectiveness was discussed in the literature (Alansari et al., 2014). The patients interviewed in this study, however, did not suggest any untoward psychological implications from their cleft or the treatment journey at this stage, but as these patients were only nine years old, they may have been too young to experience or express these emotions. In the study by Alansari *et al* (2014), the patients were interviewed as adults.

One of the patients interviewed in this study mentioned being asked at school what had happened to her following her surgery, but she seemed to be unconcerned by this attention. This would support the findings of previous research, which suggests that children are less emotionally and socially affected by their cleft than their parents estimate (Noar, 1991) and that they have equivalent

levels of self-concept and self-esteem as their peers (Klassen et al., 2012). Only a small cohort of children aged between 10 and 11 were interviewed however, so this may not be applicable to other age groups of children. The patients reported feelings of anxiety and nervousness prior to the surgery, but could acknowledge that it was not as difficult as they had anticipated. Certainly for the children, the relationship with the care team, seeing familiar faces, and the feeling of being able to trust them, helped to reduce their anxiety.

Logistical issues that parents discussed in relation to their child's treatment were the frequency of appointments for both surgical visits and orthodontics, travelling to and from Dublin for those who resided outside Dublin, as well as sleeping on hospital floors when their child was undergoing surgery. These logistical difficulties were not specific to cleft patients and are unfortunately similar to those experienced by all parents of children living with a chronic condition (Canam, 1993). The post-operative recovery period was challenging for both the parents and the patients, with some of the patients more incapacitated than was anticipated by themselves or their parents. The major concerns post treatment related to the hip and the impact it had on their mobility, as well as inability to be involved in sports for several weeks. The reports of remarkable discomfort and mobility issues from the donor site were a significant finding. While there are several factors clinicians must consider for the best surgical management of their patients, this finding of significant donor site morbidity may be factored into future decisions to consider synthetic grafting materials or bone substitutes, should their use result in comparative success and reduced donor site morbidity for patients in the future and with further clinical research on their use. Dickinson *et al* (2008) compared traditional iliac grafting to that with a bone

substitute in patients with cleft lip and palate and concluded that the bone substitute was more successful than conventional autologous grafting and resulted in reduced post-operative pain (Dickinson et al., 2008). The Cochrane review in 2011 concluded that the study by Dickinson *et al* was at high risk of bias and that there was insufficient evidence to advise if one intervention is superior to the other (Guo, 2011). However there certainly seems to be several bone-graft substitutes that demonstrate promise in alveolar cleft reconstruction (Liang et al., 2018).

5.2.5 Information seeking & Communication

The subthemes that arose under this heading were the information seeking habits of parents, the communication with the care team and the communication with other parents and children, as well as advice participants would offer others undergoing treatment. In relation to information seeking advice, previous studies suggested that most parents preferred to receive their information verbally from specialist practitioners (Byrnes et al., 2003). Other studies were in agreement that parents showed a preference for verbal information but want it to be supplemented with written leaflets (Knapke et al., 2010). This study is in agreement with these findings, that parents valued the verbal information they received at the clinic. With the increasing use of online resources, parents were asked whether they felt the need to supplement their knowledge with online research, and an interesting finding was that the majority tended to avoid online research, for fear of unreliable sources and what they may read. The exception to this was when they were directed towards specific resources by members of the

cleft team, as they then deemed these to be reliable resources. In relation to written leaflets prior to surgery, there did not appear to be any consistency in leaflets provided to patients at the MDT, however these findings must be interpreted considering the risk of recall bias.

Most parents and their children reported positive interactions with, and trust in, the cleft care team, as well as the building of rapport over the years with the various team members. In relation to multidisciplinary team clinics (MDT), the findings were in keeping with existing literature, with mixed experiences and emotions in relation to their experiences of these clinics. The results of this study were that while it was reassuring for parents to have so many experienced professionals in the room, for the children it can be overwhelming and disconcerting to have so many strangers putting their hands in their mouths without introducing themselves. This was in keeping with the findings of Myhre *et al* (2019), who carried out interviews specifically on the experiences of adult patients and parents of patients at craniofacial MDTs. They found that many participants felt a sense of gratitude toward the MDT and recognised the competence of the team, which made them feel safe. Conversely, other participants found the large team daunting and felt like an 'object on display' (Myhre et al., 2019). Pidgeon *et al* (2017) proposed a possible solution to the daunting experience of MDT clinics for cleft patients (Pidgeon et al., 2017). The study was based on a sample of craniofacial patients and found that providing parents with a leaflet containing specific information about the craniofacial MDT appointment led to lower levels of anxiety attending consultations. The importance of introducing all team members at the MDT to the patient, as well as to the parent, is highlighted also.

In relation to communication with other parents and children about their experience prior to their surgery, two of the mothers interviewed had spoken to other parents and this was through CLAPAI. They spoke positively about their experiences and recommended it for other parents of children with clefts. The other advice offered was in relation to expectation and preparation for the mobility issues associated with the hip. None of the children interviewed had spoken to other children prior to their surgery but felt it might have been helpful.

5.2.6 Coping Mechanisms

Coping with the condition was a theme that emerged through data analysis, and indeed is a common theme in qualitative literature on chronic disease (Maes, 1996). Several methods of coping arose, such as the support of immediate family, recognising other strengths and the uniqueness of the child, talking to other families affected by cleft, as well as putting the condition in perspective and the idea of 'getting on with it'. In relation to recognising the child's strength as a means of coping, the results of this study found that each of the parents, without prompting, spoke proudly of the strength of their child, their good nature or how special they were. Klein *et al* (2006) also found that parents reported recognising their child's strengths, such as a determined attitude, perseverance and sociability (Klein et al., 2006).

The ability to put the condition in perspective emerged as another method of coping. This may have been due to the upward trajectory of the condition, in that cleft patients continue to improve over time, which is dissimilar to that of other chronic conditions, where patients generally deteriorate over time. The

excellent outcomes for cleft patients in recent years contributed to the feeling of hope for the future, which parents highlight as a rewarding aspect of caring for a child with a cleft (Klein et al., 2006). This study found that parents were able to appreciate that their child was not suffering a more serious illness, and this ability to put the condition in perspective, served as a coping mechanism for some. One mother who had experience of another child with a more serious condition, seemed to cope better with the cleft, and acknowledged that everyone's perspective and ability to cope with their child's condition was relative to their own prior experiences. The coping strategy of the child in their ability to 'take it in their stride' was acknowledged by some of the parents, while for other parents, the support and interaction with other families affected by clefts served as a coping mechanism.

6.0 Conclusions

- Parents were better able to cope with their child's treatments and surgeries at the alveolar bone grafting stage of treatment compared to the earlier surgical interventions.
- Patients find the experience of both ABG and pre-surgical orthodontics challenging. Trust in and familiarity with the clinicians and care team were important factors in reducing their anxiety prior to surgery.
- Parents find the MDT clinics useful and appreciate the number of experienced clinicians and team members present. Parents found the verbal information from the team at these clinics to be the most valuable information resource.
- Patients find the MDT clinics daunting due to the number of people present in the room, particularly when they are unfamiliar with them. Therefore the importance of all team members at the MDT introducing themselves to the patient as well as the parent was highlighted.
- Parents and patients found they were unprepared for the difficulties associated with the donor site post ABG and express a desire to be better informed about this aspect of the post-surgical recovery.
- Acknowledging other strengths of the child, putting the condition in perspective and the support of family and the cleft community emerged as means of coping with the condition.

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8.0 Appendices

Appendix 1: Ethical Approval

THIS NOTEPAPER MUST NOT BE USED FOR PRESCRIPTIONS OR INVOICING PURPOSES

THE ADELAIDE & MEATH HOSPITAL, DUBLIN
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24th November 2017

Re: Patient and parental perception of alveolar bone graft surgery in children with cleft lip and palate: A qualitative study

REC Reference: 2017- 11 - Chairman's Action (11)
(Please quote reference on all correspondence)

Dear Dr. O'Brien,

The REC is in receipt of your recent application to SJH/AMNCH Research Ethics Committee in which you queried ethical approval for the above named study.

The Chairman, Dr. Peter Lavin, on behalf of the Research Ethics Committee, has reviewed your correspondence and granted ethical approval for this study.

Yours sincerely,

Claire Hartin
Secretary
SJH/AMNCH Research Ethics Committee

The SJH/AMNCH Joint Research and Ethics Committee operates in compliance with and is constituted in accordance with the European Communities (Clinical Trials on Medicinal Products for Human Use) Regulations 2004 & ICH GCP guidelines.

Appendix 2: Participant Invitation Letter

TRINITY COLLEGE DUBLIN

OSPIDÉAL DÉADACH BHAILE ÁTHA CLIATH
DUBLIN DENTAL HOSPITAL

Dear Parent/ Guardian,

Thank you for taking the time to read this.

You are being invited to partake in a research study. The study is about the experience of children with a cleft lip and palate and their parents/guardians, who have recently had alveolar bone grafting surgery.

This study is being carried out by the orthodontic department in the Dublin Dental University Hospital and Trinity College Dublin. All information will be kept confidential and only the researcher (Sinead O'Brien) will have access to the information collected. Your name or your child's name will not appear anywhere. If you are willing to participate we will arrange a time that suits you for a very informal interview over the phone, with you, your child or both of you.

To thank you for your time we will send you a One4all gift voucher, to show our appreciation for your participation.

Appendix 3: Participant Information Leaflet

PARTICIPANT INFORMATION LEAFLET

Patient and parental perception of alveolar bone graft surgery in children with cleft lip and palate: A qualitative study.

HOSPITAL: Dublin Dental University Hospital

DEPARTMENT: Public and Child Dental Health

PRINCIPAL INVESTIGATOR: Dr Sinead Emily O'Brien

You are being invited to participate in a research study. Thank you for taking the time to read this.

WHAT IS THE PURPOSE OF THE STUDY?

The aim of this study is to gain an insight in to the experience of children (and their parents/guardians) with cleft lip and palate undergoing alveolar bone graft surgery.

WHY HAVE I BEEN CHOSEN TO PARTAKE IN THIS STUDY?

You have been chosen to partake in this study because your child has had an alveolar bone graft surgery in the last 12 months.

WHAT WILL HAPPEN IF I VOLUNTEER TO PARTICIPATE?

You and your child will be asked to partake in an individual interview over the phone or in person about your experience of the alveolar bone graft surgery. There will be an audio recording made of the interview and only the principal investigator will listen to and have access to this recording and it will be stored securely.

HOW LONG WILL IT TAKE?

There is no set time limit on the interview, usually approximately 20 minutes.

ARE THERE ANY RISKS INVOLVED IN PARTICIPATING?

There are no anticipated risks involved in partaking in this study. However, if any sensitive information is brought up during the study and should the participant feel inclined, access will be made available to a counsellor.

ARE THERE ANY BENEFITS INVOLVED IN PARTICIPATING?

All participants will be given a €25 euro voucher as a token of appreciation. The study will also benefit both clinicians as well as other children with cleft lip and palate and their parents/guardians by increasing knowledge about patient experience of the surgery and potentially improving patient access to relevant information.

WHAT HAPPENS IF I DO NOT AGREE TO PARTICIPATE?

There is no consequence to not taking part in this study.

WILL MY PARTICIPATION OR WITHDRAWEL HAVE ANY IMPACT ON MY ROUTINE CARE?

No, not partaking will have no impact on your routine care.

WILL MY PARTICIPATION BE CONFIDENTIAL?

Yes, all information will be kept confidential and any data used will be anonymised. This means that neither your name nor your child's name will be used in the writing up of this research.

WHO IS ORGANISING AND FUNDING THIS RESEARCH?

The Dublin Dental University Hospital/ Trinity College Dublin

HAS THIS STUDY REVIEWED BY AN ETHICS COMMITTEE?

Yes. Ethical approval was granted for this study by the Joint Research and Ethics Committee in St James and Tallaght Hospitals.

WHAT HAPPENS NEXT?

You will be contacted by phone by the lead investigator after receiving this information leaflet to see if you are willing to participate and if you have any questions. If you are happy to participate you can sign and return the consent form in the pre-paid envelope provided and then a time can be arranged that suits you to be interviewed over the phone.

CONTACT DETAILS

Appendix 4: Participant Consent Form

TRINITY COLLEGE DUBLIN

PATIENT CONSENT FORM

Patient and parental perception of alveolar bone graft surgery in children with cleft lip and palate: A qualitative study.

INVESTIGATOR: Dr Sinead Emily O'Brien

PLEASE TICK YOUR RESPONSE IN THE APPROPRIATE BOX

- I have read and understood the Participant Information YES ☐ NO ☐
- I have had the opportunity to ask questions and discuss the study YES ☐ NO ☐
- I have received satisfactory answers to all my questions YES ☐ NO ☐
- I understand that I am free to withdraw from the study at any time without giving a reason and without this affecting my future medical care YES ☐ NO ☐

I.	I agree to take part in the study	YES ☐ NO ☐
II.	I agree for my child to take part in the study	YES ☐ NO ☐

Child's Name: _____

Parent/Guardian's Signature: _____

Date: _____

Parent/Guardian's Name in print: _____

Investigator's Signature: _____

Date: _____

Investigator's Name in print: _____

Appendix 5: Topic Guide (Parent/Guardian)

Topic Guide: Parent/Guardian

1. **Introduction**
    a. Name, position
    b. Reason for the interview
    c. Aware it will be kept anonymous
    d. Aware it is being recorded
    e. Aware you can stop at any time

2. **Participant Background Information**
    a. Name
    b. Relationship to the child, biological?
    c. Affected by cleft themselves
        i. Type of cleft
        ii. Did they have ABG?
    d. Partner affected by cleft
        i. Type of cleft
        ii. Did they have ABG?
    e. Number of children
    f. Other children affected by cleft
        i. Type of cleft
        ii. Did they have ABG?
    g. Occupation
    h. Partner's occupation

3. **Child Background Information**
    a. Name
    b. Age
    c. Type of cleft
    d. When was ABG carried out
    e. What hospital
    f. Did they have a brace on the roof of mouth beforehand

4. **Pre-operative information and communication**
    a. Can you explain to me your understanding of what an alveolar bone graft is and why it is done
        i. Facilitate canine eruption
        ii. Support base of nose
        iii. Fill in bony defect
        iv. Close fistula
    b. Tell me about the information you received prior to surgery
    c. Did you feel it was sufficient
    d. Did you carry out any further research on the surgery yourself
    e. What information resources were most valuable
    f. Did you receive:
        i. Written leaflet
        ii. Verbal information at clinic
        iii. Website – recommended by clinic or self resourced
    g. Did you meet with or talk to any other parents who had similar experience previously
    h. Did you feel involved in decision making process
    i. Did orthodontist explain the procedure or surgeon or both
    j. Aware:
        i. Why xrays needed
        ii. Risks associated
            1. Failure
            2. Further surgery
        iii. That second smaller surgery to expose canine may be necessary

iv. Future orthodontic treatment necessary

5. Experience of surgery

a. Tell me about the experience of the surgery
b. What are the things that stand out in your memory about the experience
c. Did you have any issues
d. Is there anything you would like to have known in retrospect that may have helped/made the experience easier
e. Is there anything you would tell other parents/ advice you would give
f. Effect on school/work with time off
g. Any complications from surgery
h. Would you do it again

6. Experience of orthodontic treatment

a. Always same operator or different people
b. Issues with braces
    i. Pain
    ii. Breakages
c. Understand what each type of brace is for
d. Understand connection between the braces and this surgery

7. Conclusion

a. Anything else to add?
b. Questions for me?
c. Thank you for your time

Appendix 6: Topic Guide (Child)

Topic Guide: Child

8. **Introduction**
    a. Name, position
    b. Reason for the interview
    c. Aware it will be kept anonymous
    d. Aware it is being recorded
    e. Aware you can stop at any time
    f. Its ok if you forget or are not sure about something we can move on to something else

9. **Child Background Information**
    a. Name
    b. Age
    c. Class in school
    d. How many brothers and sisters
    e. Any brothers or sisters have a cleft
    f. Hobbies

10. **Experience – Braces**
    a. Currently wearing brace?
    b. In the roof of the mouth or train tracks/ both
    c. Any issues with braces/breakages
    d. Other friends with braces in school

11. **Experience - Surgery**
    a. Do you remember having the surgery in your mouth recently?
    b. Do you know why you had the surgery done?
    c. Can you tell me about it?
    d. What happened before the surgery
        i. Nervous?
    e. What happened after the surgery in the hospital
    f. Did you stay long in the hospital
    g. What happened when you went home
    h. When did you go back to school
    i. Was it sore? (Mouth/Hip)
    j. What happens next (braces etc) do you know what the braces are for
    k. Did you talk to any other children who had the same thing done?
        i. If yes -> did it make you feel better/ did it help?
        ii. If no -> would you have liked to?
    l. What would you say to other children getting the surgery

12. **Conclusion**
    a. Is there anything else you remember or would like to say about it?
    b. Any questions for me?
    c. Thank you

Appendix 7: SRQR Checklist

No.	Topic	Item
	Title and abstract	
S1	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
S2	Abstract	Summary of key elements of the study using the abstract format of the intended publication; typically includes background, purpose, methods, results, and conclusions
	Introduction	
S3	Problem formulation	Description and significance of the problem/phenomenon studied; review of relevant theory and empirical work; problem statement
S4	Purpose or research question	Purpose of the study and specific objectives or questions
	Methods	
S5	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[b]
S6	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
S7	Context	Setting/site and salient contextual factors; rationale[b]
S8	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[b]
S9	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
S10	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[b]
S11	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
S12	Units of study	Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)
S13	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/deidentification of excerpts
S14	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 [b]
S15	Techniques to enhance trustworthiness	Techniques to enhance trustworthiness and credibility of data analysis (e.g., member checking, audit trail, triangulation); rationale[b]
	Results/findings	
S16	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
S17	Links to empirical data	Evidence (e.g., quotes, field notes, text excerpts, photographs) to substantiate analytic findings
	Discussion	
S18	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/generalizability; identification of unique contribution(s) to scholarship in a discipline or field
S19	Limitations	Trustworthiness and limitations of findings
	Other	
S20	Conflicts of interest	Potential sources of influence or perceived influence on study conduct and conclusions; how these were managed
S21	Funding	Sources of funding and other support; role of funders in data collection, interpretation, and reporting

[a]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.

[b]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.

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