

The orthodontic management of patients with cleft lip and palate: from birth to the late mixed dentition

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Key points

To understand the orthodontic management of patients with cleft lip and palate.

To understand the importance of the multidisciplinary team within decision-making.

Dental health is imperative for good orthodontic outcomes.

Abstract

This is the first of two papers outlining the orthodontic management of patients with cleft lip and palate. This paper will review orthodontic input into children with cleft lip and palate from birth to the late mixed dentition before definitive orthodontics. It will emphasise the importance of timing in alveolar bone grafting, the role of the general dental practitioner and the impact of timing on definitive orthodontic outcome.

Introduction

The orthodontist is recognised as one of the key members of the team as part of the multidisciplinary approach to cleft care.¹ They are often involved in several episodes of orthodontic intervention within a child's life and as with all specialities, it is incumbent upon them to evaluate outcomes of their interventions continually, longitudinally and those of other members of the team.

The purpose of this article is to review the orthodontic interventions required and the interactions with other members of the cleft team and the general dental practitioner. As previously mentioned, children with cleft palate and those with a cleft lip are completely different entities. Children with cleft palate have a higher incidence of dental anomalies than the general population² and often present with crowding in both upper and lower arches. For the purposes of this and the second orthodontic article, we will focus on children with clefting involving the lip, alveolus and palate, as these children present with greatest variability of dental anomalies and most significant malocclusion and therefore require the greatest orthodontic input and challenges.³

Multiple courses of orthodontic intervention place a significant burden of care on patients and families. Semb *et al.*⁴ in their longitudinal cohort study looking at the duration of treatment for children with cleft lip and palate born between 1976 and 1979 found the overall length of orthodontic treatment ranged from 3–8.5 years with 49–94 visits for the centres studied. This has led more recently to progressively simplified orthodontic protocols within the UK, although a recent retrospective study from one centre found a mean duration of 3.4 years and on average, 44 appointments for all cleft types, with the longest duration for children with bilateral cleft lip and palate. Clearly, this represents a significantly longer duration and visit expectation compared to a non-cleft population, where 18 visits and 1.6 years' duration would be considered the norm.⁵ Reducing the orthodontic burden of care for these children and young adults will be a common theme throughout the article.

The orthodontic intervention will be divided into two papers based on chronology. Paper one will consider interventions from birth to the late mixed dentition, whereas paper two will concentrate on the adult dentition and the adult returner patient.

For the purposes of this article, we will concentrate on two areas:

- Pre-surgical orthopaedics
- Alveolar bone grafting.

Pre-surgical orthopaedics

Although when present, a cleft through the alveolus appears to produce an apparent defect when measuring the alveolar volume, a deficiency hardly exists when compared to non-cleft children.⁶ The apparent defect is caused by the displacement of the alveolar shelves (Fig. 1). Pre-surgical orthopaedics plates frequently used in conjunction with lip strapping/taping aim to



Fig. 1 Child with unilateral cleft lip and palate, showing displacement and an apparent defect of the alveolar shelves

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normalise soft tissue and alveolar tissues. Lip taping or strapping produces tension across the cleft site to approximate the soft tissues and to some extent, retract the anterior alveolar segment which may be flared labially (Fig. 2).

Traditional pre-surgical or pre-operative orthopaedic appliances have been advocated for many years⁷ but have fallen out of favour in most teams in the UK. There are, however, teams around the world where they are utilised.^{8,9} The appliances are acrylic plates that are used to obturate the cleft and passively or actively encourage closer approximation of the alveolar shelves before lip and palate closure. The plates are held in place by taping to the child's face and require frequent visits for adjustment and grinding (Fig. 3).

Perceived benefits of these plates include facilitation of feeding, guidance of maxillary segment growth and development, normalisation of tongue function, facilitation of surgery, better speech, and positive psychological benefit for the patients.⁶ The Dutchcleft Study, a randomised clinical trial, comparing passive plates with controls in children with unilateral cleft lip and palate, did not support these benefits up to the age of six and was shown to be not cost-effective.^{10,11} Later follow-up to the children at 12 years further supported the lack of benefit for transverse dimension.¹² A similar conclusion for dental arch relationships has been found when utilised in children with bilateral cleft lip and palate. Bartzela *et al.*¹³ found in their retrospective longitudinal inter-centre outcome study that any small benefit obtained by infant orthopaedics in the short-term was not maintained at age nine.

In the early 1990s, Greyson and his colleagues modified the infant orthopaedic plate combining the traditional plate with nasal stents: the procedure they now term nasal alveolar moulding (NAM). The aim of NAM was to not only provide the 'benefit' of the plate but also manipulate the nasal cartilages to improve their anatomical form and aesthetics post lip closure.¹⁴

NAM comes with a significant burden of care than even compared to traditional infant orthopaedics when evaluating the number of visits required for fabrication and days worn.¹⁵ Systematic reviews and meta-analysis suggest that it may offer some aesthetic and symmetry advantages. However, no obvious dental arch development and midface growth benefits children with unilateral cleft lip and palate in the short-term.^{16,17} Studies evaluating



Fig. 2 Child with strapping in place to apply tension across the cleft site and reduce the cleft width



Fig. 3 Clinical image of a child with strapping and an orthopaedic appliance in place with metal whiskers to attach tapes for retention. Image courtesy of Professor Grant McIntyre

NAM in the longer-term show limited impact over untreated controls in internal nasal form, including septal deviation.¹⁸ All reports however cite a lack of quality of evidence in their conclusions.

Alveolar bone grafting

Prior to the advent of alveolar bone grafting, children with a cleft involving the dental alveolus often had residual oronasal fistulae, nasal reflux, food impaction and chronic periodontal disease within the region which could ultimately lead to tooth loss. Because of the associated bony defect, orthodontics to align displaced teeth within the regions was limited and prosthetic options to replace the teeth that were absent were significantly hindered.

In the early 1970s, Boyne and Sands published reports on a new technique to repair an alveolar cleft utilising cancellous bone.^{19,20} They suggested the procedure

should be undertaken in the mixed dentition before canine eruption (approximate age: 9–10 years). Modern optimisation of this treatment allows dental eruption within the region producing a continuous dental arch and has transformed the dental management and rehabilitation of children with cleft lip and palate.²¹

Timing

Timing of bone graft procedures can be broadly classified into two ranges: primary (<2 years of age) and secondary (5–11 years). Primary grafting has largely been discounted as a suitable timing in most centres around the world due to its impact of mid-facial growth.

Within the secondary grafting range, timing options advocated are 5–8 years before incisor eruption^{22,23} and 9–11 years before cleft-associated canine eruption.^{21,24,25}

Irrespective of which age range for secondary grafting, the principle is to graft to the alveolus allowing permanent tooth eruption into the

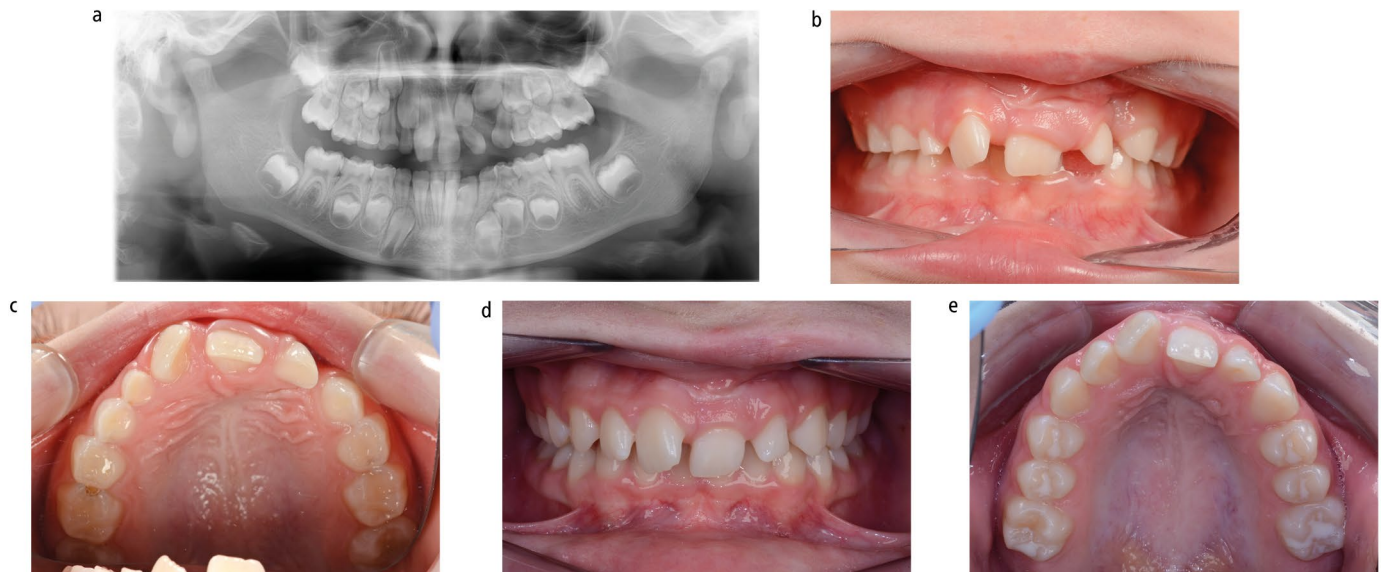


Fig. 4 a) Orthopantomogram radiograph of a child with a unilateral cleft lip showing a well-formed lateral incisor on the mesial side of the cleft, offering the possibility of early bone graft. b) Intra-oral upper occlusal image of the child showing a defect in the alveolus and a deciduous lateral incisor erupting on the mesial side of the defect. c) Intra-oral frontal image of the child showing a defect in the alveolus and a deciduous lateral incisor erupting on the mesial side of the defect. d) Intra-oral upper occlusal image of the child of post bone-graft in the adult dentition showing eruption through the graft of the upper left lateral incisor. e) Intra-oral frontal image of the child of post bone-graft in the adult dentition showing eruption through the graft of the upper left lateral incisor

graft, which is critical for tooth periodontal health and graft survival.^{26,27,28}

In the UK, the grafts are mostly carried out between the ages of 9–11 years. Proponents of grafting slightly earlier, at age 6–8 years, suggest better periodontal outcome for the central incisor adjacent to the cleft,²² without compromising facial growth²⁹ or the quality of bone radiographically when compared to those grafted in the later mixed dentition.³⁰ However, this is weighed against the maturity of the child and their ability to cope with orthodontic intervention when necessary. A recent systematic review, however, concluded it could be acceptable to graft slightly earlier, but the evidence was limited to change from the conventional gold standard of 9–11 years.³¹ Grafting at 9–11 years aims to provide adequate bone for eruption of the canine tooth associated with the cleft. When a suitable lateral incisor tooth, usually on the mesial side of the cleft, is present, a slightly earlier graft prior eruption is considered.²¹ This timing appears to minimally impact facial growth^{32,33,34} and radiographic success rates within the UK and centres around the world are excellent.^{30,35,36,37,38}

Assessment

In order to facilitate pre-surgical dental/orthodontic intervention, bone graft assessment usually takes place between 7–8 years. This timing allows early enough



Fig. 5 An orthopantomogram radiograph of a child with a unilateral cleft lip and palate showing the cleft canine associated with the cleft defect with root 50% root formation

evaluation should a viable lateral incisor, usually on the mesial to cleft, be present. If present, slightly earlier grafting is undertaken to encourage its eruption through the graft (Fig. 4). However, in most cases, timing is determined by the cleft-associated canine. This timing is critical to the outcome of the graft procedure, with a high success rate for those performed before canine eruption.^{24,32,39,40,41,42,43,44}

An orthopantomogram radiograph is usually taken for assessment and the teeth associated with the cleft site are evaluated for morphology, root development and proximity of the crown to the cleft region. Root formation of approximately $\frac{1}{4}$ to $\frac{1}{2}$ ⁴⁵ and

close approximation to the cleft and imminent eruption²⁶ are considered optimal for tooth eruption through the graft (Fig. 5). The advent of cone beam computed tomography (CT) allows a much greater visualisation of the cleft area. In practical terms, however, it offers little benefit to the surgeon, except where multiple teeth in the cleft site require locating prior removal, or where the extent of a partial cleft needs assessment and the two-dimensional plain radiograph will not provide this. Where a partial cleft exists, clinical signs can often give clues as to the extent of the cleft. Distal inclination and rotation of the incisor associated with the cleft, a notch on the palatal aspect and lack

of support for the alar base on the cleft side are often tell-tale signs of a significant cleft existing (Fig. 6).

Pre-graft preparation

Dental health and tooth removal

A recent dental review should be completed before a bone graft assessment appointment. Oral health is considered critical for bone graft success and children should have no active dental disease before grafting.⁴⁶ Poor oral hygiene is an important factor and graft infection leads to a greater incidence of graft failure.⁴¹ Issues should be dealt with pre-operatively through liaison with the local general dental practitioner and cleft dental team, including the specialist paediatric dentist where necessary.

Cleft involving the alveolus are associated with several dental anomalies, particularly hypoplasia of the adjacent central incisor (Fig. 7), which is in turn associated with dental caries and increased gingivitis.^{47,48} This again should be dealt with before graft placement.⁴⁹

Deciduous teeth associated with the cleft site are, in many teams' protocols, removed pre-operatively to increase the amount of attached mucosa for flap construction during the procedure. This may also aid access to the cleft site, with or without orthodontic intervention at the time of grafting. Although these teeth can be removed at the time of the grafting procedure, there is a suggestion that this may lead to poorer bone graft radiological result.⁴¹ As well as deciduous tooth removal, the procedure may be combined under a short anaesthetic with supernumerary removal on the distal side of the cleft, which have been shown when present to be related to increased canine impaction.⁵⁰ Where teeth are to be removed, particularly those closely associated with the cleft canine, this should be carried out approximately 3–6 months before the graft to encourage time for healing but not so distant as to encourage early canine eruption.

Pre-graft orthodontics

Although several authors suggest the indications of orthodontics pre-graft is to correct posterior crossbites,^{51,52,53} this is only appropriate where the skeletal pattern allows and requires long-term retention to maintain it increasing the burden of care for the patient.⁵¹

Rather than crossbite correction *per se* being the main indicator for orthodontic intervention, the primary reason should

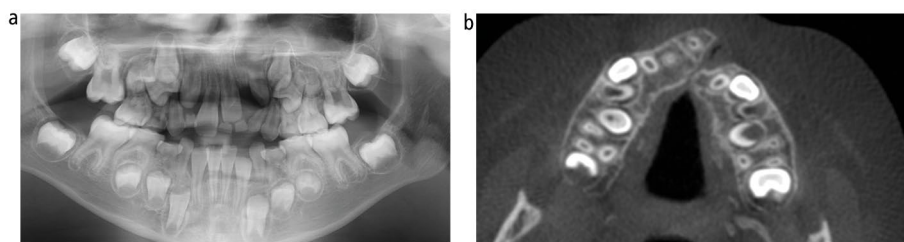


Fig. 6 a) An orthopantomogram radiograph of a child with a cleft lip, suggesting the possibility of a defect in the upper left anterior region. b) A cone beam computed tomography section confirming the diagnosis of a defect in the upper left maxillary alveolus



Fig. 7 An intra-oral maxillary occlusal image of a child with a cleft through the alveolus showing a distally inclined rotated, hypoplastic upper right central incisor adjacent to the cleft

be to aid surgical access to the cleft site. Although some have suggested orthodontic intervention leads to better outcomes,^{54,55} it is not always essential to aid surgical access, particularly where deciduous teeth associated with the cleft have been removed. Therefore, where unnecessary to aid access, orthodontic intervention should be balanced against benefit for the patient.

Where orthodontics is necessary, many protocols involve an appliance to increase the maxillary arch in a transverse direction, for example a tri- or quad-helix. This has the impact where the segments are adjusted of increasing the size of the cleft pre-surgically which has been shown to negatively impact outcomes.^{30,56}

Liao and Huang⁵⁴ suggest orthodontic input would be better focused on incisors impinging on the cleft site or expansion on a more anterior-posterior direction by proclining retroclined incisors (Fig. 8). The type of appliances used to produce this movement can be removable appliances, which can be removed post-operatively; sectional fixed appliances also don't impinge on the bone graft recipient site at the time of surgery.

Although not always necessary for a unilateral cleft, there is less debate over whether orthodontics is necessary for a bilateral cleft. This is because, not only do the tooth positions impinge on good access to the cleft site for the surgeons, but the premaxilla carrying the central incisor teeth is often mobile, requiring stabilisation in the post-operative period. From an orthodontic perspective, the premaxilla itself can be frequently displaced, creating a significant centreline shift, which can be extremely challenging to correct post bone graft if it is not corrected pre-graft (Fig. 9).

When the premaxilla is mobile and displaced it can be stabilised by utilising fixed appliances. Care must be taken particularly on the anterior teeth when placing the appliances, as little bone often exists on the distal side of the central incisors, meaning any adverse distal movement of the roots of the incisor teeth can result in fenestration, resorption, and in some cases, loss of vitality. To reduce this occurrence, the brackets on the incisor teeth are placed in such a way to prevent this. Alternatively, some have advocated other anterior appliances that avoid tooth movement all together and simply



Fig. 8 a) Intra-oral maxillary occlusal image of a child with a unilateral cleft lip and palate at the time of bone graft assessment. b) Intra-oral right lateral image of a child with a unilateral cleft lip and palate at the time of bone graft assessment showing both central incisors in crossbite and a unilateral crossbite. c) Intra-oral frontal image of a child with a unilateral cleft lip and palate at the time of bone graft assessment showing both central incisors in crossbite and a unilateral crossbite. d) Intra-oral right lateral image of a child with a unilateral cleft lip and palate after pre bone graft orthodontic input to procline the upper central incisors and extraction of the upper right deciduous canine. e) Intra-oral right lateral image of a child with a unilateral cleft lip and palate after pre bone graft orthodontic input to procline the upper central incisors and extraction of the upper right deciduous canine. f) Intra-oral frontal image of a child with a unilateral cleft lip and palate after pre bone graft orthodontic input to procline the upper central incisors and extraction of the upper right deciduous canine. g) Intra-oral maxillary occlusal image of a child with a unilateral cleft lip and palate post bone graft with eruption of the upper right canine through the graft. h) Intra-oral right lateral image of a child with a unilateral cleft lip and palate post bone graft with eruption of the upper right canine through the graft. i) Intra-oral frontal image of a child with a unilateral cleft lip and palate post bone graft with eruption of the upper right canine through the graft. j) Intra-oral maxillary occlusal image of a child with a unilateral cleft lip and palate orthodontic treatment with space closure and correction of the unilateral crossbite. k) Intra-oral right lateral image of a child with a unilateral cleft lip and palate orthodontic treatment with space closure and correction of the unilateral crossbite. l) Intra-oral frontal image of a child with a unilateral cleft lip and palate orthodontic treatment with space closure and correction of the unilateral crossbite

link the anterior teeth together in their pre-surgical positions⁵⁷ (Fig. 9).

For relatively young children, any form of fixed appliance can be challenging, and it is

the orthodontist's responsibility to limit this intervention to a minimum while also being mindful of the ideal timing for the graft. To this effect, these appliances are often started

a year or so before the optimal timing for grafting. To stabilise the graft, there is often a thick stabilising wire running across the site and therefore, there is a need for liaison within

the team between surgeon and orthodontist to make sure this is removed at the time of surgery and replaced immediately post-op to allow the stabilisation during the healing phase while still allowing access to the cleft site for the surgeon.

The appliance is often maintained for a number of months post-surgery to allow bony healing. However, this should be balanced against its impact on oral hygiene, maintenance and orthodontic treatment burden when further definitive orthodontic care will be required a few years later.

Bone graft outcome assessment

Traditionally, outcomes for alveolar bone grafting are assessed through plain radiographic examination. Studies comparing outcomes where bone grafting is performed can be subdivided into those that look at approximately six months post graft^{42,58} and those that look following tooth eruption through the graft.^{24,32,46,59,60} In both time periods, the outcomes for graft bone height/cleft bone infiltration are superior when grafting occurs before canine eruption.⁶¹

Three-dimensional radiographic assessment using CT suggests that the bone grafts are almost completely lost in the short- to mid-term unless the canine or lateral incisor tooth, if present, erupts through the graft or is moved orthodontically into the graft to close the space anteriorly (Fig. 8).^{27,61} This requirement for graft survival is also found at ten years^{62,63} and 20 years post graft.⁶⁴

The primary objective of grafting is to allow tooth eruption, most notably, the cleft canine and definitive orthodontic intervention.²¹ Although short-term (six month) radiographic follow-up and outcome is important, it doesn't appear to necessarily relate to the cleft canine tooth eruption. The canine tooth associated with the alveolar cleft has a delayed root formation compared to the general population,^{65,66} although grafting before its eruption has been found to accelerate root development.⁶⁶ Despite this, the cleft canine still has a delayed eruption compared to the population norm.^{66,67}

The cleft canine also has an increased incidence of impaction,^{21,68,69} particularly in unilateral rather than bilateral clefts.^{43,45} As with any canine impaction, the tooth may require further surgery to expose it, prolong orthodontic treatment and add to the overall orthodontic burden of care. This will be



Fig. 9 a) Intra-oral maxillary occlusal image of a child with a bilateral cleft lip and palate at the time of bone graft assessment. There is displacement of the premaxillary segment to the patients left. b) Intra-oral frontal image of a child with a bilateral cleft lip and palate at the time of bone graft assessment. There is displacement of the premaxillary segment to the patients left. c) Intra-oral maxillary occlusal image of a child with a bilateral cleft lip and palate after orthodontic intervention. A fixed appliance including Glasgow Anterior Stabilisation (GAS) (two orthodontic bands in both central incisors, connected together and a self-ligating orthodontic tube soldered to the connector, to allow archwire engagement) on the premaxillary segment has been utilised to stabilise the premaxilla and correct the premaxillary displacement. Deciduous teeth around the cleft site have been extracted. d) Intra-oral frontal image of a child with a bilateral cleft lip and palate after orthodontic intervention. A fixed appliance including GAS on the premaxillary segment has been utilised to stabilise the premaxilla and correct the premaxillary displacement. Deciduous teeth around the cleft site have been extracted. e) Intra-oral maxillary occlusal image of a child with bilateral cleft lip and palate post bone graft and removal of the fixed appliance. f) Intra-oral frontal image of a child with bilateral cleft lip and palate post bone graft and removal of the fixed appliance. g) Intra-oral maxillary occlusal image of a child with bilateral cleft lip and palate post-bone graft and eruption of both maxillary canines through the bone graft site. h) Intra-oral maxillary occlusal image of a child with bilateral cleft lip and palate post bone graft and eruption of both maxillary canines through the bone graft site

discussed in part two of this paper which will review the orthodontic and retention challenges following development of the adult dentition. It will also discuss orthodontics for the returning adult and orthodontics for orthognathic surgery.

Conclusion

This paper (part one of two) has highlighted the orthodontic input into a child with cleft lip and palate from birth to the late mixed dentition. It has emphasised the importance of timing and planning in relation to alveolar bone grafting and its ultimate impact on definitive orthodontic outcome.

Ethics declaration

The author declares no conflicts of interest.

Written consent to publish has been obtained for

Figures 1 and 2.

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