

The orthodontist, an essential partner in CLP treatment

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Abstract. *The orthodontist, an essential partner in CLP treatment. Objectives:* Patients with orofacial clefts need multidisciplinary care, which should be provided by teams of specialists working in this field. Usually the following disciplines participate in such teams: paediatrics, plastic and reconstructive surgery, orthodontics, genetics, social work or nursing, ENT, speech therapy, maxillofacial surgery, prosthetic dentistry, psychology and oral hygiene.

Design: Narrative review.

Results: An overview is given of the orthodontic treatment protocol and the role of the orthodontist in cleft palate treatment from birth until 20 years of age for a child with a complete unilateral or bilateral cleft.

Conclusions: The orthodontist has proven to be an essential partner in the cleft palate team. The orthodontist is not only responsible for the active orthodontic and facial orthopaedic treatment of the child with a cleft. More importantly, he or she is also the guardian of the child's maxillofacial growth. By nature, orthodontists have a long-term treatment perspective in mind, which will assist the team in their consideration of treatment techniques, sequence and timing in relation to the effect on maxillofacial growth. Moreover, standardised records collected by the orthodontist provide a valuable basis for retrospective studies of treatment outcome and inter-centre comparisons. Finally, in recent times, orthodontists seem to be quite often the motor behind large-scale inter-centre randomised clinical trials in the field of CLP.

Introduction

The worldwide prevalence rates of common oral clefts vary between 0.5 and 3.0 per 1,000 live births and stillbirths, with considerable variation between gender, populations and geographic regions.¹ About 10 to 20% of children with clefts have an associated malformation. This is particularly true of patients with an isolated cleft palate.²

There has been consensus since the early 1950s that a multidisciplinary team in a centre should care for children with cleft lip and palate. The team should function as an organisation with a general policy for the treatment, and each member of the team should have an understanding of the different aspects of treatment. The team provides multidisciplinary treatment and usually includes special-

ists from the following disciplines: paediatrics, plastic and reconstructive surgery, orthodontics, genetics, social work or nursing, ENT, speech therapy, maxillofacial surgery, prosthetic dentistry, psychology and oral hygiene. The aim of this multidisciplinary treatment approach is to create the conditions that allow the affected child to grow up with an aesthetically pleasing face, good hearing and speech, properly functioning and aesthetically acceptable dentition and, last but not least, harmonious social-psychological development.

In this paper, the role of the orthodontist as an essential team player in the treatment of cleft lip and palate patients will be described for three patient-age categories: 0 to 7 years, 8 to 15 years and 16 to 20 years. The treatment overview as described here can

vary widely between teams. An overview of treatment protocols of 201 cleft palate teams in Europe can be found in the description of the EUROCLEFT project.³

Age period 0 to 7 years

Shortly after birth, a child with a cleft will be seen by the CLP team, more in particular by the paediatrician, clinical geneticist, plastic surgeon and orthodontist, and, if available, by the social worker or social nurse of the team. An initial treatment plan is made focusing on reconstructive surgery of the lip and/or the palate, and neonatal maxillary orthopaedics can precede the surgical closure of the lip. Other medical conditions of the child, and the parents' psychological or social problems, will be included in the initial treatment plan.

Neonatal maxillary orthopaedics

More than half a century after its introduction by McNeil,⁴ neonatal maxillary orthopaedics still remains a controversial part of comprehensive care for cleft lip and palate patients. The therapy, also known as presurgical or early orthopaedic treatment, presurgical or infant orthopaedics, early maxillary orthopaedics, or more recently nasoalveolar moulding, was rapidly adopted by many centres around the world, although at that time there was no scientific evidence for the claimed benefits, nor were the possible adverse effects properly investigated.⁵ In Europe in the year 2000, about 54% of the operational centres used neonatal maxillary orthopaedics.³

Neonatal maxillary orthopaedics were originally introduced to restore normal anatomy and to guide the growth and development of the maxillary segments. It was only later that the discipline of orthodontics tried to justify early intervention for other reasons. Prahl *et al.*⁶ summarised the arguments of the proponents of the use of infant orthopaedics, who state that this approach allows a more normalised pattern of deglutition, prevents the twisting and dorsal position of the tongue in the cleft, improves the arch form and position of the alar base, facilitates surgery, and improves outcomes in general. Other alleged benefits in vogue are: reduction of posterior cleft width, prevention of initial collapse after surgery, prevention of cross-bite, straightening of the nasal septum, facilitation of feeding, less danger of aspiration, better speech development, better nose breathing, better middle ear

conditions, less extensive orthodontic treatment at later ages, and a positive psychological effect on the parents.

Many different types of neonatal maxillary orthopaedics have been described in the literature. A wide range of appliances have been designed for this purpose, with pin-retained active appliances at one end of the spectrum^{7,8} and passive appliances at the other end.⁹ They can be allocated arbitrarily to three main categories: active, semi-active, and passive appliances. Active appliances are constructed to apply a force to the maxillary segments to move them into the desired direction by using an active force delivery system like springs and screws. Additional anchorage can be obtained by pins that are driven into the maxillary bone holding the plate in position. Semi-active appliances are constructed by sectioning the dental cast and reorienting the maxillary segments in a more favourable position. The plate is made on the reconstructed cast and will force the palatal segments in the predetermined direction when placed in the oral cavity. External strapping across the cleft can be part of the treatment protocol. These are the McNeil⁴ and Burston¹⁰ type of appliances. Passive appliances that are combined with extra-oral strapping also fit into this category. Passive appliances are supposed to induce arch alignment during growth by grinding away material in definitive areas of the plate to ensure the proper spontaneous development of the segments. The plate is held in position by suction and adhesion only and no extra-oral strapping is applied. The "Zurich approach" as proposed is the best-known representative of this kind

of neonatal maxillary orthopaedics.^{9,11}

Finally, in the 1990s, a prospective two-arm randomised clinical trial was started to investigate the effect of neonatal maxillary orthopaedics in children with a complete UCLP. The trial was known as 'DUTHCLEFT' and it involved collaboration between three academic Cleft Palate Centres in the Netherlands, i.e. the Cleft Palate Centres of Radboud University Nijmegen Medical Centre, the Amsterdam Free University Hospital and Rotterdam Erasmus University Hospital.^{5,6} Children will be followed until 9 years of age. The following outcome variables are being studied:

- A. general effects: influence on feeding, body length and weight gain, parental satisfaction;
- B. surgical and orthodontic effects: duration of lip surgery, aesthetic outcome, maxillary arch form and dimensions, maxillofacial growth;
- C. speech and language development: prelingual sound production, early speech and language development, intelligibility;
- D. cost-effectiveness: medical and non-medical costs.

On the basis of the results of DUTHCLEFT so far, it can be concluded that neonatal maxillary orthopaedics in unilateral cleft lip and palate patients as performed in this trial is not necessary for feeding, parental satisfaction or orthodontic reasons. Regarding speech, a positive but very limited effect was found prior to the age of 2½ years, but the speech of the children with clefts was far behind that of their non-cleft peers

anyway. Relative to the total costs of the treatment of a UCLP patient, the financial investment to attain this effect was rather limited. However, it is questionable whether this limited effect is important enough to justify neonatal orthopaedics. It should also be taken into consideration that the 6-year results for speech have not been analysed yet.⁵

For children with bilateral clefts, there is no evidence-based data from randomised clinical trials about the effect of neonatal maxillary orthopaedics. On the basis of experience, it can be stated that neonatal maxillary orthopaedics results in a good alignment of the maxillary segments, which makes it possible to close the lip in one operation.

Facial mask

Despite careful monitoring of maxillary growth, mid-facial retrusion remains a common finding in operated CLP patients. Delaire *et al.*¹² have laid down the theoretical basis for the clinical application of maxillary protraction in patients with severe maxillary deficiency. Since then numerous case reports and case series studies have been published, a majority being concerned with non-cleft Class-III individuals.¹³ But studies of CLP patients in homogeneous groups with adequate sample size and long-term follow-up remain rare.

There seems to be general agreement that orthopaedic forces applied to the maxillary complex are more effective at an early age i.e. in the deciduous and early mixed dentition stages,¹³ given the patency of circummaxillary sutures. However, no comparative, well-controlled studies of timing

of maxillary protraction in CLP could be found. In a non-cleft sample, comparisons of the results of maxillary protraction started between 5 and 8 years of age and between 8 and 12 years found no difference in maxillary skeletal response, but vertical response was greater in the older age group.¹⁴

Most of the more recent studies of CLP originate from Bergen (Norway), but the results published so far cover only a three-year period after protraction.¹³ Since 1977, the Bergen Rationale involves transverse expansion, anterior protraction and fixed retention at the age of 6 to 7 years. The treatment procedure is assumed to normalise orofacial function and dimensional anatomy in order to reduce detrimental functional disturbances during further growth. A significant increase in the SNA angle of 0.9 degrees was found, together with a clockwise rotation of the mandible and an increase in lower facial height. As a result, a mean change of 2.3 degrees in the ANB angle was found. However, the treatment response was highly variable. The short-term effect of maxillary protraction was found to be a maxillary skeletal/dentoalveolar effect in a ratio of 45%/55% for UCLP and 10%/90% for BCLP.¹⁵ The changes were reflected in the soft-tissue profile as a mild increase in nose prominence and upper lip protrusion, as well as a reduction in lower lip protrusion and chin. During a three-year follow-up period, the initial growth pattern reappeared, but the upper jaw position remained stable. The maxillo-mandibular relationship was impaired due to normal downward and forward growth of

the mandible, while the maxillary position relative to the anterior cranial base remained constant. Although the effect on the soft-tissue profile diminished, some positive effect still remained.¹³

The results of the Bergen studies have been confirmed in general by studies from a limited number of centres around the world, but most published results have been short-term. The question remains, however, whether the long-term effect is stable and adequate to avoid surgical advancement of the maxilla after puberty. In this respect, cost-effectiveness analyses are also required.

Age period 8 to 15 years

Between 9 and 11 years of age and prior to the eruption of the canine of the cleft-affected side, the alveolar process is reconstructed with an autologous bone graft in most centres. In patients with bilateral clefts this operation is sometimes combined with a premaxillary osteotomy in order to correct the vertical position of the premaxilla.¹⁶ Orthodontic expansion of the maxillary arch is often necessary to align the maxillary segments. This can be easily performed in about 6 to 12 months with a removable or fixed expansion device.¹⁷

Nowadays, bone grafting of the alveolar cleft is a standard procedure in most of the cleft centres around the world. The benefits of restoration of the cleft alveolar ridge are appreciated with respect to supporting cleft-adjacent teeth, stabilising the cleft maxillary segments, eliminating the notched alveolar ridge, supporting the alar bases, and enabling expansion of the interpremaxillary suture.¹⁸ The landmark paper of Boyne and

Sands¹⁹ readdressed the bone grafting issue in mixed dentition with a technique that has become a standard still in use today at many centres. With the continued concern of orthodontists about possible scar-induced growth inhibition as a result of any surgery, it is now being asked whether early secondary bone grafting affects the subsequent growth of the maxilla. An international multi-centre cephalometric study found indications of vertical maxillary deficiencies in some of the sample.²⁰ Other studies were unable to detect any differences in maxillary size or position in a secondary grafted group when compared to a group of ungrafted matched controls.^{21,22}

In general, despite the absence of any prospective, controlled trials, the existing data from retrospective studies on secondary bone grafting would seem to indicate minimal effects, if any, on maxillofacial growth. The possible growth impairment of primary grafting resulting from the additional scar tissue in growth-sensitive areas and at a growth-sensitive age for the maxilla seems to have been circumvented by postponing the procedure until the mixed dentition stage. What remains to be evaluated is the "price to be paid" for delaying surgery, particularly with respect to the inability to provide a bony environment for the eruption and maintenance of cleft-adjacent lateral incisors, when present. In balancing these "benefits" and "burdens" of the procedure, it may be possible that, in the future, varied timing of secondary bone grafting may be optimal: earlier in patients with healthy lateral incisors or central incisors with marginal bone support requiring an intact

alveolar ridge at a younger age but risking some growth interference if done too early; later in patients with adequate support for cleft-adjacent teeth, where the risk of maxillary growth problems exceeds the benefits of additional surgery at a much younger age.²³

After bone grafting, the orthodontic treatment will be continued, with timing being dependent on the developmental stage of the dentition and especially the eruption of the canine adjacent to the cleft. Fixed appliances are always necessary and the treatment will take several years to be completed. If maxillofacial growth develops favourably, this will be the final orthodontic intervention. Unfortunately, in a considerable number of patients with clefts, a combined surgical-orthodontic correction of the skeleton is necessary after growth has ceased. After active treatment, life-long retention seems to be necessary, particularly to maintain the maxillary transverse dimension.

Age period 16 to 20 years

Despite careful growth monitoring, many patients with complete clefts of the alveolus and palate end up with a mid-facial deficiency after puberty. Correction of the jaw deformity is planned for when permanent dentition has fully erupted, the dental arches have been aligned orthodontically, and maxillofacial growth has ceased. Unfortunately, it is not so easy to determine the end point of facial growth. Chronological age and hand-wrist radiographs do not provide reliable information. The decision should be based on individual longitudinal cephalometric data of craniofacial growth. The radiographs should be taken with

an interval of at least 6 months. However, because the margin of error for the method is large, longer time intervals are to be preferred. Nowadays there is a tendency to operate earlier and not to wait until late adolescence to complete definitive surgery.²⁴ Clearly, this has psychosocial advantages as the child will not have to live through puberty with a disfigured face. Furthermore, it means that orthodontic treatment can be provided in a single stage extending from the period after bone grafting until the surgical correction of the jaw deformity. On the other hand, there is still no long-term data about the final outcome of orthognathic surgery or maxillary distraction to correct skeletal malformations in CLP patients before maxillofacial growth has ceased completely.

The final orthodontic correction should be carefully planned in consultation by all team specialists involved. At this stage the prosthodontist may want to replace missing teeth or restore hypoplastic incisors and the plastic surgeon will perform final nose and lip corrections. A good framework of well-aligned dental arches and an optimal jaw relationship will support these late corrections.

Evidence-based care?

Over the years, the orthodontist has proven to be an essential partner in the cleft palate team. The orthodontist is not only responsible for the active orthodontic and facial orthopaedic treatment of the child with a cleft. More importantly, he or she is also the guardian of the child's maxillofacial growth. By nature, orthodontists have a long-term treatment perspective in mind, which will assist the team in

its consideration of treatment techniques, sequence and timing in relation to the effect on maxillofacial growth. Moreover, standardised records collected by the orthodontist provide a valuable basis for retrospective studies of treatment outcome and inter-centre comparisons. Finally, in recent times, orthodontists seem to be quite often the motor behind large-scale inter-centre randomised clinical trials in the field of CLP.²⁵

In 1991 the National Institute of Dental Research (NIDR) sought grant applications for prospective and or retrospective clinical trials evaluating treatment procedures for non-syndromic, unilateral cleft lip and palate. The background information was that treatment sequence, timing, methods and surgical techniques were all controversial. The long-term impact on maxillofacial growth and speech were considered to be of primary importance. This initiative reflected the lack of evidence underlying treatment at this point in time. Even in 2006, not many prospective randomised clinical trials have been performed for children with clefts.²⁵ CLP research and/or the aggregation of existing data into meta-analyses is extremely difficult due to problems inherent to the CLP material itself and due to a series of inherent weaknesses of many studies published so far²³: the large phenotypic variability of the CLP population results in significant heterogeneity in the study samples; the length of the follow-up period necessary to detect growth effects also allows for confounding variables to occur, such as changes in treatment protocol, changes of surgeons, changes in operator skills, attrition of the sample, etc;

there are numerous different treatment protocols; only small sample sizes are available for all types of clefts, but samples are particularly small for the rarer conditions such as complete BCLP; inadequate follow-up period from the time of primary surgery to the time of evaluation makes the determination of growth effects difficult; there is a lack of randomised controlled clinical trials, which provide the highest level of evidence; and, finally, there is a lack of controls in many studies.

These and other methodological shortcomings make it seem as if relatively little has been accomplished over the past 50 years. However, with the negative effects of scar tissue now well accepted, the next challenge is to balance this consideration against the desire to make things as "normal" as possible, as soon as possible. To answer some of these questions in the decades to come, three research approaches need to be pursued: (a) laboratory experiments; (b) prospective randomised controlled trials; and (c) controlled inter-centre outcome studies. Finally, for outcome studies to provide valid comparisons, efforts need to be made to standardise record-taking so that growth data from different centres are consistent for age and stage of development.²³

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