

Description of transverse maxillary expansion in patients with bilateral cleft lip and palate undergoing gingivoperiosteoplasty with alveolar bone graft. A preliminary study

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ABSTRACT

Cleft lip and palate (CLP) is one of the most common facial deformities. Treatment is long and complex, requiring a multidisciplinary team, including an orthodontist, who plays a prime role in ensuring that dental phenomena occur under the best possible conditions allowed by often unfavorable anatomy and growth and healing factors. Orthodontic preparation by maxillary expansion is a key step in management, mandatory ahead of gingivoperiosteoplasty and alveolar bone graft. Since 2000, Pr Vasquez's maxillofacial and plastic surgery team in the Necker Hospital (Paris, France) has used a treatment protocol involving alveolar cleft repair by gingivoperiosteoplasty and alveolar bone graft at 4-6 years of age.

Orthopedic mechanics induces not only strictly orthopedic effects but also orthodontic dento-alveolar compensation. In CLP, physiology is exceptional in that there is no median palatine suture between fragments, and it can be supposed that the behavior of surrounding structures during maxillary expansion differs from that of a classical patient.

We therefore undertook a radiologic study of the dento-alveolar phenomena accompanying preoperative orthodontic expansion in bilateral CLP patients, in a group selected from those jointly managed by Pr Vasquez's team and Dr Vi Fane's team in the Rothschild Hospital (Paris), under the auspices of the MAFACE rare diseases reference center.

KEY WORDS

Cleft lip and palate, maxillary expansion, gingivoperiosteoplasty, secondary alveolar bone graft

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INTRODUCTION

Cleft lip and palate (CLP) is a very heterogeneous clinical entity comprising congenital deformities of mixed genetic and environmental origin. It affects the orofacial region, with involvement of varying complexity of the lip, nose, alveolar region and hard and soft palate and is due to fusion defect in the embryonic buds occurring at 4-12 weeks' gestation.

It is the most common facial abnormality. It may be isolated or associated with various deformity syndromes of varying complexity. Worldwide prevalence is estimated at about 1 per 700 live births¹⁷. Bilateral CLP is the rarest non-syndromic form, at only 10% of clefts; 20% of CLPs are bilateral. CLP shows 2:1 male predominance¹.

CHARACTERISTICS OF BILATERAL CLP

Anatomic characteristics

CLP is associated with major deformity of the central face, with marked tissue discontinuity and labio-columellar defect¹⁶. Muscular discontinuity entails dynamic imbalance; all the cartilage structures to which the affected muscles insert gradually deform, being very malleable. Deformity thus involves both soft and hard

tissue. The lip has a hypertrophic vermillion aspect. The nose is projected down and forward by the absence of columellar support and excessive traction by the orbicularis oris muscles on either side (Fig. 1). Premaxillary protrusion is the main characteristic of bilateral CLP, creating a very strong convexity profile, especially as the mandible is retrusive in infants. The volume of the

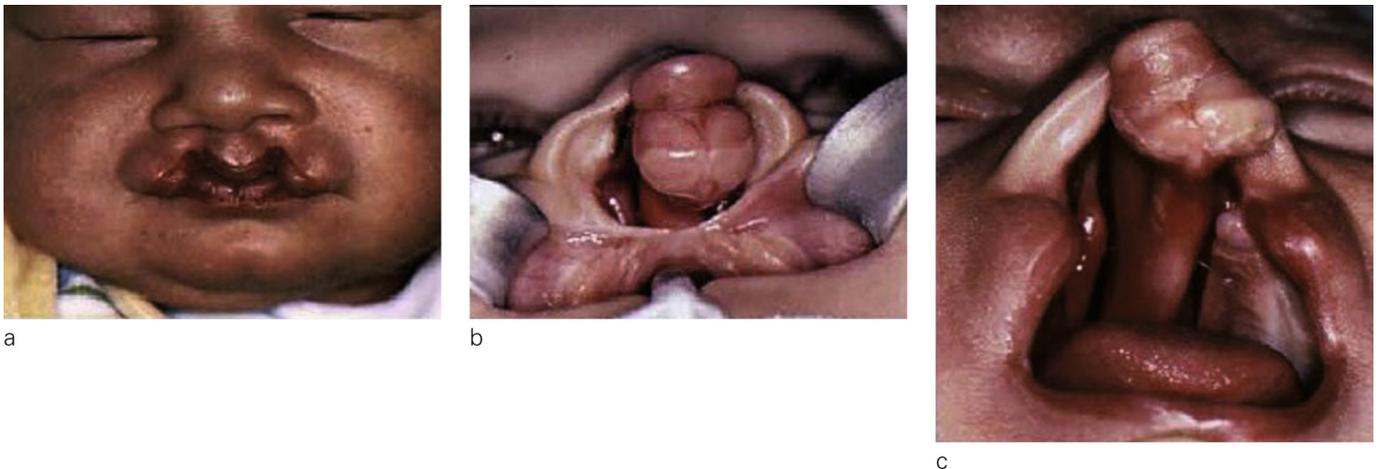


Figure 1

Various forms of bilateral CLP (from Morand and Raphael, 2004)¹⁶. a) Bilateral labial cleft. b) Bilateral labio-alveolar cleft. c) Bilateral total labio-alveolar-palatine cleft.



Figure 2
Influence of number of tooth germs on premaxillary volume (from Morand and Raphael, 2004)¹⁶.

premaxilla, and thus the number of tooth germs it contains, greatly affects local growth¹⁶ (Fig. 2).

Skeletal characteristics

Skeletal specificities are found in operated patients, resulting from a double influence: genetic (congenital) and the results of repair surgery (acquired), which inevitably has side-effects, notably scarring². Overall, there is little difference in the skull-base between bilateral CLP patients and healthy subjects, as it is remote

from the deformity and uninvolved in surgery²⁷. In the maxilla, repair technique impact is mainly premaxillary. Cheiloplasty and rhinoplasty reposition the premaxilla; if it was originally rather protruding, it may then become retrusive. Moreover, it should be borne in mind that skeletal class III is common in CLP. Premaxillary repositioning tends to normalize the anteroposterior relation with the mandible³³, which shows growth equivalent to that of healthy subjects. The direction of growth, on the other hand, tends to be divergent as mixed respiration is common, as is a low

position of the tongue due to the reduced maxilla^{27,33}.

Dental characteristics

Occlusion is severely disturbed by the disorganization of skeletal and alveolar support; dental abnormalities are much more frequent than in the general population. This is due to genetic, embryologic and anatomic interactions in the development of the lip, primary and secondary palate and tooth germs^{21,34}. Abnormality concerns the number of germs, with more frequent agenesis of the lateral maxillary incisors and second maxillary premolars^{6,12,31,34} (Fig. 3), and the size, form and structure of teeth, but also organogenesis and eruption⁶. Thus, the risk of included maxillary canine, for example, is 20 times as high as in the general population²⁴. The severity of dental abnormality is agreed to correlate

with cleft severity. This is why bilateral cleft is especially involved. Concerning inter-arcade relations, Heidbuchel *et al.*¹³ found numerous differences from healthy subjects, with a tendency for aggravation after 7 years of age. Maxillary transverse insufficiency is more marked in the canines than molars, and worsens over time if not corrected (Fig. 4a). Consequently, inter-molar diameter is often reduced at the mandible, to compensate, and the anterior supra-occlusion caused by the protruding fragment may limit mandibular growth. Anteroposteriorly, the arch angle is often reduced, leading to distal occlusion of the whole arch. Vertically, cover is often excessive, due to premaxillary repositioning (Fig. 4b). Occlusion is thus severely disturbed, with class II or III molar relations, anterior and lateral occlusion inversion, and crowding (Fig. 4c).

TREATMENT

Surgical treatment of cleft has a 3-fold objective: restoring anatomy, respecting and optimizing growth, and restoring function^{8,10,19,28,29}. It mobilizes a multidisciplinary team, which will develop its own protocol according to its therapeutic priorities. There are as many protocols as there are teams. The 2001 Eurocleft study found 194 different protocols among 201 European centers managing cleft²⁶. Respective results can only be assessed in the long term, so that there is at present no consensus as to treatment schedule or surgical technique. The protocol employed by

Pr Vazquez's team in the MAFACE rare diseases reference center is inspired by Talmant's functionalist approach, in which strictly nasal respiration is an essential aim to restore normal maxillary development^{20,28,29}. Early uranoplasty also enables speech acquisition, with the aim of mastering language by primary school. Since 2000, the team's primary treatment schedule has been as follows:

- 3-6 months: veloplasty associated to 1-step bilateral cheilorrhinoseptoplasty;
- 12 months: palate cleft closure.



Figure 3

Panoramic radiograph showing lateral maxillary incisor agenesis in a 7 year-old reference center patient with bilateral CLP.



Figure 4

Inter-arcade abnormalities. a) Maxillary arch occlusion: lateral segment collapse and severely reduced inter-canine diameter in a reference center patient aged 4 years 8 months with bilateral CLP. b) Lingual inclination and increased premaxillary cover following cheiloplasty in the same patient. c) Bilateral anterior and posterior occlusion inversion in a reference center patient aged 4 years 8 months with bilateral CLP.

- 4-6 years: gingivoperiosteoplasty (GPP) with bone graft, in 2 steps in case of bilateral cleft (Fig. 5).

The final step in the protocol, and the focus of the present study, more particularly involves the orthodontist. Since Boyne and Sands' work in the

1970s, gingivoperiosteoplasty (GPP) has usually been associated to alveolar graft^{7,11,29}. Several benefits are expected, and notably restoring alveolar bone continuity over and above mere tight closure, restoring an adequate bone support for dental

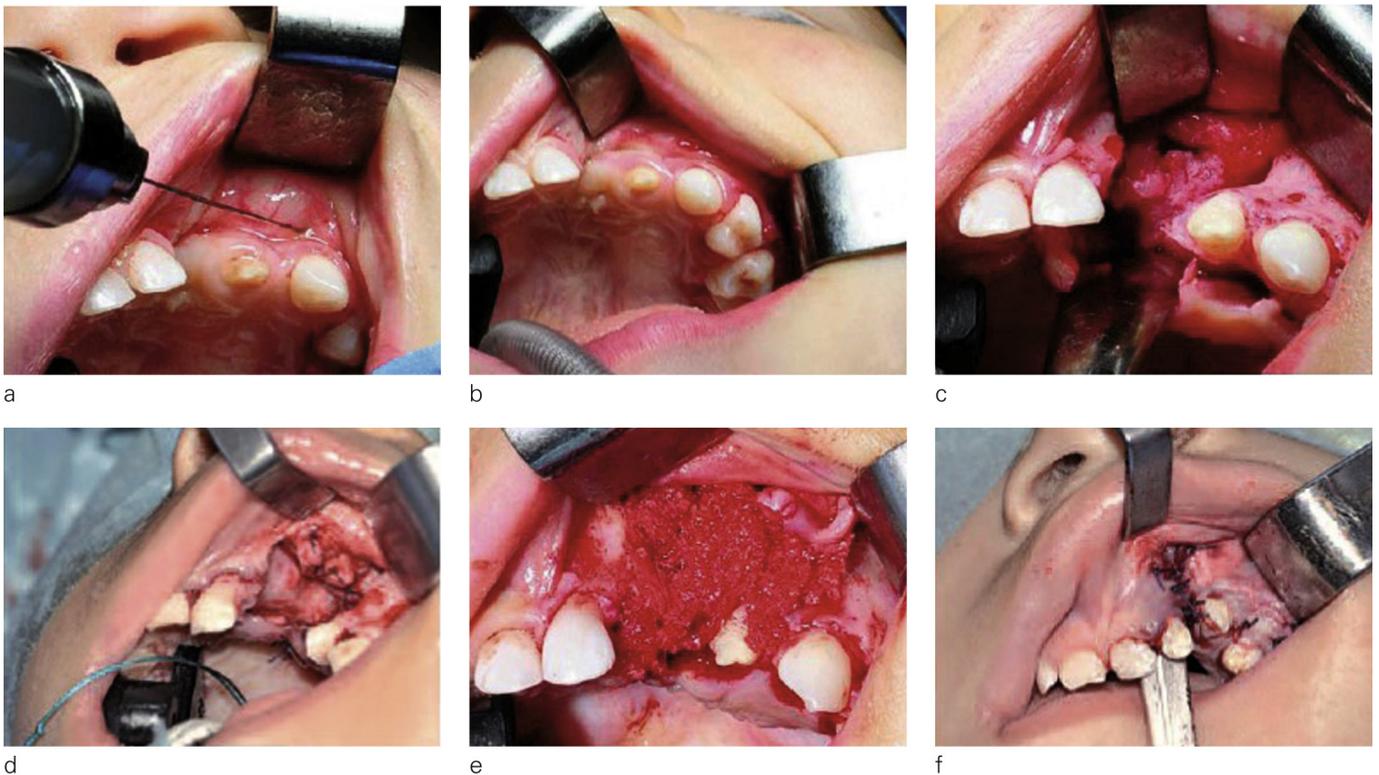


Figure 5

Gingivoperiosteoplasty in 2 cases of left unilateral CLP (from Talmant and Lumineau²⁹ and Picard et al.²⁰). a) Anesthesia. b) Incision at dental necks on vestibular side. c) Wide muco-periosteal detachment. d) Nasal and palatine suture. e) Cleft filling by iliac graft. f) Vestibular suture and postoperative aspect.

eruption, and maintaining arch width and preventing lateral fragment collapse. In the Necker Hospital maxillofacial and plastic surgery department's protocol, late primary GPP associates orthopedic treatment and bone graft²⁰. Initial orthopedic treatment (Fig. 6) of the deciduous teeth using a rapid maxillary expander on dental splints followed by a removable Quad-Helix appliance restores canine function, corrects endo-occlusion, conserves lateral incisor space and restores piriform orifice width. GPP is then performed around the age of 4 years, when the cortical bone is dense enough to protect the tooth germs while the soft tissue is

still very plastic²⁹. The cleft space is filled by cancellous iliac graft. The orthopedic system (Quad-Helix appliance) is left as contention for about 6 months, during which time arch expansion may be continued if necessary as continuity has been restored. Graft volume and success are assessed on CT or CBCT (Cone Beam Computed Tomography).

RATIONALE

Maxillary expansion can be associated to several other results³⁰: median palatine suture opening (the ideal skeletal effect sought);

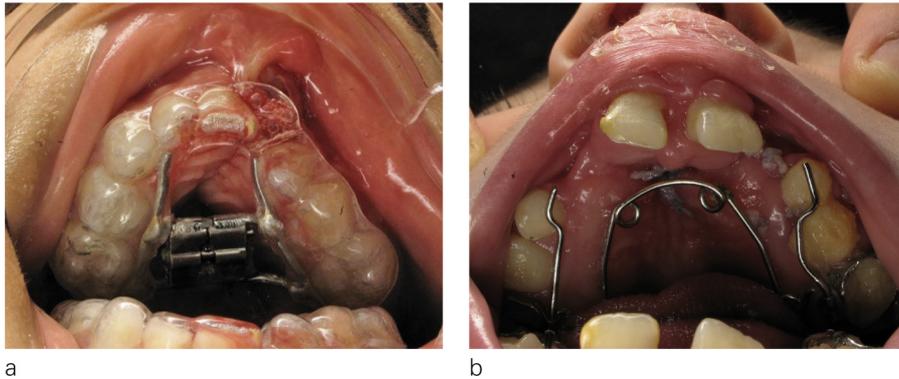


Figure 6

Reference center patient aged 6 years 6 months, with bilateral CLP. a) Orthopedic expansion by rapid maxillary expander on sealed dental splints. b) Contention phase with removable Quad-Helix appliance after left GPP.

appositional growth; vestibular inclination of the alveolar processes; and/or vestibular tipping, or tooth egression^{3,14,30}. Two types of expansion have been mainly studied: rapid expansion with rapid maxillary expander, reputed to have mainly skeletal impact, and slow expansion by Quad-Helix, with mainly dento-alveolar impact^{30,14,15,22,23,3,18,25}. Several studies have focused on alveolar growth, especially transverse growth. Björk and Skieller in particular made thorough studies of "normal" growth, especially using their endosseous implant method^{4,5}. They found that maxillary suture growth, like height and weight growth, shows peak acceleration in puberty. In the late 1970s, Dahl studied transverse maxillary growth, specifically in CLP⁹. Growth rates in a sample of patients showed that all transverse dimensions of the face displayed the same puberty peak as found for overall

growth, with the exception of maxillary width. Moreover, the expansion obtained in the suture was much less than in healthy subjects. There is in fact no suture in CLP patients, and maxillary structures are mainly displaced by the surrounding muscular structures^{9,14}. This exceptional physiology raises the question of how transverse expansion is achieved after orthodontic/orthopedic preparation.

The objectives of the present study were therefore: (i) to determine whether expansion is achieved by compensatory dento-alveolar movement under coronary vestibular version alone, or by pure distal translation; (ii) to compare expansion in canine versus molar sectors; (iii) to determine whether there is transverse relapse and, if so, of what kind and intensity; and (iv) to study occlusion plane displacement.

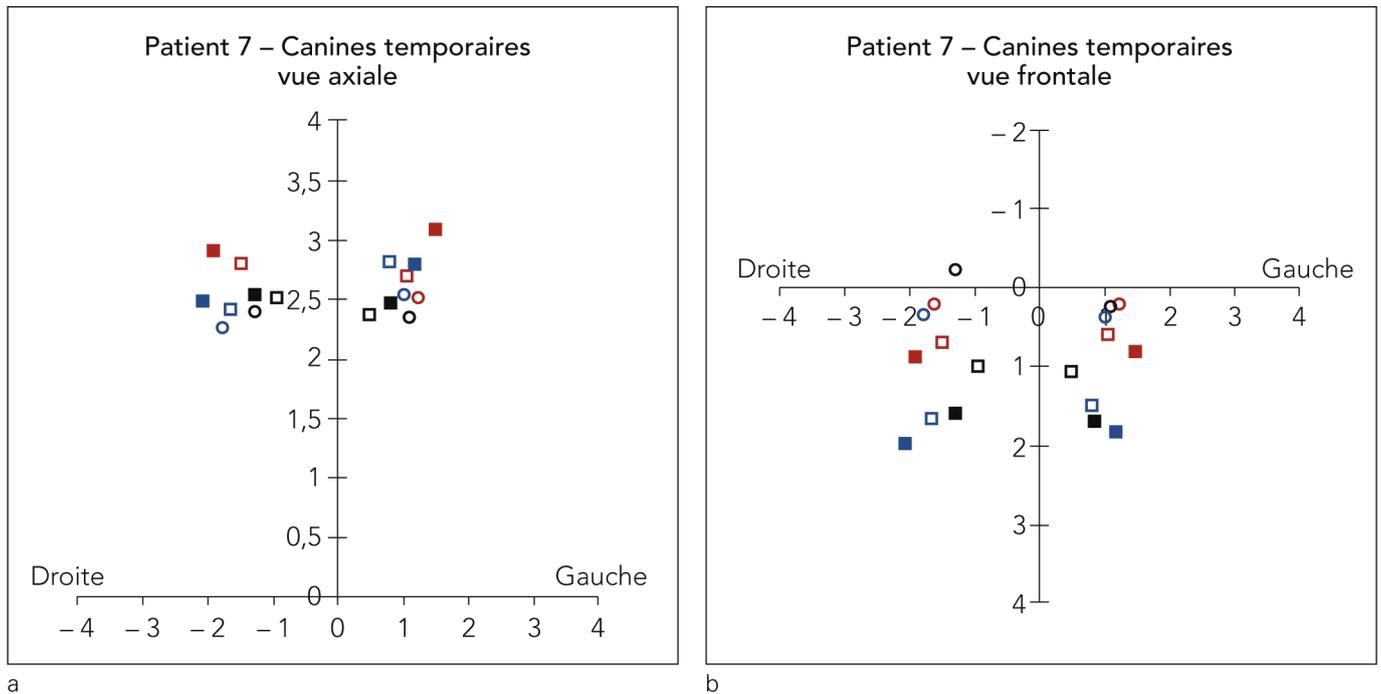


Figure 7

Example of graphic representation of results, deciduous canines, patient n° 7, (a) axial and (b) frontal view. Circle = apex; empty square = neck; full square = canine tip; black = baseline situation at T0; blue = pre-GPP situation at T1; red = post-GPP situation at T2.

MATERIAL AND METHOD

Patients were selected from among those in orthodontic treatment with Dr Vi Fane's team, in private practice or in Pr Ariane Berdal's rare diseases reference center and Pr Marie-Laure Boy-Lefèvre's functional unit in the Rothschild Hospital, and for whom primary surgery followed the 2000 Necker protocol. Inclusion criteria were: patient with bilateral CLP, and not purely alveolar cleft, and for whom native slice 3D data at 3 follow-up time points were available: baseline CBCT (or, in some cases, CT) (T0); pre-GPP CBCT at end of orthodontic preparation (T1); and post-GPP CBCT some 6 months

after the second GPP (T2). In all, 7 patients with complete bilateral CLP were included in this retrospective study: 3 boys, 4 girls; mean age at start of orthopedic treatment, 4.5 years. Orthopedic preparation systematically used a rapid maxillary expander on dental splints followed by the Quad-Helix appliance. The maxillary arch was usually considered well-prepared for GPP if the intercanine distance CC' was between 33 and 35 mm. All maxillary CBCT scans were analyzed on Osirix[®] freeware. Measurements were taken from the digital tracing, with a posterior reference axis, through the two posterior

palatine foramina. Measurements were made for the deciduous maxillary canines, definitive maxillary canines and deciduous maxillary second molars. To compare coronary and root expansion, three vertical landmarks were located: a coronary (cuspid) point, a cervical (neck) point, and a radicular (apex) point. In view of patient age, the radicular point was not studied in definitive canines:

RESULTS

Tables I to III present the movements obtained according to teeth, side and occlusion plane displacement. Mean post-GPP expansion (i.e., total final expansion) for the deciduous canines was 1.6 mm for the

as they are relatively high, they may be thought to reflect the basal effect induced by expansion. Three values, x, y and z, were attributed to each landmark, to estimate the 3D variations of each point at different stages of treatment. Dental movement was presented graphically, on frontal (Fig. 7a) and axial view (Fig. 7b), using KaléidaGraph® software.

apex, 3.6 mm for the neck and 4.6 mm for the crown; for the deciduous second molars, 0.9 mm, 3 mm and 3.6 mm respectively; and for the definitive canines, 0.9 mm for the neck and 0.6 mm for the crown.

DISCUSSION

Degree of expansion

The expansion achieved in this preliminary study was greater anteriorly than posteriorly, in agreement with other reports^{3,14,25,30}. The position of the premaxilla was basically stable over time (no lateralization of the fragment). There may be several reasons for this. Natural maxillary growth includes rotation of the two maxillary hemispheres around their posterior attachment at the pterygoid processes³⁰. The absence of inter-incisor diastema, reported by Isaacson *et al.*¹⁴ in bilateral CLP, indicates that expansion is not in the inter-incisor suture, as in healthy subjects, as the median fragment is isolated from the continuity of the arch. The two anterior alveolar clefts may thus represent a double zone of expansion.

Clinically, this is not disadvantageous, since, as seen above, transverse maxillary defect is greater in the canines and worsens with time¹³.

Nature of expansion

Expansion was greater in the crown for deciduous canines and molars: i.e., in the direction of the increase in natural vestibular version; little variation was found in definitive canines. In agreement with the literature^{3,14,23}, and in contrast to what might be expected, expansion thus seems to be predominantly dento-alveolar rather than basal. According to Isaacson *et al.*, facial resistance is increased in case of cleft, to compensate for tissular discontinuity. Thus, in bilateral cleft, where tissue

Table I: Evolution between baseline and orthodontic preparation (T0 to T1).

| PATIENTS | Deciduous canines | | | Deciduous molars | | | Definitive canines | Occlusion plane | Left/right uniformity |
|-----------|-------------------|--------------------|--------------------|------------------|--------------------|------------------------|--------------------|-----------------|-----------------------|
| | Transverse | Distal translation | Vestibular version | Transverse | Distal translation | Vestibular version | Transverse | | |
| Patient 4 | ↗ | YES, esp. right | NO | ↗ | YES ++ | YES | ↗ | ↗ | NO |
| Patient 5 | ↗ | NO | YES | ↗ | YES | YES++ | ↗ slight | → | YES |
| Patient 6 | → | | | ↗ | YES | rather lingual version | ↘ slight | → | YES |
| Patient 7 | ↗ | NO | YES++ | ↗ | YES | YES++ | ↗ | ↘ | NO |

Green: transverse increase and/or improvement; red: transverse decrease and/or aggravation; blue: stable.

Table II: Evolution between baseline and post-surgical phase (T0 to T2).

| PATIENTS | Deciduous canines | | | Deciduous molars | | | Definitive canines | Occlusion plane | Left/right uniformity |
|-----------|-------------------|--------------------|--------------------|------------------|--------------------|--------------------|--------------------|-----------------|-----------------------|
| | Transverse | Distal translation | Vestibular version | Transverse | Distal translation | Vestibular version | Transverse | | |
| Patient 1 | ↗ | YES, left | YES, left | ↗ | NO | YES++ esp.left | //// | ↗ | NO |
| Patient 2 | ↗ | NO | YES right | ↗ | NO | YES right | → | → | NO |
| Patient 7 | ↗ | YES, right | YES++ | ↗ | YES | YES++ | ↗ | ↗ | NO |

Green: transverse increase and/or improvement; red: transverse decrease and/or aggravation; blue: stable.

Table III: Evolution between orthodontic preparation and post-surgical phase (T1 to T2).

| PATIENTS | Deciduous canines | | | Deciduous molars | | | Definitive canines | Occlusion plane | Left/right uniformity |
|-----------|-------------------|--------------------|--------------------|------------------|--------------------|---------------------|--------------------|-----------------|-----------------------|
| | Transverse | Distal translation | Vestibular version | Transverse | Distal translation | Vestibular version | Transverse | | |
| Patient 3 | ↗ | YES | NO | ↗ | YES, left | YES, right esp.left | → | ↗ | NO |
| Patient 7 | → | | | ↘ | | | → | ↗ | NO |

Green: transverse increase and/or improvement; red: transverse decrease and/or aggravation; blue: stable.

discontinuity is greatest and scar areas most numerous, movement is more restricted and dento-alveolar compensation predominates over the basal effect, as compared to unilateral CLP³⁰. Although the intrinsic nature of the cleft cannot be altered, intervention to correct the transverse dimension should be early, as, during growth, expansion increasingly induces dento-alveolar compensation.

Relapse

In all patients, the transverse direction is considered especially subject to relapse. By interposing osseous material between the distracted maxillary segments, GPP may seem to be a means of maintaining expansion. However, this bone filling is purely anterior. In 1964, Matthews and Grossmann¹⁵ concluded that orthopedic expansion followed by GPP was associated with no relapse. This was in contradiction to more recent reports, systematically finding a general tendency to

relapse^{3,18,22,23,32}, even with bone graft^{18,22,23,32}.

The present results seem to show that expansion is maintained or even increases. There may be several reasons for this. The orthodontic expansion device, fitted ahead of GPP, is left in the mouth well after healing; a contention effect may thus minimize relapse. Natural transverse growth may also help maintain expansion. The type of expansion does not seem to be a predictive factor: rapid expansion is reputed to have skeletal and thus more stable impact, but slow expansion is less subject to resistance from surrounding tissue, allowing more physiological bone formation that is less at risk of relapse³. It should, moreover, be borne in mind that the clinician can reactivate the Quad-Helix appliance that is left in contention if need be. Further study is needed of the stability induced by grafting, which cannot alone maintain expansion, as demonstrated by Robertson and Fish²².

PERSPECTIVES

Taken together, the present retrospective findings provide interesting preliminary information; however, statistical analysis is not possible at this stage.

Like other teams studying bilateral cleft, we were unable to constitute a powerful sample. Bilateral clefts are the most rare. Moreover, to achieve homogeneity, not all patients seen in the rare diseases reference center were included, unless surgery had been performed by Pr Vazquez's

team following the protocol adopted in 2000.

Treatment duration for bilateral cleft posed a further problem. The interval between two GPPs was about 6 months, so that the final CBCT scan (T2) implies a much longer observation period than in unilateral CLP. For this reason, CBCT scans were not available in all cases, especially in DiCOM format. Continuing the study, however, should flesh out these preliminary conclusions,

including new patients, with longer follow-up, notably at the time of definitive canine eruption.

Radiology data collection also involved problems. The CBCT scans were intended for use at two sites, Rothschild and Necker, and priority was given to the surgery for which they had been prescribed. This geographic dispersion of data hampered collection; documents were sometimes in the hands of the patient's parents.

CONCLUSION

The initial results of the present study show that expansion was principally dento-alveolar by coronary version, rather than skeletal as we had supposed. Also, it was greater for canines than molars. Finally, the findings regarding relapse suggest that expansion is maintained, on condition that the expansion apparatus be kept in contention for a relatively long time.

The present report is part of a larger-scale and longer-term study

Furthermore, the radiology reports were not systematically accompanied by a CD-ROM with the native slices. This was especially the case for older patients and older examinations. The radiology films were in such cases analyzable clinically, which was the original objective, but not for the purposes of the present study, leading to further exclusions even when all three CBCT scans had been performed.

characterizing 3D dento-alveolar movement in both uni- and bi-lateral CLP, and also assessing the bone volume required and actually grafted and its quality and quantity after graft consolidation. Ongoing standardization of radiology documents will contribute to achieving these objectives.

Conflicts of interest: The author declares no conflict of interest.

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