## UNIVERSITE DE LAUSANNE - FACULTE DE BIOLOGIE ET DE MEDECINE

Département des services de chirurgie et d'anesthésiologie Division de chirurgie maxillo-faciale

## Influence of the primary cleft palate closure on the future need for orthognathic surgery in unilateral cleft lip and palate patients

## THESE

## préparée sous la direction du Professeur Bertrand Jaques

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Influence of the primary cleft palate closure on the future need for orthognathic surgery in unilateral cleft lip and palate patients

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Madame le Professeur Stephanie Clarke Directrice de l'Ecole doctorale

#### RESUME

**Introduction :** L'objectif de cette étude est de déterminer l'influence de la dissection du palais lors de la chirurgie primaire et le type de chirurgie orthognathique requise chez les patients porteurs d'une séquelle de fente labio-maxillo-palatine unilatérale complète

**Méthode :** Cette revue porte sur 58 enfants nés avec une fente labio-maxillo-palatine complète unilatérale et traités entre 1994 et 2008 à l'âge approprié pour une chirurgie orthognathique. C'est une étude rétrospective longitudinale mixte. Les patients avec des syndromes ou anomalies associées ont été exclus. Tous les patients ont été traités par le même orthodontiste et par la même équipe chirurgicale. Les enfants sont divisés en deux groupes : le premier comprend les patients avec une chirurgie primaire du palais conventionnelle, avec un décollement extensif de la fibro-muqueuse palatine. Le deuxième groupe comprend les patients opérés selon le protocole de Malek. Le palais mou est fermé à l'âge de trois mois, le palais dur à l'âge de six mois, avec un décollement minimal de la fibro-muqueuse palatine.

Les radiographies du crâne de profil ainsi que les données chirurgicales ont été comparées.

**Résultats :** La nécessité d'une chirurgie orthognathique est plus élevée dans le premier groupe par rapport au deuxième (60% versus 47,8%). Concernant le type de chirurgie orthognathique réalisé, des ostéotomies Lefort I en deux ou trois pièces ou des ostéotomies bi-maxillaires ont aussi été plus fréquentes dans le premier groupe

**Conclusion :** La chirurgie primaire du palais selon le protocole de Malek améliore le pronostic des patients avec une fente labio-maxillo-palatine. Avec un décollement minimal de la fibro-muqueuse palatine, le nombre d'interventions de chirurgie orthognathique a été diminué. Lorsque ces opérations étaient néanmoins indiquées, elles étaient simplifiées.

## ARTICLE

#### Introduction

Impairment of maxillary growth resulting in retrusion of the maxilla is a frequent finding in children born with cleft lip and palate. To correct these dentofacial deformities, orthognathic surgery may therefore be indicated [1]. A maxillary advancement with a Le Fort I osteotomy is the most common orthognathic procedure. Due to a possible transversal collapse of the maxillary arches on each side of the cleft, caused by the scar tissue, this advancement cannot always be achieved in one piece. In these cases, the maxilla has to be segmented in two or three pieces. The frequency of indications for a Le Fort I osteotomy in unilateral cleft lip and palate (UCLP) in the literature varies from 22% to 48.3% [2-5]. These differences may arise from different management protocols and depend also on the patient's access to adequate pre-surgical orthodontic care. Criteria used to determine the need for orthognathic surgery are also subjective to some extent, and therefore may vary between surgical teams.

Since 1989, all patients born with a UCLP have been operated in our hospital following the Malek protocol [6]. The soft palate is closed at three months of age, the hard palate and the lip at six months. During these surgeries on the palate, special care is taken to avoid elevation of the mucoperiosteum. As a result, there is only a small area of denuded palatal bone, lateral to the incisions on the palate that is left to heal by secondary intention. When compared with the classical techniques, where large areas of palatal bone are left to heal by secondary intention, the amount of scar tissue on the palate is thus greatly reduced (Fig. 1)

The aim of our study is to compare the need and type of orthognathic surgery in our children born with UCLP and treated following the Malek procedure and in those whose palate was closed with an extensive elevation of the mucoperiosteum.

#### Method

A review was performed of all children born with UCLP and evaluated for orthognathic surgery between 1994 and 2008. Children with incomplete records or for whom there was no follow-up were excluded. Children born with associated anomalies were also excluded. The required records were a description and timing of initial surgery and lateral cephalograms at ages nine and 16. This was considered to be a reasonable age to evaluate the need (or not) for orthognathic surgery. All orthodontic treatments were performed by the same orthodontist, and orthognathic surgery planned by the same maxillo-facial surgeon. The cephalometric radiographs were traced and analyzed using Quick Ceph computer software (Quick Ceph Systems, Inc.; 9883 Pacific Heights Blvd., Suite J; San Diego, CA 92121; USA). Children were divided into two groups, depending on the technique used during primary surgery for palate closure. In the first group, palate closure was realized with a large dissection of the mucoperiosteum before the age of 12 months. In the second group, children were operated first at three months for the soft palate closure and then at six months for the hard palate and lip closures following the Malek procedure [6]. The dissection of the palate was restricted to a minimum, leaving two thin lateral areas of the palate denuded and healing spontaneously by secondary intention.

An objective determination of the need for orthognathic surgery was based on the data available from the analysis of the lateral cephalograms: the anteroposterior relationship of the maxillary basal arch to the anterior cranial base uses the SNA, SNB, ANB angles (S = sella, N = nasion, A = subspinale, B = supramental); anteroposterior jaw dysplasia may be measured with the Wits appraisal (perpendiculars from points A and B onto the occlusal plane), and the distance from the upper lip to the e-plane (line drawn from the tip of the nose to the chin) were the most often used criteria. They were however not exclusive. Children with poor facial aesthetics despite a more favourable lateral cephalogram were also considered for an orthognathic correction.

#### Results

**Group I:** Thirty-five children born with UCLP were operated with extensive undermining of the palate. Sixty percent (21) (Fig. 2) met the above-mentioned criteria for an orthognathic procedure (Table 1). Of these twenty-one children, 8.6% (3) had a standard Le Fort I osteotomy; 25.7% (9) had a two or three pieces Le Fort I osteotomies; 22.8% (8)had a Le Fort I associated with a bilateral sagittal split osteotomy (BSSO) and 2.8% (1) had a transverse expansion by means of a distraction osteogenesis. (Fig. 3)

**Group II:** Twenty-three children born with UCLP were operated with minimal undermining of the palate mucosa. Orthognathic surgery was judged necessary in 47.8% (11) (Fig. 2) children (Table 2). 26.1% (6) needed a standard Lefort I, 13% (3) patients two or three pieces Le Fort 1; 4.4% (1) a Lefort 1 associated with a BSSO and 4.4% (1) a transverse distraction osteogenesis (Fig. 3).

#### Discussion

Our study shows that primary palate surgery in UCLP children following the Malek procedure results in an improved and simplified craniofacial outcome compared to a large primary surgical dissection of the palate.

Cleft lip and palate children often have midfacial growth deficiency, with a reduced upper lip support, giving a characteristic concave profile. This generally increases during adolescence. For a few authors, these growth disturbances are intrinsic to the cleft itself, as it was observed in children who were never operated for their cleft. [7-9]. However, these studies have been questioned [10] and for many authors, maxillary growth deficiency is mainly iatrogenic in nature and a consequence of the primary surgical repair of the palate [11,12]. Liao et al. [12] compared the follow-up of children born with UCLP and operated only for their lip with children born with UCLP and palate; he concluded that primary surgery on the palate was the main cause of maxillary retrusion.

Orthognathic surgery to correct facial disharmony is part of the normal followup of children born with UCLP. When planning corrective surgery, many factors are taken into account, such as facial profile, intermaxillary discrepancies and dentoalveolar relationship. Unfortunately, as there are no standardized criteria, the decision often remains subjective and this can explain the difficulty to compare results between different centres [4,5]. Ross and al, in 1995 [13] compared lateral cephalograms between UCLP children operated for their palate by different techniques and showed very similar results and measurements between children whose cleft was closed following the Malek procedure and children whose cleft was closed following a conventional technique. His study, however, did not include the surgical outcome. Figures 4 and 5 show two children operated for a Le Fort 1. In figure 4, the palate was closed during the first year of life with an extensive undermining of the mucoperiosteum. The central incisor is severely reclined, increasing the impression of maxillary retrusion. In figure 5, the palate was closed following the Malek procedure. The measurements derived from a comparison of maxillary growth impairment are similar in both children even if the profile of the second child is clearly improved.

The number of our children who had undergone surgery was high (60% and 47.8%). In the second group, the rate of surgical orthognathic procedure needed was similar to recent reports in the literature: Good et al. [4] reported 46% and Daskalogiannakis [5] (2009) 48.1%. Access to pre-surgical orthodontic treatment, adequate healthcare coverage and our preference for operative corrections for aesthetics or skeletal malpositions can explain these numbers. The demand of patients for an improved profile also seems to have increased over the last few years. Secondary alveolar bone grafting, usually performed in the mixed dentition, to enable the canines to erupt under good conditions, were performed in both groups by the same surgeon, using the same technique and following the same schedule. We therefore considered that this procedure would not interfere with our compared

results. Besides, several reports confirm that bone grafting does not impair maxillary growth [14, 15].

Finally, the most significant finding in our retrospective study concerns the type of orthognathic procedure performed. In the first group, most children underwent a more complex procedure (Table 2). Two or three pieces Lefort I osteotomies were done to correct discrepancies in the transversal as well as in the sagittal planes. This may be explained by an increase in the amount of scar tissue lateral to the incisions performed during the primary closure of the palatal cleft. In the children of the second group, we also noted a significant reduction in the frequency of indications for bimaxillary osteotomies. This finding reveals an overall improvement of the facial profile of our patients.

#### Conclusion

Over the last 20 years, all primary closures of the palate in UCLP children have been done with a minimal undermining of the mucoperiosteum. When we compare the follow-up of children who underwent a classical surgical repair of the palate with that of children operated following the Malek procedure, we find a clear reduction in the frequency of orthognathic osteotomies indications and also a less complex procedure when surgery was needed. Despite the relatively small sample size, the differences between the two groups allow us to conclude that our current protocol has improved the craniofacial outcome of our patients.

#### References

- [1] Chigurupati R. Orthognathic surgery for secondary cleft and craniofacial deformities. Oral Maxillofac Surg Clin North Am 2005; 17(4):503-17.
- [2] Ross RB. Treatment variables affecting facial growth in complete unilateral cleft lip and palate. Cleft Palate J 1987 Jan; 24(1):5-77.
- [3] DeLuke DM, Marchand A, Robles EC, et al. Facial growth and the need for orthognathic surgery after cleft palate repair: literature review and report of 28 cases. J Oral Maxillofac Surg 1997; 55(7):694-7.
- [4] Good PM, Mulliken JB, Padwa BL. Frequency of Le Fort I osteotomy after repaired cleft lip and palate or cleft palate. Cleft Palate Craniofac J 2007; 44(4):396-401.
- [5] Daskalogiannakis J, Mehta M. The need for orthognathic surgery in patients with repaired complete unilateral and complete bilateral cleft lip and palate. Cleft Palate Craniofac J 2009;46(5):498-502.
- [6] Malek R, Psaume J. New concept of the chronology and surgical technic in the treatment of cleft lip and palate. Ann Chir Plast Esthet 1983; 28(3):237-47.
- [7] Bishara SE, Jakobsen JR, Krause JC, et al. Cephalometric comparisons of individuals from India and Mexico with unoperated cleft lip and palate. Cleft Palate J 1986; 23(2):116-25.
- [8] Isiekwe MC, Sowemimo GO. Cephalometric findings in a normal Nigerian population sample and adult Nigerians with unrepaired clefts. Cleft Palate J 1984;21(4):323-8.

- [9] Shetye PR. Facial growth of adults with unoperated clefts. Clin Plast Surg 2004; 31(2):361-71.
- [10] Will LA. Growth and development in patients with untreated clefts.Cleft Palate Craniofac J 2000; 37(6):523-6.
- [11] Capelozza Filho L, Normando AD, da Silva Filho OG. Isolated influences of lip and palate surgery on facial growth: comparison of operated and unoperated male adults with UCLP. Cleft Palate Craniofac J 1996; 33(1):51-6.
- [12] Liao YF, Mars M. Long-term effects of palate repair on craniofacial morphology in patients with unilateral cleft lip and palate. Cleft Palate Craniofac J 2005; 42(6):594-600.
- [13] Growth of the facial skeleton following the Malek repair for unilateral cleft lip and palate. Ross, R.B. Cleft Palate Craniofac J 1995; 32(3): 194-8.
- [14] Daskalogiannakis J, Ross RB. Effect of alveolar bone grafting in the mixed dentition on maxillary growth in complete unilateral cleft lip and palate patients. Cleft Palate Craniofac J. 1997; 34(5):455-8.
- [15] Levitt T, Long RE Jr, Trotman CA. Maxillary growth in patients with clefts following secondary alveolar bone grafting. Cleft Palate Craniofac J 1999; 36(5):398-406.

### Tables

# Table 1: Group I average cephalometric data of operated and nonoperated children

		Operated patients (14)	Standard deviation	Non operated patients (21)	Standard deviation
SNA	(deg)	73.7	5.2	77.2	4.9
SNB	(deg)	74.8	5.2	74.1	5.2
ANB	(deg)	-1.1	3.2	3.1	3.7
Wits	(mm)	-4.65	5.7	3.3	4.6
Upper Lip E-plane	(mm)	-8.7	3.5	-4,9	3.5

Table 2: Group II average cephalometric data of operated and non-operated children

		Operated patients (12)	Standard deviation	Non operated patients (11)	Standard deviation
SNA	(deg)	75.0	3.9	75.1	2.6
SNB	(deg)	75.8	2.6	73.7	3.1
ANB	(deg)	-0.8	2.6	1.4	1.7
Wits	(mm)	-3.3	3.8	-0.1	3.0
Upper Lip E-plane	(mm)	-8.4	3.4	-6.8	3.4

## Figures

**Fig. 1.** Example of an UCLP child with a palate closed with a minimal mucoperiosteum undermining, and no lateral palatal scarring tissue



**Fig. 2**. Comparison between the total amount of orthognathic procedures between group 1 and group 2



**Fig. 3**. Comparison between the types of orthognathic procedures performed in the group 1 and group 2



Fig. 4. Lateral cephalogram of a group 1 patient, with a maxillary retrusion and severe impact on profile



**Fig. 5**. Lateral cephalogram of a group 2 patient. There is a similar maxillary retrusion, but a diminished impact of profile

