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Repositionnement chirurgical du prémaxillaire :
Incidence, indications et étude de la croissance faciale
chez 189 patients porteurs de fente labio-palatine bilatérale

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Surgical repositioning of the premaxilla: incidence, indications and growth study about a 189 bilateral cleft lip and palate population

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Abstract

Introduction: For several decades, the management of mispositioned premaxilla in bilateral cleft lip/palate (BCLP) has been a complex question with multiple answers. Surgical repositioning strategies often stray from standard orthodontic-surgical protocols and remain controversial. The aim of our study was to evaluate the rate of BCLP patients requiring surgical repositioning of the premaxilla (SRP), SRP indications and further facial growth over a 40-years period in a tertiary cleft center. **Methods:** This was a retrospective, single-center cohort study including bilateral cleft patients born between 1980 and 2019 treated in Nantes Oral and Maxillofacial Surgery department. Surgical indications and age at surgery were collected from SRP patients' medical records. Post-operative growth was evaluated on lateral cephalometries and occlusal views (at least until adolescence). Maxillary arch, maxillary and mandibular position relative to the skull base, facial convexity and Delaire cephalometric cranio-facial analysis were retrieved. **Results:** Over the whole period, 189 patients with BCLP were identified. Three patients underwent SRP, all during their primary dentition period. The indication for surgical repositioning was always premaxilla vertical overgrowth with an overbite over 10 mm. Facial growth features in the three patients were mostly comparable with a BCLP population who had no premaxillary surgery. **Discussion:** Our results showed a low incidence of SRP. No SRP was necessary during early infancy before lip repair, or during adulthood. Surgical repositioning of the premaxilla appeared to offer acceptable facial development compared with other BCLP subjects.

Introduction

Orofacial clefts (including cleft lip, cleft palate and cleft lip with cleft palate) are the most common congenital craniofacial malformations (1 in 600 births) (1). Almost ten percent are bilateral. Complete bilateral clefts are the most severe types because of their aesthetic and functional consequences. Particularly, patients with cleft lip and palate are at risk for maxillary underdevelopment. Preserving patient's growth potential is then an everyday challenge.

In complete BCLP, the premaxilla is only attached to the rest of the face by the nasal septum and the vomero-premaxillary suture (VPS). This situation may induce early misposition of premaxilla during embryogenesis or later, during infancy (2). In BCLP, early release of the premaxilla growth potential is due to the lack of premaxilla retention by lateral segments and *orbicularis oris* muscle. The VPS growth potential is then early released and favored by the tongue movements.

Misposition of the premaxilla leads to unaesthetic appearance and could result in psycho-social trouble (3,4). Traumatic injury risk (5), oncoming speech, chewing troubles and traumatic periodontal recession are observed (6). In great misposition, alveolar bone grafting can be challenging (7). Great premaxilla offset may also lead to an accentuation of the main growth pattern of BCLP patient described in previous study (3,8). This BCLP growth pattern includes maxillo-mandibular retrusion, steep mandibular slope and anterior vertical overdevelopment of the inferior third of the face. This is more likely to result in unbalanced facial profile.

Orthopedic/orthodontic and surgical options have been described to reposition the premaxilla (9).

Presurgical orthopedics (PSO) have been mainly represented by the Millard-Latham protocol performed from 1980 to 1996 in Miami (4). The protocol associated bodily retraction of the premaxilla around 2 months old with a fixed palatal orthopedic appliance followed by a

gingivo-perisosteoplasty (GPP) and a lip adhesion. The aim of this treatment was to ensure good facial and aesthetic results earlier in life, but it led to worse dento-facial results than former conservative functional protocol (4). Further types of PSO, improving columella lengthening called naso-alveolar molding (NAM) have been developed (10,11). Their long-term superiority to protocol without PSO hasn't been documented yet. NAM presents several limits including an elevated cost, the need for a high level of patient compliance, as well as the medical staff availability.

In order to relocate the premaxilla in a proper position during late childhood (between 7 and 11 y.o), Liou *and al.* described an orthopedic appliance to correct premaxilla vertical overgrowth before secondary alveolar bone grafting (12). In 2010, Meazzini *et al.* suggested a protocol to correct excessive vertical offset of the premaxilla. The authors recommended an orthopedic treatment based on Liou's technique or surgical repositioning depending on various criteria (type of dentition, previous alveolar bone grafting, amount of vertical growth) (13). These 2 studies dealing with downward displaced premaxilla didn't evaluate mid- or long-term side effects of vertical repositioning.

Concerning surgical repositioning of the premaxilla, the little epidemiology in the literature suggests SRP indications varies a lot from one cleft center to another. The incidence of SRP varied from no SRP in a 40-year period to 50 cases in 31 months (3,7,14,15).

Four main ages groups have been reported for SRP with varying decision criteria. During infancy, SRP is performed to allow primary lip closure and to avoid excessive wound tension (15,16). The surgical option is often selected when PSO (including NAM) are not technically or financially possible (17). During early childhood, SRP is performed to avoid psycho-social (3,18) and traumatic troubles (5) related to the premaxilla protrusion. During late childhood (after 8 y.o.), the goals are the same but the surgery is postponed in order to minimize the maxillary growth impact (6,8) and favor alveolar bone grafting (7). In adults, the objective is to achieve both functional and aesthetic results. Therefore, the premaxilla can be moved by the

means of a three pieces Le Fort 1 osteotomy as suggested by Freihofer *et al.* (19). This technique avoids any growth impairment related to early SRP and allows further orthodontic treatment in patients presenting with maxillary retrusion and premaxilla vertical offset.

Benefits of the surgery have to be balanced with the risk of complication such as premaxilla necrosis and pseudoarthrosis. Secondary growth impairment has also been reported in long-term follow-up studies. Vargervik compared 12 early setbacks to BCLP patients without setback (20). All SRP patients demonstrated severe midfacial retrusion with slower forward growth than unoperated BCLP patients. Friede and Pruzansky evaluated maxillary growth features in 13 patients requiring primary setbacks during infancy or at preschool years (age ranged from 3 to 7 y.o.) in comparison with control BCLP. The 2 setback groups demonstrated retrusion of the midface (21). On the opposite, after comparing maxillary features in an early, late and control groups, Padwa concluded that SRP could be done before growth completion “without compromise” (22). Marinho *et al.* reported a setback group of 10 BCLP patients operated in late childhood (mean 10,2 y.o) and followed up until late adolescence (mean 16,6 y.o.), the patients were compared to data from the Oslo Cleft Lip and Palate Archive (8). Although more concave profile were obtained in this setback study, this result wasn’t considered as an overall adverse effect on growth (8).

In our experience, neither orthopedic nor surgical sagittal repositioning of premaxilla is usually advocated. Delaire’s cleft repair technique implies a wide periosteal elevation allowing closure of the lip without excessive wound tension (23). Sagittal SRP is usually not performed during childhood, since the protrusive premaxilla in BCLP is known to spontaneously recede with growth (3,20).

Although sagittal protrusive premaxilla is generally corrected with further facial development, downward displaced premaxilla is not known to recede. Furthermore, excessive overbite may lead to orthodontic issues implying worsening of the mandibular growth pattern and long-lasting adverse outcomes in social integration. Vertical repositioning would then be required.

The aim of our study was to evaluate the rate of BCLP patients requiring SRP, SRP indications and further facial growth in a tertiary cleft center.

Methods

This was a single-center, descriptive, retrospective cohort study. This research received an approval from the ethics committee of Nantes University Hospital (2020 September 10th).

Participants

Patients born between 1980 and 2019 who had undergone cleft surgery at Nantes Hospital for bilateral cleft lip (CL), cleft lip and alveolus (CLA) and/or cleft lip and palate (CLP) were included.

Population characteristics

Patients' gender, syndrome and type of cleft were retrieved. Operative reports were screened to identify patients with SRP (including all Le Fort 1 osteotomies). SRP patients were selected for further analysis.

SRP indication and surgical schedule

The preoperative lateral cephalograms were analyzed to identify the type of premaxilla misposition. Incisal overbite and overjet were noted. Overbite was defined as the distance between each incisal edge and the occlusal plane. The latter was defined as the line joining the distal cuspid of the second premolar and the mesial cuspid of the first premolar. In temporary dentition, we determined the occlusal plane with the deciduous molars. The overjet was defined as the distance between the orthogonal projection of the upper and lower incisors edge on a perpendicular plane to the occlusal plane.

Operative criteria and surgical schedules were retrieved from medical record. We looked for early post-operative side effects: pseudoarthrosis, infection and premaxilla necrosis (7).

Facial growth study

Maxillary arch length (Pti-A'), alveolar maxillary position relative to the cranial base (SNA), alveolar maxilla-mandibular sagittal relationship (ANB) and facial convexity (NAPog) were retrieved on the preoperative cephalogram and on each cephalogram available after surgery (Figure 1).

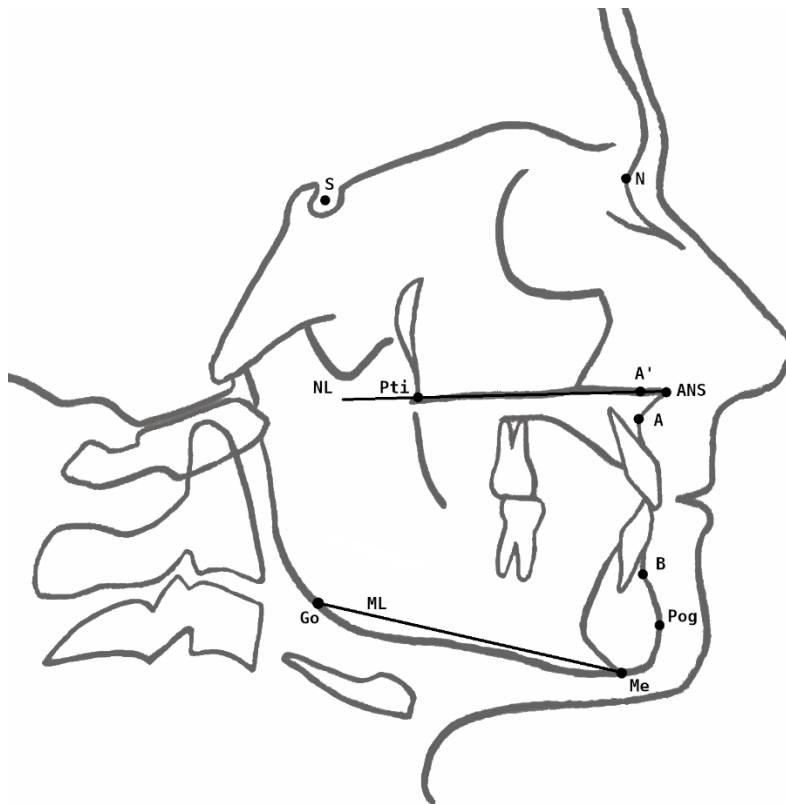


Figure 1: cephalometric landmarks and linear measurements

S: sella, N: nasion, A: A point, B: B point, Pog: pogonion, Pti: lower point of the pterygomaxillary suture, Me: menton, Go: gonion

The following linear and angular measurements were retrieved from lateral cephalograms: maxillary arch length (Pti-A'), maxillary sagittal position relative to the cranial base (SNA), upper-lower jaw alveolar relationship relative to nasion (ANB), facial convexity (NAPog), nasal line to mandibular line angle (NL^ML).

A Delaire cranio-facial analysis was performed on the most recent lateral cephalogram on dedicated software (Delaire 2015 Evolutions, JDEL, Nantes, France) to assess maxilla-mandibular relationships and jaws position relative to the optimal facial growth pattern as described by Delaire (24). Oslo complete-BCLP (c-BCLP) population was selected to compare

Pti-A', SNA, ANB, NAPog and NL^ML parameters during growth. Ninety-nine percent Student confidence intervals (CI) were generated to determine likelihood for our patients' values to belong to Oslo population (3). Oslo's 90 c-BCLP patients followed an orthodontic/surgical schedule close to ours. Especially, no presurgical orthopedic and no primary premaxilla setback was performed, resulting in a conservative functional protocol. Furthermore, Oslo c-BCLP population was described with the parameters enumerated above, available in all sex and age subgroups until early adulthood, allowing analysis even before growth completion.

All occlusal views in medical records were screened for each patient. Torsion of the premaxilla was reported from preoperative occlusal views. Anterior crossbite and incisal end-to-end occlusion were noted. Most recent occlusal features were reported: angle class, anterior or posterior crossbite, over or open bite, management of missing upper lateral incisors.

Results

Population characteristics

One hundred eighty-nine BCLP patients were operated at Nantes university hospital for a bilateral cleft lip/palate between 1980 and 2019 (table 1). No patient had SRP before lip surgery, 3 patients had SRP during early childhood (before 8 y.o). SRP decision criteria were difficulty to vertically realign premaxilla with lateral segment for all patients. No patient had SRP during late childhood and no patient had SRP (by the means of 3 pieces Le Fort 1 osteotomy) in adulthood.

Table 1. Population characteristics

BCLP	M F	Sex Ratio	Syndrome	Total
CL	11 2	5,5	0	13
CLA	8 7	1,1	0	15
CLP	106 55	1,9	22*	161
All BCLP	125 64	2,0	22	189

BCLP: bilateral cleft lip and palate, CL: cleft lip, CLA: cleft lip and alveolus, CLP: cleft lip and palate; M: male, F: female.

*7 Smith-Lemli-Opitz syndromes (SLOS), 4 Ectrodactyly Ectodermal Dysplasia Cleft lip/palate syndromes (EEC), 1 Goldenhar syndrome, 1 isolated GnRH syndrome, 1 branchio-oculo-facial (BOF) syndrome, 2 Van der Woude Syndromes, 1 fragile X-syndrome (FXS), 5 unclassified polymalformative syndromes.

Premaxilla misposition and SRP protocol

Preoperative overbite and overjet are reported in table 2. Patients with SRP had all more than 10 mm overbite. Overjet varied from 3 to 22 mm. Patient 1 presented mild torsion of the premaxilla (less than 30°). SRP patients were all in decidual dentition at the time of surgery. For patient no. 1, SRP was performed after primary GPP. Patient 2 required GPP and tibial

cancellous bone grafting 6 months after the SRP for pseudoarthrosis. The surgical schedule is detailed in table 2.

Table 2. SRP procedure and global surgical schedule

	Patient no. 1	Patient no. 2	Patient no.3
SRP			
Sex	Male	Male	Female
Pre-SRP overbite (mm)	13	12	11
Pre-SRP overjet (mm)	9	22	3
Pre-SRP torsion (degrees)	<30	No	No
Age at SRP (y-m)	5-3	2-7	5-2
Early complication	/	Pseudoarthrosis	/
Age at last follow up (y-m)	19-4	15-0	12-7
Surgical schedule			
Delaire's primary cheilo-rhinoplasty and veloplasty (25)(months)	6	5	7
Uranostaphyloraphy (months)	18	18	16
SRP (y-m)	5-3	2-7	5-2
Primary GPP (y-m)	3 -0 : Unilateral	3-5 : Bilateral	6-7 : Bilateral + ABG
Secondary veloplasty (y-m)	3-0 : Palatal fistula closure	4-6 : Palatal fistula closure	No
Pharyngoplasty (y-m)	No	No	No
Secondary GPP (y-m)	12-8 + ABG	No	No
Collumela lengthening (y-m)	12-8	6-4	9-6
Le Fort 1 osteotomy (y-m)	No (Required)	No (not required)	No (not required)

SRP : Surgical repositioning of the premaxilla, GPP : Gingivo-peristeoplasty, ABG : Alveolar Bone Grafting, y : years, m : months

Facial growth study

Maxilla relative position to the cranial base: all patients showed a trend to SNA decrease.

Absolute value at the end of the follow-up was within the 99% Oslo CI for patient no. 2. Patients no. 1 and 3 were out of their 99% Oslo CI. **Maxillary arch length:** the 3 patients had a slight Pti-A' downward trend. Final data were within the 99% Oslo CI for all patients. **Maxillo-**

mandibular relationship: ANB decreased in the 3 patients. Final values were within the 99% Oslo CI. **Facial Convexity:** Starting with fairly convex facial profile, all patients tended to develop concave facial profile. In Patients no. 1 and 3, NAPog values were within the 99% CI while patient no. 2 was beyond maximum value. **Mandibular slope:** patient no. 1 had a final $NL^{\wedge}ML$ value within the CI. Patients no. 2 and 3 were below. Details of the measures are presented in figure 2 and table 3.

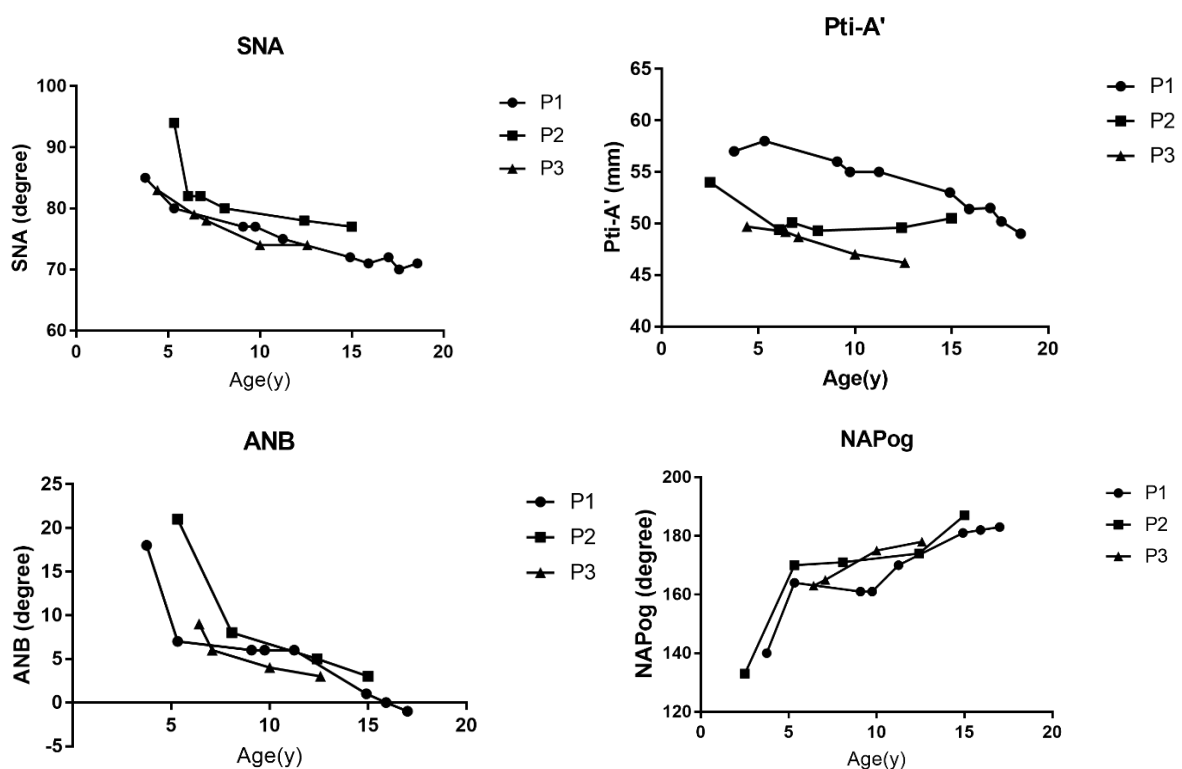


Figure 2 : SNA, Pti-A', ANB, NAPog evolution during follow-up.

Pti-ANS : maxillary arch length, SNA : maxillary sagittal position relative to the cranial base, ANB : maxilla-mandibular alveolar relationship relative to nasion, NAPog : Facial convexity, $NL^{\wedge}ML$: angle between the nasal line and the mandibular line angle.

Delaire cranio-facial analysis: Patient no. 1 displayed maxillo-mandibular retrusion with skeletal class III, without maxillary or mandibular vertical offset. No anomaly of anterior facial height was noted. The occlusal plane was fine. Patient no. 2 showed no sagittal jaw discrepancy resulting in a skeletal class I. Maxillary and mandibular heights were slightly insufficient. A counter-clockwise rotation of the occlusal plane was noted. In patient no. 3, a

balanced maxilla associated with a sagittal retrusion of the mandible resulted in a skeletal class II. There was no anomaly of the anterior facial height. The occlusal plane was satisfying. Delaire analysis drawings are available in figure 3.

Occlusal features: No patient presented anterior crossbite during their follow-up. Patient no. 1 and 2 both presented with Angle class I. Patient no. 3 presented an Angle class II. None of our patients have had lateral incisor space closure.

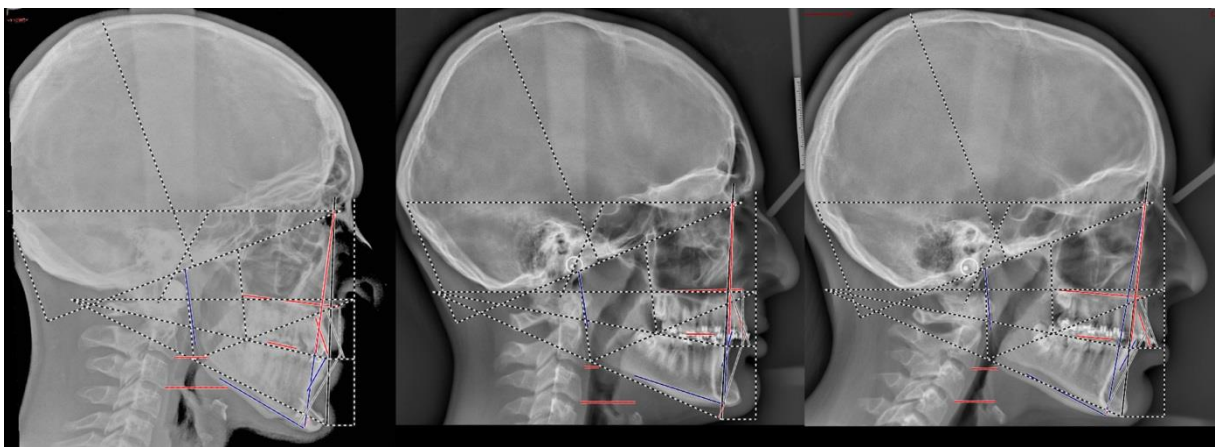


Figure 3: Patient no. 1, 2 and 3 Delaire analysis drawings (from the left to the right)

Red lines represent the skeletal maxillary reference line of the patient (except for the lower horizontal lines that indicate spinal-mandibular relationship), the blue lines represent the skeletal mandibular reference lines of the patient the dashed lines are the corresponding (maxillary and mandibular) optimal lines according to Delaire cranio-facial cephalometric analysis. The vertical black line represents the optimal location of anterior facial pillar (24).

Table 3. Facial growth characteristics at the last follow-up

	Patient		Patient		Patient	
	no. 1		no. 2		no. 3	
Growth characteristics						
Pti-A' (mm) [Oslo CI]	49	[45,7-50,5]	50,5	[47,1-50-9]	46,2	[45,4-50,23]
SNA (degrees) [Oslo CI]	72*	[72,9-77,3]	77	[75,5-79,5]	74*	[74,1-80,5]
ANB (degrees) [Oslo CI]	-1	[-1,46-1,86]	3	[0,03-3,77]	3	[0,57-4,83]
NAPg (degrees) [Oslo CI]	183	[177,9-190,1]	187*	[175,9-184,1]	178	[172,8-183]
NL^ML (degrees) [Oslo CI]	26	[21,2-30,6]	16*	[23,6-29,6]	20*	[22,9-33,3]
Delaire cranio-facial Analysis						
Skeletal Class	Class I		Class I		Class II	
Maxilla sagittal position	Retrusive		Balanced		Balanced	
Mandible sagittal position	Retrusive		Balanced		Retrusive	
Maxilla vertical position	Balanced		Insufficient		Balanced	
Mandible vertical position	Excessive		Insufficient		Balanced	
Occlusal plane	Balanced		Counter-clockwise rotation		Balanced	
Occlusal features						
Anterior crossbite during growth	No		No		No	
Angle class	Class I		Class I		Class II	
Orthodontic upper lateral incisors space closure	No		No		No	

*Values out of the confidence interval, SRP : Surgical repositioning of the premaxilla, Pti-ANS : maxillary arch length, SNA : maxillary sagittal position relative to the cranial base, ANB: maxilla-mandibular alveolar relationship relative to nasion, NAPog: facial convexity, NL^ML : angle between the nasal line and the mandibular line, [Oslo CI] : corresponding Oslo 99% confidence interval.

Discussion

The main objective of this study was to evaluate the frequency of SRP among a BCLP population in a tertiary cleft center. With only 3 SRP among 189 BCLP over a 40 year-period, it represented a low incidence rate of that procedure (3,7,14,15). In previous studies dealing with SRP decision criteria and growth consequences, the indication was always an excessive sagittal protrusion of the premaxilla, whatever the age at surgery (8,20,21). In our study the decision criterion was always a vertical offset of the premaxilla. While the protrusive premaxilla is known to recede over time with the growth of the lateral segment (3,20), vertical excess is not believed to correct spontaneously during childhood. The vertical overgrowth of premaxilla is thought to be a rare complication of the primary functional cheilo-rhinoplasty. When the lip cannot stunt the premaxilla's forward growth, the VPS growth potential is thought to be released downward and forward. The influence of individual premaxillary growth pattern has to be considered (21), likely to result in more downward than forward misposition.

The 3 studied patients presented a great premaxilla vertical offset (more than 10 mm overbite) with aesthetic and oncoming growth consequences. Since great vertical excess does not recede over time, achieving good orthodontic treatment remains unlikely. Then, mandibular growth tends to be locked forward and an anterior vertical excess of the lower third of the face [i.e. accentuation of the general steep mandibular slope of the BCLP growth pattern (3,8)] would more likely occur. Great premaxillary vertical offset may also lead to periodontal trauma (6) and the risk of dental and bony fracture has to be taken into account (5). In these rare cases of important vertical excess, current orthopedic/orthodontic treatments were not suitable to avoid SRP. Liou's appliance, described later, could have been a relevant alternative. The orthopedic intrusion is supported by bony contraction at the VPS, alveolar shortening and incisor

ingression (12). However, only 8 patients from 7 to 11 years old were treated and the follow up was stopped 1 year postoperatively. Maxillary growth and dental development were not evaluated in this study. In a general manner, no long-term data after growth completion are available, regarding both surgical and orthopedic repositioning of the premaxilla.

Even if it was not possible to assess the immediate postoperative premaxilla position due to the lack of radiological data, we can assume that vertical SRP was associated with a setback movement in two of our patients with important overjet (22 mm in patient no. 2 and 9 mm in patient no. 1). Indeed, a decrease in SNA and ANB between preoperative and post-operative times were noted (especially in patient no. 1 and 2). The decrease may be attributed to 3 factors: 1/ the counterclockwise rotation of the premaxilla to correct the incisors palatal position is likely to move point A backward relative to the cranial base; 2/ because of the oblique direction of the premaxilla, the amount of bone resection inevitably leads to a various amount of setback; 3/ some setback may have been decided during surgery in order to limit the postoperative risk of pseudoarthrosis by approaching the dental arch continuity.

Anterior crossbite after premaxillary setback has already been reported as a witness of early growth impairment (15). In the present study, no anterior crossbite occurred during follow-up and final occlusal features were in Angle class 1 or 2 without crossbite. This suggests that operated patients did not present a major growth impairment. It is to be noted that the absence of orthodontic lateral incisor space closure eliminates a potential confusion factor in the sense that orthodontic space closure is more likely to worsen interdental relationships.

Oslo BCLP population was overall comparable to our population excepted for the need of SRP. This was the only study reporting an important sample of c-BCLP (90 patients) with a lot of facial parameters available for each gender and age. This was determinant to allow growth impairment evaluation in patients who didn't reach full skeletal maturity. Our SRP patients' growth parameters were mainly within the 99% CI from Oslo c-BCLP population. Only 3 out

of 15 measures were likely to be worse than the Oslo population. This was consistent with the occlusal relationships found in our patients which didn't reveal major growth restriction.

Furthermore, we observed in patients 2 and 3 favorable values for NL^ML out of the Oslo population CI. This variation may be attributed to their individual growth pattern. Although a general BCLP growth pattern was observed in c-BCLP by Semb in 1991 (3), Friede and Pruzansky noted great variation between each individual in BCLP population as well as in non-cleft population.(21). Although the absence of surgical repositioning would have significant consequences, there is some risks to consider in SRP during early childhood. First of all, premaxilla necrosis by vascular compromise is a rare but serious complication (7). Also, pseudoarthrosis is more frequent but can be treated with GPP and alveolar bone grafting depending on the cleft size. Whatever our maxillary growth values are close to Oslo BCLP population, the added risk of growth impairment in SRP is to be outweighed by the already established risk of growth restriction in cleft lip/palate patient. Indeed, Le Fort 1 osteotomy requiring is known to increase with cleft type severity (26).

Our study presents several limits. This was a retrospective single center study. The small sample of patients did not allow statistical comparison. Moreover, 2 of the 3 patients did not reach full skeletal maturity at the study date (patient 2: 15 y.o., patient 3: 12 y.o.). At last, due to the lack of available data, it was not possible to assess the premaxilla movements performed during the SRP procedure.

Conclusion

Surgical treatment of premaxilla vertical offset presents a favorable benefit / risk balance. The orthopedic appliance described by Liou could represent a relevant alternative to SRP. Further long-term follow up studies are needed to decipher between these two options.

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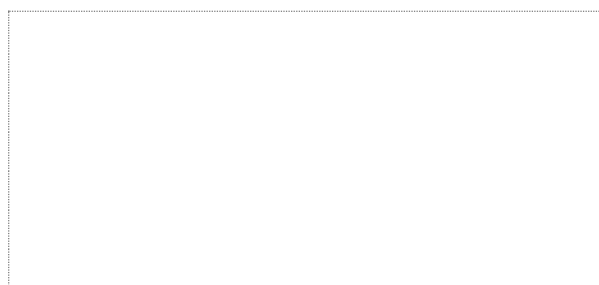
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Vu, le Président du Jury,
(tampon et signature)



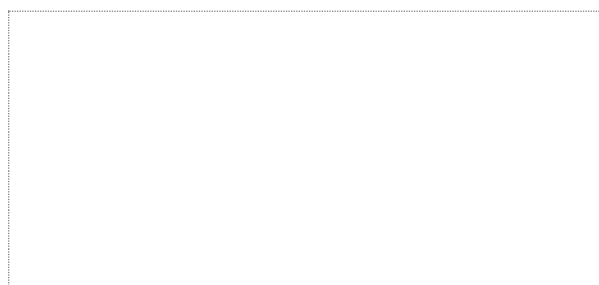
Professeur Philippe LESCLOUS

Vu, le Directeur de Thèse,
(tampon et signature)



Professeur Pierre CORRE

Vu, le Doyen de la Faculté,



Professeur Pascale JOLLIET

**Repositionnement chirurgical du prémaxillaire :
Incidence, indications et étude de la croissance faciale
chez 189 patients porteurs de fente labio-palatine bilatérale**

RESUME (10 lignes)

Les malpositions sévères du prémaxillaire dans les fentes labio-palatines bilatérales (FLPB) nécessitent le recours à des solutions orthopédiques ou chirurgicales dont les effets sur la croissance faciales sont encore mal connus. Le but de notre étude était d'évaluer la fréquence de recours au repositionnement chirurgical du prémaxillaire (RCP), ses indications et ses conséquences sur la croissance faciale. Cette étude rétrospective, monocentrique a inclus tous les patients opérés à Nantes d'une FLPB pendant 40 ans. L'indication de RCP (3 / 189 patients) était toujours un excès de croissance verticale en denture lactéale. Les données concernant la croissance étaient concordantes avec celles obtenues dans une population contrôle sans RCP. Bien qu'un effet sur la croissance ne puisse être éliminé, les conséquences en cas de non-intervention justifient le recours exceptionnel au RCP.

MOTS-CLES

Cephalometry
Child
Cleft Lip / physiopathology
Cleft Lip / surgery*
Cleft Palate / physiopathology
Cleft Palate / surgery*
Facial Bones / growth & development
Facial Bones / pathology
Humans
Mandible / growth & development
Mandible / pathology
Maxilla / growth & development
Maxilla / pathology
Maxilla / surgery
Maxillofacial Development
Skull / growth & development
Skull / pathology
Vertical Dimension