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Effects of growth on maxillary distraction osteogenesis in cleft lip and palate

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ABSTRACT

Purpose: The objective was to analyze the effects of growth on the long-term result of maxillary distraction osteogenesis (DO) in cleft lip and palate (CLP).

Patients and methods: Retrospective study of 24 CLP cases with long-term follow-up operated for maxillary DO using the Polley and Figueroa technique: 10 patients were distracted during growth, while 14 patients were operated after their growth spurt. Preoperative (T0), 6–12 months postoperative (T1), and ≥4 years postoperative (T2) cephalometric radiographs were evaluated. A classical cephalometric analysis was used to assess the treatment stability, and a Procrustes superimposition method was performed to assess local changes in the maxilla and the mandible.

Results: At T0, the mean age was of 11.9 ± 1.4 years for growing patient, and 17.9 ± 3.5 years for patient treated after their growth spurt ($P < 0.001$). Between T0 and T1, a greater increase of the SNA was shown in growing patients ($P = 0.036$), but the relapse was more important between T1 and T2, with a significant decrease of the SNA ($P = 0.002$) and ANB ($P = 0.032$) compared to the patients treated after their growth spurt. Although not significant, growing patients showed greater rotations of their palatal plane and mandibular plane.

Conclusions: Maxillary DO in CLP does not correct the growth deficit inherent to the pathology. Overcorrection of at least 20% is advised during growth.

1. Introduction

Maxillary hypoplasia leading to a class III skeletal relationship is a common developmental problem affecting patients with cleft lip and palate (CLP) (Ross, 1987; Linton, 1998). The treatment options for this deformity include conventional orthognathic surgery and internal or external distraction osteogenesis (DO) (Cohen et al., 1997; Polley and Figueroa, 1997; Karakasis and Hadjipetrou, 2004; Gedrange et al., 2006; Nada et al., 2010). Maxillary DO in CLP has been proven to have a good long-term stability (Figueroa et al., 2004; Rachmiel et al., 2005; Cho and Kyung, 2006; Aksu et al., 2010; Chua et al., 2010; Gürsoy et al., 2010; Chen et al., 2011), but it must be used judiciously for the right indications, so that its benefits can compensate its disadvantages (Cheung et al., 2006; Precious, 2007; Chua et al., 2012).

One advantage of maxillary DO in CLP is that it can be used in growing children to permit an early treatment of the severe forms of maxillary deficiency instead of waiting until skeletal maturity. Numerous authors reported good outcomes with the use of this interceptive DO (Polley and Figueroa, 1997, 1998; Cho and Kyung, 2006; Harada et al., 2006; Huang et al., 2007; Gürsoy et al., 2010; Chen et al., 2011). Unfortunately, the majority of these studies evaluated either a heterogeneous group of growing and non-growing patients, or a group comprised only of growing children, with no other group to compare them with. Therefore, it is not possible distinguish the relapse due to the distraction itself with the relapse due to the inherent growth deficit of the pathology.

The purpose of our study was then to evaluate the long-term effect of maxillary DO in growing CLP patients. Specific aims were to compare a group of growing CLP patients to a group of CLP patients treated after their growth spurt, and evaluate: (1) the

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difference in maxillary relapse, (2) the need for overcorrection; and (3) the differences on the mandible.

2. Material and methods

This retrospective study reviewed complete CLP patients treated with maxillary DO at the cleft and craniofacial and plastic pediatric surgery unit of the Lapeyronie Hospital, Montpellier, France. All of the consecutively treated CLP patients, between January 2001 and January 2008, were reviewed.

The inclusion criteria were: complete unilateral CLP (UCLP) or complete bilateral CLP (BCLP) patients with severe maxillary deficiency and class III malocclusion (requiring a horizontal maxillary advancement $\geq$ 7 mm), operated according to the same primary treatment protocol (Montoya et al., 2002) by the same surgeon, and according to the Polley and Figueroa external distraction technique (Polley and Figueroa, 1998). Cephalometric radiographs taken preoperatively (T0), 6–12 months postoperatively (T1), and more than 4 years postoperatively (T2) were required for the chart to be complete (Fig. 1). Incomplete files, internal distraction patients, and syndromic cleft patients were excluded from the study.

Included subjects were then divided into patients treated during growth (female $<$ 14 years; male $<$ 16 years), and patients treated after their growth spurt.

2.1. Maxillary distraction osteogenesis technique

The maxillary DO was undertaken using the KLS Martin rigid external distraction system® (RED, Tuttlingen, Germany) or the Walter Lorenz Blue device® (Jacksonville, Florida). Preoperatively, a custom-made intraoral orthodontic appliance was inserted in each patient to link the maxilla to the distraction apparatus. The system consisted of a double arch (vestibular and palatal arch) connected with a transpalatal arch (to increase the rigidity of the system) cemented with bands on the first permanent molars and the first primary molars or permanent premolars. Two external traction hooks were welded to the vestibular arch at the level of the lateral incisors, and were bent under and in front of the upper lip with the hook ending at the level of the palatal plane.

Intraoperatively, a LeFort I osteotomy was performed using a reciprocating saw. Pterygomaxillary dysjunction and a maxillary down-fracture were performed. Although not always realized (Yamauchi et al., 2006), this down-fracture seems useful to properly release the scar adhesions. No intraoperative repositioning of the maxilla was performed, and no bone grafting or internal skeletal fixation was utilized. The halo of the RED device was then applied and secured after closure of the intraoral wound.

After a latency period of 3–4 days, the activation period was started at a rate of 1.5 mm per day. Patients were evaluated once per week during the distraction phase until the required advancement was obtained. The overjet between the maxillary incisors and the decompensated mandibular incisors was used as the clinical guide to determine the end of the distraction. The system was then left in place for 4–6 weeks for consolidation. After removal of the halo, a removable orthodontic facemask with elastic traction was used at night for another 6 weeks for retention.

2.2. Cephalometric analysis

The standardized lateral cephalometric radiographs analyzed were taken preoperatively (T0), and postoperatively at 6–12 months (T1) and more than 4 years (T2). The Procuste software (© 2007 Procuste sarl, Caen, France) was used for the classical analysis. The bony landmarks used in this cephalometric evaluation included the points: sella (S), basion (Ba), pterygoid (Pt), nasion (N), anterior nasal spine (ANS), posterior nasal spine (PNS), A-point (A), B-point (B), menton (Me), gnathion (Gn), gonion (Go), articulare (Ar), incisal edge and apex of the maxillary central incisor, incisal edge and apex of the mandibular central incisor, occlusal point (projection of the first maxillary premolar on the occlusal plane), distal of first maxillary molar, distal of first mandibular molar, porion (Po), orbitale (Or), and pogonion (Pog).

The angular measurements analyzed at T0, T1 and T2 were (Fig. 2): SNA, SNB, ANB, Frankfort(Fr)-mandibular plane angle (FMA), facial angle of Ricketts (Fr/N-Pog), facial axis angle of Ricketts (Ba-N/Pt-Gn), occlusal plane-Fr angle (OP/Fr), interincisal angle, and superior incisor-Fr angle (Isup/Fr). The distance AoBo, representing the orthogonal projection of A-point and B-point on the occlusal plane, was also measured in millimeters (mm).

The Procrustes superimposition method (Penin et al., 2002; Penin-Lambert et al., 2003a,b) was then performed between T0 and T2 to evaluate the global effect of treatment (Fig. 3). Only the cranial base points (N, S, Ba, Pt) were used for the superimposition. This method allowed analyzing the global morphological variations of the maxilla and mandible, without introducing the size factor (eliminating the size differences between the radiographs). From these superposition images, the gonial angle (Ar-Go/Go-Me) and symphyseal angle (Go-Me/Me-B) were measured at T0 and T2 (Fig. 4). Rotations of the mandibular plane, ramus, and palatal plane were also calculated (Fig. 5). The value was negative when the rotation was counter-clockwise.

2.3. Statistical analysis

Statistical analysis of the data was performed using the Statistical Package for Social Sciences, version 17.0, software (SPSS,

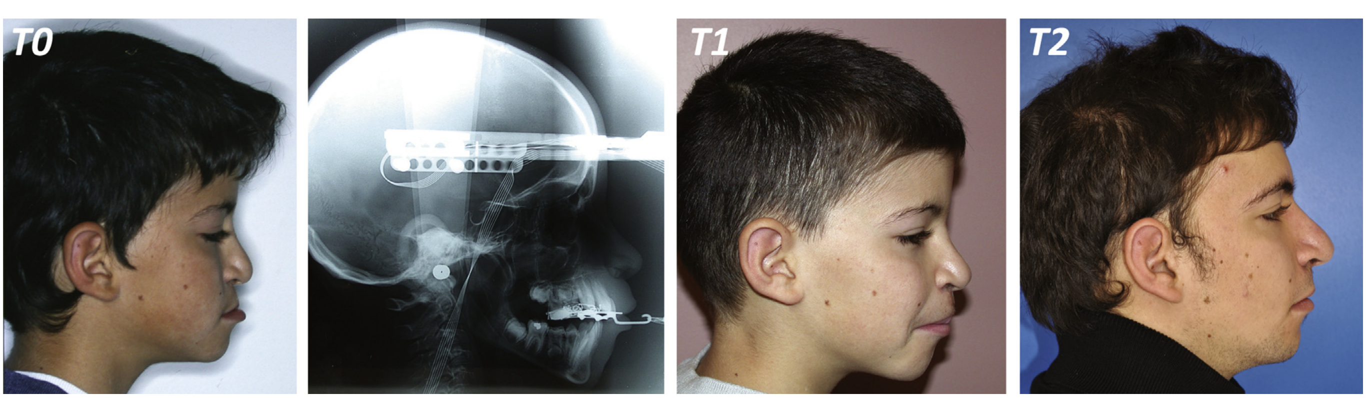

Fig. 1. Growing cleft lip and palate patient with severe maxillary hypoplasia preoperatively (T0), 6–12 months post-distraction (T1), and more than 4 years post-distraction (T2).

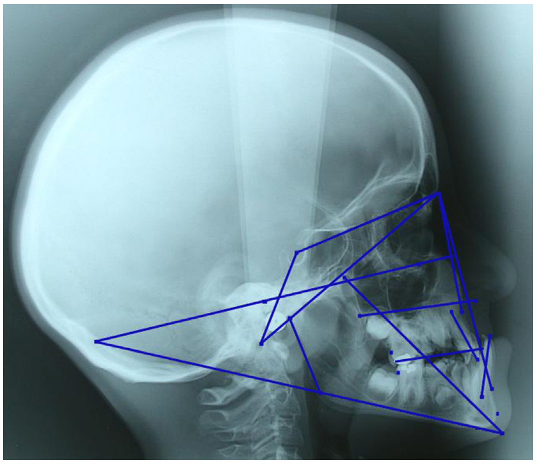

Fig. 2. Cephalometric analysis.

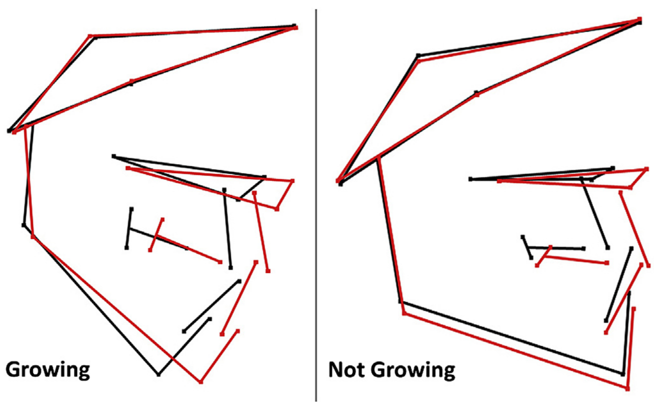

Fig. 3. Examples of Procrustes superimposition method for growing CLP patients, and CLP patients treated after their growth spurt (T0 in black, T2 in red).

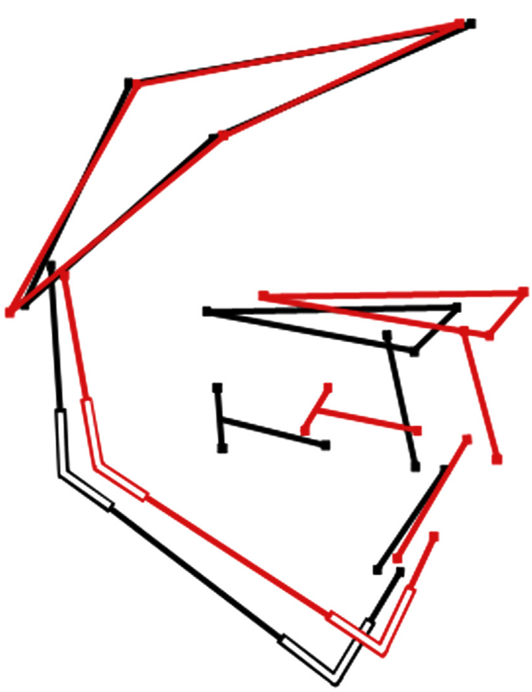

Fig. 4. Gonial and symphyseal angles (T0 in black, T2 in red).

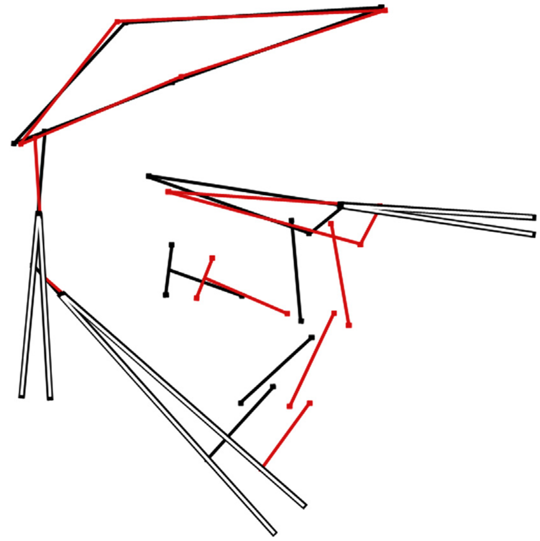

Fig. 5. Rotations of mandibular plane, ramus, and palatal plane (T0 in black, T2 in red).

Table 1
Characteristics of growing patients and patients treated after the growth spurt.

	Growing	Not growing	*P* value
Patients	10 (42%)	14 (58%)	
Age	11.9 ± 1.4	17.9 ± 3.5	<0.001*
Sex			0.242
Male	8 (80%)	8 (57%)	
Female	2 (20%)	6 (43%)	
Diagnosis			0.939
UCLP	7 (70%)	10 (71%)	
BCLP	3 (30%)	4 (29%)	
Advancement (mm)	17.4 ± 5.4	15.3 ± 5.8	0.366
Advancement > 15 mm	6 (60%)	8 (57%)	0.889
Distraction time (days)	15.5 ± 6.2	12.4 ± 3.2	0.118
Consolidation time (weeks)	5.0 ± 2.1	4.5 ± 1.5	0.498

Data presented as mean ± standard deviation or numbers, with percentages in parentheses.
*Statistically significant.

Chicago, IL). All variables were divided into continuous and categorical variables. Growing patients and patients treated after their growth spurt were compared after calculating the difference between each variables at T0–T1, T1–T2, and T0–T2. The Pearson χ^2 test was used to analyze categorical variables. The *t* test was used to evaluate a categorical variable with a continuous variable. For all statistical tests $P < 0.05$ was considered statistically significant.

3. Results

3.1. Patients

Thirty-three consecutive CLP patients with severe maxillary hypoplasia were treated with external maxillary DO, between January 2001 and January 2008. Of these, 3 syndromic patients, 2 patients with early complications (hardware loosening), and 4 patients lost to follow-up required removal from the study.

A total of 24 CLP patients met the inclusions criteria and were reviewed (Table 1). Of these patients, 10 were treated during growth, and 14 were treated after their growth spurt. At time of surgery, the mean age ± standard deviation (SD) was of 11.9 ± 1.4 years for the growing group, compared to 17.9 ± 3.5 years for the non-growing group ($P < 0.001$). At T1, the average follow-up time was of 0.6 ± 0.2 year for the younger group, and 0.7 ± 0.2 year for the older group ($P = 0.597$). At T2, the average follow-up time was of 4.6 ± 1.1 years, and 4.4 ± 0.6 years, respectively ($P = 0.583$). The mean maxillary advancement was important but similar for both groups (growing = 17.4 ± 5.4 mm, not growing = 15.3 ± 5.8 mm; $P = 0.366$). The sex, diagnosis, distraction time, and consolidation time were also all comparable for both groups (Table 1).

3.2. Maxillary changes

The maxillary changes are shown in Tables 2 and 3. Between T0 and T1, a greater increase of the SNA was shown in growing patients, with an average increase of 10.9° ± 4.4°, compared to 7.0° ± 3.7° in patients treated after their growth spurt ($P = 0.036$; Fig. 6). Between T1 and T2, a greater relapse of the SNA was shown in growing patients, with a decrease of −2.8° ± 2.5°, in contrast to 0.0° ± 1.3° for the other group ($P = 0.002$; Fig. 6). The increase of the ANB was also more important for growing patient between T0 and T1, but the differences were not significant (growing = 9.2° ± 3.1°, not growing = 7.4° ± 4.2° mm; $P = 0.268$). Again, a greater relapse between T1 and T2 was demonstrated in growing patient, with an ANB decrease of −2.9° ± 2.4°, compared to −1.1° ± 1.2° for the other group ($P = 0.032$). The distance AoBo had a similar tendency, but the differences were not statistically significant. Between T1 and T2, the distance AoBo decreased by 2.2 ± 2.3 mm in growing patient, which represent a 23.5% of the initial increase between T0 and T1. This is in comparison to a decrease of 0.8 ± 2.1 mm for the other group (9.3% of the initial increase between T0 and T1). Although not significant, the palatal plane showed a more important counter-clockwise rotation of −4.0° ± 4.1° in the growing group, in contrast to a counter-clockwise rotation of −1.9° ± 3.5° for the group treated after their growth spurt ($P = 0.183$; Table 3).

3.3. Mandibular changes

Mandibular changes are shown in Tables 2 and 3. The sagittal and vertical position of the mandible did not differ significantly between the groups, with no statistically significant change of the SNB, FMA, facial angle of Ricketts, or facial axis of Ricketts (all P's > 0.05). The gonial and symphyseal angles also demonstrated no statistically significant differences (Table 3). In growing patients, the mandibular plane and ramus showed a minimal counter-clockwise rotation of −1.4° ± 3.9°, and −1.0° ± 5.0°, respectively (Table 3). Insignificant rotations of the mandibular plane (0.6° ± 2.6°) and the ramus (−0.3° ± 3.8°) were present in patients treated after their growth spurt.

4. Discussion

Maxillary DO is an interesting option for the treatment of severe maxillary deficiency in growing CLP children. Unfortunately, it does not correct the growth deficit inherent to the pathology. Indeed, when compared to patients treated after their growth spurt, growing CLP children show a greater rate of relapse during the first to fourth post-distraction years, with statistically significant decrease of the SNA and ANB angles. Despite their growth, the relative change in mandibular position and form did not differ significantly compared to older CLP patients.

External maxillary DO in growing CLP children show a greater rate of relapse after the first post-distraction year. Between the 1st–4th post-distraction years, the SNA decreased by 25.7% of its initial increase between pre-treatment and 6–12 months post-treatment. This is in comparison to no change of the SNA in patients treated after their growth spurt. Similarly, the ANB angle and AoBo distance of growing patients decreased by 31.5% and 23.5% respectively, compared to 14.9% and 9.3% respectively for the other group. This rate of relapse was underestimated in our study because it did not account for the relapse occurring between the immediate post-distraction phase and the 6–12 months post-distraction phase.

A contradiction between the maxillary advancement (in mm) and the SNA angle values was found in our results. The impact of the advancement measured a significant higher increase in SNA angle values in the growing group between T0 and T1 (growing = 10.9° ± 4.4°, not growing 7.0° ± 3.7°; $P = 0.036$), but failed

Table 2
Cephalometric variables of growing patients and patients treated after the growth spurt.

	Growing			Not growing			P value
	T0	T1	T2	T0	T1	T2	
SNA (°)	72.2 ± 5.8	83.8 ± 7.1	80.3 ± 7.3	72.6 ± 5.1	79.6 ± 5.2	79.6 ± 5.4	0.194
SNB (°)	75.4 ± 5.8	77.7 ± 5.6	77.1 ± 5.7	77.2 ± 3.6	76.9 ± 4.2	78.1 ± 4.6	0.418
ANB (°)	−3.2 ± 1.9	6.1 ± 3.5	3.2 ± 4.0	−4.6 ± 4.8	2.7 ± 2.8	1.6 ± 2.9	0.268
FMA (°)	30.9 ± 6.5	32.0 ± 4.3	31.5 ± 5.9	29.7 ± 8.6	31.3 ± 8.0	30.7 ± 7.9	0.793
Facial angle of Ricketts (Fr/N-Pog, °)	86.3 ± 6.6	86.9 ± 6.1	86.1 ± 5.3	88.1 ± 3.2	88.1 ± 3.9	88.5 ± 3.8	0.677
Facial axis of Ricketts (Ba-N/Pt-Gn, °)	85.4 ± 6.9	86.1 ± 6.8	86.0 ± 7.1	86.6 ± 7.2	85.6 ± 6.9	86.9 ± 7.3	0.354
AoBo (mm)	−7.1 ± 3.7	2.0 ± 4.0	0.2 ± 4.7	−9.0 ± 5.5	−0.4 ± 4.4	−1.3 ± 4.9	0.799
OP/Fr (°)	8.4 ± 4.4	9.0 ± 4.2	8.6 ± 6.0	7.6 ± 4.9	6.6 ± 3.7	6.6 ± 4.0	0.522
Interincisal angle (°)	142.6 ± 14.4	135.4 ± 11.2	133.9 ± 12.1	144.1 ± 11.9	132.9 ± 8.2	135.6 ± 6.4	0.972
Isup/Fr (°)	101.1 ± 9.8	107.1 ± 8.3	109.5 ± 8.1	101.3 ± 11.0	110.6 ± 5.4	109.8 ± 5.3	0.984

Data presented as mean ± standard deviation.

Table 3
Long-term mandibular and palatal changes of growing patients and patients treated after the growth spurt.

	Growing		Not growing		P value
	T0	T2	T0	T2	
Gonial angle (Ar-Go/Go-Me, °)	131.3 ± 5.0	131.7 ± 5.5	132.0 ± 7.3	133.0 ± 8.2	0.608
Symphyseal angle (Go-Me/Me-B, °)	84.7 ± 6.7	82.7 ± 7.5	77.0 ± 6.9	74.7 ± 7.0	0.845
Mandibular plane rotation (°)	−1.4 ± 3.9		0.6 ± 2.6		0.193
Ramus rotation (°)	−1.0 ± 5.0		−0.3 ± 3.8		0.680
Palatal plane rotation (°)	−4.0 ± 4.1		−1.9 ± 3.5		0.183

Data presented as mean ± standard deviation.

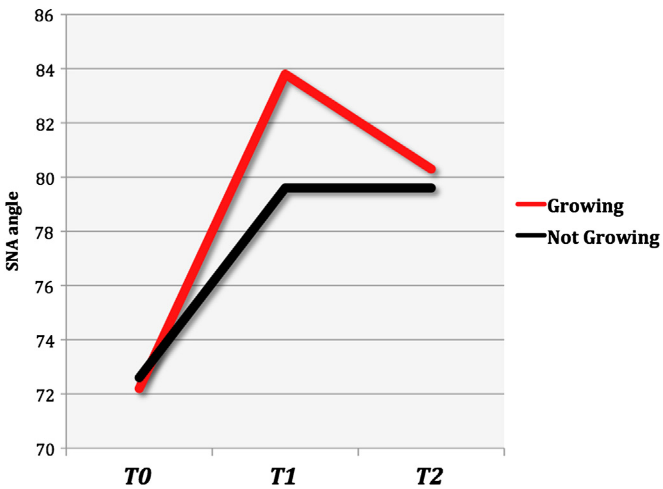

Fig. 6. Changes in SNA at T0, T1, and T2 between growing CLP patients, and CLP patients treated after their growth spurt.

to show a statistically significant difference when the advancement was measured in millimeters (growing = 17.4 ± 5.4 mm, not growing = 15.3 ± 5.8 mm; $P = 0.366$).

The anticipated maxillary relapse and mandibular growth of growing CLP children need to be compensated by overcorrection. This need for overcorrection has also been stated by numerous other studies (Cho and Kyung, 2006; Harada et al., 2006; Huang et al., 2007; Gürsoy et al., 2010; Chen et al., 2011). Cho and Kyung (2006) evaluated 9 CLP treated with RED for severe maxillary hypoplasia, with a follow-up of 1–6 years, and reported a relapse rate of 23% of the mean distraction length. They suggested that an overcorrection in maxillary advancement of 20–30% is needed in the growing child to compensate for a partial relapse and growth deficit (Cho and Kyung, 2006). Harada et al. (2006) examined 8 children with CLP who underwent maxillary external distraction, with a follow-up of 36 months, and reported a relapse of 12% of the original maxillary advancement. The maxilla also showed an inferior growth during the follow-up period (Harada et al., 2006). They reported that maxillary distraction performed during childhood needed considerable overcorrection, but that the maxilla should not be distracted to an adult position to avoid marked deterioration of their masticatory function (Harada et al., 2006). They concluded that the goal of maxillary distraction during childhood is to allow the maxilla to catch up to the mandibular growth at the treatment point (Harada et al., 2006). Huang et al. (2007) reported a horizontal relapse of the maxilla of up to 33% after evaluating 6 growing CLP patients treated with RED, with a follow-up of more than 1 year. They also reported no further forward growth of the maxilla after distraction, again indicating that maxillary DO in CLP does not correct the inherent growth deficit (Huang et al., 2007). Similarly, Chen et al. (2011), in their long-term (5 years) evaluation of 12 growing CLP patients treated with RED, reported that maxillary DO demonstrated a linear and volumetric maxillary growth during the distraction phase without clinically significant continued growth thereafter. The authors also suggested that maxillary DO may need to be overcorrected by up to 53% of the distraction length to take into account relapse and recurrence of midface retrusion (Chen et al., 2011).

Unfortunately, there is currently no predictor that would quantify the overcorrection necessary to palliate the residual maxillary growth deficit and future mandibular growth. When using our results and the results of prior studies, we can suggest that an overcorrection of at least 20% is advisable for the maxillary DO of growing CLP children. The overcorrection can be calculated by adding 20% to the maxillary advancement required to obtain a normal overjet after decompensation of the mandibular incisors. This overcorrection is necessary possibly to avoid secondary orthognathic surgery at the end of growth. It needs to be adapted to the skeletal discrepancies but also to the mandibular growth prognosis that can be calculated using Björk criteria (Björk, 1963; Björk and Skieller, 1983).

Our study failed to show a forward and downward growth of the mandible in the growing CLP group. This probably due to the use of mainly angular cephalometric measurements, and the absence of linear measurements (other than AoBo) in the horizontal or vertical plane. Nevertheless, the overall mandibular shape and rotation did not differ significantly between the two groups. Other studies have reported a downward and forward movement of the mandible when maxillary DO was used in growing CLP children (Harada et al., 2006; Gürsoy et al., 2010; Chen et al., 2011).

Limitations of our study included its retrospective design. This selection bias possibly could have been prevented with the use of a prospective clinical trial. The sample size was also small, with a higher risk of sampling error, and the number of patients in each group was unequal (10 versus 14). Further research, with a larger number of patients with long-term follow-up, would be beneficial to identify potential predictors of relapse and to clarify the amount of overcorrection required.

5. Conclusions

Maxillary DO is a reliable treatment of severe maxillary hypoplasia in growing CLP patients. Unfortunately, it does not correct the growth deficit inherent to the pathology. An overcorrection of at least 20% is recommended for the growing CLP child to compensate the relapse, residual maxillary growth deficit, and future mandibular growth. Patients and parents should be informed that secondary orthognathic surgery could be necessary at the completion of growth.

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Ethics

This research has followed the principles outlined in the Declaration of Helsinki.

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None.

Conflict of interest statement

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Guillaume Captier: no commercial associations or financial disclosures.

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