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Synchronous Premaxillary Osteotomy with Primary Cheiloplasty for BCLP Patients with Protrusion of the Premaxillae

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Background: In bilateral cleft lip and palate (BCLP) patients with protrusion and/or torsion of the premaxillae, it is difficult to achieve a good outcome. We have developed a series of procedures of premaxillary osteotomy with primary cheiloplasty for BCLP patients who did not respond well to presurgical orthodontics (PSO).
Methods: A total of 27 BCLP patients with protrusion and/or torsion of the premaxillae underwent PSO. For 3 BCLP patients in whom the protruded premaxillae could not be returned to a good position, a primary premaxillary osteotomy and gingivoperiosteoplasty (GPP) with cheiloplasty were performed simultaneously. Subsequently, Furlow palatoplasty was performed by one and a half years of age. Maxillary growth was evaluated by dental occlusion at 4 years of age.
Results: A premaxillary osteotomy and GPP with cheiloplasty were performed at 6 months. The patients' facial structures improved, their premaxillae were positioned more superiorly, and normal inclination of the incisors was achieved. They had edge-to-edge occlusions or cross bites at 4 years of age.
Conclusions: As advantages, the patients' facial structures improved, and the alveolar bones were formed by GPP. As a disadvantage, premaxillary necrosis might occur because of poor blood circulation. It is important to secure the following 2 blood supplies: from the periosteum and soft-tissue of the anterior premaxillae and from the periosteum and mucosa of the nasal septum. Synchronous premaxillary osteotomy and GPP with primary cheiloplasty are appropriate when the premaxillae cannot be properly repositioned by PSO or PSO cannot be done.
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INTRODUCTION

In bilateral cleft lip and palate (BCLP) patients with protrusion and/or torsion of the premaxillae, it is difficult to achieve a good outcome with adequate facial and maxillary development.

Some presurgical orthodontics (PSO) including nasoalveolar molding have been used to pull back protruded premaxillae,[1–5] but there have been cases in which PSO did not work. We may have no other choice but to perform only cheiloplasty as a primary procedure when PSO cannot be performed or pull back of the premaxilla by PSO fails. However, the facial appearance and occlusion would be unsatisfactory. In such cases, there are some reports of premaxillary osteotomy done in combination with secondary bone grafting or cheiloplasty at a later age,[6–11] with a few reports at an earlier age.[12–14] For the affected children, improvement of their facial appearance at an earlier age would be advantageous with regard to fitting into society, such as when attending kindergarten and elementary school.

The purpose of the present study was to establish a series of procedures for premaxillary osteotomy with cheiloplasty for BCLP patients with protrusion and/or torsion of the premaxillae.

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PATIENTS AND METHODS

This was a retrospective study conducted at Kanagawa Children's Medical Center from 2008 to 2012. During this period, 27 complete BCLP patients with premaxillary protrusion and/or torsion underwent PSO to return the protruded premaxillae to a good position. Of these patients, 3 had left premaxillary protrusion and/or torsion after PSO. The degrees of premaxillary protrusion were measured on the dental cast before and after PSO (Fig. 1; Table 1). Simultaneous premaxillary osteotomy and gingivoperiosteoplasty (GPP) with primary cheiloplasty were performed (Figs. 2, 3). Subsequently, palatoplasty using the Furlow method was performed by the age of one and a half years.

SURGICAL TECHNIQUES

Premaxillary osteotomy was performed in combination with primary bilateral simultaneous cheiloplasty and Millard type GPP (Figs. 4–6). First, the skin and mucosa were incised according to the design of Figure 2. The vomero-premaxillary suture (VPS) was identified, and an osteotomy was made with a chisel posterior to the suture. The septo-premaxillary ligament on the premaxilla anterior to the VPS was preserved.

The premaxilla was placed in its new position where its posterior edge contacts the anterior edge of the lateral maxillary segments but is not aligned with it. At that time, 2 blood supplies, from the vestibular periosteum and soft tissue of the anterior premaxillae and from the periosteum and mucosa of the nasal septum, were secured to avoid premaxillary necrosis.

GPP was carried out on both sides in a single stage with the cheiloplasty. No bone graft was added to the osteotomy site. Finally, the premaxillae were immobilized by suturing of the GPP on both sides. No wire or other type of fixation was used to stabilize the premaxillae (**see video, Supplemental Digital Content 1**, which displays GPP being performed. This video is available in the Related Videos section of the Full-Text article on PRSGlobalOpen.com. ***http://links.lww.com/PRSGO/A469***.)

The lateral lip muscles were sutured directly in the midline, and a philtrum hollow was constructed.

Subsequently, the remaining cleft of the hard and soft palates was reconstructed by one and a half months.

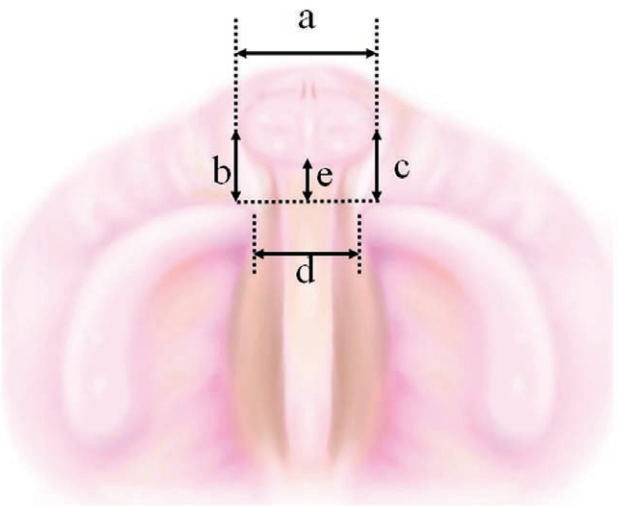

Fig. 1. Alveolar measurement before and after PSO. A, premaxillary width; B, length of the right premaxillary-lateral segment; C, length of the left premaxillary-lateral segment; D, distance between the lateral segments; E, distance between the posterior edge of the premaxilla and the anterior edge of the lateral segment.

RESULTS

For 3 BCLP patients with protrusion and/or torsion of the premaxillae in whom the protruded premaxillae could not be placed in a proper position, simultaneous premaxillary osteotomy and GPP with primary cheiloplasty were performed at 6 months of age. Proper positioning of the premaxillae was achieved in all patients. All their facial structures improved, and their premaxillae were retreated. There were no major complications, such as necrosis of the premaxillae or a fistula. However, they had edge-to-edge occlusions or cross bites (Table 2). All of them had good speech outcomes at 4 years of age. The mean follow-up for the 3 patients was 6 years 2 months.

CASE REPORTS

Case 1: A 1-Month-Old Male with Protrusion of the Premaxillae

The distance between the posterior edge of the premaxilla and the anterior edge of the lateral segment was 8 mm sagittally (Figs. 7–13). The premaxillae remained protruded and torsional, despite PSO being done for 5 months. The patient underwent premaxillary osteotomy at 6 months of age, with repositioning of the premaxillae. Furlow palatoplasty was subsequently performed at 18 months of age. At 5 years and 2 months of age, his columella was elongated, and his facial structure improved. His occlusion also improved, although his anterior teeth were edge-to-edge.

Case 2: A 1-Month-Old Male with Protrusion of the Premaxillae

The patient's premaxillae were small with congenital epulis and originally had 3 hypoplastic deciduous teeth (Figs. 13–18). The distance between the posterior edge of the premaxilla and the anterior edge of the lateral segment was 4 mm sagittally. Slight protrusion and torsion remained, despite PSO being done for 5 months. He underwent premaxillary osteotomy at 6 months of age, and his premaxillae were repositioned. Subsequently, Furlow palatoplasty was performed at 18 months of age. At 5 years of age, his columella was elongated, and his facial structure improved. However, his anterior occlusion was a cross bite.

Case 3: A 1-Month-Old Female with Protrusion of the Premaxillae

The distance between the posterior edge of the premaxilla and the anterior edge of the lateral segment was 9 mm sagittally (Figs. 18–24). Torsion remained, despite

Table 1. Alveolar Measurement before and after PSO

Case	Presurgical Orthodontics	Age of Measurement (mo)	Premaxillary Width (mm)	Right Length of Premaxillary-Lateral Segment (mm)	Left Length of Premaxillary-Lateral Segment (mm)	Distance between Lateral Segments (mm)	Distance between the Posterior Edge of the Premaxilla and the Anterior Edge of the Lateral Segment (mm)
1	Before PSO	1	17	8	12	16	8
	After PSO	6	18	6	14	23	0
2	Before PSO	1	16	11	9	18	4
	After PSO	6	17	9	5	14	0
3	Before PSO	1	14	13	14	20	9
	After PSO	6	17	2	12	19	0

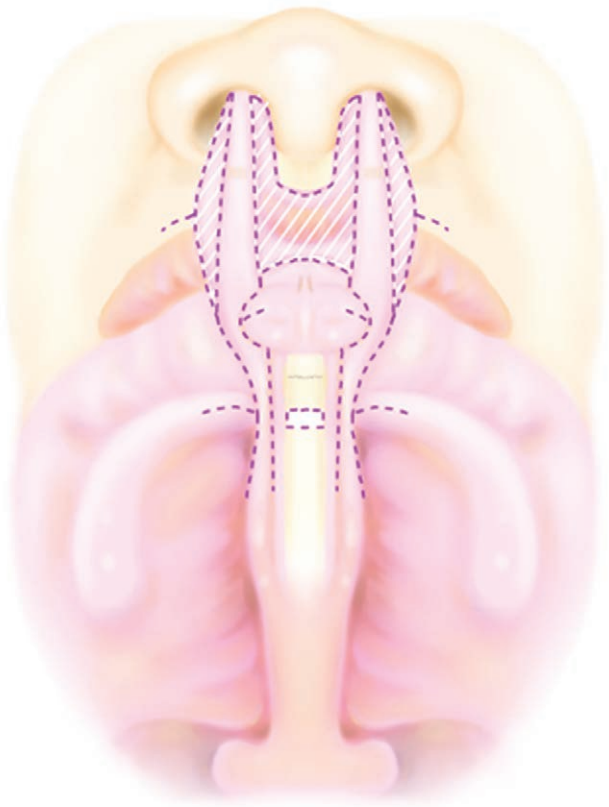

Fig. 2. Incision lines are marked along the margin of the cleft lip and alveolus for regular bilateral cleft lip and GPP. The shadow areas of skin and mucosa are removed. Then, the white area of vomer located posterior to the VPS is removed through the incision lines.

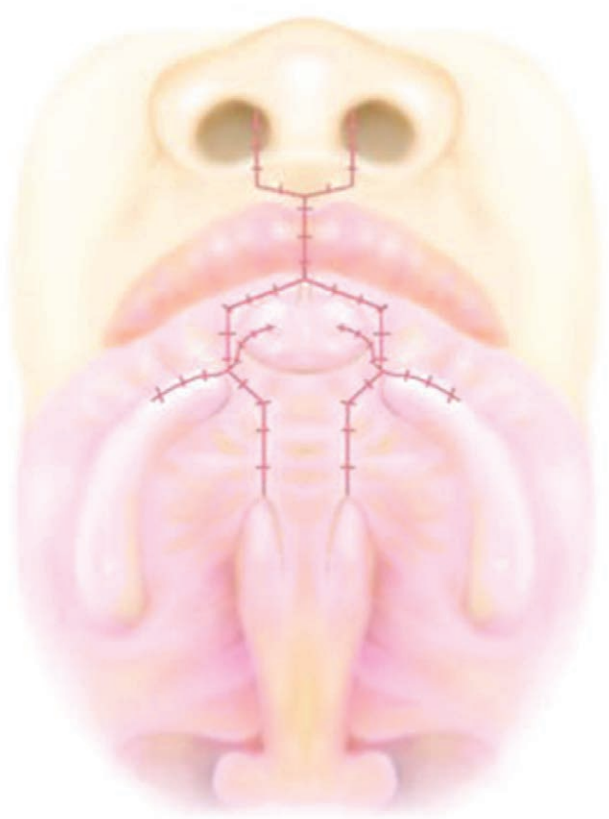

Fig. 3. After suturing; repositioning of the premaxillae before the lateral segments and placing the suture line.

PSO being done for 5 months. The patient underwent premaxillary osteotomy at 6 months of age, and the premaxillae were repositioned. Subsequently, Furlow palatoplasty was performed at 15 months of age. At 4 years of age, her columella was elongated, and her facial structure improved. Her occlusion also improved, although her anterior occlusion was a cross bite.

DISCUSSION

We have dealt with synchronous premaxillary osteotomy and GPP with primary cheiloplasty at an early age for protrusion and/or torsion of the premaxillae. The time of the osteotomy of the premaxillae, which extends from infancy to adolescence, varies. There are some reports stating that premaxillary osteotomy should be delayed until a late age.[6–11] On the other hand, there are other reports of premaxillary osteotomy performed at an early age of less than 2 years.[12–14]

Although both approaches have advantages and disadvantages, using this technique at an early age could have 3 advantages over using it at a later age. First, the patients would be better able to fit into society, such as when attending kindergarten and elementary school, as a result of the early improvement of their facial structures. Second, improving their facial structures, including their columella and philtrum hollow, would be easier at an early age than at a later age after cheiloplasty. That

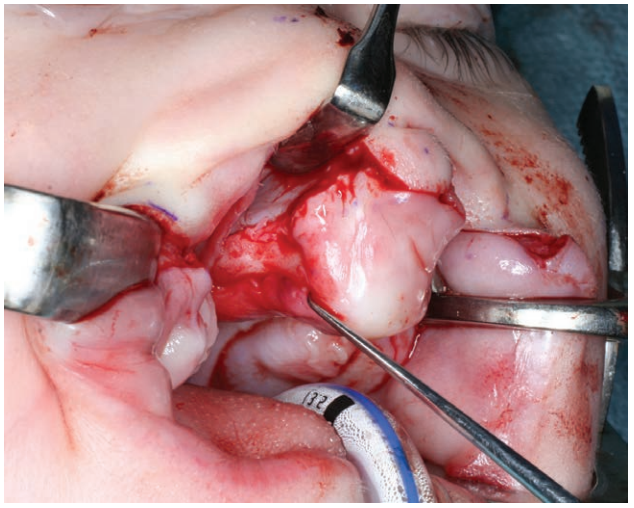

Fig. 4. The vomer behind the VPS is dissected.

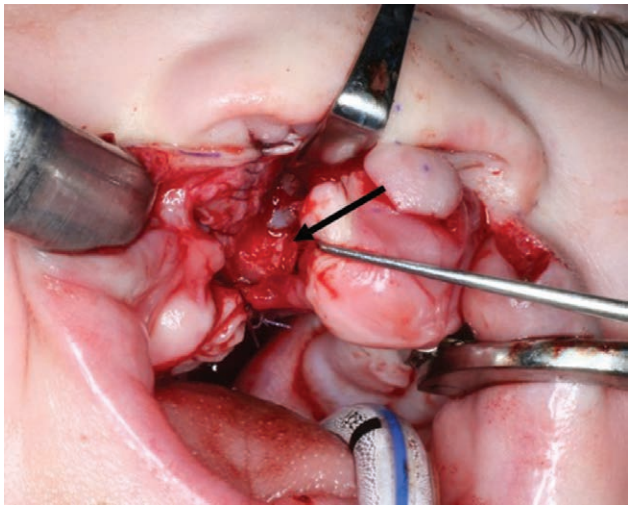

Fig. 5. After the premaxillary osteotomy, part of the vomer is removed. The vomerine stump (black arrow).

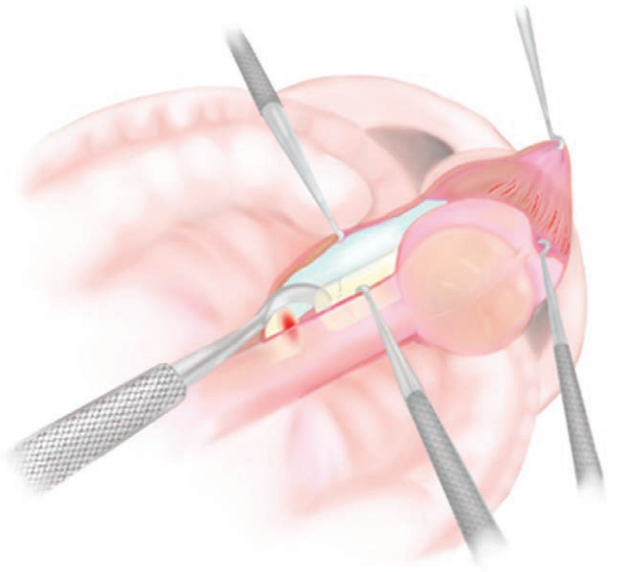

Fig. 6. Illustration of premaxillary osteotomy. The premaxillae are secured by 2 blood supplies, from the periosteum and the anterior soft tissue of the premaxillae (black arrow), as well as from the periosteum and mucosa of the vomer (black dotted arrow).

Video Graphic 1. See video, Supplemental Digital Content 1, which displays GPP being performed. This video is available in the Related Videos section of the Full-Text article on PRSGlobalOpen.com or available at, ***http://links.lww.com/PRSGO/A469***.

is why waiting until adolescence to correct the protruding premaxillae might have a negative impact on nasolabial size and shape.[15] Third, the alveolar bones were formed by GPP on both sides. The role of GPP is not only to immobilize the premaxillae but also to aid in alveolar bone formation.

On the other hand, employing this technique at an early age could have 2 disadvantages. The first issue is that premaxillary and prolabial ischemia might occur because of defective circulation of blood or direct injury. Therefore, it would be important to secure the following 2 blood supplies, from the periosteum and soft tissue of the anterior premaxillae and from the periosteum and mucosa of the nasal septum. There are 2 methods that may further reduce the risk of premaxillary and prolabial ischemia. One method is to perform GPP on only 1 side to secure the blood supply from 1 side of the nasal septum without GPP. The other is to perform premaxillary osteotomy and GPP without cheiloplasty as a first stage to secure the blood supply from the mucosa and periosteum of the anterior premaxillae.[14] Palatoplasty may be performed simultaneously when cleft palate width is narrow. The second issue is maxillary retrusion. There are reports that premaxillary osteotomy affects maxillary growth,[16,17] though there are other reports that premaxillary osteotomy does not affect maxillary growth.[18,19]

Although the procedures of premaxillary osteotomy and palatoplasty might be important elements for maxillary growth inhibition, BCLP originally might involve some underdevelopment of the lateral maxillary segments in the embryonic period. A histological study of the maxillae in fetuses has given definite support to this analysis.[20] Therefore, excessive premaxillary setback would result in much further concave faces because of congenital lateral maxillary retrusion. We tried not to

Table 2. Cephalometric Analysis

Measurements	Case 1 at 5 y and 2 mo	Case 2 at 5 y	Case 2 at 4 y	Normal Japanese Average at 5 y (mean ± SD)
SNA	73.4	77.6	71.6	81.35±2.78
SNB	69.1	75.1	70.2	76.38±2.08
ANB	4.3	2.5	1.4	0.00±0.00
Mandibular plane	37.9	29.5	28.2	31.13±5.19
Gonial angle	130.7	123.3	124.5	130.48±4.31
y Axis	71.8	63.9	62.8	63.77±3.30
Occlusal plane	15.2	86.3	15.5	14.25±4.28
N-S	65.5	66.1	64.4	62.7±2.01
N-ME	113.6	112.3	103.1	101.08±3.30
N-ANS	48.1	45.2	43.8	44.05±2.11
ANS-ME	68	68.5	59.9	59.18±2.32
S'-PTM'	15.74	18.2	13	17.43±2.31
A'-PTM'	43.8	43.2	40.8	42.45±2.47
GN-CD	97.6	98.2	92.3	91.93±2.64
POG'-GO	64.1	67.9	63.2	61.00±4.04
CD-GO	45.3	43.8	41.6	45.79±3.15

Fig. 7. Case 1. One-month-old male with protruded premaxillae.

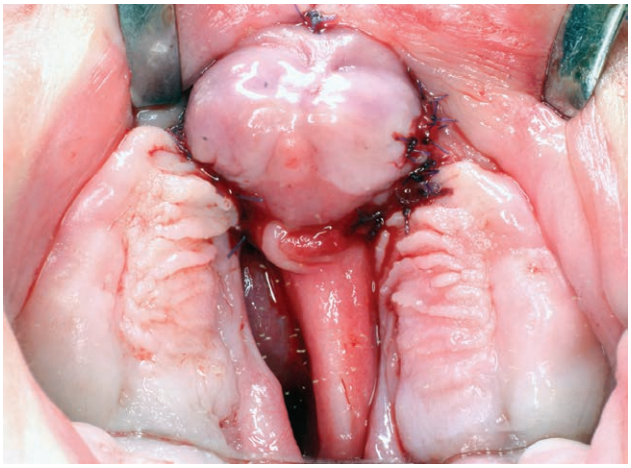

Fig. 8. His premaxillae repositioned after primary premaxillary osteotomy.

align the premaxilla with the lateral maxillary segment and to put it at the point where the posterior edge of the premaxilla could be contacted with the anterior edge of the lateral maxillary segment because the premaxilla was not placed too far back.

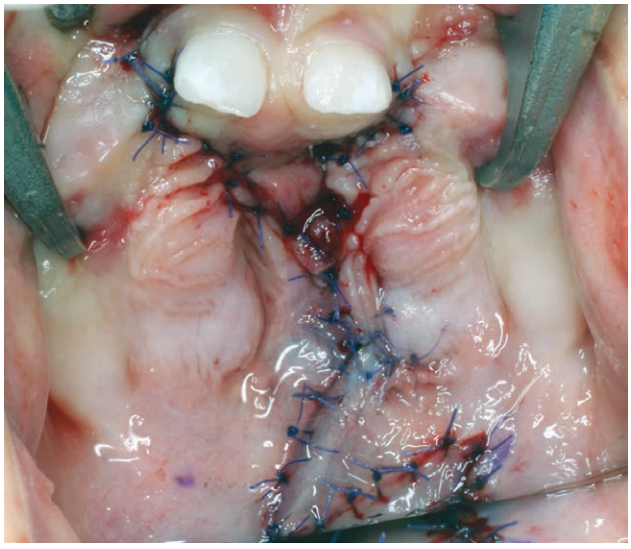

Fig. 9. Immediately after Furlow palatoplasty at 18 months of age.

Fig. 10. Submental view. Four years after primary premaxillary osteotomy.

Fig. 11. Occlusal view. Four years after primary premaxillary osteotomy.

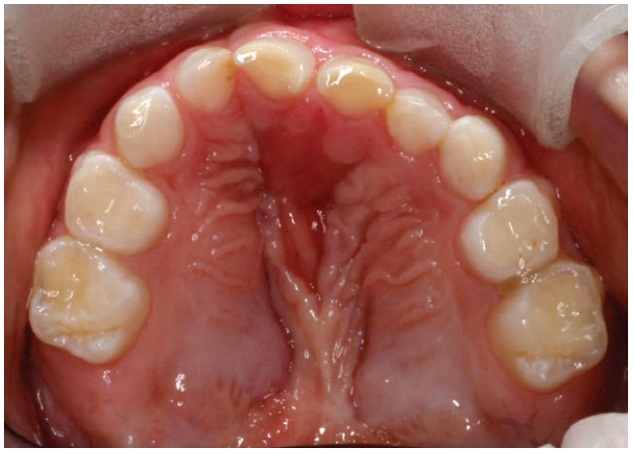

Fig. 12. Palatal view. Four years after primary premaxillary osteotomy.

Fig. 13. Case 2. One-month-old male with distorted small premaxillae.

Eventually, we could not avoid backward movement of the premaxillae, which can lead to pressure on the upper lip after the operation. However, the patients achieved good occlusion by orthodontic treatment including a

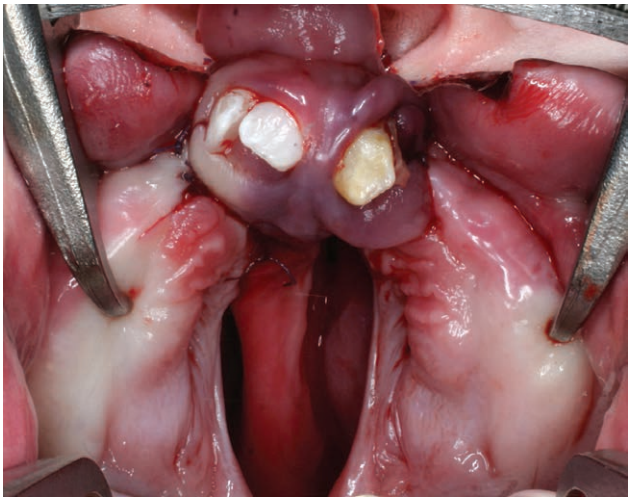

Fig. 14. His premaxillae had congenital epulis and 3 hypoplastic deciduous teeth originally. His premaxillae repositioned after primary premaxillary osteotomy.

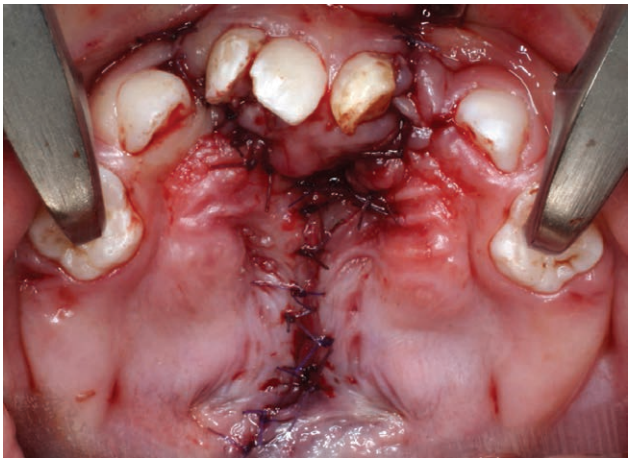

Fig. 15. Immediately after Furlow palatoplasty at 18 months of age.

Fig. 16. Submental view. Four years after primary premaxillary osteotomy.

Fig. 17. Occlusal view. Four years after primary premaxillary osteotomy.

Fig. 18. Palatal view. Four years after primary premaxillary osteotomy.

Fig. 19. Case 3. One-month-old female with distorted premaxillae.

maxillary protraction appliance because maxillary growth inhibition was held to the minimum.

In our view, early premaxillary osteotomy, whenever the premaxillae remained protruded and torsional, optimized primary cheiloplasty and minimized secondary revisions.

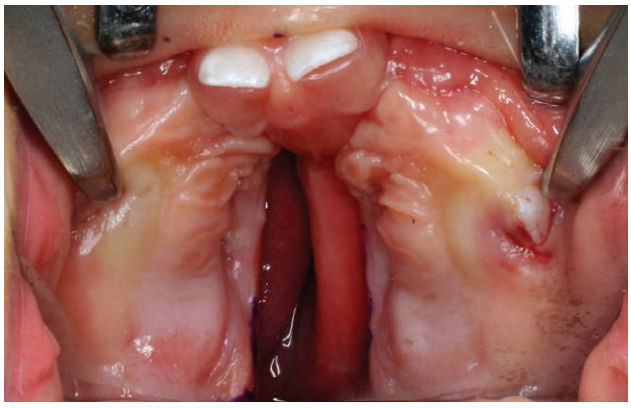

Fig. 20. Her premaxillae repositioned after primary premaxillary osteotomy.

Fig. 21. Immediately after Furlow palatoplasty at 15 months of age.

Fig. 22. Submental view. Four years after primary premaxillary osteotomy.

The present report was based on a retrospective analysis, a very small number of cases, and relatively short follow-up. Careful follow-up over a long time, especially of maxillary growth, until the patients become adults, is needed.

Fig. 23. Occlusal view. Four years after primary premaxillary osteotomy.

Fig. 24. Palatal view. Four years after primary premaxillary osteotomy.

CONCLUSIONS

Synchronous premaxillary osteotomy and GPP with primary cheiloplasty were appropriate when the premaxilla could not be pulled back by PSO or when PSO could not be performed for BCLP patients.

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PATIENT CONSENT

Patients or guardians provided written consent for the use of the patients' images.

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