Bone-anchored maxillary protraction in a patient with complete cleft lip and palate: A case report

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6 authors, including:

Daniela Gamba Garib
University of São Paulo

Marilia Yatabe
University of Michigan

Renato André de Souza Faco
University of São Paulo

Hugo J De Clerck
University of North Carolina at Chapel Hill

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Bone-anchored maxillary protraction in a patient with complete cleft lip and palate: A case report

Daniela Garib,[a] Marília Yatabe,[b] Renato André de Souza Faco,[c] Leonardo Gregório,[c] Lucia Cevidanes,[d] and Hugo de Clerck[e]
Bauru, São Paulo, Brazil, Ann Arbor, Mich, and Chapel Hill, NC

Sagittal maxillary deficiency is frequently observed in patients with operated unilateral complete cleft of the lip and palate. Treatment for moderate to severe Class III malocclusion usually relies on LeFort I surgery for maxillary advancement after the end of growth. This case report describes bone-anchored maxillary protraction in a 10-year-old white boy with unilateral complete cleft of the lip and palate. His interarch relationship was diagnosed as GOSLON index 5 before treatment with a negative overjet of 3.2 mm. The orthopedic traction was started 4 months after secondary alveolar bone graft surgery and before comprehensive orthodontic treatment. Class III elastics were used full time for 18 months. After treatment, the interarch relationship was GOSLON index 1 with a positive overjet. The SNA angle increased by 6.50° and A-Na Perp increased by 3.8 mm, leading to marked improvement in facial convexity (+14.6°). No posterior rotation of the mandible occurred with a slight closure of the gonial angle. Visualization of 3-dimensional color-coded maps showed an overall forward maxillary displacement. The bone-anchored maxillary protraction results for this patient are a promising orthopedic therapy for patients with unilateral complete cleft of the lip and palate, with the advantage of achieving much earlier improvement of facial esthetics and functional occlusion, compared with LeFort I surgery at skeletal maturity. (Am J Orthod Dentofacial Orthop 2018;153:290-7)

Maxillary growth of patients with unilateral complete cleft lip and palate (CLP) is often negatively influenced by primary surgeries of the soft tissues, leading to a Class III malocclusion.[1-3] In noncleft Class III patients, the most common orthopedic intervention is facemask therapy. The effects of 1 year of facemask therapy include an average of 2 mm of forward movement of the maxilla and downward and backward rotation of the mandible.[4,5] Maxillary protraction anchored in the teeth may also cause dental effects, including proclination of the maxillary incisors and retroclination of the mandibular incisors.[6] In children with CLP, the facemask protocol has little therapeutic effect on the maxilla associated with clockwise rotation of the mandible.[7-9]

As a new treatment option, bone-anchored maxillary protraction (BAMP) pulls the maxilla forward by using intraoral intermaxillary elastics anchored in bone plates. This therapy has been shown to cause an average maxillary protraction of 4 mm in Class III patients without oral clefts, but its effects in cleft patients are not clear because of scar tissue and often severe maxillary deficiency.[10] The purpose of this article is to report the treatment outcomes of the BAMP protocol in a 10-year-old patient with complete CLP.

DIAGNOSIS AND ETIOLOGY

A 10-year-old white boy came with a left unilateral complete CLP combined with a right incomplete cleft lip; he was receiving rehabilitation in a single center. Bilateral lip repair was performed at 4 months of age using the Spina technique. Palatal repair was performed at

[a]Department of Orthodontics, Bauru Dental School, Hospital of Rehabilitation of Craniofacial Anomalies, University of São Paulo, Bauru, São Paulo, Brazil.
[b]Department of Orthodontics, Bauru Dental School, University of São Paulo, Bauru, São Paulo, Brazil.
[c]Hospital of Rehabilitation of Craniofacial Anomalies, University of São Paulo, Bauru, São Paulo, Brazil.
[d]Department of Orthodontics and Pediatric Dentistry, School of Dentistry, University of Michigan, Ann Arbor, Mich.
[e]Department of Orthodontics, School of Dentistry, University of North Carolina, Chapel Hill, NC.
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Address correspondence to: Daniela Garib, Alameda Octávio Pinheiro Brisolla, 9-75, 17012-901, Bauru, São Paulo, Brazil; e-mail, dgarib@usp.br.
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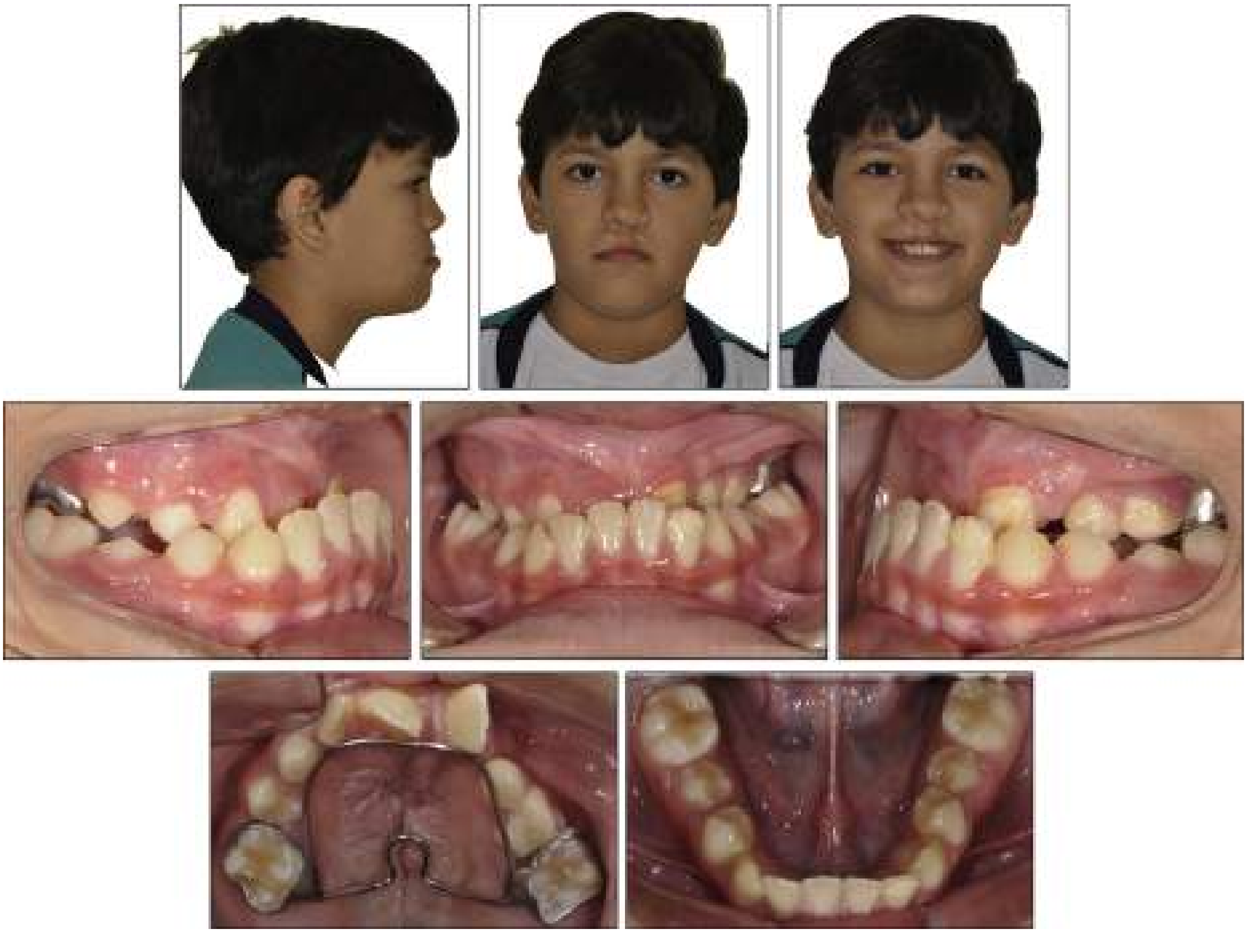

Fig 1. Initial facial and intraoral photographs. Interach relationship GOSLON index 5.

13 months of age using the Von Langenbeck technique. At the age of 6.5 years, secondary lip repair and a palatal fistula closure were performed.

At the age of 8 years, the patient demonstrated a Class III skeletal pattern and a sagittal interarch relationship of GOSLON index 5. Transverse maxillary constriction was treated with rapid maxillary expansion, and a fixed transpalatal arch was used as a retainer. Secondary alveolar bone graft surgery was performed at 10 years of age using 1.5 mg per milliliter rh-BMP2 in collagen membrane (Infuse Bone Graft kit; Medtronic, Memphis, Tenn).

At 10.6 years of age, the extraoral examination showed a Class III skeletal pattern with moderate to severe midface deficiency. The mandible was well positioned, and the face was normodivergent. In the frontal facial view, no asymmetry was observed (Fig 1). The patient was in the late mixed dentition. The intraoral examination showed an interarch relationship of GOSLON index 5 with overjet of −3.2 mm, overbite of 4.5 mm, and a complete crossbite (Fig 1). No shift between centric relation to maximal intercuspation was observed. The panoramic radiograph showed agenesis of the maxillary permanent left lateral incisor and both maxillary second premolars (Fig 2). The cephalometric analysis showed a skeletal Class III discrepancy (ANB angle, −8.4°; Wits appraisal, −6.5 mm) with a retrognathic maxilla, a well-positioned mandible, and a horizontal growth pattern (Fig 2; Table). The maxillary incisors were slightly proclined, and the mandibular incisors were retroclined (Table). At this time, the patient was at prepubertal stage CS1 of skeletal maturity according to the cervical vertebral maturation method.[11]

TREATMENT OBJECTIVES

The treatment objectives were to correct the anteroposterior maxillary deficiency and achieve a positive overbite and an adequate occlusion.

TREATMENT ALTERNATIVES

Based on the diagnosis, 2 treatment options were suggested: (1) comprehensive orthodontic treatment

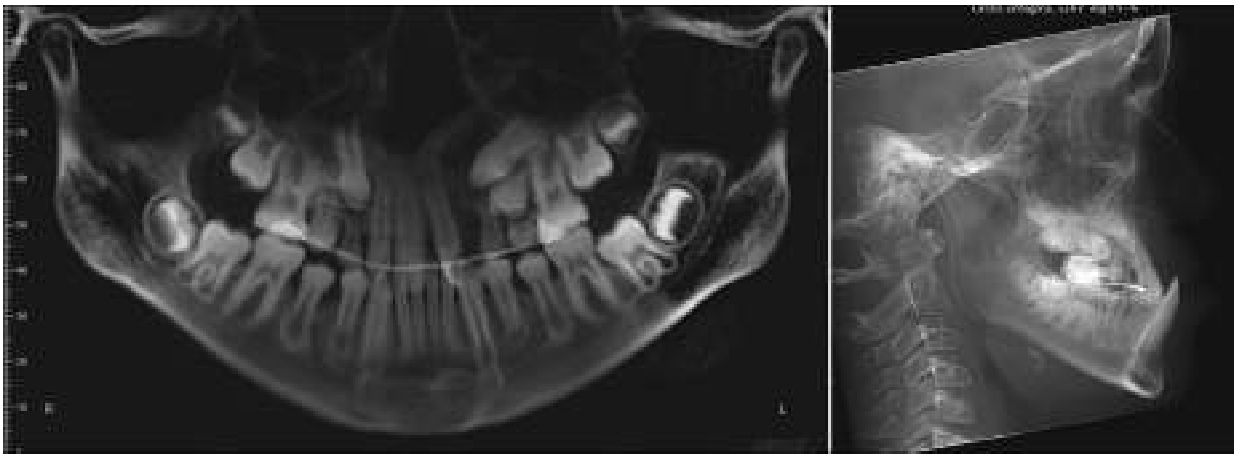

Fig 2. Panoramic and cephametric radiographs. Observe the Class III facial profile with sagittal maxillary deficiency, with a 5.8-mm negative overjet. The patient was in the mixed dentition with agenesis of the maxillary second premolars and maxillary left lateral incisors.

Table. Conventional cephalometric analysis

Variable	*T1*	*T2*	*T2–T1*
Maxilla			
SNA (°)	74.7	81.2	6.5
A-Nperp (mm)	−4.00	−0.20	3.8
Co-A (mm)	46.50	56.10	9.6
Mandible			
SNB (°)	83.1	81.2	−1.9
P-Nperp (mm)	0.80	0.40	−0.4
Mandibular length (Co-Gn) (mm)	119.8	118.1	−1.7
Co-Go (mm)	30.2	33.3	3.1
Ar-Go-Gn (°)	128.3	125.7	−2.6
Maxillomandibular relationship			
ANB (°)	−8.4	−0.1	8.3
Convexity (NA-APo) (°)	−15.9	−1.3	14.6
Wits appraisal (mm)	−6.5	−2.5	4.0
Facial pattern			
FMA (MP-FH) (°)	26.3	25.6	−0.7
SN-GoGn (°)	32.4	31.5	−0.9
Occlusal plane to FH (°)	3.8	5.1	1.3
Anterior face height (NaMe) (mm)	66.2	72.9	6.7
Teeth			
U1-SN (°)	107.3	110.4	3.1
IMPA (°)	81.2	82.5	1.3
Overbite (mm)	4.5	1.9	−2.6
Overjet (mm)	−3.2	3.1	6.3
Molar relationship (mm)	−2.1	3.6	5.7
Soft tissue			
Facial convexity (G′-Sn-Po’) (°)	−4.4	−2.2	2.2
Nasolabial angle (Col-Sn’-ULA) (°)	82.5	73.6	−8.9

T1, Pretreatment; *T2*, posttreatment.

followed by LeFort I osteotomy with maxillary advancement after growth, and (2) BAMP therapy followed by comprehensive orthodontic treatment. In the second option, the patient and his parents were informed that the therapy might not have a good response, and orthognathic surgery would still be necessary after growth.

The second option was chosen because of the potential to avoid orthognathic surgery and achieve improved facial esthetics earlier in adolescence.

TREATMENT PROGRESS

The initial cone-beam computed tomography (CBCT) examination was taken in maximal intercuspation to plan the miniplate placement. Under general anesthesia, a small horizontal incision was made at the level of the mucogingival junction. In the maxilla, it was extended from the distal aspect to the mesial aspect of the first molars where an upward oblique extension of 10 mm parallel to the infrazygomatic crest created an L-shaped flap. In the mandible, the incision extended from the distal aspect of the canine to the mesial aspect of the lateral incisor with a downward oblique extension of 10 mm. Predrillings of 1.1 and 1.6 mm of the external cortical bone, respectively, in the maxilla and mandible, were done. Four miniplates (Bollard type; Tita-Link, Brussels, Belgium) were placed in the maxilla at the left and right infrazygomatic crests and on both sides in the mandible between the roots of the permanent canine and lateral incisor. Titanium screws, 2 mm in diameter and 5 mm in length, were used for miniplate fixation. Prophylactic antibiotic therapy was initiated with the general anesthesia. Analgesics were prescribed after surgery. Hygiene was performed with a soft toothbrush. The patient was instructed not to touch the miniplates with his tongue or tools. Three weeks after surgery, Class III intermaxillary elastics (G&H Orthodontics, Franklin, Ind) connecting the maxillary and

Fig 3. Intraoral photographs after **A-C,** 6 and **D-F,** 10 months of therapy.

Fig 4. Post-BAMP facial and intraoral photographs.

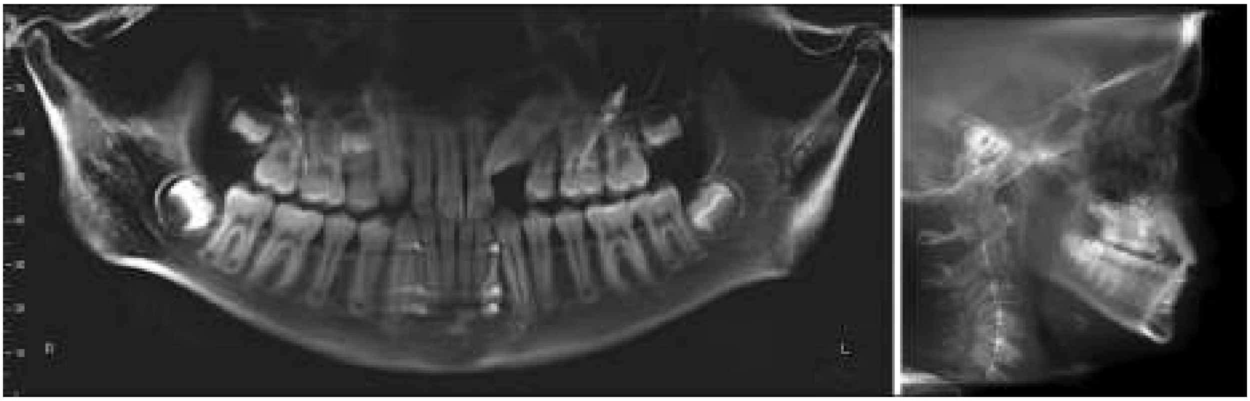

Fig 5. Panoramic and cephalometric radiographs.

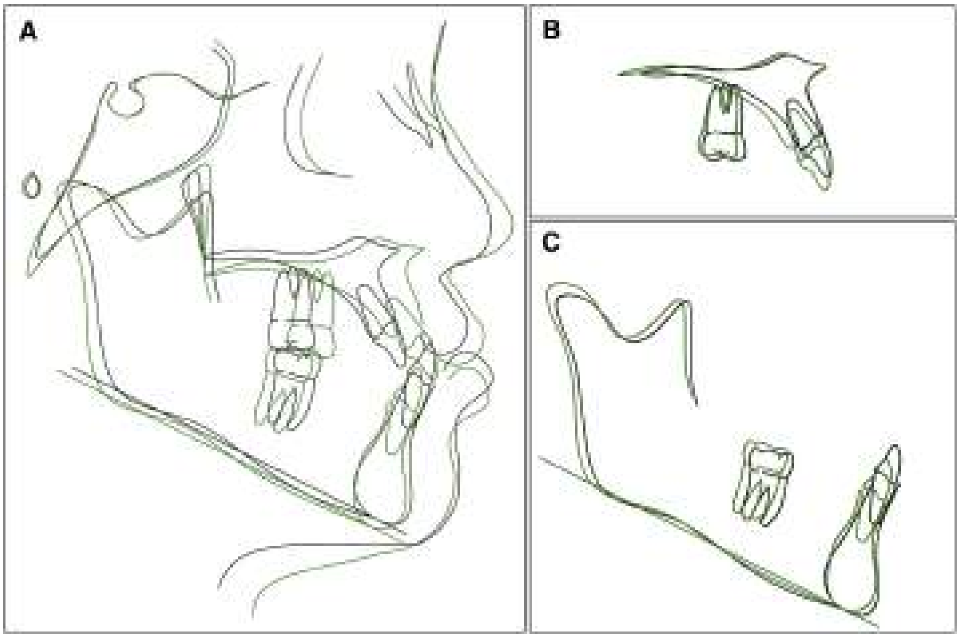

Fig 6. A, Cephalometric superimposition at the cranial base (centered on sella); **B,** maxillary superimposition (palatal plane centered on ANS); **C,** mandibular superimposition (mandibular plane centered on menton). The condyle was displaced backward after treatment as shown in the overall superimposition. BAMP therapy probably produced mandibular fossa remodeling and posterior displacement of the ramus as previously reported in noncleft patients.[12,13]

mandibular miniplates were placed with 75 g of force on each side. The patient was instructed to wear the elastics 24 hours per day and replace them twice a day, in the early morning and at night. After 1 month, the force was increased to 150 g on each side, and a removable biteplate was inserted in the maxillary arch. After 2 months, the forces were adjusted to 250 g per side and remained stable until the end of active therapy. The biteplate had finger springs lingual to the maxillary incisors to jump the bite when intermaxillary elastics reached an edge-to-edge incisor relationship. The biteplate was worn until the anterior crossbite was corrected, which occurred after 13 months of treatment. Monthly occlusal adjustments were necessary to prevent indentations of the biteplate caused by the mandibular teeth.

During treatment, both patient and parents were compliant. There were no complaints about pain at any time after surgery or during the protraction. No adverse effects were observed in the region of the bone graft. Oral hygiene was adequate during the follow-up. During the whole treatment, no local infections around the miniplates or any loosening or mobility was observed. Right after the biteplate was removed, there was a posterior open bite, but within 3 months spontaneous closure occurred. The active treatment with full-time intermaxillary elastics continued until completion of 16 months of therapy, to wait for the complete posterior intercuspation.

Although comprehensive orthodontic treatment was planned only after BAMP therapy, the patient requested to have the rotated maxillary left central incisor corrected for esthetic purposes. Orthodontic auxiliaries were used to correct the tooth rotation using the palatal arch as anchorage (Fig 3). After 16 months of active therapy, extraoral and intraoral photos and a second CBCT examination in maximal intercuspation were taken (Figs 4 and 5). The CBCT examinations were used for cephalometric analysis (Fig 6) and 3-dimensional superimpositions and visual assessments with color-coded maps of the maxillary and mandibular displacements (Figs 7 and 8). Class III elastics will be maintained as retention during the night until the end of facial growth.

TREATMENT RESULTS

After treatment, the convexity of the facial profile and the zygomatic prominence were improved (Figs 4-6). Intraorally, the anterior crossbite was corrected, and a good interarch relationship was obtained. The final panoramic radiograph showed late development of the maxillary right second premolar. Conventional cephalometrics performed on CBCT reformatted images showed considerable increases of SNA (+6.5°), A-Na Perp (+3.8 mm), ANB (+8.3°), Na-Apo (+14.6°), Wits appraisal (+4 mm), and overjet (+6.3 mm) (Table). Additionally, a slight decrease of the mandibular plane angle (−0.9°), a decrease of the gonial angle (−2.6°), and a proclination of both maxillary (+3.0°) and mandibular incisors (+1.3°) were observed (Table; Fig 6). Pretreatment and posttreatment cephalometric tracings were superimposed at the cranial base, maxilla, and mandible (Fig 6), and 3-dimensional models were superimposed at the anterior cranial base, as shown in Figures 7 and 8.[14,15] The color-coded maps of the maxilla show in dark red anterior and inferior displacements (Fig 7). Three-dimensional superimposition with semitransparency showed the marked anterior displacement of the maxilla and the small inferior displacement of the middle third, as well as backward and downward displacement of the mandible (Fig 8). Also at the level of the soft tissues, the superimposition with semitransparency (Fig 8) showed marked forward projections of the nose and surrounding tissues. The soft tissue contour of the mandible also showed backward and downward displacement. The quantitative cephalometric measurements had a reduction of the nasiolabial angle of 8.9°.

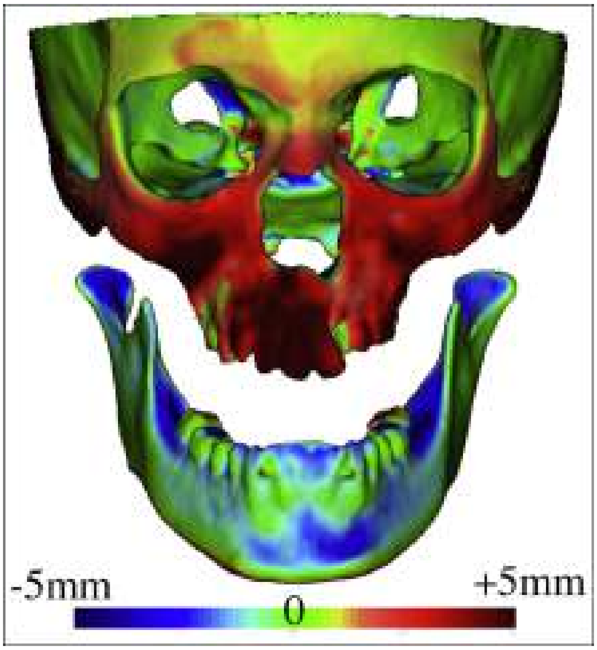

Fig 7. Closest point color map images of the maxilla and mandible created from the posttreatment over pretreatment surface models superimposed at the cranial base. The color bar ranges from −5 mm (*shades of blue*) to +5 mm (*shades of red*). The *green* color corresponds to 0 mm changes.

DISCUSSION

This is the first report on BAMP therapy in patients with oral clefts. When compared with results of other therapies in cleft patients, after 16 months of full-time Class III elastics wear, the increases in the SNA angle and A-Na Perp distance indicate anterior maxillary displacement greater than conventional facemask therapy,[7,16] and similar to the alternate rapid maxillary expansions and constrictions protocol.[17] When compared with the results of BAMP therapy in noncleft patients, the amount of maxillary advancement in this patient was similar.[10,18,19]

In our clinical protocol, we used finger springs lingual to the maxillary incisors to prevent incisor retroclination during maxillary protraction with BAMP due to lip tension and scarring. Even with activating the finger springs to jump the bite, the maxillary incisor tipping remained stable (Fig 6, *B*). These findings are different from facemask therapy that produces maxillary incisor proclination in patients with and without CLP.[20-22] The mandibular

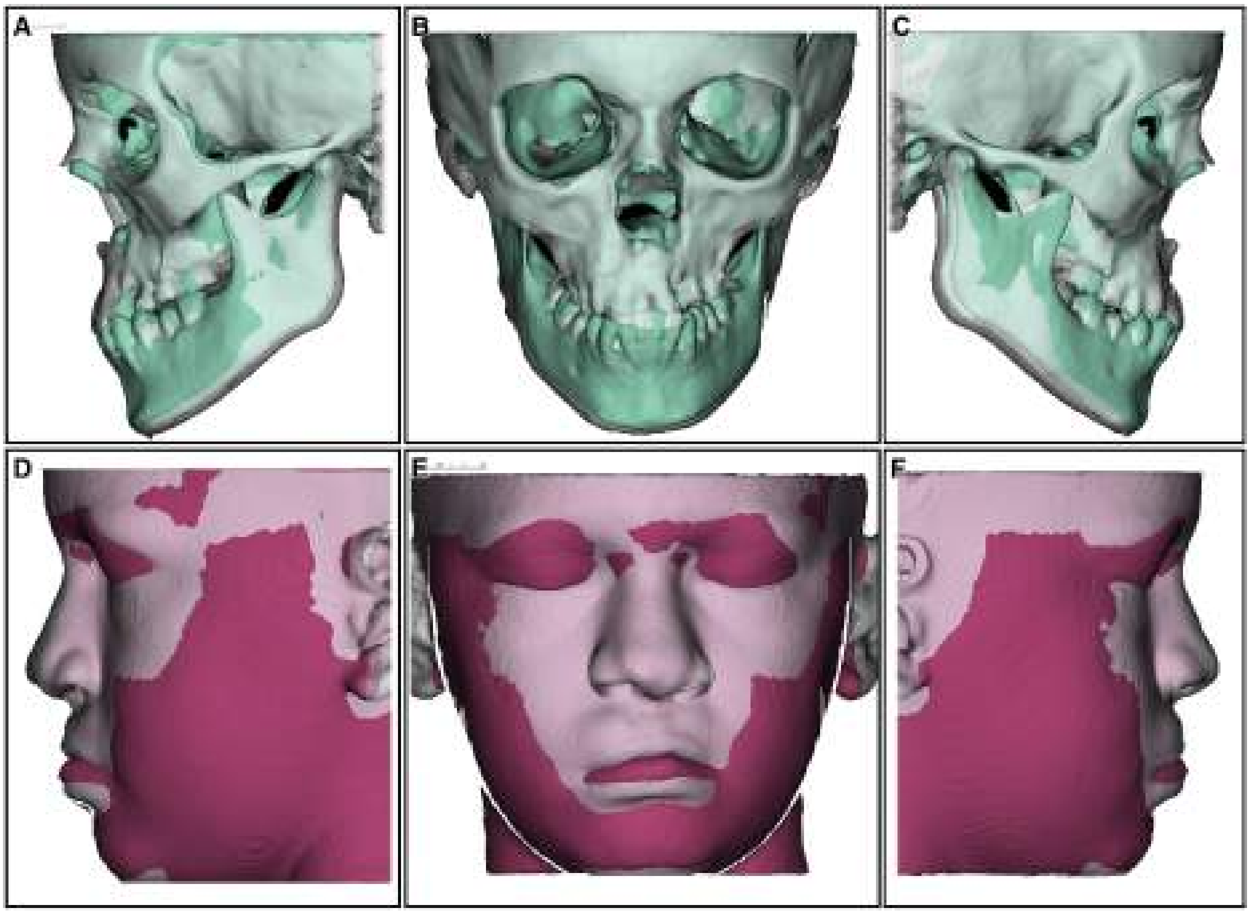

Fig 8. Anterior and lateral views of tridimensional superimposition with transparency at the anterior cranial base. Pretreatment is in *green* for the hard tissue and *pink* for the soft tissue, and posttreatment is in semitransparent *white*. **A-C,** Hard tissue, the maxilla moved forward and outward, and the mandible moved slightly down and backward; **D-F,** soft tissue, upper lip and nose tip moved forward.

incisors also showed a slight buccal tipping of 1.3° similar to the BAMP protocol in noncleft patients, but in contrast to the findings with facemask therapy in patients with clefts.[20-22] The slight buccal tipping in this patient could be explained by the overjet correction and forces generated by the tongue.

As the orthopedic BAMP forces are applied on the posterior part of the maxilla, some rotation of the right and left maxillary segments medially could have been expected, with compression of the bone graft. The clinical findings in semitransparent overlays showed a small tendency of axial rotation of the right and left maxillary segments, and no adverse effects were noted at the bone graft. Clinically, the use of a transpalatal arch in this patient may have helped to stabilize anchorage for the orthopedic traction of the maxilla.

Facemask therapy in patients with CLP can cause a significant clockwise rotation of the mandible with downward and backward movement of the chin.[7,20] These effects result from reaction forces delivered by the chincup of the facemask. In this case report, BAMP therapy produced a slight backward movement of the chin (Table), not as the result of clockwise rotation of the mandible but, rather, a decrease of the gonial angle (Table) and backward displacement of the gonial landmarks and the posterior border of the ramus and condyles (Figs 6 and 7). Studies on BAMP in noncleft patients also showed slight decreases of the mandibular plane angle and the gonial angle.[12,19]

Scar tissue and fibrosis from the primary lip and palate repairs did not seem to interfere with maxillary protraction in this patient but might influence relapse after the active phase. Therefore, active retention with nighttime wear of Class III elastics was recommended. These short-term results do not allow us to determine whether orthognathic surgery will be avoided at the end of growth. A previous study showed that patients with CLP are potential bullying victims while waiting for orthognathic surgery.[23] For this reason, the improvement of facial esthetics at an early age is a remarkable achievement in the difficult period of adolescence and may have an impact on the self-esteem of this patient.

CONCLUSIONS

Bone-anchored orthopedics caused significant maxillary advancement and restriction of the forward projection of the mandible resulting in marked improvement of the occlusion and overall facial esthetics. This new protocol seems promising for patients with CLP to reduce the need for a LeFort I osteotomy after growth. However, short-term and long-term prospective studies on larger samples are needed to further evaluate the effects of bone-anchored orthopedics in these patients and to define guidelines for clinical use.

REFERENCES

1. Semb G. A study of facial growth in patients with unilateral cleft-lip and palate treated by the Oslo CLP team. Cleft Palate Craniofac J 1991;28:1-21.
2. Capelozza L, Normando AD, da Silva 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; 33:51-6.
3. Will LA. Growth and development in patients with untreated clefts. Cleft Palate Craniofac J 2000;37:523-6.
4. Westwood PV, McNamara JA Jr, Baccetti T, Franchi L, Sarver DM. Long-term effects of Class III treatment with rapid maxillary expansion and facemask therapy followed by fixed appliances. Am J Orthod Dentofacial Orthop 2003;123:306-20.
5. Baccetti T, Reyes BC, McNamara JA Jr. Craniofacial changes in Class III malocclusion as related to skeletal and dental maturation. Am J Orthod Dentofacial Orthop 2007;132:171.e1-12.
6. Kapust AJ, Sinclair PM, Turley PK. Cephalometric effects of face mask/expansion therapy in Class III children: a comparison of three age groups. Am J Orthod Dentofacial Orthop 1998;113:204-12.
7. Buschang PH, Porter C, Genecov E, Genecov D, Sayler KE. Face mask therapy of preadolescents with unilateral cleft-lip and palate. Angle Orthod 1994;64:145-50.
8. Dogan S. The effects of face mask therapy in cleft lip and palate patients. Ann Maxillofac Surg 2012;2:116-20.
9. Susami T, Okayasu M, Inokuchi T, Ohkubo K, Uchino N, Uwatoko K, et al. Maxillary protraction in patients with cleft lip and palate in mixed dentition: cephalometric evaluation after completion of growth. Cleft Palate Craniofac J 2014;51:514-24.
10. De Clerck HJ, Cornelis MA, Cevidanes LH, Heymann GC, Tulloch CJ. Orthopedic traction of the maxilla with miniplates: a new perspective for treatment of midface deficiency. J Oral Maxillofac Surg 2009;67:2123-9.
11. Baccetti T, Franchi L, McNamara JA. The cervical vertebral maturation (CVM) method for the assessment of optimal treatment timing in dentofacial orthopedics. Semin Orthod 2005;11:119-29.
12. Nguyen T, Cevidanes L, Paniagua B, Zhu H, Koerich L, De Clerck H. Use of shape correspondence analysis to quantify skeletal changes associated with bone-anchored Class III correction. Angle Orthod 2014;84:329-36.
13. De Clerck H, Nguyen T, de Paula LK, Cevidanes L. Three-dimensional assessment of mandibular and glenoid fossa changes after bone-anchored Class III intermaxillary traction. Am J Orthod Dentofacial Orthop 2012;142:25-31.
14. Cevidanes LH, Heymann G, Cornelis MA, DeClerck HJ, Tulloch JF. Superimposition of 3-dimensional cone-beam computed tomography models of growing patients. Am J Orthod Dentofacial Orthop 2009;136:94-9.
15. Cevidanes LH, Motta A, Proffit WR, Ackerman JL, Styner M. Cranial base superimposition for 3-dimensional evaluation of soft-tissue changes. Am J Orthod Dentofacial Orthop 2010;137(Suppl):S120-9.
16. Shanker S, Ngan P, Wade D, Beck M, Yiu C, Hägg U, et al. Cephalometric A point changes during and after maxillary protraction and expansion. Am J Orthod Dentofacial Orthop 1996;110: 423-30.
17. Liou EJ, Tsai WC. A new protocol for maxillary protraction in cleft patients: Repetitive weekly protocol of alternate rapid maxillary expansions and constrictions. Cleft Palate Craniofac J 2005;42: 121-7.
18. Cevidanes L, Baccetti T, Franchi L, McNamara JA Jr, De Clerck H. Comparison of two protocols for maxillary protraction: bone anchors versus face mask with rapid maxillary expansion. Angle Orthod 2010;80:799-806.
19. De Clerck H, Cevidanes L, Baccetti T. Dentofacial effects of bone-anchored maxillary protraction: a controlled study of consecutively treated Class III patients. Am J Orthod Dentofacial Orthop 2010;138:577-81.
20. Vieira GD, de Menezes LM, de Lima EM, Rizzatto S. Dentoskeletal effects of maxillary protraction in cleft patients with repetitive weekly protocol of alternate rapid maxillary expansions and constrictions. Cleft Palate Craniofac J 2009;46:391-8.
21. Ngan P, Hägg U, Yiu C, Merwin D, Wei SH. Treatment response to maxillary expansion and protraction. Eur J Orthod 1996;18: 151-68.
22. Nguyen T, Cevidanes L, Cornelis MA, Heymann G, de Paula LK, De Clerck H. Three-dimensional assessment of maxillary changes associated with bone anchored maxillary protraction. Am J Orthod Dentofacial Orthop 2011;140:790-8.
23. Tiemens K, Nicholas D, Forrest CR. Living with difference: experiences of adolescent girls with cleft lip and palate. Cleft Palate Craniofac J 2013;50:e27-34.