Case Report

Nonsurgical orthodontic treatment using bone-anchored maxillary protraction in a patient with unilateral cleft lip and palate

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ABSTRACT

Class III malocclusion due to a retrognathic maxilla is common in patients with cleft lip and palate. Skeletally anchored maxillary protraction using screw-anchored mini-plates combined with intraoral elastics has shown promising results in achieving orthopedic changes and maintaining the outcome until the completion of the growth. This case report presents the course of treatment in a patient with unilateral cleft lip and palate and multiple congenitally missing teeth treated with bone-anchored maxillary protraction until the end of growth. Four mini-plates (Bollard plates) were used during comprehensive fixed orthodontic treatment to protract the dentition and close the space where teeth were missing, extrude the canine, and force eruption of the second premolar using extension arms and cantilevers. A 2-year follow-up at age 17 showed stable occlusion and maintenance of soft tissue results. Bone-anchored maxillary protraction treatment in a patient with cleft lip and palate demonstrates proper orthopedic results and could be a viable alternative to orthognathic surgery. (*Angle Orthod*. 0000;00:000–000.)

KEY WORDS: Maxillary protraction; Cleft lip and palate; Skeletal anchorage; Tooth agenesis

INTRODUCTION

Class III malocclusion is a common characteristic in patients with cleft lip and palate (CLP). Maxillary deficiency in the transverse and anteroposterior dimensions results from inherited growth deficiencies and postsurgical scar tissue after lip and palate repair. This maxillary growth impediment increases progressively until the end of the growth period.^{1,2}

Early orthopedic treatment geared toward growth modification using a protraction facemask aims to address maxillary hypoplasia and redirect and restrain mandibular growth.^{3,4} However, long-term stability of the treatment effects with protraction facemask remains questionable,

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and a significant variation of relapse has been reported.⁵ Despite early treatment, several studies reported that 20% to 76.5% of patients with CLP would eventually need orthognathic surgery.⁶

Heavy orthodontic forces delivered by the elastics to tooth-borne appliances elicit undesirable side effects, such as dental compensations and increased vertical facial dimension. Additionally, treatment relies mainly on unpredictable patient compliance.⁷

Over the past decade, skeletally anchored maxillary protraction has been the treatment of choice in patients with CLP. Bone anchored maxillary protraction (BAMP), described by De Clerck et al.,⁸ uses four modified miniplates as anchorage: two are placed in the infrazygomatic crest, and another two are inserted between mandibular lateral incisors and canines, paired with intermaxillary elastics.

Dental abnormalities are frequently present in cleft patients; congenitally missing and supernumerary teeth, usually at the cleft site, are the most common dental anomalies. Maxillary lateral incisors are frequently absent, peg-shaped, or abnormally positioned, mainly due to the bone defect in that area.⁹ During adolescence, when the patient is undergoing fixed orthodontic treatment, a decision must be made to manage the spaces where teeth are missing and establish a functional occlusion.

This case report demonstrates the course of treatment in a Class III patient with unilateral CLP and multiple congenitally missing and malpositioned teeth. The various

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stages of orthodontic treatment are described from childhood to postadolescence to correct maxillary hypoplasia in the transverse and sagittal dimensions and achieve functional occlusion without orthognathic surgery after growth completion or the need for extensive restorative/ prosthodontic care for the missing teeth.

Case Report Diagnosis

An 8-year-old female patient with a repaired left unilateral cleft lip and palate was referred to the Orthodontic Department at the University of Connecticut Health Center for dentoalveolar preparation of the cleft for a secondary alveolar bone graft. Lip repair was done at 10 weeks. Soft and hard palatal repair was performed with doubleopposing Z-plasty at 10 months of age. Extraoral examination showed a concave profile with severe midface deficiency (G'-SN-Po': -13°), depressed left ala of the nose, retruded upper lip, and a repaired cleft lip on the left side (Figure 1a). The upper lip was symmetric in length, and no other visible asymmetry was observed in the frontal view. Intraorally, the patient had a Class III molar relationship and an interarch relationship of GOSLON (Great Ormond Street, London, and Oslo) Index 5,¹⁰ with a severe negative overjet of approximately -10 mm, deep overbite, and bilateral posterior crossbite with significant arch collapse at the canine area. The maxillary arch had a V shape with slight disruption of the arch form at the cleft site. At age 8, the central incisors and first molars had erupted. A residual left oronasal fistula and the alveolar cleft were present (Figure 1a, b). Her panoramic radiograph revealed a complete alveolar cleft between the maxillary left central incisor and canine, missing maxillary lateral incisors and maxillary right second premolar, and a peg-shaped maxillary left second premolar. The maxillary left first premolar was positioned horizontally in the alveolar bone (Figure 1a).

The lateral cephalometric x-ray (Figure 2a and Table 1) taken at age 11 demonstrated a severe skeletal Class III malocclusion (ANB: -9.8°, Wits appraisal: -11.8 mm) due to maxillary hypoplasia (SNA: 69.5°), reduced height of the midface, with a steep mandibular plane angle (FMA: 33°). The mandible was well-positioned in the sagittal plane (SNB: 79°) and the upper and lower incisors were retroclined (U1 to SN: 89°, IMPA: 78°). Conebeam computed tomography (CBCT) taken 18 months after alveolar bone grafting and at the beginning of comprehensive fixed orthodontic treatment (age 11), demonstrated continuity of the maxillary dental arch at the cleft site with the permanent canine erupting into the bone grafted area (Figure 2a).

Treatment Objectives

The treatment objectives were determined based on the stage of the orthodontic treatment. The primary aim before the secondary alveolar bone graft was to expand the upper arch anteriorly, increasing the width of the cleft for graft placement. The objectives of the comprehensive orthodontic treatment were to correct the sagittal maxillary deficiency, improve facial esthetics, establish an orthognathic facial profile, and achieve functional occlusion.

After maxillary expansion and subsequent alveolar bone graft, orthopedic maxillary advancement was planned via BAMP, followed by fixed orthodontic appliances to address multiple agenesis and impacted teeth. The mini-plates were used to achieve maximum anchorage in the maxilla to protract the entire maxillary dentition and close all missing tooth spaces.

Treatment Alternatives

Based on the severity of the malocclusion, two treatment options for phase II of orthodontic treatment were presented. The first option was maxillary orthopedic correction using the BAMP technique, followed by orthodontic treatment to substitute the canines for the missing laterals, protract the maxillary right molars, and close the space of the missing maxillary right premolar. The second option was orthodontic treatment, followed by orthognathic surgery to address the skeletal discrepancy after the completion of growth. Regarding the treatment options for the missing teeth, extracting the ankylosed maxillary right primary second molar, maintaining space and replacing it with an implant was presented; this option would keep the occlusion with the antagonist teeth for the mandibular second molar at the end of treatment. After explaining the prognosis and risks of each plan, the patient and her parents opted for the first option to avoid orthognathic surgery and dental implants as much as possible.

Treatment Plan and Progress

To prepare the cleft site before the secondary alveolar graft, at the age of 8 and 6 months, a fan-shaped expander was cemented on the maxillary arch to differentially expand the anterior area with minimal effects in the molar region. The expander fulcrum was placed slightly distal to the first molars. The active expansion was 5 weeks (0.25 mm or one turn daily), followed by 6 months of retention with the appliance. A total of 8 mm of expansion at the primary canines was achieved. Six months later (at the age of 9), the patient received a secondary alveolar bone graft from the Iliac crest. A transpalatal arch with anterior arm extensions was placed as a fixed retainer. The patient was followed every 6 months and, at age 11 (Figure 2a, b), she was referred to Boston Children's Hospital for the placement of the Bollard mini-plates (Bollard, Tita-Link, Brussels, Belgium).

The Bollard mini-plates were placed bilaterally at the infrazygomatic crest of the maxilla above the permanent maxillary first molars and between the roots of



Figure 1. (a) Intraoral and extraoral photographs and panoramic X-ray at age 8 before the alveolar bone graft. (b) Study models at age 8 before the alveolar bone graft.



Figure 2. (a) Intraoral and extraoral photographs, and x-rays at age 11, at the beginning of the BAMP treatment. (b) Study models at the beginning of BAMP treatment. BAMP indicates bone-anchored maxillary protraction.

 Table 1.
 Cephalometric Analysis: Pretreatment (Before BAMP) and Posttreatment

Measurement	Norm	Pretreatment (Before BAMP)	Posttreatment
SNA (°)	82	66.5	74.0
SNB (°)	80	75.0	71.7
ANB (°)	3	-9.2	2.3
Wits (mm)	-1.0	-11.3	-0.9
NA-APg (°)	8.0	-19.0	2.4
SN-GoMe	33.0	38.1	38.9
U1 to SN	102.4	81.0	92.0
L1 to GoGn	90.0	82.4	89.4
Upper lip to E-line	-4.0	-7.7	-6.0
Lower lip to E-line	-2.0	3.0	0.4
Facial convexity (°) (G'-SN-po')	12	-13.0	3.9

the permanent mandibular canines and lateral incisors. Three weeks after surgery, full-time Class III elastics connecting the maxillary and mandibular mini-plates were initiated with 150 g of force on each side. The patient was instructed to change the elastics at least twice daily.

After one month, the force level was increased to 200 g per side. Three months after the surgery, maxillary protraction continued with 250 g of force until the end of active orthopedic treatment. When the patient reached an incisor edge-edge relationship 6 months after surgery (Figure 3), a removable biteplate with anterior springs was delivered to remove the incisor interferences and facilitate bite jumping. After 10 months of treatment, the patient achieved positive overjet. At this point, maxillary incisors were bonded with preadjusted 0.022×0.028 -inch slot brackets (Ormco, Brea, CA), and alignment of the anterior maxillary teeth was initiated. The maxillary left first premolar was exposed surgically, and the ankylosed

right second primary molar was extracted. Two weeks after exposure, a transpalatal arch was cemented on the first molars. An anteriorly extended arm with a hook on the mesial aspect was fabricated on the right side using 0.017×0.025 stainless steel wire. The arm was engaged in the mini-plate tube and used as skeletal anchorage for protracting the right posterior teeth using an elastomeric chain (Figure 4). On the left side, a 0.017×0.025 titanium molybdenum alloy cantilever engaged in the mini-plate tube was used to extrude the partially erupted left canine. The maxillary left first premolar was brought into the arch and protracted into the canine position with a stainless-steel extension arm from the miniplate. The same mechanics were used to protract the left peg-shaped second premolar and the first molar.

Spaces were successfully closed after one year, and the patient continued wearing Class III elastics from the mini-plates. The fixed orthodontic appliances were removed temporarily 20 months after bonding per the neurologist's request, as a diagnostic magnetic resonance imaging scan needed to be taken after a grand mal seizure. Treatment resumed 5 months after to complete the space closure and finalize the occlusion. After 34 months, the fixed orthodontic appliances were debonded. However, the patient continued to wear Class III elastics at night as part of the active retention regimen. A fixed retainer was bonded in the mandibular arch, and a thermoplastic retainer was delivered for maxillary retention. The patient was referred for prosthetic restoration of the peg-shaped maxillary left second premolar (Figure 5a).

RESULTS

The total treatment time, including BAMP and fixed orthodontic treatment, was 46 months. The initial phase



Figure 3. Intraoral photos six months after placement of Bollard plates with the removable bite plate.

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Figure 4. Cantilevers were used from the Bollard plates to protract the maxillary molar on the right side and to extrude the maxillary left canine.

before bone grafting was 6 months, followed by 2 years of retention before the comprehensive phase. Posttreatment records (Figure 5a, b) showed significant improvement in the soft tissue profile, showing a convex profile with a normal upper lip position and contour. The concavity of the paranasal and the malar area improved significantly. On smiling, increased incisal show and reduced buccal corridors were evident due to forward movement of the maxilla. The upper lip and nose were translated forward, and the facial convexity (NA-APg) improved by 21.4° (Table 1).

After removing the fixed orthodontic appliances, a second CBCT was taken for cephalometric analysis and three-dimensional (3D) superimposition. Initial CBCT, taken before mini-plate placement (T1) and the final CBCT (T2), were superimposed to evaluate the skeletal changes due to treatment. Data from each CBCT were stored as Digital Imaging and Communications in Medicine files. Model construction, cranial base registration, and visualization were performed using methods described by Bazina et al. Three-dimensional surface models of the anatomic region of interest were constructed from T1 (before treatment) and T2 (posttreatment) images using ITK-SNAP (open-source software; http:// www.itksnap.org).¹¹ The 3D models were then superimposed using 3D Slicer software (https://www.slicer.org). The "region of interest" method was adopted, and the cranial base was used as reference. Quantifying the differences was then done by measuring the distance between the two surface models using closest-point color maps. A final 3D color map (Figure 5) illustrated the skeletal and dental changes. The heatmap (Figure 5a) shows a significant forward movement of the maxilla (approximately 5 mm, 6.9 mm at A point) and a moderate posterior displacement of the mandible (<5 mm).

Mild anterior displacement was also noticed at a higher level of the midface with anterior translation of the zygomatic arches and inferior orbital rim (about 5 mm). Clockwise rotation of the mandible with posterior displacement of the ramus was evident in the superimposition.

Intraorally, a Class I canine, Class II molar relationship (a full-tooth Class II on the right side and full-cusp Class II on the left side) with ideal overjet and overbite was achieved. All of the spaces for the congenitally missing teeth were closed. Due to the space closure in the maxillary arch, the mandibular right second molar was left without an antagonist; a retainer with occlusal coverage was used to prevent extrusion. The esthetic outcome of canine substitution was favorable; the canine gingival level resided at the zenith level of the central incisors. Enameloplasty was performed on the incisal edges and buccal surfaces of the canines to create the lateral incisor shape. A porcelain crown was placed over the peg-shaped left maxillary second premolar.

The posttreatment panoramic x-ray showed adequate bone height around the left canine and incisors without any evidence of bone resorption. Proper root parallelism was evident, except for minor mesial tipping of the maxillary right second molar, with no sign of root resorption.

Two-year follow-up at age 17 (Figure 6) demonstrated stable results, maintained soft tissue convexity, and good interdigitation with a slight reduction in the overjet of about 0.5 mm. The patient had the mini-plates removed after the completion of growth at age 18.

DISCUSSION

BAMP was the treatment of choice for this patient, considering the severe Class III malocclusion associated with CLP and the patient's age. The BAMP approach was selected to avoid extraoral appliances and enhance



Figure 5. (a) Posttreatment extraoral and intraoral photographs, x-rays, lateral cephalometric radiograph, and 3D superimpositions. (b) Final study models after debonding.



Figure 6. Extraoral and intraoral photos at 2-year follow-up.

patient compliance, allowing continuation of the orthopedic treatment over the adolescent growth period. The amount of maxillary forward displacement in the patient was significantly greater compared to that reported in previous studies; the range of reported maxillary forward movement in patients with CLP was between 1.5 mm^{12,13} and 3.17 mm.¹⁴ The 3D superimposition and the constructed color map showed significant forward movement (approximately 5 mm) of the maxilla and maxillary dentition; point A moved forward 6.9 mm, the upper incisor displaced anteriorly about 9 mm, and proclined 10°. Nguyen et al. reported a range of 1.45 to 8.5 mm forward displacement of the maxilla in noncleft patients.¹⁵ The significant forward movement of the maxilla, in this case, could be attributed to the duration of BAMP treatment, as the patient continued nighttime wear of the elastics during active fixed orthodontic treatment.

The forward displacement was also evident in the midface and zygomatic arch, which significantly improved

the patient's malar eminence projection and paranasal hollowing (Figure 5). The inferior orbital rim and zygomatic arch moved forward significantly, ranging from 4 to 6 mm on the cleft and noncleft sides (Figure 5). As described by Heymann et al., due to the proximity of the mini-plates to the zygomaticomaxillary sutures, the applied force was closer to the zygomatic and infraorbital area, which resulted in more significant changes compared to the conventional facemask treatment.¹⁶

The superimposition showed a clockwise rotation of the mandible, which resulted in a 3.2° increase in steepness of the mandibular plane. Studies evaluating the effect of BAMP in cleft patients reported clockwise rotation of the mandible corpus as a contributing factor favoring the improvement of facial convexity.¹⁷ Despite the increase in the mandibular plane angle, the gonial angle decreased by 4°; other authors have reported this effect after BAMP treatment in cleft patients.^{8,13} As described by Nguyen, patients treated with BAMP exhibit a swing-back

effect of the mandible due to a distal inclination of the ramus combined with closure of the gonial angle and counterclockwise rotation of the mandible.¹⁸ The backward chin movement, in this case, was a combination of the swing-back effect, noticeable in CBCT superimposition (Figure 5), and slight clockwise rotation of the mandibular plane. Considering the significant deep overbite and an over-closed appearance of the mandible at the beginning of the treatment, the clockwise rotation was favorable esthetically and helped further improve the profile convexity. The maxilla moved downward, and the palatal plane rotated counterclockwise (CCW) 5° due to the force vector passing occlusal to the maxillary center of resistance. The CCW rotation is more significant in facemask treatment due to the additional chin cup effect of the appliance on the mandible.

After the alveolar bone graft, the left canine erupted into the grafted area. The final CBCT showed a normal bone crest level and adequate cortical bone thickness. In patients with CLP with agenesis of the lateral incisors, the eruption of canines into the grafted area is considered the gold standard for maintaining the alveolar graft.¹⁹

Expanding the boundaries of conventional tooth movement was possible due to the versatility of skeletal anchorage. Significant protraction of the maxillary dentition was achieved without compromising incisor position by designing different auxiliaries and reinforcing the anchorage directly and indirectly from the mini-plates. Considering the ankylosis of the maxillary right primary second molar, extraction to prevent a vertical bone defect was recommended; however, protracting the maxillary molars left the mandibular right second molar without an antagonist which increased the chance of extrusion. To prevent this, the patient was instructed to wear the retainer with occlusal coverage in the lower arch, and implant placement for the maxillary right second molar after growth is completed was recommended.

The panoramic x-ray showed bodily movement of the posterior teeth with normal alveolar bone architecture. The auxiliary arm on the right side was designed to apply the protraction force at the level of first molar furcation to prevent tipping. On the left side, the maxillary left first premolar was protracted through indirect anchorage provided by the ipsilateral mini-plate.

Another advantage of BAMP treatment was the possibility of maintaining the protraction force until the end of mandibular growth and managing the relapse tendency. In this patient, the mini-plates were removed after 2 years of follow-up, at age 18.

CONCLUSIONS

 Early treatment with BAMP can have a significant impact on facial esthetics and the profile by improving severe maxillary deficiency in patients with cleft lip and palate.

- This treatment should be considered as an alternative to orthognathic surgery later in life, especially in patients with CLP.
- Mini-plates can be used as anchorage to close spaces efficiently where teeth are missing, without compromising the position of the incisors.

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DISCLOSURES

The authors declare that there is no conflict of interest.

The first author's (Niloufar Azami) role was conceptualizing and drafting the case report; she led manuscript writing and will address reviewers' feedback. The second author's (Philip Farha) role was to prepare the first draft of the manuscript and create 3D superimpositions. The third author (Flavio Uribe) supervised the orthodontic treatment of the case and contributed significantly to its clinical aspect and the critical review of the manuscript.

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