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Case Report

Maxillary distraction osteogenesis for correction of maxillary retrognathia in cleft lip and palate patients: a case series

Burçak Kaya[1], Seçil Çubuk[2]*, Sıdıka Akdeniz[2], Burak Bayram[2]

[1]Department of Orthodontics, [2]Department of Oral and Maxillofacial Surgery, School of Dentistry, Ankara, Turkey

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***Correspondence:**
Dr. Seçil Çubuk,
E-mail: secilcubuk@hotmail.com

ABSTRACT

The treatment protocol of patients having maxillary retrognatia is presented in this case series. Maxillary expansion, secondary alveolar bone grafting and/or oronasal fistula closure were performed in 6 cleft lip and palate patients. Preoperative simulation of Le Fort I osteotomy and adaptation of maxillary distractors were realized with stereolytographic cranium models. Intraoral maxillary distraction was applied under general anesthesia. A single unit acrylic occlusal splint was ligated to maxillary dental arch and worked as a guide during entire activation and consolidation period of distraction osteogenesis. Rigid internal fixation plates were placed in the osteotomy sites at the end of consolidation period. The amount of maxillary advancement was between 5-14 mm. Satisfying occlusal and skeletal relationships were obtained in all patients. Advancement of maxilla by distraction osteogenesis resulted in gradual formation of bone at the osteotomy line and enhanced treatment outcome in patients with cleft lip and palate.

Keywords: Cleft lip palate, Maxillary retrognathia, Distraction osteogenesis, Orthodontics

INTRODUCTION

Maxillary retrognathia is a frequent symptom observed in patients with cleft lip and palate (CLP). Advancement of maxilla to a large extent is required for elimination of maxillary retrognathia and can be realized with different techniques. However, advancement of maxilla using Le Fort I osteotomy with single step orthognathic surgery is challenging in CLP patients because of palatal scar contracture, upper lip tension and decreased postoperative stability due to large amount of anteroposterior discrepancy.[1,2] Distraction osteogenesis (DO) allows gradual lengthening of bone and soft tissue structures. Therefore, it is advantageous in these patients, since orthognathic surgery results with relapse when great amount of surgical movement is performed.

Intraoral distraction devices are more esthetic and tolerable compared to extraoral ones. Additionally, their long-term stability was found higher than the external distractor appliances. These advantages make intraoral distractors more feasible in CLP patients, despite the difficulties to maintain the 3D vector control by intraoral distractors.[3]

The aim of this case series is to present the treatment protocol of 6 cleft lip and palate patients using intraoral distraction devices to correct maxillary retrognatia.

CASE REPORT

Six nonsyndromic patients are included in this case series. All treatments started with maxillary expansion and followed by alveolar bone grafting if required. Patients who had oronasal fistulas received palatal repair surgeries. Cone beam computed tomography images of the patients were obtained and 3D cranium models were fabricated. Adaptation of the distractors and simulation of

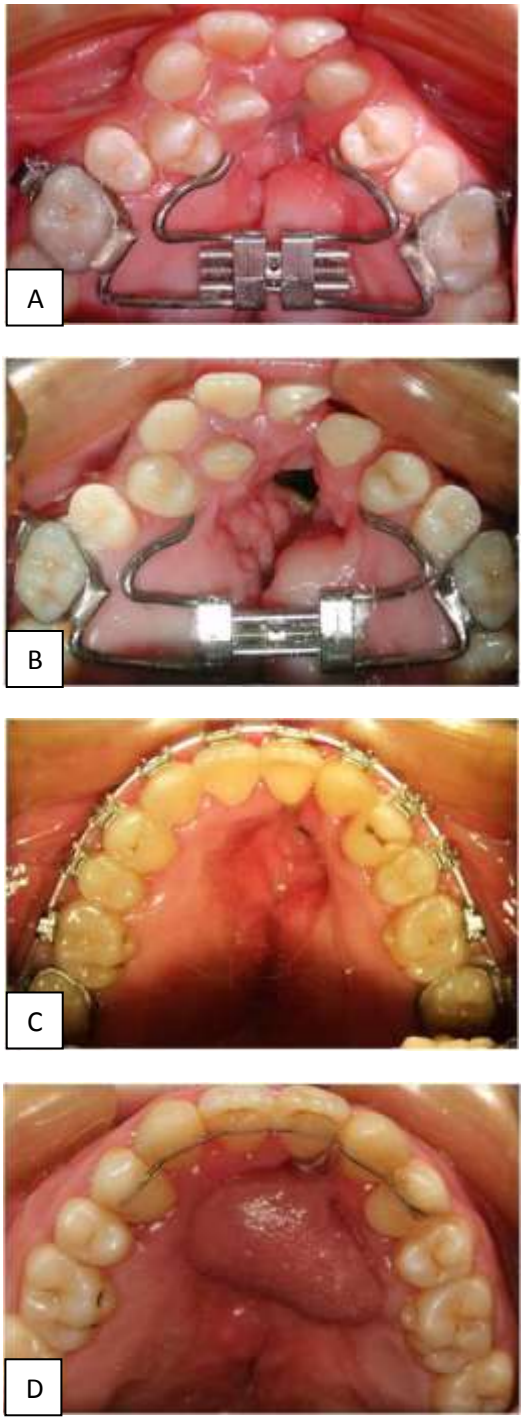

Figure 1: Maxillary occlusal view: (A) At the beginning of maxillary expansion. (B) At the end of maxillary expansion. (C) After extraction of right lateral incisor followed by alignment of maxillary teeth and the first palatal repair surgery. (D) After completion of orthodontic treatment and palatal tongue flap surgery.

Le Fort I osteotomy were realized with these models. An acrylic splint was attached to upper teeth to prevent medial deviation of lateral maxillary segments. Then, internal bone-borne maxillary distraction device (Synthes GmbH, Switzerland) was inserted under general anesthesia. A modified incision preserving an anterior vascular pedicle was performed. Alignment rods were used to check the advancement vector. The intraoral distractors were activated after a latency period of 7 days. The rhythm and rate of distraction was 2 times a day, obtaining 1 mm of advancement each day. The consolidation period lasted for 3 months and rigid fixation plates (Synthes GmbH, Switzerland) were placed in the osteotomy sites at the end of this period.

Figure 2: Facial profile of the patient: (A) Before distraction osteogenesis. (B) After distraction osteogenesis.

Case 1

Case 1 was a 15 years old female with unilateral complete cleft lip and palate. Her major problems were maxillary retrusion, narrow anterior maxilla, upper anterior crowding and oronasal fistula.

Her treatment started with maxillary expansion using a hyrax appliance. It was followed by alveolar bone grafting from iliac crest and two palatal repair surgeries involving a tongue flap (Figure 1). The treatment was followed by distraction osteogenesis by using intraoral distractors to advance the maxilla. The maxilla moved 5 mm forward, a positive overjet was obtained and the facial profile improved.

Case 2

Case 2 was a 17 years old female with bilateral complete cleft lip and palate. Her major problems were severe maxillary retrusion, absence of premaxilla, anterior openbite, missing lower posterior teeth and a strained upper lip.

The maxillary arch was expanded and the teeth were aligned with fixed orthodontic appliances. The maxilla was advanced by distraction osteogenesis using intraoral distractors. The maxilla moved 13 mm forward and a positive overjet was obtained after placement of fixed dentures. The facial profile improved, but was still defective, as the upper lip remained strained (Figure 2).

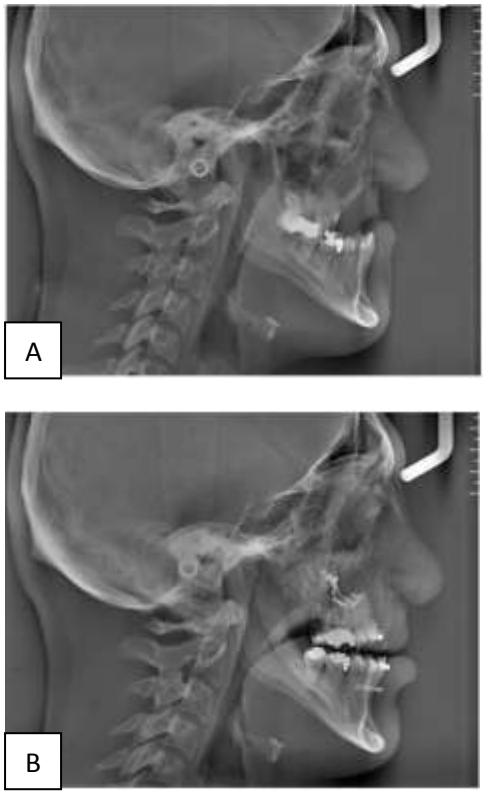

Figure 3: Lateral cephalometric radiograph of the patient: (A) Before orthodontic treatment and distraction osteogenesis. (B) After distraction osteogenesis.

Case 3

Case 3 is a 25 years old male with bilateral complete cleft lip and palate. His major problems were severe maxillary retrusion, absence of premaxilla, severe maxillary transvers deficiency and a strained upper lip.

The maxillary arch was expanded with hyrax and the teeth were aligned with fixed orthodontic appliances. Similar with the former patients the maxilla was advanced with distraction osteogenesis by using intraoral distractors. The maxilla moved 14 mm forward, however a positive overjet and overbite could not be obtained as minor maxillary retrusion was still present and the maxilla was rotated anteriorly (Figure 3). Despite the facial profile improved, it is still concave and imperfect. The orthodontic treatment of the patient proceeded with orthodontic compensation to obtain a positive overjet, overbite and proper occlusal functions.

Case 4

Case 4 is a 19 years old female with unilateral complete cleft lip and palate. Her major problems were maxillary retrusion, narrow anterior maxilla, missing maxillary incisors.

The maxillary arch was expanded and the teeth were aligned. The same intraoral distraction protocol used in other patients was applied and the maxilla was advanced. The maxilla moved 13 mm forward, positive overjet was obtained and the facial profile improved (Figure 4).

Figure 4: Facial profile of the patient: (A) Before distraction osteogenesis. (B) After distraction osteogenesis.

Case 5

Case 5 is a 15 years old female with bilateral complete cleft lip and palate. Her major problems were maxillary retrusion, missing upper incisors, a mobile premaxilla and maxillary transvers deficiency.

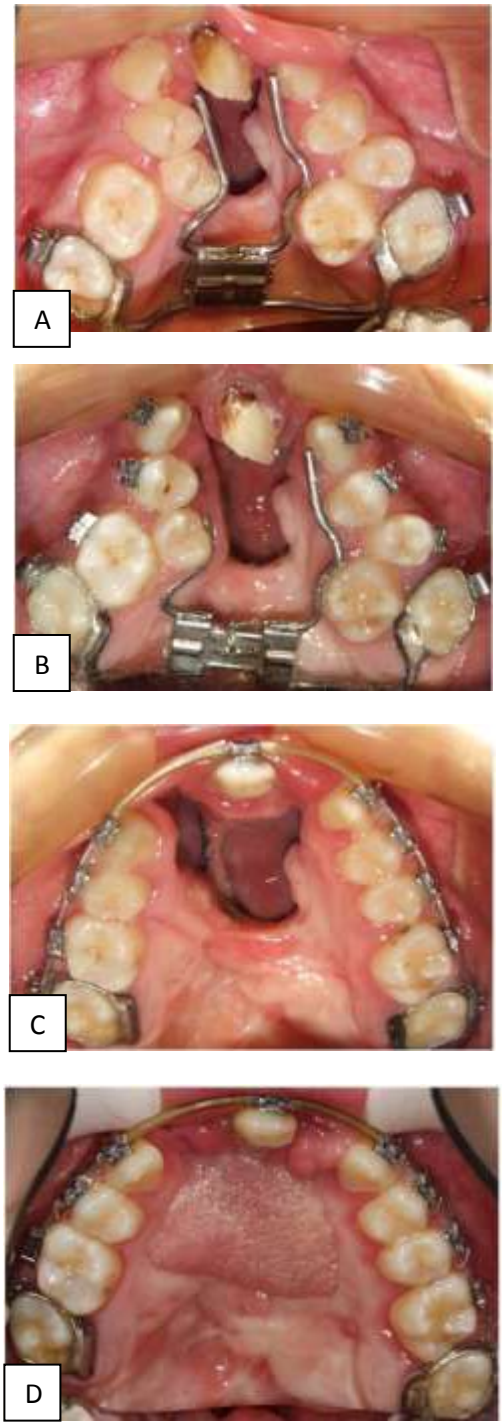

Figure 5: Maxillary occlusal view: (A) At the beginning of maxillary expansion. (B) At the end of maxillary expansion. (C) After extraction of right second premolar and alignment of maxillary teeth. (D) After palatal tongue flap surgery.

A hyrax appliance was used for expansion and the teeth were aligned. The wide palatal fistula which enlarged after maxillary expansion was eliminated using a tongue flap and bone graft obtained from iliac crest was placed in the alveolar cleft area (Figure 5). Intraoral distractors were placed and the maxilla was advanced by distraction osteogenesis similar with the other patients. The maxilla moved 10 mm forward, a positive overjet and a balanced facial profile was obtained.

Case 6

Case 6 is an 18 years old female with unilateral complete cleft lip and palate. Her major problems were severe maxillary retrusion, missing upper incisors and severe transvers deficiency in both two arches.

The maxillary transvers deficiency was corrected with a hyrax appliance. The teeth were aligned with expansion of upper and lower arches. The maxilla was advanced with distraction osteogenesis by using the similar treatment protocols explained for the other patients. The maxilla moved 14 mm forward, a positive overjet and a balanced profile was obtained (Figure 6).

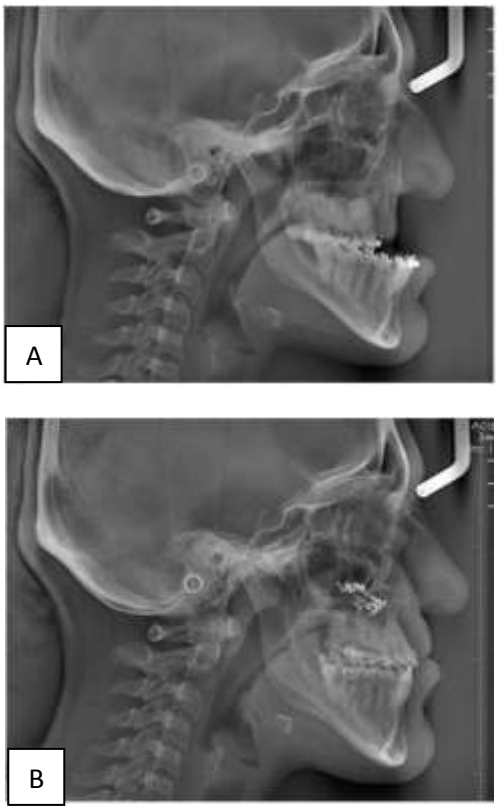

Figure 6: Lateral cephalometric radiograph of the patient: (A) Before distraction osteogenesis. (B) After completion of orthodontic treatment.

Maxillary retrusion was eliminated and sagittal skeletal discrepancy was corrected or reduced at the end of the distraction protocols in all patients. The amount of maxillary advancement was between 5 to 14 mm. Satisfying occlusal relationships were obtained and soft tissue profiles were improved as a result of the skeletal corrections achieved by distraction osteogenesis. Advancement of maxilla by distraction osteogenesis resulted in gradual formation of bone at the osteotomy line and enhanced treatment outcome in patients with cleft lip and palate.

DISCUSSION

Individuals with CLP receive many surgical interventions during their lifetime. Class III malocclusions requiring surgical intervention was reported as 25-50% in CLP patients.[4]

A randomized clinical trial conducted by Chua et al reported 37% horizontal and 65% vertical relapse in point A in CLP patients who had undergone LeFort I osteotomies for maxillary advancement.[1] The relapse rate of DO was also evaluated in their study and reported to be 8.24%, which was significantly lower than the relapse rate reported after orthognathic surgeries.

Meazzini et al reported that DO is the most appropriate treatment modality for adult CLP patients who need maxillary advancement more than 10 mm.[5] On the other hand, DO is also recommended for less than 10 mm advancement of maxilla in CLP patients who have extreme scarring. In this case series, DO for less than 10 mm maxillary advancement was applied in only one patient. The reason for preference of DO rather than orthognathic surgery was the numerous previous hard palate repair surgeries she had undergone which led excessive scar formation in her palate.

External distractors are generally used for treatment of severe mid-facial deficiencies in craniofacial syndromes. However, application of external distraction devices in most cleft lip and palate patients is stated to be unnecessary. Internal distraction devices are reported to be more tolerable for patients with their relatively smaller size. They are also more stable in structure compared to external distraction devices which are less rigid due to the wires they involve. Consequently, internal devices act like rigid fixation plates during consolidation period.[6]

One of the disadvantages of internal distraction devices is their lack of ability to move maxilla truly in 3 dimensions. Clinicians must make meticulous planning of maxillary advancement and adjust the distractors on 3D cranium models preoperatively to determine the vector of distraction precisely.[6] In our case series, the treatment plans of all patients were simulated on 3D cranium models. Bending and adaptation of the distraction devices were completed on 3D models preoperatively. Acrylic splints were attached to upper teeth preoperatively to prevent medial deviation of lateral maxillary segments and avoid collapse of maxillary arch. Additional stab incision was made to place the rods of the distractors and move maxilla forward with an ideal sagittal direction vector in the male patient, as his mustache can easily mask the scar tissue.

Drew et al suggested that adequate ossification between two bone segments must be confirmed with CT scan before removal of distractors at the end of consolidation period.[6] They applied bone grafts and placed rigid fixation plates at the time of distractor removal in patients who had poor bone formation and bone stock. On the other hand, Tabrizi et al evaluated the impact of rigid fixation plates on stabilization rate and they observed no statistical difference in stabilization rate regarding horizontal and vertical relapse after 18 month follow up.[7] In our case series, L miniplates were applied bilaterally to all patients during removal of the distractors at the end of consolidation period to prevent any relapse risk.

In conclusion, advancement of maxilla by distraction osteogenesis resulted in gradual formation of bone at the osteotomy line and enhanced treatment outcome in patients with cleft lip and palate in this case series.

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