Case Report

A case of cleft lip and palate with severe maxillary retrognathism treated by distraction osteogenesis and custom made intraoral rapid maxillary expansion appliance: A 2-year follow-up of retention

ABSTRACT

Herewith, a case is reported of an adolescent female patient with maxillary retrognathism due to restricted growth arising out of the previous cheiloplasty and palatoplasty surgeries treated for cleft lip and palate. She also presented an oroantral fistula in the scarred tissues of the palatal region. There was anterior crossbite and distorted occlusion in the anterior segment with crowding and open bite. There is maxillomandibular discrepancy of 6 degrees . The distraction osteogenesis was performed so as treat the maxillary hypoplasia. This allows undermanding adaptation of the soft-tissue structures to the modification in the skeletal structures as a result of surgical procedures and ensures long-term stability. A custom made intraoral rapid maxillary expansion device was prepared utilizing the hyrax screw for the distraction of the bony segments. At the end of the treatment and a retention period of 24 months, the patient exhibited improved facial profile and hence esthetics.

Keywords: Distraction osteogenesis, HYRAX, maxillary retrognathism, secondary grafting

INTRODUCTION

Patients with primary cleft lip and palate (CLP) and their subsequent surgical interventions leads to disturbed maxillary growth resulting in deformities of the jaw and malocclusion.[1] Skeletal and dental growth in the transverse and anteroposterior planes is affected and especially the maxillary arch is collapsed because of these early surgical procedures leading to Class III malocclusions.[2] In such cases, the functional and esthetic demands in CLP patients are achieved through surgery by advancing the maxilla.[3-5]

Distraction osteogenesis (DO) by gradual mechanical traction of bone segments at an osteotomy site is being used to treat abnormalities such as maxillomandibular hypoplasia, facial asymmetry, and congenital micrognathia in the craniofacial complex.[6,7]

In the present case, the patient with a skeletal Class III malocclusion resulting from hypoplastic maxilla with a normal mandible was presented and corrected with an anterior maxillary DO, and a custom made rapid maxillary expansion tooth-borne device using a rapid expansion screw.

CASE REPORT

A 12-year-old female patient presented to the Department of Orthodontics in with a chief complaint of impaired esthetics. She presented with a dental history of feeding problem due to oronasal communication. Cheiloplasty and

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palatoplasty were performed at 5 months and 12 months of age, respectively. Her prognosis was poor according to Goslon Yardstick.[8,9] Extraoral examination revealed a concave profile with a Class III skeletal pattern having a maxillary hypoplasia. Intraoral examination revealed that there is a prominent oronasal fistula perforation seen on the left side within palatal scar tissue. She had a Class I molar relationship on the right and left sides, anterior crossbite with reverse overjet of 4 mm, Unilateral open bite on the left side, palatally positioned 12, partially erupted 22 and incisors are in Class III relationship [Figure 1]. The mandibular arch is relatively undisturbed.

Cephalometric analysis showed Class III skeletal pattern with relatively retrognathic maxilla, vertical growth pattern, clockwise rotation of maxilla and mandible, retroclined and retruded upper incisors, normally inclined lower incisors and Class III soft-tissue profile [Figure 2 and Table 1].

Figure 1: Initial facial and intraoral photographs of our patient who was diagnosed with angles Class I malocclusion, a skeletal Class III jaw relationship with a concave profile, unilateral anterior crossbite, open bite, distorted Anterior segment with crowding, occlusal distortion, a left-sided cleft lip and palate and oronasal fistula at 13 years of age

Treatment plan

The main treatment objective was to improve the facial esthetics and profile by correcting the Class III skeletal pattern and alignment of maxillary and mandibular dentition. Anterior maxillary DO was planned using an intraoral device to obtain correct skeletal relationship and adequate arch length to align the maxillary teeth. The main advantage of this procedure is decrease in the amount of relapse, because of soft-tissue histogenesis along with incremental traction of anterior maxilla.

Treatment alternatives

Le Fort I osteotomy and anterior maxillary osteotomy are discussed as treatment alternatives but ruled out considering the amount of bone shift required to correct the crossbite.

Treatment progress

The treatment was started when the patient is at 12 years 7 months of age. Surgery was aimed to provide alveolar bone support in the region of 22, 23 through autogenous bone graft raised from anterior iliac crest. The cleft region was

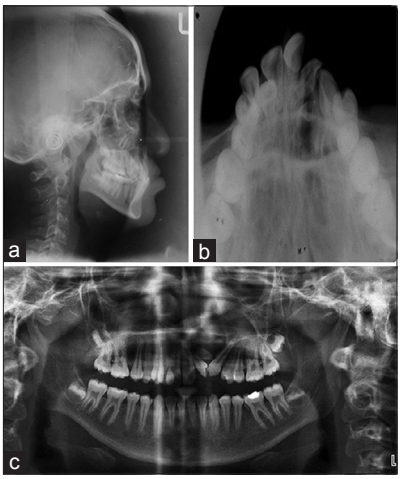

Figure 2: Pretreatment radiographs. (a) Lateral cephalogram. (b) Occlusal radiograph. (c) Orthopantomogram

Table 1: Comparison of pre-, post-treatment and post-retentive cephalometric parameters

		Skeletal		
Landmarks	**Parameter**	**Pretreatment**	**Posttreatment**	**Postretention**
Maxilla	SNA (°)	74	80	80
	N perpendicular to Point'A" (mm)	−8	−3	−3
	Effective maxillary length (mm)	74	81	80
Mandible	SNB (°)	78	79	79
	N perpendicular to Pog (mm)	−10	−5	−6
	Effective mandibular length (mm)	105	107	107
Skeletal pattern	ANB (°)	−4	1	1
	Convexity at pt A (mm)	9	3	4
	Wits appraisal (mm)	−9	−2	−3
Growth pattern	FMA (°)	32	31	33

S: Sella, N: Nasion, A: A-point, B: B-point, FMA: Angle between mandibular plane and Frankfort Horizontal (FH) plane

untouched for 6 months for the graft take up [Figure 3]. After this secondary graft placement orthodontic correction was carried out to align upper and lower the teeth. The period of presurgical alignment and leveling was about 6 months. The surgical site for DO was decided in the interdental region between the maxillary first and second premolars on both sides. This surgical site provides the advantage of avoiding maxillary sinus exposure and taking more anchorage support from second premolars and molars on either side. Treatment simulation was made using dolphin digital imaging software (Paterson and Kelly, version 11.2) showed 7 mm advancement is required to achieve the adequate amount of overjet. To avoid the relapse after the surgery, it is planned to advance up to 9 mm [Figure 4].

After the treatment simulation was finalized, a tooth-borne distraction device was custom made using a hyrax screw positioned in anteroposterior direction. The posterior anchorage unit was modified into an occlusal bite plane with cold cure acrylic. Anterior segment was consolidated by making a cap splint to prevent the cleft segment from parting away [Figure 5].

Osteotomies for the anterior maxillary distraction were carried out under general anesthesia. Horizontal corticotomy cut was sited from premolar to premolar region 5 mm above the root apices. Vertical corticotomy cut was made interdentally between premolars on either side. Vertical cuts are extended to the palatal region on either side, and a greenstick fracture of the anterior maxillary segment had been done. Mucoperiosteal flap was closed primarily, custom-made distraction device was cemented to the teeth using glass ionomer cement and checked for its activation [Figure 6].

After a latency period of 5 days, distraction was started from the 6th day after the surgery and was carried out at a rate of 1 mm per day with a rhythm of 0.5 mm twice a day. Distraction is done for 9 mm and the consolidation period took 2 months. Bone formation in the distraction site was confirmed on radiographs. At the end of this consolidation period, the distractor is removed [Figure 7].

Postsurgical orthodontics was performed by placing the brackets on the maxillary teeth. Lateral open bite was developed in the region of 22, 23, and 24. This lateral open bite was corrected using the box elastics. The first premolar on either side is completely distalized and is included in the anchor unit. This is followed by distalization of canines on both sides. Palatally blocked out 12 is slowly aligned into this space. Left lateral incisor was in the cleft region; hence, very mild forces are used to derotate it [Figure 8].

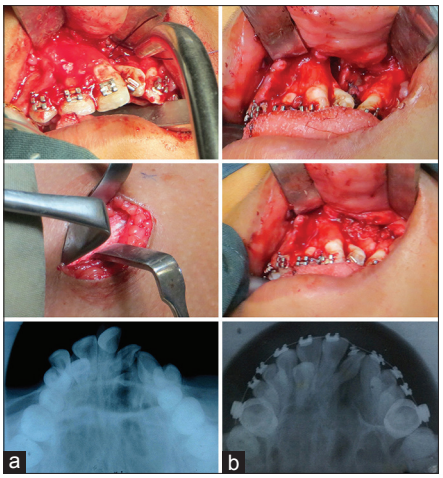

Figure 3: Secondary alveolar bone grafting. (a) Occlusal radiograph at pre grafting stage and (b) Occlusal radiograph 6-month postgrafting stage

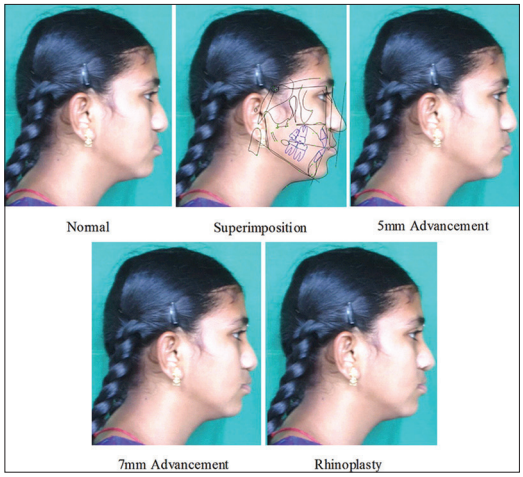

Figure 4: Treatment simulation using dolphin digital imaging software (Paterson and Kelly, version 11.2)

Figure 5: Fabrication of Intraoral distraction appliance with hyrax screw and mock surgery

Retention is provided using upper fixed retainer from premolar to premolar and a Begg wrap on retainer, lower

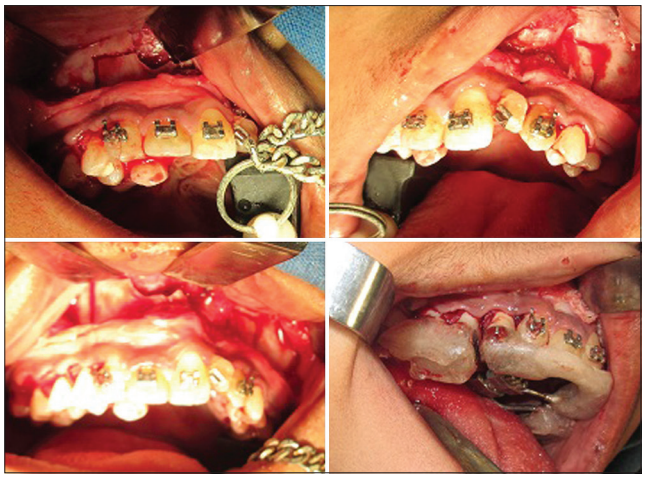

Figure 6: Surgical corticotomy and fitting of intraoral appliance for distraction

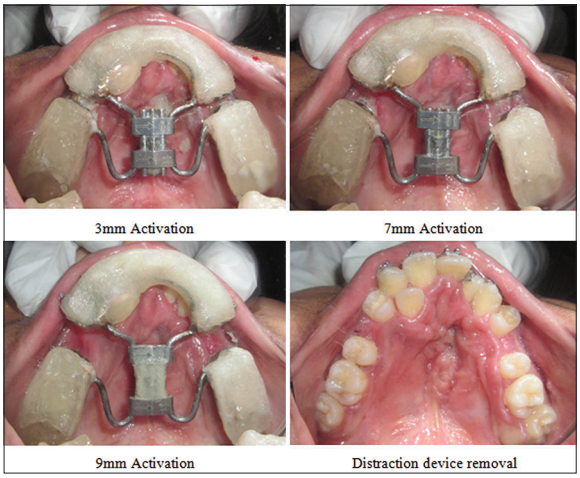

Figure 7: Distraction device activation and removal

Figure 8: Posttreatment facial and intraoral photographs of the patient with well-aligned maxillary and mandibular arches, change in facial profile, and improved esthetics

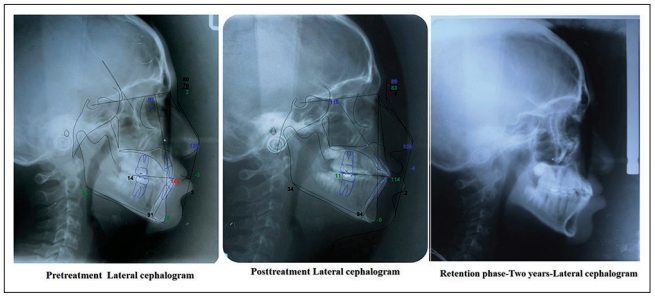

Figure 9: Comparison of pretreatment, posttreatment, and retention phase lateral cephalograms

retention is attained by fixed retainer from canine to canine. Superimposition of the pre- and post-treatment lateral cephalograms showed a linear movement of maxillary anterior segment by 7 mm [Figure 9 and Table 1].

Treatment results

The results showed an improvement in skeletal, dental, and soft-tissue parameters. Anterior crossbite has been corrected. Overjet is improved from −4 mm to +2 mm. ANB angle was increased from −4° to 1°. Angle of inclination increased from 74° to 87°. Superimposition of the pre- and post-treatment lateral cephalograms showed a linear movement of maxillary anterior segment by 7 mm. Upper incisor was proclined little more than the ideal inclination to achieve acceptable interincisal relationship. The follow-up retention after 24 months showed a stabilized skeletal pattern with no signs of relapse [Figures 9 and 10].

DISCUSSSION

Le Fort I osteotomy is the most commonly used surgical technique to treat the maxillary hypoplasia to reestablish facial proportion and occlusion in patients who have completed their growth.[10,11] Scarred soft tissue caused by the preceding operation makes it difficult to mobilize the complete maxilla in CLP patients. Relapse and occlusal instability is the most common disadvantage associated with Le Fort I maxillary advancement in CLP patients when compared with noncleft patients who have maxillary hypoplasia.[12,13] DO advanced the field of maxillofacial surgery because of its versatility, simplicity, and possibility of avoiding bone grafts, infections, blood transfusions, or intermaxillary fixation for long periods of time.[7] In addition, DO has an advantage of soft-tissue augmentation simultaneously with the bone.[7]

DO, provides skeletal advancement as well as soft-tissue histogenesis simultaneously.[14] Due to these advantages, over other surgical techniques DO became an effective and efficient surgical method for CLP patients.[15]

Long-term stability after maxillary advancement with DO in CLP patients appears to be a fairly stable procedure.[13,14]

Figure 10: Retentive phase 24-month extraoral and intraoral radiographs

The relapse rate is higher in DO with an external distracter than DO with an internal distracter. The other advantage with DO with internal devices is, it does not require much of anchorage support from teeth to advance the maxillary anterior segment, some of the disadvantages are difficulty in vector control and minimum advancement of maxilla.[15]

CONCLUSION

DO technique can effectively transpose the maxilla forward and downward in moderate and severe maxillary retrusion. In the present case, custom made intraoral device was designed for the advancement of the maxilla. Accordingly, the patient after a retention period of 24 months showed well-consolidated maxillary bone with improved soft-tissue adaptation and esthetics.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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