Original Article - Prospective Study

Maxillary Distraction Osteogenesis in Unilateral Cleft Lip and Palate Patients with Rigid External Distraction System

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Abstract

Aim: Distraction osteogenesis (DO) is a treatment option for patients with maxillary hypoplasia secondary to cleft lip and palate (CLP). **Purpose:** The aim of this study is to present a technique for maxillary DO using Le Fort I osteotomy with rigid external distraction (RED) system. **Subjects and Methods:** The patient presented in this paper was an Asian female with CLP aged 13 years and 6 months. She presented with severe midfacial deficiency with a Class III dental malocclusion with a negative overjet and concave facial profile. Cone-beam computed tomography images were recorded preoperatively and the operation performed involved a high Le Fort I osteotomy. The appliance fabricated was banded to upper first molars used for anchorage of the RED system. Distraction of the maxilla was initiated after 7-day latency period. **Results:** Postoperative cephalometric analysis showed maxillary advancement anteriorly and superiorly, the total distraction treatment period was 10 days. The maxillary advancement was 10.5 mm and the SNA angle increased from 67.5° to 77.9°. Furthermore, the ANB angle changed from −9.8° to 1.6° and the occlusion changed from Class III to Class I. The profile of the face changed from concave to convex and a much better esthetic result was achieved. **Conclusion:** The study suggests RED system to be a reliable alternative procedure for the treatment of midfacial hypoplasia with or without cleft. Furthermore, it minimizes the risk of the surgical procedure and shortens the operating time.

Keywords: Cleft, orthodontics, rigid external distraction, surgery

Introduction

Cleft lip and palate (CLP) is a birth defect that happens when the fetus' tissue of the lip and/or the palate do not fuse between the 5th and 12th week of pregnancy.[1] Clefts affect approximately 1 in every 600 new born babies.[2] However, the prevalence varies across geographic areas and ethnicity. A nonsyndromic CLP incidence occurs approximately 1.30 per 1000 for Asians, 1.41 per 1000 among Japanese, and 1.21 per 1000 among Chinese population[3] whereas 1.12 per 1000 live birth in European and American communities.[4] CLP affects the face and oral cavity not only in terms of esthetic and functional problems but also psychological aspects.[5] Multidisciplinary treatment and wide range of supportive care are employed on CLP patient to restore this defect to the normal range as possible, during patient lifetime.

Maxillary hypoplasia in CLP deformities results from congenital reduction in midfacial growth and the effects of the surgical scar from CLP repair.[2] Turvey *et al*. suggested that this disproportionate jaw growth is the biologic consequence of prior surgical intervention for closure of the soft tissues and is not related to the congenital cleft deformity.[6]

Midface hypoplasia has the following characteristics: concave facial profile, inverted nasal tip, wide alar base, acute nasolabial angle, and excessive exposure of sclera. Intraoral findings are anterior and posterior crossbite, CLP, accentuated curve of Spee, Class III dental malocclusion, multiple missing teeth, oronasal communication, and residual cleft. Speech disturbances are also usually present due to velopharyngeal incompetency and oronasal communication.[7]

Midfacial hypoplasia is commonly treated by performing conventional Le Fort surgery to displace the maxilla anteriorly and stabilization afterward with rigid fixation. However, several limitations have been reported with this procedure.[8] Therefore,

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Figure 1: Frontal and lateral views of 13 years and 6 months Asian female

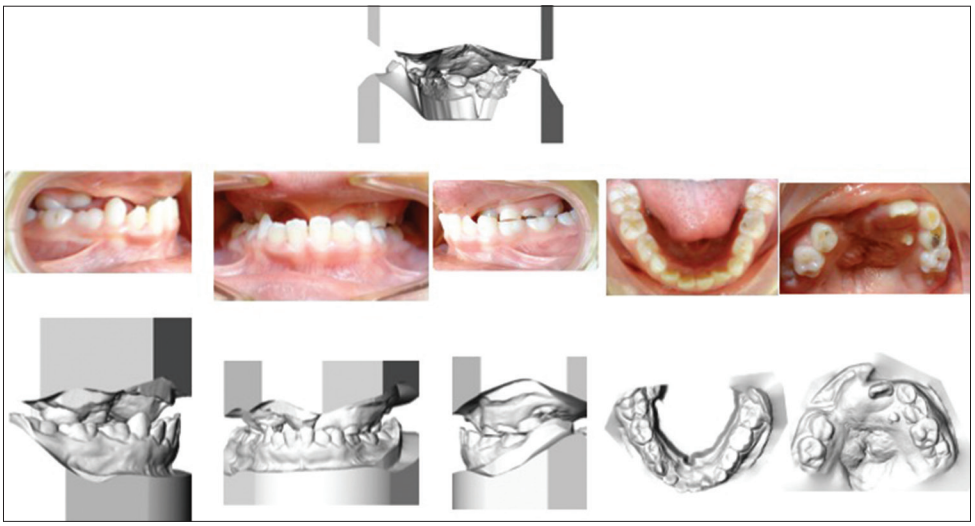
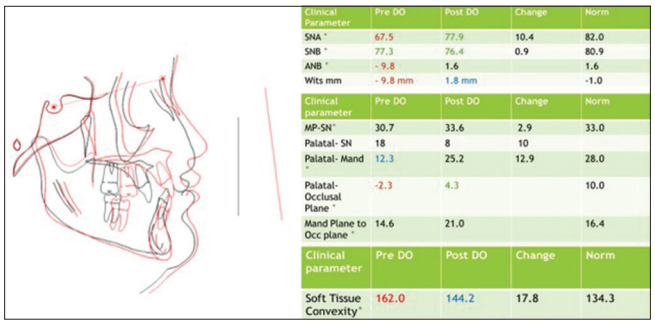


Figure 2: Models and intraoral photograph



Clinical Parameter	Pre DO	Post DO	Change	Norm
SNA °	67.5	77.9	10.4	82.0
SNB °	77.3	76.4	0.9	80.9
ANB °	- 9.8	1.6		1.6
Wits mm	- 9.8 mm	1.8 mm		-1.0

Clinical parameter	Pre DO	Post DO	Change	Norm
MP-SN °	30.7	33.6	2.9	33.0
Palatal- SN °	18	8	10	
Palatal- Mand °	12.3	25.2	12.9	28.0
Palatal-Occlusal Plane °	-2.3	4.3		10.0
Mand Plane to Occ plane °	14.6	21.0		16.4

Clinical parameter	Pre DO	Post DO	Change	Norm
Soft Tissue Convexity °	162.0	144.2	17.8	134.3

Figure 3: Superimposition and analysis table

distraction osteogenesis (DO) has become an alternative technique to treat craniofacial dysplasia to overcome some of this limitations. DO is a surgical technique for the growth of new bone through the application of tensile stresses to a preexisting tissue lying between two bone ends. This technique of skeletal regeneration generally involves four stages: (1) creation of a full osteotomy or corticotomy, (2) latency period or callus formation between two bones ends, (3) distraction period or bone formation by callus stretching, (4) consolidation or new bone tissue maturation.[6,9]

Polley and Figueroa introduced maxillary distraction osteotomy in 1997 using a rigid external distraction (RED) device.[10] RED system has provided a significant alternative to treat severe midface hypoplasia with minimally invasive procedure, especially those secondary to orofacial clefting with the absence of maxillary and alveolar bone and the presence of scaring.[1] The gradual advancement and lengthening of the soft tissue in distraction osteogeneses procedure offer lower relapse rates 5.56%–10%[1] and more advancement.

In this case report, we describe the treatment with the combination of DO using RED system and orthodontics for a girl with a midfacial hypoplasia associated with a nonsyndromic unilateral CLP.

Subjects and Methods

A 13 years and 6-month-old Asian female visited the orthodontics department at UAB, with a chief complaint of feeding and speaking difficulties with a compromised esthetics and occlusion. The clinical and radiographic examination (all records obtained with patient consent) indicated that she had unilateral repaired CLP associated with midface hypoplasia. Her medical history included right-sided CLP grafting surgery at age of 2 years, left iliac bone graft to maxilla for reconstruction, and closure of the oronasal fistula at 7 years old. She had unknown familial history since the patient was adopted.

The pretreatment facial photographs presented asymmetrical face with inverted nasal tip toward the left side, a chin point that was deviated 2 mm toward the right-side, wide alar base, decrease the lower face height, short upper lip, scar on the right-side of the upper lip due to early closer to the cleft lip, everted upper and lower lip, concave facial profile, acute nasolabial angle, no incisal display in the smile frontal photograph, lower dental midline deviated to the right from the facial midline [Figure 1].

Functional examination presented with normal temporomandibular joint function, speech disturbances, feeding difficulties due to velopharyngeal incompetency, and oronasal communication.

Intraoral examination showed that the dental arch form was ovoid, asymmetrical, and small upper arch in the three dimensions, retroclined upper left incisor, supernumerary tooth, deficiency in the alveolar bone due to palatal cleft, Class III molars relationships, negative 10 mm overjet, 8 mm overbite, a transversal and sagittal deficiency maxillary arch [Figure 2]. The lateral cephalometric analysis [Figures 3 and 4] showed an SNA angle of 67.5°, an SNB angle of 77.3°, and ANB angle of −9.8°. The mandibular plane angle was 30.7°. The maxillary left incisor was palatally inclined at an angle of 92.9° to the SN plane. Interincisors angle was 147.6°; soft tissue convexity was 162.0°.

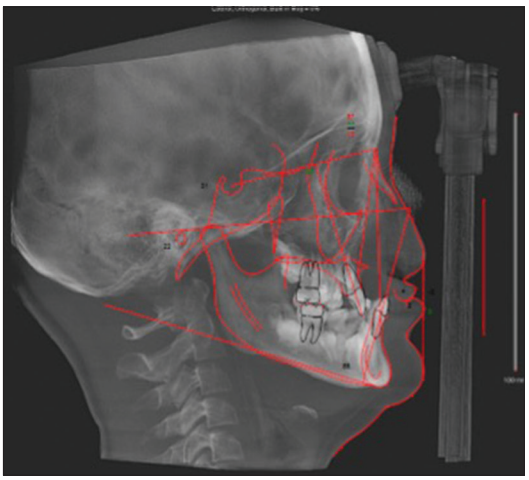


Figure 4: Predistraction lateral cephalometric

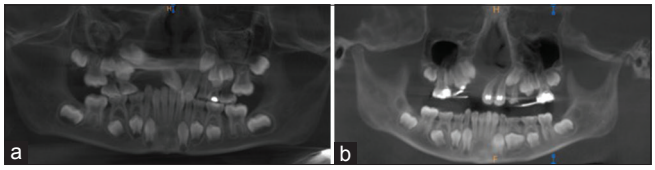


Figure 5: (a) Initial panoramic radiograph. (b) Predistraction progress panoramic radiograph

The panoramic radiograph presented the right-side cleft palate with fistula, ectopic eruption of upper permanent first molars, undermined distal of upper primary second molars due to ectopic eruption of upper permanent first molars, alveolar bone deficiency due to CLP, supernumerary tooth at the upper left side, congenitally missing of upper laterals [Figure 5].

Clinically, the patient's periodontal tissues were healthy. Three-dimensional computed tomography images showed anteroposterior deficiency of maxilla with narrow and short upper arch [Figure 6].

Treatment objective

Because of the midface, hypoplasia patient and her family concern about facial appearance. Thus, the treatment objectives were established as follows; (1) correct the patient's facial concavity by advancing the maxilla and coordinate the facial, maxillary, and mandibular relationship; (2) correct the skeletal Class III anteroposterior jaw relationship; (3) correct the occlusal plan to achieve dental Class I canine and molar; (4) correct the retroclined incisors position and achieve an ideal overjet and overbite relationship.

Treatment alternatives

Based on the treatment objectives, the following treatments' alternatives were considered: (1) Le Fort I maxillary orthognathic surgery advancement with rigid fixation, (2) Le Fort I osteotomy

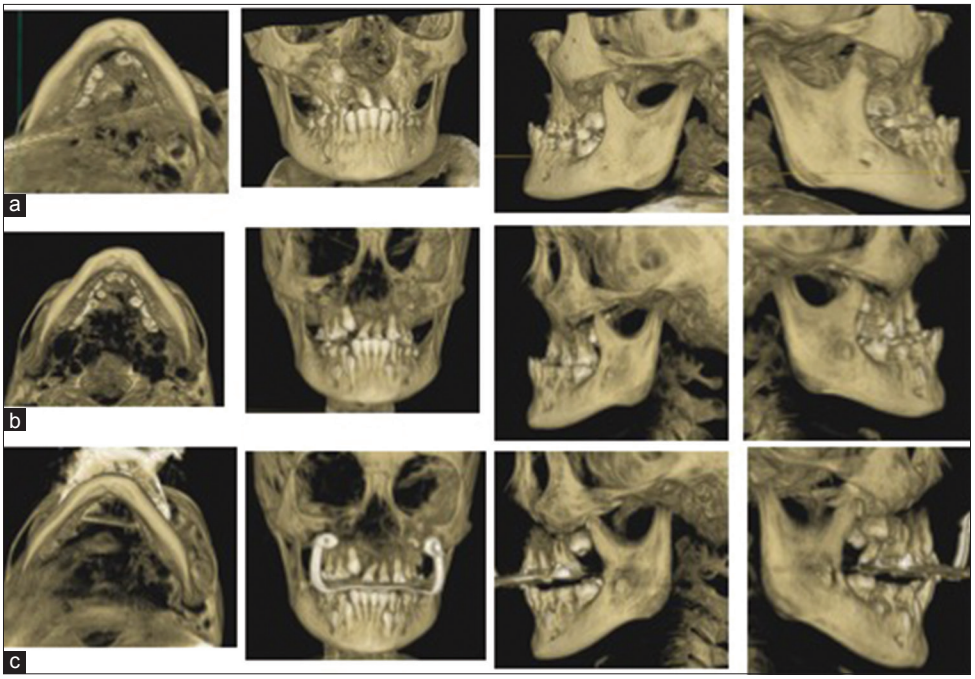


Figure 6: Cone-beam computed tomography images (a) predistraction. (b) Predistraction progress. (c) Postdistraction

Figure 7: Predistraction extraoral and intraoral photographs

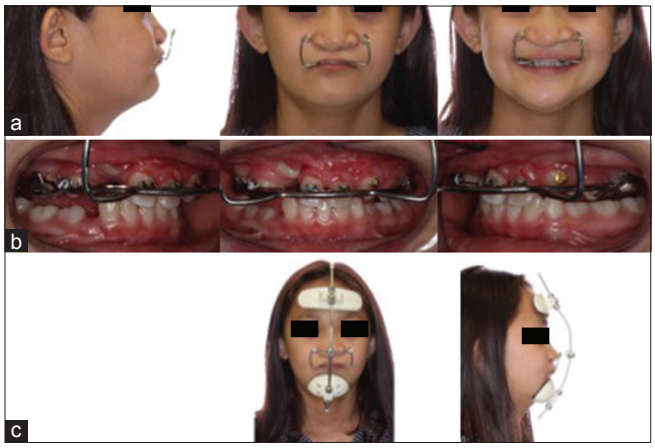


Figure 9: (a) Postdistraction extraoral view. (b) Postdistraction intraoral view. (c) Retention period by reverse headgear

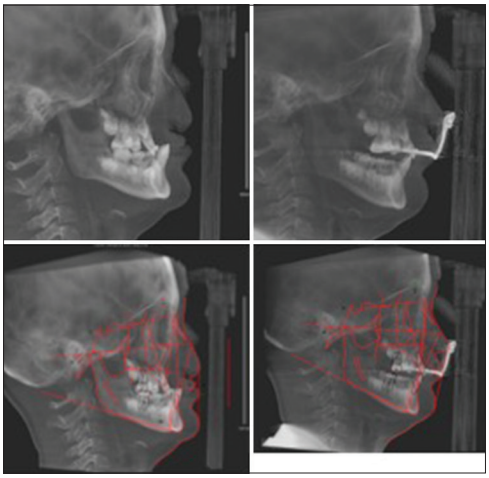


Figure 11: Pre- and post-treatment lateral cephalometric

associated with intraoral distractor device, (3) Le Fort I osteotomy associated with RED system. The parents did not want to undergo extensive orthognathic surgery. Therefore, the RED device to advance the maxilla was chosen.

Treatment progress

The treatment plan was to facilitate eruption of permanent teeth to anchor the traction force of the RED system, followed by correction of midface hypoplasia and create normal relationship between upper and lower jaws by DO of the maxilla. The procedure started with leveling and aligning upper central incisors, extract the primary second molars

Figure 8: Custom-made simi-rigid splint

Figure 10: Posttreatment intraoral photos

to facilitate eruption of the ectopic upper permanent first molars. Transverse palatal arch wires between U6s extract supernumerary tooth, expose, and align impacted upper canines and upper right first premolars [Figure 7]. After aligning the erupted teeth, transpalatal arch wire removed, 050" headgear tubes are fitted on the first permanent molar, an impression is obtained of the maxillary to fabricate accustom-made device. Head gear (HG) face bow and palatal wires are around the perimeter of the dental arch with considering the incisors brackets and soldered to the upper first molars bands [Figure 8]. The external arms of HG face bow are bent over and anterior to the upper lip and eventually bent into an eyelet figure from which the splint and the RED device are connected by means of surgical wire. The position of the eyelet of the external arms is significantly important to exert control over the anteroposterior movement of the maxilla. The center of the resistance of the maxilla has been estimated to be at the level of the apices of the second of the bicuspids.[11] This guideline can be used to determine the position of the traction hooks. A force vector above the center of mass will create a clockwise rotation, whereas a force vector below it creates counterclockwise rotation, and one through the center of the mass will advance the maxilla in a linear fashion. Therefore, the placement of the traction hooks slightly above the root apices of the teeth and the osteotomy cut will provide a biomechanical favorable position. Surgery and RED system placement: The patient was hospitalized, and surgery of DO was performed under general nasotracheal anesthesia. The maxilla was injected with 1% xylocaine with epinephrine for

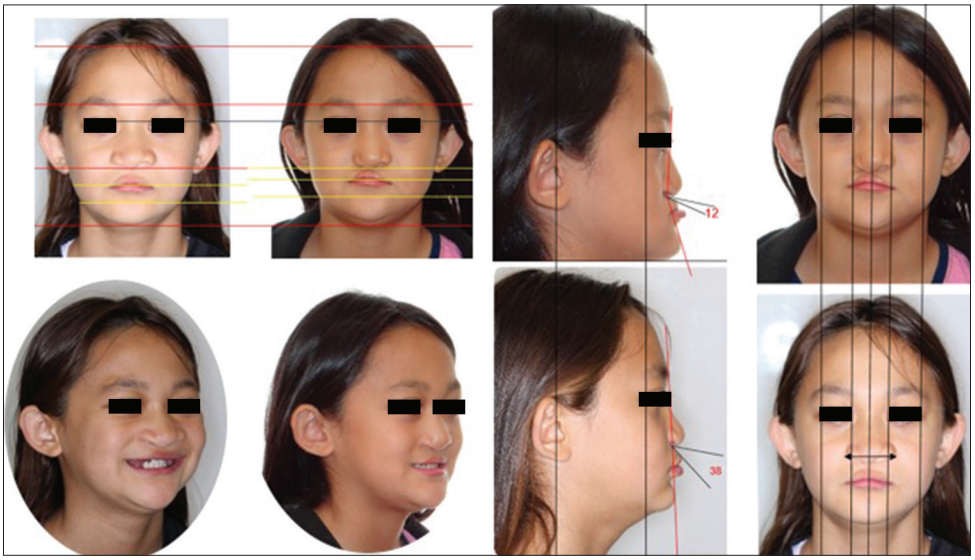


Figure 12: Pre- and post-distraction extraoral photograph

Figure 13: Smile change pre- and post-treatment

hemostasis and the scalp was injected with 0.25% marcaine for postoperation pain, vestibular incision was performed, and mucoperiosteum was elevated. A sagittal saw was used to make a Le Fort I osteotomy through the pyriform aperture and the pterygomaxillary fissure. The maxilla was then downfractured and mobilized. The semirigid splint was then cemented with glass ionomer cement to the upper first molars and wired to upper central incisors. The RED frame was then attached to the cranium with three sharp pins bilaterally. This was secured and the lower member of the device was attached to the head frame and 24-gauge wires were used to connect to the extraoral splint arms. The device was activated to 10 mm and the maxillary moved fairly well. It was placed in the latency position. Merocel was packed in the nose and the incision was closed with 3–0 chromic suture. Patient informed to start activation after 1 week latency, 1 turn a day for 7 days with additional 3 days was essential for overcorrection. After achieving correction of sagittal maxillary deficiency, the RED system was maintained for 6 weeks without activation to permit bone consolidation. After the removal of RED device, reverse pull HG fitted and delivered with 3/16in, 6.5 oz face mask elastics. The patient informed to wear it full time to retain the distraction. Using a Facemask after removing the RED has significant stable results.[12] Same fabricated splint was used to apply the elastics force [Figure 9]. The protraction face mask was discontinued after a month and the upper splint was removed. Brackets bonded to upper first molars and full arch wire were placed to continue the dental malocclusion treatment [Figure 10]. The patient moved to another state and she was provided with a referral to continue the orthodontics treatment in Texas.

Results

The posttreatment photographs and radiographs showed that midface advancement with proper overjet and overbite was achieved, and no insufficiency of velopharyngeal or speech function deterioration was observed [Figure 11]. The linear horizontal changes were measured relative to a line perpendicular to S-N plane passing through Sella, and the vertical changes were measured perpendicular to the S-N plane. The cephalometric analysis presented that there is pretty large improvement for maxilla as the overjet was increased by 10.5 mm. The value of SNA improved by 10.4° (A point advanced 10.5 mm anteriorly and 2 mm superiorly), while the nasal tip advanced 4 mm anteriorly and 5 mm superiorly. The upper lip also advanced 6 mm anteriorly and 2 mm superiorly. The deep bite was opened from 7.9 mm to 1.3 mm and the mandibular plane was opened by 3° [Figure 3]. The facial profile changed from concave to convex figure, anterior lower height improved by 9 mm; therefore, the facial balance and esthetics improved, the NLA improved by 26°, the maxillary incisors were proclined and 4 mm of upper centrals incisor displayed in the frontal smile photograph, increased alar base width [Figures 12 and 13], corrected posterior crossbite [Figure 14]. Posterior open bite presents after maxillary distraction. The patient and her family were satisfied with the result.

Figure 14: Pre- and post-treatment intraoral change

Midfacial hypoplasia is commonly treated by conventional maxillary orthognathic surgery, which included some kind of Le Fort surgery depending on the severity that results in the displacement of the maxilla anteriorly and stabilization afterward with rigid fixation. However, several limitations have been reported with this procedure;[8] the literature documents 25%–40% relapse,[7] excessive blood loss, and edema;[8] it may induce an undesirable fracture, especially in the patient with abnormal bon structure such as CLP patient.[8] The frequent surgical intervention creates a tight surgical scar which is considered as a factor for growth retardation and relapse of the midface after conventional orthognathic surgery. In addition, maxillary advancement more than 6 mm is difficult to achieve using this technique due to maxillary scar.[13] Patients may have pharyngeal flaps that affect stability after conventional maxillary.

McCarthy *et al*., 1992 reported the first clinical application of distraction osteogeneses, they showed the elongation of hypoplastic mandible by biological process generating new bone between the bony segments.[14] Traction force applied to bone also creates tension in the soft tissue, initiating a sequence of adaptive change termed distraction histogenesis.[15,16] DO can be performed either by intraoral distractor device or RED system. The major drawbacks of internal distractors are poor victor control, differential movement within the midface, discomfort, infection, and trauma during activation.[7] RED system has provided a significant alternative to treat severe midface hypoplasia with minimally invasive procedure. The gradual advancement and lengthening of the soft tissue in distraction osteogeneses procedure offers proper adaptation and lowers relapse rates 5.56%–10%, with more advancement.[17] This technique can be used to treat skeletal dysplasia at a young age (compared with the need to wait for skeletal maturity if conventional orthognathic surgery is to employed). The surgical procedure affected maxilla without having to operate mandible[18] and it is very simple, no need for blood transfusion or bone grafting nor hard fixation hardware. 2–5 mm overcorrection is very necessary to compensate anticipated relapse after DO procedure. The flexibility of the traction hooks allows for energy storage. This continuous tension is favorable event and is conductive to osteogenesis.[6,19] However, after ostomizing the maxilla, less force is required to advance it; therefore, the effects on individual tooth position have been minimal.[20] The intraoral splint is custom designed, easily constructed by the orthodontics, which is helpful in patients with unusual arch forms, especially in cleft patients, inexpensive, hygienic, comfortable, none traumatic. It does not interfere with speech and eating. The main advantages of the designer of the intraoral splint are the ability to control the direction of the force vectors at any time during the distraction process without discomfort the patient, adjusting the force to pass through or above the center of the mass of the maxilla alters the rotation movement direction, activating one side more than other allows for midline correction when the maxilla skewed to one side.[19] The limitation of the rigid distraction technique relate to those patients with complete absence of teeth or lack of adequate bone in the cranial vault. Limited patient acceptance of the halo frame due to unaesthetic external component, incompliant patients, or parents may contribute to postoperative problems such as turning the screw to the wrong side, avoidance of daily turning due to pain, and forgetting daily turning of the screw when activating the traction screw.[21] Malocclusion may occur due to traction force through the teeth. The cost of the device is only a minor disadvantage.[12] Some complications related to the use of the cranium fixation have been reported such as device failure, pin loosening after trauma, and infections surrounding the pins.[22] However, patients do not complaint of the pain after placement the halo pins. Patients can shampoo their hair in the usual fashion. The device is removed in the clinic without local anesthesia after the rigid retention (consolidation) phase. Thus, we believe that it is mandatory to the professions to offer special care for patients with CLP such as innovative techniques, regular supervision, motivation to achieve the desired outcome with minor complications.[1]

Conclusions

Our results demonstrate that DO using rigid external distractor allows moving the maxilla in multiple planes, simple, highly effective, stable, and predictable option for treat midface hypoplasia associated with unilateral CLP, especially when maxillary advancement more than 10 mm is required. The therapeutic results showed a significant improvement of the patient's esthetic and occlusal balance.

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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