

# Maxillary Advancement Using External Distraction Device Internally: a Case Report and Review of the Literature

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**Introduction:** Advancing the dysplastic maxilla in cleft palate patients is a challenging issue. This study aimed to report a case of maxillary advancement in a cleft palate patient by distraction osteogenesis technique. **Materials and Methods:** A 21 year old woman with the chief complaint of retruded maxilla was planned to be treated by distraction method. An external craniofacial distraction device was used in a total different way to advance the hypoplastic maxilla. In the second section of the study a review table of advancing techniques in such cases was designed and presented. **Results:** The amount of maxillary advancement was 10 mm horizontally and 5 mm inferiorly. The distraction procedure gave the ability of moving the maxilla in two directions. The tooth show was improved and paranasal area and A point were advanced to help in gaining a straight profile. The remained discrepancy was corrected by mandibular setback procedure. **Conclusions:** Distraction osteogenesis may be a reliable treatment plan in advancing hypoplastic midface in cleft palate patients. The results of this method may be more sufficient than routine orthognathic surgery.

**Keywords:** Cleft palate; Distraction Osteogenesis; Maxilla

## Introduction

Class III malocclusion is the most common skeletal deformity observed in cleft palate patients (1). This kind of malformation is occurred due to maxillary hypoplasia following scar formation on the palate. Scar formation following several surgical procedure of palate inhibits the normal growth of the maxilla (2). Esthetic and functional problems as the results of maxillary hypoplasia necessitate choosing an appropriate surgical procedure to overcome these malfunctions.

Management of maxillary hypoplasia resulted from the cleft palate is a challenge in maxillofacial reconstructive field (3). Choosing the surgical procedure depends on the severity of maxillary deficiencies (4). Maxillary advancement by lefort I osteotomy has been performed routinely in correction of hypoplastic maxilla and cleft palate patients are not an exclusion (4). However stability of advanced maxilla and the risk of relapse are always major consideration in cleft palate patients. Some studies have indicated that the relapse tendency in cleft palate patients is dependent on the amount of advancement (5). In that order researchers attempt to overcome this problem by performing other kind of surgeries such as distraction methods.

Distraction osteogenesis (DO) has been presented as helpful alternative method in treating facial dysplasia especially cleft-

related maxillary hypoplasia (6). Gradual activation of the distraction device gives the soft tissue to be adapted during progressive bone formation. DO technique gives the surgeon the ability to advance the dysplastic maxilla much more than he could perform by traditional orthognathic surgery. On the other hand relapse tendency has benn reported to be less than orthognathic surgery in the literature (7). Reducing the risk of relapse and providing a greater stability of advanced maxilla is an important issue in cleft palate patients where the palatal scar is an interfering factor for advancement. Maxillary distraction devices are divided into internal and external categories. Advantages of each group make the surgeons to decide which one in different cases (8).

Current study aimed to present a case of severe maxillary retrusion due to cleft palate managed by DO technique and reviewing the similar treated cases presented in the literature.

## Case Report

A 21-year old female patient with mastication difficulty was referred to the department of oral and maxillofacial surgery, Taleghani hospital, Tehran, Iran. The patient mentioned the history of bilateral cleft lip and palate. The patient was underwent surgical procedures five



**Figure 1.** Profile view of the patient before the surgery. Concave profile is obvious in this view



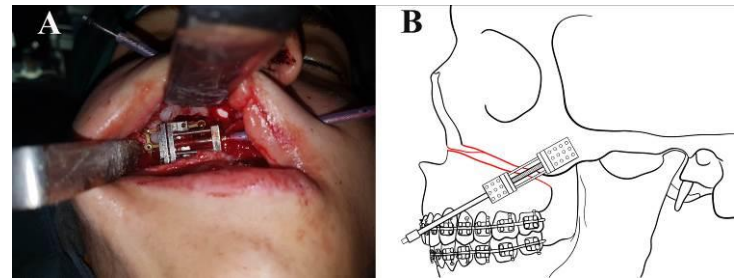
**Figure 3.** Frontal view of the patient before the surgery. Whistle



**Figure 2.** Intraoral view of the patient before the surgery. Retruded maxilla is observed

times to correct cleft lip and palate (cheilorrhaphy, staphylorrhaphy, palatorrhaphy, alveolar cleft reconstruction, and repairing soft palate by pharyngeal flap). The patient was under orthodontic treatment for almost two years before being ready for orthognathic surgery.

Concave profile indicated the maxillary hypoplasia (Figure 1). During intraoral examination, reversejet of 12mm was observed (Figure 2). Class III molar relationship was obvious in intraoral examination. Cephalometric analysis showed severe retrusion of the maxilla. The orthodontist was not able to correct the inclination of upper incisors due to crown restoration and tendency of anterior maxillary segment not able to correct the inclination of upper incisors due to crown restoration and tendency of anterior maxillary segment bone loss.



**Figure 4.** Application of the distraction device. A, The activator rod is passed through the paranasal area. B, Schematic picture of the device

Whistle deformity was observed. Upper and lower dental midlines were matched to the facial midline. Upper lateral incisors were missed following the bilateral alveolar cleft. There was no tooth show presented in the rest position of the lips (Figure 3).

In order to be able manage the vectors of maxillary advancement perfectly it was decided to use an external craniofacial distraction device internally. Lefort I osteotomy was performed. The distraction device was fixed on the posterior maxilla (Figure 4). The activator parts of the devices were passed through the paranasal area (Figure 4). The oral mucosa was sutured completely to cover the device for better oral hygiene. After five days of latency period the devices were activated twice (1mm) per day. After the consolidation period the devices were removed and sagittal split osteotomy of mandible was used for mandibular setback and correction of the occlusion.



**Figure 5.** Frontal view following the distraction process. The increased tooth show was about 5 mm.



**Figure 6.** Intraoral view following distraction process showed the advancement of about 10 mm and remained reverse jet of 2mm

### Ethical considerations

The patient was informed of the purpose and design of the treatment and signed an appropriate consent form. The procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation of Shahid Beheshti University of Medical Sciences as well as Helsinki Declaration.

### Review of the literature

The PICO (population, intervention, comparison, and outcome) question was stated as follows: In cleft palate patients with maxillary hypoplasia (P) does DO technique (I) in comparison to usual orthognathic surgery (C) increase the stability and success rates (O)?



**Figure 7.** Profile view of the patient. A, concave profile view before the DO surgery. B, Slightly convex profile following the two surgeries showed sufficient results



**Figure 8.** Frontal view of the patient three years after surgery

Electronic databases (Medline and Embase) were searched by the authors to find the articles which were published up to March 2020.

The search terms used in this review included “maxillary hypoplasia” OR “maxillary dysplasia” OR “maxillary retrusion” OR “cleft palate” [Mesh] AND the Mesh terms related to the maxillary advancement including “orthognathic surgery” AND “Distraction osteogenesis” OR “Lefort I osteotomy”.

The abstracts of all articles were evaluated to exclude the unrelated papers. After initial review the full texts of retrieved articles were assessed to select the relevant references to the following inclusion criteria:

**Table 1.** Categorized data of investigations on maxillary advancement in cleft palate population

Authors	Sample size	Mean age (year)	Method of advancement	Mean advancement amount (mm)	Considerations
Tong H. et al. (2019) (13)	26	11.5 ± 2.1	Trans-sutural distraction osteogenesis	13.99 ± 4.90	This method depends on both the secondary displacement promoted by sutural bone formation mainly in the pterygomaxillary suture and the primary displacement by growth of maxillary tuberosity
Tobolowsky W. et al. (2019) (14)	21	16.8	DO followed by bone grafting	14	Bone grafting and rigid fixation as a second procedure may help ameliorate relapse risk
Moran I. et al. (2018) (7)	79	Over 17 year old patients	Lefort osteotomy	I 10	This procedure is associated with a wide range of postoperative complications.
Zheng Y. et al. (2018) (15)	73	11.2	trans-sutural distraction osteogenesis	9.9 ± 4.2	Traction hook failure as a complication
Burstein F. et al. (2015) (16)	22	10.24	Internal DO	18	The intraoral nature of this device makes it much more acceptable in many patient populations
Rao S. et al. (2013) (17)	5	16.8	Anterior segment advancement by custom-made tooth-borne distraction device	8.56	There was no change in the velopharyngeal sphincter
Baek S.H. et al. (2007) (11)	14	21.7	Lefort osteotomy	I 6.42	No significant difference in the amount of anteroposterior relapse between Lefort I osteotomy and external DO groups
Baek S.H. et al. (2007) (11)	11	16.3	External DO	10.79	The amounts of vertical relapse of PNS displayed significant difference between Lefort I osteotomy and external DO groups
Rachmiel A. et al. (2005) (10)	12	15	Internal DO	13.66	Suitable in moderate or severe retrusion
Figuerola A.A. et al. (2004) (6)	17	12.6	External DO	9.5	This method is stable and effective.
Heliovaara A. et al. (2002) (12)	14	27.2	Lefort osteotomy	I 4.7	postoperative relapse was 9.4% (0.5 mm) horizontally and 17.8% (1.3 mm) vertically
Hirano A. et al. (2001) (5)	71	19.8	Lefort osteotomy	I 6.9	A relapse was more likely to occur in patients with bilateral cleft.
Figuerola A.A. et al. (1999) (18)	14	Older than 5	External DO	11.6	The technique allows for vector control of the osteotomized maxilla throughout the distraction process
Holina F. et al. (1998) (19)	38	9	External DO	8	Velopharyngeal function remains unchanged
Hochban W. et al. (1993) (4)	31	26.1	Lefort osteotomy	I 8.3	More relapse in cleft palate cases (20 to 25%)
Posnick C.J. (1990) (20)	30	18	Lefort osteotomy	I 6.7	0.8 mm relapse after 1 year
Houston W. et al. (1989) (3)	30	17	Lefort osteotomy	I 9	7% mean horizontal relapse and 23% mean vertical relapse
Luyk N.H. et al. (1985) (9)	3	21.6	Lefort osteotomy	I 3.7	No significant relapse

- Studies published up to March 2020
- Clinical trials and human studies
- Assessing the effects of DO technique or orthognathic surgery on Maxillary advancement
- Studies executed on the patients with cleft palate
- Studies executed on systemically healthy and non-syndromic patients

The exclusion criteria:

- Case reports
- Letters to the editors
- Technical notes
- Grey literature
- Reviews
- Animal studies

The following data were extracted from each study and categorized in tables:

- Number of patients
- Mean age of the patients
- The mount of reversejet (discrepancy)
- The type of advancing technique
- The mean advancement rate
- The follow up period
- The other important results and findings
- The major complications of the certain technique

## Results

### Case findings

The patient was under this treatment for 2 months. Maxilla was advance 10 mm horizontally and 5 mm inferiorly. Photographs obtained following DO process showed excellent of maxilla and good fullness of paranasal area. Fives millimeters tooth show was observed after DO treatment which was suitable since it was decided to correct the lip deformity later than (Figure 5). Cephalometric evaluation indicated enough advancement of maxilla (SNA 78°). Although there was still 2 mm discrepancies activation of distraction device was not possible due to the interference of mandibular incisors (Figure 6). So it was decided to perform mandibular setback surgery to correct the occlusion and facial profile. Slightly convex profile was suggestive of the optimum esthetic result (Figure 7). Overjet was acceptable and class I molar relationship was achieved. Follow up after three years showed a good stability of the advancement and esthetic results (Figure 8).

### Literature review

Eighteen articles were retrieved after screening the abstracts. The findings of investigation designed on the maxillary advancement of cleft palate patients are categorized in Table 1.

## Discussion

Achieving to an ideal profile and Angle class I relationship is hard to get in cleft palate patients due to multiple surgical scars. Fibrotic tissue occurs following surgeries on the palatal tissue is a major inhibitor of maxillary advancement during orthognathic surgery (9). DO has been suggested as a good alternative for maxillary advancement in cleft palate patients (10). Using distraction device is various in different cases and the every surgeon may choose different techniques up to achieve ideal goals. The current survey attempted to present a cleft palate patient treated by DO technique in new different way.

The greatest advantage of DO is the good stability and preventing relapse of maxillary advancement. It is possible to gain more advancement by using DO than usual orthognathic surgery (11). This advantage is very important in cleft palate patients which are difficult cases of maxillary reconstruction. Some researchers proposed that it is possible to advance the maxilla in such cases less than 6 mm and the amount of advancement reversely correlates with the stability of fixed maxilla (5). Several investigations showed the enough stability of advanced maxilla following distraction technique in a long time follow up.

There are some published articles in the literature which investigated the influence of bone grafting on the stability of maxillary advancement (9, 12). Although they showed more stability than routine orthognathic surgery the relapse rate is not less than DO methods.

The current study reported a severe retruded maxilla due to the palatal cleft treated by distraction method. Using various kinds of distractions device is up to the surgeon's opinion in different cases. Internal and external devices are available and each of them are supported by different researchers. In present case it was decided to use an external craniofacial distraction device in a different way. According to the clinical and cephalometric examinations, lack of tooth show and severely dysplastic midface was observed. So it was designed to move the maxilla inferiorly and horizontally advanced. The distraction which was used in present case originally is used for advancement of madifacial compartment. The fixed plates of the device will be kept under the skin on zygomatic buttress and the activator rod is passed through the skin. In the current case, maxillary reconstruction were achived by performing the Lefort I osteotomy and placing the device on posterior maxilla. The activator rod was passed through the skin of paranasal region to be available for easy activation. This technique helped the mentioned achievements to reach to an advanced maxilla and improved tooth show.



Previous unpleasant cheilorrhaphy of the patients resulted in deformity of the upper lip. The whistle deformity indicated the lack of orbicularis oris muscle in the middle of the upper lip. The lack of muscle presence in the midpart of the lip led into the excessive retrusion show of the maxilla. This was the reason of remained retruded appearance of the midface despite 10 mm advancement of the maxilla. So the upper lip was planned to be corrected by secondary cheiloplasty to reorient the orbicularis oris muscle and improve the maxilla protrusive show.

## Conclusion

In conclusion, distraction techniques may lead to stable results of maxillary advancement in cleft palate patients. The vector of maxillary movement is under control by using the suitable kind of distraction device. Simple application and sufficient results of DO method has made this procedure the treatment of choice in such cases. Correction of slightly discrepancies remained following DO technique is afforded by routine orthognathic surgery with no considerable complication.

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