Orthognathic Treatment with Distraction Osteogenesis in Two Asymmetry Cases

İki Asimetrik Vakada Distraksiyon Osteogeneziyle Ortognatik Tedavi

Ayse Gulsen[1], Serhat Sibar[1], Safa Manav[2], Kemal Findikcioglu[1]

[1]Gazi University Hospital, Department of Plastic, Reconstructive and Aesthetic Surgery, Ankara, Turkey
[2]Afyonkarahisar State Hospital, Department of Plastic, Reconstructive and Aesthetic Surgery,Afyonkarahisar, Turkey

ABSTRACT

Distraction osteogenesis (DO) is commonly performed procedure in craniofacial surgery and especially useful in treating craniofacial anomalies. Hemifacial microsomia (HFM) and unilateral Tessier no 7 cleft anomalies are congenital anomalies and creates facial asymmetry due to hypoplastic bones and soft tissues but each had unilateral maxillomandibular hypoplasia. DO in HFM cases are well-known technique however there is a lack of information about DO in Tessier cleft 7 in literature. This report describes the use of DO in HFM and Tessier cleft 7 however the characteristics of the mandible and also maxillomandibular deviation in two cases were similar so two cases were reported in our study. Gonial, occlusal, maxillary, nasal base and lip cants were measured. In both case with DO is combined with fat grafting methods so more symmetric and balanced facial contours were obtained in postoperative 5 years follow-up period.

Key Words: Craniofacial abnormalities, distraction osteogenesis, hemifacial microsomia

Received: 07.13.2020 **Accepted:** 09.30.2020

ÖZET

Distraksiyon osteogenezi (DO) kraniyofasiyal cerrahide özellikle kraniyofasiyal anomalilerin teavisinde kullanılan bir uygulamadır. Konjenital anomalilerden olan hemifasiyal mikrozomi (HFM) ve tek taraflı Tessier 7 no'lu yarık anomalileri, kemikte ve yumuşak dokuda hipoplazilere, her ikisinde de tek taraflı olacak şekilde maksillomandibuler hipoplaziler oluşturarak yüz asimetrilerine neden olmaktadır. HFM vakalarında DO iyi bilinen bir tekniktir ancak literatürde Tessier 7 no'lu yarık vakalarında DO kullanımı hakkında bilgi eksikliği vardır. Bu çalışma, mandibula karakteristik özellikleri ve maksillomandibular deviasyonları benzer olan iki farklı vakada (HFM ve Tessier yarık 7 no'lu yarıkta) DO'nun kullanımını açıklamaktadır. Gonial, oklüzal, maksiller, nazal taban ve dudak kanatları ölçülmüştür. Her iki vakada DO'ya yağ grefti uygulaması eklenmiş olup ameliyat sonrası 5 yıllık takipte daha simetrik ve dengeli yüz konturları elde edilmiştir.

Anahtar Sözcükler: Kraniofasiyal anormallikler, distraksiyon osteogenezi, hemifasiyal mikrozomi

Geliş Tarihi: 13.07.2020 **Kabul Tarihi:** 30.09.2020

ORCID IDs: A.G. 0000-0003-3222-8002, S.S. 0000-0002-7533-7877, S.M. 0000-0002-6037-3509, K.F. 0000-0002-8276-5730

Address for Correspondence / Yazışma Adresi: Serhat Sıbar, MD Gazi University Hospital, Department of Plastic Reconstructive and Aesthetic Surgery, 14th floor Besevler, Ankara, Turkey E-mail: serhatsibar@hotmail.com

doi:http://dx.doi.org/10.12996/gmj.2021.58

INTRODUCTION

Distraction osteogenesis (DO) that based on Ilizarov's technique is widely used in craniofacial surgery since its introduction by McCarthy et al. (1,2). Hemifacial microsomia and unilateral Tessier no 7 cleft anomalies are congenital craniofacial anomalies and creates facial asymmetry due to the unilateral maxillomandibular hypoplasia in bones and soft tissues. Hemifacial microsomia primarily is a congenital syndrome of the first branchial arch with an extremely variable phenotype and, characterized with maxillomandibular hypoplasia and facial asymmetry with occlusal canting involving underdevelopment of the ear, mandible, maxilla, zygoma, temporal and audiotory bones, and the associated musculature and soft tissues and also more retruded mandibles and maxilla and a more vertical morphology compared to the reference population were seen in this group (3,4). Tessier no. 7 clefts are one of the rare clefting and are different from hemifacial microsomia, which is the lateral facial cleft extends from the oral cavity towards the tragus, involving both soft-tissue and skeletal components like absence of the zygomatic arch, variable deformity of the mandibular ramus, condyle, and coronoid process; and hypoplastic maxillary alveolus; maxillary cleft in the molar region, some vertical maxillary hypoplasia very rarely maxillary jaw duplication soft-tissue abnormalities including macrostomia, ear abnormality, temporalis abnormality, and absence of preauricular hair (6-8).

In adult congenital facial asymetry cases, bimaxillary conventional orthognathic surgery especially in mild cases, costachondral grafts or microsurgical reconstruction with flaps, DO, and DO of costachondral graft for ramus, and soft tissue augmentation with fat graft or vascularized tissue flaps for soft tissue hypoplasia are some of the treatment options of facial asymmetry.

Distraction osteogenesis in HFM cases was reported in literature as only in mandible with Lefort 1 osteotomy and IMF, as only in maxilla with mandible osteotomy, or as distraction of both maxilla and mandible and as DO of costachondral grafting in some grade III cases. In this study, orthognathic treatments with DO were presented in two cases with hemifacial microsomia (HFM) and unilateral Tessier cleft 7. HFM and Tessier cleft 7 anomalies are different anomalies but each had maxillomandibular unilateral hypoplasia.

CASE REPORT

Two patients; one with hemifacial microsomia and the other with unilateral tessier cleft 7, diagnosed as Pruzansky type 1, treated with DO was introduced in our study. All patients informed consents were taken and due to the retrospective nature of this study, it was granted an exemption in writing by the Gazi University Medical School institutional review board. Lateral, posteroranterior and panaromic radiographs were obtained in two cases. The landmarks and parameters used for cephalometric diagnosis and follow-up analysis are showed in Figure 1.

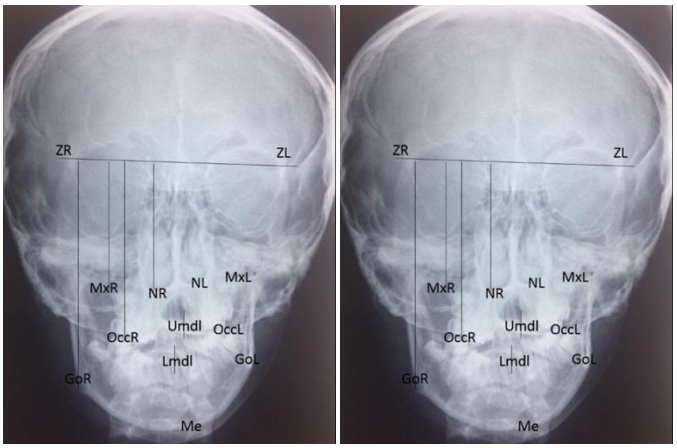

Figure 1A **Figure 1B**

Figure 1A and B Anatomic landmarks, parameters on the posteroanterior cephalogram

Z, intersection between zygomaticofrontal suture and orbita; Mx, maxillary point on intersection between zygomatic arch and maxilla; N, the most inferiot point of nasal cavity; Occ, the intersection of upper and lower molar teeth; Go, gonion; Me, menton, Umdl, the midline between upper central incisors; Lmdl, the midline between lower central incisors; (Me: Distance between midsagital plane and mentum; L-mdlD: Distance between midsagital plane and midline of lower incisors; UmdlD: Distance between midsagital plane and midline of upper incisors; N.Cant: cant of nasal base (the angle between horizontal reference plane and nasal base); M.Cant: Cant of maxillary base (the angle between horizontal reference plane and maxillary plane) ; O. Cant: Cant of occlusal plane (the angle between horizontal reference plane and occlusal plane); Go.Cant: Cant of gonial plane (the angle between horizontal reference plane and gonial plane); VNLD: difference between the heights of right and left nasal base points (measured distance in millimeters from horizontal reference plane to nasal points); VOccLD: Difference between right and left occlusal heights (distance in millimeters from horizontal reference plane to contact point between maxillary and mandibular molars); VMxLD: Difference between the heights of right and left maxillary points (distance in millimeters from horizontal reference plane to maxillary point); VGoLD: Difference between the heights of right and left gonial points (distance in millimeters from horizontal reference plane to gonial points)

Case 1

A 17-year-old female with left hemifacial microsomia had an asymmetrical long face with a mildly underdeveloped left side with lip canting and with asymmetric gummy smile (Figure 2A).

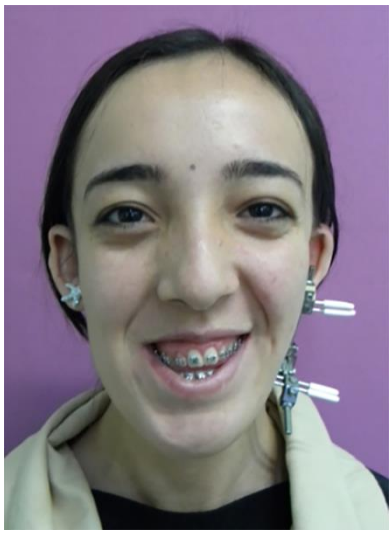

Figure 2A Preoperative frontal view of case 1.

Table 1.

	SNA(°)	SNB(°)	ANB(°)	GOGNSN(°)	N-Me(mm)
Case 1					
Before DO	72	70	2	43	119
After DO	72	74	-2	43	119
After Le Fort1	76	74	2	39	119
After 5 years	75	73.5	1.5	41	119
Case 2					
Before DO	68.5	69	-0.5	41	112
After DO	70	69	1	45	114
After 5 years	70	69	1	45	114

Mandibular distraction with a single-sided multidirected extraoral distractor to lengthen short ramus and to correct asymmetry and midline coordination was planned as first phase. In second phase, Le Fort 1 osteotomy to level the maxillary canting during the removal of mandibular distractor and in third phase, fat grafting to the affected side was planned. After 11 months of orthodontic treatment, surgical intervention was performed. A mandibular horizontal ramus osteotomy above the lingula on the affected side was done and multi-vector distractor (Synthes CMF, West Chester, PA) was placed to both sides of the osteotomy**.** Following a 7-day latency period, the distractor was activated during 47 days vertically and 14 days horizontally until the mandibular midline deviation was overcorrected by up to one third of the initial discrepancy (5). During the distraction period, intermaxillary elactics were used to direct the movements of distracted mandible. Following a 3 months consolidation period, mandibular distractor was removed, and Le Fort 1 surgery (four millimeter impaction in the unaffected side and six millimeter elongation in the affected side) was performed to correct the maxillary cant and occlusion. (Table 1). Six months later, the patient was operated for rhinoplasty and 10 cc fat grafting was added to the affected side three times.

Landmark locations and parameters on the lateral cephalogram: *S, sella*; N, nasion; A, anterior maxillary point; B, Anterior mandibular point; Gn, gnathion, most anterior point of chin; Go, the intersection of ramus and corpus of mandible; Me, the most inferior point of chin; SNA, Angle that defines maxillary anteroposterior position; SNB, angle defines that mandibular anteroposterior position; ANB, angle defines the position of maxilla in reference to mandible or vice versa; GoGnSN, the angle between Sella-Nasion and GoGnN-Me, the height between nasion and menton that defines anterior facial height. DO, Distraction osteogenesis.

The postoperative lateral cephalometric radiographs showed that mandible moved forward and the facial height didn't change after DO. Following the maxillary surgery, Class 1 relationship was kept. The results after four years follow-up were stable (Table 1). In the follow-up posteroanterior cephalometric analysis, mentum was overcorrected and this over correction was kept following four years. Gonial levels were equal. Occlusal and lip cantings were corrected. It was not obtained equalization in right and left nasal base levels but was better following distraction and rhinoplasty operation (Table 2). The occlusal canting was more balanced and gummy smile on the right was corrected (Figure 2B). Maxillary and occlusal canting was corrected without facial elongation and she had better smile. Although equalization in underlying skeletal structures in vertical plane was obtained, the gonial region was still has less volume so fat grafting was added to overcome the hypoplastic soft tissue and left side appeared more fully appearance. Left gonial level seems decreased after Le Fort 1 surgery but didn't change in follow-up period (5 years).

Figure 2B Postoperative result of case

Case 2

A 18-year old female with a history repaired unilateral Tessier 7 clefts had deviated occlusal cant, deviated mentum to the left (Figure 3A).

Maxillary distraction with a single-sided multidirected extraoral distractor to lengthen short ramus and to correct asymmetry and midline coordination together with unilateral mandibular distraction osteogenesis was planned as phase first. Fat grafting to the affected side was planned as phase second. A double-sided vertically oriented alveolar distractor was placed in maxilla and a multivectoral distractor was placed to the mandible at the same operation. Mandibular distractor was placed like first case. A complete horizontal Le fort 1 type osteotomy was performed. Separation of pterigomaxillary junctions was done in both side. The maxilla was mobilized without downfracture. In the right side maxilla lowered four milimeter. Bilateral alveolar distractors (Synthes CMF, West Chester, PA) were placed vertically and alveolar distractor was placed opened in unaffected side. The mandibular distraction was started 7 days later and the distraction on the left side of the maxilla was started on the 9th day. Distraction period took 33 days. Seventh and 27. day distractor was opened horizontally. Maxillar left alveolar distractor was opened for 14 days. Consolidation period continued for 16 weeks. The patient was given totally 50 cc fat graft to the affected side four times in two years. The postoperative lateral cephalometric radiographs showed that skeletal class 1 relationship was obtained with an minor increment of facial height. The results after 5 years follow-up were stable (Table 2).

Table 2.

	Me (mm)	L- mdl (mm)	U-mdl (mm)	N.Cant(°)	M.Cant (°)	O.Cant (°)	Go.Cant (°)	VNLD (mm)	VMLD (mm)	VOccLD (mm)	VGoLD (mm)
Case 1											
Before DO	8 (L)	3 (L)	5 (L)	11 (LU)	10 (LU)	9 (LU)	7.5 (LU)	-5	-9	-9	-10
After DO	2.5 (R)	2 (R)	4 (L)	11 (LU)	10 (LU)	7 (LU)	5 (LU)	-5	-9	-9	+7
After Le Fort1	2 (R)	3 (R)	3 (R)	6 (LU)	0	0	0	-3	0	0	+1
After 5 years	2 (R)	3 (R)	3 (R)	6 (LU)	0	0	0	-3	0	0	+1
Case 2											
Before DO	9 (L)	3 (L)	6 (L)	6 (LU)	7 (LU)	9 (LU)	8 (LU)	-5	-9	-8.5	-16
After DO	4 (R)	4 (R)	0	4 (LL)	-	5 (LU)	5.5 (LL)	+3	+2	+2	+7
After 5 years	2 (R)	0	0	3 (LL)	1 (LU)	3 (LU)	2 (LU)	+0.5	+0.5	+1	+3

L (left); R (right); LU (canting toward left upper direction); LL (canting toward left lower direction)
Me: menton; Lmdl: the midline between lower central incisors; Umdl: the midline between upper central incisors (Me: Distance between midsagital plane and mentum; L-mdlD: Distance between midsagital plane and midline of lower incisors; UmdlD: Distance between midsagital plane and midline of upper incisors); N.Cant: cant of nasal base (the angle between horizontal reference plane and nasal base); M.Cant: Cant of maxillary base (the angle between horizontal reference plane and maxillary plane); O. Cant: Cant of occlusal plane (the angle between horizontal reference plane and occlusal plane); Go.Cant: Cant of gonial plane (the angle between horizontal reference plane and gonial plane); VNLD: difference between the heights of right and left nasal base points (measured distance in millimeters from horizontal reference plane to nasal points); VMLD: Difference between the heights of right and left maxillary points (distance in millimeters from horizontal reference plane to maxillary point); VOccLD: Difference between right and left occlusal heights (distance in millimeters from horizontal reference plane to contact point between maxillary and mandibular molars); VGoLD: Difference between the heights of right and left gonial points (distance in millimeters from horizontal reference plane to gonial points). DO: Distraction osteogenesi

Figure 3A Preoperative frontal view of case 2

In the follow-up posteroanterior cephalometric analysis, mentum was overcorrected four milimeters to the right and this over correction decreased following 5 years. Maxillary and mandibular midline coordinated midfacial reference. Gonial levels were equal. Occlusal cantings were corrected but lip canting continued due to the previous scar tissue. It was obtained equalization in right and left nasal base levels following DO. Although equalization in underlying skeletal structures in vertical plane was obtained, the gonial region and left cheek had still less volume so an additional fat grafting was added (15 cc) to overcome the hypoplastic soft tissue and left side appeared more fully appearance (Figure 3B).

Figure 3B Postoperative result of case 2.

DISCUSSION

In facial asymmetry cases with hypoplasia of ramus, surgical treatment strategies was performed according to the severity of deficiency. Conventional bimaxillary surgery, mandibular distraction and orthodontic treatment, autologous bone grafting with facial soft tissue augmentation, distraction of mandible with maxilla together with IMF, simultaneous maxillo-mandibular distraction or simultaneous costachondral graft with mandibular distraction are the some of the effective techniques (9-11). In this kind of anomalies maxillo-mandibular treatment is required to correct facial asymmetry in three dimension and to correct occlusal and aesthetical appearance. Conventional orthognathic bimaxillary surgery is possible in mild cases but in very hypoplastic cases, orthognathic surgery with or without bone grafts is difficult.

DO is widely accepted treatment technique to obtain enough skeletal structure without donor need.

In this case report, two adult with congenital hemifacial hypoplastic maxilla and mandible with small and short ramus were presented. And, maxillary and occlusal cantings and chin were deviated to the left side and soft tissues of affected side was deficient. Oral commisure in HFM cases was deviated to the upper left but in Tessier case was lower left due to the scar of soft tissue. In two cases, skeletal and soft tissue ramal volume was enhanced vertically with DO.

Maxilla-mandibular DO with a single mandibular-distractor using an interdental acrylic splint made preoperatively and intermaxillary fixation in adults not to have occlusion problems was introduced first by Monasterio (12). This technique is very handy method to keep the patient's original occlusion and to immediate correction of occlusion and occlusal canting and widely accepted method may be needed (13,14). The disadvantages are elongation of face and long period with intermaxillary fixation without chewing in consolidation period and also in gummy smile cases, this method is not a good option. In our first case, there was gummy smile and maxillary impaction was needed in non-affected side and Le Fort 1 osteotomy including maxillary impaction in right and maxillary leveling was postponed to distractor removal time following three months consolidation period. In the second surgery fixation plate was also used not to relapse of distracted segments due to three months. Two phased surgery is disadvantage of this method but the patient had obtained more balanced face and maxillary impaction could be done in second distractor removal surgery and also intermaxillary fixation was not used and the patient could feed easily. In the second case, we used both mandibular and maxillary distractors in the same operation. Scolossi et al. used maxillary-mandibular distraction with separate distractors including one distractor on the affected side and wire fixation on the unaffected side for rotation (15). This method doesn't require IMF so the patients can go a normal oral function and dental hygiene and feeding.

Both HFM and Tessier cleft 7 cases had also hypoplastic soft tissue volume. With DO, skeletal and soft tissue volume can be corrected in one dimension but in mediolateral direction are still deficient and three-dimensional restoration of facial symmetry is not possible due to in one directed enlargement. So fat grafting was performed in both cases to reconstruct the soft tissue contour. Fat grafting is an easy method but it requires multiple interventions due the loss of volume. In two cases, three and four times fat grafting was performed to obtain enough volume.

Congenital facial asymmetry cases with mandibular hypoplasia can be reconstructed with DO and fat grafting methods. In first case, maxillary surgery postponed to the distraction removal durgery and impaction on the right and extrusion on the left side were performed. In second cases, maxillary and mandibular distraction was performed same time with different distractors. DO method is very valuable and effective technique in some cases. According to the the severity of anomaly, and to the possibilities available (the socioeconomic condition of patient), the methods may vary. In both case with distraction more symmetric and balanced facial contours were obtained.

Conflict of interest
No conflict of interest was declared by the authors.

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