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REVIEW

Le Fort III Osteotomy and Distraction for Midface Hypoplasia Secondary to Radiotherapy-Clinical Report and Review of The Literature

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Received: 20 February 2017, Accepted: 6 March 2017, Published online: 29 March 2017

ABSTRACT

A successful reconstruction of severe mid-facial hypoplasia secondary to chemo-radiotherapy with Le Fort III osteotomy and distraction osteogenesis with Rigid External Distraction device is reported. A 24 years old boy with mid-facial hypoplasia and class III malocclusion caused by chemo-radiotherapy of expanding giant cell granuloma of nasal, peri-orbital and cranial base is presented. However the distraction osteogenesis has some disadvantages originated from impairment on bone regeneration on irradiated bone, the distracted segments revealed favorable bone healing. This procedure resulted with good clinical results. In conclusion, The Le Fort III osteotomy might be conducted to the individual cases with mid-facial hypoplasia secondary to irradiation. Bony reconstruction of the radiation induced maxillofacial hypoplasia with distraction osteogenesis might be considered as first step treatment method for patients with good quality covering soft tissue in cranio-maxillofacial region.

Key words: Le Fort III, distraction osteogenesis, radiotherapy, facial hypoplasia

INTRODUCTION

Multimodality treatment, including chemo-radiotherapy and surgery is an inevitable requirement for most of head and neck cancer in pediatric population. However, radiotherapy is an important factor for survival expectations, diminishes facial bone growth in the young growing child. Radiation has been shown to affect the facial skeleton in 93% of patients treated for cancer with mild to severe radiation damage to soft tissue and bone [1]. Radiotherapy induced facial asymmetry was shown primarily in young children treated before craniofacial maturation [2]. Maxillofacial reconstruction by distraction osteogenesis following tumor surgery and radiotherapy is a useful treatment method in patients with facial deformities [3,4]. However, the technique has widespread application in craniomaxillofacial reconstruction, has numerous disadvantages including impairment on bone regeneration [5], fibrous union [6], reduced biomechanical quality of the regenerate [7], when conducted on radiated bones. The aim of the study is to investigate the literature about distraction osteogenesis of the irradiated maxillofacial skeleton and to compare the features and results of the cases with this particular patient.

CLINICAL REPORT

A 24-years-old male patient presented with severe mid-facial hypoplasia without additional health problem. The patient developed a rapidly expanding, hemorrhagic tumor in mid-facial and peri-orbital region, which was protruding from bilateral nasal orifices and causing exoftalmus. He underwent partial resection of the tumor, chemotherapy including vinkristin and actinomycin D and unknown dose of radiotherapy when he was 2 years old. The ophthalmologic examination showed bilateral optic atrophy, superomedial deviation of left glob and cataract formation. The patient presented to our clinic 5 years after this initial treatment with maxillofacial deformity. The depression secondary to initial

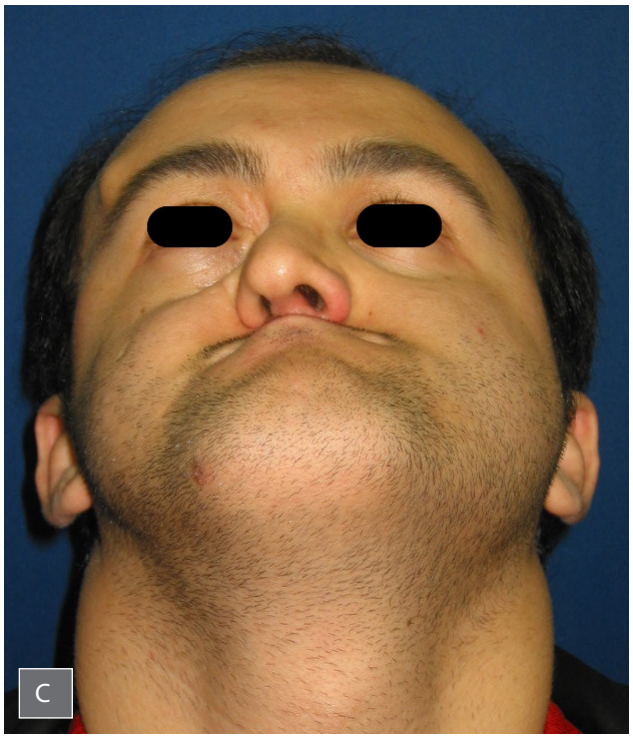
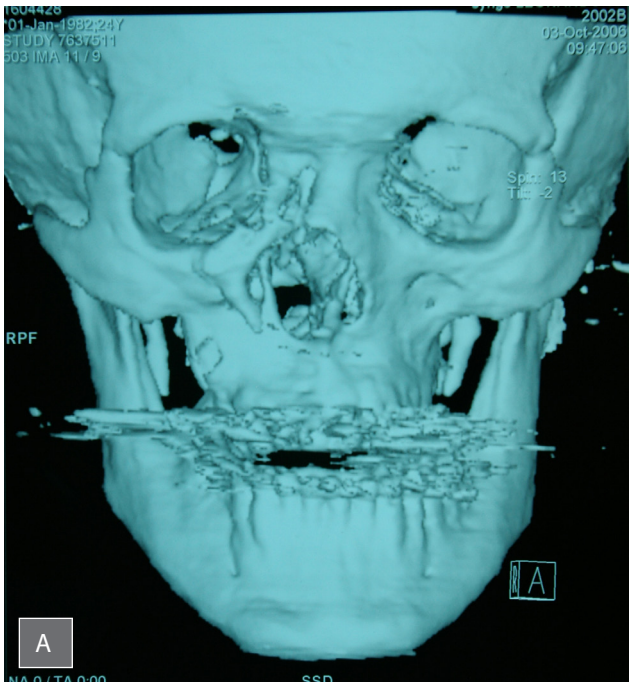


Figure 1. Mid-facial hypoplasia of 24 year old men secondary to radiotherapy.
A) Anterior, B) Right lateral, C) Inferior views.

therapy on the right infra-orbital area was reconstructed with bone graft. The patient hospitalized again for reconstruction of mid-facial hypoplasia 17 years after bone grafting. The physical examination (Figure 1A, 1B, 1C) and computed tomography scan revealed nasal deformity secondary to the bilateral severe zygomatic and maxillary hypoplasia and unfavorable result of right infra-orbital bone grafting (Figure 2A, 2B, 2C). The lateral cephalometric analysis demonstrated maxillary retrusion and mandibular protrusion with increased lower facial height. Upper incisors were labially inclined and lower incisors were uprighted.

TECHNIQUE

Standard Le Fort III osteotomy has been performed with bicoronal approach. After pterigo-maxillary junction separation with gingivobuccal insicion, infra-orbital rims are explored for the wire traction points for the Rigid External Distraction System (RED; KLS Martin, Jacksonville, FL). Upper traction wires were anchored directly around the infra-orbital rims, as they seemed strong and durable enough to handle the distraction forces. After completion of the Le Fort III osteotomy RED System mounted to the calvarium and lower traction wires attached to the applied intraoral splint to deliver distraction forces to the maxilla through the dentition (Figure 3). At the end of the 5 days of latent period the distraction has been initiated with the rhythm of once, 1mm per day. At the end of the first week of the distraction, right infra-orbital rim was fractured from the traction point. Right infra-orbital rim revealed avulsion from anchoring site, which was the previous bone grafting area during the exploration.



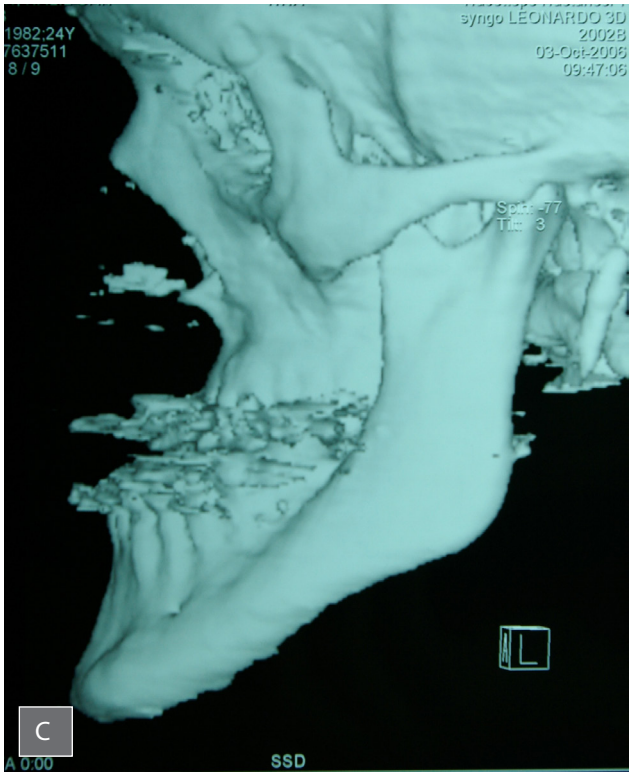


Figure 2. Tridimensional reconstruction of computerized tomography scans. Severe mid-face retrusion due to poor development of maxillary and zygomatic bones can be observed. The remnant of previous bone graft on the medial border of the infra-orbital rim can be detected,

A) Antero-posterior, B) Right lateral and C) Left lateral preoperative views.

Fracture was fixated with plate and screws and traction wire was anchored to the plate. The distraction procedure completed without any additional complication. The patient was followed weekly with lateral cephalograms and clinical examinations until satisfactory skeletal convexity, over-jet, over-bite, and relative stable occlusion was achieved. At the end of the eighth week of consolidation period, favorable radiological (Figure 4A, 4B, 4C) and clinical (Figure 5A, 5B) results obtained. Patient's profile was

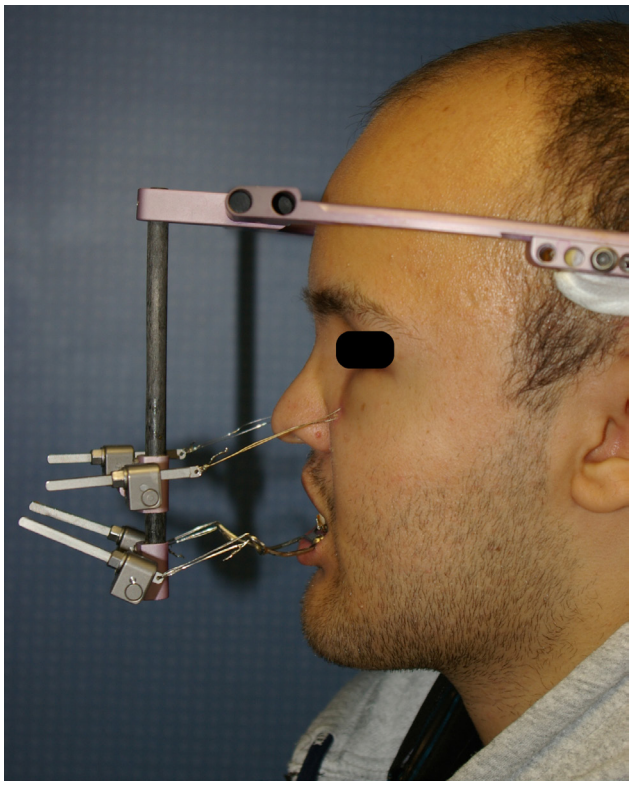


Figure 3. Application of RED system and traction vectors during distraction period.

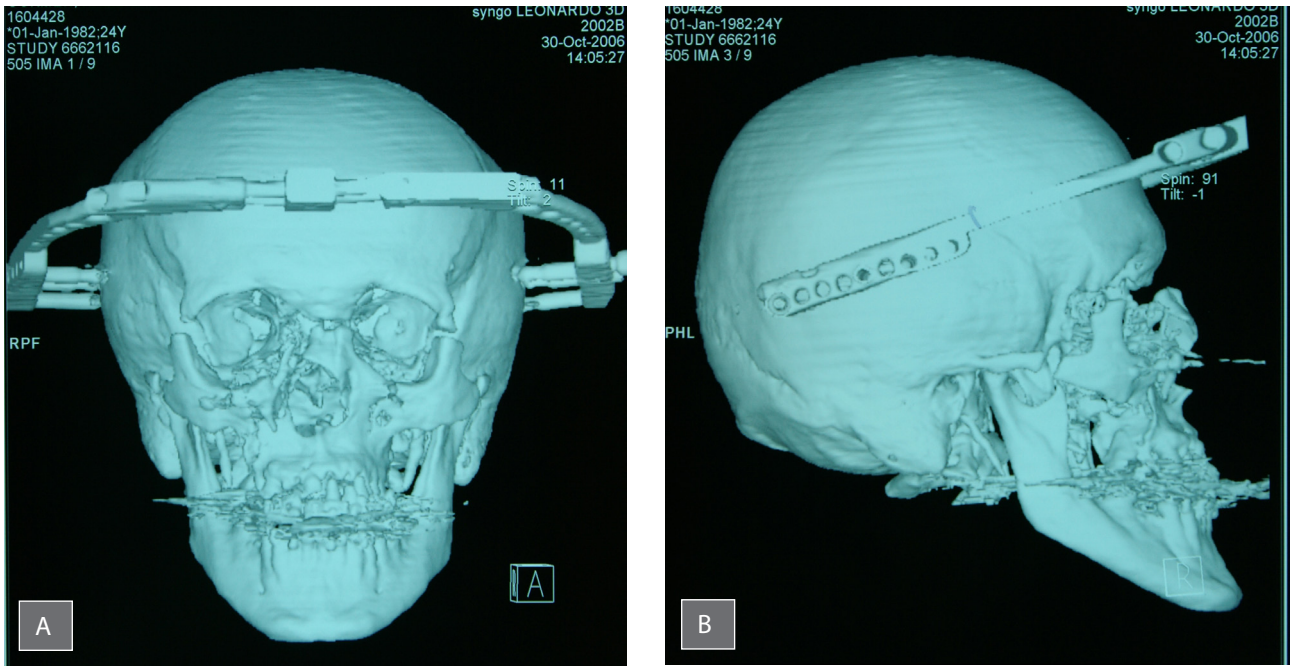


Figure 4. Tridimensional view after distraction period during consolidation stage.
Frontozygomatic (A), temporozygomatic and pterigomaxillary osteotomies (B, C) and distraction gaps can be observed.

esthetically pleasing with the advancement of maxilla and mid-face at the end of the treatment. The patient's complaint had been addressed. Due to bad oral hygiene patient lost many teeth and orthodontic treatment terminated earlier. Removable prosthesis was fabricated for the lost teeth. According to the lateral cephalometric analysis, 10.5mm of maxillary advancement was achieved at the level of point A according to FH⊥PTV. SNA and ANB angles increased. Convexity changed from -21mm to -7mm. Increase in FMA, GoGnSN angles and decrease in SNB, facial axis, facial depth angles supported the clockwise rotation of mandible. The soft tissue profile showed that nasal projection and nasolabial angle increased with upper lip retrusion reduction. These changes result from the effects of maxillary advancement by the RED system (Figure 6).

DISCUSSION

Head and neck malignancies of pediatric population are commonly treated by high dose chemo-radiotherapy protocols. As a result of these treatment modalities soft tissue and bony growth alterations often require unconventional surgical techniques. The facial skeleton appears most susceptible to high radiation doses before age six and at puberty, which are critical times of skeletal development [8]. Final deformity appears after puberty with the completion of the skeletal growth. The deformity differs from mild soft tissue damage to severe facial hypoplasia. Usually the deformity includes mandible and occurs in asymmetric manner [9, 3] where as our patient reveals symmetric mid-facial hypoplasia and nasal deformity secondary not only to radiotherapy, but also to surgical intervention. The patient seems to be the first case, which has midfacial deformity secondary to radiotherapy reconstructed with Le Fort III osteotomy and distraction osteogenesis with RED system in the literature. Maxillofacial reconstruction following tumor

Figure 5. Anteroposterior (A) and right lateral (B) views 6 mounts after operation.

surgery is a standing and challenging obstacle for the surgeons. Distraction osteogenesis might contribute to solve problem, related with ablative surgery or radiotherapy under certain circumstances. The technique has widespread clinical applications in treatment of hypoplastic skeleton of craniofacial anomalies, in treatment of defects due to cancer surgery, in management of both acute trauma treatment, in management of chronic traumatic growth disturbances, limb deformities and non-union. The Distraction osteogenesis has several advantages over conventional techniques; it is a less invasive intervention, easier control of infection and has no donor site morbidity when compared with reconstruction by autologous bone grafting or free flaps. In addition one of the most important advantages of DO is the expansion of the surrounding soft tissues that accompanies the bony regeneration without scar formation. However the patient has been attempted to treat with autologous bone grafting in 1989 before the application of distraction osteogenesis in craniomaxillofacial area, the results of the operation was far from being favorable. This poor result and severe mid-facial hypoplasia of the patient directed the authors to conduct distraction osteogenesis for establishing facial convexity and occlusion. However it has been offered to perform a rhinoplasty and mandibular set-back to achieve superior occlusion and aesthetic outcome at the beginning of the treatment, the patient refused to have additional surgical intervention due to satisfactory results of the mid-face distraction according to him. There are several reports on distraction osteogenesis in irradiated facial areas. These papers are generally about distraction osteogenesis of irradiated mandibles or facial asymmetries secondary

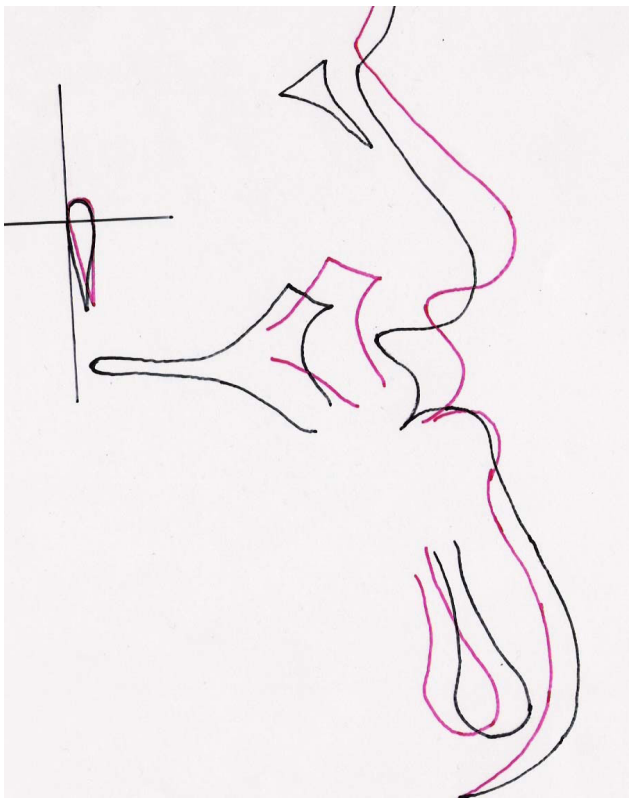


Figure 6. Superimposition of the lateral cephalometric analysis illustrating changes before and after treatment.

Table. Description of the 15 Cases of irradiated Cranio-maxillofacial reconstruction with distraction osteogenesis. (SCC: Squamous Cell Carcinoma, HBO: Hyper Baric Oxygen, LD: Latissimus Dorsi Musculocutaneous flap, REM: Rectus Abdominus Musculocutaneous flap)

Authors		Main Disease	Total Dose of Irradiation (Gy)	Distraction Area	Distraction Strategy	Reported bone quality	Latency Period (Day)		Complications	Additional intervention
Grover et al	1		Undeclared	Right Orbito-zygomatic		Excellent contour and projection	9		None	Prosthesis
Taub et al	1	SCC	45	Palate	Transport Distraciton	New bone	2	1	None	Mucoperiosteal flep repair
	3	SCC	60 to 70	Mandible		Acceptable Good Excellent	10	0.5	One Partial intra-oral exposure Onee hemimandibu-lar exposure One none	
Holmes et al	2	Clear cell odontogen-ic tumor Rabdomyo-Sa	60 Unknown	Mandible Mandible	Horizontal Vertical	No bone for-mation No bone formation	5 5	1 1	Distractor fracture Distracture fracture	Plate fixation Bone graft+Plate fixation
Lazar et al	1	SCC	40	Mandible	Vertical	New Bone	7	1	None	Dental İmplants
Elsalanty et al	1	Rhabdomyo-Sa	50	Mandible	Trifocal	New bone		0.6 and 1.2	Non union at dock-ing site of distract-ing segments Pin infections	HBO therapy Bone Graft İnternal fixation
Kashiwa et al	5	Rhabdomyo-Sa	50	Mandible	Transport	Good	7 to 10	0.25 to 1		Le Forte I LD with rib and scapula
		Sarcoma of in-fratemporal fossa	50	Mandible	Horizontal	One side Good and One side Poor			Fibrous union	LD with rib and scapula Bone graft and Plate fixation
		Oral Carcinoma	40	Mandible	Horizontal	One side Good and One side Fair			Fracture of new bone	RAM with rib Plate fixation of fracture sites
		Oral Carcinoma	30	Mandible	Horizontal	Good				RAM with rib
		Oral Carcinoma	30	Mandible	Horizontal	Good				RAM with rib
	1	SCC	60	Mandible	Horizontal	No bone formation	7	0.5		HBO Bone Greft+
Konas et al	1	Clear Cell Granuloma	unknown			New Bone formation	5	1	Infraorbital rim frac-ture (Traction point)	Infraorbital rim Plate and screw fixation

to unilateral irradiations [3,4,6,10,11,12,13]. Only one case was related with distraction of unilateral peri-orbital and zygomatic region with RED system [9] and one case was related with palatal advancement for oro-nasal fistula repair secondary to tumor abla-tion and irradiation [14]. However we found limited numbers of the cases in the literature, the survey of reports seems to indicate that complication rates in-creases with the elevation of total irradiation doses (Table). Raghoebar also suggested that there might be a radiation threshold above which mandibular distraction will fail [10]. The applied distraction regi-ments seem to be a little bit different than standard distraction osteogenesis procedures. The surgeons

tend to lengthen the latency period and lower the distraction rate due to avoid complications about bone regeneration (Table). The hypothesis of diminution in cell function and impairing optimal bone regeneration by radiation has been shown by Fregene et al. [5] with the discovery of significant increase of low mineralized, immature bone and significant decrease of highly mineralized, mature bone in irradiated regenerate. Despite these negative effects of irradiation on bone regeneration, distraction osteogenesis seems to be one of the major treatment modality for skeletal augmentation of irradiated cranio-maxillofacial skeleton. The expanding effect the technique on soft tissue coverage also provides additional advantages like avoiding new scar formation when compared to alternative treatment methods like free tissue transfers. Although, Holmes et al. [11] and Raghoebar et al. [10] reported unsuccessful distraction of irradiated mandibles, in our patient we have not inspected any problem with bone regeneration and soft tissue coverage on irradiated area except for poor right infraorbital rim bone quality which led us to use plate and screw fixation to create stronger anchorage point for traction force. The favorable bone regeneration of our patient may be related with superior vascularization of mid-face and larger contact area of the maxilla with the surrounding well-vascularized soft tissue.

CONCLUSION

Application of standard Le Fort III osteotomy in irradiated area might be conducted for individual cases resembling mid-facial hypoplasia. Bony reconstruction of the radiation induced maxillofacial hypoplasia with distraction osteogenesis might be considered as first step treatment method for patients with good quality covering soft tissue in cranio-maxillofacial region.

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