Short Communication

Simultaneous maxillomandibular distraction osteogenesis in hemifacial microsomia: Report of 7 cases

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ABSTRACT

Introduction: The goal in treating Hemifacial microsomia (HFM) is to improve facial symmetry, allow functional jaw movement, obtain stable occlusion, and achieve patient satisfaction. This study was planned with an aim to assess the outcome of simultaneous maxillomandibular distraction osteogenesis in hemifacial microsomia.

Material & methods: Seven patients with hemifacial microsomia were included in this study and demographic data was obtained. Simultaneous maxillo-mandibular distraction was planned for correction of maxillary cant, vertical ramal lengthening and midline, by performing unilateral mandibular osteotomy in ramus and distractor fixation, a Le Fort I osteotomy, with fixation at contralateral zygomatic buttress and intermaxillary elastics. Derriford Appearance Scale was used to measure individual responses to address problems of appearance.

Results: 6 males and 1 female, with age range 18–26 years, mean 22.14 were included in the study. Maxillary occlusal cant varied from 4–16°, mean 9.85° and distraction achieved ranged from 12–19 mm, mean 15.57. Preoperatively DAS score ranged from 42–61, mean 51.7 and improved to 10–27, mean 18.4. 100% success was achieved in 5 cases, while other two showed 96% and 99% success.

Conclusion: Simultaneous maxillomandibular distraction osteogenesis improves facial esthetics and obviates the need for postoperative orthodontics, minimizing the treatment period.

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1. Introduction

Hemifacial microsomia (HFM) is a term often used interchangeably with otomandibular dysostosis, lateral facial dysplasia, malformation syndrome of the first and second arches, temporal oculoauricular dysplasia, or craniofacial microsomia. HFM includes some degree of developmental abnormality of the facial skeleton involving mandible, maxilla, zygoma, temporal bone, ear, and soft tissues.¹ HFM presents with mild to gross facial asymmetry, retracted mandible, microtia, slanted lip commissure as well as occlusal canting.

Treatment of HFM poses a challenge as both soft and hard tissue deficiencies are to be addressed. The goal in treating HFM is to improve facial symmetry, allow functional jaw movement, obtain stable occlusion, and achieve patient satisfaction.

This study was planned with an aim to assess the outcome of simultaneous maxillomandibular distraction osteogenesis in hemifacial microsomia.

2. Material & methods

Seven patients with hemifacial microsomia, who visited our outpatient clinic of oral & maxillofacial surgery between jan 2015–july 2016, were included in this study. Institutional ethical clearance was obtained. All patients were informed about the surgical procedure and possible complications, and consent for the treatment was obtained and recorded.

Demographic data was obtained (Table 1). Postero-anterior and lateral cephalograms with computed tomography and its three-dimensional reconstruction were done for surgical planning to objectively delineate asymmetries, measure maxillary and mandibular deviations from midline, differences in ramus height, and occlusal cant.

Simultaneous maxilla-mandibular distraction was planned for correction of maxillary cant, vertical ramal lengthening and midline. All patients were operated by a single surgeon. The surgical protocol included a unilateral mandibular osteotomy with

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Table 1

Demography and treatment outcome of hemifacial microsomia patients.

S No	Age (yrs)	Sex	HFM Type	Tt Plan	Max cant (°)	DO (mm)	Device EO/IO	Device L/MV	Complication	Success %
1	21	M	Ila	SMDO	5	12	IO	L	None	100
2	24	M	IV	SMDO, G	16	17	EO	MV	None	100
3	18	F	IV	SMDO, G	11	15	EO	MV	none	100
4	26	M	Ila	SMDO, G,M,D, S	7	14	IO	L	none	100
5	22	M	IV	SMDO, G,M,D, S	14	19	IO	L	Medpore angle exposed, removed	96
6	20	M	Ila	SMDO	4	14	EO	L	parotid fistula, managed by compression bandage 2 months	99
7	24	M	Ila	SMDO	12	18	EO	L	none	100
Mean	22.14				9.85	15.57				99.28

SMDO: simultaneous maxillo-mandibular distraction osteogenesis, M: Medpore, D: dermal fat, G: Genioplasty, S: contralateral lower border shave, IMF: intermaxillary fixation, EO/IO: extraoral/intraoral, L/MV: linear/multivector. HFM types: **I**: Hypoplastic TMJ, **Ila**: Hypoplastic, abnormal shape of mandibular ramus, condyle, TMJ, **Ilb**: mandibular ramus hypoplastic and markedly abnormal in form and location, being medial and anterior, **III**: Absence of mandibular ramus & **IV**: Mandibular body hypoplasia.

fixation of the distractor device to the ramus and a complete Le Fort I osteotomy, but no down fracture. Fixation at the contralateral zygomatic buttress was done using a 4 hole titanium miniplate and intermaxillary elastics were maintained during the distraction phase.

DAS: Derriford Appearance Scale² (Table 2) was used to measure individual responses to address problems of appearance, and was recorded pre and postoperatively at 1 year follow up.

Success rate was defined as 100% if the planned goal of cant correction and ramal lengthening was accomplished.

3. Results

Six males and one female patient of hemifacial microsomia were included in the study. Their age ranged from 18 to 26 years, mean 22.14 years. Four cases were Pruzansky Kaban type Ila, and

Table 2

Changes in DAS score with treatment in hemifacial microsomia patients.

	1			2			3			4			5			6			7			Mean pre	Mean post
	Pre	Post	change	Pre	Post	change	Pre	Post	change	Pre	Post	change	Pre	Post	change	Pre	Post	change	Pre	Post	change		
1. Feel confident	2	5	3	1	4	3	0	5	5	1	5	4	0	4	4	2	5	3	2	5	3	1.4±0.8	4.7±0.48
2. Distress at reflection	3	0	3	4	1	3	5	0	5	4	0	4	5	2	3	4	1	3	4	0	4	4.16±0.75	0.51±0.78
3. Irritable at home	2	1	1	4	1	3	4	0	4	3	0	3	4	1	3	2	1	1	1	0	1	2.8±1.2	0.57±.53
4. Feel hurt	2	0	2	4	1	3	5	0	5	4	0	4	4	1	3	4	2	2	3	0	3	3.7±.95	0.57±.78
5. Self consciousness affects work	4	1	3	4	2	2	5	1	4	4	0	4	4	1	3	5	3	2	2	0	2	4±1	1.1±.07
6. Distressed outside home	3	1	2	4	1	3	5	1	4	3	0	3	5	2	3	3	1	2	4	0	4	3.8±.89	0.85±.7
7. Misjudged due to appearance	3	2	1	5	1	4	5	0	5	5	0	5	5	2	3	3	2	1	4	1	3	4.3±.95	1.3±1.1
8. Self conscious of appearance	5	1	4	5	2	3	5	1	4	5	0	5	4	1	3	5	2	3	4	0	4	4.7±.48	0.81±.66
9. Irritable at work	2	1	1	4	0	4	4	1	3	3	0	3	4	1	3	4	2	2	0	0	0	3±1.5	0.71±.75
10. Adopt concealing gestures	5	1	4	5	2	3	4	0	4	4	0	4	2	0	2	5	2	3	4	0	4	4.1±1	0.71±.95
11. Feel rejected	5	0	5	5	1	4	5	0	5	4	0	4	4	0	4	5	0	5	4	0	4	4.5±.53	0.14±.37
12. Close into shell	5	0	5	4	1	3	5	1	4	3	0	3	3	0	3	5	2	3	4	0	4	4.1±.89	0.57±.78
13. Feel normal	0	5	5	0	4	4	0	5	5	1	5	4	0	4	4	0	3	3	0	4	4	0.14±.37	4.3±.75
14. Avoid leaving house	2	0	2	4	1	3	5	1	4	3	0	3	4	1	3	2	0	2	1	0	1	3±1.4	0.42±.53
15. Avoid restaurants	4	1	3	4	0	4	4	0	4	3	0	3	4	1	3	4	1	3	5	4	1	4±.57	1±1.4
TOTAL	47	19	44	57	22	49	61	16	65	50	10	56	52	21	47	53	27	38	42	14	42	51.7±6.26	18.4±5.6

Each scored 0–5, where 0 is least and 5 is maximum.

Fig. 1. a. Preoperative, b. mandibular distractor in place, c. Lefort I osteotomy & contralateral fixation, d. post distraction, e. Preoperative PA cephalogram, f. postdistraction PA cephalogram, g. 1 year follow up PA cephalogram.

Fig. 2. a. Preoperative front view, b. preoperative profile view, c. preoperative 3D CT front view, d. preoperative 3D CT profile view, e. postdistraction front view with mandibular multivector distractor in place, f. postdistraction profile view with distractor in place, g. preoperative occlusion, h. post operative occlusion.

three were type IV. Maxillary occlusal cant varied from 4–16°, mean 9.85°. Distraction distance ranged from 12–19 mm, mean 15.57 mm. External distractor was used in 4 cases, while three had internal distractors (Fig. 1). Linear vertical distraction was achieved in five cases, but two required multivector distraction (Fig. 2). Simultaneous maxillo-mandibular distraction osteogenesis was carried out in all seven along with a LeFort I osteotomy and its fixation on contralateral side. Medpore augmentation on the angle region, dermal fat graft (Fig. 3) and contralateral lower border shave were done additionally in two cases, while genioplasty was performed in four cases.

In one case the medpore implant got extruded because of tension in the overlying skin and had to be removed at 1 month postoperatively. In another patient, during the distraction phase there was a parotid fistula, which was later managed by compression bandage and healed within 2 months. All patients showed improvement in facial appearance with correction of maxillary cant and vertical ramal lengthening.

Preoperatively DAS score ranged from 42–61, mean 51.7 and improved to 10–27, mean 18.4. The mean change in DAS score was 48.7. 100% success was achieved in 5 cases, while other two showed 96% and 99% success.

4. Discussion

HFM is the second most common craniofacial birth defect and is estimated to effect 1 in 3600–5600 live births in the United States each year. It is more prevalent in boys with a 3:2 reported ratio.¹ The Pruzansky–Kaban classification of HFM consists of four types. Type I is a small mandible with normal morphology. Type IIa is a mandibular ramus abnormal in both size and shape; type IIb is a mandibular ramus and temporomandibular joint abnormal in size,

morphology, and location. A type III deformity consists of an absent ramus, condyle, and TMJ.³ Kaban outlined four main treatment objectives for such patients: increase the size of mandible and associated soft tissue, create temporomandibular joint if lacking, foster vertical maxillary growth, and obtain a stable occlusion.

The first report of surgery in children with HFM dates back to 1941.⁴ Up to 1980s, reconstruction of the severely hypoplastic mandible was achieved by mandibular osteotomy and costochondral cartilaginous rib graft, although rib, iliac crest, fibula, and temporal bone were also used.⁵ However, most of these procedures were associated with poor aesthetics, relapse, functional problems, graft resorption, and unpredictable growth.⁶ Soft tissue deficiencies were usually not addressed until after restoration of bony symmetry.⁷

Several cases of mandibular corpus distraction osteogenesis (DO) were reported in the literature during the first half of the twentieth century, but the technique did not gain popularity until the early 1990s when McCarthy⁸ at New York University began to perform the procedure. Advent of bidirectional distraction devices took the technique further ahead. Shaw⁹ published a critical appraisal of 88 publications on DO over the period 1995–2000. The Eurocran Distraction Study was an important landmark in obtaining more insight into the use of DO and its clinical results.¹⁰

Compared to costochondral cartilage graft, mandibular distraction osteogenesis shows an advantage as it allows stable expansion of mandible with concurrent lengthening and expansion of surrounding muscle and soft tissue, increased vertical length of the mandible, greater bone stock, improved soft tissue asymmetry and less relapse. Additionally, shorter operative times; fewer complications; and simpler postoperative course with less blood loss, greater vector control, no donor site morbidity, and an ability

Fig. 3. a. Preoperative front view, b. preoperative 3D CT front view, c. PA cephalogram with distractor in place, d. medpore onlay implant at angle fixed with screws, e. dermal fat graft, f. genioplasty, g. postdistraction front view.

to lengthen the mandible at a younger age are the other reported advantages. DO can be employed at any age from infancy to adulthood, applied extraorally or intraorally, and is repeatable. It stretch-expands the soft-tissue envelope concurrently with bone-generation, and can even distract bone grafts.¹¹ However, there is a need for a second surgery for removal of the intraoral distractor at the end of the distraction and bony overgrowth on the device, makes its removal difficult.¹² Other potential problems reported are patient noncompliance, pin tract infection, osteomyelitis, nerve injury, tooth injury, nonunion/malunion, and possible relapse.

In HFM patients, with existing costochondral graft, a three-dimensional device was used to correct malocclusion and facial asymmetry using a dovetail geometrical pattern osteotomy to increase the osteotomy surface area and offer protection to the tension-sensitive structures in the center of the distraction zone, allowing early bone formation and mineralization adjacent to the center zone in the distraction gap.¹³ Mandibular distraction osteogenesis has been seen to improve facial asymmetry and retrognathia (50.1%), correct the slanted lip commissure (24.7%), and improve or level the mandibular occlusal plane (11.1%).⁵ Shetye¹⁴ suggested use of functional orthodontic appliances to counterbalance the muscular forces acting on the distraction regenerate to minimize relapse. Sagittal split ramus osteotomy and intraoral curved distractor were reported to be efficacious to lengthen the mandibular body and ramus, as the sagittal split permitted immediate rotation of the proximal segment and lengthening while still providing a large surface interface for the regenerative bone.¹⁵ Reports also stress on multi-stage correction of the facial deformity using both DO and grafts.¹⁶ Ow⁵ realized the requirement of additional soft-tissue augmentation procedures, as soft-tissue expansion from distraction was not fully able to correct the lateral depression at the mandibular angle region. Nagy¹⁷ reported a lack of statistical evidence to support use of DO as a single treatment modality before the permanent dentition stage.

Monasterio¹⁸ was the first to introduce simultaneous mandibular and maxillary DO with a single mandibular-distractor in adults with HFM to overcome the problem of malocclusion and the unacceptably prolonged and difficult orthodontic treatment. He left the pterygomaxillary junction of the unaffected side, the nasal septum and the nasal spine intact, to act as a pivot point and improve the stability of the midface. Padwa¹⁹ carried out pterygomaxillary disjunction bilaterally but no down fracture and no detachment of nasal septum. IMF was done in distraction as well as consolidation phase. Anna²⁰ demonstrated improved facial balance and symmetry in eight patients (4 preadolescents, 4 adolescents) of grade II mandibular deformity with simultaneous maxillary and mandibular distraction following a LeFort I osteotomy, wire fixation at uninvolved side, ipsilateral horizontal mandibular ramus osteotomy, a semiburied distractor placed over ramus, and IMF. Shehata²¹ successfully treated 6 hemifacial microsomia with simultaneous maxillary and mandibular distraction following LeFort I osteotomy without doing any down fracture. We performed detachment of nasal septum and pterygomaxillary disjunction bilaterally but no down fracture in our series. Choi²² too reported successful treatment of facial asymmetry, and a skeletal Class II profile in a period of 27 months, by lengthening mandibular ramus with distraction osteogenesis, and correcting maxillary transverse deficiency with miniscrew-assisted rapid palatal expansion.

In contrast, Scolozzi²³ performed simultaneous distraction using two separate distractors for maxilla and mandible. This eliminated the need for IMF and hence decreased postoperative discomfort, earlier normal jaw function, easier maintenance of oral hygiene, better nutrition, and higher patient acceptance and satisfaction. Using same protocol, Lu²⁴ demonstrated good lasting outcomes at completion of growth with single-stage simultaneous

maxillo-mandibular distraction using double distractors in maxilla and mandible and no IMF.

Postoperative orthodontic treatment is required to achieve tooth alignment and closure of the posterior open bite when mandibular distraction alone is performed. However, bimaxillary DO obviates the need for postoperative orthodontic treatment.²⁵

Without any evidence based studies, there still exists a disagreement on the choice of osteotomy site (ramus, body or angle), and on the choice of DO device, as several devices and techniques are available.¹⁰

5. Conclusion

Simultaneous maxillomandibular distraction osteogenesis in hemifacial microsomia improves facial esthetics and obviates the need for postoperative orthodontics, minimizing the treatment period.

Conflict of interest

The authors have none to declare.

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