Case Report

# An Interdisciplinary Management of Severe Facial Asymmetry Due to Hemifacial Microsomia


## Abstract

This case report outlines the importance of an interdisciplinary approach and a patient centric outcome for the treatment of facial asymmetry resulting from hemifacial microsomia. Different treatment modalities are available to treat asymmetries. However, the best treatment outcome can be achieved only when the treatment plan is individualized for every patient. This report portrays an adult patient with hemifacial microsomia, who had facial asymmetry from the level of supraorbital region with a Class II skeletal base. Orthodontic treatment, along with surgical management, was required to transform the patient into a symmetrical profile, which is esthetically pleasing. A sound knowledge of the various technologies and resources that are available to us and making the best use of it to bring out a drastic change in the patient's life. Combined effort of the orthodontists and oral surgeons are required to manage patient's with hemifacial microsomia that has caused the severe facial asymmetry. Moreover, special attention has to be given to a patient-centric outcome.


**Keywords:** *CADCAM splint, facial asymmetry, hemifacial microsomia, NemoCeph Software, orthognathic surgery*


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## Introduction

The term "asymmetry" refers to variations among homologous elements, causing an imbalance between structures. Significant facial asymmetry resulted in the patient with not only functional problems but also cause esthetic concerns.[1] Severt and Proffit reported frequencies of facial laterality of 5%, 36%, and 74% in the upper, middle, and lower thirds of the face, respectively.[2]

Hemifacial microsomia may be the cause of facial asymmetry in 30%–79% of patients [Table 1].[3] It is an asymmetric development of the craniofacial structure, which results from hypoplasia of the first and second branchial arch structures.[4] It is the second-most prevalent defect of craniofacial structures after cleft lip and palate.[5] Hemifacial microsomia also known as first and second brachial arch syndrome, oral-mandibular-auricular syndrome, lateral facial dysplasia, or oto-mandibular dysostosis is often a congenitally developed disorder.[6-9]

The degree of asymmetry determines the treatment modality. Slight facial asymmetries are common and treated using conventional orthodontic mechanics. However, a more severe form requires a combined effort of an orthodontist and oral maxillofacial surgeon to resolve it.

This case report deals with a patient who had a congenitally developed asymmetry from the supraorbital level due to hemifacial microsomia, which consequently led to a skeletal, dental and soft tissue asymmetry. To correct; extraction of 4 first premolars, bi-jaw surgery and genioplasty were sequentially performed on the patient.

## Case Report

An adult patient sought treatment with the major complaint of facial deformity and noticeable jaw deviation to the left side. The patient was born full term to non-consanguineous parents. The patient had no significant prenatal history. Family history revealed that her younger brother had delayed milestones and was dumb.

The patient had a previous history of incomplete cleft palate, which was treated by primary palatoplasty at the age of 3 years and bifid uvula, which was treated by uvuloplasty 3 months ago.



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On general examination, the patient was found to be moderately nourished and built with psychological impairment.

On extraoral examination [Figure 1], gross facial asymmetry from the supraorbital region was noted. The eyebrow, eye, ala of the nose, and corner of the mouth were placed at a higher level on the left side compared to the opposite side. The chin was shifted toward left by 6 mm at rest, which deviated further on mouth opening. The left side of the face showed a primitive ear, Grade III microtia,[10] with hearing deficit. The fullness of the face on the right was seen. Lip incompetency with 90% of incisal exposure at rest was noted. The examination of muscles of mastication revealed no significant findings.

On intraoral examination [Figure 2], the patient had Katz's Class I premolar relationship with proclined upper anterior, rotation of 22,13,43, spacing in 13, 11, 21, lingually placed 33, lower dental midline shift of 3 mm to the left. Severe canting of the occlusal plane in the transverse dimension was noted [Figure 3]. Arrested caries were seen in 26, 36, 37, and 46 along with generalized enamel hypoplasia.

Radiographic evaluation with lateral cephalogram revealed a skeletal class II malocclusion with backwardly rotating mandible on the high mandibular plane angle with a retrusive chin [Figure 4]. The Three-dimensional (3D) reconstruction of the computed tomography (CT) reveals dissimilarity between the left and right orbit, zygoma, maxilla, and mandible [Figure 5]. OPG analysis[11] revealed asymmetric levels of the glenoid fossa, ramal height, and irregularly shaped left condylar morphology with chin shift towards left by 6 mm.

The patient was diagnosed as Hemifacial microsomia with Katz's Class I premolar relation with skeletal class II malocclusion attributing to backwardly rotating mandible with proclined upper anteriors, rotation of 22.13 43, spacing in 13, 11, 21, lingually placed 33 and transverse canting of the occlusal plane with a retrusive chin.

## Treatment objectives

The treatment objectives were to: (1) correct the asymmetric facial appearance, (2) correct the convex facial profile, (3) resolve the dental malalignment, (4) establish normal overbite and overjet, and (5) achieve an ideal occlusion.

## Treatment plan

Orthodontic treatment associated with combined orthognathic surgery in the maxilla and mandible, with the

**Figure 1: Pretreatment extra oral photograph**

**Figure 3: Occlusal can't examination**

**Figure 2: Pretreatment intra oral photograph**

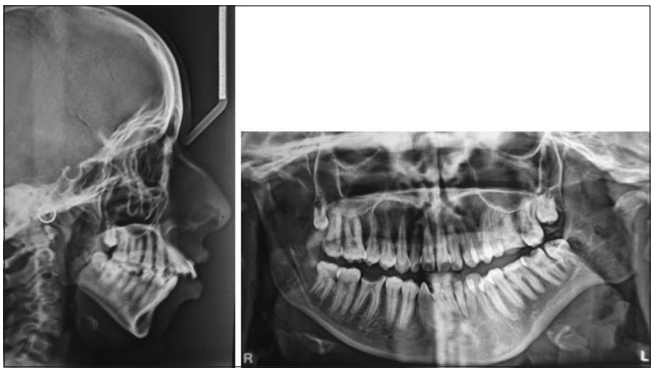

**Figure 4: Pretreatment radiographs**

extraction of 14, 24, 34 and 44, followed by Le-Forte I maxillary osteotomy with differential impaction of 7 mm and 3 mm on the right and left sides, respectively, and BSSO cut with 3.5 mm of superior positioning on the right side and 5 mm of inferior positioning on the left side of the mandible with an iliac bone graft on the deficient area on the left. The orthodontic treatment would be finished in a Class I molar, premolar, and canine relationship and dental midlines coincident to each other and to the facial midline.

Following orthodontic and surgical management, plastic surgery was proposed for the correction of the left ear to restore aesthetics. The patient is referred to an otolaryngologist for the hearing deficit. The patient was advised restorative management of the arrested caries teeth and oral prophylaxis.

## Treatment progress

Based on the diagnostic data, the ideal treatment plan was performed. It contained three phases of management.

### *Phase I: Presurgical orthodontic phase*

The principle step of this phase is to align the arches and achieve leveling of all the teeth. The case was started by bonding 0.022" ×0.028" MBT pre-adjusted edgewise prescription appliance, and all the first premolars were extracted. Initial leveling and aligning were carried out with 0.016" NiTi, 0.016" ×0.022" NiTi, 0.017" ×0.025" NiTi and 0.019" ×0.025" NiTi followed by 0.019" ×0.025" SS for retraction of upper and lower anterior and 0.021" ×0.025" SS as final stabilizing wires. After 4 weeks of final stabilizing archwire, upper and lower arch impressions and other pre-surgical records were taken for surgical planning.

The dental cast was articulated into a semi-adjustable articulator with the help of a face bow transfer [Figure 6]. The mock surgery was done manually and also digitally using NemoCeph Software [Figure 7].

The articulated casts [Figure 8] also helped in the fabrication of the surgical computer-aided design & computer-aided manufacturing (CADCAM) splints. An intermediated CADCAM splint [Figure 9] was made after the superior impaction of the maxilla as according to the treatment plan. The mandibular cast was then repositioned accordingly [Figure 10], and the final occlusal splint was fabricated [Figure 11].

### *Phase II: Surgical phase*

Le-Forte I osteotomy cut was carried out with superior maxillary impaction on the right by 7 mm and on the left by 3 mm. Bilateral sagittal split osteotomy cut was made, and the mandible was repositioned superiorly by 3.5 mm on the right and inferiorly positioned on the left by 5 mm (thus achieving a total of 10.5 superior positionings of the right side and 2 mm inferior positioning of the left side). The deficient area on the left side of the mandible was grafted

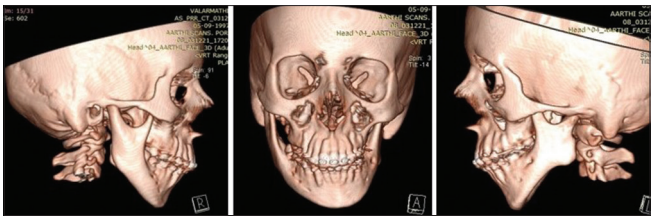

**Figure 5: Three-dimensional reconstruction of computed tomography**

**Figure 6: Facebow transfer**

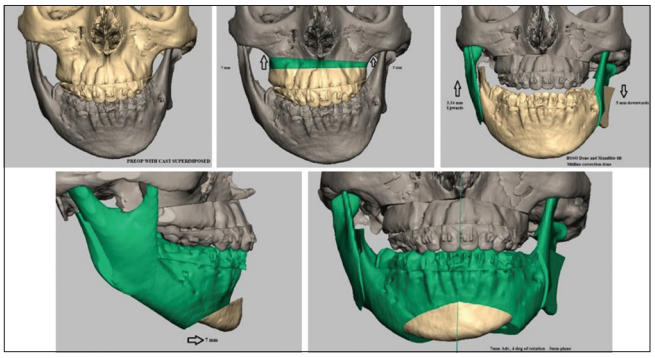

**Figure 7: Digital mock surgery using nemoceph**

with a monocortical bone graft (3 mm × 2 mm), which was harvested from the right iliac bone. This was followed by genioplasty, wherein it was rotated to the right by 3 mm and advanced by 7 mm. Rigid type fixations were used in both jaws using mini plates and screws on both sides. The patient was hospitalized for 2 days and was advised to maintain a soft diet for the first 2 weeks postsurgery. By 8 weeks, the patient was back on a normal diet.

### *Phase III: Postsurgical orthodontic phase*

Once initial healing was satisfactory, and the patient had attained an average range of motion, active orthodontic treatment was resumed. The heavier 0.021" X 0.025" SS wires were replaced with a smaller dimension 0.017" X 0.025" TMA in the upper arch and 0.016" SS round wire in the lower arch. The patient was instructed to wear elastics for settling, which were weaned off as further detailing of occlusion took place. Once the settling was completed, the fixed orthodontic appliance was debonded. Retention protocol with upper Begg's wrap around and lower lingual

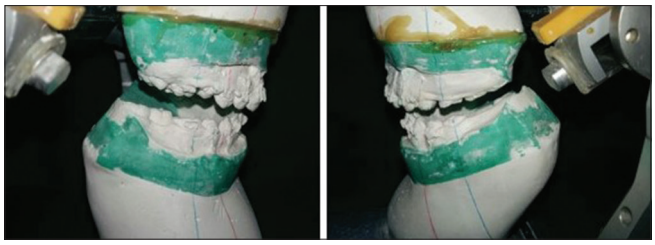

**Figure 8: After maxillary mock surgery**

**Figure 10: After bi jaw mock surgery**

**Figure 9: Intermediate CADCAM splint fabrication**

**Figure 11: Final CADCAM splint fabrication**

bonded retainer was advocated. Post-treatment records were taken [Figures 12-14].

Treatment was completed in 15 months. Acceptable facial symmetry was achieved, the profile was improved, facial heights were made average, which cumulatively resulted in a pleasing profile. A symmetrical facial midline, harmonious facial profile and dental occlusion were obtained from treatment.

The patient reported back to the department after 9 months with a complaint of a swelling on the left side of the face and pus discharge near the lower back tooth region. On examination, it was detected that she developed an infection around the mandibular left bone plate. Immediate surgery was performed under general anesthesia to remove the infected bone plates. Pus sample collected from the site was sent for culture test to decide the required treatment regimen.

The patient was recalled 3 months after the removal of the infected plate (1 year after debonding) for a review. The overall treatment resulted from orthodontic and surgical management were found to be stable [Figures 15-18]. She had a pleasing profile and a more confident smile with an overall improvement in her social and psychological well-being.

## Discussion

The etiology of facial asymmetry can be grouped into three main categories[12-15] (I) Congenital, originating prenatally; (II) Acquired, resulting from injury or disease; and (III) Developmental, arising during growth with inconspicuous etiology [Table 2].

Cohen used the term "hemi-asymmetries" in discourse of craniofacial asymmetry. He further classified these conditions into hemi-hyperplasia, hemi-hypoplasia, hemi-atrophy, and other miscellaneous entities.[3] Bishara classified it according to the structures involved in the asymmetry [Table 3].

**Table 1: Signs and symptoms of hemifacial microsomia**

| |
|---|
| Facial asymmetry |
| Abnormalities of the outer ear such as absence, reduced size (hypoplasia), and/or displacement |
| Small and/or flattened maxillary, temporal, and malar bones |
| Deafness due to middle ear abnormalities |
| Ear tags |
| Abnormalities (in shape or number) of the teeth, or significant delay of tooth development |
| Narrowed mandible (jaw) or absence of half of the mandible |
| Cleft lip and/or palate |
| Reduced size of facial muscles |
| Abnormalities of the eyes (extremely small or absent) |
| Skeletal abnormalities including problems of the spine or ribs |
| Absence of cheek muscles or nerves supplying those muscles (resulting in an uneven smile) |

Hemifacial microsomia is characterized by maxillomandibular hypoplasia and facial asymmetry, which varies from a mild asymmetry in the face, to severe under-development of one facial half, with orbital implications, a partially formed ear or even a total absence of the ear.[4]

The Pruzansky-Kaban classification of hemifacial microsomia describes three mandibular types based on the status of the condyle-ramus-glenoid fossa unit: Type I (temporomandibular joint and ramus are well formed but smaller than normal), type II (temporomandibular joint, ramus, and glenoid fossa are hypoplastic and malformed, and sometimes malpositioned), and type III (temporomandibular joint, ramus, and glenoid fossa are absent).[16,17] In this case report, the patient had a severe form of type II hemifacial microsomia along with unilateral ear deformity.

Although there are numerous theories for the development of hemifacial microsomia based on embryological, clinical, and laboratory studies, the exact etiology is unknown. Laboratory studies suggest an early loss of neural crest

**Table 2: Classification based on etiology**

| Dental | Skeletal | Muscular | Functional |
|---|---|---|---|
| 1. Congentially missing tooth or teeth | 1. Involving | 1. Hemifacial microsomia | 1. Centric prematurities causing a lateral mandibular displacement on full closure from initial tooth contact position to habitual occlusal position |
| 2. Premature loss of deciduous teeth | Maxilla, or/and Mandible | 2. Mobius syndrome | |
| 3. Deleterious oral habits such as digit sucking resulting in asymmetric open bite | Number of skeletal structures on one side of the face, as in hemifacial microsomia and Treacher-Collins syndrome | 3. Cerebral palsy | 2. Presence of malpositioned tooth, dental crossbite, constricted maxillary arch or anteriorly displaced articular disc usually results in functional deviations |
| 4. Midline discrepancies | | 4. Unilateral masseter or temporal muscle hypertrophy | |
| 5. Occlusal discrepancies in first, second or third order plane | | 5. Long-term untreated cases of torticollis causing fibrosis of the sternocleidomastoid muscle | |

**Figure 12: Extraoral photograph on the day of debonding**

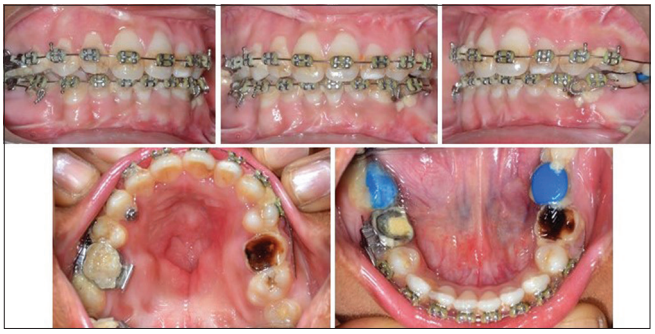

**Figure 13: Intraoral photographs on the day of debonding**

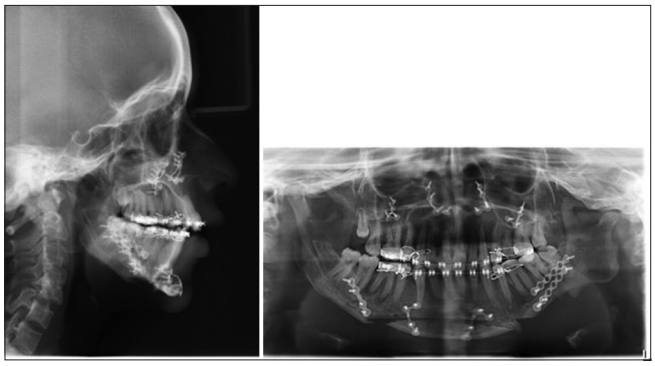

**Figure 14: Radiograph on the day of debonding**

**Figure 15: One year follow-up extraoral photograph**

cells may be a specific factor.[18] However, pathogenically, the etiology is said to be heterogeneous.[14]

Nowadays, modern dentistry pays more attention to a patient-centric outcome; hence, the treatment plan should consider not only the treatment result and duration but also the possible physical and psychological enhancement.

Treatment of hemifacial microsomia varies depending on the features present and the severity in each affected person. In this patient, asymmetry above the level of maxilla did not produce major esthetic or functional predicament, whereas the asymmetry in the lower third of the face was a major apprehension. Thus, the asymmetry present in the maxillo-mandibular region was given prime importance for rectification.

During growth, orthopedic or functional appliance[17] can be used to control growth and minimize the extent of orthognathic surgery required. Maxillary cant can be corrected with bite block or high pull headgear.[19-21] Both of these methods require significant patient cooperation, and it is difficult to control the direction and quantity of tooth movement.[21-23] However, for adults with a severe skeletal canting, as seen in our case, this treatment option was invalid.

In 1992, McCarthy *et al*.[24] did distraction osteogenesis in patients with hemifacial microsomia. However, studies have not shown stable results after mandibular ramus lengthening by distraction osteogenesis, with relapse often occurring on the elongated side.[17,25-27] Moreover, it worsens an already elongated face. Thus, this treatment modality could not be considered for this patient.

Hyo-Won Ahn *et al*. used a combination of corticotomy and orthopedic force using TADs for correction of maxillary cant and asymmetrical mandibular setback for mandibular asymmetry. We could not advocate this modality as corticotomy should be performed in two stages, which increased treatment timing. Moreover, intrusion with

**Figure 16: One year follow-up intraoral photograph**

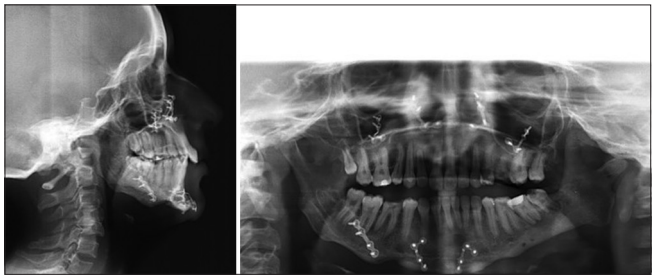

**Figure 17: One year follow-up radiograph**

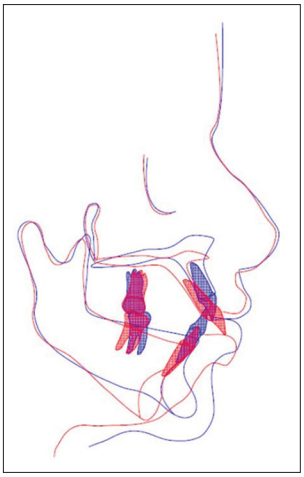

**Figure 18: Pre- and post-treatment superimposition**

corticotomy is acceptable up to 6 mm, but we required more amount of intrusion than what is clinically acceptable.[28]

Young Jin Jeon advocated the use of mini plates and screw assisted correction of the maxillary cant followed by BSSO surgery for correction of mandibular asymmetry. However, this could not be an acceptable treatment alternative as the maxillary canting was too severe to be corrected by intrusion with mini plates (extrusion cannot be advocated as it would cause clockwise rotation of the mandible, producing a long face).[20] Moreover, the treatment time was extended and increased the risk of side effects in using skeletal anchorage for correction of maxillary cant.[29]

For hemifacial microsomia patients, mild skeletal deformities, such as mandibular hypoplasia and facial asymmetry,

**Table 3: Classification based on craniofacial structures involved**

| Congenital factors | Acquired factors | Developmental factors |
|---|---|---|
| 1. Cleft lip and palate | 1. Temporomandibular joint | 1. Unknown causes |
| 2. Tessier clefts | 2. Ankylosis | |
| 3. Hemifacial microsomia | 3. Facial trauma | |
| 4. Neurofibromatosis | 4. Children's radiotherapy | |
| 5. Congenital muscular torticollis | 5. Fibrous dysplasia | |
| 6. Craniosynostoses | 6. Facial tumors | |
| 7. Vascular disorders | 7. Unilateral condylar hyperplasia | |
| 8. Others | 8. Parry-Romberg syndrome | |
| | 9. Others | |

have conventionally been treated with bimaxillary surgery, whereas autogenous costochondral grafting is used for more severe malformations.[30] Hence, we used iliac bone graft.

Good vertical stability can be observed after surgical maxillary impaction, with only 6.5% of patients experiencing 2 mm or more relapse 1 year after surgery[31] with results being stable even after 5 years, thus stating its long-term stability.[32] According to Proffit *et al*., the impaction of the maxilla is among the osteotomies that provide greater stability.[33]

The most widely used osteotomy techniques in surgical correction of mandibular asymmetries are bilateral sagittal split osteotomy and intraoral vertical ramus osteotomy.[34] In this patient, we opted for bilateral sagittal split osteotomy as[35-37] the features characterized in this case did not warrant more invasive procedures such as alloplastic or autogenous reconstructions of the ramus or condyle. Furthermore, the BSSO option is well documented in the literature.[33,38-40]

The patient voiced no complaints regarding the TMJ disorder-related symptoms, such as pain on palpation, popping, clicks, or crepitus in the temporomandibular joints, despite the mandibular asymmetry present pretreatment. After 15 months of treatment, the goals were achieved: improved oral health, dental and facial aesthetics, occlusion, mandibular functions, and proper temporomandibular joint function.

Superimposition of the lateral cephalometric tracings showed the superior positioning of the maxilla followed by counterclockwise rotation of the mandible with the advancement of the genium and an overall improvement in the soft tissue drape. Examination of the extraoral and intraoral photographs confirms that the facial deformity was corrected with improved facial symmetry and a balanced occlusal plane.

## Conclusion

Whatever the dentofacial deformity, there should be a protocol for individualized attention, by listing the important

needs of the patients, to resolve them promptly. For the success of a treatment involving orthodontic and surgical management, an interdisciplinary approach is always a mandate. This case report highlights a patient-centered outcome and a multidisciplinary interaction for rational management of a severe dentofacial deformity.

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### Conflicts of interest

There are no conflicts of interest.

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