Adult patient with hemifacial microsomia treated with combined orthodontics and distraction osteogenesis

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Distraction osteogenesis is an alternative treatment option for patients with facial asymmetry and mandibular hypoplasia. New bone is formed between bone segment surfaces that are gradually separated by incremental traction. The purpose of this article is to report the treatment of a 22-year-old man with hemifacial microsomia, facial asymmetry, and a skeletal Class II profile. The patient’s left mandibular ramus was lengthened with distraction osteogenesis, and miniscrew-assisted rapid palatal expansion was used to correct the maxillary transverse deficiency. Postoperative orthodontic treatment achieved tooth alignment and closure of the posterior open bite. The total treatment period was 27 months. These therapeutic treatments improved the patient’s facial appearance. (Am J Orthod Dentofacial Orthop 2014;145:72-84)

Hemifacial microsomia is the second most frequent form of isolated facial birth disorder, after cleft lip and palate. Hemifacial microsomia is a congenital skeletal malformation in which there is a deficiency on 1 side of the face; characteristic features include maxillomandibular hypoplasia and facial asymmetry. It is primarily a syndrome of the first brachial arch, involving underdevelopment of the ear, mandible, maxilla, zygoma, temporal bone, and the associated musculature and soft tissues.1 The exact etiology of hemifacial microsomia has not yet been determined, but laboratory studies suggest that the clinical features might be caused by early loss of neural crest cell-related formation of the first brachial arch.2,4

In adults with hemifacial microsomia, mild skeletal deformities—such as mandibular hypoplasia and facial asymmetry—have traditionally been treated with bimaxillary surgery,4 whereas autogenous costochondral grafting is used for more severe malformations.5 In 1988, Ilizarov6,7 first described distraction osteogenesis, an alternative treatment process in which new bone is formed between bone segment surfaces that are gradually separated by incremental traction. In 1992, McCarthy et al8 used mandibular lengthening by distraction osteogenesis in patients with hemifacial microsomia. Because of the slow lengthening procedure, the soft tissues adapt gradually; this is advantageous because it minimizes the risk of relapse for hemifacial microsomia patients who lack both bone and soft tissues.9,10

In this case report, we describe the treatment with the combination of distraction osteogenesis and orthodontics, as well as miniscrew-assisted rapid palatal expansion, for a man with a skeletal Class II malocclusion, hemifacial microsomia, and a maxillary transverse deficiency.

DIAGNOSIS AND ETIOLOGY

A 22-year-old man visited the orthodontic department at Yonsei University Dental Hospital in Seoul, Korea, with a chief complaint of facial asymmetry. The clinical and radiographic examination results indicated that he had Pruzansky-Kaban type I left hemifacial microsomia with well-formed but small temporomandibular joint and ramus. His medical history included a
Fig 1. Pretreatment facial and intraoral photographs.

Fig 2. Pretreatment cast models.
right orbital wall fracture 3 years previously and removal of a skin tag on his left ear during childhood. He had no family history of craniofacial deformities and no temporomandibular joint-related symptoms.

The pretreatment facial photographs showed that his face was asymmetrical with a mildly underdeveloped left side, a chin point that was deviated 8.0 mm toward the affected side, and a retrusive chin in profile view, in addition to lip canting and incompetence. There was no significant difference between the levels of his right and left eyes. The intraoral examination showed frontal canting of the occlusal plane and a mandibular midline deviation of 2.0 mm to the left. He had severe Class II canine and molar relationships, with an 11.0-mm overjet, a 3.0-mm overbite, and a transverse deficiency with a narrow V-shaped maxillary arch (Figs 1 and 2).

The lateral cephalometric analysis showed an SNA angle of 82.2°, an SNB angle of 75.0°, and an ANB angle of 7.2°. The mandibular plane angle was 47.6°, and the ramus height was 45.4 mm (Fig 3, Table I). The maxillary incisors were labially inclined at an angle of 120.6° to the SN plane. The upper and lower lips were protrusive with respect to the E-line. The posteroanterior cephalometric analysis showed that the maxillary molars were extruded by 3.1 mm more on the right side compared with the left, and the chin was deviated by 12.0 mm to the left. The panoramic radiograph showed differences in the sizes of the ramus, condyle, and coronoid between the left and right sides as well as impaction of all 4 third molars with complete root formation. Clinically, the patient’s periodontal tissues were healthy. Three-dimensional computed tomography images showed deficiencies of the left condylar and coronoid processes, along with ramus hypoplasia and chin deviation. The angle of the mandible was also underdeveloped (Fig 4).

![Fig 3. Pretreatment radiographs: lateral and posteroanterior cephalograms and panoramic radiograph.](image)
### Table I. Lateral cephalometric measurements

<table>
<thead>
<tr>
<th>Measurement</th>
<th>Norm</th>
<th>Pretreatment (22 y 3 mo)</th>
<th>Postdistraction (22 y 10 mo)</th>
<th>Posttreatment (24 y 6 mo)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Skeletal</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>SNA (°)</td>
<td>82.4 ± 3.2</td>
<td>82.2</td>
<td>82.4</td>
<td>83.6</td>
</tr>
<tr>
<td>SNB (°)</td>
<td>80.4 ± 3.1</td>
<td>75</td>
<td>80.1</td>
<td>79.3</td>
</tr>
<tr>
<td>ANB (°)</td>
<td>2.0 ± 1.7</td>
<td>7.2</td>
<td>2.3</td>
<td>4.3</td>
</tr>
<tr>
<td>SN-GoGn (°)</td>
<td>22.0 ± 5.0</td>
<td>47.6</td>
<td>41.9</td>
<td>42.1</td>
</tr>
<tr>
<td>Gonial angle (°)</td>
<td>117.1 ± 6.7</td>
<td>139.5</td>
<td>138.6</td>
<td>134.9</td>
</tr>
<tr>
<td>Ramus height (mm)</td>
<td>57.6 ± 5.2</td>
<td>45.4</td>
<td>54.5</td>
<td>53</td>
</tr>
<tr>
<td>Go-Me (mm)</td>
<td>79.0 ± 5.0</td>
<td>72.6</td>
<td>76.6</td>
<td>75.9</td>
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<tr>
<td><strong>Dental</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>U1-SN (°)</td>
<td>107.0 ± 5.0</td>
<td>120.6</td>
<td>121.8</td>
<td>113.6</td>
</tr>
<tr>
<td>U1-NA (°/mm)</td>
<td>25.0/7.0</td>
<td>38.3/13</td>
<td>39.4/13</td>
<td>30/8.9</td>
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<td>L1-NB (°/mm)</td>
<td>27.0/6.9</td>
<td>36.6/12.6</td>
<td>34.2/11.9</td>
<td>37.4/12.3</td>
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<tr>
<td>L1-GoGn (°)</td>
<td>95.0 ± 5.0</td>
<td>94</td>
<td>92.2</td>
<td>96</td>
</tr>
<tr>
<td><strong>Soft tissues</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>E-line (mm)</td>
<td>1.0 ± 2.0</td>
<td>6.3</td>
<td>2.1</td>
<td>2</td>
</tr>
<tr>
<td>Upper lip/lower lip</td>
<td>2.0 ± 3.0</td>
<td>9.8</td>
<td>6.1</td>
<td>6.7</td>
</tr>
</tbody>
</table>

![Three-dimensional computed tomography images before distraction osteogenesis.](image-url)
**TREATMENT OBJECTIVES**

Because of the mild Pruzansky-Kaban type I hemifacial microsomia on the left side, the patient expressed concern about his facial asymmetry. Thus, the treatment objectives were established as follows: (1) correct the patient’s facial asymmetry by lengthening the affected ramus, and coordinate the facial, maxillary, and mandibular dental midlines; (2) correct the skeletal Class II anteroposterior jaw relationship; (3) coordinate the widths of the dental arches; (4) correct the canted occlusal plane and achieve dental Class I canine and molar relationships; and (5) relieve the proclined incisor position and achieve an ideal overjet and overbite relationship.

**TREATMENT ALTERNATIVES**

Based on the treatment objectives, the following treatment alternatives were considered: (1) maxillary and mandibular orthognathic surgery with anteroposterior impaction, with or without segmental surgeries; (2) maxillary orthognathic surgery with anteroposterior impaction, with or without segmental surgeries, and distraction osteogenesis to advance the mandibular body and lengthen the shorter ramus; (3) an iliac bone graft to level the canting of the maxillary left side, and distraction osteogenesis to advance the mandibular body and lengthen the shorter ramus; or (4) combined orthodontic treatment, with or without extraction of the 4 first premolars, and distraction osteogenesis.

The patient did not want to undergo orthognathic surgery and wanted minimal surgeries and bone grafting. He did not agree to extraction of the 4 first premolars because he was not concerned about lip protrusion. Therefore, the fourth option without extractions was chosen.

**TREATMENT PROGRESS**

The treatment plan was to correct the mandibular asymmetry and the occlusal plane canting by distraction osteogenesis of the ramus on the affected side, followed by orthodontic treatment to align the teeth and establish a functional occlusion. The patient was hospitalized, and surgery for the distraction osteogenesis was performed under general nasotracheal anesthesia. Through an intraoral incision, a horizontal ramus osteotomy was performed on the left at the level of the occlusal plane. The distractor was placed with a vector parallel to the posterior border of the ramus. After 7 days of latency, the distraction device was activated by 1 mm per day for 24 days, until the mandibular midline deviation was overcorrected by up to one third of the initial discrepancy. The distraction device was maintained for 90 days (Fig 5). In the consolidation phase, a resin occlusal bite-block was placed on each arch because of a surgically created posterior unilateral open bite on the elongated side. At the end of this consolidation phase, the patient had a posterior open bite and a crossbite on both sides (Figs 6-8).

After removal of the distractor, orthodontic treatment with miniscrew-assisted rapid palatal expansion was initiated. Appropriately sized bands were selected and placed on the maxillary first premolars and first molars. A custom hyrax-type rapid palatal expander with an 0.8-mm stainless steel wire hook for placement of miniscrew implants was delivered. Under local anesthesia, 4 miniscrews were placed perpendicular to the palate in the parasagittal area, where sufficient bone width could
Fig 6. Intraoral photographs after the distraction and consolidation phases.

Fig 7. Lateral and posteroanterior cephalograms and panoramic radiograph after the distraction and consolidation phases.
be obtained. After miniscrew placement, light-cured resin was used to connect the miniscrew heads and hooks to form a single unit (Fig 9). The appliance was activated at 0.25 mm per day. After achieving correction of the transverse maxillary deficiency, the appliance was maintained for 3 months without activation. After removal of the appliance, brackets were placed on the maxillary and mandibular arches for tooth alignment and correction of the posterior open bite. During fixed orthodontic treatment, vertical elastics were used to close the posterior open bite (Fig 10).

After 18 months of postdistraction orthodontic treatment, the fixed orthodontic appliances were removed (Figs 11-13). The total treatment period lasted 27 months. Fixed lingual retainers were bonded to the lingual surfaces of the anterior teeth in both arches. Maxillary and mandibular circumferential retainers were delivered with instructions to use them 24 hours per day for the next 6 months.

RESULTS

The posttreatment photographs and radiographs showed that facial symmetry and an ideal occlusion with proper overjet and overbite were achieved. The mandibular dental midline coincided with the facial and maxillary midlines, and the occlusion was finished to Angle Class I canine and molar relationships. The panoramic radiograph showed that new bone with the same characteristics as adjacent bone was created on the affected side by gradual separation of the 2 bony segments that were surgically severed (Figs 11-13).

Fig 8. Superimposition of the 3-dimensional computed tomography images at pretreatment and after the consolidation phase.
Superimposition of 3-dimensional computed tomo-
graphs showed that after distraction osteogenesis and
the consolidation phase, the shorter ramus was length-
ened to 54.5 mm, the mandible was advanced compared
with pretreatment, and the ANB angle decreased to 2.3°
(Fig 8). However, the overall superimposition showed
that some relapse occurred during orthodontic treat-
ment (Fig 14): an ANB angle of 4.3° and ramus height
of 53.0 mm were observed at the end of treatment (Fig 13, Table I). At 1 year after debond, the results were stable, and the patient was satisfied with his facial esthetics (Figs 15 and 16).

DISCUSSION

Hemifacial microsomia manifests in various clinical presentations, for which many classification systems have been developed.11,12 The Pruzansky-Kaban classification describes 3 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).13 The mode of treatment (typically a combined surgical-orthodontic approach) for these patients aims to intercept and correct the asymmetries in both the maxilla and the mandible.14,15 During growth, an orthopedic or a functional appliance can be used to encourage growth and minimize the extent of orthognathic surgery required. In mild and moderate type I or II patients, distraction osteogenesis is postponed until growth has ended because this can prevent a second, later surgical intervention.16 In nongrowing adults, presurgical orthodontic treatment is often followed by mandibular and maxillary surgery; however, the inherent risk of relapse caused by the inability of the muscles to be stretched often compromises the results.17 The use of costochondral grafting to treat more severe deformities leads to important postoperative concerns regarding infection, pain, and donor-site morbidity.5

The process of distraction osteogenesis was developed to lengthen long bones that load mechanical compressive stress from the surrounding soft tissues. Traction forces applied to bone also create tension in the soft tissues, initiating a sequence of adaptive changes termed...
Fig 12. Posttreatment cast models.

Fig 13. Posttreatment radiographs: lateral and posteroanterior cephalograms and panoramic radiograph.
Fig 14. Superimposition of cephalometric tracings.

Fig 15. Facial and intraoral photographs show the treatment results 1 year after debonding.
immediately after distraction in our patient, the length of the mandibular ramus increased and his face appeared more symmetrical (Fig 8). However, as is typical after distraction, our patient had a posterior open bite on the distracted side and a bilateral crossbite because of a pre-existing V-shaped narrow maxillary arch.20

To correct this maxillary transverse deficiency, we used miniscrew-assisted rapid palatal expansion, which is a variant of the normal, tooth-supporting hyrax-type of rapid palatal expander.21,22 The primary objective of using this appliance was to gain maximum skeletal effects at the patent sutures but not necessarily at the obliterated sutures. Four miniscrews ensured expansion of the underlying basal bone and maintained the separated bones during the consolidation period. Because of structural restrictions in the maxilla and the design of the device, we achieved greater dentoalveolar expansion than basal bone expansion; this pattern was considered to be an inevitable consequence in this patient (Fig 8).

Previous studies have not shown stable results after mandibular ramus lengthening by distraction osteogenesis, with relapse often occurring on the elongated side.16,23 In our patient, the skeletal relapse was mainly observed during fixed orthodontic treatment, possibly caused by soft-tissue tension. Furthermore, backward movement of the chin might have been caused by the relapse in mandibular ramus height (Fig 14). The relapse tends to slowly mold the elongated bone back to the original affected structure, even though the increased mandibular volume and projection are maintained. The genotype and musculoskeletal structures must be considered when planning distraction.24,25 Because we overcorrected the affected ramus by lengthening up to one third of the initial discrepancy, the distraction osteogenesis results were reasonably stable at the 1-year follow-up (Figs 15 and 16).
At the end of treatment, the patient’s periodontal condition was not ideal and included labial gingival recession and attachment loss of the mandibular lateral incisors (Fig 11); this might have been a result of age, oral hygiene, orthodontic proclination of the mandibular incisors in a patient with a thin gingival biotype, a narrow zone of keratinized gingiva, or a combination of these factors. The patient chose not to undergo mucogingival surgery after the treatment. To reduce plaque and gingival inflammation, he follows a strict oral hygiene program including brushing, rinsing, and professional cleaning at regular visits. The influence of the width of keratinized gingiva and gingival biotype on the result of orthodontic treatment makes it advisable to perform mucogingival surgery before orthodontic proclination of the mandibular incisors in at-risk patients.

CONCLUSIONS

Our results demonstrate that the combination of distraction osteogenesis and orthodontic treatment is an effective option for an adult with hemifacial microsomia. The therapeutic results showed improvement of the patient’s facial appearance.

REFERENCES