Treatment of skeletal open-bite malocclusion with lymphangioma of the tongue

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Lymphangioma of the tongue causes massive tongue enlargement, leading to difficulties in swallowing and mastication, speech disturbances, airway obstruction, and skeletal deformities such as open-bite malocclusion. Early reduction of tongue volume improved the excessive open bite in a young girl, but it was not sufficient to redirect the original hyperdivergent growth pattern. Orthodontic camouflage treatment was therefore rendered. Long-term evaluation after tongue-reduction surgery and orthodontic treatment is presented. (Am J Orthod Dentofacial Orthop 2012;141:627-40)

An open bite develops from a combination of dental, skeletal, and environmental factors.1,2 Although simple open bites caused by dental factors are relatively easy to correct with favorable outcomes, open bites with skeletal components are usually more difficult to treat with less stable results.2,3 Environmental factors, including aberrant neuromuscular function of the lip or tongue, abnormal tongue posture, and airway obstruction, can also increase the risk of open bite.1,2,4,5 Therefore, proper identification of the etiology and careful diagnosis are important in achieving optimal treatment results. Here, we report the long-term outcome of an open-bite malocclusion with macroglossia due to tongue lymphangioma and hyperdivergent growth treated with serial tongue-reduction surgeries and orthodontic camouflage treatment.

DIAGNOSIS AND ETIOLOGY

A girl, aged 3 years 5 months, came to the Department of Orthodontics, Gangnam Severance Dental Hospital, Yonsei University, in Seoul, Korea, seeking orthodontic treatment. She had been diagnosed with lymphangioma of the tongue. She had a dental history of abnormal tongue posture. She had a convex profile with increased lower facial height and severe anterior open bite (−13 mm). The deciduous second molars were the only teeth that were occluding. Several carious lesions were present in the maxillary arch (Figs 1 and 2). The panoramic radiograph showed a supernumerary tooth between the maxillary central incisors. Lateral cephalometric analysis confirmed a skeletal Class II open bite with increased lower facial height. The posteroanterior cephalometric radiograph showed slight mandibular asymmetry but otherwise was within normal limits (Figs 3 and 4, Table).

The primary cause of the open bite was evidently related to the macroglossia of the tongue due to lymphangioma.6-8 Lingual lymphatic malformation is frequently localized in the anterior two thirds of the tongue and enlarges to a great extent after an episode of upper respiratory tract infection.9 As the tongue swells, it can cause difficulties in swallowing and mastication, speech disturbances, airway obstruction, and deformities of maxillofacial structures.10,11 Surgical removal is the treatment of choice when the lymphangioma interferes with esthetics or function.11,12 Spontaneous regression of anterior open bite after glossectomy has been reported in several patients,8,13 and self-correction has also been described throughout the growth period.14 We believed therefore that a decrease of the anterior open bite could be expected after reduction of the tongue volume in our patient. However, the effects of a glossectomy on the vertical skeletal pattern or changes during facial growth are not yet fully understood.

TREATMENT OBJECTIVES

The main treatment objectives were to regain proper function and to improve the occlusion early as possible.
For these goals, reduction of excess tongue volume was of paramount importance. If the hyperdivergent growth pattern remained, growth modification followed by camouflage or surgical orthodontic treatment would also be necessary to achieve ideal esthetics and function.

**TREATMENT ALTERNATIVES**

To improve perioral function and to correct the open bite caused by the enlarged tongue, surgical excision of the lesion and tongue-reduction surgery were indicated. Spontaneous changes in occlusion after tongue reduction

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**Fig 1.** Initial photographs, age 3 years 5 months.

**Fig 2.** Initial dental casts, age 3 years 5 months.
Fig 3. Cephalometric and panoramic radiographs, age 3 years 5 months.

Fig 4. Cephalometric tracing, age 3 years 5 months.
and continued eruption of the permanent dentition were expected, so careful follow-up of growth was planned before considering any active orthodontic treatment. Surgical extraction of the supernumerary tooth was also advised before the eruption of the maxillary central incisors.

If the hyperdivergent growth pattern remained during the follow-up period, several methods for growth modification could be considered. In patients with long-face syndrome combined with a Class II skeletal growth pattern, a high-pull headgear or a maxillary splint could be used to prevent excessive vertical growth. Another option would be to use functional appliances combined with a high-pull headgear for maximum vertical growth control.

Based on the severity of the vertical skeletal problems and the amount of remaining open bite after growth observation and modification, camouflage treatment with a fixed appliance accompanied by either nonextraction or extraction or surgical correction could be considered to achieve ideal esthetics and function.

After a series of tongue surgeries, the patient and the parents were clearly reluctant to accept additional surgical treatment. Therefore, growth modification with headgear during the growth period was emphasized, followed by orthodontic camouflage treatment.

### TREATMENT PROGRESS AND RESULTS

The patient had several tongue-reduction surgeries before she was 9 years old. Progressive open-bite closure could be seen after the tongue-reduction surgeries; this implied that the macroglossia was a major etiologic factor for the extreme open bite. The supernumerary tooth was also surgically extracted.

By the time the patient was 10 years 4 months old, all permanent teeth except the maxillary and mandibular right canines had erupted. The skeletal age of the patient reached stage 3 of the skeletal maturity index of Fishman at this point. Since a hyperdivergent growth pattern was evident even after the decrease in open bite, orthopedic growth modification with a maxillary splint and high-pull headgear was attempted before the growth peak started at skeletal maturity stage 4. There was dental midline deviation to the right (2 mm) with a lack of canine space on the right side, leaving both maxillary and mandibular canines impacted (Fig 5). Considering the amount of space needed for the canines with correction of the midline and the large overjet (8.0 mm), first premolar extractions were indicated. The maxillary and mandibular right first premolars were extracted first to guide the canines to erupt along with continuous use of the high-pull headgear. However, the patient did not comply well with the appliance, and there was little orthopedic effect, if any. Cephalometric superimposition throughout the period confirmed the persistent hyperdivergent skeletal growth pattern (Fig 6).

At 12 years 1 month of age, the patient was reexamined for the second phase of orthodontic treatment. She had a long face, mentalis muscle strain, right shift of the dental midlines of both arches, and a large overjet of 8.5 mm (Fig 7). According to the dental cast analysis, there were a 1-mm arch-length discrepancy in the maxillary arch and a 6.5-mm discrepancy in the mandibular arch (Fig 8). Cephalometric analysis indicated that the patient was skeletal Class II with a hyperdivergent growth pattern and proclination of the maxillary incisors. The posteroanterior radiograph showed a right shift of the dental midline in both arches. According to the hand-wrist radiograph, the patient was in stage 8 of skeletal maturity, and menarche had already occurred (Figs 9 and 10, Table).

### Table. Cephalometric analysis

<table>
<thead>
<tr>
<th>Age</th>
<th>Initial 3 y 5 mo</th>
<th>Pretreatment 12 y 1 mo</th>
<th>Posttreatment 13 y 10 mo</th>
<th>1-year retention 14 y 10 mo</th>
</tr>
</thead>
<tbody>
<tr>
<td>SNA (°)</td>
<td>84.5</td>
<td>78.0</td>
<td>76.0</td>
<td>77.0</td>
</tr>
<tr>
<td>SNB (°)</td>
<td>74.5</td>
<td>73.5</td>
<td>72.0</td>
<td>73.0</td>
</tr>
<tr>
<td>ANB (°)</td>
<td>10.0</td>
<td>4.5</td>
<td>4.0</td>
<td>4.0</td>
</tr>
<tr>
<td>Wits (mm)</td>
<td>–2.5</td>
<td>3.0</td>
<td>–1.5</td>
<td>–3.0</td>
</tr>
<tr>
<td>Gonial angle (°)</td>
<td>112.0</td>
<td>135.5</td>
<td>135.5</td>
<td>135.0</td>
</tr>
<tr>
<td>SN-MP (°)</td>
<td>55.0</td>
<td>55.5</td>
<td>55.5</td>
<td>54.0</td>
</tr>
<tr>
<td>FMA (°)</td>
<td>45.8</td>
<td>45.3</td>
<td>46.3</td>
<td>46.2</td>
</tr>
<tr>
<td>Posterior facial/anterior facial height</td>
<td>52.3</td>
<td>54.7</td>
<td>55.4</td>
<td>55.3</td>
</tr>
<tr>
<td>Ramus height (mm)</td>
<td>31.9</td>
<td>41.4</td>
<td>45.6</td>
<td>46.4</td>
</tr>
<tr>
<td>Overbite depth indicator</td>
<td>52.8</td>
<td>55.5</td>
<td>58.3</td>
<td>64.5</td>
</tr>
<tr>
<td>U1 to SN (°)</td>
<td>96.0</td>
<td>120.0</td>
<td>98.5</td>
<td>102.0</td>
</tr>
<tr>
<td>IMPA (°)</td>
<td>83.0</td>
<td>66.5</td>
<td>72.0</td>
<td>76.0</td>
</tr>
</tbody>
</table>
Fig 5. Intraoral photographs and panoramic radiograph at age 10 years 4 months. Canines on the right side are impacted.

Fig 6. Cephalometric superimpositions from 3 to 12 years, showing the results of tongue reduction.
The treatment plan for the second phase of orthodontic treatment was established. Additional extractions of the maxillary and mandibular left first premolars were planned for correction of the midline, alignment, and overjet. A high-pull headgear was advised to control the remaining vertical growth. The patient once again did not cooperate with the high-pull headgear, and vertical growth control was compromised. After premolar extraction, fixed appliances were bonded. Both arches were aligned, and space closure and midline correction were continued. After 1 year 9 months of orthodontic treatment, overbite and overjet improved, resulting in Class I molar and canine relationships. Lip closure became much more natural after orthodontic treatment, but the mentalis muscle strain still remained (Figs 11 and 12). Maxillary anterior proclination was corrected, and the dental and facial midlines were coincident. From the hand-wrist radiograph, radius fusion had started, indicating that the growth was almost complete (Figs 13 and 14). According to the superimposition, the open bite had been successfully managed with the camouflage treatment; however, vertical growth control showed limited results because of the lack of patient cooperation (Fig 15).

After treatment, maxillary and mandibular fixed retainers were bonded, and removable circumferential retainers were used full time for the next 6 months. The occlusion remained stable after 1 year of retention without any relapse (Figs 16 and 17). Occlusal force measured by using the prescale system had also increased after treatment and even during the retention period indicating functional improvement (Fig 18). Vertical growth still continued throughout the retention period, but overjet and overbite were maintained (Fig 19).

**DISCUSSION**

Open bite develops by interaction of many etiologic factors, and malfunction of the tongue might be a primary reason for a skeletal open bite. For this patient,
Fig 8. Pretreatment dental casts.

Fig 9. Pretreatment cephalometric, panoramic, and hand-wrist radiographs.
Lymphangioma of the tongue was a major factor that caused the open bite; tongue-reduction surgeries assisted in the progressive open-bite closure. Lymphangioma is a benign tumor of the lymphatic vessels. A majority of cases are congenital, and 95% of the tumors occur before age 10. Lymphangioma of the tongue is a rare condition, but it is reported to be the most common cause of congenital macroglossia. According to our long-term evaluation, the early reduction of the tongue volume was effective in the improvement of the excessive open bite, but it was not sufficient to redirect the original hyperdivergent growth pattern.

Many reports have mentioned difficulty in treating skeletal open bites in association with long-face syndrome, which involves a dolichocephalic facial form, high mandibular plane angle, hyperdivergent skeletal growth pattern, and anterior open bite. Our patient demonstrated the typical traits of a Class II skeletal pattern and long-face syndrome. Facial pattern is reported to develop even before the first molar erupts. Also, patients with an open bite and increased lower facial height are reported to have earlier onset of puberty. This indicates that early orthopedic intervention is necessary for favorable craniofacial growth, and randomized clinical trials have also reported the effectiveness of treatment in the mixed dentition. Therefore, attempts were made to redirect the growth pattern before and during puberty. However, it was unfortunate that we could not determine the effect of early growth modification treatment using high-pull headgear, because of the lack of patient compliance.

The mandibular growth pattern should also be a consideration for favorable treatment results. The condyles of open-bite subjects are reported to grow posteriorly, leading to clockwise rotation of the mandible. Therefore, the second phase of orthodontic treatment involving extractions should be postponed until after puberty to prevent unnecessary extrusion of the posterior teeth and further bite opening. In this patient, orthodontic treatment involving fixed appliances and premolar extractions was postponed until after she had menarche to prevent further clockwise rotation of the mandible. If the patient had cooperated with the high-pull headgear, treatment results could have been more favorable.

Skeletal open bite with excessive vertical growth can be a challenge to the practitioner and patient alike, since long-term stability is a major concern regardless of the treatment modality. The final occlusion was well maintained regardless of the remaining vertical growth in this patient. Improvement in function indicated by the gradual increase in occlusal force could be taken as a positive sign of stability. However, considering the patient’s skeletal age, minor growth might still remain, and it requires careful follow-up.
Fig 11. Posttreatment photographs, age 13 years 10 months.

Fig 12. Posttreatment dental casts.
**Fig 13.** Posttreatment cephalometric, panoramic, and hand-wrist radiographs.

**Fig 14.** Posttreatment cephalometric tracing.
Fig 15. Cephalometric superimposition before and after phase 2 treatment.

Fig 16. One-year retention photographs, age 14 years 10 months.
Fig 17. One-year retention dental casts.

Fig 18. One-year retention lateral and panoramic radiographs and changes in occlusal force.
CONCLUSIONS

Macroglossia caused by lymphangioma can be a major cause of an open-bite malocclusion. Thorough evaluation of the etiology and its correction, proper diagnosis, and timely intervention produced a favorable functional outcome.

REFERENCES


