Case Report

Severe Dental Open Bite Malocclusion With Tongue Reduction After Orthodontic Treatment

Hitoshi Hotokezaka, DDS, DDSca; Takemitsu Matsuo, DDS, DDScb; Maki Nakagawa, DDSc; Akio Mizuno, DDS, DDScd; Kazuhide Kobayashi, DDS, DDSce

Abstract: We treated a 21-year-old woman with a severe open bite and macroglossia with a standard edgewise appliance and without partial glossectomy. This was followed by retention using a Begg-type plate retainer for the upper dental arch and a fixed canine-to-canine for the lower arch. A crib was added to the upper plate retainer for suppression of a tongue thrust. The lower arch relapsed during the retention period, with a widening of the intermolar distance, flaring of the anterior teeth, and increased mobility of the teeth. We chose tongue reduction to resolve these problems and one-third of the middle dorsal part of the tongue was excised. After the tongue reduction, the patient experienced no functional problem in mastication, swallowing, and gustation, but she complained of mild speech difficulty and slight pain on the dorsal portion of her tongue. These symptoms disappeared 6 months after surgery. At this time, the mandibular dental arch was markedly improved. The flared lower dental arch had returned to an upright position and the tooth mobility reduced to normal. No appliance was used after surgery. Most of the recovery changes occurred within 4 months. This case highlights the importance of the teeth tending to move toward a balance between the tongue pressure from the inside and labio-buccal pressure from the outside. (*Angle Orthod* 2001;71:228–236.)

Key Words: Glossectomy; Dental arch contraction; Relapse; Equilibrium

INTRODUCTION

There is much controversy among orthodontists about the prognosis of treatments for open bite. The causation is difficult to identify since a wide range of etiological factors exists. These factors include skeletal or dental causes, congenital or acquired causes, and combinations of these forms. From the morphological point of view, open bite is basically classified as skeletal or dental. In the case of dental open bite, the etiology is roughly subclassified as acquired, caused by a habit such as thumb sucking or tongue thrusting, and congenital, caused by macroglossia, ankylog-

- ^a Instructor, Department of Orthodontics, Nagasaki University School of Dentistry, Nagasaki, Japan.
- ^b Assistant Professor, Department of Oral and Maxillofacial Surgery I, Nagasaki University School of Dentistry, Nagasaki, Japan.
- ^c Resident, Department of Orthodontics, Nagasaki University School of Dentistry, Nagasaki, Japan.
- d Professor, Department of Oral and Maxillofacial Surgery I, Nagasaki University School of Dentistry, Nagasaki, Japan.
- ^e Professor, Department of Orthodontics, Nagasaki University School of Dentistry, Nagasaki, Japan.

Corresponding author: Dr Hitoshi Hotokezaka, Department of Orthodontics, Nagasaki University School of Dentistry, Sakamoto 1-7-1, Nagasaki 852-8588, Japan

(e-mail: hotoke@net.nagasaki-u.ac.jp).

Accepted: December 2000. Submitted: September 2000. © 2001 by The EH Angle Education and Research Foundation, Inc.

lossia, etc. There also are indistinguishable dental open bite subclasses caused by airway obstruction or neurological problems.

Although it may be relatively easy to treat the causative factor of open bite directly when the factor is clearly identified, it is rather difficult to do so in daily practice because the etiology is often obscure and combined. Although the etiology may bring a consideration of tongue reduction when the tongue volume is excessive, the diagnosis is not always easy. This is because the diagnosis of macroglossia is not fully established and also because the tongue sometimes adapts to the contracted narrower space after orthodontic treatment.

Here we report a patient with dental open bite that we first treated orthodontically without a surgical operation, which invited a relapse during the retention period. Following tongue reduction, the flared lower dental arch recovered to an upright position.

CASE REPORT

Pretreatment evaluation

A 21-year-old woman with no history of a congenital abnormality such as Down Syndrome, Beckwith-Wiedemann Syndrome, or hypothyroidism showed an excessive open bite with an Angle Class III malocclusion (-4.5-mm



FIGURE 1. Intraoral photographs. (A) Pretreatment. (B) One year after tongue reduction.

overbite and -2-mm overjet) (Figure 1A). Although her tongue was rather large, she showed no difficulty in the speech production of the words team or sank. Patients with macroglossia or mandibular protrusion are prone to pronounce these words as theme or thank.

The lateral cephalometric analysis showed the upper central incisor angulation near average (U1-SN = 103.6° , mean = $104.5 \pm 5.6^{\circ}$) and the lower central incisor inclined labially (L1-mandibular plane = 101.0° , mean = $96.3 \pm 5.8^{\circ}$). A mild Angle class III skeletal discrepancy between the upper and lower jaws was observed (ANB = 1.5° ; mean = $3.4 \pm 1.8^{\circ}$), with a mandibular plane angle of 28.6° (mean = 27.8 ± 5.2), and the lower lip slightly protruded (2.5 mm) to the E-line (Figure 2A). The upper incisors were a little small, with an anterior ratio of 0.87 (ideally 0.78), lacking 2.3 mm in width. Our diagnosis was a dental open bite caused by macroglossia.

Initial treatment objectives and plan

In order to correct the open bite and to establish a Class I canine relationship, the lower left first molar was extracted, followed by placement of edgewise appliances and Class III mechanics. Prosthetic treatment was planned after orthodontic treatment to improve the tooth size ratio.

To stabilize the altered dental arch after orthodontic treatment, a plate-type retainer with a tongue crib was worn and myofunctional therapy was performed. In the case of instability after orthodontic treatment, the treatment plan was to be reevaluated to determine whether tongue reduction would be necessary.

Treatment progress

Because the patient presented no signs of a functional problem caused by the large volume of tongue, and also because the tongue was expected to adapt to the postorthodontic dental arch, the orthodontic treatment was initially carried out only with a $0.018'' \times 0.025''$ edgewise appliance without tongue reduction.

Because the lower right first molar was missing and the lower left first molar was suffering from a carious lesion, the lower left first molar was extracted and the first molar spaces were utilized to retract the anterior teeth. The open bite was corrected using short bilateral Class III elastics between the distal portion of the upper canine and the mesial portion of the lower canine at 30–40 g on each side for 18 months. After the appliance was removed, removable retainers were worn (Figures 3A, 4A and 5B).

The upper incisors, upper and lower premolars, and lower left first molar were restored by prosthodontic treatment. Although the long axes of the crowns were not changed by the prosthetic treatment, the widths of the crowns of the upper incisors and upper left second premolar were made wider for correction of the tooth size ratio and the crowns of the lateral incisors were slightly corrected vertically in order to align the upper anterior teeth. Extracted lower first molar spaces could not be closed completely at the end of orthodontic treatment, and the lower left second premolar and first molar were joined just before tongue reduction. After orthodontic treatment, the patient learned tongue positions at resting and swallowing.

Since the patient complained about vomiting caused by a lower plate-type appliance, the lower retainer was re-

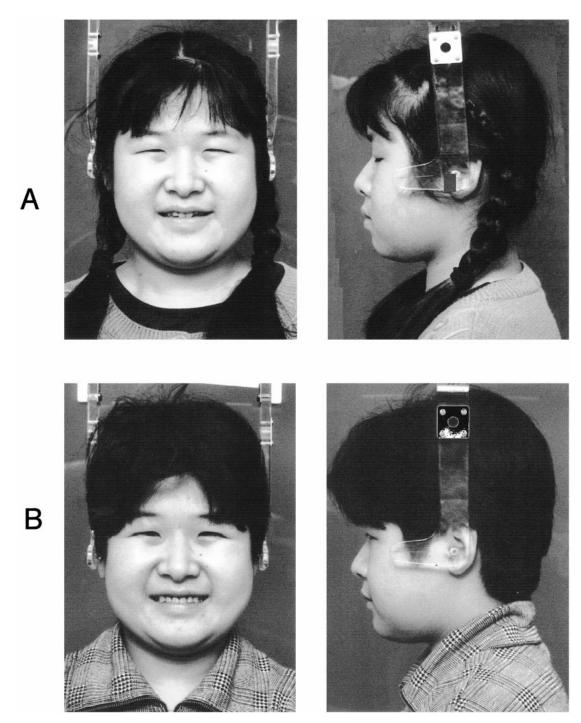


FIGURE 2. Facial photographs. (A) Pretreatment. (B) One year after tongue reduction.

placed with a fixed canine-to-canine retainer after 1 month. A crib was added to the upper retainer to suppress the tongue thrust (another plate without a crib was used during the day). The lower dental arch flared during the retention period (Figures 3B, 4B and 5C) and the closed lower first molar extraction spaces opened. The premolar to molar regions resulted in a cusp-to-cusp occlusion after 1 year of retention. At this time, the teeth mobility (M2-M3) was

greater than that (M1) after orthodontic treatment (Figure 6).

Wolford and Cottrell¹ described 18 clinical features of macroglossia in order to determine whether a tongue reduction is necessary. Eight of these features were observed in this case: (1) a wide, broad, and flat tongue; (2) open bite; (3) mandibular prognathism; (4) class III malocclusion; (5) chronic posturing of the tongue between the teeth

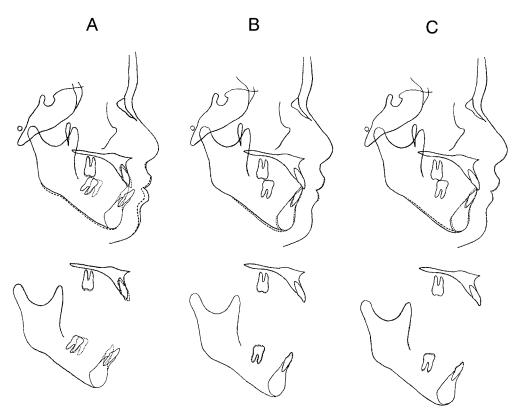


FIGURE 3. Superimpositions of lateral cephalograms. (A) Pretreatment (solid line, May 1990) to immediately after orthodontic treatment (dotted line, March 1995). (B) Immediately after orthodontic treatment (solid line) to before tongue reduction (dotted line, December 1995). (C) Before tongue reduction (solid line) to 1 year after tongue reduction (dotted line, December 1996). The superimpositions were performed with Nasion and S-N plane, ANS and palatal plane, and Menton and mandibular plane, respectively.

at the rest; (6) buccal tipping of posterior teeth; (7) increased transverse width of dental arch; (8) instability in orthodontic treatment. In this case (3) and (4) were mild, and (2) and (4) were improved after orthodontic treatment, but (6), (7), and (8) were still significant after orthodontic treatment. At that point, we rediagnosed the problem and recommended tongue reduction.

The middle dorsal part of the tongue was resected by the method of Harada and Enomoto (Figure 7 and 8). The total weight of the excised tissue was 11 g, and the inflammatory swelling disappeared by 1 month after surgery. The upper plate retainer was not used after surgery. The mobility of the lower anterior teeth and molars increased transiently 2 months after tongue reduction, which is assumed to be caused by a postsurgical inflammatory swelling of the tongue. The mobility of these teeth decreased gradually thereafter (Figure 6, upper). The contraction of the lower dental arch and the reduced mobility of teeth were observed gradually and simultaneously in the first 6 months after surgery (Figure 6, lower). Thereafter, the dental arch and the tooth mobility gradually became stable. The upper and lower molar cusp-to-cusp relationship improved to an upright position (Figure 4C and 5C,D), and the tooth mobility was reduced to M0-M1. The most prominent change of tooth mobility was that of the

lower second molars, which was M3 before tongue reduction and M0 after tongue reduction (Figure 6).

Though the total Class I relationship could not be fully attained because of the smaller width of upper posterior teeth and the opening of space at the lower first molar extraction site, acceptable overjet and overbite were obtained (Figure 9 and 10).

DISCUSSION

In this case, excessive open bite caused by macroglossia was initially corrected orthodontically without tongue reduction. The first attempt, consisting mainly of lower anterior retraction and transverse contraction, resulted in a collapse of the dental arch during the first treatment period. Following tongue reduction, most of those problems recovered remarkably without any further orthodontic treatment. How did these changes occur? Teeth that are moved orthodontically have a potential to return to their original position. This potential is assumed to result mainly from soft tissues, including the dentoalveolar ligaments, gingival fibers, and buccolabial and tongue muscles. While the tongue in this case is believed to have played a major role in the relapse after orthodontic treatment and also in the recovery after tongue reduction, dentoalveolar and gingival tissues

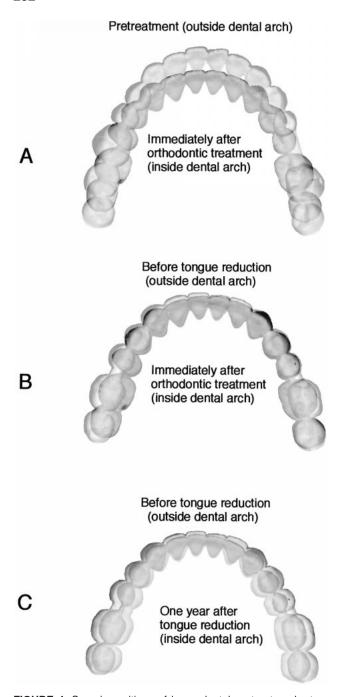


FIGURE 4. Superimpositions of lower dental casts at each stage. The pictures of the dental casts were taken at 50 cm vertically to occlusal plane and above from the center of the right and left first premolars. Superimposition was performed by referring transversally to the PA cephalogram and sagittally to the lateral cephalograms with Me and Go as reference points. (A) Pretreatment (May 1990, outside dental arch) to immediately after orthodontic treatment (March 1995, inside dental arch). (B) Immediately after orthodontic treatment (March 1995, inside dental arch) to before tongue reduction (December 1995, outside dental arch). (C) Before tongue reduction (outside dental arch) to 1 year after tongue reduction (December 1996, inside dental arch).

also may have played some role, as is seen in the distal movement of lower second molars after orthodontic treatment (Figure 3B).

Teeth are subjected to a variety of forces from mastication and the actions of the lips, cheeks, and tongue. Whether intermittent or continuous, these forces are large enough to produce tooth movement.^{2,3} Because these forces are usually in equilibrium, the tooth position is usually stable. When we treat Japanese patients, we often encounter patients with Class III skeletal pattern, anterior open bite, and bimaxillary dentoalveolar protrusion caused by excessive tongue volume as well as by a habit of tongue thrusting.⁴ In these cases, myofunctional therapy for the large tongue is not enough for the retention after orthodontic treatment, and it is difficult to expect a balanced tooth position after the tongue position is changed by the treatment.

In this patient, the lower dental arch width showed significant relapse during the retention period whereas the lower dental arch length was not significantly changed. The crib of the upper retainer may have contributed to the stability of lower anterior teeth and had an effect on the retension of the lower dental arch length. These actions could result from the tongue crib taking away anterior space in the oral cavity from the tongue, which may have compressed the tongue posteriorly with an increment of lateral pressure at the molar region as the result. Other retention methods such as a lingual arch should have been used for the retention of lower intermolar width.

Partial glossectomy is sometimes performed as part of the total treatment for a Class III skeletal problem, anterior open bite, and bimaxillary dentoalveolar protrusion. These cases have a rather large tongue size for the space available, resulting in speech problems, tooth flaring, and abnormal skeletal growth. Many methods of tongue reduction have been reported and all are still controversial. Mixter et al5 and Harada and Enomoto6 reported that standard tongue reduction may result in an ankylosed, globular tongue with an insensitive tip. In our patient, we used the method of Harada and Enomoto, which was reported to resolve these problems. After surgery, the patient complained of minor functional problems in speech, mastication, swallowing, and gustation. However, she did experience a mild paralysis and pain on the dorsal portion of her tongue. The postoperative pain caused her difficulty in pronouncing the word get, which was pronounced as jet. These problems were completely resolved within 6 months postoperatively.

In the case of patients with a sagittal split mandibular set back, the tongue pressure would not usually change because the tongue and its origins connecting to the supporting bone also move back and the tongue adapts to an altered position.⁷ On the other hand, in the case of patients treated with

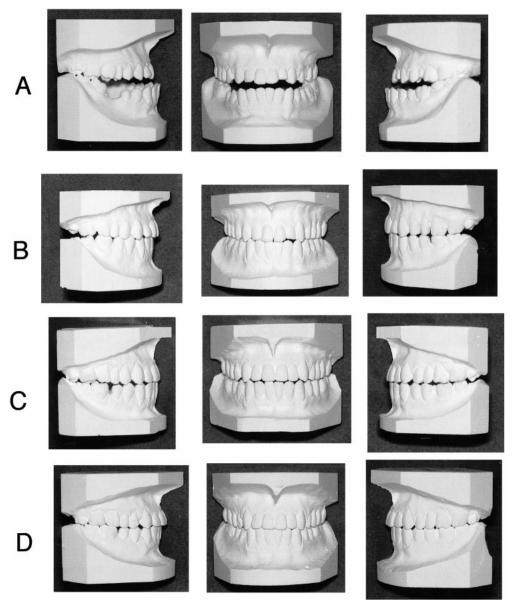


FIGURE 5. Dental casts. (A) Pretreatment (May 1990). (B) Immediately after orthodontic treatment (March 1995). (C) Before tongue reduction (December 1995). (D) One year after tongue reduction (December 1996).

orthodontic tooth retraction without a mandibular set back, the origins of the tongue muscles are not changed. This may cause the tongue difficulty in adapting to the altered position, as in this case.

The success of orthodontic treatment procedures combined with tongue reduction varies from case to case in the literature. The youngest patient to be treated was the reported treatment of a 3-month-old infant with Beckwith-Wiedemann syndrome.⁸ Tongue reduction at a young age is often required in patients with craniofacial abnormalities such as Down Syndrome,^{8,9} Beckwith-Wiedemann Syndrome,^{8,10-13} and lymphangioma.¹⁴ These disorders can lead

to respiratory or speech problems due to excessive tongue volume. From the etiological point of view, the causative factors should be treated first. Therefore, macroglossia should ideally be treated with partial resection early if it causes a dental arch deformity such as open bite. Although the clinical features of macroglossia requiring tongue reduction have been reported, most of those features are not digitally measurable yet and the objective diagnosis has not yet been fully established. At present, comprehensive diagnosis and treatment planning at an early age, including the timing of tongue reduction and the volume of the excision, is not always possible.

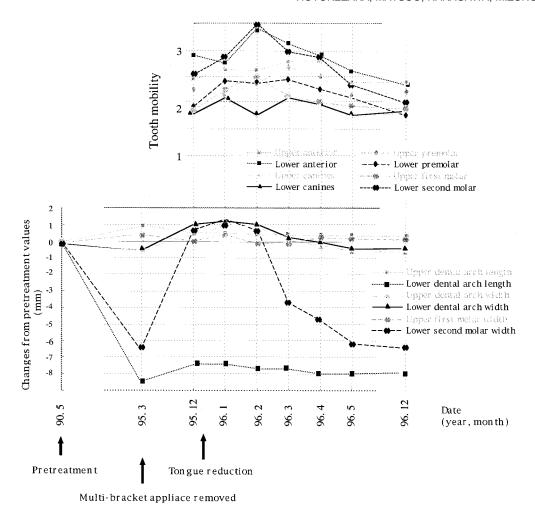


FIGURE 6. Alteration of tooth mobility and dental arch measurements. The mobility of each tooth was measured 3 times by tapping from the labial or buccal center of the tooth crown perpendicularly to the dental arch using Tooth Mobility Checker (Yoshida Co Ltd, Tokyo, Japan). The average of the mobility scores in each tooth was plotted. Indexes: 1.5–2.3 approximately corresponds to M0 of clinical evaluation, 2.3–2.8 to M1, 2.8–3.5 to M2, and >3.5 to M3. The transverse width between the upper first molars and one lower second molar width indicate the intermesio buccal cusps width. The subtracted amount (mm) from the pretreatment value was plotted. The lower interdental arch length was measured from the mesial edge of the lower second molars to the lower central incisal edge. Because the lower left second premolar and second molar were joined by prosthodontic treatment before tongue reduction, only the right second premolar and second molar were measured thereafter.

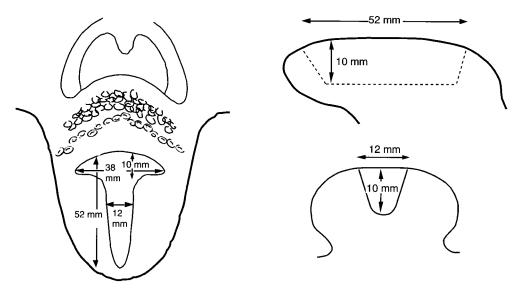


FIGURE 7. Excised region of the tongue. Left, overview of Harada and Enomoto-method; right upper, sagittal view; right lower, transverse view.

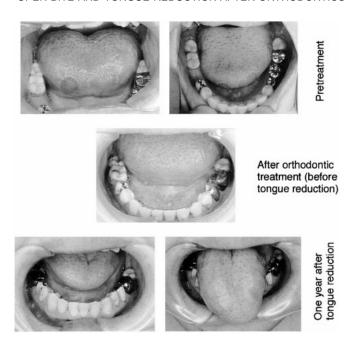


FIGURE 8. Comparison of the tongue at each stage of treatment. Upper, pretreatment; middle, before tongue reduction (December 1995); bottom, 1 year after tongue reduction.





FIGURE 9. Panoramic images of pretreatment (upper, May 1990) and 19 months after tongue reduction (lower, July 1997). Mild to moderate resorption of root and alveolar bone ridge around anterior teeth to premolars are observed.

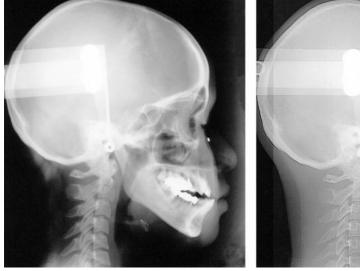




FIGURE 10. Lateral x-ray cephalograms of pretreatment (left, May 1990) and 1 year after tongue reduction (right, December 1996).

ACKNOWLEDGMENT

This work was partly supported by grants-in-aid (09771845) from the Ministry of Education, Science, Sports, and Culture of Japan.

REFERENCES

- Wolford LM, Cottrell DA. Diagnosis of macroglossia and indications for reduction glossectomy. Am J Orthod Dentofac Orthop. 1996;110:170–177.
- Proffit WR. Equilibrium theory revisited: factors influencing position of the teeth. Angle Orthod. 1978;48:175–186.
- Proffit WR. The etiology of orthodontic problem. In: Proffit WR, ed. Contemporary Orthodontics. St Louis, Mo: CV Mosby; 1986: 105–115
- Deguchi T. Case report: three typical cases of glossectomy. Angle Orthod. 1993;63:199–207.
- Mixter RC, Ewanowski SJ, Carson LV. Central tongue reduction for macroglossia. *Plast Reconstr Surg.* 1993;91:1159–1162.
- Harada K, Enomoto S. A new method of tongue reduction for macroglossia. J Oral Maxillofac Surg. 1995;53:91–92.
- Wickwire NA, White RJ, Proffit WR. The effect of mandibular osteotomy on tongue position. J Oral Surg. 1972;30:184–190.

- Kveim M, Fisher JC, Jones KL, Gruer B. Early tongue resection for Beckwith-Wiedemann macroglossia. *Ann Plast Surg.* 1985;14: 142–144.
- Margar BF, Witzel MA, Munro IR. Speech intelligibility after partial glossectomy in children with Down's syndrome. *Plast Re*constr Surg. 1987;79:44–49.
- Menard RM, Delaire J, Schendel SA. Treatment of the craniofacial complications of Beckwith-Wiedemann syndrome. *Plast Re*constr Surg. 1995;96:27–33.
- Sirinelli D, Silberman B, Baudon JJ, Sinnassamy P, Gruner M, Montagne JP. Beckwith-Wiedemann syndrome and neural crest tumors. A report of two cases. *Pediatr Radiol*. 1989;19:242– 245.
- Friede H, Figueroa AA. The Beckwith-Wiedemann syndrome: a longitudinal study of the macroglossia and dentofacial complex. J Cran Genet Dev Biol. 1985;1:179–187.
- Sokoloski PM, Ogle RG, Waite DE. Surgical correction of macroglossia in Beckwith-Wiedemann syndrome. *J Oral Surg.* 1978; 36:212–215.
- Kanai M, Horimoto Y, Yoshioka H, Fujino S, Takano T. Perioperative management for partial resection of a lymphangioma of the tongue. *Jpn J Anesth.* 1996;45:869–872.