The role of tongue reduction

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Macroglossia may occur as a congenital or acquired condition. The enlarged tongue has both functional and cosmetic deformity, which may affect the oral airway, speech, and the development of the jaws. We discuss the various tongue-reduction procedures and present cases to illustrate the uses of this procedure, its potential complications, and the results. (Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2003;95:269-73)

Large tongue, or macroglossia, is an uncommon congenital condition caused by vascular malformation, congenital syndromes such as Beckwith-Wiedemann, and muscular hypertrophy.1-13 It is also found as a rare acquired state in conditions such as amyloidosis, neurofibromatosis, acromegaly, hypothyroidism, and chronic edema.1,5,14-18 Surgical reduction of the tongue is only occasionally required, but it may be indicated in cases of congenital macroglossia,1-5,8 Beckwith-Wiedemann syndrome,9-13 and before or after orthodontic treatment or orthognathic surgery to prevent relapse caused by an enlarged or dysfunctional tongue,19,20 the macroglossia of amyloid,14-16 or chronic edema. We describe the surgical techniques, complications, and outcomes.

Many surgical incisions have been proposed to accomplish tongue reduction (Fig 1). Peripheral excisions such as those described by Butlin5 and Ensign15 (Fig 1, A) or by Harris21 and Dingman and Grabb7 (Fig 1, D) leave the tongue globular and immobile. A V-shaped wedge taken from the tip of the tongue, as reported by Harris,22 Blair,23 and Hendrick3 (Fig 1, B), will shorten but not narrow it, whereas an ellipse taken from the midline of the tongue will narrow it but not shorten it (Fig 1, C).24 Pichler and Trauner25 proposed a combination of an ellipse from the midline posteriorly combined with a wedge from the tip, and various other incisions have been recorded,10,26-31 but the so-called keyhole excision allows both narrowing and shortening of the tongue as required (Fig 1, L).32 Care should be taken to avoid the lingual arteries, but bleeding may be brisk despite this. Even if both lingual arteries are severed, the tongue will not normally necrose because of the rich collateral blood supply and anastomoses. Postoperative edema is often quite substantial, and sutures should not be too small or too tight because the swelling may cause them to cut through. This particular technique is illustrated by the 3 following cases.

CASE REPORTS

Case 1

A 32-year-old man presented with a 3-year history of massive lymphedema of the tongue after acute respiratory distress syndrome and renal failure secondary to necrotic pancreatitis. The macroglossia and associated right-sided 12th nerve palsy (caused by compression injury from the edema) resulted in a speech impediment and marked proclination of the anterior teeth, in addition to potential airway embarrassment (Fig 2). A tongue-reduction procedure was requested before the commencement of orthodontic treatment. A large keyhole-type excision resulted in a great improvement in tongue size and allowed orthodontic treatment to proceed. Despite the good result from initial surgery (Fig 3) and orthodontic correction of the maxillary teeth, orthognathic surgery was required to close the residual anterior open bite. A further tongue-reduction procedure was required; again the keyhole excision was used. This was followed by a Le Fort I osteotomy to correct the residual anterior open bite without relapse (Fig 4).
Case 2

Beckwith-Wiedemann syndrome (BWS) is characterized by macroglossia, umbilical hernia, exomphalos, nephromegaly, and Leydig cell hyperplasia. The prevalence of BWS has been estimated to be 0.07 per 1000 births. However, it is felt that this number is underestimated because of the early death of children who have yet to manifest the signs of BWS or because less-affected patients may not have been classified. Patients with BWS come to the attention of the oral and maxillofacial surgeon for the treatment of upper airway obstruction secondary to macroglossia. There are several temporary treatment options for airway management of these children, such as the use of oral splints or laryngeal mask airways. The definitive treatment usually consists of partial glossectomy with or without tracheostomy. Respiratory distress in these patients may be complicated by concurrent abdominal masses such as hepatoblastoma or insulinoma that can result in decreased total lung capacity and tidal volume.

A 14-week-old male infant (34-week gestational age) whose amniocentesis had shown a normal karyotype was diagnosed with BWS after transfer from a Northern California hospital. Before transfer he was dehydrated and not thriving, and he had persistent hypoglycemia. On admission he was found to have macroglossia (Fig 5) and increased abdominal girth secondary to hemorrhage from a hepatoblastoma.

Five days after admission, the infant had increased respiratory distress requiring surgical intervention. He underwent...
a tracheostomy and a keyhole tongue reduction. Despite the friable nature of the tissue, which made approximation difficult, the tongue was closed with several layers. After surgery, the patient was sedated. Perioperative and postoperative methylprednisolone was given to reduce surgical edema. The tongue was well healed at 6 weeks, and the patient was able to keep his tongue in his oral cavity (Fig 6).

**Case 3**

An 18-year-old girl with developmental delay, facial dysmorphism, and polycystic ovarian disease was referred for craniofacial assessment of an anterior open bite and generalized anterior spacing. A tongue guard had been placed 3 months before presentation to alter tongue function; although this allowed closure of the anterior open bite, a bilateral posterior open bite resulted. It was felt that the tongue posture and size had caused this change. After the removal of the tongue guard, the posterior open bite closed, but the anterior open bite recurred. To secure a stable orthodontic environment, a keyhole-type reduction glossectomy was performed with the patient under general anesthesia (Figs 7-9), which allowed her orthodontic treatment to proceed without relapse.

**DISCUSSION**

Macroglossia was first described by Galen in the second century and is illustrated in medieval carvings. A number of cases are also recorded through the 16th and 17th centuries. The first report of congenital macroglossia secondary to a lymphatic hamartoma was in 1854. Surgical treatment of macroglossia caused by mercury poisoning was reported as early as 1658, and Bar-
Tholin operated on a patient with macroglossia in 1680. Early surgical techniques (illustrated in Fig 1, A-D) were aimed at the reduction of tongue bulk. Those procedures described more recently were aimed at the preservation of tongue function, along with the reduction of tongue mass. The incisions shown in Fig 1, I-K, were designed to preserve the tip of the tongue. The keyhole procedure allows for greater reduction of the tongue mass, and the loss of the tip is not reported to result in a marked sensory deficit, which is confirmed in our experience. Other procedures have been proposed: Davalbhakta and Lambert described a modification of the Köle incision (Fig 1, F) with horizontal wedges taken from the muscle laterally to ensure the reduction of tongue thickness, and Austerman and Machtens described 2 methods for the reduction of asymmetric tongue enlargement (Fig 1, G and H). With central tongue-reduction procedures, such as the keyhole excision, the resection must extend to just anterior of the circumvallate papillae for adequate reduction, but particular care must be taken to avoid the neurovascular bundles, which run inferolaterally.

Taste and tongue mobility are rarely affected by tongue reduction. Speech can be affected, but this complication usually corrects itself within a matter of weeks and formal speech therapy is rarely needed. Speech improvement has been reported as a result of tongue reduction. Sounds made primarily by the tip of the tongue against the upper or lower teeth or the anterior palate are affected.
The sensitivity of the tongue tip is also sometimes altered, but, again, this usually resolves spontaneously within a few weeks. Revision surgery may be required if wound breakdown and subsequent secondary healing caused by postoperative edema cause either a bifid tongue tip or clefting of the tongue.

The indications for tongue reduction are relatively few. In connection with orthodontics and orthognathic surgery, most authorities recommend proceeding with the treatment and carry out a tongue reduction after treatment only if the tongue appears to be instrumental in causing relapse. In these cases, the tongue reduction is carried out at the first sign of relapse but is actually required relatively infrequently. However, in our first patient, the tongue enlargement was so extreme that orthodontics could not be commenced until the tongue was reduced. Several articles illustrate the spontaneous correction of anterior open bite after tongue reduction. In BWS, tongue reduction can allow decannulation and more normal deglutition and carry out a tongue reduction after treatment only if the tongue appears to be instrumental in causing relapse but is actually required relatively infrequently. However, in our first patient, the tongue enlargement was so extreme that orthodontics could not be commenced until the tongue was reduced. Several articles illustrate the spontaneous correction of anterior open bite after tongue reduction.

CONCLUSION

Tongue reduction is an uncommon procedure that may be required for functional and esthetic reasons. The aim of this surgery is to produce a tongue that lies within the lower dental arch and yet can protrude to wet the lips. In our experience, the keyhole excision provides a flexible template to narrow or shorten the tongue as required.

REFERENCES


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