

## Chapter 22 – Tongue Reduction Procedures

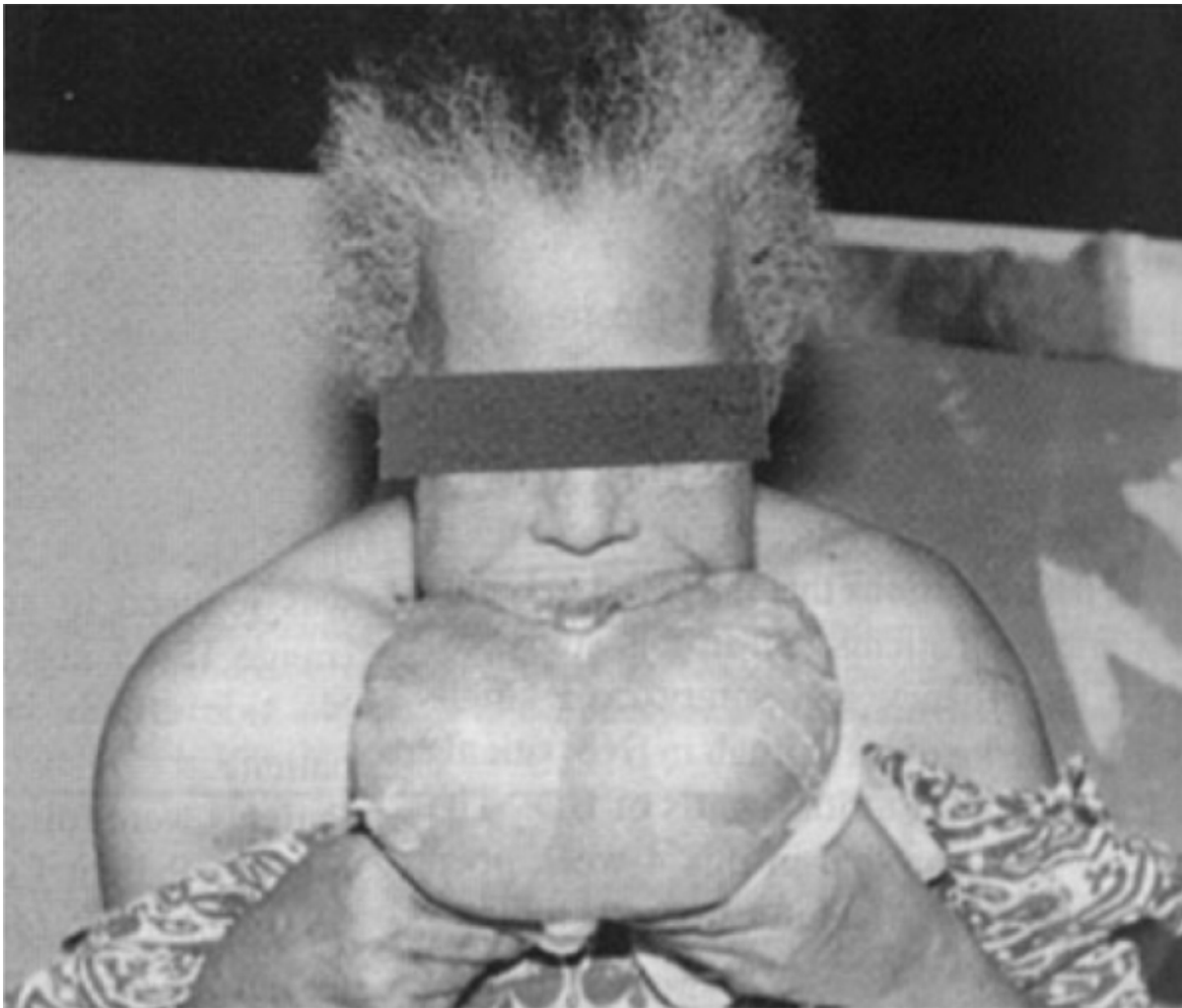
### Eugene N. Myers

Macroglossia was first described by Galen in the second century.<sup>[1]</sup> Other cases were recorded during the 16th and 17th centuries. Surgical treatment of macroglossia was first reported in 1658.<sup>[2]</sup> Macroglossia may be classified as true macroglossia when the tongue is enlarged or as relative macroglossia when there is insufficient space in the oral cavity. Tongue reduction is an uncommon procedure that may be required in such patients for functional or aesthetic reasons. Ring<sup>[2]</sup> presented a fascinating overview of the treatment of macroglossia before the 20th century that should be read by all surgeons interested in the management of patients with this condition.

The most common conditions giving rise to macroglossia are lymphangioma, neurofibroma, amyloidosis, hemangioma, Beckwith-Wiedemann syndrome, mucopolysaccharidosis, hypothyroidism, cretinism, rhabdomyosarcoma, gigantism, acromegaly, muscular dystrophy, Down syndrome, lipid protein triploid syndrome, and idiopathic muscular hypertrophy.<sup>[3]</sup> Neurofibroma, in a localized manner, may give rise to problems with articulation and may promote malocclusion. The tongue may be the only site of neurofibroma or may be part of neurofibromatosis. Lymphangiomatous macroglossia may interfere with articulation and deglutition. These lymphatic malformations increase in size with upper respiratory tract infections, and the enlarged tongue may cause partial airway obstruction.<sup>[4]</sup> Patients with hemangioma or a mixture of hemangiomatous and lymphangiomatous macroglossia may also have problems with intermittent bleeding.

### **PATIENT SELECTION**

Patients with symptoms of difficulty articulating or swallowing, malocclusion, and bleeding and those in whom physical examination confirms the presence of generalized or localized enlargement of the tongue or hemangioma should be considered for this procedure. These patients may have extreme macroglossia that precludes closing of the mouth. If biopsy reveals amyloidosis and its associated signs and symptoms, the diagnosis should be systemic amyloid. An exuberant form of amyloidosis has been reported by Moura and coauthors (Fig. 22-1).<sup>[5]</sup>



**Figure 22-1** Patient suffering from exuberant macroglossia.

(Reprinted with permission from Moura CG, Moura TGG, Durães AR, Souza SP: Exuberant macroglossia in a patient with primary systemic amyloidosis. *Clin Exp Rheumatol* 23:428, 2005.)

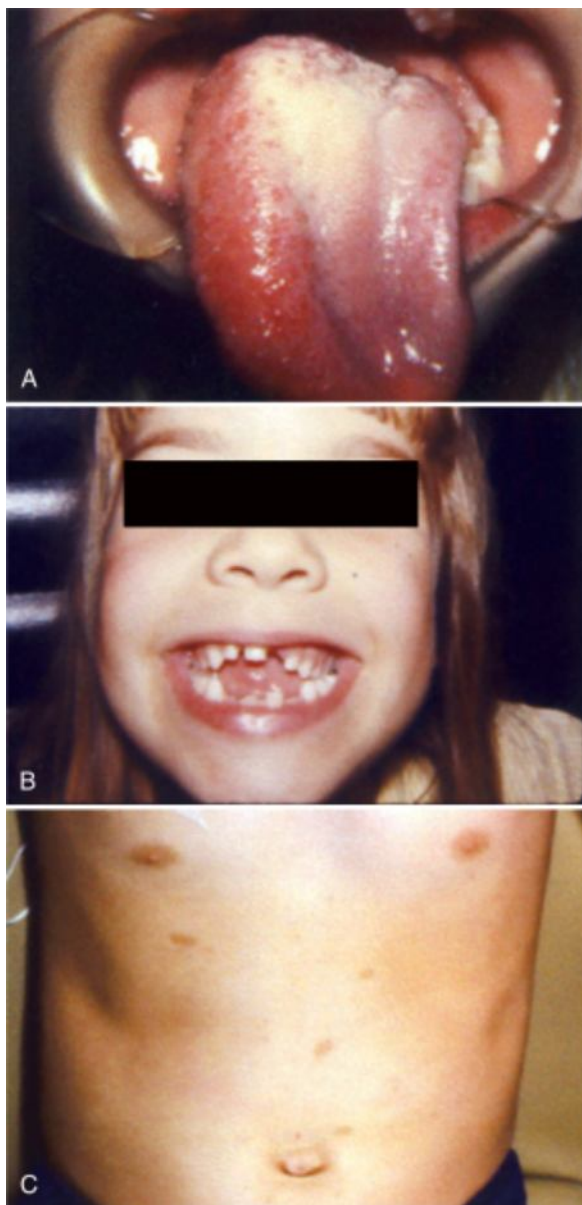
The decision to treat macroglossia surgically should be based on the presence and degree of three types of problems: functional deficits, such as deglutition, drooling, phonation difficulty, and airway obstruction; dental-skeletal alterations from excessive action of the tongue on surrounding structures, such as an increase in the gonial angle and the anterior facial height with the development of an anterior open-bite and excessive vestibularization of the inferior incisors; and psychological consequences of the physical appearance of the patient that give the false impression of mental deficiency, such as lingual protrusion, dyslalia, and drooling. In cases in which uniform tongue reduction is not necessary, such as in patients with cystic neoplasms, we perform partial glossectomy with the aim of resecting the entire mass. In every case surgical treatment must preserve the vital functions of taste, sensitivity, and mobility.[4]

Beckwith-Wiedemann syndrome consists of macroglossia, exomphalos, somatic gigantism, neonatal hypoglycemia, organomegaly, hemihypertrophy, and risk of tumor development.[6] Macroglossia is due to hyperplasia of the muscle fibers of the tongue. It may be symmetrical, or hemihypertrophy may be present. Of the many consequences of macroglossia, the most important is obstruction of the upper airway at rest or during feeding. It may also interfere with normal articulation and feeding. A major consequence of macroglossia is protrusion of dentoalveolar structures resulting in an anterior open bite and the appearance of a prognathic mandible. Laroche and associates[7] described two patients with Beckwith-Wiedemann syndrome associated with a cleft palate. Treatment is complex, but the authors prefer to not repair the cleft palate before performing tongue reduction procedures. This sequence would enable the patient to heal more efficiently.

### **PREOPERATIVE PLANNING**

A history and physical examination are of vital importance in evaluating patients for tongue reduction procedures. In most of these patients the history will reveal a lifelong presence of localized or generalized enlargement of the tongue. Patients with neurofibroma should undergo a complete physical examination because in some of these

patients, neurofibroma involving the tongue may be a localized manifestation of neurofibromatosis (von Recklinghausen's disease). The presence of café au lait spots is pathognomonic of this condition (Fig. 22-2). A large epidermal cyst of the tongue will produce the same symptoms as a localized form of macroglossia (Fig. 22-3).



**Figure 22-2** A, Neurofibroma of the tongue. B, Malocclusion caused by an enlarged tongue. C, Café au lait spots confirm the diagnosis of von Recklinghausen's disease.



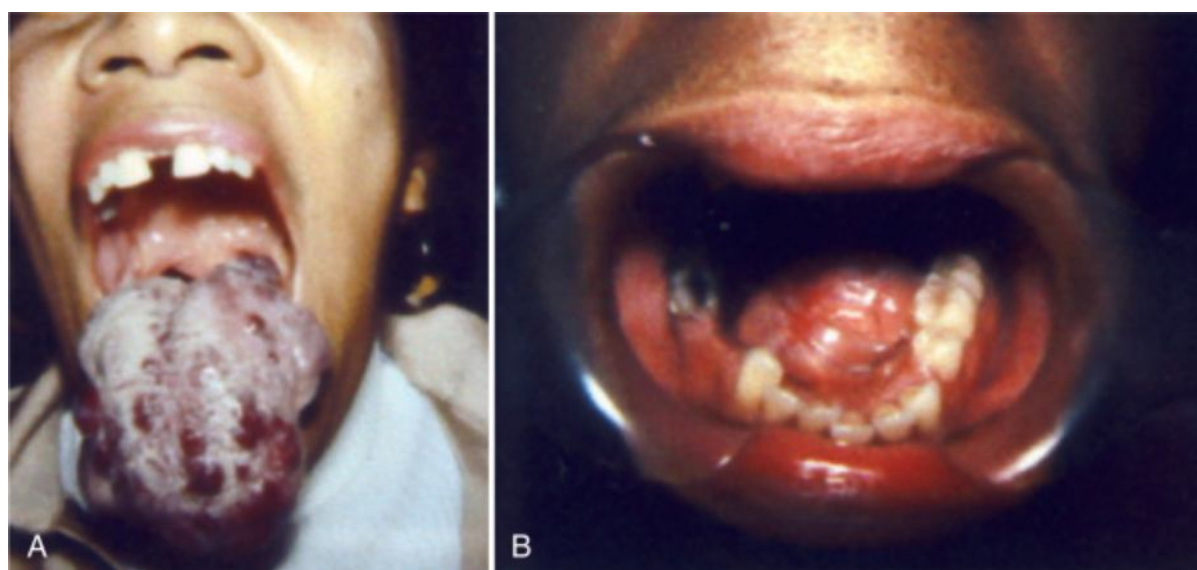
**Figure 22-3** Patient with a lifelong history of difficulty articulating because of a large epidermal cyst of the tongue.

Patients with lymphangiomatous macroglossia will not only have a history of lifelong difficulty with enlargement of the tongue (Fig. 22-4A) but may also have marked enlargement and ulceration of the tongue as a result of upper respiratory tract infection or minor trauma (see Fig. 22-4B). These patients respond to antibiotic therapy. In young children, the mandibular incisor and canine teeth may fail to erupt from pressure of the enlarged tongue. After tongue reduction procedures these teeth will erupt (see Fig. 22-4D). Patients with lymphangiomatous macroglossia often have involvement of the floor of the mouth and submental space (see Fig. 22-4C), and unless these areas are included in the tongue reduction procedure, the outcome will be less than satisfactory.



**Figure 22-4** Lymphangiomatous macroglossia. **A**, Usual appearance. **B**, Appearance during an upper respiratory tract infection. **C**, Involvement of the submental space with lymphangioma. **D**, Appearance after a tongue reduction procedure, including the submental space. The incisor teeth erupted after the decrease in size of the tongue.

Macroglossia is also thought to be an etiologic factor in open bite. Bimaxillary protrusion and special dental arches may cause instability after orthognathic treatment.<sup>[8]</sup> Patients with hemangioma may have a history of not only enlargement of the tongue but also bleeding from the tongue. Physical examination will reveal localized or generalized areas of obvious hemangioma (Fig. 22-5A). Patients who have the previously mentioned symptoms and are troubled by bleeding, swelling, or interference with eruption of dentition or occlusion should be considered appropriate candidates for these procedures.

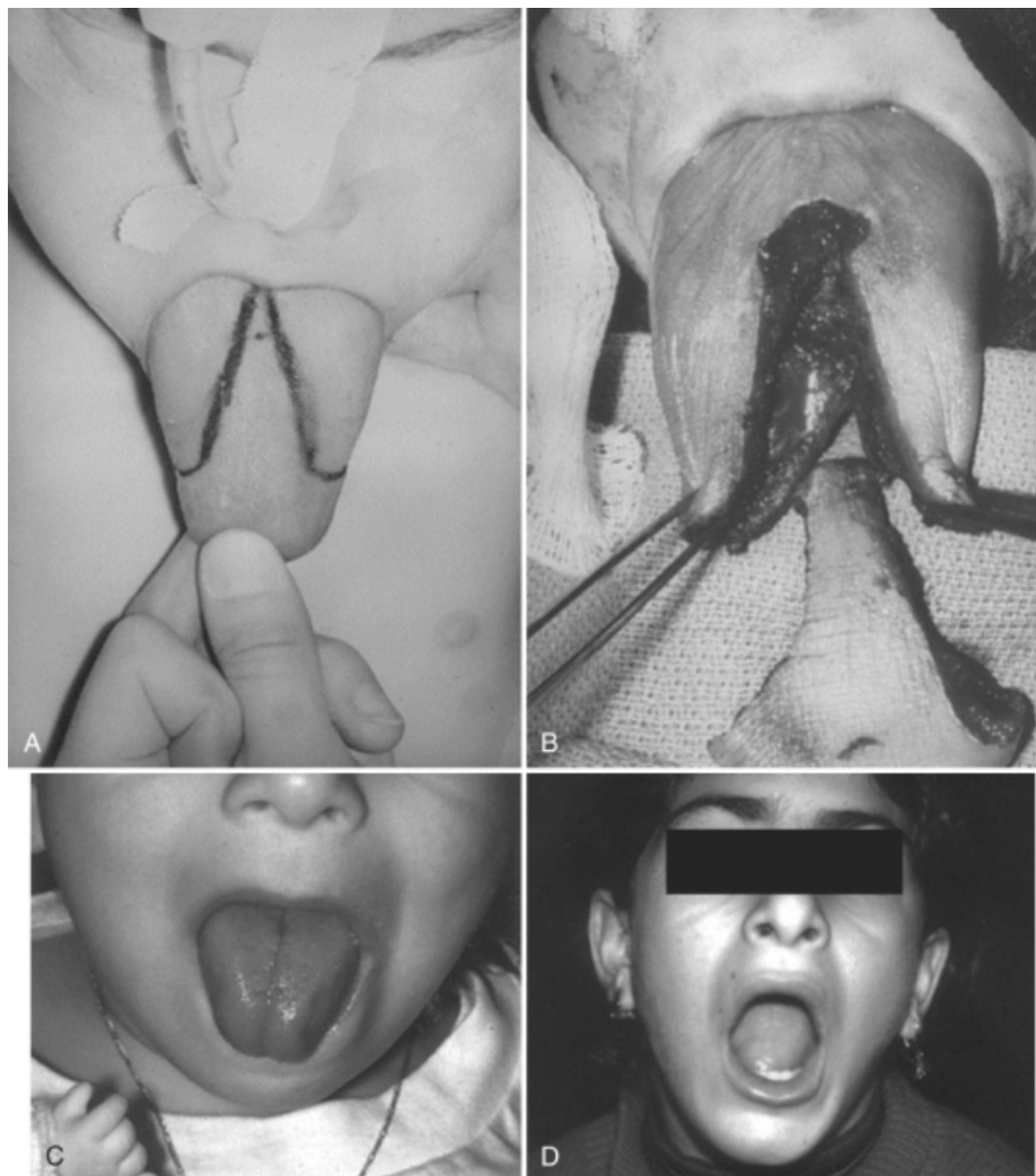


**Figure 22-5** **A**, Hemangioma of the tongue that was unresponsive to repeated partial surgery, sclerosing agents, and radiation therapy. **B**, Appearance after a tongue reduction procedure (note the anterior displacement of the central and lateral incisions).

Physical examination should include a thorough inspection of the tongue, floor of the mouth, and submental space, as well as bimanual palpation of these structures. Palpation will give the examiner an idea of the extent of these lesions, particularly in the case of more localized lesions. Computed tomography or magnetic resonance imaging may provide an estimate of the size and location, particularly of smaller tumors, which is helpful in planning.

### **SURGICAL TECHNIQUE**

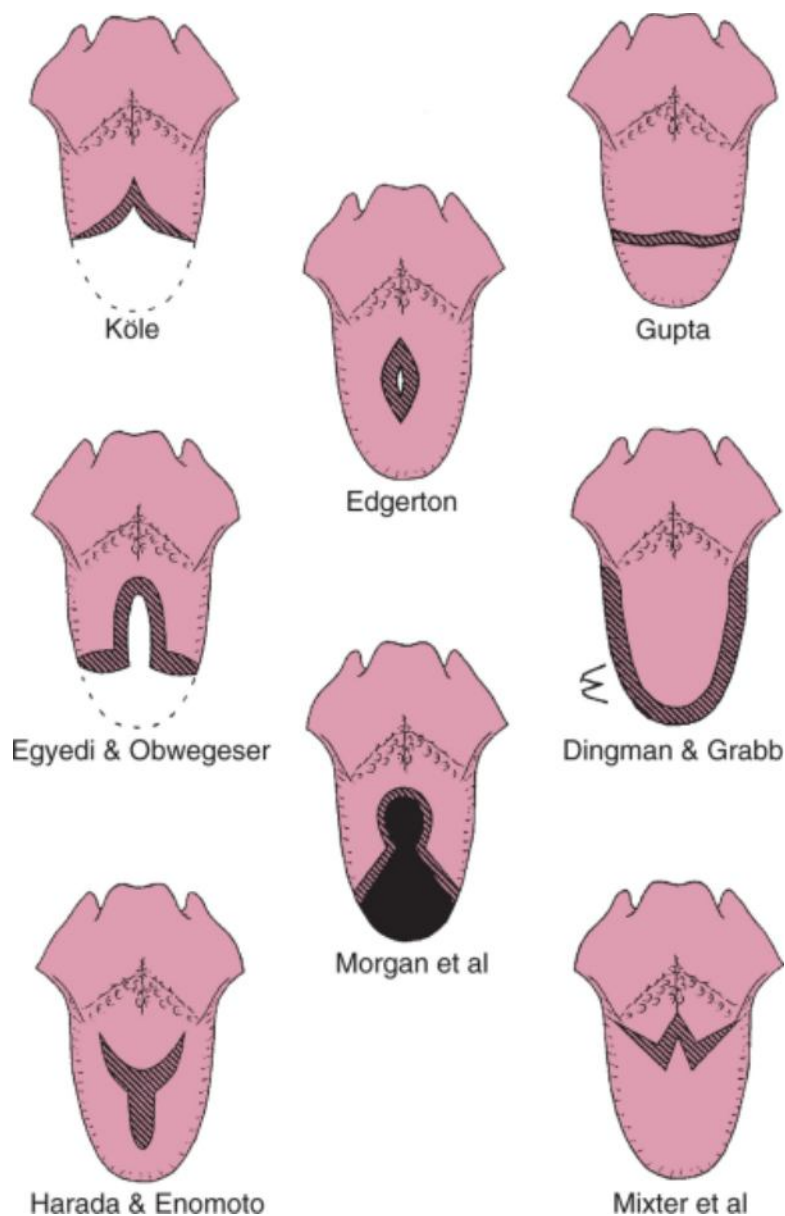
Wang and colleagues<sup>[6]</sup> presented an interesting array of surgical procedures but prefer the keyhole procedure. Kacker and associates<sup>[9]</sup> described a wide variety of tongue reduction procedures, some of which they used in their series of six cases of macroglossia. These authors also prefer the anterior keyhole procedure, which was able to uniformly reduce the tongue and provide good function and mobility (Fig. 22-6A to D).



**Figure 22-6** A and B, Intraoperative pictures showing a keyhole tongue reduction. C, Postoperative picture showing the reconstructed tongue. D, Postoperative patient at 3-year follow-up with good tongue reduction and an intraoral tongue.

(Reprinted with permission from Kacker A, Honrado C, Martin D, Ward R: Tongue reduction in Beckwith-Weidemann syndrome. *Int J Pediatr Otorhinolaryngol* 53:1-7, 2000.)

Symmetrical macroglossia can be corrected with a variety of techniques well illustrated in a paper by Davalbhakta and Lamberty (Fig. 22-7).<sup>[8]</sup> With the excision kept in a medial location, the hypoglossal nerves are preserved anteriorly and the vascular supply more posteriorly. A new tongue tip is created by approximation of the sides of the wedge. This technique not only narrows but also shortens the tongue. Approximation should be performed in layers with absorbable suture to close the dead space.

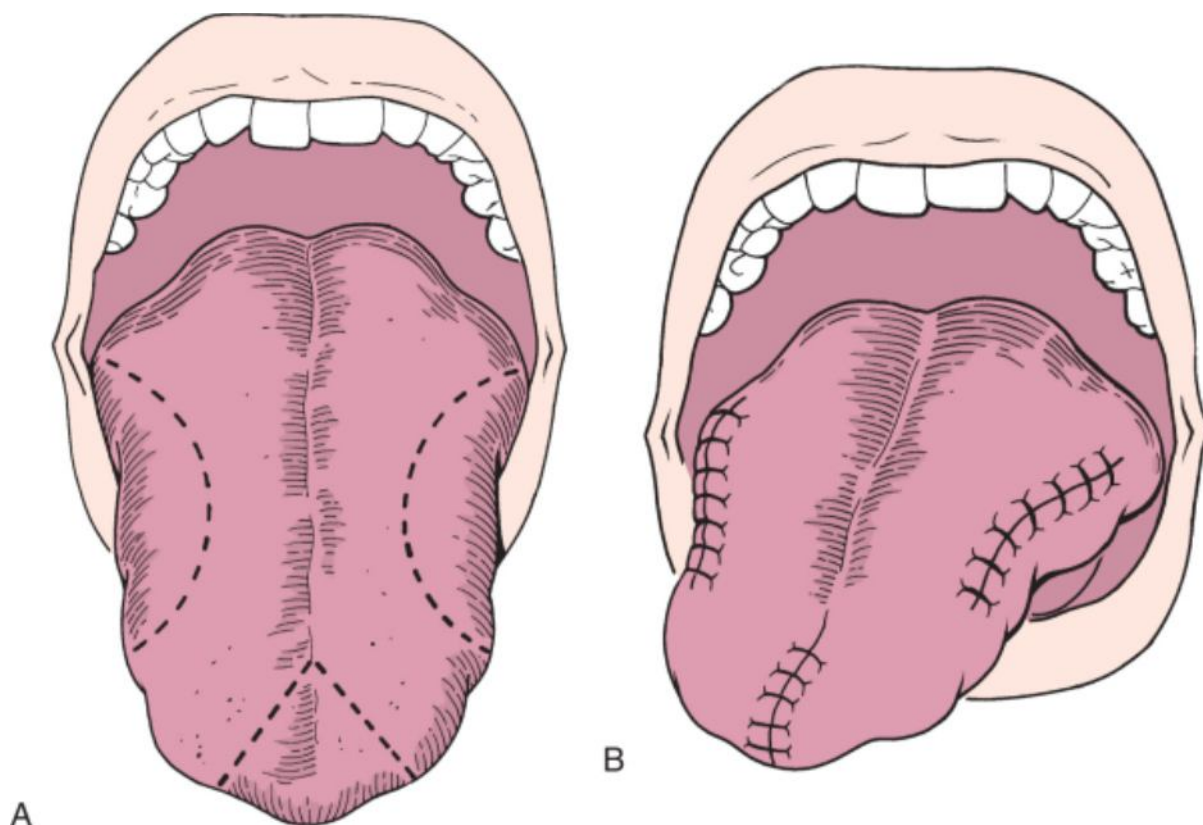


**Figure 22-7** Various techniques of tongue reduction.

(Reprinted with permission from Davalbhakta A, Lamberty BGH: *Technique for uniform reduction of macroglossia*. *Br J Plast Surg* 53:294-297, 294-297, 2000.)

Tongue reduction procedures, particularly in patients with involvement of the entire tongue, have the potential for airway obstruction. Therefore, a preliminary tracheotomy should be performed because the large size of the tumor may also preclude the possibility of easy endotracheal intubation. Antibiotics are administered perioperatively. Some surgeons routinely administer perioperative steroids to reduce the immediate postoperative edema that commonly accompanies tongue surgery.

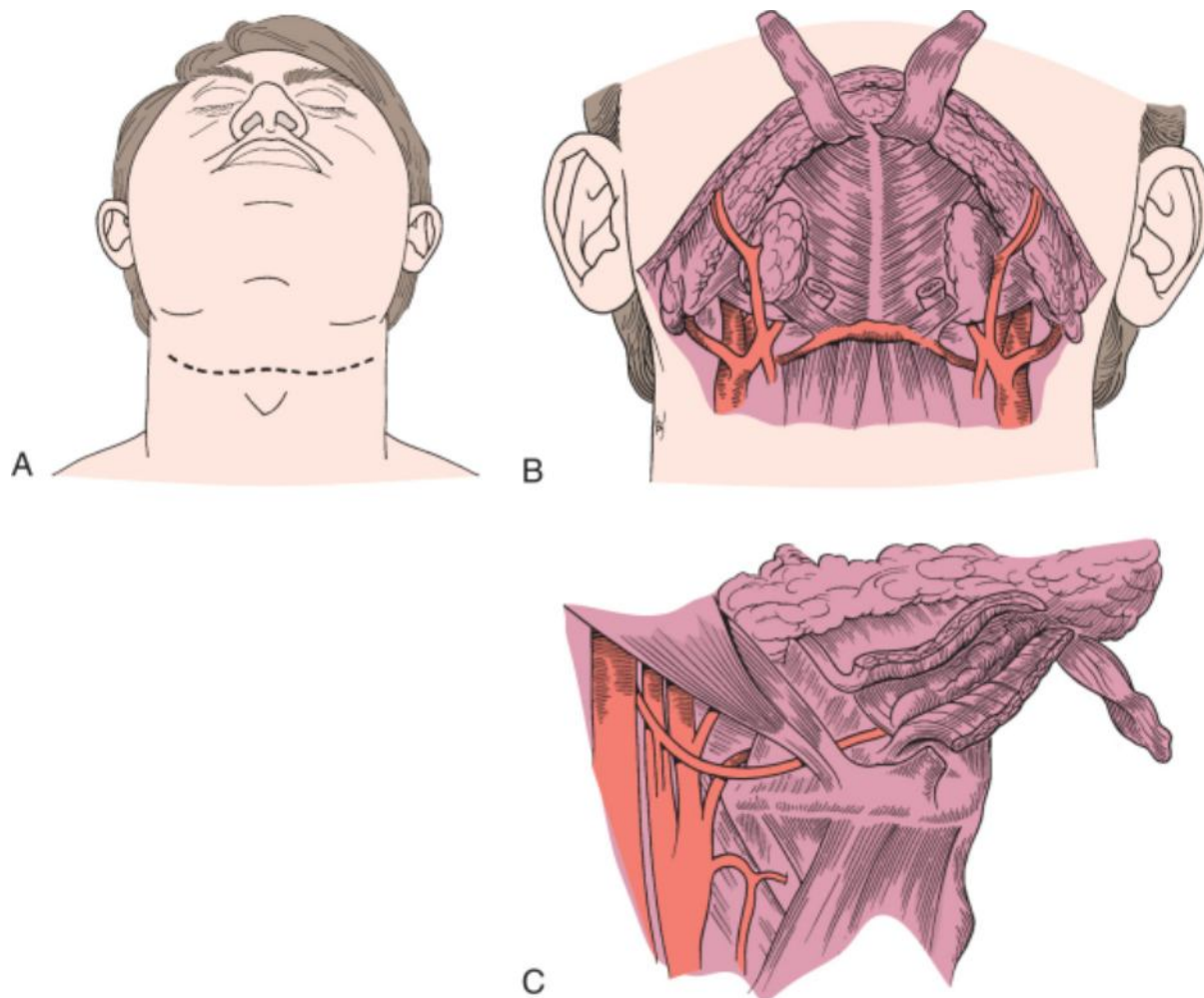
Patients with localized neurofibroma may be approached in a way similar to that described for partial glossectomy. A Jennings mouth gag is inserted, and a suture is placed approximately 1 cm posterior to the tip of the tongue in the exact midline. Using right-angled retractors to retract the buccal mucosa, the lesion in question is identified. The tongue is dried with gauze, and based on physical examination and imaging (if used), marks are made that describe an ellipse on the tongue overlying the lesion. An ellipse of mucosa and the underlying lesion are excised to facilitate closure (Fig. 22-8).



**Figure 22-8** A and B, Partial excision of the tongue.

Patients with lymphangiomatous macroglossia that involves the submental space and floor of the mouth must be approached extraorally through the neck, as well as intraorally. An incision is made in a skin crease just below the submental space (Fig. 22-9A). Skin flaps are elevated, and the anterior belly of the digastric muscle is identified (see Fig. 22-9B). An incision is made in the fascia between the two anterior bellies. The mylohyoid muscle is often very thin because of stretching by the lymphangioma and may be difficult to see. Before any resection in this area, the lingual arteries and hypoglossal nerves are identified and dissected up into their entrance in the anterior portion of the tongue (Fig. 22-9C). A small Penrose drain or vessel loop is placed around each hypoglossal nerve to protect it from injury and retract them out of the way of the incision. The mylohyoid muscles are transected to gain entry into the floor of the mouth (Fig. 22-9C). The area of lymphangioma is identified and excised as completely as possible. These tumors are usually infiltrative and not encapsulated, which makes it very difficult to know precisely when to stop. In such cases the excision is “sculpted” rather than attempting complete excision. Hemostasis is obtained, and the wound is irrigated and closed in layers over a Hemovac drain.





**Figure 22-9** A, Incision made in the submental space. B, Retraction of the anterior bellies of the digastric muscle. C, The lingual arteries and hypoglossal nerves are identified and retracted, the mylohyoid muscles are transected, and the submental space is entered.

Jian<sup>[10]</sup> reported a series of seven patients with lymphangioma or hemangioma of the tongue from 1992 to 2002 who were treated by a technique designed to significantly reduce the entire volume of the tongue. Endotracheal intubation was used in these cases to protect the airway. Cosmesis and function were improved after surgery, and none of the patients had a recurrence.

### **POSTOPERATIVE MANAGEMENT**

The perioperative antibiotics are continued for one week. Steroids, if used, are generally discontinued in 24 hours. Local irrigation with half-strength hydrogen peroxide is used to mechanically débride the suture lines and promote good healing. If a tracheostomy has been performed, the patient is decannulated as soon as the potential for airway obstruction has passed. Aspiration will usually be reduced or eliminated by plugging the tracheostomy tube or by decannulation. Patients should be fed by nasogastric tube during this time. Those who have undergone localized excision may be fed intravenously for several days, with oral feeding begun once the pain in the tongue has decreased to the point at which swallowing may be resumed. Involvement of a speech and swallowing therapist may be helpful in assessing the patient's readiness to eat.

### **COMPLICATIONS**

Complications include edema, airway obstruction, paralysis, anesthesia, and devascularization of the tongue. Edema and airway obstruction may be prevented by adequate hemostasis in the deep musculature of the tongue. Even though airway obstruction may be prevented in this manner, it may be wiser to perform a preliminary tracheostomy in patients who will undergo major tongue reduction procedures, particularly those with hemangiomas. The use of perioperative steroids and antibiotics is helpful in reducing or preventing postoperative edema.

One of the potential pitfalls of this procedure is injury to the neurovascular bundle, which may produce either partial or complete paralysis of one or both hypoglossal nerves. Although function is only minimally impaired by

interruption of one hypoglossal nerve, total paralysis caused by interruption of both hypoglossal nerves is devastating and will result in the patient becoming an "oral cripple." If the hypoglossal nerve is inadvertently cut, neurorrhaphy with 8-0 or 9-0 nylon should be performed by microscopic technique. Similarly, interruption of a lingual nerve will result in unilateral anesthesia of the oral tongue. Nerve injury should be repaired via neurorrhaphy. Devascularization of the tongue with resultant necrosis and loss of tongue substance may occur if both lingual arteries are lost.

Although tongue vascularity can be maintained with one artery, loss of both arteries is incompatible with survival of viable tongue. This complication can be prevented by preliminary identification of the arteries and avoidance of bilateral posterolateral tongue resection.

#### PEARLS

- Macroglossia may be a local disorder or a local manifestation of a systemic disease.
- Many techniques have been described for tongue reduction, all of which require careful patient selection and meticulous surgical technique.
- Edema and postoperative bleeding are usually preventable with meticulous hemostasis in the deep musculature of the tongue.
- Preservation of important anatomic structures, such as the lingual arteries and hypoglossal nerves, will ensure maintenance of the viability and function of the tongue.
- Speech and swallowing therapists, as well as psychologists, are valuable team members in the more exuberant form of macroglossia (see Fig. 22-1).<sup>[5]</sup>

#### PITFALLS

- Systemic disease, such as primary amyloidosis, should not be overlooked as a cause of macroglossia.
- Failure to identify and preserve the hypoglossal nerves will result in paralysis of the tongue with the patient becoming an oral cripple.
- Loss of both lingual arteries will result in necrosis of the tongue.
- Potential airway obstruction should be anticipated by performing a tracheostomy in cases in which large amounts of tongue may be removed.
- The possibility of cleft palate in association with macroglossia (Beckwith-Wiedemann syndrome) should not be overlooked.

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