

# Subtotal glossectomy by modified keyhole lingual resection technique for the treatment of true macroglossia

*Glossectomia subtotal pela técnica de ressecção lingual em orifício de fechadura modificada como tratamento de macroglossia verdadeira*

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## ABSTRACT

We describe a case of true macroglossia in a 6-year-old child that was surgically treated with a midline elliptical excision and anterior wedge resection along a Gothic-shaped arch. The postoperative result was considered satisfactory with regard to both the volume of the tongue, which now remains entirely within the mouth, and the aesthetic appearance; the ability to taste has been maintained.

**Keywords:** Macroglossia/congenital. Macroglossia/surgery, Glossectomy. Tongue/surgery.

## RESUMO

Neste artigo é apresentado o caso de macroglossia verdadeira em criança de 6 anos de idade, tratada cirurgicamente, com excisão elíptica na linha média da língua e ressecção ampla na ponta da língua em arco gótico. O resultado pós-operatório foi considerado satisfatório, tanto sob o aspecto de volume da língua, que passou a se manter totalmente dentro da boca, como sob o aspecto estético, mantendo a capacidade de gustação normal.

**Descritores:** Macroglossia/congênito. Macroglossia/cirurgia, Glossectomia. Língua/cirurgia.

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## INTRODUCTION

Macroglossia is an unusual pediatric condition with a variety of etiologies<sup>1,2</sup>. It is classified as true macroglossia when the tongue size is larger than normal, and as relative macroglossia when the oral cavity is not large enough to accommodate the tongue.

Macroglossia may be congenital or acquired<sup>2</sup>. The diagnosis is based on the presence of tongue protrusion with reference to the teeth, open lips, and maintenance of open-mouth posture<sup>3</sup>.

Myer et al.<sup>4</sup> classified the etiologies of macroglossia – congenital, inflammatory, traumatic and neoplastic – as either

generalized or localized (Table 1), according to the extent of the disease process<sup>5-24</sup>.

The tongue is involved in many functions, including swallowing, phonation, breathing, and normal development of the alveolar process and facial bone structure. Because of this, macroglossia may cause a number of problems, including difficulty in speaking, chewing, and swallowing as well as airway obstruction. In addition, tongue protrusion predisposes to tongue trauma, dry mouth, and repeated infections of the upper airways<sup>25</sup>.

The evaluation of macroglossia begins with a careful consideration of the patient's history and physical examination, with particular attention to changes in breathing, swallowing,

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**Table 1** – Summary of macroglossia causes.

Etiology		Extent of disease	Example
Congenital		Localized	Hemangioma, lymphangioma
		Generalized	Down syndrome, Beckwith-Wiedemann syndrome, triploid syndrome, Behmel syndrome, gargoylism, mucopolysaccharidosis, gangliosidosis type 1, Tollner syndrome
Inflammatory		Localized	Tuberculosis, syphilitic gum, dental infection, ranula, Riga disease, actinomycosis
		Generalized	Chronic glossitis
Metabolic		Generalized	Amyloidosis, lipoid proteinosis, chronic treatment with steroids, acromegaly, myxedema
Traumatic		Localized	Hematoma, dental irritation, postoperative edema
		Generalized	Postoperative edema
Neoplastic	Benign	Localized	Carcinoma, sarcoma
	Malign	<b>Generalized</b>	Granular cell tumor, neurofibroma, leiomyoma, lipoma

phonation, and alteration of dental occlusion. Effort should also be made to try to identify any undiagnosed syndrome.

The therapeutic approach should include follow-up observations, treatment of any underlying systemic condition that may be causing tongue enlargement, orofacial therapy, and proper surgical intervention that preserves the vascularization and innervation of the tongue.

Surgical resection is considered the most appropriate treatment in the pediatric age group. Keyhole subtotal glossectomy is an optimal technique for these patients because it allows normal body development and prevents dentoalveolar complications. This technique was originally described by Morgan et al.<sup>26</sup> and differs as to the reduction of muscle size in the 3 planes.

Surgical success should be evaluated by many factors, such as the disappearance of teeth marks on the edges of the tongue; proper positioning of the tongue within the oral cavity; improvement in or resolution of respiratory disorders; improvement in swallowing and phonation; preservation of taste, heat pain, epicritic and protopathic sensitivity; and improvement in tongue mobility.

Currently, priority is given to multidisciplinary treatment, including orthodontic, surgical, and phonoaudiological evaluation.

### CASE REPORT

The patient, a 6-year-old girl, presented with a tongue with increased length and width. The tongue was resting and was interposed between the teeth, interfering with phonation and causing difficulty in chewing. Assessment of the skeletal profile showed absence of any alterations related to dentofacial deformities. A diagnosis of true macroglossia was made (Figure 1).

Glossoplasty was performed under general anesthesia, according to the technique of modified keyhole subtotal glossectomy with Gothic arch-shaped extensive resection of the tongue ends (Figures 2 and 3).

The incision was carried out using a surgical scalpel; the diaeresis was completed with electrocautery, for better hemostasis. In this manner, the excess tongue muscle tissue was removed (Figures 4 and 5). The tongue was closed using absorbable Vicryl sutures (Figure 6).



**Figure 1** – Preoperative condition.

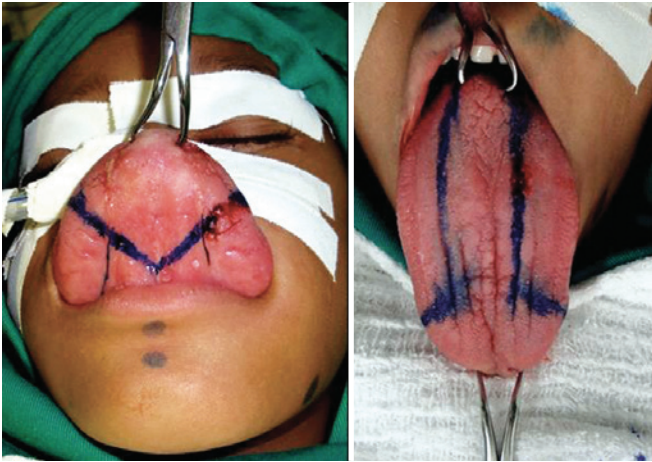


Figure 2 – Glossoplasty planning.

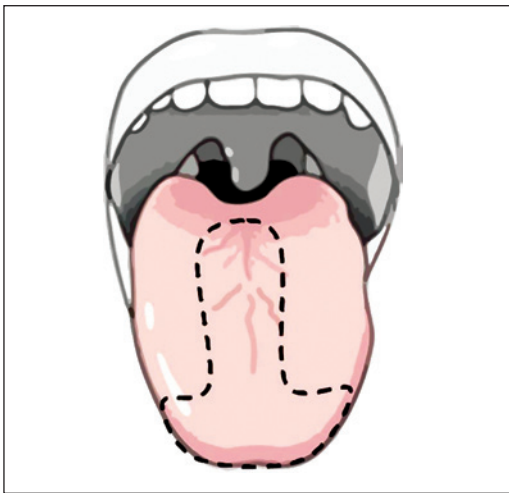


Figure 3 – Keyhole subtotal glossectomy, with gothic arch-shaped extensive resection of the tongue ends.

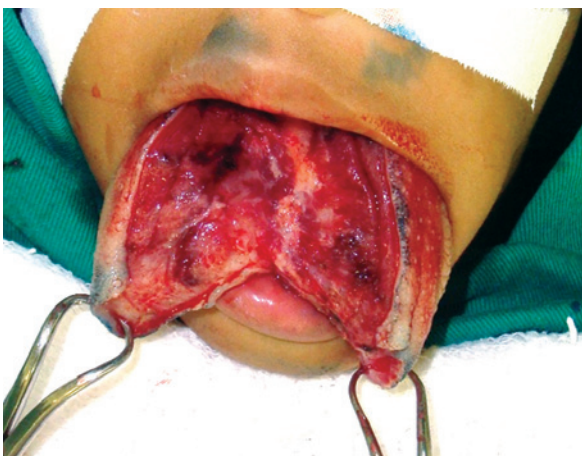


Figure 4 – Excess tongue tissue removal.

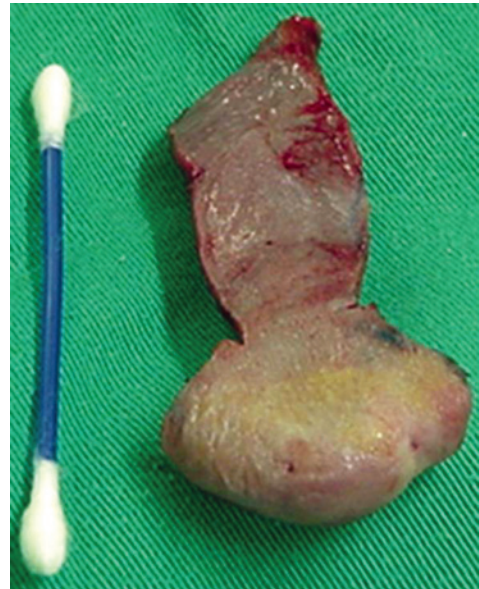


Figure 5 – Large rhomboid resection of the anterior third of the tongue.



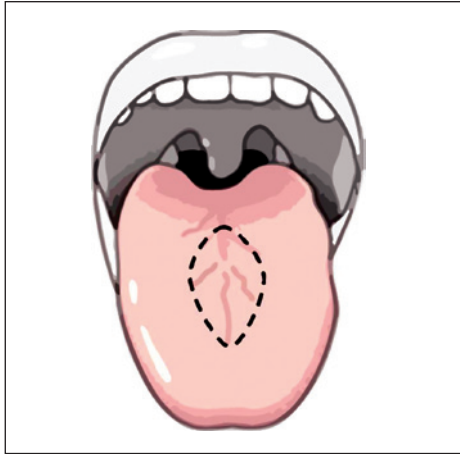
Figure 6 – Approximation of planes and sutures.

## DISCUSSION

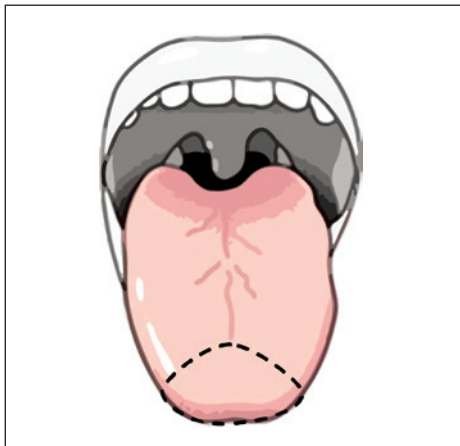
Subtotal glossectomy is the appropriate surgical treatment in most cases in patients with airway obstruction, difficulty in speaking, dysphagia, and aesthetic damage.

Several surgical techniques are described for the correction of true macroglossia; these are subdivided into median line and peripheral glossectomies<sup>27</sup>.

In 1950, Edgerton<sup>28</sup> described a technique consisting of excision along the center of the tongue with preservation of blood vessels, nerves, and taste buds, obtaining as result



**Figure 7** – Elliptical excision in the center of the tongue, as proposed by Edgerton<sup>28</sup>.



**Figure 8** – Triangular incision at the end of the tongue, as proposed by Kole<sup>29</sup>.



**Figure 9** – Anatomical presentation of the tongue at 9 months postoperatively.

getting the strengthening of the tongue without changing the length (Figure 7). In 1965, Kole<sup>29</sup> proposed a triangular incision at the end of the tongue, reducing the length and thickness of the anterior third of the tongue (Figure 8).

The surgical technique presented in this article is a variant of keyhole glossectomy, originally described by Morgan et al.<sup>26</sup>, and involves a large excision of the distal end of the tongue, thus further reducing its length.

The realization of this technique has shown good results in the case shown (Figure 9), as in others described in the literature<sup>27-30</sup>.

In cases of macroglossia with idiopathic muscle hypertrophy, such as the reported in this paper, there is risk of relapse, thus, the surgical indication must be individualized, taking into consideration the risks and possible benefits for the patient<sup>31,32</sup>.

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