CONGENITAL MACROGLOSSIA: A RARE CASE OF TONGUE HYPERTROPHY

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ABSTRACT
We are reporting a case of 2-year-old male child with congenital macroglossia presented with increased tongue volume and protrusion since birth, drooling of saliva, difficulty swallowing and phonation. The patient was submitted to partial glossectomy under general anesthesia using the ‘keyhole’ technique, with objective to provide reduction of the lingual length and width. Post-op results were good, without taste and motor alterations and without any discrete paraesthesia at the apex of the tongue. We are presenting this case because of the rarity of the disease and the successful outcomes of ‘keyhole’ technique in the treatment of this rare disease.

Keywords: Macroglossia, Keyhole, Partial Glossectomy

INTRODUCTION
The first report of macroglossia was a description of oral lymphatic malformation, in 1854, by Virchow and Uber. Macroglossia is an uncommon condition with variety of etiologies that can lead to several alterations like dental-muscle-skeletal deformities, orthodontic treatment instability, masticatory and breathing and phonation problems, characterized by increased size of the tongue, can be caused by congenital malformations or acquired diseases (Vogel et al., 1986).

Several techniques have been proposed in the literature to enable the reduction of the tongue. Peripheral incisions with marginal resection of tissue have as complications hypomobility and change in the form of the tongue that becomes globular (García et al., 2009). Incisions V-shaped positioned in the midline of the tongue are effective in reducing the length but are ineffective in reducing the width of the tongue (Hendrick, 1956). Elliptical incision positioned in the midline without reaching the apex of the tongue contributes to reducing the width with little influence on its length (Pichler et al., 1948). Incisions in the form of keyhole combine characteristics of elliptical and V-shaped incisions and are indicated when the reduction of the width and length of the tongue are desirable and its design can be changed according to the specific needs of each case (Kacker et al., 2000). Keyhole subtotal glossectomy allows normal body development and prevents dentoalveolar complications. This technique was originally described by Morgan et al., (1996) and differs as to the reduction of muscle size in the 3 planes.

CASES
A 2-year-old male child presented with increased tongue volume and protrusion since birth associated to drooling is saliva, difficulty swallowing and phonation. Upon examination, he had considerably enlarged tongue resting outside his mouth and was interposed between the teeth. Assessment of the skeletal profile showed absence of any alterations related to dentofacial deformities. Upon further investigations no other congenital abnormality detected. A diagnosis of true macroglossia was made. Partial glossectomy was performed under general anesthesia, according to the technique of modified keyhole subtotal glossectomy with 'T' shaped extensive resection of the tongue ends. The incision was carried out using a surgical scalpel; the dissection was completed with electrocautery, for better hemostasis. In this manner, the excess tongue muscle tissue was removed. The tongue was closed using absorbable Vicryl sutures. In the immediate postoperative period there was slight swelling in the floor of the mouth and on belly anterior tongue, without taste or motor alteration and with good tissue repair, perfect symmetry, and no tongue interposition. The histo-pathological examination revealed a cavernous hemangioma. Currently he has
excellent cosmesis, with spontaneous reduction in prognathism and is having improved phonation. The patient was referred to orthodontic and phonoaudiology treatment.

**DISCUSSION**

Regardless of the cause, macroglossia is a significant functional problem that can lead to acute airway obstruction in newborns, excessive drooling, feeding difficulties, dentoskeletal deformity, and psychosocial concerns. Unfortunately, macroglossia is often mistakenly associated with developmental delay or mental retardation. Though some children with enlarged tongues have syndromes that affect the neurologic system, many do not. This can lead to teasing, discrimination, and social isolation.
Surgery for the correction of macroglossia has been performed for hundreds of years. It began without an understanding of the cause and proceeded with crude forms of resection till the past century. With more understanding of the neuroanatomy of the tongue, vasculature, innervations, healing of muscles after surgery, understanding of surgical approach, and design has undergone rapid evolution.

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; improvement in tongue mobility and self-esteem of the patient.

The surgical technique presented in this article is a variant of keyhole glossectomy, originally described by Morgan et al., (1996) 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 as in others described in the literature (Gasparini et al., 2002, Kaufman et al., 2008). Excellent aesthetic and functional results were obtained with this technique, with improvement of ronchi, appearance, feeding and speech after surgery. The child could develop speech and breath adequately, thus avoiding future craniofacial problems. The surgical technique employed in our case recommends keyhole resection of tongue. It is a versatile resection that may be used in most cases of macroglossia.

We recommend a multidisciplinary treatment plan for these patients, including orthodontic, surgical, and phonoaudiological treatment.

REFERENCES

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Case Report
