MACROGLOSSIA : A RARE CLINICAL ENTITY

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Introduction

Macroglossia is defined as a resting tongue that protrudes beyond the teeth or alveolar ridge [1]. In studying macroglossia, no one has described normal tongue size or applied direct measurement to the pathologically enlarged tongue. Although it is a relatively uncommon disorder, it may cause significant morbidity. Macroglossia causes a variety of signs and symptoms. These include tongue protrusion which exposes the tongue to trauma. The exposure also leads to mucosal drying and recurrent upper respiratory tract infections. Other symptoms include speech impediment, swallowing difficulties, airway obstruction, mandibular deformities, drooling and failure to thrive. Macroglossia in the absence of any systemic disorder or outgrowth of tongue due to tumours etc in an otherwise normal child is very rare entity and considered to be the result of true hypertrophy of the tongue muscles [2]. Such a case presented to our hospital and was taken up for surgery. This case report is to highlight the importance of preanaesthetic assessment to exclude or identify any underlying systemic disease and to prepare for the difficulty in intubating the patient.

Case Report

A 13 year old son of a retired soldier, a case of macroglossia was posted for anterior and lateral wedge resection of tongue to reduce the tongue bulk. He reported with generalized, painless, gradual enlargement of tongue of more than three years duration. He had no difficulty in chewing, swallowing or change in speech. There were no constitutional symptoms. Examination revealed an adolescent boy with normal vitals. Routine investigations were found to be normal. Patient was euthyroid. Left submandibular and parotid gland were enlarged and soft. No bruit or thrill could be appreciated over the mass or carotids.

In this case the mouth was constantly open and tongue occupied almost the entire mouth. Patient had prognathism. Anterior half of the tongue was hypertrophied more so on the lateral sides (Fig-1). Plaque were present over lower teeth. Further mouth

opening allowed visualization of hard palate only, putting the patient in Class IV of Mallampatti classification (according to airway assessment score by Mallampatti it is the most difficult case for intubation). Other tests to assess the airway like Patil test (thyromental distance > 6.5 cm), Savva test (sternomental distance > 12.5 cm) were within acceptable limits. A difficulty in intubation was anticipated. A blind awake tracheal intubation was planned. All steps in the management of predicted abnormal airway were taken. Endotracheal tubes of different sizes, 2 laryngoscopes with different size blades, laryngeal mask airway (size 2 and 3), Mini-trach set were kept ready to tackle failed intubation. Patient was monitored with electrocardiogram, non-invasive blood pressure monitor and pulse oximeter. Topical anaesthesia using 4% lignocaine gargles was given. Bilateral superior laryngeal nerve block was induced by injecting 3 ml of 2% lignocaine below each

Fig. 1 : Macroglossia - preoperative profile

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Fig. 2 : Post-operative improvement

greater cornu in the thyrohyoid membrane. Patient was placed in classical Magills position.

Nasal passage to be used was selected. A well lubricated tracheal tube was then blindly passed into the trachea by listening and feeling the air breathed out from the proximal end of the tube without any difficulty. The throat was packed. Anaesthesia was maintained with O₂, N₂O and muscle relaxants. Haemostatic ligatures were first applied on the undersurface of the tongue. Hypertrophied tongue was excised and haemostatic sutures were applied on the cut surface. Due to excision of the hypertrophied tongue and the obstruction being relieved there was no respiratory obstruction post extubation. A Ryle's tube was passed and residual paralysis was reversed and patient was extubated when he was fully awake and oriented. He was maintained on Ryle's tube feed for three days and then put on oral feeds. Oral mouth gargles were also given. Patient had an uneventful post operative period. He has shown definite improvement in appearance after surgery as shown in (Fig-2).

Discussion

The term macroglossia is referred to an enlarged tongue. Macroglossia may be classified in several ways. A classification by Myer et al divides the causes into generalized and localized based on the extent of tongue involvement [3]. An aetiological classification has been proposed by Kharbanda et al [4].

a. Primary macroglossia (muscular enlargement)

- i. Isolated macroglossia due to muscular hypertrophy
- ii. Isolated macroglossia

aa. Relative macroglossia due to disproportionate growth of tongue and jaws.

ab. True macroglossia due to isolated true muscular enlargement.

b. Secondary macroglossia

i. Congenital

aa. Local causes - lymphangioma, hemangioma, cystic hygroma

ab. Systemic - Beckwith Wiedman syndrome, Hurlers syndrome, Mongolism.

ii. Acquired - Acromegaly, Amyloidosis.

Postoperative macroglossia can occur after prolonged surgery in prone position if due precautions are not taken [6].

Evaluation begins with a careful history and physical examination to identify any undiagnosed syndrome. Lab tests are guided by clinical suspicion. Thyroid function tests are used to rule out hypothyroidism. Absence of clinical symptoms, normal thyroid function tests and radiograph excluded the above mentioned possibilities. The tongue appeared normal in shape, colour, consistency and movements hence isolated true macroglossia though not a common entity was considered the etiological cause in this case as has also been reported earlier by Kharbanda et al and Sacco et al [2,4].

Treatment options include observation, orofacial therapy and surgery. Patients with minimal symptoms may be observed since changes in tongue position can improve the disorder. Orofacial therapy uses a palatal device to stimulate muscular tone and proper tongue position. The majority of cases of macroglossia are treated surgically. The technique used is an anterior wedge resection with posterior key hole. The lateral incisions are beveled out to decrease tongue bulk. The tongue is then closed in T [5].

Since a difficult intubation was anticipated blind awake tracheal intubation was planned. The patient being an adolescent boy the procedure could be explained to the patient so that he was fully cooperative.

The blind nasal intubation technique is not a routinely used procedure and may fail in the best of hands. Its other complications include epistaxis and avulsion of nasal polyp. However the main indication for blind intubation is the patient in whom direct laryngoscopy is likely to be difficult or has failed. The technique should be fully explained and the patient's cooperation is vital. Extreme gentleness should prevail. If the tube does not enter the larynx its direction may be adjusted by rotation of the tube, rotation of the neck or digital movement of the larynx to meet

the advancing tube. Normal free breath sound at the proximal end of the tube shows that it is in the trachea. Successful insertion of tube in the trachea is confirmed by capnography. If unsuccessful, blind intubation should not be persisted or trauma may result. Persistent attempts at intubation of a difficult case is often nonproductive and can even be disastrous. In such a case oxygenation of patient without aspiration should be the main aim.

Any macroglossia represents a potential problem for intubation however a careful pre anaesthetic evaluation and careful planning of the procedure ensures that anaesthesia and surgery of the patient is completed smoothly as has been done in this case.

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