

Lymphangioma of Tongue: A Case Report

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Abstract: *Lymphangioma is a benign tumour involving the lymphatic channels and is mostly confined in the head and neck region in about majority of cases. Although lymphangiomas commonly occur in the head and neck region, intraoral lymphangiomas are rare. Lymphangioma of the tongue may cause gross structural deformity of the face and interfere with speech and swallowing. Anaesthetic concerns include difficulty in mask ventilation, intubation, bleeding, extrinsic and intrinsic pressure on the airway causing distortion and enlarged upper respiratory structures like tongue, lips and epiglottis. We present a case of a 2 year old male child with the lymphangioma of tongue.*

Keywords: Lymphangioma, Macroglossia

1. Introduction

Lymphangioma is a benign tumour involving the lymphatic channels and is mostly confined in the head and neck region in about majority of cases.^{1,2}Virchow in 1854 showed that lymphangioma arise from the dilatation of lymphatic spaces. Ewing in 1942 has further remarked that, being composed of lymph vessels, they consist of endothelial cells and supporting connective tissue, both of which take part in the neoplastic process. Although lymphangiomas commonly occur in the head and neck region, intraoral lymphangiomas are rare.³In the oral, cavity anterior two-third of the tongue is most commonly involved site. Lymphangioma of the tongue may cause gross structural deformity of the face and interfere with speech and swallowing.⁴ Lymphangiomas are the most common cause of macroglossia in infancy. They are known to be associated with Turner's syndrome, Noonan's syndrome, Trisomies, Cardiac anomalies, Fetalhydrops, Fetal alcohol syndrome and Familial pterigiumcolli. Anaesthetic concerns include difficulty in mask ventilation, intubation, bleeding, extrinsic and intrinsic pressure on the airway causing distortion and enlarged upper respiratory structures like tongue, lips and epiglottis. We present a case of a 2 year old male child with the lymphangioma of tongue.

2. Case Report

A 2 year old male child weighing 10 kg reported to paediatric surgery OPD with generalized swelling of the tongue. A pea sized swelling over tongue was noticed by the parents immediately after birth but was left untreated. There was gradual increase in the size of the swelling. History of progressive difficulty in feeding the child was given by the parents but there was no breathing difficulty. There was no significant antenatal or family history. No history of developmental delay, jaundice or cyanotic spells was present. Immunization of the child was complete till date. On general examination, child was conscious and active, with vitals within normal range. Local examination revealed an enlarged tongue protruding and keeping the mouth open permanently. Systemic examination was normal. Mouth opening, inter-incisor gap and Mallampatti grading could not be elicited due to enlarged tongue. No other congenital abnormalities were present. All routine investigations were

within normal limits. Ultrasound showed hypertrophy of tongue with multiple cystic areas seen in parotid and submandibular gland. Magnetic resonance imaging revealed enlargement of the tongue in the anterior 2/3 region. Lymphangiomas seen in bilateral parotid and submandibular glands with enlarged right parotid gland. Child was premedicated with intranasal midazolam 0.5 mg, 30 minutes prior to shifting to the operation theatre. On arrival to the operating room, routine monitors, including a pulse oximeter, noninvasive blood pressure cuff and electrocardiographic monitor were applied. Difficult airway cart, tongue stitch and tracheostomy set were kept ready. Child was induced with sevoflurane 2-6% in increments using size 3 transparent facemask. (fig 3) It was difficult to assist ventilation in supine position so he was turned to the right lateral position. Intravenous line was secured with 24 G cannula in the right hand. Injection glycopyrrolate 50 µg and fentanyl 20 µg were given IV. After checking for ability to ventilate, check laryngoscopy done and succinylcholine 20 mg given IV. Child was successfully intubated orally with 4.5 mm uncuffed tube in a single attempt. Anaesthesia was maintained with N₂O 67%, oxygen 33%, isoflurane 1% and atracurium. Injection paracetamol 150 mg IV, hydrocortisone 20 mg IV was given intraoperatively. The inverted V shape anterior half of the tongue was removed. The tumour had large cystic spaces filled with lymph like fluid. After completion of the surgery, patient was extubated without any adverse event. The intra-operative and recovery period were uncomplicated and child was shifted to the ward after being observed for 2 hours in the recovery room.



Figure 1: A child with lymphangioma of tongue



Figure 2: Mask ventilation with size 3 mask



Figure 3: Child with ETT in situ.

3. Discussion

Lymphangiomas are hamartomatous, congenital malformations of the lymphatics.⁵Lymphangioma can be classified into four categories-Lymphangioma simplex (lymphangioma circumscriptum): composed of small, thin-walled lymphatics. Cavernous lymphangioma: comprised of dilated lymphatic vessels with surrounding adventitia. Cystic lymphangioma (cystic hygroma): consisting of huge,

macroscopic lymphatic spaces with surrounding fibrovascular tissues and smooth muscles. Benign lymphoendothelioma (acquired progressive lymphangioma), lymphatic channels appear to be dissecting through dense collagen bundles. Cervical lesions in a child can cause dysphagia and airway obstruction which is rare in adults.⁶The anterior two-thirds of the dorsal surface of tongue is the most common site for intra-oral lymphangiomas leading to macroglossia. Macroglossia resulted in lesions on the dorsal surface of tongue, improper phonation and poor oral hygiene. It rarely arises on palate, gingival, buccal mucosa and lips. Awake intubation may be the first option but is not easily performed in children since cooperation is quiet essential. Adult fiberoptic bronchoscopes have an outer diameter of around 3.5-4.0 mm and thus can take realistically a size 4.0-4.5 mm endotracheal tube loaded onto them. Ultra thin fiberscopes have an outer diameter of 2.2 mm so a 2.5mm endotracheal tube can be railroaded over them. The optical quality of these scopes is good but it has no suction channel and secretions have to be aspirated with a suction catheter.⁷Though an ideal technique, pediatric bronchoscopic intubation is time consuming and requires expertise, skill and expert assistance proper size of FOB, smooth inhalational induction, deep plane of anaesthesia and maintenance of spontaneous ventilation.⁸Premedication and pre-oxygenation should be followed by inhalation of either halothane or sevoflurane in a spontaneously breathing patient. It is better to withhold muscle relaxants until the airway is secured. Intubation should be performed under deep inhalational anaesthesia. Cooperation of the child and support of the parents will be a factor in the induction decision-making process. The laryngeal mask airway in pediatric patients with difficult airway is an excellent aid and can be used as a conduit for intubation. Video laryngoscopes are new addition to airway armamentarium. A variety of videolaryngoscopes like King Vision are available for use in paediatric difficult airway and are a good alternative to fiberoptic bronchoscope as a first choice. Pediatric airway management poses unique challenges to the anaesthesiologist. Anesthesia for the child with lymphangioma involving the oral cavity and oropharynx requires thorough preparation and vigilance. Knowledge of the disease and the affected airway structures will help the anaesthesia provider prepare for the case by having the correct equipment and the safest plan for the anesthetic.

Conflicts of interest: Nil

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Volume 10 Issue 3, March 2021

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