

Failed Tracheal Extubation Due to Transient Isolated Macroglossia in a Child

To the Editor,

Massive tongue swelling has been reported in adult patients after neurosurgical procedures earlier.^[1,2] We report unanticipated macroglossia of an indeterminate etiology in a child after a successful on-pump cardiac surgery. This resulted in a failed on-table tracheal extubation. A 10-month-old boy underwent an uncomplicated bidirectional Glenn operation. The child had a history of snoring, presumably resulting from tonsillar and adenoidal hypertrophy. The child received general anesthesia for the Glenn procedure, and the trachea was intubated with a 4.0 mm un-cuffed endotracheal tube [Cormack-Lehane scale: Grade-1 laryngeal view]. The drugs administered as part of the general anesthesia regimen included fentanyl, thiopentone, dexmedetomidine infusion, sevoflurane, and cisatracurium for muscle relaxation. The child also received tranexamic acid, methylprednisolone, and two doses of cefazoline. A pediatric transesophageal echocardiography probe was advanced through a bite block. After the surgery, the child's trachea was extubated [Glenn pressure: 15 mm Hg]. Postextubation, the child developed signs of upper airway obstruction. There was a loud snoring sound and visibly increased work of accessory muscles of respiration and arterial oxygen desaturation [$<80\%$]. There was no evidence of laryngospasm, as the child could breathe spontaneously with minimal neck adjustment. After propofol administration [2 mg/kg], a video laryngoscopy was repeated, which showed tonsillar enlargement. The child's trachea was reintubated with a 4.0 mm endotracheal tube smoothly, and the child was transferred to the pediatric intensive care unit for further management. After about 30 mins of arrival at the intensive care unit, the tongue was seen to protrude out of the child's mouth [Figure 1]. There was no appreciable bronchospasm nor any noticeable skin rash. The edema appeared to be isolated to the tongue alone. Considering the possibility of an allergic reaction to the drugs administered intraoperatively, a blood sample was sent to estimate serum tryptase and C1 Esterase Inhibitor Antigen levels. Simultaneously, intramuscular adrenaline [0.06 mg] was given along with an intravenous steroid. With the child on mechanical support by a synchronized intermittent mandatory ventilation mode, the Glenn pressure was below 16 mm Hg. A color Doppler blood flow map by transthoracic echocardiography showed laminar flow through the Glenn shunt [Videoclip 1]. The tongue edema subsided within 6



Figure 1: The macroglossia in the child

hours, and the child's trachea was extubated successfully after 12 hours of assisted mechanical ventilation. The levels of serum tryptase [3.2, normal range: $<11 \mu\text{g/l}$] and C1 Esterase Inhibitor Antigen [0.37, normal range: 0.21-0.39 $\mu\text{g/l}$] were reported to be normal. Institutional ethical committee approval [CR#2023/28] and informed consent from parents were obtained for the publication of this report.

The manifestation of isolated tongue edema may be immediate when the drainage of the lingual veins is compromised.^[1] It may make a delayed appearance and takes time to resolve when there is a reperfusion injury due to lingual artery compression.^[1] Mechanical compression due to an endotracheal tube or a bite block, a tight throat pack, and even transesophageal echocardiography in children may result in tongue swelling.^[1-4] An angioedema characterized by an acute nonpitting generalized swelling and obstructed venous flow through the bidirectional Glenn shunt was excluded.^[5] The discernable tongue swelling manifested slowly but resolved within 6 hours. It was not possible for the authors to definitively establish the etiology of the patient's macroglossia.

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Conflicts of interest

There are no conflicts of interest.

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