Case Report

Massive lingual swelling following cleft palate repair

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Abstract

We report two cases of massive tongue edema in routine palatoplasty. All patients had uneventful recovery. We postulated that the macroglossia was secondary to ischemia and venous congestion after prolonged use of Killner Dott mouth gag with slotted tongue blade exaggerated by hyperextension of neck and Trendelenberg position.

Key words: Killner dott mouth gag, palatoplasty, tongue edema

Introduction

It is a most unwelcome experience and challenge for an anesthesiologist to find a potentially difficult airway at the end of procedure. The main concerns in palatoplasty, at the time of extubation are hemorrhage and upper airway obstruction.[1,2] It is well-known that patients with Franceschetti syndrome and Pierre Robin sequence have increased risk for developing airway obstruction following palatoplasty. This phenomenon can be explained as due to shallow nasopharyngeal airway and insufficient maxillofacial growth at the time of repair.[3] However, massive swelling of tongue following palatoplasty is rarely reported in literature[4] and could be a source of great concern and challenge to the anesthesiologist. There are case reports of massive tongue edema following other procedures too.[5,6] Much of the literature suggests that prolonged duration of surgery and long-term oral intubation has a direct correlation with increased incidents of tongue swelling and airway related complications.[4,7]

Case Report

A 13-month-old female baby weighing 10 kg with no previous history of any significant medical problem was taken up for palatoplasty. She had an uneventful Cleft Lip procedure at the age of 3 months. Basline investigations showed hemoglobin as 12.5 g.dl/1. She was premedicated with Midazolam syrup (5 mg) along with 0.4 mg atropine orally 1 h before procedure. After sedation anesthesia was induced with sevofluarne in oxygen. Atracurium was used to facilitate tracheal intubation using 4.5 non cuffed (Ring, Adair, and Elwyn) tube. Anesthesia was maintained on O_2, Nitrous Oxide and sevoflurane with atracurium as muscle relaxant. Electro CardioGram, non-invasive blood pressure, oxygen saturation, capnography, precordial stethoscope and airway pressure (attached to Fabius GS machine) were monitored. Palatoplasty was done by Bardach technique and the procedure lasted for 220 min. At the end of the procedure, after removing the mouth gag the concerned anesthesiologist noticed swelling in the tongue, which was still progressing. Tongue sutures were *in situ*, which is a standard practice in our institution. There was no excessive bleeding. Hemodynamic status of the child was within acceptable limits, bilateral air entry was good. Dexamethazone 2 mg IV bolus was given, arterial oxygen saturation was satisfactory. Child was retained in Operation theatre and extubated with re-intubation and tracheotomy setup on the stand by. Gentle laryngoscopy was done before and after extubation to suction out secretions and asses airways. The decision for trial extubation was made after observing the child on spontaneous with endotracheal tube for 30 min when the child showed no evidence of respiratory distress and maintained adequate oxygen saturation. Child was kept in prone position with head turned to one side and oxygen was given by mask. After observing the child for another couple of hours in the operation theatre, she was shifted to post-operative ward and kept in propped up position for the next 48 h, where she was closely monitored clinically for any

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evidence of airway obstruction in addition to continuous pulse oxymetry. Steroids (Dexamethasone 1 mg IV once in 6 h) and oxygen mask were continued in post-operative ward. Child started taking glucose water after 6 h. Swelling reduced gradually in size and tongue regained near normal proportion by 3rd post-operative day.

Following this we had another similar experience with an 18-month-old baby, which was managed along similar lines and had an uneventful recovery. Duration of procedure and anesthesia technique was similar to the previous case.

Discussion

Since it was our first encounter with macroglossia following palatoplasty, a thorough literature search was under taken. Sporadic reports of similar episodes were noted to cleft palate repair[1,3,4,8-11] Even in the reported cases, most of them were in Franceschetti's syndrome and Pierre Robin sequence where, the surgical intervention is more demanding due to micrognathia and glossoptosis and to have a proper surgical access surgeon injudiciously applies more forceful and lengthy tongue retraction.[3]

Bell *et al*. 1998 suggested excessive pressure exerted on the base of tongue by the retractor producing glossal hematoma, ischemic necrosis of tongue muscles, venous stasis or lymphedema[3,9] Hyperextension of the head and Tredelenberg position[12] may also contribute to impaired arterial flow and decreased venous drainage of the tongue.[1,3,12] Extreme Tredelenberg is highly detrimental especially when combined with high retractor pressure in prolonged surgery. Most of the authors believe that lingual edema is time dependent and Lee and Kingston suggest periodic release of the mouth gag to prevent prolonged ischemia of the tongue.[1,13] Other suggested mechanisms of tongue swelling are trauma, allergy, infection and massive fluid load perioperatively.[7]

Massive lingual swelling contributes to airway compromise. For the safety of child it is important to maintain airway patency once they develop tongue edema. Twigg *et al*. suggest to attempt extubation only after resolving tongue edema.[13] Occlusion of the posterior oropharyngeal space in palatoplasty may cause delayed acclimatization by the child. Chan *et al*. suggest inserting nasopharyngeal airway of appropriate size under the direct vision by the surgeon before extubation.[1] As part of our institutional protocol, we routinely put tongue sutures.

Conclusions

We present two cases of massive tongue edema following palatoplasty, which were satisfactory managed by delayed extubation, propped up position, steroids and prolonged Intensive care monitoring. In both scenarios, swelling came down drastically by 72 h post-operatively. Neither of the two required further airway intervention. We believe the mechanism is due to cumulative effect of prolonged compression by Killner Dott mouth gag, extreme Trendelenberg position and hyperextension of neck. We suggest periodic release of mouth gag in prolonged procedures and inserting nasopharyngeal airway of proper size in the unfortunate event of similar episodes in future.

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