

Anesthetic challenges in managing a case of type III laryngo-tracheo-esophageal cleft

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Abstract

Laryngo-tracheo-esophageal cleft (LTEC) is a rare congenital anomaly characterized by failure of fusion of the cricoid cartilage posteriorly and incomplete development of the tracheo-esophageal septum. Securing the airway during anesthesia in patients with LTEC, especially in the severe forms is a challenge. We describe the anesthetic management and the airway challenges in a neonate with type III LTEC who underwent bronchoscopy and repair of LTEC.

Key words: Aspiration pneumonia, bronchoscopy, cleft, difficult airway, larynx

Introduction

Congenital laryngeal cleft, first reported by Richter in 1792, is a rare developmental disorder of upper airway which accounts for 0.3% to 0.5% of all congenital anomalies of the larynx.^[1,2] It demands greater skills and high clinical judgment to diagnose as well as to manage airway perioperatively in patients with congenital laryngeal cleft. Aspiration pneumonia and other postoperative complications are more in severe forms of laryngeal cleft.^[1-3]

Case Report

A 2.8 kg, 4-day-old boy, the second child of non-consanguineous parents, was referred to our institute for the management of respiratory distress and septicemia. He was born by normal vaginal delivery after an uneventful antenatal period, and was mildly depressed at birth with Apgar score of 1 and 6 at 1 and 5 minutes respectively. He was resuscitated with bag and mask to which he responded and then was shifted to the

intensive care unit on oxygen hood. His condition improved slowly and he was weaned off oxygen over 72 hours. He was administered expressed breast milk feeds on day four but developed choking, aspiration of feeds and respiratory distress.

On arrival, the baby was floppy, had a weak cry and shallow breathing. The neonate's trachea was intubated with a 3.5 mm endotracheal tube (ETT) and fixed at a length of 9 cm. Lungs were ventilated with help of a pressure controlled ventilator with FiO₂ 0.7 and moderate pressures. Chest X-ray showed aspiration pneumonitis in the right upper lobe. Patient was started on second line antibiotics, Cefotaxim and Amikacin. Blood culture grew *Klebsiella pneumoniae* and antibiotics were changed to meropenem according to the sensitivity. Patient developed multiple episodes of desaturations associated with dislodgement of ETT to the esophagus. It was observed that when ETT was pushed further in the trachea, the baby ventilated better and there was no ETT dislodgement [Figure 1]. This raised the possibility of a congenital anomaly of the airway.

Further evaluation confirmed a small patent ductus arteriosus and bilateral retinal coloboma. Computerized tomography (CT) scan chest showed evidence of a defect in posterior larynx at the level of vocal cords and inflammatory changes in the left upper lingual and right lower lobe. A diagnostic bronchoscopy was done under general anesthesia to confirm the diagnosis. Patient was given fentanyl 4mcg and atropine 0.4mg intravenous (IV). Anesthesia was induced by inhalation of air, oxygen and sevoflurane. Muscle relaxation was achieved with atracurium 1 mg IV. Rigid bronchoscopy with a 2.5 mm sized scope showed a large LTEC extending up to the middle

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of the trachea (Type III LTEC) [Figure 2]. The bronchi were normal.

Repair of cleft was performed after two days under general anesthesia. Before the surgical repair, a large bore (10F) nasogastric tube was placed and the ETT was re-enforced at the angle of the mouth with a stay suture. Monitoring included pulse oximetry, capnography, electrocardiogram, temperature, noninvasive blood pressure and urine output. There was a 2.5 cm long posterior laryngeal cleft extending from just below the vocal cords till the suprasternal notch with a common tracheo-esophageal passage. The cleft was repaired by placing long strips of sternomastoid muscle between trachea and esophagus. The trachea was repaired over the 3.5 mm ETT. Esophagus was repaired over the nasogastric tube [Figure 3]. We did not encounter any tube displacement during surgery. A tracheostomy tube was placed through the third tracheal ring. The ETT was pulled up above the tracheostomy and was fixed. It was kept as a stent for one week. Analgesia was provided by intermittent IV doses of fentanyl. The surgery was completed uneventfully over 2 hours. The oxygen saturation and hemodynamic parameters were stable during this period. Blood loss was minimal and 25ml of 5% dextrose with half normal saline was given as infusion during surgery.

Postoperatively, the baby was stable and total parenteral nutrition followed by expressed breast milk feeds were started via the nasogastric tube. The baby was weaned off ventilator after 4 days and given oxygen supplementation via T-piece over the tracheostomy. Two weeks after the surgery, patient developed copious drooling of secretions. Bronchoscopy under general anesthesia ruled out the possibility of a fistula but endoscopy revealed esophageal stricture which was dilated using Savary-Gilliard dilators and feeds were resumed. Patient had swallowing difficulties due to pharyngo-esophageal dysfunction postoperatively. Later patient developed significant aspiration of feed and respiratory distress which required re-intubation; however, he deteriorated further and developed disseminated intravascular coagulation with sepsis and died at two and a half months of age, despite supportive measures.

Discussion

Laryngo-tracheo-esophageal cleft (LTEC) is a rare pathology with a male to female ratio 5:3.^[1] An LTEC occurs when there is incomplete fusion of the posterior lamina of the cricoid cartilage and the tracheo-esophageal septum. The Benjamin Inglis classification of LTEC is the simplest and most commonly used [Table 1].^[1,2]



Figure 1: Endotracheal Tube is pushed up to the Carina during ventilation and is supported through a large tube placed in the esophagus

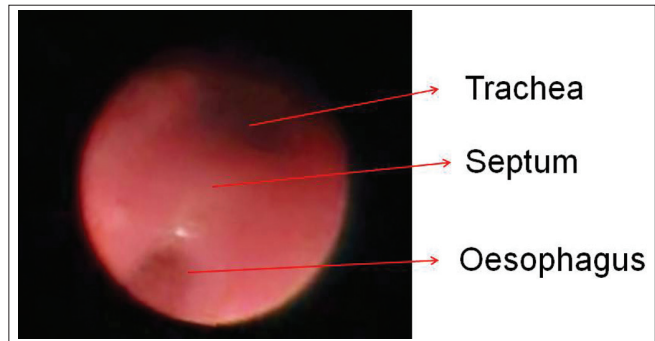


Figure 2: Rigid bronchoscopy showing a large tracheoesophageal cleft extending up to the middle of trachea (Type III LTEC)

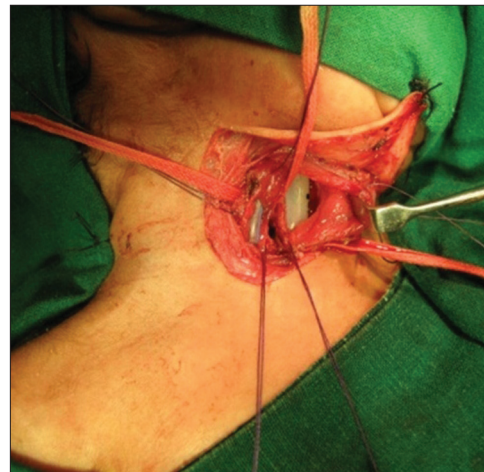


Figure 3: Trachea repaired over the ETT and the esophagus over the nasogastric tube. Strips of sternomastoid muscle placed between the trachea and esophagus

Our patient had feeding difficulties, respiratory distress and aspiration. The repeated slipping of the orotracheal tube into the esophagus, alerted us to the possibility of a congenital anomaly of the airway. Microlaryngoscopy and bronchoscopy are the definitive means of diagnosing laryngeal cleft and assessing adjacent native tissue for reconstruction.^[2]

Table 1: Benjamin Inglis classification of laryngo-tracheo-esophageal cleft

Type I	Interarytenoid cleft of the soft tissue without extension inferiorly into the cricoid cartilage.
Type II	Cleft extending into the cricoid cartilage but not through the inferior lamina of the cricoid.
Type III	Extends through the entire posterior lamina of the cricoid with or without extension into the cervical trachea.
Type IV	Extends into the thoracic trachea and may extend down to the carina.

There are reports of repeated accidental extubations, esophageal intubation and posterior displacement of ETT into the esophagus especially in severe forms of LTEC.^[1] Ruder managed the airway of type III LTEC with a tracheal tube and a Foleys catheter passed into the stomach and the inflated balloon retracted into the gastro-esophageal junction.^[4] The Foleys catheter provided a seal in preventing gas from entering the stomach during positive pressure ventilation, besides helping to maintain an anterior position of the ETT and preventing it from being displaced posteriorly into the esophagus. We found that a large bore nasogastric tube (10 F) passed into the esophagus was useful in decompressing the stomach and keeping the ETT near the carina prevented its displacement.

Tracheostomy,^[1-3] two tracheal tubes into each main stem bronchus,^[3] custom-made bifurcated ETT,^[1,5] endobronchial intubation, one lung ventilation,^[6] pediatric bilumen tube,^[7] laryngeal mask airway,^[8] ECMO and cardiopulmonary bypass^[9] have been used for airway management of LTEC. Maintaining the airway and prevention of aspiration during reconstruction is also difficult.^[3] An ETT anchored at the angle of mouth, a large bore nasogastric tube and close vigilance to detect and rectify any tube displacement helped us to tide over the intraoperative period. Tracheostomy after the reconstruction helped to further manage the airway patency during the recovery period.

Nutritional status has to be maintained for the wound healing and neonatal growth.^[1] Gastric transection with double gastrostomies, gastrostomy, and total parenteral nutrition has been reported.^[1] Gastro-esophageal reflux leads to aspiration pneumonia, sepsis and increased mortality. Aggressive antireflux protocols such as prokinetics, reverse Trendelenberg positioning, protection of the airway were tried on our baby. Proton pump inhibitors, histamine type 2

antagonists, proximal gastric drainage, Nissen fundoplication, high gastric transection, double gastrostomy have also been advocated.^[1,2] Other complications include tracheo-esophageal fistula, laryngeal nerve injury, granulation tissue formation, tracheomalacia and dysmotility of reconstructed esophagus.^[2]

LTEC requires high clinical judgment to diagnose, and team approach and planning is essential for its successful management.

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