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Anesthetic Considerations in Rare Type of Congenital Lobar Emphysema of Left Lower Lung Lobe

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Running Title: CONGENITAL LOBAR EMPHYSEMA OF LEFT LOWER LUNG

***Abstract**: Although congenital lobar emphysema (CLE) is a rare lung disease, it can cause severe respiratory distress in the newborns due to compression atelectasis. It is most often associated with mediastinal shift and subsequent hypoxia. Its incidence is 1/20,000-30,000 live births with male to female ratio of 3:1. The prenatal incidence is unknown due to diagnostic difficulties with prenatal ultrasonography (USG). We have reported a case of 17 days old male neonate posted for left lower lung lobectomy, who was presented with the sudden onset of respiratory distress related to CLE affecting the left lower lung lobe, a rarest form of CLE.*

***Key-words**:Congenital lobar emphysema, positive pressure ventilation, intercostal nerve block.*

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I. Introduction

Congenital lobar emphysema (CLE) is a rare developmental lung malformation characterized by hyperinflation of one or more pulmonary lobes leading to respiratory distress in newborns.[1] One-third among them are symptomatic at birth and nearly all of them are diagnosed in the first 6 months of life. Anesthetic management for surgical excision of the affected lobe of the lung in these patients is quite challenging in terms of their age group and the severity of cardiorespiratory distress.[2] The maintenance of spontaneous respiration may not be successful and the institution of positive pressure ventilation (PPV) has the potential of worsening the underlying condition.[3]

Case History: A 17 days old male neonate, weighing 2.9 kg, was brought by the parents with a history of respiratory distress and rapid breathing rate. He was antenatally diagnosed as congenital diaphragmatic hernia and had peripheral cyanosis at birth. He was breast-fed and suffered no other medical problems.

On examination, the neonate was pale with capillary refilling time (CRT) >5 sec and SpO_2-60% at room air with tachypnea (68-70 breaths/min), intercostal retractions and tachycardia (180 beats/min). On chest auscultation, air entry was reduced bilaterally, more so on left side and heart sounds were heard on right side. Chest radiography demonstrated emphysematous changes (hyperlucency) in left lung with marked right sided mediastinal shift leading to compression atelectasis of right lung [Figure1]. Routine hematological and biochemical investigations were normal. A clinical diagnosis of CLE was made and treatment started in the neonatal intensive care unit (NICU) with immediate endotracheal intubation using single lumen uncuffed endotracheal tube (size 3.5 mm) followed by gentle manual assisted ventilation resulting in increase in SpO_2-98% with oxygen @ 5 litres/min, intravenous (iv) antibiotics and bronchodilators. Fluid resuscitation was commenced with 20 ml/kg N/5 dextrose solution that restored CRT to normal, although tachypnea persisted.

Computed tomography (CT) scan confirmed the diagnosis of CLE of left lower lobe along with compression of left upper lobe and right lung with marked right sided mediastinal shift [Figure 2]. Posterolateral thoracotomy followed by left lower lobectomy was planned under general anesthesia. The child was kept fasting according to standard guidelines. In the operation theatre, American Society of Anesthesiologists (ASA) basic standard monitoring was instituted which included pulse oximetry (SpO_2), electrocardiogram (ECG), non-invasive blood pressure (NIBP), and temperature monitoring. The hemodynamic parameters recorded were HR-174/min, NIBP- 62/42 mm Hg and SpO_2-100% with oxygen. Intravenous access was checked and iv ringer's lactate started. Pre-operative arterial blood gas analysis (ABG) demonstrated pH-7.30, PaO_2-68 mm Hg and $PaCO_2$-54 mm Hg. Injection glycopyrrolate (0.03 mg) and fentanyl (6 mcg)were given intravenously. Patient was induced with sevoflurane 2.5% in 100% oxygen and endotracheal tube was repositioned just above the carina and confirmed by auscultation and end-tidal CO_2 ($EtCO_2$). Patient was gently ventilated manually (low tidal volume) using Jackson-Rees circuit to avoid hyperinflation of left lower lobe further aggravating the mediastinal shift and thus cardiovascular collapse.

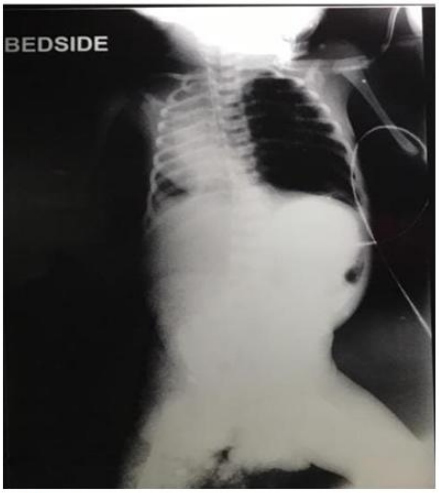

Figure 1

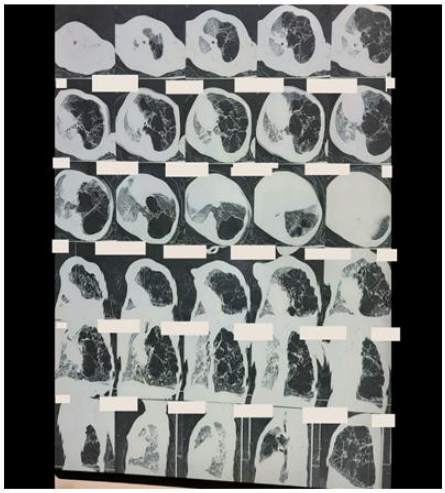

Figure 2

The child was placed in right lateral position followed by intercostal nerve block with 2.5 ml of 0.25% bupivacaine in left fourth intercostal space. Gentle assisted manual ventilation was continued until the emphysematous lobe popped out through left thoracotomy incision [Figure 3].Anesthesia was maintained with oxygen, sevoflurane (1-2%), intermittent iv boluses of atracurium with intermittent positive pressure ventilation (IPPV) using pressure-controlled mode keeping peak inspiratory pressure around 20 cm H_2O. The left lower lobectomy was performed by the cardiothoracic surgeon. After this resection, left upper lung was expanded using manual recruitment maneuver before closure and presence of stump leak was checked with saline. The procedure lasted for three hours with stable hemodynamic parameters. An intercostal drain (ICD) was placed on the left side of the chest. Fluids (Ringer lactate and dextrose 5%) and blood were replaced judiciously as per losses. Residual neuromuscular blockade was reversed with iv glycopyrrolate 0.02 mg and neostigmine 0.15 mg. A paracetamol suppository (80 mg) was inserted before shifting the patient.

The patient was shifted to NICU on ambu bag for elective ventilation for 24 h. Post-operative chest radiograph done on next day revealed expanded right lung as well as remaining left lung and normal position of mediastinum [Figure 4]. As post-operative serial ABG reported to be in normal limits, child was gradually weaned off the ventilator followed by extubation onfirst post-operative day and was kept on nasal bubble continuous positive airway pressure (CPAP) for next two days. ICD was removed at 72 hours. Post-operative period remained uneventful and the infant was discharged on day eighteen.

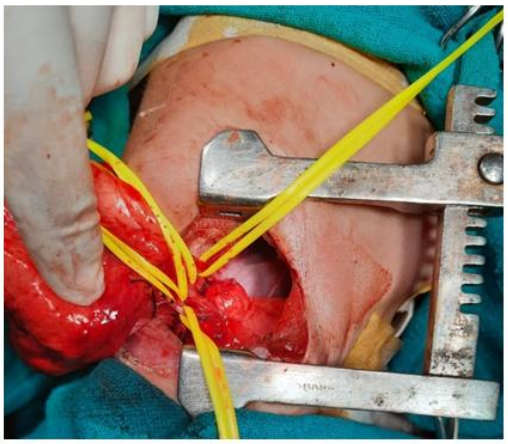

Figure 3

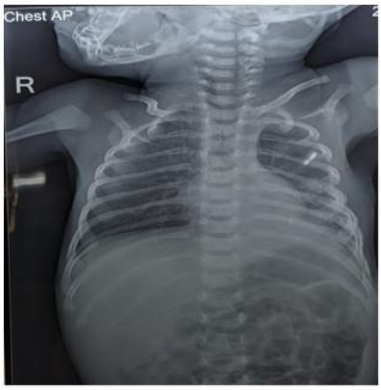

Figure 4

II. Discussion

CLE is a rare cause of sudden respiratory distress in infants. It usually involves the left upper lobe (43%), followed by right middle lobe (32%) and right upper lobe (21%). Lower lobe involvement (2%) is the rarest form as present in our case.[4] The cause of obstruction is unknown, although literature revealed evidences of deficient and disordered bronchial cartilage. CLE usually appears clinically in the first 6 months of life with tachycardia, tachypnea, and chest retractions.

Induction of anesthesia is challenging in these children as excessive crying and struggling can increase the amount of trapped gas due to ball valve effect and lowers pulmonary reserve.[3] On the contrary,PPV leads to over-inflation of the emphysematous lobe resulting in mediastinal shift, hemodynamic compromise and even cardiac arrest. Thus, smooth inhalational induction must be preferred technique of anesthesia in these patients.[5,6] During induction, nitrous oxide should be avoided because it can increase the mediastinal shift in the lung owing to its rapid spread in the closed cavity.

The key anesthetic concern in such patients is to avoid overdistension of the emphysematous lobe by avoiding PPV and providing gentle assisted manual ventilation when required, while maintaining airway pressure at 20-25 cm H_2O until thoracotomy.[7] Prabhu and Joseph[3] used mild IPPV following muscle sparing. During this phase, monitoring of vital parameters is must.Single-lumen endotracheal tubes are usually preferred for tracheal intubation as double-lumen tubes are not commercially available in this age group. Moreover, in young infants, lung isolation is technically difficult and is not widely applied. In addition to this, the correct placement of bronchial blockers is a challenging task.

Unlike adults, the physiological principles of ventilation and perfusion for thoracic surgery in lateral decubitus position do not apply in these patients. In infants with unilateral lung involvement, oxygenation does not improve with the diseased lung being non-dependent and healthy lung being dependent, due to their more lung compliance. The FRC is also closer to the residual volume and airway closure becomes more likely in the dependent lung. The advantage of the abdominal hydrostatic pressure gradient to the dependent diaphragm is lesser in infants. The favorable increase in perfusion to the dependent lung is also lesser due to their small size. All these considerations leads to their decreased ability to tolerate one-lung ventilation (OLV).[3]There are reported cases of isolated healthy lung endobronchial intubation, until lobectomy is performed but it may not be sufficient to maintain adequate oxygenation in all the patients.

Single-lumen tube may expose the affected lobe to positive pressure. This may result in excessive hyperinflation and cardiac compression. Therefore, the surgeon should open the thoracic cavity as soon as possible after induction of anesthesia. When the thoracic cavity is opened, the emphysematous lobe herniates out of the thorax reducing the compression of mediastinal structures.

High-frequency ventilation is another ventilation strategy described by Goto et al.[8] It allows the use of low ventilation pressures and hence achieving betteroperating conditions for surgeons.

The preferred options for maintenance of anesthesia are inhalational agents along with intermittent boluses of neuromuscular blocking agents. Pre-emptive analgesia in the form of infiltration of local anesthetic along the line of incision, blunts the incision response thereby decreasing the requirement of inhalational agents and facilitates rapid recovery.

We preferred intercostal nerve block for intraoperative and postoperative analgesia as it can be easily administered. Longer acting opioids should be avoided due to the risk of post-operative respiratory depression. Caudal thoracic epidural catheter, described by Raghavendra et al,[9] provides stable hemodynamics along with excellent analgesia without any risk of post-operative respiratory depression but there is risk of kinking of catheter.

Although extubation is preferred at the end of surgery but we had shifted the baby to NICU for elective ventilation till adequate expansion of the healthy lung was achieved. Lung protective strategies should be continued, if elective ventilation is needed.[2] Adequate analgesia is warranted for the infant to breathe spontaneously.

In conclusion, anesthetic management of these patients poses various challenges for an anesthesiologist from induction till extubation. An infant presenting in emergency with progressively increasing respiratory distress should be viewed with a suspicion for CLE. Its early recognition and surgical intervention can be life-saving. Even today, despite advanced diagnostic techniques, pitfalls in diagnosis and management are not uncommon.

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Figures' Legends:

Figure 1: Pre-operative X-ray chest (PA view) showing emphysematous changes in left lung with compression atelectasis of right lung with right sided mediastinal shift

Figure 2: Pre-operative Computed tomography (CT) scan chest showing emphysematous left lower lung lobe along with compression atelectasis of left upper lobe and right lung and marked right sided mediastinal shift

Figure 3: Posterolateral thoracotomy showing emphysematous left lower lobe resection

Figure4:Post-operative X-ray chest showing re-expansion of right lung and left upper lung with normal mediastinum

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