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Case Report

Surgical cause of respiratory distress in a newborn

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ABSTRACT

Common causes of respiratory distress in a term infant include transient tachypnoea of newborn, sepsis/congenital pneumonia and congenital heart diseases. Surgical causes of distress in a newborn are rare and should be considered when other causes have been ruled out. We report a case of an early term female neonate who developed respiratory distress immediately after birth which was gradually progressive and required respiratory support. On evaluation with chest radiography and CT, a diagnosis of congenital lobar emphysema (CLE) was made. Immediate surgical resection was planned and done on day 8 of life. Post operatively she had no respiratory distress and discharged within a week. Baby is now healthy with normal growth and development. Half of the cases of CLE are diagnosed within the first 6 months of life but very few cases present with respiratory distress since birth. Timely diagnosis and early surgical excision in neonates <2 months improve the outcome, as seen in our case.

Keywords: Respiratory distress, Hyperlucency, CLE

INTRODUCTION

CLE is one of the rarest causes of respiratory distress in newborns.[1] During the third week of gestation, the development of the respiratory system begins and aberrations during any stages of lung and airway embryogenesis may cause parenchymal lung malformations such as CLE.[2] Congenital alveolar overdistension, congenital large hyperlucent lobe, and congenital lobar overinflation are other terms synonymous with CLE. It is more common in males, and the male to female ratio is 3:1.[3] The incidence is 1/20,000-30,000 live births.[4] Left upper lobe is most frequently affected (42%), followed by right middle lobe (35%) and right upper lobe (21%) involvement. Lower lobe involvement (1%) is the rarest form.[5,6] One-third of cases are symptomatic at birth and nearly all of them are diagnosed in the first 6 months of life.[7-9] Diagnosis is by chest X-ray and CT, although initial radiographs in the immediate newborn period may appear like pneumonia, thus delaying the diagnosis.[10,11] We are reporting a newborn presenting with respiratory distress soon after delivery, which progressively worsened requiring surgical excision of the affected lobe.

CASE REPORT

A female baby is born at 37 4/7 weeks gestation by emergency LSCS I/v/o maternal request to a 25 years old gravida-4, abortions-2 and ectopic-1 woman. Present pregnancy is IVF conception and born to non-consanguineous parents with marital life of 5yrs. She received adequate prenatal care and had an uneventful antenatal course. No h/o GDM PIH, poly/oligohydramnios and leaking PV. Level II scan done at 20 weeks of GA was normal. Birth weight was 3.198 kgs and baby cried immediately after birth. The Apgar scores were 8/10, 9/10 at 1 min, 5 min respectively and did not require any form of resuscitation. Immediately after delivery baby developed mild tachypnea (Downe's score-2/10) with preductal oxygen saturation of 88% even after 10 minutes, hence was shifted to NICU for further care.

Course of illness

Respiratory distress was increasing hence baby was connected to high flow nasal cannula (HFNC) with FiO_2-0.3 and flow-6 litres/min. Initial chest radiograph was normal. Suspecting sepsis, septic screening and blood culture was sent and started on empirical antibiotics. Septic screening and blood culture was negative, hence antibiotics were given for 5 days and stopped. Initial differential diagnosis considered were-Transient tachypnea of newborn, congenital pneumonia, congenital heart diseases, persistent pulmonary hypertension of newborn, air leak syndromes and congenital malformations of lung

Respiratory distress was worsening with Downe's score-4/10. FiO_2 requirement also increased to 0.4. Despite increasing respiratory distress, baby was active and tolerating full feeds. On examination heart sounds were more prominent on the right side of precordium. Repeat chest radiography was done and was suggestive of hyperlucency and hyperinflation of left upper lobe with herniation to the right side and shift of superior mediastinal structures with compression of right lung (Figure 1), suggestive of CLE. Findings were then confirmed by CT chest (Figure 2).

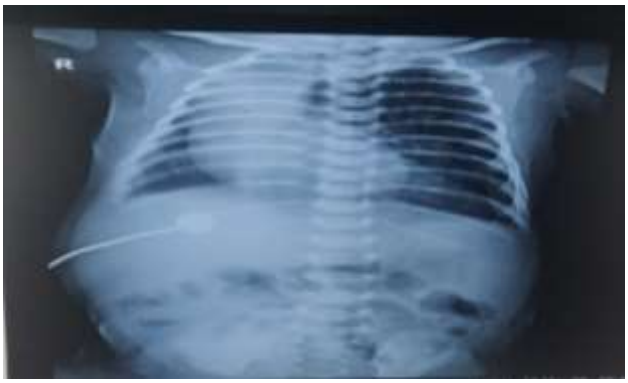

Figure 1: Chest radiography of hyperlucent left lung fields with mediastinal shift and herniation of left upper lobe to the right.

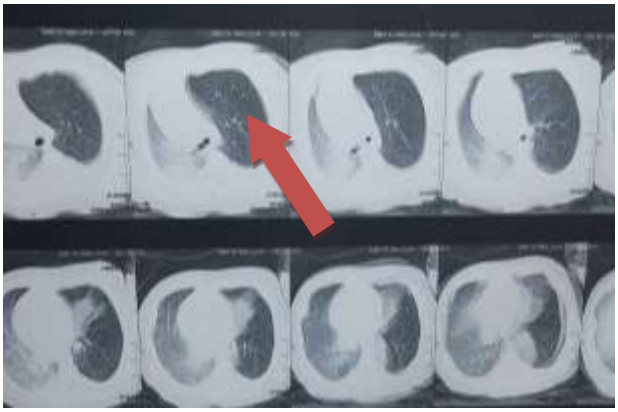

Figure 2: CT chest of hyperinflated left lung with mediastinal shift.

2D echo was done to rule out congenital heart diseases and was normal. In view of worsening respiratory distress requiring persistent respiratory support pediatric surgery opinion was sought and planned for surgery.

Parents were counseled regarding diagnosis and management. Under general anesthesia, thoracotomy with left upper lobectomy was done on day 8 of life (Figure 3 and 4).

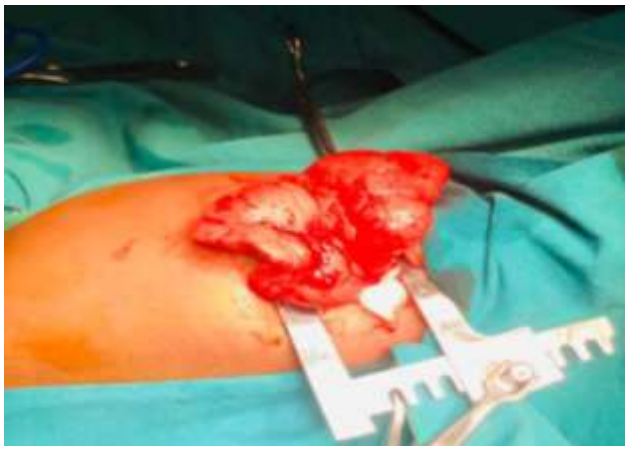

Figure 3: Emphysematous left lobe popped out through left thoracotomy.

Figure 4: Resected left upper lobe.

Intraoperatively emphysematous left upper lobe popped out through left thoracotomy. Specimen microscopy showed lung tissue with distortion of the architecture and the alveoli appeared dilated with the break in the wall of the alveoli. Interstitium showed focal fibrosis and capillary proliferation. There was no evidence of inflammation or dysplasia

Post operatively baby received mechanical ventilation for 24 hours followed by 48 hours of HFNC (Figure 5). By post-operative day 3 baby was in room air and full feeds. Now, baby is 15 months old with normal growth and development.

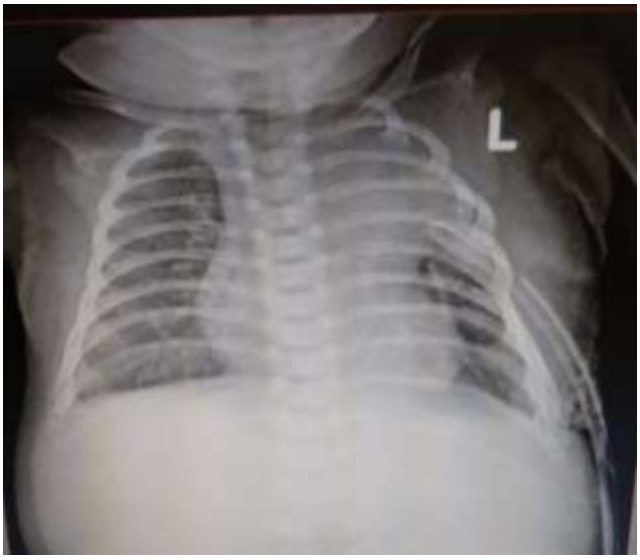

Figure 5: Chest radiography post left upper lobectomy.

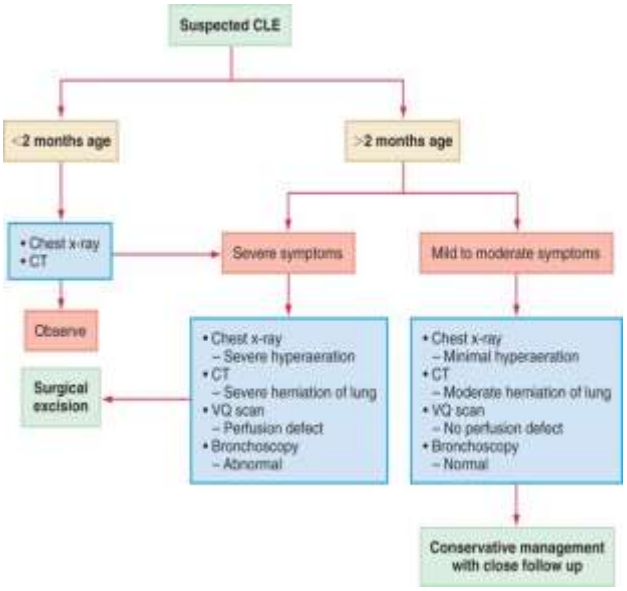

Figure 6: Management of CLE.

DISCUSSION

CLE is a rare developmental lung malformation mechanisms underlying CLE is intrinsic or extrinsic obstruction of the airways resulting in air trapping, overdistension and subsequent emphysema. Multiple mechanisms have been described including dysplastic bronchial cartilage, inspissated mucus, aberrant cardiopulmonary vasculature and infection.[12] Half of the cases no underlying pathology is identified. CLE can be detected on fetal ultrasonography, although it is difficult to make correct diagnosis, as in our case where all antenatal scans were normal.[13] Clinical reports have described the appearance of lesion by ultrasound as cystic and/or echogenic with mediastinal shift present and subsequent regression with advancing gestation. These lesions are suspected to be congenital pulmonary airway malformation/ bronchopulmonary sequestration (CPAM/BPS). Fetal MRI can be helpful in characterising these lesions although it is not diagnostic.[13]

Majority of these children present with cyanosis, respiratory distress and recurrent pulmonary infections in first 6months of life, with 13 out of 33 (39%) were symptomatic on the first day of life in a single case series, similar to our case.[14] Severity of symptoms depends on the size of the affected lobe, extent of mediastinal shift and compression of surrounding tissues.

Chest radiography demonstrates a hyperinflated lung (transitioning from a fluid filled to air filled lung) over initial post-natal days with compression of other airways of lung and mediastinal shift. Less severe cases, CT chest and bronchoscopy is helpful in managing the decision.[15]

Treatment of CLE depends on the severity of symptoms (Figure 6).

Conservative and supportive management should be considered in milder cases. Surgical resection of the affected area should be considered in patients with life-threatening progressive pulmonary insufficiency from compression of the adjacent normal lung. Once the emphysematous area is resected there is compensatory lung growth on the affected side and the long-term prognosis is excellent.[16]

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