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Case report

Early neonatal lobectomy for ILE; is it feasible?☆

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ABSTRACT

Introduction: Congenital lobar emphysema (CLE) also called congenital lobar overinflation and infantile lobar emphysema is a rare congenital anomaly of lung often presents in the neonatal period, with hyperinflation of one or more pulmonary lobes.
Importance: Author is reporting here an uneventfully done earliest ever neonatal lobectomy for ILE.

1. Introduction

Nelson RL in 1932 first described congenital lobar emphysema (CLE). Its prevalence is 1 in 20,000 to 1 in 30,000. Though its aetiology is not well understood yet, CLE is a rare condition causing over inflation of lung lobes, compressing the ipsilateral, as well as the contra lateral lung; causing acute respiratory distress. Often detected in neonates or young infants but, some even present late at childhood. The upper lobes are affected in over 95% of cases, with multiple lobes in about 15% of cases.

2. Clinical presentation

A day 1 male neonate, born full term by lower segment caesarean section with a birth weight of 2.68 kg was noticed to have respiratory distress immediately after birth. Chest radiography revealed hyperlucent left lung with mediastinal shift; contrast enhanced computed tomography (CECT) showed left hyperinflated lung with gross mediastinal shift (Fig. 1A, B, C). Baby was clinically stable requiring oxygen of 2 l per minute without ventilator support. Having normal haematological parameters as well as reverse transcriptase polymerase chain reaction (RT PCR) test negative for severe acute respiratory syndrome corona virus 2 (SARS-CoV2), baby was taken up for left lobectomy under general anaesthesia.

Induction done with thiopenone, fentanyl, propofol, atracurim and intubed with 3 mm micro cuffed endotracheal tube; maintained with oxygen, air, isoflurane and intermittent positive pressure ventilation (IPPV) with bilateral lungs kept ventilated throughout the procedure. Left posterolateral thoracotomy and left upper lobe ILE was exposed, its supplying vessels and bronchus were ligated at the hilar level, cut and resected ILE sent for histopathology, which revealed ILE showing massive distension of alveoli without tissue destruction (Fig. 2E,F,G,H, I).

Post operatively baby improved uneventfully without any ventilator support and hence discharged on post operative day 5.

3. Clinical discussion

Infantile lobar emphysema (ILE) occurs due to over distension of a pulmonary lobe, result of a partial postnatal obstruction of the associated bronchus with air trapping. Hence baby presents with respiratory distress, 50% in the first week and in 80% first 6 months of life [1,2].

Imaging studies usually show hyperlucent lobe or a lobe of normal lucency that occupies a disproportionate part of the hemithorax, with mediastinal shift and compression of the uninvolved lobes. Associated anomalies are present in 5% to 40% of patients, with 70% of these anomalies are cardiovascular. The supplying bronchus may reveal stenosis, atresia, or intrinsic obstruction, although many cases may be caused by extrinsic bronchial obstruction, so the bronchus may appear grossly normal [1–3].

Microscopically the lung shows a uniform over distension of apparently normally developed acini with alveolar saccules and alveoli 3 to 10 times the normal size (Fig. 2H, I) [1,3,4].

Congenital lobar emphysema (CLE) may be clinically confused with tension pneumothorax. Hence the chest tube insertion may further worsen the distress and lead to further lung injury. In CLE the pulmonary vessels extend to the periphery of the hyper inflated lobe and there is no visualization of a pleural line compared to pneumothorax [2,3].

The differential diagnosis includes congenital cystic adenomatoid malformation, sequestration, bronchogenic cyst, unilateral hyper lucent

☆ Ethical standard statement-parents gave informed consent to the publication of the case report.

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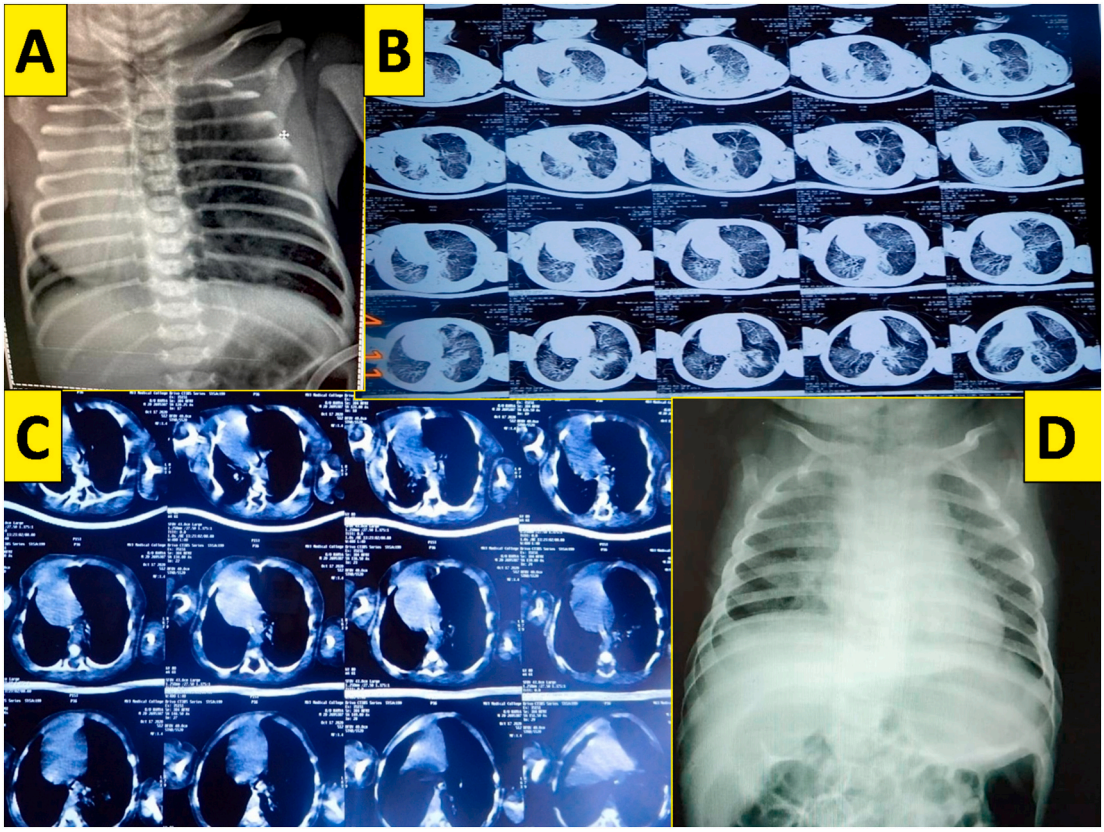

Fig. 1. A-Chest radiography of left infantile lobar emphysema with left emphysema along with mediastinal shift., B and C-computed tomographic images of chest showing left upper lobe ILE, D-post operative chest radiography.

lung syndrome, and pulmonary interstitial emphysema.

Though conservative management has supported by some studies due to its potentially reversible status in few cases which is very rare event but difficult to predict, hence surgery is the most definitive and curative; either via segmentectomy or lobectomy [1–5]. Nevertheless symptomatic respiratory distress in a neonate needs definitive surgical management to avoid morbidity and mortality.

Though the literature is abundant with ILE with varied time for its definitive resection of the affected lobe, either by open or by minimally invasive approach, author is reporting the first ever, lobectomy done for a case of ILE on a post natal day 5 uneventfully; in a tertiary care neonatal and paediatric centre.

4. Conclusion

Having adequate facilities with normal haematological parameters, early neonatal lobectomy is feasible option in a tertiary care centre.

Provenance and peer review

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None.

Ethical approval

Yes taken.

Consent

Written informed consent was obtained from the parents for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Research registration

Not yet done.

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Dr Jayalaxmi Shripati Aihole has contributed sole to the artilce.

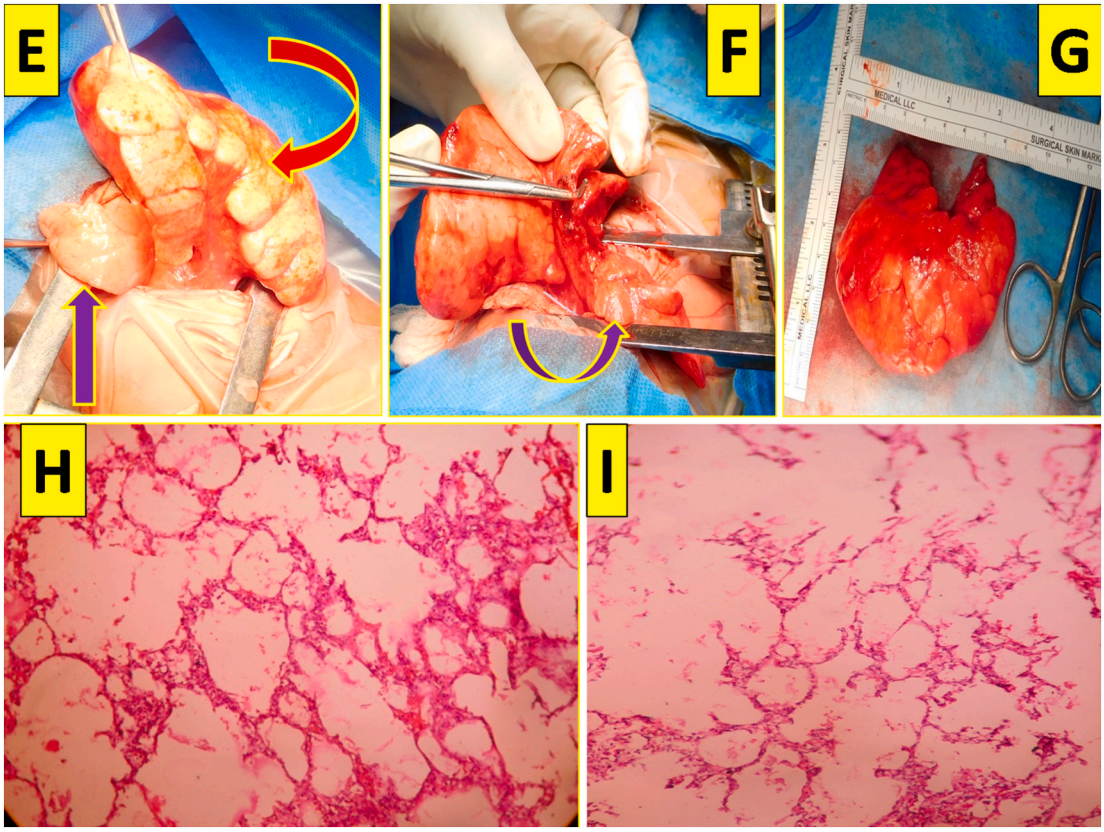

Fig. 2. E- Intra operative pictures—red curvilinear arrow –pointing at left upper lobe ILE. Purple straight arrow pointing at normal left lower lobe.
F- Hilar dissection of left upper lobe ILE. Purple curvilinear arrow pointing at left lower lobe.
G-Excised left upper lobe ILE.
H and I-Histopathological images Low power view10X—massive distension of alveoli without tissue destruction.

Declaration of competing interest

None.

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Manuscript has not been previously published in whole or in part or submitted elsewhere for review.

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