



Sequential Presentation of Bilateral Congenital Lobar Emphysema

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To the Editor: Congenital lobar emphysema (CLE) is a lung malformation, caused presumably by extrinsic or intrinsic airway obstruction and ball-valve effect [1]. CLE of two lobes is rare with only around 15 cases reported [2, 3].

A term baby boy presented on day 23 with severe respiratory distress and hypoxemia. Chest radiograph showed left lung hyperinflation and collapse of right upper lobe, which were worsening in serial radiographs despite invasive ventilation and nebulisation. HRCT chest showed left upper lobe CLE and partial collapse of other lobes. Left upper lobectomy was performed on day 27. Pathological examination of excised lobe suggested CLE, polyalveolar type.

Chest radiograph after 48 h of surgery showed hyperinflation of right middle lobe, which was worsening in subsequent radiographs. Repeat HRCT chest showed right middle lobe CLE. CT angiography and bronchoscopy were normal.

As baby improved clinically, expectant management was planned and baby was discharged. He presented again at 6 mo with respiratory distress, when right middle lobectomy was performed. On follow-up until one year, he had no respiratory distress and had normal growth and development.

Unlike previously reported cases of bilateral CLE where CLE of both lobes were identified simultaneously, our baby had sequential presentation. The first HRCT showed only left upper lobe CLE. Right middle lobe was collapsed probably due to the mass effect of left upper lobe. Right middle lobe started overinflating after removal of left upper lobe.

Lei et al. reported the only other case of sequential presentation of bilateral CLE [4]. The baby had right middle lobe CLE initially. CLE of left lower lobe developed 5 d after right middle lobectomy.

Since the presumed underlying pathology is airway malformation, involvement of bilateral airways and lungs is possible in CLE. We suggest that infants with unilateral CLE should be followed up until we ensure the other lobes are normal.

Declarations

Conflict of Interest None.

References

1. Cataneo DC, Rodrigues OR, Hasimoto EN, Schmidt AF Jr, Cataneo AJ. Congenital lobar emphysema: 30-year case series in two university hospitals. *J Bras Pneumol.* 2013;39:418–26.
2. Maiya S, Clarke JR, More B, Desai M, Parikh D. Bilateral congenital lobar emphysema: How should we proceed?. *Pediatr Surg Int.* 2005;21:659–61.
3. Zinaye A, Zeray A. Bilateral congenital lobar overinflation (CLO) - A rare presentation of an uncommon condition. *Radiol Case Rep.* 2022;17:3481–4.
4. Lei Q, Zeng W, Ju R. Congenital lobar emphysema in bilateral lung lobes: A case report. *Transl Pediatr.* 2020;9:266–71.

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