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Congenital Lobar Emphysema Mimics Foreign Body Aspiration in Infancy

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ABSTRACT

Congenital lobar emphysema (CLE) is a rare cystic lung malformation derived from the foregut and often presented in the neonatal period with a life-threatening respiratory distress. This rare entity may mimic other medical conditions of respiratory distress. Appropriate diagnosis and treatment of CLE could improve its prognosis. We described a case of 45-days male infant presented with an episode of respiratory distress which was wrongly interpreted as a result of foreign body (FB) aspiration. The findings of chest x-ray, multi-detector computed tomography (MDCT), and bronchoscopy were suggestive for CLE at the left upper lobe. Thoracotomy was performed for left upper lobectomy with uneventful postoperative course.

Keywords: *Congenital disorders, lungs, congenital lobar emphysema, infant, thoracic surgery*

INTRODUCTION

Congenital lobar emphysema (CLE) is a rare developmental anomaly of the lung which occurs in 1 case per 20000–30000 births [1].Most of the cases with CLE appear in the neonatal period as a hyperinflation of one or more pulmonary lobes [2]. The emphysematous lobe may compress the surrounding lobes and in severe cases it results in mediastinal shift [1].

The etiology of CLE is idiopathic in 50% of cases; however other intrinsic and extrinsic causes of air-trapping were suggested [3].Therefore, CLE should be differentiated from other causes of from bronchial obstruction (lymph nodes, vessels, masses, foreign body, or cysts). The left upper lobe is most commonly involved in cases with CLE followed by the right upper lobe and right middle lobe [1].

Early diagnosis and treatment of CLE improves outcome and reduces mortality. Surgical removal is the most common treatment choice with operative mortality in up to 13% of cases [4].

CASE PRESENTATION

A 45-days male infant presented with dyspnea and wheezy chest for 2 days. The infant was referred to our department of thoracic surgery from a pediatrician for a suspicion of foreign body (FB) aspiration. There was a history of acute upper respiratory tract infection at the age of 3 weeks which was resolved within 5 days after medical therapy. The baby was delivered vaginally at full-term withaverage body weight and usual post-natal course.

On physical examination, there was a respiratory distress, tachypnea (respiratory rate of 50/min), oxygen saturation (SaO2) of 88% on room air, and diminished air entry on the left side of the chest with scattered rhonchi. There were unremarkable results of laboratory tests with no abnormal findings or congenital cardiac anomalies on echocardiography.

On chest x-ray, there was hyper inflated left upper lung lobe with trans-mediastinal herniation to right side and increased its lucency and subsequent mediastinal shift to the right side(Figure 1).On multi-detector computed tomography (MDCT) of the chest, the left upper lung lobe was hyperinflated and hyperextended with trans-mediastinal herniation into the right side as well as partial volume reduction of the right lung and preserved intra-parenchymal vascular pattern(Figure 2).On bronchoscopy, there was no evidence of FB or bronchial obstruction buta slit like opening of the left upper lobe bronchus during expiration (Figure 3). Left thoracotomy was performed with left upper lobe lobectomy (Figure 4). The postoperative course was uneventful with no further episodes of respiratory distress.

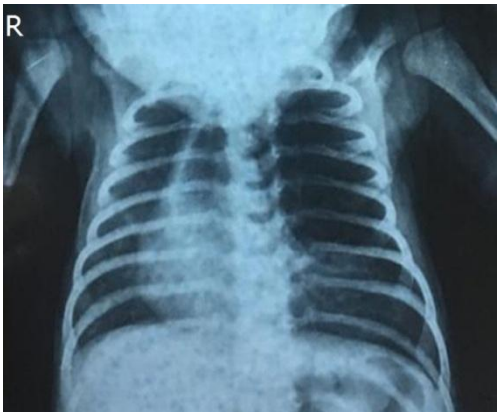

Figure1. *Plain chest x-ray showinghyperinflated left upper lung lobe with hyper-lucency and mediastinal shift to the right side.*

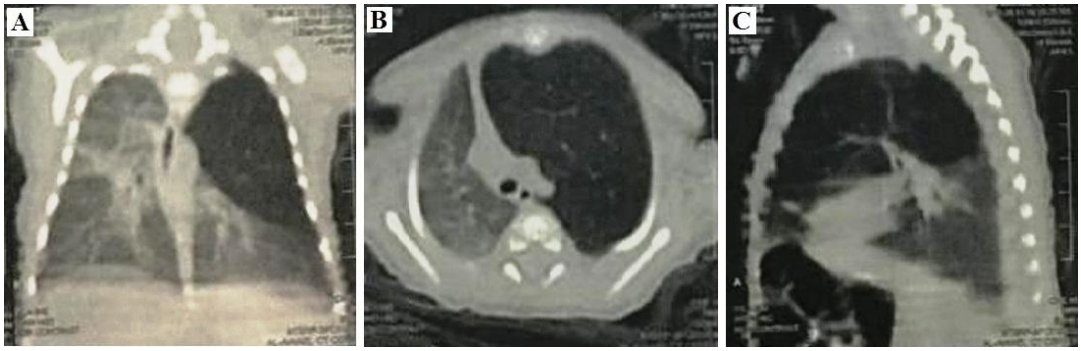

Figure2. *Views of multi-detector computed tomography (MDCT) of the chest showing hyperinflatedleft upper lung lobe and hyperextended with trans-mediastinal herniation into the right side: (A) Coronal view, (B) Axial view, and (C) sagittal view.*

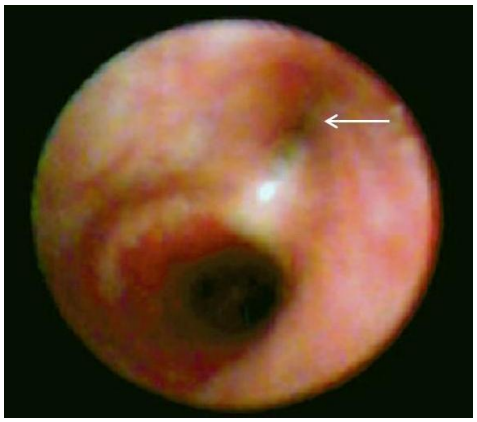

Figure3. *A bronchoscopic view showing a slit-like opening of the bronchus of left upper lobe during expiration (arrow).*

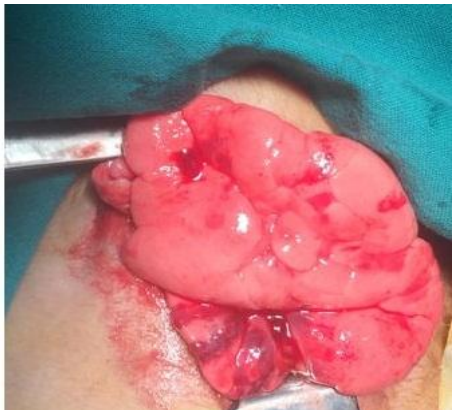

Figure4. *Intra-operative view of hyperinflated (emphysematous) left upper lobe*

DISCUSSION

As one of the developmental lung malformations, CLE occurs due to abnormalities in the bronchopulmonary foregut starting at the third week of gestation [5]. The pathogenesis of CLE is related to hyperinflation of one or more of lung lobesas a result of partial bronchial obstruction. This rare entity is frequently presented in the neonatal period and infancy often during the first year of life [6]; however some cases have been reported in the childhood and adult age [7].

The cases of CLE may manifest a life-threatening respiratory distress due to compressive consequences and mediastinal shift; however asymptomatic cases have been reported [8].Therefore, an early diagnosis of CLE in the neonatal period is important depending on clinical high index of suspicion as it may be confused by other conditions particularly pneumonia and pneumothorax [9].

Initially, CLE may appear on chest x-ray as over inflated lobe, tracheal or a mediastinal shift to the opposite side, atelectasis of the adjacent lobes or herniation of the expanded lobe across the midlines [10].However, chest CT provides more information than chest x-ray and it is indicated in cases with CLE when respiratory distress or lung expansion not improved. Moreover, CT is a useful tool to exclude the presence of pneumothorax and to avoid inadvertent chest tube insertion [10, 11].

Bronchoscopy can be used in cases with CLE to excludeintrinsic or extrinsic airway obstruction, but it is not performed as a routine. The bronchoscopic appearance of a patent airway during inspiration and dynamic airway collapse on expiration suggests that the bronchial collapse contributes to lung hyperinflation in cases with CLE [12].Also, echocardiography is an important diagnostic tool in the cases with CLE to exclude theassociated congenital cardiac malformations, such as patent ductusarteriosus, atrial septal defect, ventricular septal defect, total anomalous pulmonary venous return, and Tetralogy of Fallot [13]. The presence and severity of cardiac anomalies may change the timing and extent of surgery for the concomitant congenital heart disease [14].

The presence of severe respiratory distress is the main indication for surgical excision of CLE. Evidence of mediastinal shift with subsequent compression of the unaffected lung lobes indicates surgical excision. Some surgeons recommended surgery in all infants younger than 2 months or older than 2 months with severe respiratory symptoms [1, 4, 15]. Lobectomy of the affected lung lobe is the preferred surgical option for CLE. Some surgeons suggested segmental resection with an advantage to preserve the healthy lung parenchyma; however, no difference in the postoperative complication rates has been reported between lobectomy and segmentectomy [16].

CONCLUSION

The diagnosis of rare cases of CLE requires a high index of clinical suspicion, as it may mimic other conditions of respiratory distress and bronchial obstruction. Early diagnosis is crucial to avoid life-threatening consequences. Chest CT is the most useful diagnostic modality for CLE. The treatment depends on the severity of the clinical symptoms, with a need for surgical resection before the age of 2 years or when the patient presented with respiratory distress.

REFERENCES

[1] Andrade CF, Ferreira HP, Fischer GB. Congenital lung malformations.J Bras Pneumol. 2011;37 (2):259-71.

[2] 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(4):418-26. doi: 10.1590/S1806-37132013000400004.

[3] Durell J, Lakhoo K. Congenital cystic lesions of the lung. Early Hum Dev. 2014;90(12):935-9. doi: 10.1016/j.earlhumdev.2014.09.014.

[4] Kamal YA. Management of Congenital Lobar Emphysema: The Current Challenges. ARC Journal of Surgery.2018; 4(1):20-25. doi: dx.doi.org/ 10.20431/2455-572X. 0401005.

[5] Mendeloff EN. Sequestrations, congenital cystic adenomatoid malformations, and congenital lobar emphysema. Semin ThoracCardiovasc Surg. 2004; 16(3): 209-14.

[6] Ozcelik U, Gocmen A, Kiper N, Dogru D, Dilber E, Yalcin EG. Congenital lobar emphysema: evaluation and long-term follow-up of thirty cases at a single center, Pediatr. Pulmonol. 2003;35 (5):384-91.

[7] Pike D, Mohan S, Ma W, Lewis JF, Parraga G. Pulmonary imaging abnormalities in an adult case of congenital lobar emphysema. J Radiol Case Rep. 2015;9(2):9-15.

[8] Latif I, Shamim S, Ali S. Congenital lobar emphysema. J Pak Med Assoc. 2016; 66(2): 2102.

[9] Quinton AE, Smoleniec JS. Congenital lobar emphysema--the disappearing chest mass: antenatal ultrasound appearance. Ultrasound Obstet Gynecol. 2001;17(2):169-71.

[10] Kumar S, Debata PK, Gupta R. Congenital lobar emphysema and intercostal drainage tube insertion: the common fate of an uncommon disease. J ClinDiagn Res. 2012;6(9):1568-70.

[11] Prabhu SM, Choudhury SR, Solanki RS, Shetty GS, Agarwala S. Inadvertent chest tube insertion in congenital cystic adenomatoid malformation and congenital lobar emphysema-highlighting an important problem. Indian J Radiol Imaging. 2013;23(1):8-14.

[12] Doull IJ, Connett GJ, Warner JO. Bronc hoscopic appearances of congenital lobar emph ysema.PediatrPulmonol.1996;21(3):195-7.

[13] Chinya A, Pandey PR, Sinha SK, SarinYK. Congenital lobar emphysema: Pitfalls in diagnosis.Lung India. 2016;33(3): 317-9.

[14] Dogan R, Dogan OF, Yilmaz M, Demircin M, Pasaoglu I, Kiper N, Ozcelik U, Boke E. Surgical management of infants with congenital lobar emphysema and concomitant congenital heart disease. Heart Surg Forum. 2004; 7(6):E644-9.

[15] Kanakis M, Petsios K, Bobos D, Sarafidis K, Nikopoulos S, Kyriakoulis K, Lioulias A, Giannopoulos N. Left Upper Lobectomy for Congenital Lobar Emphysema in a Low Weight Infant. Case Rep Surg. 2016; 2016:4182741.

[16] Bagrodia N, Cassel S, Liao J, Pitcher G, Shilyansky J. Segmental resection for the treatment of congenital pulmonary malformations. J Pediatr Surg. 2014 Jun; 49(6): 905-9. doi: 10.1016/j.jpedsurg. 2014.01. 021.

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