

A case of endobronchial polyp mimicking congenital lobar emphysema in an infant

Rahele Mehrain (MD)^{*1, 2}
Abbas Hadipur (MD)¹

1- Non-Communicable Pediatric Diseases Research Center, Amirkola Children's Hospital, Babol University of Medical Sciences, Babol, Iran.
2- Department of Radiology, Amirkola Children's Hospital, Babol University of Medical Sciences, Babol, Iran.

* **Correspondence:**
Rahele Mehrain, Non-Communicable Pediatric Diseases Research Center, Amirkola Children's Hospital, Babol University of Medical Sciences, Babol, Iran.

E-mail:
sany_monzavi@yahoo.com
Tel: 0098 111 3242151
Fax: 0098 111 3253061

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Abstract

Background: Congenital lobar emphysema, also called infantile lobar emphysema, is a respiratory disease that occurs in infants when air enters the lungs and cannot leave easily. It results from cartilage deficiency and bronchomalacia causing distal air trapping and respiratory distress. In this study, we present a case of endobronchial polyp mimicking congenital lobar emphysema.

Case presentation: An 18-month old infant was admitted to the hospital due to sudden respiratory distress. Chest x- ray showed left hemithorax lucency, mediastinal shift to the right side, left hemidiaphragm flattening, and left oblique fissure bulging to the superior. A bronchoscopy showed polyp in the left lower bronchus and the biopsy of the lesion confirmed to be the polyp.

Conclusion: The findings of this case emphasize the value of bronchoscopy prior to lobectomy in cases of congenital lobar emphysema.

Key words: Congenital lobar emphysema, Endobronchial polyp, Air trapping, Bronchial Obstruction, Bronchoscopy.

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Congenital lobar emphysema (CLE) is an uncommon but potentially life threatening abnormality affecting infants (1, 2). It is caused by the hyperinflation of the lung lobe with compression of the normal lung parenchyma and contra lateral displacement of the mediastinum (3). Over distension of the pulmonary lobe is secondary to partial bronchial obstruction (4).

The most common affected lobe is the left upper lobe followed by the right upper lobe and the right middle lobe but any of these lobes may be affected. There is not only an inspiratory air entry but also a collapse of the narrow bronchial lumen during expiration. Bronchial defect results in lobar air trapping. Most patients often have recurrent respiratory distress within the first 6 months of life (2).

Various suggestions have been put forward as to the cause of CLE, but approximately half of the cases are of unknown etiology. A further 25% cases are due to bronchial cartilage defect, which can range from hypoplasia to absence of tissue. Thirteen percent cases are due to endobronchial obstruction, such as a fold, web, mucus plug, or tumor and tumor like conditions. Other causes include bronchial compression by a vascular structure such as a patent ductus arteriosus or aberrant left pulmonary artery (5). Among the benign endobronchial growths, the chance that the obstruction is produced by an inflammatory polyp arising from the bronchial mucosa is indeed very small and their clinical presentation in the pediatric population is extremely rare. Nonetheless, its existence should not be ignored (6, 7).

In this paper, we report an endobronchial polyp as a cause of lobar hyperinflation in an infant that mimicked congenital lobar emphysema.

Case Presentation

An 18-month old boy was referred to Amirkola Children's Hospital because of sudden cough, wheezing and respiratory distress. He was the couple's first child and was a full term baby with normal delivery. The post-natal period was uneventful until he developed the above symptoms. There was not any history of related diseases in his family. On physical examination, the infant had tachycardia, tachypnea, and wheezing on auscultation on the base of the left lung. The routine white blood cell and biochemical investigations were normal.

Chest x-ray showed left hemithorax lucency, mediastinal shift to the right side, left hemidiaphragm flattening, and left oblique fissure bulging to the superior (figure 1).



Figure 1. CXR shows left hemithorax hypertranslucency, mediastinal shift to right, left hemidiaphragm flattening and left oblique fissure bulging to the superior.

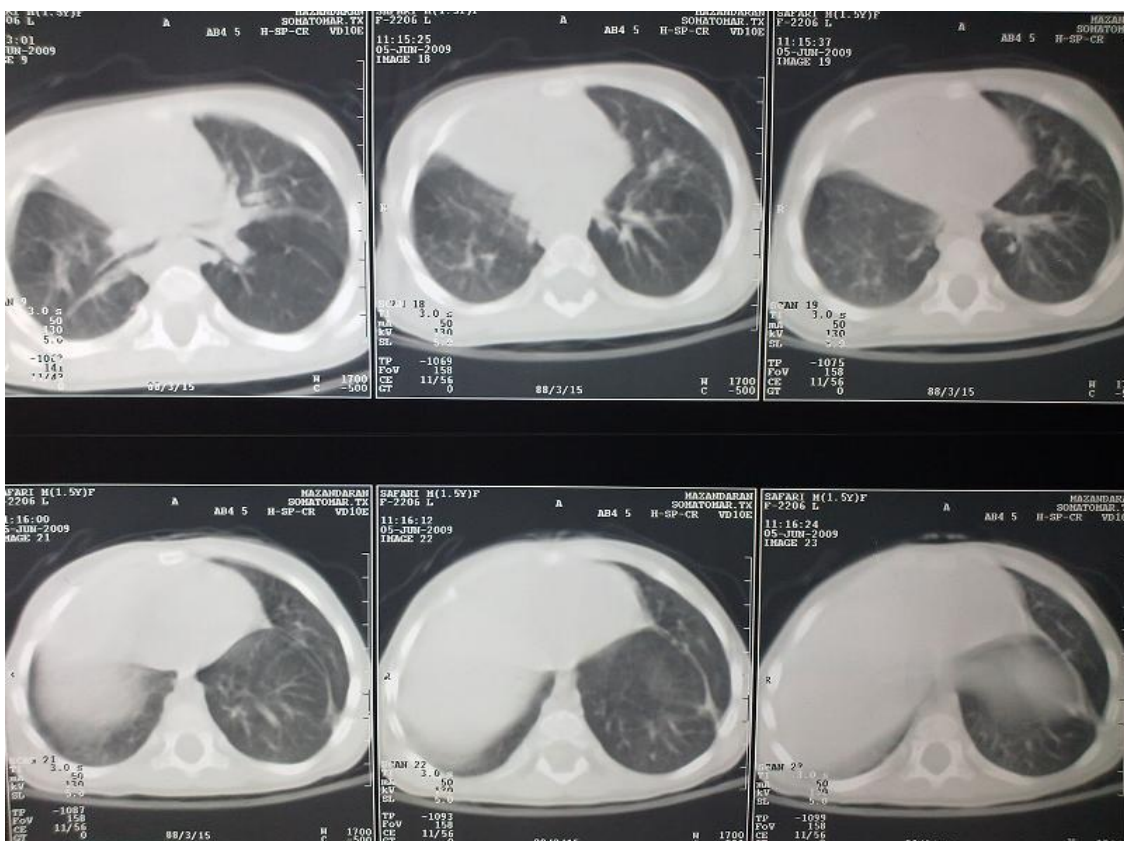


Figure 2. Chest CT scan shows air trapping in left lower lobe, mediastinal shift to right, oblique fissure bulging to the superior and compression atelectasis of adjacent lobe.

Because of the latter sign in chest x-ray, we decided to perform a chest CT scan, to differentiate pneumothorax, pulmonary bulla and lobar hyperinflation. The findings on CT scan without contrast were air trapping in left lower lobe,

mediastinal shift to right, oblique fissure bulging to the superior and compression atelectasis of adjacent lobe (figure 2). On the basis of CT findings, the pneumothorax and bulla were ruled out. To differentiate lobar

hyperinflation and congenital lobar emphysema (CLE) especially CLE, foreign body and other endobronchial lesions bronchoscopy was performed. In bronchoscopy, a pinkish, bright, pedunculated polyp was seen moving up and down in the left lower bronchus that almost totally obstructed it. The bronchoscope was impossible to pass behind the polyp to assess its segmental origin. No endobronchial foreign body was found.

A biopsy was performed with great difficulty because of the extremely tough nature of the polyp and risk of bleeding. The histopathologic evaluation of the sample was reported as inflammation and the biopsy specimen was non malignant in nature. It was not possible to remove the polyp because of the risk of bleeding. The patient recovered relatively and was discharged from the hospital one week later.

Discussion

Our patient had respiratory distress and lobar air trapping, mostly suggestive of the CLE. The differential diagnosis of lobar hyperinflation is “congenital lobar emphysema” that is more common in boys than in girls like our patient, but it is more common in infants less than six months and in left upper lobe unlike our patient that was 8 months and the air trapping was in the left lower lobe. Another cause of lobar hyper translucency, especially in an infant was the foreign body that was more common in the right lower lobe bronchus.

With investigation facilities available, the baby with moderate respiratory distress like our case can be diagnosed easily. Chest x-ray and CT scan of thorax are diagnostic and show the hyperlucent affected lobe (8). Bronchoscopy can rule out the foreign body as an important cause of air trapping in an infant like our patient and shows another cause of endobronchial obstruction that was bronchial polyp.

This case showed that endobronchial polyp can be presented by hyperinflation. Their clinical presentation in the pediatric population is extremely rare. McShane et al. reported that three out of 4 patients had a history of mechanical ventilation in the neonatal period. Presentation in 3 of the 4 patients was with lobar collapse and the fourth patient presented with hyperinflation (7).

The significant point is that once the nature of the process is ascertained by biopsy, the lesion can be eradicated completely by the endoscopic route, avoiding recourse to unnecessary major surgery that is the traditional management of

congenital lobar emphysema. Although, controversy exists regarding the surgical and conservative management of this malformation, one opinion is in favor of the conservative management for mild cases but stringent follow-up is necessary (9).

Our case, together with a few case reports of others, form a distinct group of endobronchial polyps characterized by rapidly progressing signs and symptoms of bronchial obstruction in the absence of preceeding manifestations of tracheobronchial or pulmonary disease.

The findings of this case emphasize the value of bronchoscopy prior to lobectomy in cases of congenital lobar emphysema.

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References

1. Clublely E, England RJ, Cullinane C, Crabbe DC. Ball valve obstruction of a bronchus causing lobar emphysema in a neonate. *pediatr surg int* 2007; 23: 699-702.
2. Chao MC, Karamzadeh AM, Ahuja G. Congenital lobar emphysema. *Int J Pediatr Otorhinolaryngol* 2005; 69: 549-54.
3. Cunha Fatureto M, Pinheiro Ferreira D, Amaro Ferraz D, Vieira Dos santos JP, Andrade Maia S. Congenital lobar emphysema: Study of a case. *Rev Port Pneumol* 2008; 14: 893-6. [In Portuguese]
4. Konan Blé R, Coste K, Blanc P, et al. Congenital lobar emphysema: a rare etiology of hyperechoic lung. *Gynecol Obstet Fertil* 2008; 36: 529-31.
5. Joazlina ZY, Wastie ML, De Bruyne FA. Congenital lobar emphysema: a diagnostic challenge. *J HK Coll Radiol* 2005; 8: 105-8.
6. Caldarola VT, Harrison EG Jr, Clagett OT, Schmidt HW. Benign tumors and tumor-like conditions of the trachea and bronchi. *Trans Annu Meet Am Bronchoesophagol Assoc* 1964; 44: 46-69.
7. McShane D, Nicholson AG, Goldstrow P, et al. Inflammatory endobronchial polyps in children: Clinical

- spectrum and possible link to mechanical ventilation. *Pediatric Pulmonol* 2002; 34: 79-84.
8. Khemiri M, Ouederni M, Ben Mansour F, Barasaoui S. Bronchogenic cyst: an uncommon cause of congenital lobar emphysema. *Respir Med* 2008; 102: 1663-6.
9. Mei-Zahav M, Konen O, Manson D, Langer JC. Is congenital lobar emphysema a surgical disease? *J Pediatr Surg* 2006; 41: 1058-61.