

## Spontaneous regression of left upper lobe emphysema after division of large ductus arteriosus in an infant

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**SUMMARY:** Doğan ÖF, Duman Ü, Yorgancıoğlu C, Böke E. Spontaneous regression of left upper lobe emphysema after division of large ductus arteriosus in an infant. Turk J Pediatr 2006; 48: 269-271.

Congenital lobar emphysema is cause of infantile respiratory distress and diagnosed by lobar overaeration, mediastinal shift and/or compression of the adjacent lobe. However, concomitant congenital heart disease with this clinical condition is not uncommon, and there is no uniform consensus about the treatment strategy in these particular cases. We present a child who was treated with division of large patent ductus arteriosus without the lobectomy.

*Key words:* compressive lobar emphysema, spontaneous regression, patent ductus arteriosus, surgery.

Congenital lobar emphysema (CLE) is a rare clinical condition that often requires lobectomy in early infancy. Bronchial cartilaginous dysplasia is often associated with this condition, but in half the affected infants, the etiology remains undetermined. An extrinsic obstruction, such as dilated pulmonary arteries, or left atrium associated with ventricular septal defect and patent ductus arteriosus (PDA), or tetralogy of Fallot (TF) with dilated pulmonary arteries may cause this condition. Here, we report an infant with left upper lobar emphysema caused by a dilated ductus arteriosus compressing the left main bronchus. Two months after the division of the ductus arteriosus, spontaneous complete regression of the emphysema was observed. We should explore the cause of lobar emphysema thoroughly before lobectomy, especially when it is extrinsic. The emphysema may regress by eliminating the extrinsic factor.

### Case Report

An eight-month-old girl was admitted to emergency service with a lower airway infection, persistent fever and progressive respiratory distress. She exhibited symptoms of severe cyanosis and hypoxia, and a systolo-diastolic murmur was also heard from the second intercostal space. She was intubated immediately after admission to the emergency

service. Arterial blood gas analysis showed deep hypoxia and pCO<sub>2</sub> increase. Hyperlucency of left upper zone and bilateral infiltration were seen on her chest roentgenogram. Dilated PDA and moderate pulmonary hypertension were detected by transthoracic echocardiography. Extubation of patient was unsuccessful. The left upper lobe emphysema and pneumonic infiltration were diagnosed by a thoracic computed tomography (CT) (Fig. 1). Additionally, thoracic 3-D CT demonstrated a dilated ductus arteriosus, located just on the left main bronchus and thus compressing it due to its curving toward the bronchus (Fig. 2). Airway infection was regressed. In bronchoscopic examination, severely narrowed left main bronchus was seen due to extrinsic compression. PDA ligation was decided, but operation was postponed because of development of septic complication due to recurrent lower airway infection. Although airway infection regressed two weeks later, the patient could not be weaned from the respirator, and she underwent an operation for the closure of PDA.

Left thoracotomy incision was made and the large ductus arteriosus was clearly visualized. The division of the large ductus arteriosus that leaned onto the left main bronchus was performed without an accompanying lobectomy. Postoperative thoracic chest roentgenogram

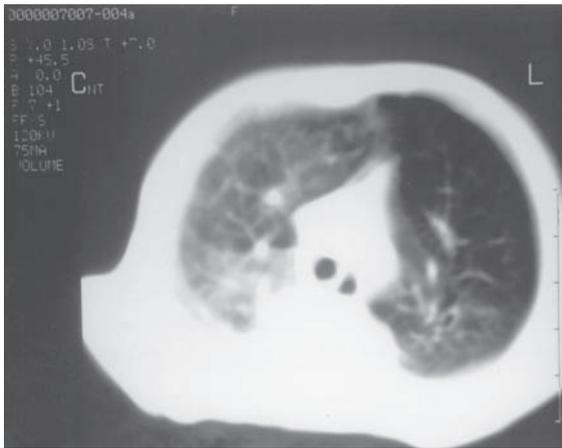


Fig. 1. Chest computed tomographic (CT) scan revealing left upper lobe emphysema and shifting of mediastinum to ipsilateral site.

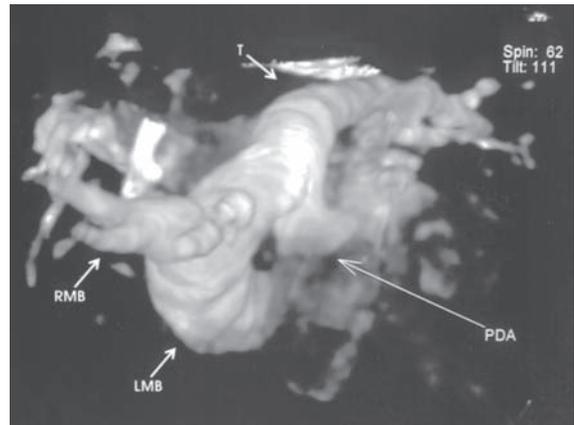


Fig. 2. 3-D thoracic CT illustrating compression of the left main bronchus by a large ductus arteriosus. Long arrowhead shows large ductus arteriosus compressing the left main bronchus. T: Trachea. LMB: Left main bronchus. RMB: Right main bronchus. PDA: Patent ductus arteriosus.

illustrated no abnormality and the patient was weaned from the respirator successfully one day following surgery. The patient was discharged from hospital in good clinical condition. The emphysema was not seen in the follow-up CT two months after surgery (Fig. 3).

### Discussion

Congenital lobar emphysema (CLE) is an uncommon cause of infantile respiratory distress diagnosed by the evidence of lobar overaeration, mediastinal shift and compression of the adjacent lobe. Concomitant congenital heart disease (CHD) and CLE is not uncommon. In the literature, a 12-20% concomitance rate is given<sup>1-3</sup>. The optimal treatment for

respiratory symptoms associated with CLE and CHD is not clear; however, there has been a great deal of progress in relation to the treatment of CLE and CHD. In 1954 Gross and Lewis<sup>4</sup> published the first case report of CLE in which the patient was treated by lobectomy. A few studies have reported cases treated conservatively with medicine<sup>5-8</sup>; those usually resulted from hypertensive or dilated pulmonary arteries associated with ventricular septal defect (VSD) or PDA. Several cases have been reported in whom regression of CLE occurs after open-heart surgery<sup>9,10,11</sup> and/or after a given medical treatment<sup>8</sup>. One of the patients had PDA and concomitant TF with large pulmonary arteries and underwent

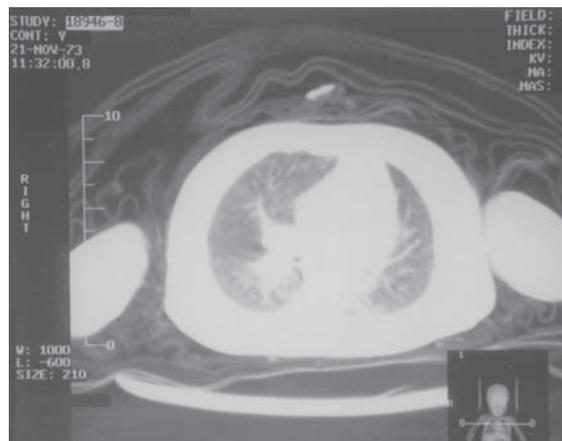


Fig. 3. Postoperative thoracic computed tomographic (CT) scan showing spontaneous regression of the left upper lobe emphysema two months after surgery.

division of PDA and pulmonary arterioplasty with cardiopulmonary bypass. The other patient had VSD and underwent surgical closure of the VSD. Our patient had no need for pulmonary arterioplasty and had no another CHD such as VSD or TF. Furthermore, Doğan et al.<sup>11</sup> suggested lobectomy in patients with onset of severe symptoms who are resistant to medical treatment due to CLE.

The most frequent site for emphysema is the right middle lobe, where the right middle bronchus is often compressed by dilated pulmonary vessels<sup>2</sup>. In the present patient, emphysema was found in the left upper lobe, which is a rare site in cases of lobar emphysema with CHD. Our patient had no excessive dilated pulmonary vessels but had a dilated left ductus arteriosus, which is located near the left main bronchus. If emphysema is found in the left upper lobe in patients with PDA and there is no sign of vascular ring, the possibility of large ductus arteriosus compressing the left bronchus should be considered. Follow-up thoracic CT revealed complete regression of the emphysema two months after the division of ductus arteriosus. While urgent lobectomy may be necessary for the infant with severe respiratory distress, in other cases it must be considered carefully because lobar emphysema from an extrinsic cause can regress without lobectomy following the removal of the primary cause.

In summary, we have presented a case of lobar emphysema due to compression of the left main bronchus by a large ductus arteriosus. This case shows that lobar emphysema due to a large ductus arteriosus may regress in the postoperative period. We should explore the cause of lobar emphysema thoroughly before

lobectomy, especially when it is extrinsic. The emphysema may regress by eliminating the extrinsic factor.

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