Mediastinal Esophageal Cyst Causing Unilateral Hyperlucent Lung

Unilateral emphysema secondary to bronchial obstruction by a foregut-derived mediastinal cyst is rare. Here we describe an infant with a unilateral hyperlucent lung due to compression on the left main bronchus by an esophageal cyst, visualized by chest computed tomography and magnetic resonance imaging. A chest roentgenogram and a perfusion scan presented the normalized left lung after resection of the cyst. (JJTCVS 1999; 47: 141-143)

Index words: hyperlucent lung, mediastinal cyst

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The distinctive radiographic signs of the Swyer-James syndrome are a unilateral hyperlucent lung of normal or decreased size with diminished lung markings, a smaller hilar shadow than the contralateral lung and slight displacement of the mediastinum to the affected side. A unilateral hyperlucent lung, however, can be caused by other conditions. A partially obstructing lesion in the main bronchus can produce three of the roentgenographic signs of Swyer-James syndrome. Here we report a rare infantile case of a unilateral hyperlucent lung caused by an esophageal cyst.

Case

An 11-month-old male infant with a history of repeated pulmonary infection was admitted to the Department of Pediatrics of our University. Left lung emphysema and a decreased size of the left pulmonary arterial trunk on a chest roentgenogram had been noted at 2 months old. On admission, a physical examination revealed hyper-resonance and diminished respiratory sounds of the left hemithorax. A systolic murmur on the left sternal border was noted. Chemistry profile, complete blood cell count, and room-air blood gas values on admission were all normal except for an increased serum SCC concentration at 2.98 ng/ml.

A chest roentgenogram on admission revealed
the left lung hyperlucency with sparse pulmonary vessels (Fig. 1). A pulmonary angiogram revealed thin and attenuated vessels throughout the left lung and no stenosis in the left pulmonary arterial trunk (Fig. 2). A pulmonary scan showed a marked decrease in the perfusion of the left lung (Fig. 3). A chest CT scan confirmed an unexpected mediastinal mass located inferiorly to the aortic arch (Fig. 4). The findings from MRI, of a low intensity lesion on T1-weighted imaging (Fig. 5) and a high intensity on T2-weighted imaging, suggested that the mass contained protein-rich fluid. Fiberoptic bronchoscopy revealed a nearly obstructed left main bronchus compressed by the surroundings. These findings suggested that the mediastinal cyst was responsible for the severe stenosis in the left main bronchus.

Left thoracotomy revealed a well-encapsulated mediastinal soft mass, strongly adhering to the esophagus and compressing the left main bronchus. The excised mediastinal mass was 3.2 × 2.3 × 2.2 cm in size, consisting of a thin-walled cyst, filled with brownish turbid fluid. The cyst fluid contained CEA (439 ng/ml), CA19-9 (74.5 × 10^7 U/ml) and SCC (30.1 ng/ml). Histologically, the cyst was covered with stratified ciliated epithelia, and had two layers of smooth muscle. These findings were compatible with an esophageal cyst.2

He was discharged from our department on the 13th postoperative day without any complications. At 6 months after the operation, a chest roentgenogram showed improvement in the left lung hyper-