INTRAPULMONARY BRONCHOGENIC CYST: A CASE REPORT

İntrapulmoner bronkojenik kist: Bir vaka takdimi

Yiğit Akçalı[1]

Summary: *On February 6, 1987, in our department, 11-year-old boy who complained cough, dyspnea, and sputum production was managed surgically for intrapulmonary bronchogenic cyst.*

Key Words: *Bronchogenic cyst, Intrapulmonary*

Özet: *6 Şubat 1987'de, kliniğimizde öksürük, dispne ve balgam çıkarımından yakınan 11 yaşında bir erkek çocuk intrapulmoner bronkojenik kist nedeniyle cerrahî olarak tedavi edildi.*

Anahtar Kelimeler: *Bronkojenik kist, Akciğer içi*

Bronchogenic cysts (BCs) are thought to result from the primitive respiratory system between the third and sixt weeks of gestation and consequently may be mediastinal, intrapulmonary or diaphragmatic in location (6,16). BCs have varied clinical presentation and course, and present in one of three ways: (1) as asymptomatic roentgenographic findings; (2) with symptomatic compression of adjacent intrathoracic structures; or (3) as infectious complications related to cyst (13,18). Infants typically presents with respiratory compromise due to airway compression (8) whereas older children present with superimposed infection (9,16,18). We describe a case of an intrapulmonary bronchogenic cyst in 11-year old boy in whom the presenting symptoms were due to infected cyst plus pneumonia.

CASE REPORT

An 11-year-old boy was admitted with cough, dyspnea, and sputum production to the Department of Thoracic and Cardiovascular Surgery.

He had recurrent fever, productive cough, and had been treated medically at the Department of Pediatrics. Physical examination revealed rales in the left lung field. The chest roentgenogram showed a cystic cavity with air-fluid level in the left upper lobe (Figure 1). The remainder of both lungs and mediastinum were normal. An antibiotic therapy was administered for a week period then patient underwent a thoracotomy. The excessive pleural adhesion was resected, the cystic mass which was 80 mm in diameter, full of white gelatinous fluid, dissected from the bronchus on the left upper lobe. Bronchial air leak was sutured and cystic cavity was capitonnaged. The pathological diagnosis was reported to be bronchogenic cyst. The postoperative course was uneventful. The patients was discharged one week after the operation.

Erciyes University Medical Faculty 38039 Kayseri, TÜRKİYE Department of Thorocic and Cardiovascular Surgery. Assist.Assoc. Prof[1].

DISCUSSION

Bronchogenic cysts (BCs), which arise from nests of cells that become isolated from the main pulmonary branching when the lung bud is seperating from the primitive gut, are found most frequently in the lungs or mediastinum, but they can develop in extrathoracic locations, such as cervical, pericardial, paravertebral, or infra-and intra- diaphragmatic sites (6,11,17). In 1948, Maier classified BCs by location in the thorax and mediastinum: (1) paratracheal (right and left); (2) carinal (subcarinal); (3) hilar (right and left), intrapulmonary; (4) paraesophageal (right and left). BCs have also been reported to occur in the supraclavicular or presternal spaces or within the pericardium (6,17,18).

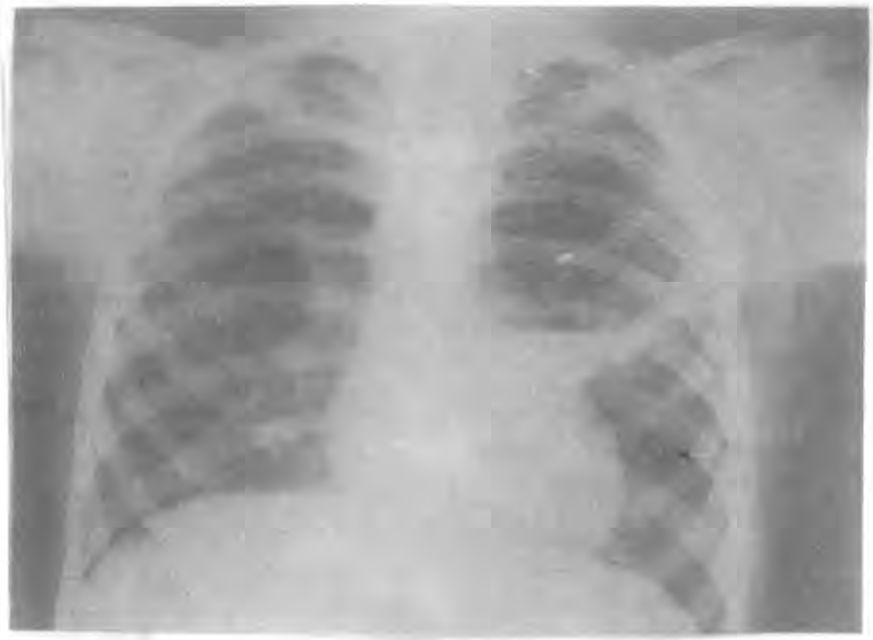

Figure 1. Posteroanterior chest roentgenogram demonstrating a rounded, thin-walled, and air-fluid levelled mass.

The relative occurence rates of mediastinal and intrapulmonary BCs are controversial. Studies by some authors (5,14) indicated the frequency of mediastinal BCs was greater, whereas the series reported by some authors (11,15) showed a higher frequency of pulmonary lesions. The intrapulmonary BC is equally distiributed between the two lungs but is about twice as common in lower lobes as it is in upper lobes.

Past reports emphasize that BCs may be asymptomatic. In some series (5,18), 27-28 % of the patients were asymptomatic and 72-73 % presented with respiratuar symptoms such as cough, dyspnea, pain, fever and pneumonia, whereas in other series (11), nearly all patients were symptomatic, presenting with such signs and symptoms as fever, recurrent pneumonia, cervical mass, respiratory distress, frequent upper respiratory infections, and empyema. The most BCs are or will ultimately become symptomatic. Table 1 summarizes clinical symptoms and signs in the patient with BCs.

The onset of symptoms is progressive in 80 % of the patients, and the severity of symptoms is moderate in 56 % of the patients (18). The vast majority of patients have more than one symptom. Most reported BCs have been in the childhood, where they often are seen as life threatening emergencies with airway obstruction resulting in atelectasis, air trapping, and respiratory distress (3,8,11,18). Matzinger, et al (9) disclosed a spontaneous pneumothorax caused by rupture of an intrapulmonary BC. Kirwan, et al (7) reported catastrophic spontaneous rupture of an

Table 1. Clinical symptoms and signs in the patients with BCs (1-18)

Chest pain	Purulent sputum **	Recurrent pneumonia
Cough*	Anorexia and weight loss	Cervical mass
Dyspnea*	Dysphagia*	Frequent upperrespiratory infect.
Fever	Hemoptysis	Empyema

** They are caused by compression or irritation of major airways and esophagus by the cyst.*
*** It may be indicate infection of the cyst with fistulization or more likely pneumonia in the adjacent compressed lung*

Table 2. Complications (7,9,12,13,18,19)

Arrhytmias	Hemorrhage	Superior vena cava syndrome
Bronchial atresia	Infection without fistula	Ulcerations of cyst wall alone
Cardiac tamponade	Recurrency	Malign transformation of
Fistulization with airways	Spontaneous pneumothorax	the cyst wall

upper mediastinal BC with leakage into the pericardium and cardiac tamponade. Complications in the patients with BC are summarized in table 2.

Traumatic pulmonary cyst and congenital lung cyst are the major considerations in the differential diagnosis of the patients with BC. Traumatic cysts typically follow blunt chest injury with sudden compression of the elastic thoracic wall in young persons. Radiologically appearent within 12-24 hour of injury, traumatic cysts undergo complete resolution in 2-16 weeks (16). Congenital cysts tend to be multiple, either limited to a lobe, or generalized, involving one or both lungs. They may be asymptomatic, or the patient may present with complications of either infection or air trapping and tension (16).

Other possible causes of thin-walled, air-containing lung cysts include cavities resulting from mycotic, tuberculosis, or Echinococcus infections and pneumotoceles caused by Staphylococcus aureus or hydrocarbon ingestion, or lipoid pneumonia presenting cavitation (1,2,16).

Chest roentgenography is the usual imaging method for initial detection of BC, although the findings are not always diagnostic. On a chest radiograph, an intrapulmonary BC typically appears as a sharply defined, thin-walled, round or oval lesion that may be air filled, have homogeneous water densitiy, or contain an air-fluid level. CT provides optimal demonstration of cyst location, morphology, and contents (9). Because the walls of uncomplicated BCs are relatively thin and are interposed between the cyst contents and the adjacent structures, the imaging characteristics of these lesions are essentially those of the contents of the cyst. Roentgenograms show a soft-tissue mass, frequently in other mediastinal locations or within the lung. CT scans show sharp margination and no contrast enhancement (10).

St-Georges et al (18) thought that most, if not all, patients who had BCs either in the mediastinum or in the lung were operated on. Indications for operation in the patients with BCs are outlined in table 3.

Table 3. Indications for operation (18).

Mass previously known	
	Stable
	Increase in size and/or change in symptomatology
Mass not previously known	
	Asymptomatic
	Symptomatic
	Incidental finding during operation for reflux esophagitis

Surgical excision, which has minimal morbidity and mortality, is the treatment of choice for both the infected and uninfected cyst to firmly establish the diagnosis and prevent future complications (5,16). The most frequent operative approach is a posterolateral thoracotomy. Others approachs are an axillary incision, a median sternotomy and a cervical mediastinoscopy incision for a subcarinal BC. Surgical excision of mediastinal BC can be hazardous because of dense pericystic adhesions with adjacent organs traheobronchial tree, esophagus, pericardium, or lung. In a series (18), major operative difficulties or intraoperative complications were encountered in 44 % of the patients. Table 4 outlines the intra-and post-operative complications.

St-Georges et al (18) recommended that all presumed BCs seen in the adult be resected because the majority will ultimately become symptomatic or complicated. Because cyst recurrence, which carries a substantial morbidity, can occur, transtracheal and cyst aspirations (18,20), which have been proposed

Table 4. Intra-and post-operative complications (4, 18).

Intraoperative	Postoperative
Arterial laceration	Respiratory failure requiring tracheotomy for respiratory support
Vagal trunk division	Hemothorax
Segmental bronchus laceration with subsequent segmental resection	Atelectasis
Esophageal mucosal laceration	Pleural fluid accumulation
	Wound infection
	Transient Horner's syndrome

as alternatives to operation, are not recommended. Because definitive tissue diagnosis can only be established by means of surgical excision, early surgical intervention is also important.

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