

Bronchogenic cyst: A congenital cystic malformation of the lung*

Alaaddin ÇELİK, Hüseyin ÖZBEY, Cem BONEVAL, Selim AKSÖYEK,
Gülnaz KERVANCIOĞLU, Tansu SALMAN

Department of Pediatric Surgery and Pathology, University of İstanbul, İstanbul Faculty of Medicine, Türkiye

Summary

Bronchogenic cyst, cystic adenomatoid malformation, bronchopulmonary sequestration and lobar overinflation are rare congenital pulmonary malformations, which may sometimes be rapidly fatal if not recognized and treated promptly. Among those, bronchogenic cyst is the most common type of the lung cyst. Between 1985 and 1992, 3 infants and a 6 year old girl with bronchogenic cysts were treated surgically in our clinic.

The most common presentations were respiratory distress, cyanosis, and a pulmonary cystic mass found incidentally on a routine chest x-ray. This pulmonary malformation must be included in the differential diagnosis of the respiratory distress of the newborn, infant and child.

Key words: Bronchogenic cyst, bronchopulmonary foregut malformation

Introduction

Bronchogenic cysts are considered as the result of abnormal budding of the tracheobronchial tree at any point before the alveolus (5,7). They are usually located in the mediastinum but may also be intrapulmonary. Cysts may be filled with air and mucoid and/or infected material (8). A retrospective review of four patients with bronchogenic cysts was carried out and the importance of the clinical recognition of this malformation is pointed out.

Material and Methods

The case notes of four patients with bronchogenic

cysts were reviewed retrospectively. Patients were investigated by one or more of the following imaging techniques; plain chest x-ray, computerized tomography, radionuclide scintigraphy and esophagography (Figure 1).

Results

Clinical findings and the radiological features are outlined in Table 1. The main clinical finding was found to be respiratory distress with cyanosis, mostly prominent during feeding or crying.

The radiography of the chest in case 2, suspected a diaphragmatic hernia but a barium enema and esop-

Table 1. Clinical and radiological findings

Case no	Sex&Age at admission	Signs&symptoms	Radiological findings
1	F, 1 month	resp. distress, cyanosis during feeding since birth	CXR:large air cyst at left hemithorax, heart&esophagus deviated to the right (Fig. 1)
2	M, 3.5 months	resp. distress, fever and cyanosis since 40 days of age	CXR: an air filled cyst of the left lung, CT:cystic mass in the left upper lobe
3	F, 10 months	cyanosis during crying and fever since birth	CXR:cystic mass of the right lung, CT:right intrapulmonary cystic mass with minimal fluid at base
4	F, 6 years	chronic cough	CXR:mass like mediastinal lesion

*resp. distress: respiratory distress
CXR: chest x-ray
CT: computerized tomography*

* Presented at the 7th International Congress of Pediatric Surgery (1992, Hamburg), Germany
Address: Hüseyin Özbey, Tatlıpınar Cad. Akyiğit Ap. No:20/4, Topkapı, İstanbul, Türkiye

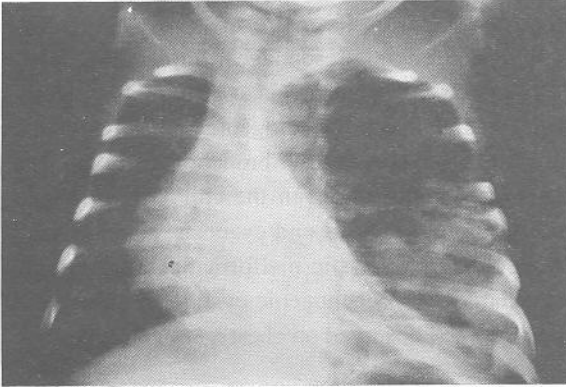


Figure 1. Posteroanterior chest x-ray in case 1. Heart and esophagus deviated to the right, due to the compression of the large air cyst in the left hemithorax.

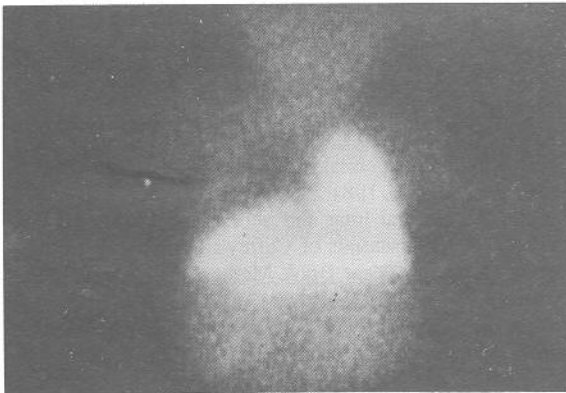


Figure 2. Perfusion scintigraphy in case 2; which shows no uptake in the left hemithorax.

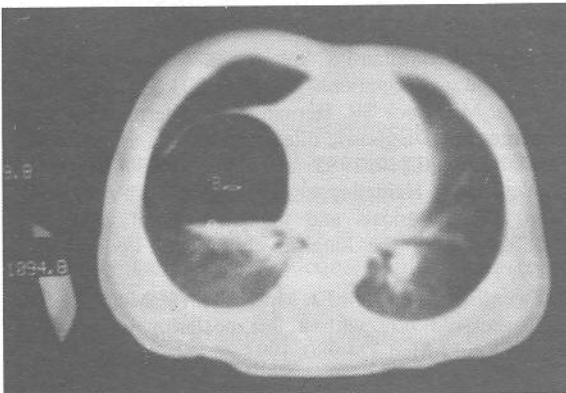


Figure 3. Intrapulmonary bronchogenic cyst, with minimal fluid at base (case 3).

hageal contrast study revealed normal gastrointestinal configuration. Perfusion scintigraphy displayed no uptake in the left upper lobe (Figure 2).

In case 3, the cyst was found intrapulmonary and infected probably after an attempt for drainage (Figure 3).

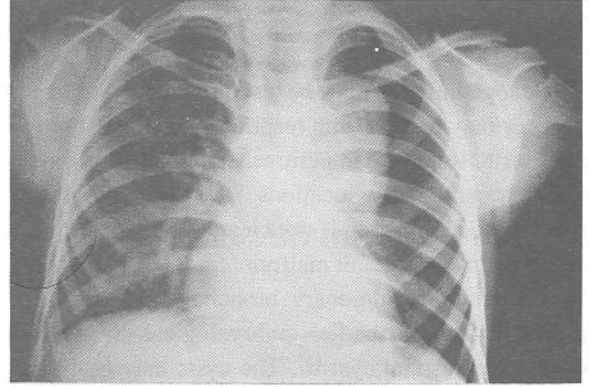


Figure 4. Posteroanterior chest x-ray in case 4. Note the mass image of left peritracheal bronchogenic cyst.

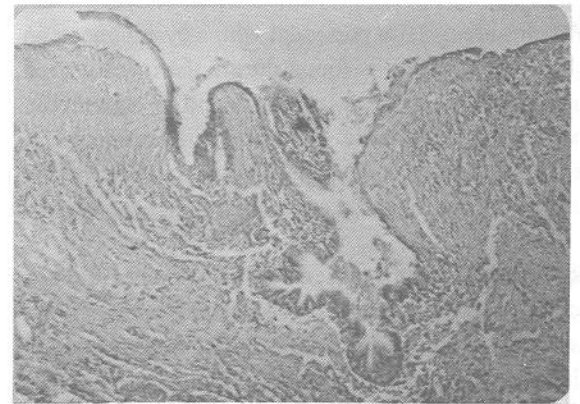


Figure 5. Squamous metaplasia at the junctional point of the bronchus and the cyst wall (case 2).

Case 4 (Figure 4) was misdiagnosed as mediastinal lymphoma at another medical center. At operation, a cyst, 5 cm in diameter, filled with mucoid material was found between the arcus aorta, pulmonary artery and trachea.

Three patients (case 1,3,4) were treated with cyst excision and one patient (case 2) was treated with lobectomy due to the gross involvement of the left upper lobe. All of the patients are symptom-free on their follow-up.

The histological examinations confirmed the diagnosis of bronchogenic cyst, with the findings of respiratory epithelium, fibrous tissue wall and nests of cartilage. In one of the cases (case 2), squamous metaplasia was observed at the junctional point of the bronchus and the cyst wall (Figure 5).

Discussion

Bronchogenic cyst may be asymptomatic for over years or may present with respiratory distress shortly after birth. It may be in peritracheal, hilar, subcarinal and intrapulmonary locations (11). Although their precise mode of origin is not clear, they are considered as primitive foregut malformations (1,10). It is believed that intrapulmonary bronchogenic cysts are probably from an earlier embryological error than does a mediastinal cyst (5). The cysts may be filled with clear or mucoid material and when they become infected the clinical symptoms of infection appear.

Patients are usually presented with a cystic image on chest x-ray, which is found during the investigation of respiratory distress, cyanosis and fever of unknown origin. Periodic episodes of dyspnea, wheezing and cyanosis, especially aggravated by crying or feeding are the usual symptoms (1,2,6,8,9).

Radiography of the patients show an either fluid or air filled cystic mass. Tension pneumothorax and infection are the complications of the diagnostic or therapeutic needle aspiration. Tension pneumothorax may cause rapid deterioration of the patient due to sudden mediastinal shift. Esophageal contrast study is the most useful and simple investigation in localizing the cyst and revealing any compression of the adjacent structures. CT must be the further investigation method which may also help in the differential diagnosis of the cystic image, among other cystic malformations of the lung. Bronchoscopic evaluation should not be performed due to the risk of airway obstruction (9). Incidentally found asymptomatic pulmonary cysts should also be removed because of their propensity for infection and association of malignancy (3).

Other congenital cystic malformations of the lung, diaphragmatic hernia, esophageal duplication cysts, pneumothorax, pneumatocele, pulmonary abscess, round pneumonia and neurogenic mass are in the differential diagnosis of the bronchogenic cyst. The newborns, infants and even the children, presented with respiratory distress and cyanosis, should create the suspicion of a cystic malformation of the lung, particularly of a bronchogenic cyst, if an air or fluid filled cyst was observed on chest x-ray.

References

1. Bailey PV, Tracy T, Connors RH, deMello D: Congenital bronchopulmonary malformations. *J Thorac Cardiovasc Surg* 99:597, 1990
2. Eraklis AJ, Griscom NT, McGovern JB: Bronchogenic cyst of the mediastinum in infancy. *N Engl J Med* 281:1150, 1969
3. Kraus HF, Sexauer CL: Embryonal rhabdomyosarcoma arising within a congenital bronchogenic cyst in a child. *J Pediatr Surg* 16:506, 1981
4. Kuhn JP: Normal lung and anomalies. Silverman FN, Kuhn JP (eds) "Essentials of Caffey's Pediatric X-ray Diagnosis". Chicago, Year Book Medical Publishers, 1990, p. 241
5. Luck SR, Reynolds M, Raffensperger JG: Congenital bronchopulmonary malformations. *Curr Probl Surg* 23: 245, 1986
6. Marshall CP, Cookson HA: Tracheobronchial cyst. *Lancet* 1:305, 1943
7. McMullin N, Doi O, Kent M: The spectrum of bronchopulmonary foregut malformations. *Pediatr Surg Int* 2:304, 1987
8. Rodgers B, Harman PK, Johnson AM: Bronchopulmonary foregut malformations. *Ann Surg* 203:517, 1986
9. Synder ME, Luck SR, Hernandez R, Sherman JO, Raffensperger JG: Diagnostic dilemmas of mediastinal cysts. *J Pediatr Surg* 20:810, 1985
10. Wesley JR, Heidelberger KP, DiPietro MA, Cho KJ, Coran AG: Diagnosis and management of congenital cystic disease of the lung in children. *J Pediatr Surg* 21:202, 1986
11. Zwerdling RG, Mark EJ, McNeely WF, McNeely BU: A 17-week-old boy with a left posterior intrathoracic mass. *N Engl J Med* 324:980, 1991