Atypical lung parenchymal bronchogenic cyst complicated by tuberculosis infection

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A bronchogenic cyst (BC) is a rare congenital lesion that may cause life-threatening organ compression in children, but is generally asymptomatic in adults unless there are other complications. In the present report, a 34-year-old woman in whom a BC was complicated by tuberculosis infection is described. Due to the small size of the BC, it was asymptomatic and could not be diagnosed until she was treated with antituberculosis medications.

Key Words: Bronchogenic cyst; Lung complication; Surgery; Tuberculosis

Learning objectives
● To recognize the characteristics of bronchogenic cysts (BCs).
● To understand that infection can make the diagnosis of a BC difficult.

Pretest
● What are the clinical characteristics of BCs?
● How are BCs diagnosed?

CASE PRESENTATION

A 34-year-old female presented to an outside hospital with chills, fever and left-sided chest pain for one month. Her highest temperature was 39.2°C and chest pain worsened with deep inspiration. She denied any cough, shortness of breath, hemoptysis or night sweats. Computed tomography (CT) revealed an indeterminate soft-tissue lesion in the left upper lobe. The patient was treated with ampicillin/sulbactam combined with levofloxacin. Her temperature returned to normal and chest pain was relieved after three days of antibiotics. After two weeks of antibiotics, however, there was no change in the mass on CT and she was transferred to the Shanghai Pulmonary Hospital (Shanghai, China). On admission, the patient denied any complaints, and physical examination revealed only coarse respiratory sounds. Laboratory studies revealed the following: a hemoglobin level of 132 g/L, a white blood cell count of 5.3 x 10^9/L (neutrophils 55%, lymphocytes 42%) and an elevated erythrocyte sedimentation rate of 27 mm/h. Levels of the tumour markers alpha-fetoprotein, carcinoembryonic antigen, neuron-specific enolase, carbohydrate antigen (CA) 242, and CA211, CA153, CA199 were normal. Purified protein derivative skin test was positive (induration, 18 mm with a blister). Bronchoscopy and pulmonary function testing revealed no abnormalities.

Bronchoscopy and pulmonary function testing revealed no abnormalities. Examinations of sputum and bronchoscopy brush samples for acid-fast bacilli, along with fungal, bacterial and tuberculosis cultures were negative. Chest radiography and CT (Figure 1A and 1B) revealed an indeterminate soft-tissue lesion in the left upper lobe, with fuzzy edges and uneven density (mean Hounsfield units [HU] 40; range 10 HU to 90 HU). No lymphadenopathy was noted. CT-guided fine-needle aspiration revealed necrosis and epithelial cell and lymphocyte infiltration, and cultures were positive for Mycobacterium tuberculosis. The patient was treated with isoniazid, rifampin, ethambutol and pyrazinamide for two months, and isoniazid and rifampin for an additional four months. After six months, CT demonstrated absorption and decrease in the size of the lesion; however, a 2 cm cavity was noted (Figure 1C).

Thoracotomy revealed a thick-walled cyst, 2 cm in size, with internal grayish-yellow caseous crisp necrosis in the lingual of the left upper lobe. The wall thickness was 0.5 cm, and there was no evidence of bronchial communication. The cyst was excised, and histopathological examination showed the cyst wall to be covered with pseudostratified columnar epithelium, with areas of squamous metaplasia (Figure 2A). Coagulation necrosis filled the cyst cavity with numerous epithelioid and multinuclear giant cells. A granuloma was noted on the cyst wall (Figure 2B), which was positive on acid-fast staining (Figure 2C). Staining for fungal hyphae and spores was negative. The final diagnosis was bronchial cyst (BC) with tuberculosis granuloma. At 4.5 years follow-up, no recurrence of tuberculosis (TB) or the cyst was noted.

DISCUSSION

A BC is a rare congenital cystic lesion occurring in the mediastinum (65% to 90%), and rarely occurs in extramediastinal areas such as lung parenchyma (1). They are usually diagnosed in neonates, infants and...
children because of organ compression caused by the cyst. In adults, compression is much less likely and BCs generally remain asymptomatic unless an infection occurs (1).

The BC in the present case was unique in location and size, and there was no air or air-fluid level in the cyst as is commonly found (2-6), making the diagnosis difficult. BCs of the lung are rare, they typically range in size from 2 cm to 10 cm, and can be easily detected on chest radiograph or CT scan (1). On radiographic studies, a BC typically appears as a mass with homogeneous density and well-defined margins (5). Normally, the attenuation value within the mass on CT is <20 HU (7). BCs can be misdiagnosed as a malignancy when they appear as a solid mass on CT (8).

A total of six case reports of intraparenchymal BCs complicated with mycobacterial infection have been published (2-4,6,9). In a report of two cases by Liman et al (3), cysts were detected by radiograph or CT, and the diagnoses of TB were made after thoracotomy. Detection of TB also only occurred after examination of a specimen of the cyst in three of the other reported cases (2,4,6). From these cases, it is clear that the symptoms of TB were not typical, and the mycobacterium was only isolated from cyst specimens. Unlike the cases reported previously, the BC in the present case could not be detected from imaging studies until the patient had been treated with anti-TB medications.

In summary, BCs complicated by infection may not be readily diagnosed with imaging studies. In cases of pulmonary TB or other infections in which a lesion remains after treatment, a BC may be considered in the differential diagnosis.

REFERENCES