



SHORT COMMUNICATION

Mycobacterial infection of intraparenchymal bronchogenic cysts

Serife Tuba Liman^{a,*}, Yesim Dogan^b, Salih Topcu^a,
Nevzat Karabulut^c, Nese Demirhan^d, Zehra Keser^e

^aThoracic Surgery Department, The Faculty of Medicine, Kocaeli University, Kocaeli, Turkey

^bThoracic Surgery Department, Denizli State Hospital, Denizli, Turkey

^cRadiology Department, The Faculty of Medicine, Pamukkale University, Denizli, Turkey

^dPathology Department, The Faculty of Medicine, Pamukkale University, Denizli, Turkey

^ePathology Department, Denizli State Hospital, Denizli, Turkey

Received 23 January 2006; accepted 3 March 2006

KEYWORDS

Bronchogenic cyst;
Surgery;
Complication;
Tuberculosis;
Mycobacterium

Summary Bronchogenic cysts (BCs) may rarely cause some interesting and unusual complications. Although infection is a common complication of BCs, there are only two patients with BC infected with mycobacterium in English literature. Two intraparenchymal BCs infected with mycobacterium are presented here as unusual complications. Cystectomy was performed for the cysts. They were given antituberculosis treatment. No complication or recurrences were detected in follow up period.

© 2006 Elsevier Ltd. All rights reserved.

Introduction

Bronchogenic cysts (BCs) are congenital anomalies, with originates from primitive foregut in embryonic life. Although they may cause life-threatening complications, they may be asymptomatic for years. Generally half of the BCs are symptomatic.

In the literature, there are great number of reports about complications of BC. Here we have presented very unusual complication in two BCs.

Case 1

A 20-year-old male patient was admitted to the hospital with the complaints of fever, cough, purulent sputum. Body temperature was 39 °C. Respiratory sounds were diminished in right lower zone. White blood cell count was 7000/00³. PPD tuberculosis skin test, sputum culture for nonspecific microorganisms were negative. Sputum smear and culture for tuberculosis were negative. In chest X-ray a mass was detected in right lower zone. Thorax tomography revealed a multiloculated cystic mass suspicious for cystic adenomatoid

*Corresponding author. Tel.: +90 262 303 7407.

E-mail address: tubaliman@yahoo.com (S.T. Liman).

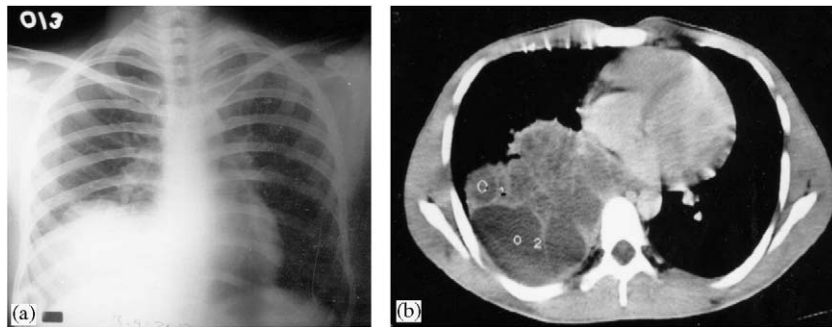


Figure 1 (a,b) Posterior anterior chest X-ray and thorax tomography demonstrating paracardiac multiloculated cystic mass in right lower lobe.

malformation or complicated hydatid cyst in the right lower lobe (Fig. 1). The patient underwent posterolateral thoracotomy. The $10 \times 8 \times 8$ cm cystic mass was found in the lower lobe. Necrotic material was aspirated from inside the cyst. There was no bronchial communication. The cystic lesion was excised totally via cystectomy. Culture for aerobic and anaerobic bacteria of necrotic material were negative. No specimen was submitted for mycobacterial culture. Pathological examination revealed a typical BC, the inner wall covered by ciliated columnar respiratory epithelium. Granuloma formation with epithelioid histiocytes, caseification and Langhans type giant cells were detected in the cyst wall. The patient had a history of BCG vaccination and neither he nor his family had any known previous tuberculosis exposure. He was given 9-month-tuberculosis treatment and had no further complications.

Case 2

A 32-year-old female patient was admitted to the hospital for cholecystectomy due to gallstones. She had no respiratory symptoms. Her physical and laboratory examinations revealed no abnormalities except an elevated sedimentation rate (35 mm/h). In preoperative chest X-ray, a solitary pulmonary nodule was detected in the right lung. On computed tomography, a round well-circumscribed cystic lesion was detected (Fig. 2). Microscopic examination of sputum for tuberculosis was negative and sputum culture was performed. The patient underwent right thoracotomy. Cystic lesion was located in the major fissure of the right lung. Necrotic material was emptied from inside the cyst and bronchial communication was observed. Bronchial opening was sutured and cystectomy was performed. Pathologic examination revealed BC

with granulomatous infection with caseification and Langhans type giant cells. She was given isoniazid treatment. After discharge from the hospital her sputum culture for tuberculosis was reported as positive. Culture showed bacterial resistance to isoniazid. Her tuberculosis treatment was changed. It was learned that there was no tuberculosis exposure in her past medical history. Her TB therapy continues and no complications have been observed.

Discussion

BCs are congenital lesions arising from the abnormal budding of the primitive foregut. The inner surfaces are lined by columnar ciliated epithelium. The cyst walls may contain cartilage, smooth muscle, elastic tissue and mucous glands. They can be located in mediastinum, pulmonary parenchyma, posterior sulcus and hilum of the lung and uncommon extrathoracic locations. They are frequently unilocular and may contain clear or hemorrhagic fluid, proteinaceous mucus, calcium or air.

Although some BCs are asymptomatic and are incidental findings upon radiography, at least half of cysts are symptomatic and complications are more common in symptomatic patients. Complications depend on cyst localization, size and communication with the tracheobronchial tree. The most frequent symptoms are cough, fever, pain and dyspnea.¹⁻³ Tracheobronchial compression and pulmonary infections are the most common complications. Pneumothorax, hemoptysis, vena cava syndrome, pleurisy, arrhythmias, stenosis of pulmonary artery, compression of airway, infection, hemorrhage, dysphagia, malignant transformation and tension BCs have been reported as uncommon complications of BCs.²⁻⁷

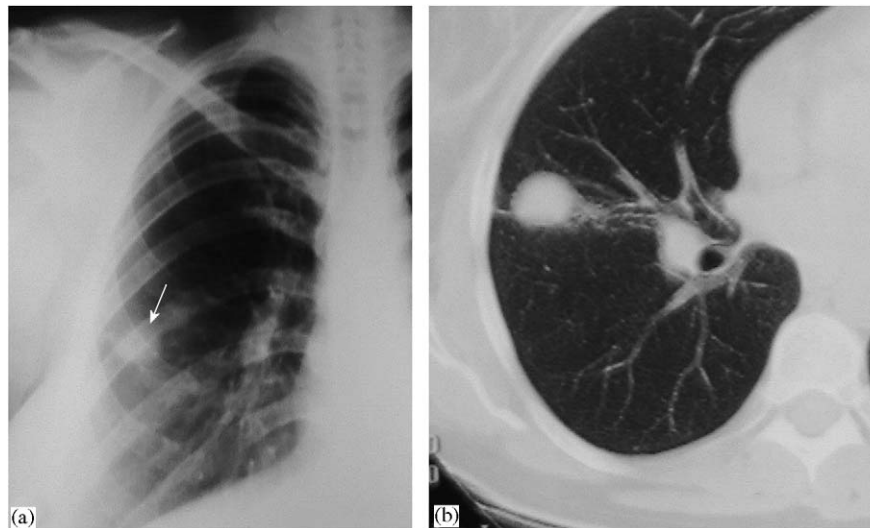


Figure 2 (a,b) Chest radiography and thorax tomography showing a mass with homogeneous density and well-defined margin, located major fissure.

Infection is a common complication especially in cysts with bronchial communication. There are many reports of infected BCs but only two reports on mycobacterium infected BCs.^{8,9} In one report, mycobacterium avium was isolated from specimen. In the other report, mycobacterium was demonstrated in the wall of the BC but the microorganism was not isolated. In both reports there were no other foci of tuberculosis in the lung or other tissues. In one of our cases, the sputum culture was positive and INH-resistant mycobacterium tuberculosis was isolated. Unfortunately, we could not culture mycobacterium in the other case but we were able to identify bacillus in acid fast staining of the specimen. Tuberculosis treatment was given to the patient even though mycobacterium tuberculosis was not identified.

The source of the infection has not been identified in either case. We found bronchial communication in one of two cysts. The possible route of mycobacterial infection of BC for these with communication to the lung may be direct ventilation. The communication may be microscopic or may close. Communication could not be demonstrated in our other case. Hematologic and lymphatic spread are improbably since there were no foci of mycobacterial infection in the other part of the bodies.

In conclusion, mycobacterium infection is a very rare entity for BCs but according to our experiences, it should be kept in mind in infected cystic

lesions and tuberculosis culture should be performed from both the specimen and from the material inside the cyst in all cases. Excision of asymptomatic BC should prevent further complications and allow definite diagnosis after pathological examination.

References

1. Kanemitsu Y, Nakayama H, Asamura H, Kondo H, Tsuchiya R, Naruke T. Clinical features and management of bronchogenic cysts: report of 17 cases. *Surg Today* 1999;**29**:1201–5.
2. Sarper A, Ayten A, Golbasi I, Demircan A, Isin E. Bronchogenic cyst. *Tex Heart Inst J* 2003;**30**:105–8.
3. Aktogu S, Yuncu G, Halilcolar H, Ermete S, Buduneli T. Bronchogenic cyst: clinicopathological presentation and treatment. *Eur Respir J* 1996;**9**:2017–21.
4. Ribet ME, Copin MC, Gosselin B. Bronchogenic cysts of the mediastinum. *J Thorac Cardiovasc Surg* 1995;**109**:1003–10.
5. Lardinois D, Gugger M, Ris H. Bronchogenic cyst of the left lower lobe associated with severe hemoptysis. *Eur J Cardiothorac Surg* 1999;**16**:382–3.
6. Altinok T, Topcu S, Kurul IC, Yazici U. Bronchogenic cyst with milk of calcium. *Eur J Cardiothorac Surg* 2002;**22**:311.
7. Dogan R, Cetin G, Moldibi B, Kaya S, Alp M, Ucanok K, et al. Pulmonary and mediastinal bronchogenic cysts. *Rev Mal Respir* 1988;**5**:123–7.
8. Lin SH, Lee LN, Chang YL, Lee YC, Ding LW, Hsueh PR. Infected bronchogenic cyst due to mycobacterium avium in an immunocompetent patient. *J Infect* 2005;**51**:131–3.
9. Houser WC, Dorff GJ, Rosenzweig DY, Aussem JW. Mycobacterial infection of a congenital bronchogenic cyst. *Thorax* 1980;**35**:312–3.