Case report

Bronchogenic cyst of the left lower lobe associated with severe hemoptysis

Didier Lardinois*, Matthias Gugger, Hans-Beat Ris

Department of Thoracic and Cardiovascular Surgery, Division of Pneumology, University of Bern, 3010 Bern, Switzerland

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Abstract

Bronchogenic cysts result from congenital disorders, are often asymptomatic at diagnosis, but complications are not uncommon. We report the case of a 19-year-old woman with severe hemoptysis. This rare presentation of an intrapulmonary bronchogenic cyst should be considered as differential diagnosis in patients with cavernous lesion of a lobe. Surgery was performed as a diagnostic and therapeutic measure. © 1999 Elsevier Science B.V. All rights reserved.

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1. Case report

A 19-year-old nurse from Sri Lanka presented with acute, massive hemoptysis with hypotension and collapse. She lived in Switzerland since 1986. The medical history revealed a pneumonia on the right side treated with antibiotics in 1985 but no tuberculosis or other infectious diseases. The history of her family was also uneventful. The patient had neither fever, cough, sweating nor dyspnea. On the chest X-ray, a sharply defined, solitary cavernous lesion of the left lower lobe was found. The white blood count and blood chemistry showed no pathological findings, especially no signs of infection. The hemoglobin value fell from 13.7 to 10.1 g/dl. The patient was transferred to the intensive care unit with suspicion of active tuberculosis. An antituberculous chemotherapy with four drugs was started. CT scans showed a 6 × 5 × 4 cm large, thin-walled cavernous lesion of the left lower lobe without any parenchymal infiltrations (Fig. 1); a bronchoscopy showed no pathological findings, except blood clots in the left lower lobe bronchus. A thoracotomy with resection of the left lower lobe bronchus was performed on the next day to prevent bleeding recurrence (Fig. 2). There were no postoperative complications and the patient was discharged 8 days later. Histology revealed a cyst communicating with the bronchial tree. The wall of the cyst contained smooth muscle, bronchial mucus glands and cartilage. No epithelioid granulomas and no abscesses were found. Cultures showed no fungi, no mycobacteria, no parasites.

2. Discussion

Bronchogenic cysts are lesions of congenital origin derived from the primitive foregut [1]. Fifty to 80% of the patients are symptomatic on presentation but an incidental finding on radiographic examination is not rare. They are slightly more common in men and are mainly discovered in the third or fourth decade of life [1,2]. The most frequent symptoms are pain, cough, fever or dyspnea, but severe hemoptysis is rarely reported [3,4]. Although more commonly located in the mediastinum, intrapulmonary cysts represent about 15–20% of all the bronchogenic cysts and usually occur in the lower lobes [5]. Most of them have no patent communication with the tracheobronchial tree. Most of the complications of bronchogenic cysts result from compression of mediastinal structures, infection, or rarely, changing into a malignant tumor [6,7]. The differential diagnosis of the parenchymal form includes lung abscess, hydatid cyst, fungal diseases, tuberculosis, infection with nocardia, infected bulla, vascular malformation and neoplasm [8]. In our case, the preoperative chest X-ray showed no calcification (hydatid cyst) and no air fluid level (abscess). Contrast fluid did not fill the lesion (vascular malformation). The location in the lower lobe, the thin wall of the cyst without surrounding parenchymal pathology, and the absence of acid-fast bacilli in the bronchial secretions were atypical for tuberculosis. However, because of the
origin of the patient and the increased prevalence of tuberculosis in immigrants, a mycobacteriosis could not be excluded completely. CT scan revealed a homogeneous content of a part of the cavernous lesion, suggesting blood, but the content was indistinguishable from tuberculous pus or aspergilloma. It is well known that aspergillomas can produce a massive hemorrhage, making this diagnosis possible as well. The resection was mainly performed to prevent a second episode with massive hemoptysis, but also to obtain a definitive diagnosis. In the case of tuberculosis or an aspergilloma with severe hemoptysis, the operative procedure seems to be the therapy of choice. In our case, an intrapulmonary bronchogenic cyst was histologically diagnosed without isolation of pathogens. A review of the literature reveals that surgery represents the therapy in symptomatic bronchogenic cysts and some authors recommend the resection of all bronchogenic cysts, asymptomatic or not [1,3,9]. The resection must be complete because recurrence has been observed after incomplete surgical removal. On the basis of the size of the lesion, a segmentectomy was not possible and the whole lower lobe had to be resected. The therapy of asymptomatic bronchogenic cysts remains controversial but the majority of authors seem to advocate a surgical approach to obtain a diagnosis and to prevent complications, which occur in up to 80% in patients with symptomatic cysts [1,3].

In conclusion, severe hemoptysis is a rare complication of intrapulmonary bronchogenic cysts and may be the only symptom at presentation time. Bronchogenic cysts should be considered in the differential diagnosis of patients with severe hemoptysis, especially because a definitive confident preoperative diagnosis is not always possible on the basis of standard investigations.

References