

Case Report

Impact of the bronchopulmonary sequestration on endobronchial tuberculosis: the case report and the review of literature

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Abstract

Introduction: We describe the rare case of endobronchial tuberculosis (EBTB) and chronic pulmonary atelectasis with mediastinal distortion. Finding of the concomitant venous anomaly of inferior vena cava revealed the diagnosis of bronchopulmonary sequestration.

Case Report: A 22-year-old Caucasian woman presented with a history of chronic cough, initially treated as bronchial asthma for a year. Chest X-ray showed fibrocavicular tuberculosis on the right lung. Acid Fast Bacilli (AFB) were found in sputum samples. Patient was treated for 6 months with usual antituberculous regimen. Control chest X-ray showed subatelectasis of the upper right lobe. Six months later the first thorax computed tomography (CT) showed complete atelectasis of the right lung. Patient was admitted to the hospital again after 6 years due to the persistent fever and cough. Endoscopic finding and histopathological analysis confirmed EBTB. Thoracic CT scan revealed duplication of inferior vena cava which led to profound vascular analysis and aberrant arterial vascularization of aortic origin that contributed to the diagnosis of bronchopulmonary sequestrations. Antituberculous treatment was initiated (streptomycin, isoniazid, rifampicin, ethambutol and pyrazinamide) and lasted for 8 months. After 8 months a follow-up fiberoptic bronchoscopy showed the progression of endoscopic finding with 60-70% tracheal stenosis. Histopathological finding of the mid-trachea showed non-specific granulations. During 7 years of follow-up repeated bronchoscopy and thoracic CT scans were unchanged and patient was well-shaped.

Conclusions: The clinician should consider bronchopulmonary sequestration in the cases of recurrent EBTB.

Key words: Pulmonary sequestration; endobronchial tuberculosis; treatment.

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Introduction

In rare cases of recurrent tuberculosis (TB) including endobronchial tuberculosis (EBTB), bronchopulmonary sequestration may be a predisposing factor [1]. Only five cases of TB and bronchopulmonary sequestration have been described in the literature [2-6], while there is none any published data on EBTB complications. We present the rare case of recurrent EBTB, initially treated as bronchial asthma for a year. A chest computed tomography (CT) revealed signs of active and chronic TB infection, and pulmonary sequestration with aberrant arterial supply of aortic origin and duplication of inferior vena cava (IVC).

Case Report

A 22-year old female patient, a non-smoker, was admitted to the hospital due to fever (38.5 °C), cough, and sweating. She was complaining of dyspnea and cough she had for a year, which had been treated as bronchial asthma at the regional hospital. She lived in a good social-economic environment, and was vaccinated at birth against TB.

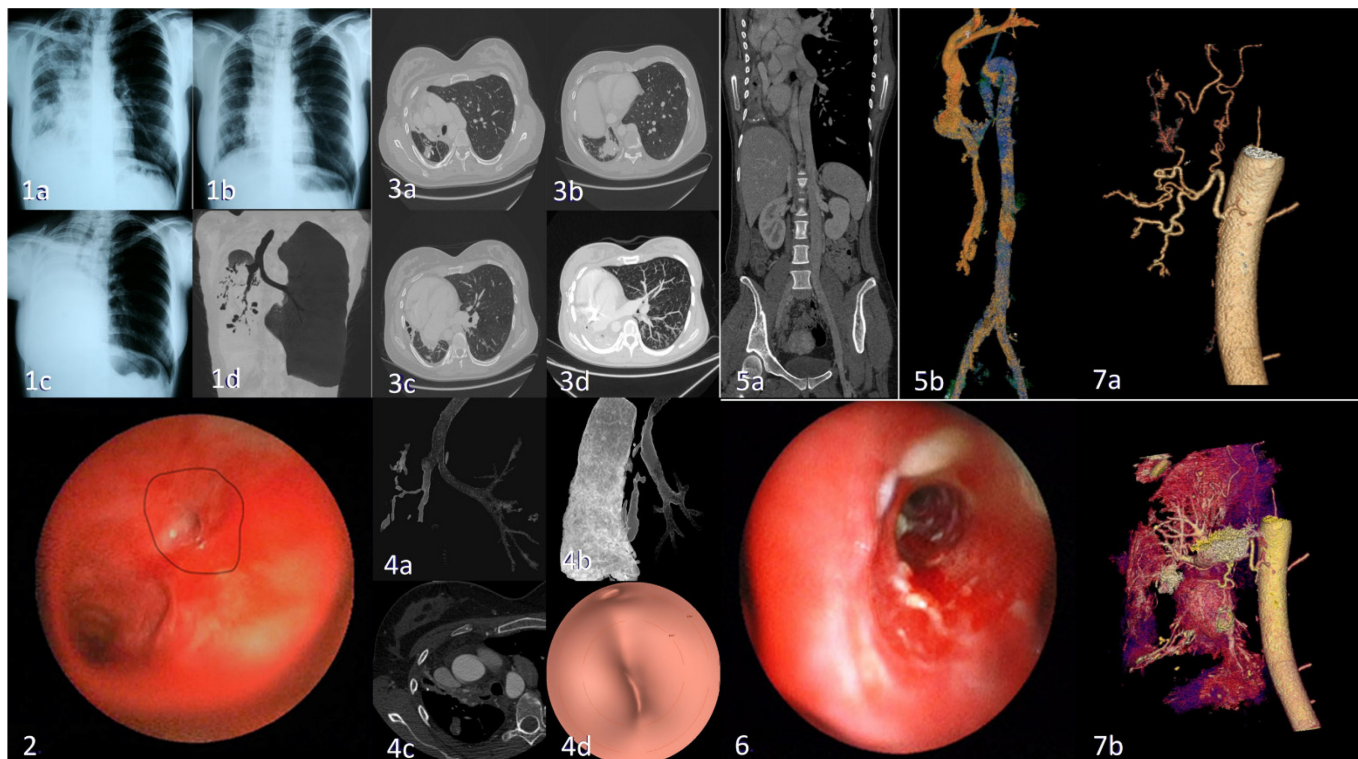
At presentation, the patient was in good clinical condition, with blood pressure 120/80 mmHg, heart rate of 80 bpm, respiratory rate 12 bpm, and body temperature 37.8°C. Ear, nose, and throat findings were normal. Decreased breath sounds over the right lung were noticed. A routine laboratory test, including blood cell count, was within the reference range, and

erythrocyte sedimentation rate was mildly elevated. Initial electrocardiogram showed sinus tachycardia with normal axis, segments, and intervals. Chest X-ray showed patchy confluent lung changes on the right lung with opaque elements corresponding to fibrocavous-cavernous tuberculosis (Figure 1-1a). Direct sputum microscopy confirmed acid-fast bacilli (AFB), Lowenstein-Jensen cultures confirmed the presence of *Mycobacterium tuberculosis*, sensitive to all first-line antituberculous drugs. Treatment with 4 antituberculous drugs was initiated in the standard dose regiment (isoniazid, rifampicin, ethambutol, and pyrazinamide) and lasted 6 months. At the end of the treatment, the control chest X-ray showed subatelectasis of the upper right lobe (Figure 1-1b). No further diagnostic procedures were done.

Six months later, the patient presented to the regular check-up. AFB and Lowenstein-Jensen cultures were negative. Lung function tests showed: FVC 54% (1.97L), FEV1 55% (1.75L), FEV1/FVC 89%, indicating restrictive ventilator disorder. Chest X-ray

showed atelectasis of the right lung (Figure 1-1c). The first chest CT was done and confirmed complete atelectasis of the right lung (Figure 1-1d). Fiberoptic bronchoscopy was performed, and endoscopic findings showed stenosis of the right main bronchus, impassable to flexible bronchoscope (Figure 1-2). The patient skipped her regular check-ups for the next 6 years. After more than 6 years, she was admitted to the hospital for the second time due to the persistent fever and cough. AFB was found in the sputum samples again and Lowenstein-Jensen cultures were positive for the presence of *M. tuberculosis*. Chest X-ray was unchanged. A new chest CT found depicted loss of volume of right lung accompanied by traction of mediastinal structures. Zones of atelectasis of lung tissue, described at the chest CT 6 years ago, had developed to mass lesions presented with calcifications and necrotic centers, surrounded by “tree-in-bud” opacities (Figure 1-3a, 1-3b), with developing cavern in the middle lobe (Figure 1-3c). Also, the “tree-in-bud” pattern and thickening of a bronchial segmental branch (Figure 1-3d) suggested activation of TB infection; 4a, b, c, d) A loss of lower lobar bronchus branching was present and all visible right bronchial walls were irregular due to fibrosis; 5a, b) Abdominal CT scan revealed the presence of two inferior vena cavae (IVC) and 6) Control fiberoptic bronchoscopy revealed deviation of the tracheobronchial tree axis to the right, right main bronchus stenosis, while tracheal mucosa showed prominent inflammation, covered with whitish nodules and necrotic debris.

Figure 1. 1a) Initial chest X-ray showed patchy confluent lung changes on the right lung with opaque elements corresponding to fibrocavous-cavernous tuberculosis; 1b) The control chest X-ray showed subatelectasis of the upper right lobe; 2) Fiberoptic bronchoscopy was performed, and endoscopic findings showed stenosis of the right main bronchus, impassable to flexible bronchoscope; 3a, b, c) The control CT scan revealed mass lesions presented with calcifications and necrotic centers, surrounded by “tree-in-bud” opacities with developing cavern in the middle lobe; 3d) The “tree-in-bud” pattern and thickening of a bronchial segmental branch of the left lower lobe suggested activation of TB infection; 4a, b, c, d) A loss of lower lobar bronchus branching was present and all visible right bronchial walls were irregular due to fibrosis; 5a, b) Abdominal CT scan revealed the presence of two inferior vena cavae (IVC) and 6) Control fiberoptic bronchoscopy revealed deviation of the tracheobronchial tree axis to the right, right main bronchus stenosis, while tracheal mucosa showed prominent inflammation, covered with whitish nodules and necrotic debris.



of the left lower lobe suggested activation of TB infection (Figure 1-3d). The right main bronchus was narrowed. There was a loss of lower lobar bronchus branching (Figure 1-4a, 1-4b, 1-4c, 1-4d). All visible right bronchial walls were irregular due to fibrosis.

Section of retrorural space showed dilated hemiazygos vein, and additional CT of the abdomen detected the presence of two inferior vena cavae (IVC). The left-sided IVC was a dominant vein that drained the blood of the lower extremities, pelvis, and left kidney, following the pathway of hemiazygous vein. Right-sided IVC carried out the blood of right kidney and hepatic veins and drained to right atrium normally (Figure 1-5a, 1-b).

Another fiberoptic bronchoscopy was performed and showed deviation of the tracheobronchial tree axis to the right. Tracheal mucosa showed prominent inflammation, covered with whitish nodules and necrotic debris (Figure 1-6) with right main bronchus stenosis. Left main bronchus and left bronchial tree were unchanged. Biopsy of the tracheal mucosa was performed with histopathological findings of specific granulomatous bronchitis and caseous tuberculosis.

Due to TB reactivation and development of tracheal EBTB electrocardiographic (ECG) gated chest CT was done, with aim to investigate vascular supply of lungs and heart, considering anatomic variations of IVC. CT showed progression of existing chronic inflammatory lung lesions. There was irregular cavern of the middle lobe without contact to bronchial tree as well as stenosis of right main bronchus and trachea. Vascular supply of pulmonary artery origin and venous drainage by pulmonary veins were intact. In addition, aberrant blood vessel of thoracic aortic origin was detected that carried out vascular supply to all persistent mass lesions and cavern of right lung and arose suspicion of existence of bronchopulmonary sequestrations (Figure 1-7a, 1-b). Microbiology analysis showed that *M. tuberculosis* was sensitive to all standard antituberculous drugs and category II regimen of antituberculosis treatment was initiated (streptomycin, rifampicin, isoniazid, pyrazinamide, ethambutol) and lasted 8 months.

After 8 months, a follow-up fiberoptic bronchoscopy showed the progression of endoscopic finding. Approximately 2 to 3 cm from vocal cords, 60-70% tracheal stenosis was found with hyperemic, vulnerable mucosa. The rest of the trachea, starting from this point, was also stenotic and deformed, while the right main bronchus was impassable for the bronchoscope. There was no progression of the inflammation to the left bronchial tree. The histopathological finding of the biopsy specimen from the mid-trachea showed non-specific granulations. AFB was not detected. A thoracic surgeon was consulted for possible surgical intervention. Although right pneumonectomy was a possible solution, it was not considered as the first choice because of the nonspecific inflammation of the trachea and there was a high risk of fistula as the adverse consequence of the surgery.

In the following period of 6 years, the patient was in good general condition. Her lung function tests were practically unchanged FVC 57% (2.08L), FEV1 47% (1.49L), FEV1/FVC 71.6%, with the same radiographic findings of atelectasis of the right lung and unchanged endoscopic findings. Blood gass analyses very in a normal range, with SaO₂ 97%.

Discussion

We describe a rare case of EBTB and bronchopulmonary sequestrations. To our knowledge, there are five case reports with pulmonary TB and bronchopulmonary sequestrations, but no any describing complications by EBTB (Table 1). Usually, there is one infected pulmonary sequestration, while in our case there are at least three (upper, middle, and lower lobe). All except one were mass-forming, while that one in the middle lobe was in a form of a cavern. We could not detect whether they were congenital or acquired, as her first imaging was at the age of 22, when the disease was manifested. Usually, bronchopulmonary sequestrations are diagnosed when becoming symptomatic due to infection. The limitation of the present case is the absence of surgical findings. Although the initial idea was to treat patient surgically, the development of tracheal EBTB disabled forming of

Table 1. Review of the literature on reported cases of bronchopulmonary sequestration infected with tuberculosis.

Reference	Gender	Age	Localization	Lung lesion revealed with X-ray or CT scan	Position	Treatment
2	M	36	Left lower lobe	Mass lesion	Abdominal aorta	medicament
3	M	61	Left lower lobe	Mass lesion	Abdominal aorta	surgical
4	F	22	Right lower lobe	Mass lesion	Descending thoracic aorta	medicament
5	M	17	Left lower lobe	Consolidation with cavitation	Descending thoracic aorta	surgical
6	M	19	Left lower lobe	Consolidation	Descending thoracic aorta	surgical

proper anastomosis due to the existence of granulation tissue and high localization. In favor of our decision are studies showing a high incidence of anastomosis complications, such as bronchopleural fistula or dehiscence in patients with EBTB [7, 8]. Bronchopulmonary sequestration is a rare disorder, frequently unrecognized until becoming infected [9]. Independent of etiology, congenital or acquired, continuous infection such as TB may lead to serious consequences [1]. Standard first-line treatment of pulmonary TB consists of isoniazid, rifampin, pyrazinamide, and ethambutol. The combination of medicaments is given for 2 months in the initial phase of treatment while the continuation phase for drug-susceptible TB includes isoniazid and rifampin for a further 4 months. Disease relapse extends treatment for 2 months. Factors known to be associated with TB reactivation are extensive lung changes like cavitation, positive sputum culture after 2 months of treatment failing to gain weight during treatment [10]. TB reactivation in presence of venous anomaly, not so uncommon due to intricate development, [11] raises suspicion of accompanying lung anomaly. Patients suffering from EBTB usually have persistence of some degree of bronchial obstruction and this may be the explanation why the patient was treated for bronchial asthma at the beginning. Bronchoscopy revealed stenosis of the right main bronchus and it was confirmed by a chest CT. Additional imaging showed the presence of venous and arterial anomaly consisting of the aberrant arterial supply of aortic origin to few mass-like focuses at right lung and multiple pulmonary sequestrations.

In conclusion, we point out that the clinician should pay attention to the rare cases of pulmonary TB with bronchopulmonary sequestration in order to avoid maltreatment.

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