Tuberculosis presenting as isolated bronchonodal fistula in a patient with systemic lupus erythematosus

Case report
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Abstract

Rationale: Lymph node is a preferred site for extrapulmonary tuberculosis (TB). In the thorax, mediastinal tuberculous lymph nodes can erode adjacent structures such as heart, aorta, and esophagus, forming fistula, and causing fatal consequences. However, tuberculous bronchonodal fistula as a complication of lymph node TB in adults is rarely known in terms of imaging or clinical findings. Here, a case of isolated tuberculous bronchonodal fistula appearing as the first presentation of TB in a 74-year-old male with systemic lupus erythematosus (SLE) is reported.

Patient concern: A 74-year-old male with SLE visited the hospital with dry cough. In family history, his son was treated for pulmonary TB 9 years previously. Laboratory test revealed increased C-reactive protein level and erythrocyte sedimentation rate. Chest computed tomography (CT) scan revealed a necrotic lymph node in the right hilar area connected to the inferior wall of the right upper lobe bronchus and the lateral wall of bronchus intermedius.

Diagnoses: On bronchoscopy performed under guidance of 3-dimensionally reconstructed CT image, fistula formation between the right hilar lymph node and 2 bronchi (the right upper lobe and intermediate bronchus) was confirmed. Sputum culture revealed growth of Mycobacterium tuberculosis.

Intervention: Anti-TB medication with isoniazid, ethambutol, pyrazinamide, and moxifloxacin for 9 months.

Outcome: The patient’s symptom was gradually improved. Follow-up bronchoscopy performed at 3 months after starting the medication revealed decreased size of the fistula.

Lessons: This is a rare case of bronchonodal fistula appearing as the first presentation of TB in a 74-year-old male patient with SLE. CT provided useful information regarding the origin and progress of the disease.

Abbreviations: CT = computed tomography, SLE = systemic lupus erythematosus, TB = tuberculosis.

Keywords: bronchonodal fistula, endobronchial tuberculosis, lymph node, SLE, tuberculosis

1. Introduction

Patients with systemic lupus erythematosus (SLE) have increased risk of pulmonary and extrapulmonary tuberculosis (TB) due to intrinsic and extrinsic immune suppression.\textsuperscript{[1]} Lymph node TB is the most common form of extrapulmonary TB. In the chest, Mycobacterium tuberculosis can enter the lymphatic system from parenchymal lesions and cause mediastinal and hilar lymphadenopathy due to caseation necrosis and granulation tissue formation.\textsuperscript{[2]} Tuberculous lymph nodes in the mediastinum can erode adjacent structures, including esophagus, heart, aorta, and airways through inflammation and necrosis, causing fatal consequences sometimes.\textsuperscript{[3-6]}

Airway involvement by enlarged TB lymph nodes occurs mostly in children who have small-caliber airways. Bronchial compression and bronchonodal fistula formation by enlarged mediastinal/hilar TB lymph nodes in children with active pulmonary TB have been reported previously.\textsuperscript{[7,8]} However, to the best of our knowledge, tuberculous bronchonodal fistula in adult patient, particularly in those without concomitant parenchymal lesion, has not been reported yet.

Herein, a case of tuberculous bronchonodal fistula occurring in a 74-year-old male patient with SLE as the first presentation of TB
is reported, emphasizing the important role of chest computed tomography (CT) in diagnostic process and understanding the pathogenesis of tuberculous bronchonodal fistula.

2. Case report

Informed consent for publishing this report and any accompanying images was obtained from the patient. A 74-year-old male farmer visited our hospital due to stiffness and progressive deformity of finger joints dating back 10 years. His father had the same problem. He thought their symptoms were due to long-term farm working. On physical examination, his conjunctivae appeared pale. There were ulcers in the oral cavity. Laboratory examination showed leukopenia (3900/mm³) with increased C-reactive protein level (40.7 mg/L) and erythrocyte sedimentation rate (104 mm/h). Tests for detecting autoantibodies such as fluorescent antinuclear antibody (1:160), anti-ds DNA IgG, and anti-cardiolipin IgG were positive. After all evaluations, the patient was diagnosed with SLE. Chest CT scan showed emphysematous change in lungs and a few borderline-sized or enlarged lymph nodes in the mediastinum and hilar area (Fig. 1). There was no interstitial lung disease or active parenchymal lesion. He started medications for SLE with oral corticosteroid (prednisolone), 10mg daily and hydroxychloroquine 200 mg daily.

Three months later, the patient visited the hospital with dry cough that lasted 2 weeks. Laboratory test revealed increased C-reactive protein level and erythrocyte sedimentation rate. The physician suspected pneumonia. Since there were no obvious infiltrates on chest radiographs, chest CT scan was performed. Chest CT scan showed a cavity in the right hilar area in which an enlarged lymph node was noted initial chest CT. The cavity was connected to the inferior wall of the right upper lobe bronchus and the lateral wall of bronchus intermedius on 3-dimensionally reconstruction image (Fig. 2). There was no parenchymal lesion suggestive of active pulmonary TB draining into the bronchus. Bronchoscopy was recommended with a suspicion of bronchonodal fistula. However, one of the fistulas was missed on the initial examination. On repeat bronchoscopy performed after reviewing CT images, focal mucosal defects covered with necrotic tissue were noted at the right upper lobe bronchus and bronchus intermedius, corresponding to the location of bronchial fistula on CT (Fig. 3). Caseous material draining into bronchus was seen. Bronchial mucosa was clear in other areas. Polymerase chain reaction using bronchial aspirate and sputum culture revealed *M tuberculosis*. In family history, his son was treated for pulmonary TB 9 years previously. Since drug sensitivity testing showed resistance to rifampin, anti-TB medication was started with isoniazid 300 mg daily, ethambutol 800mg daily, pyrazinamide 1500mg daily, and moxifloxacin 400mg daily. Dry cough was improved in 2 weeks after beginning the treatment. Now, the patient is under treatment for 6 months on a 9-month course of medication. Follow-up bronchoscopy performed at 3 months after starting the medication revealed decreased size of fistula. Although medication for SLE has stopped with the starting of anti-TB treatment, SLE-associated symptoms are stable.
3. Discussion

SLE is a chronic inflammatory autoimmune disease. Its estimated prevalence ranges from 16 to 70 per 100,000 persons.[] Infection is a major cause of morbidity and mortality among patients with SLE. In particular, the risk of TB has been reported to be 5- to 60-fold higher in patients with SLE than that in those without SLE. Their susceptibility to TB can be explained by immunologic disturbance intrinsic to SLE, long-term administration of immunosuppressive drugs such as corticosteroids, and cross-reactivity between mycobacterial cell wall glycolipids and antinuclear antibody.

This is a rare case of bronchonodal fistula appearing as the first presentation of TB in a 74-year-old male patient with SLE. Initial or follow-up chest CT of the patient did not show pulmonary TB. TB involving lymph nodes showed nonspecific findings on initial CT for this patient. In addition, the patient did not complain of any TB-related symptoms. Therefore, the diagnosis was established relatively late during the course of disease, when the fistula had already formed between the right hilar lymph node and 2 bronchi. Relatively long course of disease might have been reactivated and rapidly progressed, ultimately forming fistula with adjacent bronchi.

Diagnosis of extrapulmonary TB and related complications can be delayed in immunocompromised patients due to nonspecific clinical presentation and frequent concomitant medical problems. In this study, initial and follow-up CT provided useful information regarding the origin and progress of the disease. Especially, 3-dimensionally reconstructed CT image of tracheobronchial trees helped the physician understand the disease status and guided bronchoscopic examination. On initial bronchoscopy, the physician missed the lesion in the bronchus intermedius without understanding imaging findings. Therefore, the physician had to repeat bronchoscopic examination after reviewing and discussing CT findings. According to Park et al, bronchoscopist can overlook the presence of fistula because the fistulous tract is often concealed and covered by exudate or necrotic tissue. They might consider the disease as a mere endobronchial TB. Thus, the presence and precise location of fistula should be documented through CT scan before bronchoscopy.

Tuberculous bronchonodal fistula can be treated with anti-TB medication. According to microbiological examination of sputum and bronchial aspirates, anti-TB medication was started with isoniazid, ethambutol, pyrazinamide, and moxifloxacin. Follow-up bronchoscopy showed decreased size of fistula.

In summary, a rare case of isolated bronchonodal fistula in an elderly male patient with SLE which occurred as the first presentation of TB is reported here. In immunocompromised patients, imaging studies may play crucial role in patient care. Serial chest CT scans with multiplanar reconstruction and 3-dimensional reconstruction can help physicians understand disease progression and current status, thus aiding a definite diagnosis.

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