Two cases of bronchial anthracofibrosis combined with tuberculosis

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Abstract:

BACKGROUND: Bronchial anthracofibrosis is a common disease that has been reported in the past. We aim to summarize the clinical characteristics of bronchial anthracofibrosis combined with tuberculosis infection to reduce missed diagnosis.

METHODS: The clinical features of two cases of bronchial anthracofibrosis combined with tuberculosis were analyzed retrospectively, and relevant studies were reviewed.

RESULTS: The two patients were both elderly individuals who presented with chronic cough and expectoration. Pigmentation in the bronchus mucosa and stenosis of lumen were observed during bronchoscopy. Tuberculosis infection was confirmed by biopsy. The symptoms were remarkably relieved and no recurrence was found after anti-tuberculosis treatment.

CONCLUSION: Bronchial anthracofibrosis may be combined with tuberculosis. To avoid misdiagnosis, we should be aware of possible tuberculosis infection when patients are diagnosed with bronchial anthracofibrosis.

Keywords:
Bronchial anthracofibrosis, clinical characteristics, tuberculosis

Bronchial anthracofibrosis is a common disease that has been frequently reported in the past. Chung et al. named it in 1998. It refers to bronchial stenosis or obstruction with anthracotic pigmentation in the mucosa in patients who are not exposed to dusty environments and without smoking history. Lymph nodes adjacent to the bronchus can be enlarged and calcified; computed tomography (CT) often reveals atelectasis of adjacent lung tissue. Bronchial anthracofibrosis also presents as proliferation of fibrous tissue in mediastinum. This proliferation can cause stenosis of the adjacent trachea, esophagus and pulmonary vessels, possibly leading to superior vena cava syndrome. At first, many researchers reported it as an independent clinical entity (i.e., independent clinical, pathological, radiological, and bronchoscopic characteristics). These patients began to be noticed by researchers and such reports gradually increased. A segment of the patient population has concomitant tuberculosis infections. Researchers realized that bronchial anthracofibrosis is closely related to active or latent pulmonary tuberculosis. At present, there are few reports of bronchial anthracofibrosis combined with tuberculosis in China and abroad. In recent years, we encountered two cases in clinical practice. Here, we review the clinical features of these patients, aiming to raise awareness of disease.

Case Report

Case 1 is a female, 68-year-old. She complained chronic cough, expectoration, and wheezing for more than 10 years,
with recurrence associated with intermittent fever for 1 month. She was hospitalized. The patient denied particular exposure history and tuberculosis infection history. Symptoms were cough, expectoration, and wheezing with large amount of white mucilage phlegm that occurred intermittently more than 10 years ago. Symptoms tended to reoccur during cold weather in autumn and winter. She was diagnosed with chronic bronchitis at a local hospital. Her symptoms were relieved after treatment in the local hospital for an exacerbation. No bronchoscopy had been performed during the course of her illness. Due to the aggravation of symptoms, she was treated in a local hospital 7 months prior to admission. The usual treatment had poor effects this time, and she came to our hospital for further diagnosis and treatment. Chest CT revealed large exudative foci and consolidation in the left lower lung. Bronchoscopy revealed stenosis in the left upper and lower lobes with pigmentation. The initial diagnosis was community-acquired pneumonia [Figures 1 and 2]. Cefoperazone sulbactam and levofloxacin were given empirically. The patient was discharged after 10 days’ treatment. Symptoms were greatly relieved at discharge.

One month before the second admission, the patient presented with intermittent low fever in the afternoon. Levofloxacin were given in the local hospital for 1 week. The patient was hospitalized in our hospital again because her body temperature was unstable. Physical examination showed slight dullness to percussion in the left lower lung, dry rales scattered in both lungs, wet rales in the left lower lung, no pleural friction. Chest CT showed that there remained a large exudative lesion and consolidation in the left lower lung, which had no significant improvement compared with previous chest CT [Figure 3]. Bronchoscopy showed that the left bronchus was narrow with pigmentation, the lumen of the left lingual lobe and the dorsal segment of the lower lobe were obstructed. During brush biopsy, there was quite some bleeding in the left lower lobe. Acid-fast bacilli were negative in the smear and sputum.
The initial diagnosis of second admission was left lower lung obstructive pneumonia and chronic obstructive pulmonary disease. Cefoperazone sulbactam and etimicin was given for anti-infective treatment, but with poor effect. With no obvious inducement, the patient presented sudden hemoptysis of 300 ml on the 5th day of hospitalization. Vasopressin was given along with other hemostatic treatment. Because of the recurrent fever, the poor effect of anti-infection treatment and the sudden hemoptysis, our department discussion concluded that tuberculosis was highly suspected. CT guided lung biopsy was performed with the consent of family members. Pathological reported granulomatous inflammation of the left lower lung with coagulative necrosis. Taking the clinical manifestation into account, the final diagnosis was secondary tuberculosis (lower left, smear negative, and primary treatment). Cough and shortness of breath were relieved, body temperature returned to normal without hemoptysis and her weight increased 5 kg following initiation of anti-tuberculosis treatment (isoniazid, rifampicin, pyrazinamide, and ethambutol). During 2 years’ follow-up, she showed no cough or wheezing. The large exudative consolidation shadow of the left lower lung was absorbed and substantially dissipated [Figure 4].

Case 2 is a male, 74-year-old. He was hospitalized for repeated cough, expectoration, and wheezing for 8 years, chest pain and short of breath for 7 days. The patient denied particular exposure history, tuberculosis infection history or smoking history. He was diagnosed with chronic bronchitis at a local hospital. Symptoms were relieved after treatment of an exacerbation. Therefore, no bronchoscopy had been during the course of his disease. Because of the aggravation of symptoms, he came to our hospital for further diagnosis and treatment 9 months prior to this admission. Chest X-ray revealed right middle lobe atelectasis. Bronchoscopy showed obstruction in the right middle lobe with pigmentation. Acid-fast bacilli were negative on brush biopsy smear [Figure 5]. The initial diagnosis was acute attack of chronic bronchitis.
and bronchial anthracofibrosis in the right middle lobe. Anti-infection treatment, bronchodilation and expectoration were given to relieve the symptoms. The patient was discharged with relief of symptoms. He was re-admitted due to chest pain and progressive short of breath of 1 week duration.

Physical examination showed tachypnea but no cyanosis of lips, dullness to percussion in the right lung, right lower lung breath sound disappeared. There were no dry or wet rales. Chest CT showed right pleural effusion [Figure 6]. He was admitted to rule out malignant pleural effusion. Thoracoscopy revealed a large amount of yellow pleural effusion. Multiple white nodules of various sizes were seen on the parietal pleura after suction [Figure 7]. Biopsy was performed. Acid-fast bacilli were negative. Pleural effusion carinoembryonic antigen was 1.37 ng/mL. Biopsy of pleura showed granulomatous inflammation of right parietal pleura [Figure 8]. He was diagnosed with tuberculous pleuritis. After anti-tuberculosis treatment (isoniazid, rifampicin, pyrazinamide, and ethambutol), the pleural effusion reduced remarkably, and his chest pain and shortness of breath were relieved. The pleural effusion disappeared 2 months after the end of anti-tuberculosis treatment, consistent with the diagnosis of tuberculous pleuritis.

**Discussion**

Bronchial anthracofibrosis is a typical manifestation on bronchoscopy in patients who had pneumoconiosis or were severely exposed to the environment of atmospheric dust in the past. However, Chung et al. reported a group of patients in South Korea who had no history of coal dust exposure or smoking who showed pigmentation deposition in the bronchial mucosa (most of them were multilobular); the corresponding bronchi showed luminal stenosis or even complete obstruction, leading to periodic atelectasis and obstructive pneumonia.[1] Because there were multiple calcified lymph nodes in the mediastinal window, Chung named the condition bronchial anthracofibrosis. The main manifestations of the disease on bronchoscopy are pigmentation in bronchial mucosa and stenosis of the corresponding segments.

It has been reported that the two prominent characteristics of bronchial anthracofibrosis on bronchoscopy are hard texture tissue at the lesion site and tendency to bleed during biopsy.[1,2] In recent years, we treated 18 cases of atelectasis with unknown reasons. Further bronchoscopy showed that there were varying degrees of bronchial anthracofibrosis accompanied by stenosis of the lumen.[3] Most of these patients had no smoking history, no exposure history such as coal dust. In the present report, neither the patient had a tuberculosus exposure history, and the imaging features were atelectasis. On bronchoscopy, bronchi of several lobes showed varying degrees of pigmentation and stenosis. In the first case, there was more bleeding during brush biopsy that was consistent with previous report. On July 2014, Paulin et al. described a 76-year-old female complaining of cough, fatigue, and substantial inspiratory wheeze for several months,[4] chest CT revealed bilateral inferior lobe ground glass infiltrated shadow with a high-density shadow seen in the right main branch. Bronchoscopy revealed stenosis of the right main bronchus with pigmentation and mucocutaneous fragility. The right middle lobe bronchoalveolar lavage fluid was positive for acid-fast bacilli. Symptoms were relieved after treatment with anti-tuberculosis drugs. This case is very similar to the two cases, we reported. Both are bronchial anthracofibrosis combined with tuberculosis. Paulin et al. speculated that inflammation of lymph nodes in the lesion site led to pigmentation and stenosis of adjacent bronchus. Chung et al. reported that 20%–60% of patients with bronchial anthracofibrosis were found positive for *Mycobacterium tuberculosis*. In our report, both patients were confirmed positive for tuberculosis infection by histology and treatment effect, consistent with the findings of the previous report. This suggests that patients with bronchial anthracofibrosis are more susceptible to tuberculosis.[3] Smith et al. reported a 75-year-old patient with endobronchial tuberculosis.[6] Black lymph nodes could be seen protruding in the lumen on rigid bronchoscopy and these were removed. Black tumor body could be seen on bronchoscopy in another patient with endobronchial tuberculosis confirmed by bacteriology. The tumor body disappeared after anti-tuberculosis treatment, leaving pigmentation in the mucosa. These findings suggest that we should be aware of the possibility of active tuberculosis infection when bronchial anthracofibrosis is observed during bronchoscopy.

In case 1, the long history of chronic respiratory disease was initially considered as the result of obstructive pneumonia caused by pigmentation and stenosis. The left main bronchus is long and narrow, and is susceptible to poor drainage and repeated infection of local tissues in the context of tracheal stenosis. This resulted in repeated aggravation of symptoms and refractory infection.

Case 2 was also an elderly patient with history of chronic cough and expectoration. He was treated for chronic bronchitis in the past. Symptoms were relieved when given treatment for infection and expectoration. No further attention was paid for years. Bronchoscopy revealed that the bronchus mucosa of the right middle lung had pigmentation and the right middle lung was obstructed. Acid-fast bacilli were negative on brush biopsy smear. Tuberculosis was never considered.
as a possible diagnosis because the symptoms were relieved after routine treatment. He was hospitalized for the right pleural effusion finally, and was diagnosed with tuberculous pleuritis following thoracoscopy and pathology. The pleural effusion became stable after anti-tuberculosis treatment. We believe that, because of atelectasis of the right middle lobe, the lobe became repeatedly infected, resulting in the decrease of immunity and active tuberculosis infection.

Both cases were misdiagnosed. They were both elderly patients who suffered chronic cough, expectoration, and wheeze over the years. The symptoms were consistent with the symptoms of chronic obstructive pulmonary disease. They were relieved by anti-infectives and symptomatic treatment of every exacerbation. Therefore, pigmentation and stenosis were missed during the course of the disease. In the recent course of the disease, active tuberculosis was also missed. For patients with histories of chronic respiratory diseases whose imaging manifestations are atelectasis or inflammatory changes, bronchoscopy should be performed to identify the airway condition. Meanwhile, bronchoalveolar lavage or brush biopsy should be performed as well. If necessary, tissue biopsy should be performed to screen for the possibility of active tuberculosis infection. Both cases were diagnosed after pathological examination, and symptoms were alleviated by anti-tuberculosis treatment. This is consistent with the clinical diagnosis of tuberculosis.

Combined with the literature and our clinical experience, we summarize the characteristics of bronchial anthracofibrosis as follows: (1) It mainly occurs in elderly population. (2) It has no relationship with pneumoconiosis and smoking history. (3) The clinical characteristics are cough, expectoration and/or dyspnea; some patients may suffer from hemoptysis, chest pain, fever (which may be related to active tuberculosis infection). (4) Chest CT can appear as multiple lesions of bronchus stenosis. There are soft-tissue shadow or enlarged lymph nodes or calcified lymph nodes adjacent to the lesions. Bronchoscopy can show pigmentation in the bronchial mucosa. (5) Some patients have evidence of active tuberculosis, or history of tuberculosis infection. Both the present cases were characterized by these features.

**Conclusion**

It critical to improve the understanding of bronchial anthracofibrosis combined with tuberculosis. Clinicians must perform bronchoscopy when encountering patients who suffer from chronic cough with mediastinal or hilar masses, atelectasis, especially pulmonary consolidation adjacent to narrow or obstructed bronchi. Clinicians should remain vigilant to the possibility of tuberculosis infection when bronchoscopy indicates bronchial anthracofibrosis so as to avoid unnecessary thoracotomy.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**

There are no conflicts of interest.

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