Case Report

Pulmonary Sequestration with Tuberculosis Confined to the Sequestrated Lung

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Introduction

Pulmonary sequestration is a congenital anomaly in which a portion of the pulmonary tissue is detached from the normal lung and is supplied by anomalous systemic arteries.1) The sequestered aberrant lung tissue has no connection with the normal bronchial tree or pulmonary arteries.2) In previous reports, pulmonary sequestration may be combined with pyogenic infection, but few are combined with tuberculosis. We present a rare case of intralobar pulmonary sequestration in a patient infected with tuberculosis without any symptoms including respiratory or systemic. This case report may provide some information about the diagnosis of pulmonary sequestration combined with tuberculosis. Moreover, we also present some discussion of the causes of misdiagnosis and the lessons we can learn from this rare case.

Case Report

A 19-year-old man, presenting with no symptoms, was reported to have consolidation and infiltration in the left lower lobe on a routine chest X-ray (Fig. 1a) during a health examination. He had no history of medical illness. No symptoms attributed to his respiratory system, such as...
cough, hemoptysis or dyspnea, were noted. Constitutional symptoms, including malaise, fever, night sweats, anorexia, and weight loss, were also lacking. Although he presented with no symptoms and had no history of pulmonary infection, he was treated with azlocillin (3.75 ivgtt bid) plus levofloxacin (0.4 ivgtt qd) for one week, in the outpatient department of a local hospital. A few days after the treatment, the patient underwent a chest computed tomography (CT), and the chest CT showed infiltration and consolidation in the left thoracic paravertebral area. (Fig. 1b and 1c). Thus, the patient was admitted to a local hospital, where his antibiotic treatment was changed to piperacillin / tazobactam sodium, (6.75 ivgtt bid) plus azithromycin (0.5 ivgtt qd). His physical examination was unremarkable. Results of laboratory examinations were all negative: skin test with a purified protein derivative-standard (PPD-S) was 0 mm, sputum was negative for acid-fast bacilli, and sputum culture was negative. Blood counts were normal: RBC 4.88×10^{12}/L; PLT 158×10^{9}/L; WBC 4.88×10^{9}/L; HB 148g/L;N% 0.681. Erythrocyte sedimentation rate (ESR) was normal at 2 mm/h. After these 12 days of antibiotic treatment and before admission, he underwent a second the chest CT. The scan showed no improvement in the left lower lobe. Based on the chest CT scan, the local hospital changed the antibiotics again toceforazone / sulbactam sodium 2.0 ivgtt bid for 9 days. At the end of this period of antibiotic therapy, he underwent a bronchoscopy. However, it showed no endobronchial abnormality. Then, the patient underwent a chest CT for a third time. Unfortunately, the image of the left lower lobe was still the same (Fig. 1d and 1e).

Because the image of the left lower lobe was not improved, he was referred to our hospital. During the history taking, the patient revealed that he had worked as a waiter in a karaoke box during the past 6 months. He has been suffered with left chest pain two times. The chest pain relieved spontaneously without any treatment: one time was ten years ago, and the other was 6 months ago. On admission to our division, the physical examination was still unremarkable. Laboratory examination including blood chemistry, blood count, c-reactive protein, ESR, anti nuclear antigen (ANA), elutable nuclear antigen (ENA), Antineutrophil cytoplasmic antibody (ANCA), arterial blood gas analysis, blood tumor markers, liver and renal functions were all within normal range; Lung functions were all unremarkable; Tuberculin skin test (PPD) showed a 4-mm induration at 48–72h; sputum for acid-fast bacilli and sputum culture were all negative. Bronchoscopy was normal; acid-fast stains, bacterial and mycobacterial cultures of the left lower lobe bronchial washings were all negative. A chest contrast-enhanced CT with multiplanar reconstruction was taken. The image showed that an aberrant artery that originated from the left side of the descending aorta, crossed through the left lower lobe into the territory of the consolidation patch, but the venous drainage could not be visualized clearly (Fig. 2a–2c). The patient was diagnosed with pulmonary sequestration, and he underwent a left lower lobectomy by open thoracotomy. The thoracotomy showed an aberrant artery that had originated from the left side of the descending aorta with branches inside the sequestrated segment. The diameter of this aberrant artery was approximately 6 mm. Sequestrated lung tissue had the same visceral pleura as that of the left lower lobe. There was a lump mass in the sequestrated lung; the size of the lump was 5.0×6.0×6.0 cm. In addition, the bronchopulmonary hilar lymph nodes and interlobar lymph nodes were found enlarged during thoracotomy. The surgeons resected the entire left lower lobe, including the aberrant tissue after clipping of the aberrant artery. Microscopy showed a clear boundary between sequestrated and normal lung. Microscopy revealed resected sequestrated lung tissue that was mainly composed of caseous necrosis with Langhans cells (Fig. 3a–3c). Acid-fast stains of the resected lung were negative, but we found one mycobacterium tuberculosis in resected lung by immunofluorescent stain. Based on these findings, we confirmed the diagnosis of pulmonary sequestration combined with tuberculosis. The patient recovered well after the operation, and he was discharged on the 15th day after the operation. The patient was treated with 300 mg isoniazid, 1500 mg ethambutol, and 450 mg rifampicin orally daily. His clinical status was excellent, 4 months after the operation.

Discussion

Pulmonary sequestration is a relatively rare congenital malformation defined by non-functional and dysplastic pulmonary tissue with abnormal or absent communication with the tracheobronchial tree and arterial blood supply from the systemic circulation. As report, the incidence of this anomaly ranging from 0.4 percent to 1.8 percent. Sequestrations of the lung are classically divided into two types: extralobar and intralobar. More detailed classifications include those of Pryce. Imaging techniques, especially helical CT angiography can identify a sequestered or dysplastic lung mass, aberrant arterial supply, and even anomalous venous drainage. Intralobar sequestrations are usually found within the lower
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Fig. 1
(a) Posterior–anterior chest X-ray, showing a mass adjacent to the left cardiac border.
(b), (c) Lung and Mediastinum window of chest CT scan, by local hospital (5.11). Irregularly shaped mass in the left thoracic paravertebral area, and with a large patchy infiltration shadow in the left lower lobe.
(d), (e) Lung and Mediastinum window of chest CT, by local hospital (6.2), infiltration in the left lower lobe.

lobes.\textsuperscript{5} The arterial supply usually originates from the aorta or its branches.\textsuperscript{6} The sequestrated lung is non-functional, dysplastic tissue, and once the sequestrated lung has communicated with normal lung tissue, the patient is apt to have a repeat lung infection. Sequestrated lung also may affect the function of adjacent organs and tissues, by compressing the organs and tissues nearby. So an operation is recommended, once the diagnosis of sequestration
is confirmed. An early operation can avoid a repeat lung infection and decrease the difficulty of the operation, caused by the repeat lung infection.

Sequestrated lung is a frequent site for common pyogenic infection.\(^7,8\) Infected pulmonary sequestration due to mycobacteria tuberculosis has been reported, but only rarely.\(^9,10\) Yatera and Izumi\(^9\) described a 22-year-old female who presented with persistent, non-productive cough and repeated exacerbations of a right, lower-zone infiltration. Right lower lobectomy revealed lower lobe sequestration. Microscopy demonstrated a clear boundary between sequestrated and normal lung, as well as caseating epithelioid granulomas confined to the sequestration. No acid-fast bacilli were observed, but homogenates of the sequestration were positive for Mycobacterium tuberculosis by polymerase chain reaction. Ayesha and Mo-been\(^10\) described a 20-year-old man who presented with cough, hemoptysis and on and off fever for 3 years. He was initially misdiagnosed as tuberculosis and received anti-tuberculous therapy (ATT) for 8 months. CT pulmonary angiography revealed an arterial branch arising from lower thoracic aorta cross the lesion. Left lower lobectomy revealed Left lower lobe intralobar sequestration with adjacent pulmonary inflammation.

In our case, unlike the cases above-mentioned, the patient had no history of medical illness, had no symptoms attributed to his respiratory system, and had no constitutional symptoms, either. Even the Laboratory examination and tuberculin skin test (PPD) were all negative. He was just found persistent left lower zone infiltration and consolidation in chest CT, and was misdiagnosed pneumonia, treated with a series of antibiotics for nearly one month. There were some factors might contribute to the misdiagnosis. First, in the patient’s chest CT scan, the lesion is not a single lump of consolidation, but multiple infiltrated and consolidated area connected with each other, and there was low density area in the center of the consolidation. This kind of image commonly was considered as signs of infection. Second, his chest CT is not a typical image of sequestration. The consolidation and infiltration were not merely confined to the paravertebral area, but distributed, comparatively dispersed. Third, the doctors in the local hospital did not suspect the primary diagnosis when antibiotic therapy was not effective. They did not consider other diagnosis, such as sequestration. They didn’t order contrast-enhanced CT, or angiography, but just change the antibiotics again and again, which resulted in the abuse of antibiotics, lengthening the time of therapy.

Although the diagnosis was confirmed as sequestration associated with tuberculosis, our patient presented with no symptoms. As Savic\(^6\) reported, most of the patients suffered with sequestration (>50%) become symptomatic after the second decade of life, and still 15% of patients’ anomaly caused no symptoms. Perhaps our patient will demonstrate symptoms of tuberculosis with the time passing by. How could the tuberculosis reach the

Fig. 3 (a) Histopathology of the resected lung. Only the sequestrated lung showed caseous necrosis with Langerhans’ cells. (b) The resected lung showed caseous necrosis, including Langerhans’ cells. Hematoxylin and eosin stain. (c) Microscopic section of the lung parenchyma showing accumulation of lymphocytes and granulomatous inflammation.
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sequestration? This is another question we want to discuss. Mycobacteria tuberculosis might reach the sequestrated lung through the bloodstream, through the lymphatic system, or from adjacent normal lung tissue via Kohn's pores. Because the patient’s bronchopulmonary hilar lymph nodes and interlobar lymph nodes were found enlarged during thoracotomy, we presume lymphatic system was the most probably way through which mycobacterium tuberculosis reach the sequestrated lung. In addition, Johnston11) suggested that abundant arterial blood flow from the systemic circulation makes the sequestration more susceptible to tuberculosis, an obligate aerobe. Moreover, the sequestrated aberrant lung tissue has no connection with the normal bronchial tree; therefore, the tuberculosis was just confined to the sequestered lung tissue.

We can learn some lessons from this case, which can help us avoid the misdiagnosis and unnecessary treatment in the future. First, infiltration and consolidation in a chest CT scan are not only attributed to bacterial infection. If a patient just has infiltration and consolidation in chest CT, no any other signs of infection including symptoms, laboratory evidence, but could not recover after standard antibiotic treatment, we should suspect the primary diagnosis, consider other cause to this abnormality, rather than just simply change antibiotics. Next, this case reminds us that if the CT scan shows persistent infiltration or consolidation in the same segment of the lung, especially, the lower lobe near vertebral column, we should suspect the possibility of sequestration, even though the lesion is not a single one. Third and the most important, even if the diagnosis is confirmed as a pulmonary sequestration, we should still consider the complication of tuberculosis, especially in tuberculosis prevalent regions.

**Conclusion**

Pulmonary sequestration is a relatively rare malformation. Imaging techniques: contrast-enhanced CT, especially helical CT angiography, can identify sequestration. If the CT scan shows persistent infiltration or consolidation in a same segment, especially, in the lower lobe near the vertebral column, a followed-up with contrast-enhanced CT, or even CT angiography, is necessary. Sequestration associated with tuberculosis may be symptomless, so when the diagnosis of sequestration is confirmed, we should still suspect the complication of tuberculosis, especially in a tuberculosis prevalent region.

**Disclosure Statement**

No conflicts of interest exist in the submission of this manuscript, and the manuscript is approved by all authors for publication. The hospital committee of Medical Research Ethics approved the report.

**References**

2) Feison B. The many faces of pulmonary sequestration. Semin Roentgenol 1972; 7: 3-16.