Case Report

Pulmonary arterial pseudoaneurysm as an unusual complication of pulmonary actinomycosis: Two case reports

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Abstract

We report 2 cases of pulmonary actinomycosis complicated by a pseudoaneurysm. In Case 1, a 67-year-old man visited a hospital 7 months ago because of hemoptysis. CT revealed a suspected lung abscess in the left lingular segment; however, no diagnosis was confirmed by bronchoscopy. A CT scan taken after heavy hemoptysis showed a pseudoaneurysm within the consolidation of the same segment. On the same day, embolization of the left bronchial and intercostal arteries was performed. Left lingulectomy was performed 5 days later, and pulmonary actinomycosis was diagnosed histologically. Case 2 was a 51-year-old man with a 2-year history of cough and intermittent hemoptysis. CT showed a lesion with a cavity suggesting an abscess 3 months previously, and antibiotic treatment was started. After the appearance of massive hemoptysis, embolization was performed for a pseudoaneurysm seen on bronchial arteriography. Four days later, a left lower lobectomy was performed, and pulmonary actinomycosis was histologically diagnosed. Pseudoaneurysms are commonly associated with tuberculosis; however, only one report of pseudoaneurysms has been associated with pulmonary actinomycosis. Appropriate treatment should be selected according to the type of pseudoaneurysm and the risk of recurrent hemoptysis. Angiography and

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embolization are essential tools in diagnosing and treating pulmonary arterial pseudoaneurysms; however, surgical intervention may also be an option in some cases to ensure a good long-term outcome.

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Introduction

Actinomycetes are gram-positive rods of the genus Actinomyces and are common in the oral cavity and intestinal tract. Actinomycosis is a chronic granulomatous disease caused by the Actinomyces species. The thoracic form of the disease accounts for approximately 15% of actinomycosis cases [1]. Pseudoaneurysms are commonly associated with tuberculosis; however, to the best of our knowledge, there has been only one report of pseudoaneurysms associated with pulmonary actinomycosis [2]. We report 2 cases of pulmonary actinomycosis complicated by a pseudoaneurysm.

Case presentation

Case 1

A 67-year-old man presented to our hospital with a complaint of blood sputum. He was an ex-smoker with a history of pleuritis 14 years prior. His body temperature was 36.7°C, blood pressure was 148/77 mmHg, WBC count was 9.5 (μL × 10⁹), and the level of C-reactive protein was 3.67 (mg/L). Contrast-enhanced computed tomography (CT) showed fluid collection in the lingular segment of the left lung, suggesting a lung abscess (Fig. 1A and B). Bronchoscopy did not confirm the diagnosis; further, antibiotic therapy with ampicillin/sulbactam (ABPC/SBT) was provided.

The blood sputum had reduced 3 months later, and a follow-up contrast-enhanced CT showed that the lesion with suspected lung abscess had subsided. However, the patient presented at the hospital 2 months later with massive hemoptysis. Contrast-enhanced CT demonstrated a 15-mm hyperenhancing nodule with the same attenuation as the aorta within a segmental consolidation with an internal low-density area in the left lingular segment (Fig. 1C and D). This hyperenhancing nodule was considered as an intrapulmonary pseudoaneurysm related to the patient’s hemoptysis. There was also pleural thickening outside the lesion and a small amount of left pleural fluid. On the same day, antibiotic treatment with ABPC/SBT was initiated, and an angiogram was performed to confirm the diagnosis and treatment. Although the left pulmonary and bronchial angiograms were performed, no pseudoaneurysm was observed. A selective angiogram of the left pulmonary artery after embolization of the left bronchial artery and the seventh to ninth intercostal arteries with a gelatin sponge showed a faint saccular dilatation of the left pulmonary artery (Fig. 1E). However, there was residual blood flow from the intercostal arteries to the pseudoaneurysm; additionally, embolization of all of these could have resulted in the excessive use of contrast media. There was also a risk of residual lesions. Therefore, a decision was made to add a surgical procedure. Five days later, a left lingulectomy was performed for the abscess and pseudoaneurysm. Pulmonary actinomycosis was diagnosed as the resected specimens showed numerous microabscesses and Actinomyces colonies in the bronchial lumen (Fig. 1F). A CT scan obtained 7 days after the surgery showed residual pleural effusion between the left upper and lower lobes. However, his general condition was stable, and his inflammatory parameters had improved, and he was discharged 8 days after surgery. Since then, no recurrence of hemoptysis has been observed.

Case 2

A 51-year-old man visited the hospital and presented with a cough. He was a never-smoker and had no particular medical history other than childhood asthma. The WBC count was 7.8 (μL × 10⁹), and the C-reactive protein was 0.75 (mg/L) on this day. He was diagnosed with pneumonia as a chest radiograph showed infiltration in the left lung and was treated with levofloxacin. However, the cough persisted 2 months later. A chest CT scan revealed consolidation in the lower lobe of the left lung, suggesting a lung abscess (Fig. 2A). The patient was treated with antibiotics using ABPC/SBT and levofloxacin and recovered. Two years later, the patient presented to the hospital with a complaint of cough and hemoptysis. Contrast-enhanced CT demonstrated consolidation with a cavity in the left lower lobe of the lung, suggestive of an abscess (Fig. 2B and C), and antibiotic treatment with garenoxacin was administered. Three months later, the patient was admitted to the hospital with fever and approximately 100 mL of hemoptysis. A chest radiograph showed infiltration of the left lower lung, and an angiogram was performed on the same day for diagnostic and therapeutic purposes. A selective angiogram and computed tomography (CT) angiography of the left bronchial artery revealed a 7 mm pseudoaneurysm at the peripheral part of the bronchial artery in the left lower lobe of the lung (Fig. 2D, E). Embolization of the feeding bronchial artery with absorbable gelatin sponges stopped the bleeding. However, residual pseudoaneurysm could cause recurrent massive hemoptysis in the later years. Therefore, resection of the lung lobe containing the pseudoaneurysm was considered as optimal. Four days later, a left lower lobectomy was performed, and a histological diagnosis of pulmonary actinomycosis was made (Fig. 2F). The immediate postoperative period was uneventful, and the patient was discharged on postoperative day 5. Two weeks and 6 months after discharge, the patient was observed to perform well without cough or symptoms of hemoptysis.
Fig. 1 — A 67-year-old man with a pulmonary arterial pseudoaneurysm (PAP) due to pulmonary actinomycosis. (A,B) Contrast-enhanced CT images obtained with lung and mediastinal window settings show a fluid collection in the lingular segment of the left lung, suggesting a lung abscess. (C) Contrast-enhanced CT image obtained with mediastinal window settings almost at the same level as A and B shows a 15-mm hyperenhancing nodule with the same attenuation as the aorta within a segmental consolidation with an internal low-density area in the left lingular segment. (D) Maximum intensity projection (MIP) image of c demonstrates the PAP (arrow). (E) Selective angiogram of the left pulmonary artery after embolization of the left bronchial artery and the seventh to ninth intercostal arteries shows a faint bronchial artery-pulmonary artery fistula with a saccular dilatation (arrow). (F) A Gram stain of the specimen shows numerous Gram-positive filamentous branching bacteria in the bronchial lumen.

Discussion

Pulmonary involvement of actinomycosis is caused by the aspiration of oropharyngeal or gastrointestinal secretions into the airways. The symptoms include blood sputum, hemoptysis, cough, fever, and chest pain, with bloody sputum and hemoptysis being the most common in approximately 30%-60% of the cases [1,3]. The 2 cases reported here were all associated with blood sputum or hemoptysis, and some had chest pain and cough. Chest CT of pulmonary actinomycosis generally shows air space consolidation with a central low-attenuation area [4]. It is also characterized by thickening of the pleura in the adjacent area, suggesting the spread of inflammation to the visceral pleura [4]. As Actinomycetes are anaerobic, their culture isolation rates are low. Moreover, it is difficult to confirm the diagnosis of pulmonary actinomycosis by culture because they are endemic to the oral cavity [3]. Diagnosis by bronchoscopy or needle biopsy is also cumbersome because the biopsy forceps cannot penetrate the thick granulation tissue surrounding the actinomycete mass. Consequently, several cases of actinomycosis have been diagnosed by surgical resection [3]. In both cases reported here, actinomycosis could not be diagnosed until surgical resection. As for the treatment, empirical antibiotics are usually used before the diagnosis of pulmonary actinomycosis is confirmed. Several antibiotics are chosen for the treatment of pulmonary actinomycoses, such as penicillin G, cephalosporins, ampicillin, and amoxicillin [5]. Surgical resection is required in cases that are refractory to long-term antibiotic therapy or in cases with massive hemoptysis. Notably, half of the 94 patients with pulmonary actinomycosis eventually required surgery, many of whom had refractory hemoptysis [5].

Pulmonary arterial pseudoaneurysms (PAPs) are associated with various conditions, including trauma, lung tumors, and chronic lung diseases, such as bronchiectasis. However, PAPs resulting from non-tuberculous infections are rare [6]. In particular, there has been only one report of mycotic pseudoaneurysms associated with actinomycosis [2]. Furthermore, to our knowledge, there have been no multiple case reports, including CT images of PAPs associated with pulmonary actinomycosis.

PAPs are important, as they can lead to life-threatening ruptures. An aneurysm is defined as permanent dilatation of a blood vessel involving all the vessel wall layers. In contrast, a pseudoaneurysm is a hematoma involving the destruction of
Fig. 2 – A 51-year-old man with a pulmonary arterial pseudoaneurysm (PAP) due to pulmonary actinomycosis. (A) CT image obtained with lung window settings shows consolidation in the lower lobe of the left lung, suggesting a lung abscess. (B, C) Contrast-enhanced CT images obtained with lung and mediastinal window settings show consolidation with a cavity in the left lower lobe of the lung suggestive of an abscess. (D, E) Selective angiogram and CT angiogram of the left bronchial artery revealed a 7 mm pseudoaneu ysrm at the peripheral part of the bronchial artery in the left lower lobe of the lung (arrow). (F) Photomicrograph of the histopathologic specimen shows filamentous structures that are positive for Grocott–Gomori methenamine-silver staining.

the entire vessel wall and surrounding tissues and is therefore considered to have a higher risk of rupture.

Shin et al. classified PAPs into 4 groups according to the pattern of blood supply from the pulmonary arteries and bronchial and non-bronchial systemic collateral arteries [6]. Type A PAPs were visualized by non-selective pulmonary angiography. Type B PAPs were visualized by selective pulmonary angiography. Type C PAPs are demonstrated on bronchial and nonbronchial systemic collateral arteriography through bronchopulmonary shunting without visualizing feeding pulmonary arteries on selective pulmonary angiography. Type D PAPs are depicted only on pulmonary CT angiography and not on catheter-directed angiography. Both cases 1 and 2 were considered to be type C.

As the PAPs in cases 1 and 2 are type C, embolization of the bronchial and nonbronchial systemic collateral arteries is said to be effective [6]. In Case 1, embolization of the left bronchial artery with shunting to the left pulmonary artery and the seventh to ninth intercostal arteries was performed. However, the blood flow from the intercostal artery to the pseudoaneurysm remained. Because of the possible complications of additional emboli, a left lingulectomy was performed for the abscess and the pseudoaneurysm 5 days later. Meanwhile, in Case 2, embolization of the feeding bronchial artery of the PAP was performed. Although the bleeding had stopped, the remaining pseudoaneurysm could cause recurrent massive hemoptysis in later years. Therefore, a left lower lobectomy was performed 4 days later.

There have been cases of repeated hemoptysis after embolization and deaths due to repetitive hemoptysis after embolization [7]. Thus, the treatment options for PAPs are still under discussion. The advantage of embolization is that it is less invasive than surgery and can be performed following diagnostic angiography if the source of the bleeding is not apparent. From this point of view, it seems to be a reasonable option to perform angiography first, as in cases 1 and 2, and then to perform surgery if repetitive hemoptysis is expected even after embolization.
Here, we report 2 cases of pulmonary actinomycosis complicated by a pseudoaneurysm. Knowing the type of PAPs can help in choosing the appropriate treatment. Angiography and embolization are the essential tools in diagnosing and treating PAPs; however, surgical intervention may also be an option in some cases to ensure a favorable long-term outcome.

Data availability

Imaging and clinical data may be obtained by contacting the corresponding author.

Patient consent

Written informed consent was obtained from the patients for publication of this case report, including the accompanying images.

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