Case reports

Pulmonary artery pseudoaneurysm caused by Streptococcus constellatus

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Running title: S. constellatus-induced Pulmonary artery pseudoaneurysm

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We report a rare case of mycotic pulmonary artery pseudoaneurysm (PAP) secondary to lung abscess due to *Streptococcus constellatus*. PAP was confirmed by the pathological findings of the pseudoaneurysm, the presence of bacteria, and the microbiological analysis. PAP is uncommon, but it is important to recognize this condition because PAP can lead to fatal hemoptysis.

**Key words**: Pulmonary artery pseudoaneurysm, *Streptococcus constellatus*, hemoptysis
**Introduction**

Life-threatening massive hemoptysis accompanied by infection sometimes occurs in patients with bronchiectasis and pulmonary tuberculosis, but infection-associated hemorrhage from pulmonary artery pseudoaneurysms (PAPs) is rare. Here, we report a case of infectious PAP confirmed by pathological and microbiological examinations in a patient who presented with hemoptysis.

**Case report**

A 63-year-old Japanese man with hypertension had a 10-day history of cough and fever. He was an ex-smoker and had had pneumonia when he was 58 years old. Chest computed tomography (CT) revealed an infiltration in the right upper and middle lobes. He was diagnosed with pneumonia and recovered well after taking oral levofloxacin for 2 weeks. However, a follow-up CT scan performed a month later revealed a persistent mass lesion, and he was scheduled for admission to our hospital for an extensive examination that would include investigations for malignancy. However, before admission, he visited our emergency department because of sudden massive hemoptysis.

His body temperature was 37.3°C, blood pressure was 128/81 mmHg, pulse rate was 126 beats/min, and oxygen saturation was 96% in room air. Rhonchi were heard in the right middle chest during auscultation. Laboratory analysis determined a white blood cell count of
A hemoglobin level of 10.1 g/dL, and a C-reactive protein level of 11.83 mg/dL. To prevent further hemoptysis, intravenous hemostatic agents were immediately administered and complete bed rest was prescribed.

An enhanced CT scan showed an enlarged mass lesion enclosing an aneurysm (Fig. 1), but embolization was not performed because the aneurysm was considered inaccessible. Although no bacterial infection was detected by microbiological examinations, including sputum and blood cultures, intravenous tazobactam/piperacillin (13.5 g/day) was administered for the possibility of lung abscess. Improvement of laboratory inflammatory parameters was seen, but surgical management was required because of recurrent hemoptysis. The patient underwent right middle lobectomy and right upper lobe segmentectomy.

The major pathological findings were pneumonia with organization and multiple small abscess formations (Fig. 2). Detailed examination uncovered destruction of vessel walls at the site of the severe inflammation and around the abscess, and fibrin deposition from fresh blood around a hematoma. These findings indicated pseudoaneurysm associated with inflammation.

There was no evidence of malignancy.

The surgical tissue in anaerobic transport medium was inoculated onto HK semisolid medium (Kyokuto Pharmaceutical Industrial Co., Tokyo, Japan) and cultured under anaerobic condition. On postoperative day 8, the culture became positive. The pathogen was identified as *Streptococcus constellatus* by a biochemical identification method using the Phoenix100
system (Becton Dickinson, Franklin Lakes, NJ) and exhibited good susceptibility to penicillins.

After 20 days’ treatment with tazobactam/piperacillin, the patient was discharged and made an uneventful recovery.

Discussion

Pulmonary artery aneurysms (PAAs) and PAPs are uncommon but important, as diagnostic and therapeutic procedures can affect the associated morbidity. An aneurysm is defined as a permanent dilatation of blood vessels that involves all layers of the vessel wall. In contrast, a pseudoaneurysm is a hematoma involving destruction of the entire vessel wall and surrounding tissues, and is therefore associated with a higher risk of rupture.

PAAs are commonly associated with congenital anomalies, and are usually observed in a major pulmonary artery such as the pulmonary artery trunk and major branches. Acquired PAAs are associated with pulmonary hypertension and infections, including tuberculosis, syphilis, and endocarditis. However, the incidence of infection-associated PAAs has decreased due to progress in the treatment of infectious diseases. In contrast to PAAs, PAPs are typically caused by traumatic events, including catheterization procedures, and in some cases are associated with chronic lung diseases, such as cavitary pulmonary disease and bronchiectasis (1). PAPs associated with tuberculosis are known as Rasmussen aneurysms (1), but PAPs caused by non-tuberculous infections are very rare.
Lung abscess is a less common cause of massive hemoptysis than active pulmonary tuberculosis and bronchiectasis. In cases of hemoptysis associated with lung abscess, radiologically visualized PAPs can be observed within the abscess (2-3). Non-tuberculosis microorganisms that have been implicated in PAAs include *Staphylococcus aureus* (4), viridans Streptococci (4-5), *Enterococcus* species (4), and *Candida albicans* (6). However, these previous reports included microorganisms that were not detected directly at the site of the infection. Therefore, our case differs in that it is a confirmed case of infectious PAP based on the pathological findings of the pseudoaneurysm, the presence of bacteria, and the microbiological analysis.

*S. constellatus* is a component of the normal flora of the human oral cavity, and has been recognized as a cause of pulmonary infections (7). Identification of *S. constellatus* is sometimes difficult for the clinical laboratory because of requirement of anaerobic condition and long culture. The anginosus group, formerly the *S. milleri* group, includes *S. anginosus*, *S. intermedius*, and *S. constellatus*, and these organisms are often isolated from abscesses. However, the frequency with which each species of the anginosus group is associated with abscess varies. Claridge reported that approximately 80% of *S. intermedius* and *S. constellatus* isolates, but only 19% of *S. anginosus* isolates, were associated with abscess (8). Thus, continuous invasion of *S. constellatus* could lead to the destruction of the pulmonary arterial wall.
The case we report was initially suspected to be lung cancer because of the persistent mass opacity after antibiotic therapy. Therefore, hospital admission was scheduled so that pathological diagnosis could be performed by endoscopic biopsy. As we detected an aneurysm on contrast CT after his arrival at the emergency department with massive hemoptysis, we did not need to perform that invasive procedure. This is indeed fortunate, as a fatal case of pulmonary artery aneurysm hemorrhage after endobronchial lung biopsy has been reported (9).

PAPs are uncommon but important, as PAP can lead to fatal hemoptysis. In cases of suspected lung cancer accompanied by prolonged inflammatory findings, non-invasive examinations, such as contrast CT scanning, which can provide information on the coexistence of pseudoaneurysms, should be performed before biopsy.

Conflict of Interest

None.
References


The mass lesion in the right middle lobe contains a low-density area with a pulmonary artery aneurysm (white arrow).

In the Elastica van Gieson–stained sections (A, original magnification ×100), multiple small abscess formations with complete destruction of the vessel wall layers (arrowheads) were observed. In the Gram-stained sections (B, original magnification ×1000), Gram-positive cocci (white arrows) were observed in the abscess lesions around the pseudoaneurysmal changes.
Figure 1