Case Report

A case of pulmonary aspergilloma and actinomycosis

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Pulmonary aspergilloma and pulmonary actinomycosis are rare pulmonary infectious diseases. Clinical manifestations of pulmonary aspergilloma and pulmonary actinomycosis include chronic cough, fever, chest pain, haemoptysis and other pathologies, but some patients may be asymptomatic. We report a case of a healthy 33-year-old woman without any underlying diseases, who was admitted to Zhongxing Branch of Taipei City Hospital, Taiwan, for intermittent haemoptysis and right upper chest pain, which had persisted for several months. A chest radiograph revealed a focal consolidation in the right upper lobe (RUL) of the lung, which grew in size over time. A sputum study and bronchoscopy revealed no positive findings, although malignancy could not be ruled out. Thus, the patient received a wedge resection of the RUL lesion. Subsequent, pathological examination demonstrated the presence of pulmonary aspergilloma and pulmonary actinomycosis. The patient's symptoms resolved after resection of the RUL lesion.

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Introduction

Aspergilloma is a disease caused by members of the genus *Aspergillus*. This condition presents with a spectrum of illnesses, ranging from allergic reactions to colonization of pre-existing pulmonary cavities to invasion and destruction of lung tissue with pyaemic spread to the brain, skin and other organs (Kradin & Mark, 2008). Aspergilloma often occurs in patients with cavitory pulmonary disease. Clinical presentations of pulmonary aspergilloma include chronic cough, fever, chest pain, haemoptysis and some symptoms associated with chronic wasting diseases. However, some individuals may have no symptoms (Fujiuchi *et al.*, 2004). Chest imaging may provide some unique patterns to allow the diagnosis of this disease, but it often needs an analysis of pathological tissue for a definitive diagnosis. Current treatments include antifungal regimens and surgical resection (Zmeili & Soubani, 2007).

Pulmonary actinomycosis is caused by Gram-positive filamentous anaerobic-to-microaerophilic bacteria, which do not form spores and are not acid-fast stained (Brook, 2008). The *Actinomycetaceae* family are normally harmless commensals in the oropharynx but can cause subacute and/or chronic pulmonary infections. Pulmonary actinomycosis

often occurs following aspiration of oropharyngeal material. Most patients are between 30 and 60 years old, and male patients outnumber female patients by four to one (Brook, 2008). Chronic lung diseases, including bronchiectasis and other chronic obstructive pulmonary diseases, are common underlying conditions associated with pulmonary actinomycosis (Tsubochi *et al.*, 2007). Clinical symptoms such as fatigue, weight loss and low-grade fevers may be present for weeks to months prior to a correct diagnosis, and often mimic the presentation of other pulmonary malignancies or infections. Eventually, most patients gradually develop a productive cough and pleuritic chest pain, but haemoptysis is relatively uncommon (Mabeza & Macfarlane, 2003). Chest imaging most commonly demonstrates fibrotic infiltrates confined to a single lobe with one or more small cavitory lesions. In advanced cases, the findings are more distinctive and are characterized by penetration through the chest wall, destruction of adjacent bone tissue, or direct extension through an interlobar fissure (El Ghannam *et al.*, 2010). The drug of choice for the treatment of actinomycosis is penicillin G and a variety of surgical treatments (Brook, 2008).

Case report

A 33-year-old non-smoking married woman with no previous significant medical history was admitted to

Abbreviations: CT, computed tomography; RUL, right upper lobe.

Zhongxing Branch of Taipei City Hospital, Taiwan, after suffering from intermittent haemoptysis and right upper chest pain for several months. The patient's history indicated no fever, chills, cough, dyspnoea, nausea, vomiting, diarrhoea, abdominal pain or special travel history. Her family history was also unremarkable.

At the hospital's outpatient department, a chest radiograph was taken and revealed a nodular lesion of the right upper lobe (RUL) of the lung (Fig. 1a). The patient was calm and clear minded, and had the following vital measurements: a body temperature of 36 °C; a pulse of 80 beats min⁻¹; a respiratory rate of 16 breaths min⁻¹; and blood pressure of 120/60 mmHg. Auscultation of the chest indicated that the bilateral lung fields were clear and physical examination of the patient was otherwise unremarkable. Laboratory studies revealed the following: a white blood cell count of 7410 cells mm⁻³ (78.5 % neutrophils, 15.8 % lymphocytes); a haematocrit level of 40.5 %; haemoglobin level of 13.8 g dl⁻¹; and a platelet count of 263 000 platelets mm⁻³. The results of biochemistry tests such as those of liver function, electrolytes and C reactive protein were normal. A chest computed tomography (CT) scan was performed and revealed an ill-defined nodular lesion with a pleural tail in the RUL (Fig. 1b). Subsequent studies, including those of sputum acid-fast stain, sputum cytology and blood cryptococcal antigen, were all normal. Sputum samples were cultured in Löwenstein-Jensen medium and 7H10 agar, and were negative for *Mycobacterium tuberculosis*. Tumour markers including carcinoembryonic antigen and squamous cell carcinoma antigen were normal, while bronchoscopy revealed no endobronchial lesion or haemorrhage. We suggested that the patient undergo a follow-up close chest radiograph.

Approximately 1 month later, the patient was examined and a chest radiograph was taken. The lesion appeared denser, but then the patient was lost for follow up examinations for another 3 months. After this time, the patient's symptoms still persisted and she was admitted to Zhongxing Branch of Taipei City Hospital again. We repeated the chest CT examination and confirmed that the RUL nodular lesion had grown in size. Furthermore, one new nodular lesion was found in the left upper lung field (data not shown). Because tumour malignancy could not be ruled out and the patient's symptoms had persisted, we opted for surgical resection. The patient received a right upper lung wedge resection and was treated with amoxicillin-clavulanate (1 g orally every 12 h) for 5 days for a chest-tube wound infection. The operated lung tissue culture yielded *Haemophilus influenzae* and *Aspergillus flavus*. The pathology examination of the lung biopsy after haematoxylin and eosin staining revealed characteristic findings of pulmonary aspergillosis and pulmonary actinomycosis, which included a cavity filled fungal ball that was composed of numerous acute angled branching septated hyphae (Fig. 2a) and actinomycosis characterized by multiple sulfur granules (Fig. 2b). The adjacent lung showed fibrosis,

Fig. 1. (a) Initial chest X-ray, revealing a RUL of the lung field consolidation (indicated by an arrow). (b) Lung window of chest CT scan, revealing an ill-defined nodular lesion with a pleural tail over the RUL (indicated by an arrow).

granulation tissue formation, as well as acute and chronic inflammatory cell infiltration. The patient's symptoms improved after resection of the RUL lesion and the left upper lobe lesion was not treated. The patient was discharged on the 14th hospital day (7 days after surgery), but she was then lost for further follow up examinations.

Discussion

We report an extremely rare case of simultaneous pulmonary aspergilloma and pulmonary actinomycosis. Only two similar cases have been reported in the literature: a 79-year-old patient who presented with haemoptysis and severe anaemia (Hartmann & Keller, 2000), and a 56-year-old patient who presented with haemoptysis and coughing (Herrak *et al.*, 2007). In our study, this combined infection presented as a slowly progressing lung lesion; hence, we had to take several differential diagnoses into consideration, including a lung tumour, as well as inflammatory and non-inflammatory processes (Barikbin *et al.*, 2007). Nevertheless, for undetermined lung lesions, an examination of tissue pathology is usually required for a definitive result. Although, chronicling course pathology can be used as a last resort in the absence of other available diagnostic methods. This case belongs to the aspergilloma type of *Aspergillus*-related diseases (Buckingham & Hansell, 2003),

which is usually asymptomatic and 40–60 % patients have haemoptysis, while cough and fever are less common. Pulmonary actinomycosis often results in subacute or chronic infection, and the clinical presentation includes cough, haemoptysis, fever, anorexia and chest pain (Mabeza & Macfarlane, 2003). The patient in this case study had haemoptysis and chest pain, and although these symptoms were consistent with a wide range of pulmonary pathologies, they were also consistent with the co-infection of aspergilloma and actinomycosis (Kitasato *et al.*, 2010).

The treatment of pulmonary aspergillosis is typically with anti-fungal agents such as voriconazole (Jain & Denning, 2006) or micafungin (Izumikawa *et al.*, 2007), and while the management of aspergilloma is often through surgical resection, it can also be treated medically. By comparison, the treatment for actinomycosis is fairly effective with common antibiotics (Tsubochi *et al.*, 2007). Hence, surgical intervention for the later of these infections is usually reserved for complex or medically refractive cases. However, in this case study, the surgically resected material was critical to establishing the definitive diagnosis.

This patient had no previous significant medical history and was a homemaker living in the mountain area of Taipei County, Taiwan. As a result, this study was limited by the fact that the patient did not receive long-term outpatient follow up. Importantly, we cannot conclude that the resection of the lesion was curative, nor can we assess the patient's long-term prognosis or response to long-term pharmacological regimens of postoperative antibiotic and antifungal therapies to prevent subsequent infections and unnecessary morbidity.

Conclusion

Pulmonary aspergilloma and pulmonary actinomycosis are relatively rare pulmonary infectious diseases, and their occurrence simultaneously is even rarer. Chest imaging may provide some clues as to how to diagnose these diseases, but a definitive diagnosis often relies on a pathological examination of the infected tissue.

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Fig. 2. (a) Photomicrograph of resected lung tissue showing a picture of aspergilloma with many fungal balls composed of massive acute angle branching septated hyphae. (b) Photomicrograph of resected lung tissue showing actinomycoma with many sulfur granules (indicated by an arrow).

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