Opportunistic actinomycosis in pulmonary alveolar proteinosis

Sze Shyang Kho 1,2, Suhashini Ganapaty,3 Noorjehan Omar 1,3, Shang Ze Tan,4 Mona Zaria Nasarudin,1 Jamalul Azizi Abdul Rahaman1

CASE REPORT

A 58-year-old woman with major depressive disorder presented with an insidious onset of fever and haemoptysis for a 1-month duration. She was also troubled with chronic cough over the past 2 years. Her weight was stable and there was no family history of malignancy. She was an ex-smoker of 15 pack years. She worked in the textile industry involved in stonewashing of denim fabric in which she had unprotected exposure to silica dust from pumice stones for the past 30 years. On examination, she was febrile with temperature of 37.8°C and heart rate of 109 beats/min. Blood pressure was 154/86 mm Hg with a saturation of 96% on room air. Chest auscultation revealed reduced air entry over the right upper zone anteriorly. There was no cervical lymphadenopathy, and her fingers were not clubbed.

A plain chest radiograph revealed right upper lobe consolidation with patchy ground glass opacity over the left hemithorax (figure 1). Blood investigations were normal, and the initial workup was not suggestive of any acquired immunodeficiency state. CT thorax revealed necrotic consolidative changes in the anterior segment of right upper lobe with multiple air pockets. Interestingly, patchy ground glass opacities were also seen interspersed with smooth thickening of interlobular and intralobular septa in all lobes, giving rise to a crazy-paving pattern which was distributed centrally (figure 2A).

As microbiological analyses were negative and presentation with haemoptysis, rigid bronchoscope was performed in anticipation of uncontrolled bleeding and this revealed a yellowish necrotic endobronchial lesion within the anterior segment of the right upper lobe with overlying inflamed mucosa (figure 2B). Endobronchial cryobiopsy showed granulation tissue formation with filamentous bacterial colonies, favouring Actinomycete species. The bacteria are highlighted by Periodic acid-Schiff (PAS) and Grocott-Gömöri’s methenamine silver stain. Bronchial washing was negative for bacterial, fungal and mycobacterium. Cytological examination showed numerous neutrophils consistent with acute inflammatory process. The patient was treated with intravenous benzylpenicillin for a clinical diagnosis.
of pulmonary actinomycosis with possible underlying pulmonary alveolar proteinosis (PAP) from prolonged unprotected silica exposure.

However, despite a month of intravenous therapy, her haemoptysis persisted, and the right upper lobe consolidation only improved minimally with an unchanged crazy-paving pattern, thus leading to the decision to perform right upper lobectomy. Histopathological examination of the resected specimen shows Actinomycotic granules within the bronchus and bronchioles surrounded by acute inflammatory exudates (figure 2C). The background alveolar spaces are filled with coarse, granular eosinophilic PAS-positive material containing acicular spaces and eosinophilic globules, consistent with clinical suspicion of PAP (figure 2D). A final diagnosis of opportunistic pulmonary actinomycosis in secondary PAP was clinched. The patient remained well post surgery and was discharged with oral amoxicillin for a total duration of 1-year antimicrobial therapy. She remains asymptomatic and is currently on surveillance for her PAP.

**DISCUSSION**

Actinomycosis is a chronic supplicative granulomatous infection caused by *Actinomyces israelii*, a gram positive, facultative anaerobic actinobacteria. Thoracic involvement is uncommon and maybe more prevalent in patients with underlying lung disease with impaired local host defence, such as PAP.1

PAP is a rare pulmonary disease caused by defective macrophage surfactant clearance resulting in accumulation of lipoproteinaceous material in the alveolar spaces giving rise to crazy-paving pattern radiologically. Although a crazy-paving pattern is a common radiological feature, it is not pathognomonic of PAP as this pattern can also be found in infection, lipid pneumonia and bronchoalveolar adenocarcinoma.2 Hence, as demonstrated in our case, pathological evidence of PAS-positive acellular eosinophilic material on surgical specimen or bronchoalveolar lavage remains essential in the diagnosis of PAP. Silica dust inhalation is a known cause of secondary PAP.3 Hence, vigilant history on possible occupational or recreational silica exposure is essential as demonstrated in this case. Although we did not assess anti-GM-CSF (granulocyte-macrophage colony-stimulating factor) antibody level as this test was not available in our region, her demographic does not fit into the typical presentation of autoimmune PAP.4

Patients with PAP commonly presented with dyspnoea and chronic cough although one-third of patients can be asymptomatic.4 Atypical symptoms such as chest pain and haemoptysis in our patient are usually suggestive of opportunistic infection which is common in PAP and remain one of the main causes of mortality. Common pathogens that have been reported include nocardia, fungi and mycobacterium.1 However, opportunistic infection due to actinomycosis in PAP has not hitherto been described. Treatment of actinomycosis generally include initial 4–6 weeks of intravenous penicillin therapy followed by 6–12 months of oral amoxicillin.1 Clinical and radiological monitoring remain essential throughout the treatment period especially in pulmonary actinomycosis which may frequently require longer treatment duration.1 In conclusion, this case highlights the potential association of actinomycosis with PAP as well as the importance of early recognition of potential opportunistic infection to ensure a favourable long-term outcome.

**REFERENCES**