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Case report

A case of pulmonary actinomycosis presented with endobronchial involvement

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ABSTRACT

Pulmonary actinomycosis is a rare infectious disease, which is characterized by a wide range of symptoms and no specific imaging findings and may be confused with neoplasia, tuberculosis or pneumonia. Endobronchial involvement of actinomycosis may be caused by aspiration of foreign bodies or broncholithiasis and may bronchoscopically masquerade as malignancy. A case of 68-year-old man is reported, who presented with productive cough and fever and had no response to antibiotic therapy with moxifloxacin. Patchy air-space consolidation on left lower lobe was demonstrated on CT and flexible bronchoscopy revealed an endobronchial white necrotized mass, causing partial occlusion of bronchus and masquerading as lung cancer. Endobronchial actinomycosis was confirmed by biopsies of lesion, which revealed radiating filamentous colonies of Actinomyces and no evidence of malignancy. The patient was successfully treated with intravenous penicillin G for two weeks, followed by doxycycline per oral for six months, achieving full resolution of lesion on follow-up CT and bronchoscopy and no recurrence of symptoms.

1. Introduction

Actinomycosis is an uncommon infectious bacterial disease caused by Actinomyces species. Bacteria of actinomyces species belong to normal human flora, having been detected in oral cavity, gastrointestinal tract and female genital tract. The development of actinomycosis is caused by disruption of mucosal barrier, which allows spreading of bacteria, causing endogenous infection and affecting numerous organs. It is estimated that cervicofacial and abdominopelvic actinomycosis are the most common locations of actinomycosis, while the pulmonary form of actinomycosis is the third most frequent type of actinomycosis and constitutes approximately 15% of the total burden of the disease. Pulmonary form of actinomycosis may arise from aspiration of oropharyngeal or gastrointestinal secretions [1]. The clinical presentation of pulmonary actinomycosis is varied, including cough, fever, hemoptysis and weight loss. In addition, imaging findings of pulmonary actinomycosis present low specificity, making the diagnosis difficult and leading to confusion with other diseases, such as lung cancer, tuberculosis, pneumonia and aspergillosis [2]. Consequently, actinomycosis is often misdiagnosed or diagnosis is delayed. In this case, pulmonary actinomycosis was presented with endobrochial involvement and endoscopically masqueraded as lung cancer. Finally, diagnosis was confirmed with bronchoscopic biopsies.

2. Case presentation

A 68-year-old man was admitted to the emergency department due to productive cough with purulent sputum that had worsened over the previous 2 weeks, with fever up to 38.3 °C during the latter 6 days and generalized body weakness. Owing to symptoms, the patient had been examined by general practitioner and received clarithromycin 500 mg twice per day the last three days without remission of symptoms. At admission the patient's oxygen saturation was 95%. His past medical history consists of diabetes mellitus type II and arterial hypertension and the patient took medications for both diseases. He was a smoker with a history of 35 pack-years. The chest X-ray revealed a small opacity of left lower lobe and laboratory findings showed leucocytosis (WBC: 12.700/mL) and an elevated CRP of 7mg/dL. Due to laboratory and imaging findings and clinical picture, pneumonia of community was suspected and treatment with moxifloxacin 400 mg once a day started. Despite the antibiotic therapy, no clinical and laboratory improvement was observed on the first three days, while the results of blood cultures were

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negative for bacteria, parasites and fungi. Therefore, CT thorax was performed, revealing patchy air space consolidation in the posterior segment of the left lower lobe, suggesting obstructive pneumonia (Fig. 1) and the patient was underwent flexible bronchoscopy, so as to investigate the underlying cause. Bronchoscopy revealed an endobronchial white necrotized lesion within the left lower lobe (B6), causing an obstruction of 60% of the left lower bronchus and mimicking malignancy (Fig. 2). Bronchial secretions were collected by washing. Aerobic cultures of samples were negative. However, gram stain of bronchial secretions demonstrated gram positive bacteria, while Ziehl-Neelsen stain was negative. In addition, biopsies of the mass obtained with forceps and bronchoscopic biopsy specimens showed infiltration from neutrophils, plasmocytes and eosinophils and no evidence of malignancy. Furthermore, staining with Periodic acid-Schiff (PAS) revealed radiating filamentous colonies of actinomyces. After the results of the biopsies, the treatment switched to 24 million units G penicillin daily and remission of symptoms and laboratory improvement were observed during the first week after switching. In the second week of hospitalization, the patient refused to continue the intravenous treatment and he wanted to be discharged due to personal reasons. Therefore, treatment was switched to doxycycline 100 mg twice daily. Two months after starting treatment, a follow-up CT thorax and bronchoscopy were performed. Imaging and bronchoscopic findings demonstrated the complete resolution of lesion (Figs. 3 and 4). Concomitantly, there was almost full improvement in symptoms. He continued to receive doxycycline 200 mg per day for a total of four months without recurrence of the symptoms.

3. Discussion

Chronic lung diseases, such as bronchiectasis and chronic obstructive lung disease, alcoholism, diabetes, hematologic diseases, such as leukemia, human immunodeficiency virus infection and the use of immunosuppressive agents, such as corticosteroids and anti-TNF agents have been associated with the development of pulmonary actinomycosis. Furthermore, in cases of endobrochial development of actinomycosis, broncholithiasis and endobronchial foreign bodies seem to be predisposing factors [3]. In the above case, the patient was immunocompetent and no history of lung disease or aspiration foreign body preexisted, however, the patient did suffer from diabetes. Diagnosis of pulmonary actinomycosis is often a diagnostic challenge due to no specific features of actinomycosis in imaging procedures and a significant number of features on CT in pulmonary actinomycosis have been described, including air-space consolidation, lobar consolidation, ground glass opacification, pleural effusions, pleural thickening, hilar lymphadenopathy and necrotic mass [4]. Bronchoscopic findings of endobronchial actinomycosis are also no specific. In past reports, endobronchial

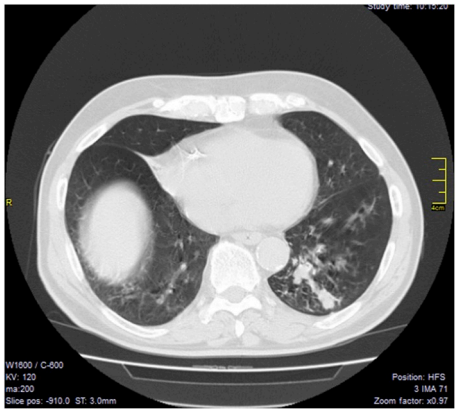

Fig. 1. Computer tomography of chest shows patchy air space consolidation in the posterior segment of the left lower lobe.

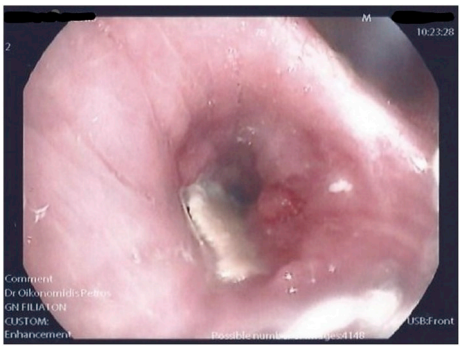

Fig. 2. Bronchoscopy demonstrates an endobronchial white necrotized lesion within the left lower lobe (B6), causing partial occlusion of bronchi.

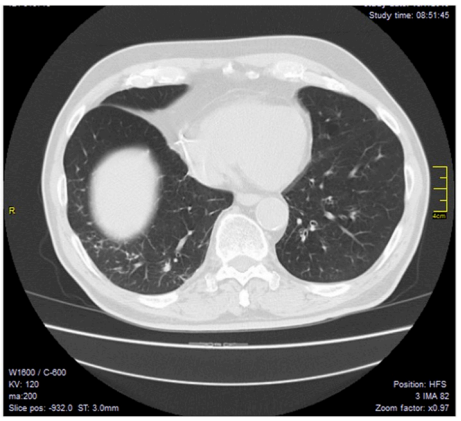

Fig. 3. Computer tomography of chest shows full resolution of consolidation in left lower lope.

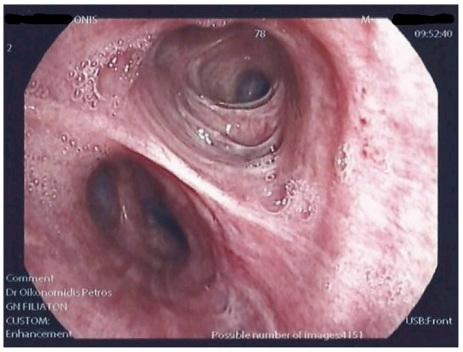

Fig. 4. Bronchoscopy shows the resolution of endobronchial mass and a normal bronchial mucosa.

actinomycosis have been reported to manifest bronchoscopically as granular thickening and partial occlusion of bronchi, submucosal or exophytic mass with or without necrotic material, causing partial or full obstruction of bronchus and mimicking as lung cancer or tuberculosis [5–7]. Therefore, endobronchial involvement of actinomycosis should be included in differential diagnosis of an endobronchial lesion. Hence, microbiological or pathological investigation of tissue specimens, obtained with bronchoscopy, surgical resection or percutaneous biopsy guided by CT is required for diagnosis of pulmonary actinomycosis. Microbiological diagnosis of actinomyces species requires immediate specimen transport and prolonged bacterial culture (5–20 days) under anaerobic conditions. In most cases of actinomycosis, bacterial confirmation is not achieved as a result of the overgrowth of associated

bacteria, inadequate short-term incubation or previous antibiotic therapy suppresses the growth [8]. On the other hand, histopathologic evaluation of tissue specimen plays in most times a critical role on diagnosis of Actinomycosis. Typical histopathologic features include polymorphonucleates, plasma cells and fibroblasts. Furthermore, staining by hematoxylin and eosin and period acid-Schiff may demonstrate the presence of sulphur granules and radiating filamentous colonies [8]. In addition, molecular methods, including real time PCR, DNA-DNA hybridization, 16S rRNA gene sequencing and fluorescence in situ hybridization (FISH), have recently been developed for the correct identification of actinomyces species [9]. The prognosis of the disease is usually favorable and patients with actinomycosis should be treated with antibiotic therapy for a prolonged period of time. High doses of intravenous penicillin G (18–24 million units daily) over two or six weeks, followed by oral penicillin V for 6–12 months is the recommended treatment. In addition, treatment with β-lactams, such as amoxicillin and ceftriaxone, doxycycline and clindamycin have often been successfully used [10]. On the other hand, metronidazole, aminoglycosides, co-trimoxazole, aztreonam and fluoroquinolones have poor or no activity against actinomyces species [9]. Furthermore, despite the recommendations for prolonged treatment, many patients with actinomycosis have been cured with a <6-month antibiotic therapy [11]. Noteworthy, in cases of endobronchial involvement of actinomycosis, a follow-up bronchoscopy after starting antibiotic treatment and imaging improvement is essential in order to exclude a foreign body, given the fact that many cases of foreign body-induced endobronchial actinomycosis have been reported and in 45% of cases the foreign body was detected after starting antibiotic therapy. In event of the existence of a foreign body, extraction should be performed [12]. Also, management of the disease should include treatment of existing predisposing factors, such as improvement of dental status or management of aspiration syndromes [13]. Untreated pulmonary actinomycosis may cause serious complications, such as empyema necessitans, rib destruction and mediastinal invasion, which may progress into the heart, causing pericarditis [10].

4. Conclusion

Pulmonary actinomycosis is an uncommon infectious disease with no specific symptoms and endobronchial involvement of pulmonary actinomycosis may masquerade as lung cancer or tuberculosis. Consequently, pulmonary actinomycosis may hold accountable in the differential diagnosis of patients suspected of having lung cancer, tuberculosis or unresolving pneumonia. This issue raises the necessity for increased awareness in the management of endobronchial lesion and in cases of suspected endobronchial actinomycosis; histopathologic examination and anaerobic cultures under appropriate conditions are fully warranted.

Conflicts of interest

The authors report no conflict of interest and no financial and non financial interest in the subject matter or materials discussed in this manuscript. The authors alone are responsible for the content and writing of this article.

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Ethical approval

Informed consent was obtained from the patient and it is available upon request.

Author contribution

FF, DD: writing and data selection, **PK, IP, GG:** writing, **PO:** design of study and supervision. All authors read and approved the final manuscript.

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