

Recurrent endobronchial actinomycosis following an interventional procedure

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ABSTRACT

Actinomycosis is an indolent, slowly progressive infection caused by anaerobic or microaerophilic bacteria, primarily from the genus *Actinomyces*. Thoracic involvement is observed in approximately 15% of cases of infection with actinomycosis. Here, we present a case of a 61-year-old male who presented with recurrent endobronchial actinomycosis. The case is being presented because of its rarity on three counts – endobronchial involvement, which is uncommon, recurrence in different sites in the bronchial tree, which is even rarer and development of the disease following an endobronchial procedure.

KEY WORDS: Endobronchial, pulmonary actinomycosis, recurrence

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INTRODUCTION

Actinomycosis is a chronic suppurative pulmonary infection caused by genus *Actinomyces*. They are the normal inhabitants of the human oropharynx and frequently found at gingival margins of persons with poor oral hygiene.^[1] Cervicofacial infection is the most common manifestation, and thoracic actinomycosis accounts for approximately 20% of cases. Abdominal and pelvic manifestations are less commonly seen. Pulmonary actinomycosis is acquired mainly through aspiration of the organism from the oropharynx. Infection via inhalation, hematogenous dissemination and direct extension from adjacent tissues can also occur.^[2] The classical presentation of this disease is as thoracic mass lesions which spread without respecting the anatomical barriers, rib destruction, and pleural empyema. Lesions which mimic malignancy or tuberculosis also have been reported.^[3] Correct diagnosis is frequently missed until invasive procedures are performed to get tissue samples. We hereby present a case of a 61-year-old gentleman with a history of carcinoid tumor

of the bronchus in the past, who presented with recurrent endobronchial actinomycosis which was found to affect different segments of the lung.

CASE REPORT

A 61-year-old gentleman, a private banker by profession, who had never smoked but a regular pan chewer, presented to our hospital in 2009 with symptoms of high-grade fever and cough with mucoid sputum of 2 weeks duration. He had received treatment with a cephalosporin from a local health facility, but since fever was persisting, came to us for detailed assessment. His vital signs were normal and he was afebrile at presentation.

One year back, he was diagnosed with a slowly resolving pneumonia of the right upper lobe and found to have an endobronchial carcinoid (biopsy proved). He had undergone Argon Plasma Coagulation for removal of the same. He was asymptomatic after that and 6 months

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later had undergone a surveillance bronchoscopy to rule out recurrence, which did not reveal any intrabronchial lesion. He was not an alcoholic and did not have any other comorbid illness including diabetes mellitus, hypertension, or chronic liver disease. He was also not on any regular medications.

Clinical examination of the respiratory system revealed the presence of a few wheezes bilaterally as well as fine crepitations in the left mammary area. He had poor oral hygiene, with dental caries and extensive tobacco stains were found in the teeth. His total white cell count was 11,000/cumm with 69% polymorphs, 24% lymphocytes, and 5% eosinophils. The erythrocyte sedimentation rate was 48 mm/h and his random blood sugar value was 108 mg/dL. Other investigations including renal and hepatic functions were normal. Chest X-ray posteroanterior view revealed the presence of a patchy segmental consolidation abutting the left cardiac border [Figure 1]. He was started on intravenous cefoperazone-sulbactam and azithromycin, but even after 3 days, he remained to be febrile. Sputum culture and blood culture did not grow any organisms and Ziehl-Neelsen staining for acid fast bacilli was also negative. Hence, a bronchoscopy was done which revealed the presence of two yellowish mass lesions with granularity on the surface, one in the lingular bronchus of the left upper lobe [Figure 2] and the other in the apical segmental bronchus of the right upper lobe. Punch biopsies were taken from the lesions. Histopathological examination revealed the presence of bacterial filaments positive for Grocott's Methenamine Silver staining surrounded by lymphocytes and neutrophils [Figure 3]. The bronchoalveolar lavage fluid also showed the presence of fungal filaments with Giemsa stain [Figure 4]. This was consistent with a diagnosis of pulmonary actinomycosis. He was advised to take intravenous penicillin for 2 weeks, but since he refused to take it, in consultation with the infectious diseases specialist, he was started on oral amoxicillin 500 mg 4 times daily. He became afebrile within a week and was continued on the same regimen for 6 months under regular supervision. He was also advised to have a dental treatment to improve his oral hygiene. At the completion of treatment, a surveillance bronchoscopy was done, which showed both bronchial trees to be remarkably free of disease.

The patient again developed features of lingular consolidation in June 2012. Fiber optic bronchoscopy was done which showed yellowish, necrotic mass with sulfur granules at the level of bifurcation vision of left main bronchus into upper lobe and lingular segments [Figure 5]. The right bronchial tree was totally disease-free. As before, he again refused to take intravenous medications and hence was given a 6 months course of oral amoxicillin once more.

A surveillance bronchoscopy was done in March 2014 for follow-up. Although he denied having any respiratory symptoms, a similar looking yellowish mass lesion was

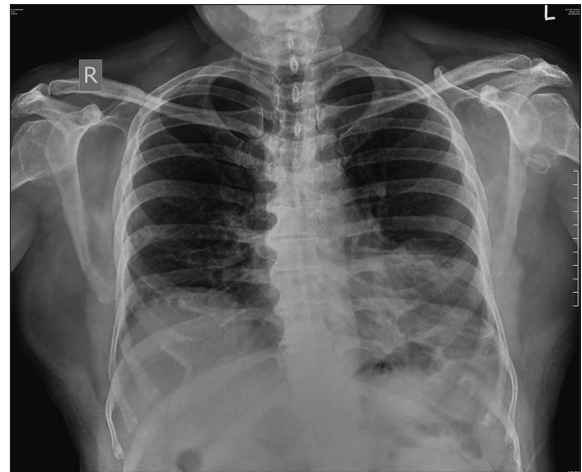


Figure 1: Chest X-ray posteroanterior view showing the presence of lingular consolidation

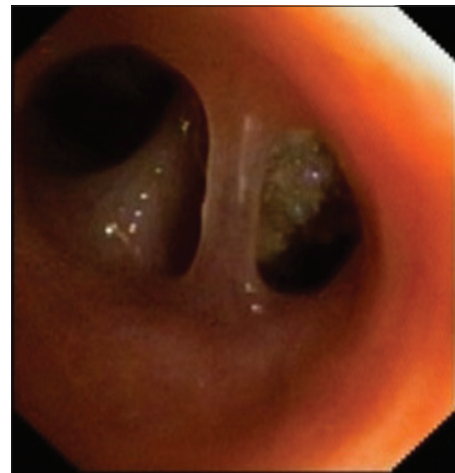


Figure 2: Flexible fiber optic bronchoscopy showing the presence of a yellowish mass with sulfur granules in the lingular bronchus

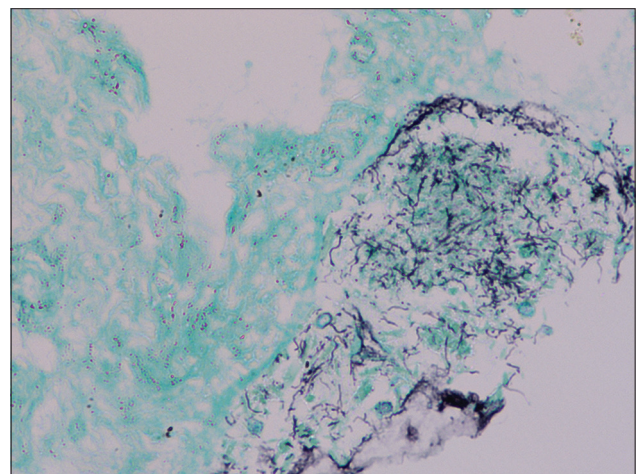


Figure 3: Fungal hyphae in histopathology specimen (Grocott's Methenamine Silver, x400)

detected to be present in the anterior segment of the right upper lobe. The left bronchial tree was entirely free of disease.

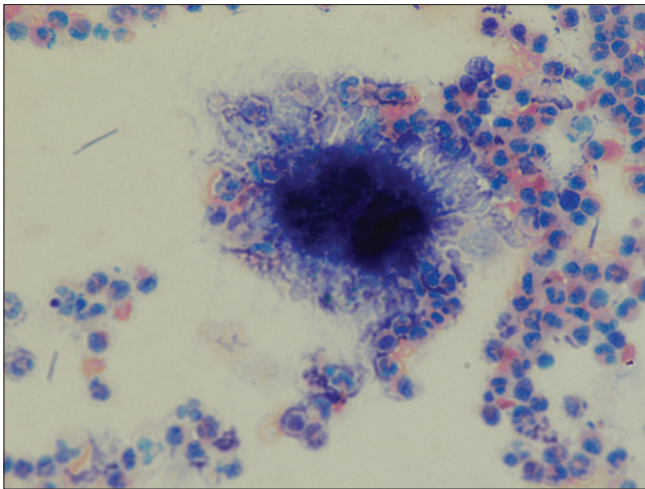


Figure 4: Fungal colonies in bronchoalveolar lavage fluid (Giemsa, $\times 400$)

The patient continued to have poor oral hygiene and this, along with his habit of pan chewing, was thought to be the reason for his developing recurrent actinomycosis. He was advised extraction of all his teeth and with much persuasion and reluctance, he underwent dental extraction. As the treatment for the newly detected actinomycotic lesion, he was administered doxycycline for 4 months. A repeat bronchoscopy, done after 6 months later for surveillance, revealed both the bronchial trees to be disease-free. Postdental extraction, 2 years later, he remains asymptomatic and disease-free till this point of time.

DISCUSSION

Endobronchial actinomycosis is a rare manifestation of actinomycosis.^[4] A thorough review of literature was done and as far as we could find out, there are hardly any case reports of recurrent endobronchial actinomycotic infections. Little is known about predisposing host conditions and the role of the immune system in actinomycosis. Poor dental hygiene, as was found in our patient, seems to be a disease-promoting and perpetuating factor. Presentation as opportunistic infections in immunocompromised patients is rarely observed. The inflammatory host response may play an additional role in the pathogenesis of clinical actinomycosis.

The most common clinical presentation of thoracic Actinomycosis is an indolent, slowly progressive process that involves some combination of the pulmonary parenchyma and pleural space. Chest pain, fever, weight loss, and less commonly, hemoptysis are prominent symptoms, and a cough, when present, is variably productive. There are no specific radiographic patterns for this infection. Although any lobe may be involved, upper lobe involvement is considered to be rare, since aspiration is the most common way in which pulmonary involvement occurs.^[5] The usual appearance is either as a mass lesion or pneumonia with or without pleural involvement.

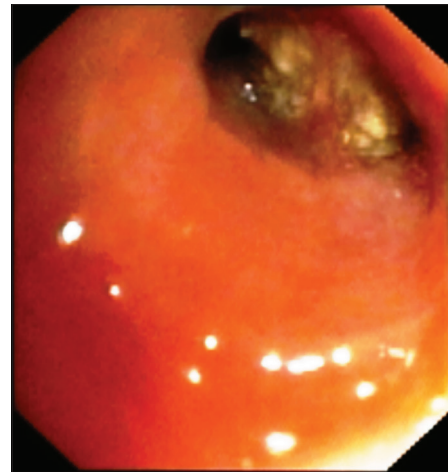


Figure 5: Flexible fiber optic bronchoscopy showing the presence of a yellowish mass with sulfur granules at the bifurcation of the left main bronchus

Pulmonary disease that extends across fissures or pleura, or which involves the mediastinum, or has contiguous bony disease should suggest actinomycosis and is also more readily appreciated by computerized tomography of the thorax. Extension to the chest wall with the development of a soft tissue mass, a draining sinus, or both is a telltale sign when present. In the absence of this classic scenario, however, thoracic actinomycosis is almost never suspected. It is mistaken for either malignant disease, with the diagnosis being made by histopathological examination post resection or for an empyema or pneumonia secondary to other more usual causes. In areas where tuberculosis is endemic, as in our country, actinomycosis may closely mimic tuberculosis. It is necessary to treat this disease both with high doses of antibiotics and for a prolonged period of time. Although therapy needs to be individualized, 18–24 million units of penicillin given intravenously for 2–6 weeks, followed by oral therapy with penicillin or amoxicillin for 6–12 months, is a reasonable guideline for serious infections and bulky disease.^[6] If the duration of therapy is extended beyond the resolution of measurable disease, then relapse, one of the clinical hallmarks of this infection, will be minimized. If the patient is allergic to penicillin, then tetracyclines are the next best alternative.^[7]

It has been postulated that prior endobronchial manipulations by disrupting mucosal barriers could predispose to the development of endobronchial actinomycosis.^[8] This could well explain the development of the disease in our patient, who started experiencing symptoms a year after undergoing argon plasma coagulation.

This case is being presented to highlight a few learning points. Recurrence of endobronchial actinomycosis is an extremely rare condition. It was observed that new endobronchial lesions kept appearing at different sites and that the previously detected lesions had completely disappeared at the completion of the antibiotic course.

The previous intervention in the airway could have led to the development of the disease, which was kept perpetuating despite optimal antibiotic therapy by the poor oral hygiene the patient had. The modern era of interventional pulmonology could well see the development of such indolent diseases much more frequently. The close resemblance to tuberculosis in endemic regions would often result in the prescription of empiric antitubercular therapy if a tissue diagnosis is not attempted. Again, since it mimics malignancy, early diagnosis is important as it would spare the patient of drastic surgeries and other expensive treatment modalities. Moreover, the elimination of the source of infection is of utmost importance to prevent recurrence as has been observed in our case.

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Conflicts of interest

There are no conflicts of interest.

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