Case Report

# Endobronchial actinomycosis with broncholithiasis presenting with hemoptysis—A case report

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## Abstract

Pulmonary actinomycosis is a rare chronic pulmonary infection caused by actinomyces. Pulmonary involvement is uncommon and often leads to a misdiagnosis of pulmonary tuberculosis or lung cancer. Endobronchial involvement is very rare, and broncholithiasis has occasionally been reported in association with pulmonary actinomycosis. Herein, we report a case of a 50-year-old male patient, who presented with a history of cough and hemoptysis diagnosed to be endobronchial actinomycosis with broncholithiasis by transbronchial biopsy.

**Keywords:** Actinomycosis, broncholithiasis, hemoptysis

## Introduction

Actinomycosis is a rare chronic, slowly progressive granulomatous disease caused by filamentous Gram-positive anaerobic or microaerophilic bacteria of the family Actinomycetaceae, genus Actinomyces,[1] with Actinomyces israelii as the most common causative agent.[2,3] It normally inhabits oropharynx, urogenital, and gastrointestinal tract;[3,4] however, interruption of the mucosal barrier predisposes individuals for developing actinomycosis. Cervicofacial infection is the most common manifestation, and pulmonary actinomycosis accounts for approximately 15–20% of cases,[1] wherein the infection is caused by aspiration of oropharyngeal secretions.[5] Poor dental hygiene, alcohol abuse, underlying structural damage because of pre-existing tuberculosis (TB), or foreign body aspiration are the usually reported risk factors. Broncholithiasis secondary to pulmonary actinomycosis is a rare condition.[5] It is often mistaken as malignancy or TB due to its atypical presentation. Herein, we report a case of biopsy-proven pulmonary actinomycosis with radiological features suggestive of mass with an endobronchial calcification.

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## Case Report

A 50-year-old diabetic non-smoker, non-alcoholic male patient presented with a 5-month history of cough and scanty mucoid expectoration associated with two episodes of blood-streaked sputum on presentation. He had history of multiple episodes of blood-streaked sputum in the past. He also had weight loss and loss of appetite since 5 months. He had no history of trauma, dental issues, or surgeries conducted in the past. On examination, he had good oral hygiene. Physical examination revealed normal vitals. General physical examination and systemic examination were unremarkable.

Laboratory examination revealed elevated erythrocyte sedimentation rate (ESR) levels with normal blood parameters. Sputum culture, cytology, and Ziehl-Neelsen (ZN) staining were inconclusive. Chest X-ray [Figure 1a] was normal. For further evaluation, cone beam computed tomography (CECT) chest showed spiculated mass lesion [Figure 1b] with an endobronchial



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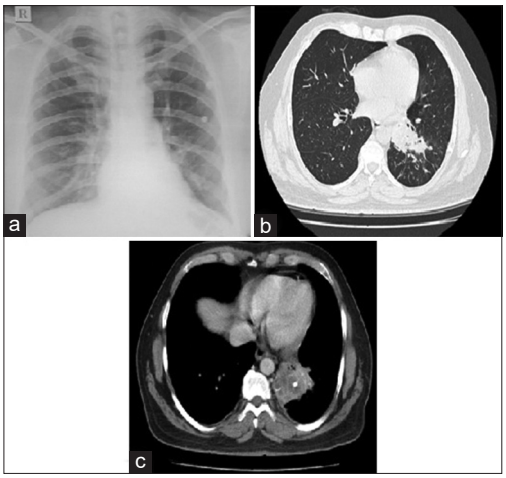


**Figure 1:** (a) Normal chest X-ray. (b) Chest showing left lower lobe mass with spiculated margins. (c) Endobronchial calcification within the mass invading into the left lower lobe

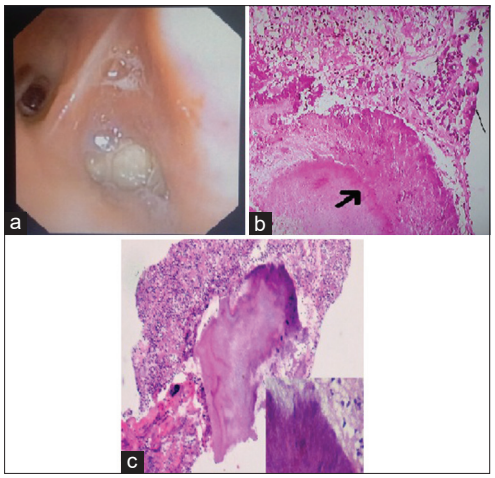


**Figure 2:** (a) Bronchoscopic image showing the mass occluding left lower lobe bronchus. (b) Histopathology-lymphoid follicles and areas of suppuration 100 × H&E. (c) Bacterial columns exhibiting a filamentous appearance at the periphery morphologically resembling actinomycosis 400 × H&E

calcification [Figure 1c] within the mass invading into the left lower lobe. Flexible bronchoscopy was conducted which showed a growth in the left lower lobe completely occluding the bronchus [Figure 2a]. Bronchoalveolar lavage (BAL) fluid was collected and sent for culture, ZN stain for acid-fast bacilli (AFB), and punch biopsy was taken from the growth.

Histopathological examination revealed lymphoid follicles and areas of suppuration with inter-spread bacterial columns exhibiting a filamentous appearance at the periphery morphologically resembling actinomycosis [Figure 2b, c]. BAL gram stain also revealed gram-positive filamentous bacilli suggestive of actinomycosis. As per recommendations, he was started on IV penicillin G 20 million units IV per day (Q6h) for 2 weeks and was discharged on oral penicillin V 2 gm/day (Q6h) for 6 months. On follow-up after 2 months, he is doing well with no further episodes of hemoptysis and planned to continue antibiotics for at least 6 months.

## Discussion

Pulmonary actinomycosis remains a challenging diagnosis as it is a rare entity with atypical clinical manifestations. Pulmonary actinomycosis may occur at any age and commonly affects patients between 30 and 60 years of age. Literature suggests that men get this infection more often than women do.[6] Symptoms of actinomycosis are nonspecific and include cough, productive sputum, hemoptysis, chest pain, weight loss, and fever which may mimic a variety of other lung diseases, such as malignancy, TB, pneumonia, and pulmonary abscesses, making the diagnosis complicated. Also, endobronchial actinomycosis may present with dyspnea due to airway obstruction. The most common radiological finding is consolidation followed by mediastinal or hilar lymph node enlargement, atelectasis, cavitation, bronchial obstruction, broncholithiasis, bronchiectasis, ground-glass opacity, pleural thickening, effusion, and empyema.[2] Lungs and pleura are infected either by aspiration or by direct spread from the pharynx or the neck or even upward through the diaphragm from the abdomen and rarely by hematogenous spread.

Endobronchial actinomycosis with broncholithiasis is a rare manifestation. Formation of broncholithiasis can be primary wherein the infection itself leads to the formation of broncholith, when the colonies of actinomyces are demonstrated throughout the broncholith not merely confined to the periphery. The formation can be from secondary infection of a pre-existing broncholith.[3,5] Inflammatory processes that occur aggravate the endobronchial lesion, resulting in progressive obstruction of the airway which shows consolidation picture. In most cases of the endobronchial form of actinomycosis, a broncholith was reported to be caused by a previous TB infection.[2] In our case, there was no history of TB. Hence, it could be primarily due to pulmonary actinomycosis infection.

Diagnosis is often challenging due to nonspecific symptoms. Also, it is common for the actinomyces organisms to colonize devitalized tissues, necrotic neoplasms, and foreign bodies; thus, the underlying lesion can be easily missed. Histopathology of lung tissue biopsies obtained by CT-guided transthoracic needle biopsy, bronchoscopic techniques, or even surgical resection shows sulfur granules—colonies of branching organisms that appear as round or oval basophilic masses with radiating eosinophilic terminal clubs on staining with hematoxylin-eosin representing the typical feature of pulmonary actinomycosis.[7] Grocott-Gomori stain is preferred as it differentiates actinomyces from nocardia or eumycetoma (other organisms producing sulfur granules). Cultural identification should be conducted in all cases even though bacterial confirmation has been achieved in only a minority of cases due to empirical antimicrobial pretreatment, overgrowth of associated bacteria, and the fastidious nature of actinomycetes. Molecular methods, such as polymerase chain

reaction (PCR) with 16S rRNA gene sequencing and mass spectrometry of the affected tissue, also seem to provide rapid and accurate microbiological confirmation of the disease.

This infection carries a good prognosis, and antibiotic treatment is generally curative. IV administration of 10–20 million units of penicillin for 4–6 weeks, followed by oral therapy with penicillin V (2-4 g/day) or amoxicillin (1.5-3 g/day) for 6–12 months, is a reasonable guideline for extensive infections.[8] If allergic to penicillin tetracycline, erythromycin, clindamycin, and ceftriaxone are alternative drug of choice.[9,10] Response to the therapy should be monitored, and imaging with CT is necessary to check for the resolution of the disease. In experienced centers, broncholiths can be removed by endoscopic techniques.

## Conclusion

When a broncholithiasis is associated with distal postobstructive pneumonia/mass lesion in places with endemic TB, one should consider a differential diagnosis of actinomycosis if biopsy is negative for TB.

## Abbreviations

TB = tuberculosis
AFB = acid-fast bacilli
PCR = polymerase chain reaction
CT = computed tomography

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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## Conflicts of interest

There are no conflicts of interest.

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