A Rare Bug and Recurrent Bleed

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Abstract

Isolated pulmonary actinomycosis is a rare entity. Its clinical features and radiological findings are nonspecific, making early diagnosis difficult for clinicians. We report a case of 40-year-old nonsmoker, immunocompetent male without an underlying structural lung disease who presented to us with recurrent hemoptysis and was diagnosed to have Actinomycosis after multiple readmissions.

Keywords: Pulmonary actinomycosis, recurrent hemoptysis, solitary pulmonary nodule

INTRODUCTION

Pulmonary actinomycosis is the third-most common form of the disease comprising only 15% of all forms of actinomycosis,^[1] after cervicofacial (60% cases), and abdominopelvic location (20% cases).^[2] Incidence has come down over the past few decades due to improved oral hygiene. Due to a lack of specific clinical and radiological features, it is often misdiagnosed as tuberculosis, lung abscess or malignancy,^[2,3] leading to incorrect treatment.

CASE REPORT

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A 40-year-old male, a bus driver by profession, presented with complaints of frequent episodes of blood-tinged sputum from the past 4 years with intermittent bouts of massive hemoptysis (>150 ml). There was no history of any comorbid illnesses, immunocompromised state, respiratory illnesses, smoking, or alcoholism in the past. Laboratory parameters were within the normal limits. The serology for HIV was negative. Clinical examination was inconclusive. He was admitted twice in the past for similar complaints.

During his first admission on March 2017, Chest X-ray showed a right lower lobe lesion. Subsequently, the high-resolution computed tomography (CT) of the chest was done, which was suggestive of right lower lobe basal segment solitary pulmonary nodule (SPN) measuring 2.1 cm \times 1.6 cm, raising the possibility of neoplastic etiology. CT-guided Fine-needle aspiration cytology (FNAC) was inconclusive. Biopsy could

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not be performed due to its small size and intraparenchymal location. Sputum culture grew Streptococcus pneumoniae. He was managed with IV antibiotics and antifibrinolytics.

Following a few months, on August 2017, he presented with intermittent bouts of massive hemoptysis. There was no radiological worsening seen on the chest-X ray and CT chest. Repeat FNAC showed infective/inflammatory etiology. Sputum culture showed heavy growth of pseudomonas. He was managed with antibiotics and antifibrinolytics. On follow-up, after a month patient was stable with no further hemoptysis, but the lesion persisted radiologically. The patient was offered the option of resection surgery, but he declined it.

He was symptomatically better during the interim until January 2020 when he presented again with similar complaints. Persistent hemoptysis led to repeat contrast-enhanced CT of the thorax, which showed an increase in the size of SPN to 2.3 cm \times 2.3 cm \times 3.1 cm [Figure 1a].

After stabilization, he underwent fiberoptic diagnostic bronchoscopy. Bronchoscopy showed endoluminal narrowing of right lower lobe bronchus. Bronchoalveolar lavage culture

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grew Pseudomonas; Acid-fast bacilli by Ziehl–Neelsen stain and Gene Xpert was negative; fungal cultures were negative as well. Bronchoscopic brushing was negative for malignant cells. Repeat CT-guided FNAC was done which showed a tangled cluster of filamentous organisms with cotton ball appearance on Papanicolau (PAP) and May Grunwald-Giemsa stain (MGG) [Figure 1b]. These Gram-positive, nonacid fast, Periodic Acid–Schiff negative, branching, and filamentous bacteria were identified as colonies of Actinomyces on special stain of Gomori Methenamine silver [Figure 1b].

He was eventually diagnosed to have pulmonary actinomycosis as the cause for his recurrent hemoptysis and nonresolving right lower lobe lesion 4 years after the onset of his symptoms. After 1 week of intravenous penicillin, the patient was symptomatically better, but however streak hemoptysis persisted. In view of recurrent hemoptysis for 4 years, lack of definitive tissue diagnosis, and gradual increase in the size of lesion radiologically patient was referred to cardiothoracic surgeon and resection surgery was advised. After 6 weeks of oral antibiotics, he underwent wedge resection [Figure 1a]. Histopathology of the lung parenchyma confirmed the presence of actinomyces on routine H and E and special



Figure 1: (a) Contrast enhanced Computed Tomography of the chest showing well-defined lobulated soft tissue density lesion of 2.3 cm \times 2.3 cm \times 3.1 cm can be seen in the posterobasal segment of right lower lobe (Right); Gross specimen of resected lung revealing numerous sulphur granules (Left). (b) Tangled clumps of organisms with cotton ball appearance (MGG \times 10 and PAP, \times 20) (Top); Colony of Filamentous organisms highlighted by GMS stain (\times 40) (Bottom)

stains. He is asymptomatic postsurgery and is under regular follow-up.

DISCUSSION

Actinomyces species is a nonmotile, nonspore-forming, nonacid fast, obligate anaerobe, and Gram-positive filamentous bacteria. The most common form of infection is cervicofacial followed by abdominopelvic and pulmonary form.^[2] Pulmonary actinomycosis is a chronic, suppurative granulomatous disease. It has a bimodal age distribution in the second decade and fourth-fifth decade. Men are twice more affected than women.^[4] Poor oral hygiene, alcoholism, immunosuppressive conditions, diabetes, and underlying structural lung diseases such as emphysema, chronic bronchitis, and bronchiectasis can predispose for infection.^[5,6] They are commensals seen in the mucosal lining of the oropharynx, gastrointestinal tract, and female genitalia.^[2] It results from aspiration of oropharyngeal and gastrointestinal secretions.^[7] Clinical manifestation is like that of typical pneumonia, i.e., cough, fever, and breathlessness.^[6] Hemoptysis as a presenting symptom is very rare.^[2,8-10] The radiological spectrum can vary from pulmonary infiltrates to cavitary mass. Pneumonia secondary to actinomyces are more commonly located in the right middle lobe and left lower lobe.^[2] Clinical features and radiological imaging are equally inconclusive, hence it is often misdiagnosed.^[5] The average duration of diagnosis is around 6 months.^[2] Diagnosis is performed microbiologically by the demonstration of bacteria, using special stains (such as MGG, GMS, Brown-Brenn, and PAP) in sputum, bronchial lavage, or tissue biopsy. As a positive sputum culture is seldom seen, a tissue biopsy is often needed.[11] These organisms are difficult to culture as they as fastidious in nature. Pulmonary actinomycosis is treated with antibiotics with or without surgery. Surgery is sometimes indicated not only to expedite the diagnosis but also for optimizing the treatment as well.^[2] High-dose intravenous penicillin for 2-6 weeks is the drug of choice; followed by long-term therapy of oral penicillin for 12 months.^[2]

Pulmonary actinomycosis can be a difficult condition to diagnose. Diagnosis of actinomycosis hinges on clinicohistopathological correlation and response to a specific treatment. Pulmonary actinomycosis should be considered in patients with recurrent hemoptysis of obscure etiology when routine workup for infections, inflammatory, and neoplastic etiologies are inconclusive. Microbiological isolation and identification of actinomycosis from sputum or bronchial lavage sample are difficult and rare^[11] as in our case repeated sputum and lavage cultures were negative. Positive cultures are only seen in about 50% of cases, which may be attributed to either prior use of antibiotics or an imperfect environment for the bacteria to grow.^[2] If clinically suspicious, it is important to alert the pathologist and microbiologist so that special stains can be performed and disease can be diagnosed early. In quite a few cases the diagnosis is so elusive that it necessitates resection surgeries. Diagnosis is often performed on postoperative resected lung specimens.^[8,10] Our case is an eye-opener as actinomycosis with pulmonary involvement and hemoptysis should be considered in patients even in the absence of immunocompromised and chronic lung disease states.

Actinomycosis as a cause recurrent hemoptysis and nonresolving pneumonia in an otherwise immunocompetent patient with no underlying structural lung disease was a clinical surprise.

Declaration of patient consent

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

Research quality and ethics statement

The authors followed applicable EQUATOR Network (http:// www.equator-network.org/) guidelines, notably the CARE guideline, during the conduct of this report.

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Conflicts of interest

There are no conflicts of interest.

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