

Broncholithiasis secondary to pulmonary actinomycosis

Author list:

Nicholas R. Henry, MSc, RRT-NPS, AE-C
Assistant Professor
Department of Respiratory Care
Texas State University
San Marcos, TX

John D. Hinze, DO, FCCP, FACP
Pulmonary and Critical Care Consultants of Austin
Austin, TX

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Abstract

We report a case of broncholithiasis secondary to pulmonary actinomycosis. Broncholithiasis has occasionally been reported in association with actinomycosis but it is unclear if actinomycosis leads to lymph node calcification with subsequent erosion into the airway producing a broncholith or if an existing generic broncholith is secondarily colonized with *Actinomyces*. We describe a patient with post-obstructive pneumonia whose computerized tomography findings demonstrated calcified nodules obstructing the bronchus intermedius and distal necrotizing pneumonia. Histopathologically, the nodules demonstrated sulfur granules containing gram-positive branching filamentous organisms consistent with *Actinomyces*. The finding of *Actinomyces* throughout the broncholith is strong evidence that the etiology of the broncholithiasis in this patient is a primary pulmonary infection of *Actinomyces*.

Key Words: Actinomycosis; Actinomyces; broncholithiasis; broncholith; hemoptysis; lithoptysis.

Introduction

Broncholithiasis secondary to pulmonary actinomycosis is considered to be a rare condition.^{1,2} Actinomycosis is a chronic pulmonary infection caused by an *Actinomyces* species, with *Actinomyces israelii* being the most common pathogen.³⁻⁶ *Actinomyces* are gram-positive, filamentous branching rods that are not acid-fast and produce colonies that are round to oval in shape.^{2,4,5,7} *Actinomyces* colonies produce “sulfur granules” that are yellowish in color.^{4,5} *Actinomyces* species are considered normal flora of the human oropharynx, gastrointestinal tract and urogenital tract^{5,7} but can cause infection by invading structures and organs through a mucosal lesion.⁸⁻⁹ Actinomycosis is considered to be a rare infection with an incidence rate of approximately 1 in 300,000 people.⁹ Pulmonary infection due to *Actinomyces* is presumed to be caused by the aspiration of oropharyngeal secretions into the lungs^{1,5} and pulmonary actinomycosis is estimated to account for approximately 15-20% of actinomycosis cases.⁸ Actinomycosis is confirmed by the isolation and culture of *Actinomyces* colonies and/or the histopathologic findings of sulfur granules.^{4,7}

Broncholithiasis is a condition in which calcified nodules or lymph nodes are identified within or migrating into a bronchial lumen.¹⁰⁻¹² The erosion of the calcified nodule into an airway is a result of continuous movement of the thoracic structures during active breathing or deglutition,^{5,11,13} thereby causing partial or complete obstruction of the airway potentially leading to an obstructive pneumonia.¹³ Chronic inflammatory processes or granulomatous diseases such as tuberculosis, histoplasmosis,

coccidioidomycosis, nocardiosis, cryptococcosis, aspergillosis and silicosis are common causes of broncholithiasis.^{1,10-12}

We describe a rare clinical case of broncholithiasis secondary to pulmonary actinomycosis. Literature regarding broncholithiasis coupled with pulmonary actinomycosis is limited and the description of this rare occurrence may add more knowledge for clinical use. Furthermore, it depicts an additional form of airway obstruction.

Case Summary

A 61-year-old Caucasian female presented to the emergency department with weakness, fatigue, increased sputum production, and a four-month history of hemoptysis. She stated that she smoked tobacco from age 15 to 25 years old with less than 10 package-years.

Eleven years prior to presentation, the patient developed a chronic productive cough. Eight years later, she developed a protracted pneumonia resulting in significant weight loss and was diagnosed with a large pulmonary abscess in the right lung that was treated with a percutaneous drain removing approximately one liter of fluid. Unfortunately no other information was obtained regarding the pulmonary abscess because it was diagnosed and treated at another institution. The patient states that the abscess fluid cultures were negative for tuberculosis.

Upon presentation, the patient appeared chronically ill with a productive cough, producing yellow sputum with occasional blood streaks. Her highest reported body

temperature over the previous 24 hours was 102.2° Fahrenheit. Upon presentation, the patient had a heart rate of 88 beats/min, respiratory rate of 24 breaths/min, blood pressure of 135/62 mmHg, and a SpO₂ on room air of 96%. An examination of her neck revealed a mildly enlarged anterior cervical lymph node on the right side. Auscultation revealed diminished breath sounds over the right base with a regular heart rate and rhythm. There was no evidence of digital clubbing, edema or cyanosis of the extremities. The remainder of her physical exam was unremarkable.

A contrast-enhanced computerized tomography (CT) scan of the patient's chest was performed in the emergency department revealing calcified right hilar lymphadenopathy (Figure 1), near complete consolidation of the right lower lobe with cavitation and necrosis, and tubular bronchiectasis in the right lower lobe (Figure 2). The chest CT scan further revealed an enlarged lymph node anterior to the superior vena cava that measured 14 mm in the short axis, and atelectasis and consolidation in the right middle lobe.

The patient did not possess MRSA risk factors but was started on a course of vancomycin and piperacillin/tazobactam upon hospital admission by the primary team. During the patient's admission, three sputum samples were obtained. Each smear was negative for acid-fast bacillus and acid-fast bacillus was not isolated after 6 weeks via culture.

Fiberoptic bronchoscopy revealed the bronchus intermedius completely filled with creamy purulence. Once the bronchus intermedius was suctioned clear, several large

impacted stones, consistent with broncholithiasis, were encountered (Figure 3). A bronchoalveolar lavage (BAL) of the bronchus intermedius was performed and two small bronchololiths were extracted by suction. The BAL had a dense neutrophilic infiltrate with normal respiratory flora and no anaerobes were isolated after 5 days via culture. The BAL was negative for yeast and fungus via stain and culture, negative for pneumocystis carinii via stain and negative for acid-fast bacillus via stain and culture.

The remaining bronchololiths were extracted in pieces by rigid bronchoscopy with the assistance of Neodymium-doped Yttrium Aluminum Garnet laser photoablation. The bronchololiths were described as tan-white, irregularly shaped fragments ranging from 0.1-0.8 centimeters in diameter. Microscopic examination of the bronchololiths revealed polypoid fragments of granulation tissue, hemorrhage and calcified material. The biopsy of the bronchololiths consisted of purulent exudate containing actinomycotic sulfur granules and gram-positive branching rods throughout the bronchololiths (Figure 4). A Gomori's Methenamine Silver stains of the bronchololiths were negative for fungal organisms. There was no evidence of neoplasm.

The patient's empirical antibiotic treatment was then switched to a continuous infusion of intravenous penicillin. Clinically improved with symptoms resolving, the patient was discharged home on a continuous infusion of 12 million units penicillin per day for four weeks followed by a P.O. dosage of 500mg amoxicillin TID. The patient continued to take the P.O. amoxicillin, as ordered, for 4 months but then stopped taking it due to neuropathy. Throughout outpatient treatment, her symptoms abated and a follow-

up chest computerized tomography scan demonstrated modest bronchiectasis in the right lower lobe (Figure 5) and additional right calcified hilar lymph nodes that have not eroded into an airway (Figure 6).

Discussion

The clinical and radiological findings of the reported case shares many characteristics of previously reported cases of pulmonary actinomycosis. Previously reported cases have described the presence of productive cough, hemoptysis, fever and chest pain,^{2,13} which is consistent with the described case except for chest wall pain. Radiologic findings of previous literature include air-space consolidation with cavitation, lymphadenopathy, bronchiectasis within the consolidation, localized pleural thickening, and pleural effusion.^{2,4} which is also consistent with the described case except for pleural thickening and effusion. The described case also shares CT findings described in previous case reports of endobronchial actinomycosis associated with broncholithiasis, which includes a calcified density occluding an airway with distal airspace consolidation, bronchiectasis, and enlarged hilar lymph nodes.^{1,5}

In our described case, we suggest that the actinomycosis caused the lymphadenopathy with subsequent calcification and eventual erosion into the airway. We assert that her prolonged course, subsequent evidence of pulmonary actinomycosis and the finding of gram-positive branching rods and sulfur granules throughout the broncholiths provide support that the broncholiths were secondary to the infection, and not vice versa. In addition, fungal infections, tuberculosis and bronchogenic carcinoma

were ruled out via negative studies. Previous literature has described cases of endobronchial actinomycosis associated with broncholithiasis,^{1,5} however there was no evidence of endobronchial involvement in the described case. Sulfur granules were only isolated within the broncholiths and *Actinomyces* was not isolated via BAL. As a result, the broncholiths were most likely caused by a parenchymal infection leading to lymphadenopathy, calcification and erosion into the airway.

Kim et al. retrospectively reviewed nine cases from their institution who were diagnosed with endobronchial actinomycosis and concluded that in their cases, preexisting broncholiths were likely secondarily infected with *Actinomyces*, which further caused the broncholiths to enlarge.⁵ We do not believe this occurred in the described case because the sulfur granules were identified throughout the broncholith and were not exclusively identified in the periphery of the broncholith. In the authors' opinion, it would be difficult and implausible for *Actinomyces* to penetrate and produce sulfur granules throughout a preexisting broncholith due to the broncholith's calcium concretion.

The presented case is clinically important because the evidence of sulfur granules throughout the broncholiths provides support that pulmonary actinomycosis can be an etiology of broncholithiasis leading to airway obstruction. The proper management of this particular case was dependent upon the definitive diagnosis and treatment for both pulmonary actinomycosis and broncholithiasis, concurrently.

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Figure Legends

Figure 1. A contrast-enhanced computed tomographic scan of the chest displaying right hilar lymphadenopathy and a broncholith obstructing the bronchus intermedius.

Figure 2. A contrast-enhanced computerized tomographic scan displaying consolidation and necrosis of the right lower lobe distal to the broncholith.

Figure 3. Bronchoscopy image displaying two large impacted broncholiths occluding the bronchus intermedius.

Figure 4. A hematoxylin-eosin stain of a broncholith revealing sulfur granules and branching rods consistent with *Actinomyces*.

Figure 5. A contrast-enhanced computerized tomographic scan displaying bronchiectasis of the right lower lobe following antimicrobial treatment.

Figure 6. A computerized tomographic coronal reconstruction of the chest following antimicrobial treatment displaying additional right calcified hilar lymph nodes that have not eroded into an airway.

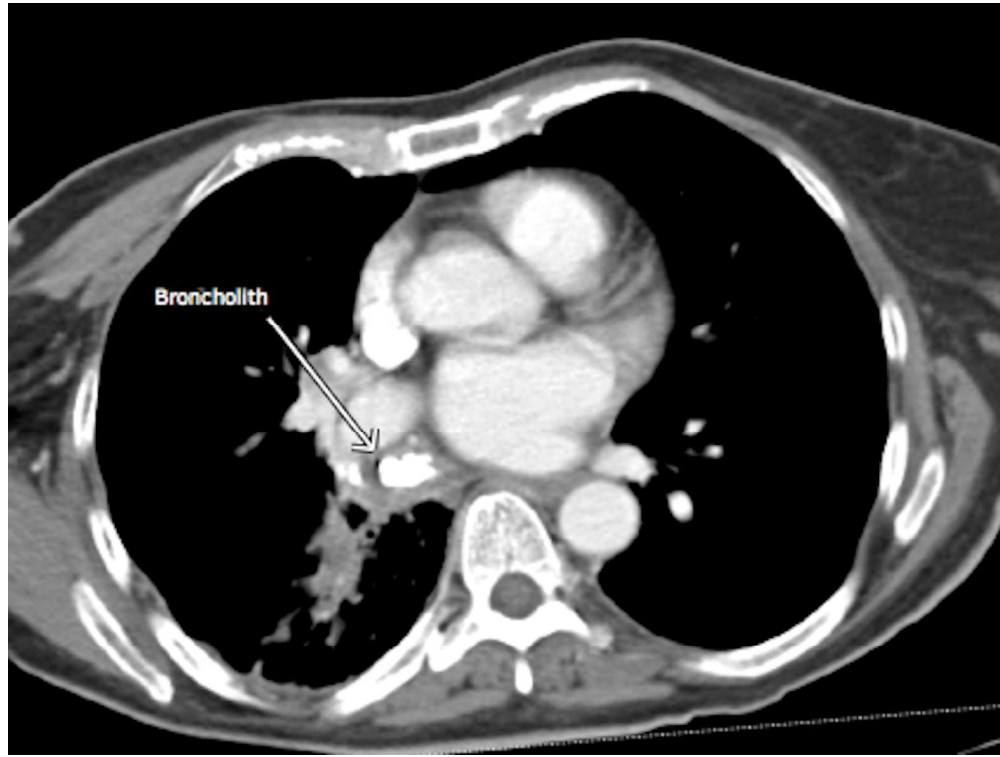


Figure 1. A contrast-enhanced computed tomographic scan of the chest displaying right hilar lymphadenopathy and a broncholith obstructing the bronchus intermedius.
150x112mm (300 x 300 DPI)



Figure 2. A contrast-enhanced computerized tomographic scan displaying consolidation and necrosis of the right lower lobe distal to the broncholith.
130x101mm (300 x 300 DPI)

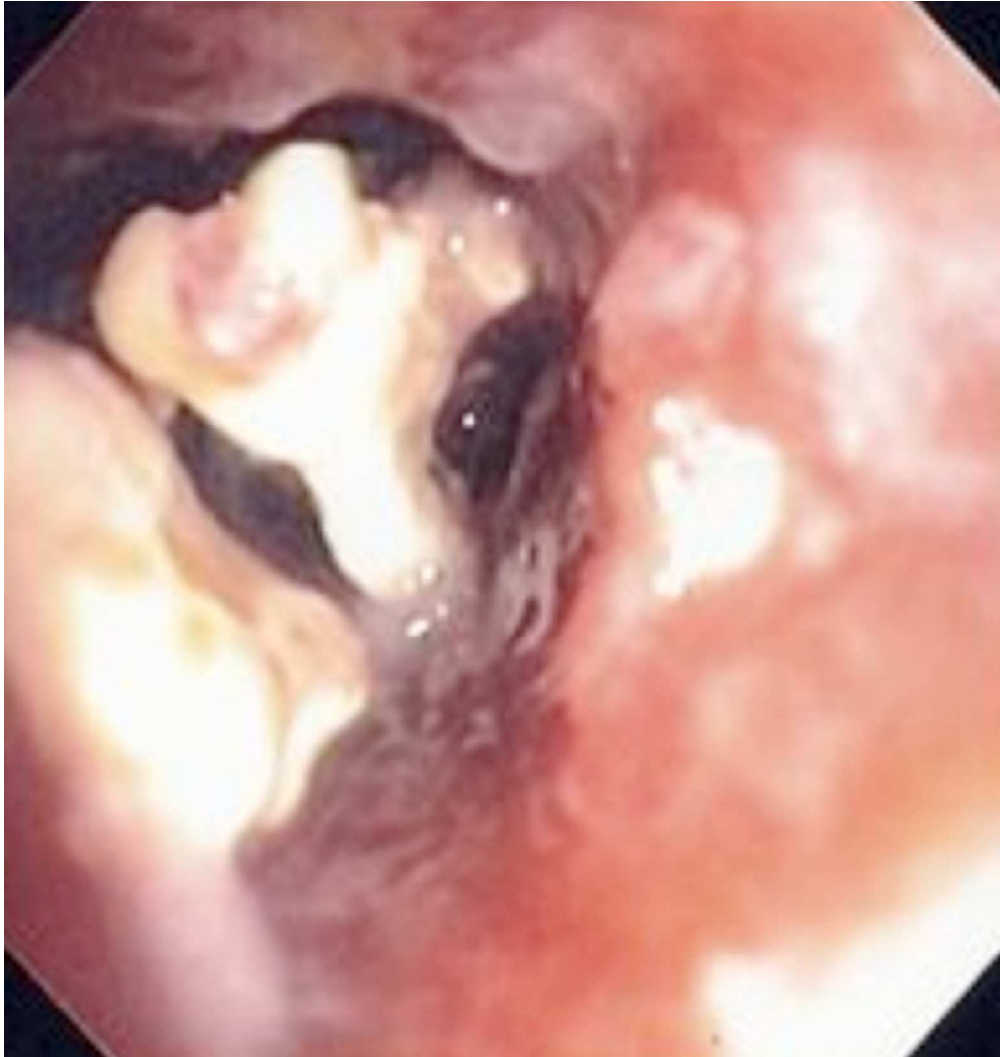


Figure 3. Bronchoscopy image displaying two large impacted bronchololiths occluding the bronchus intermedius.

134x141mm (300 x 300 DPI)

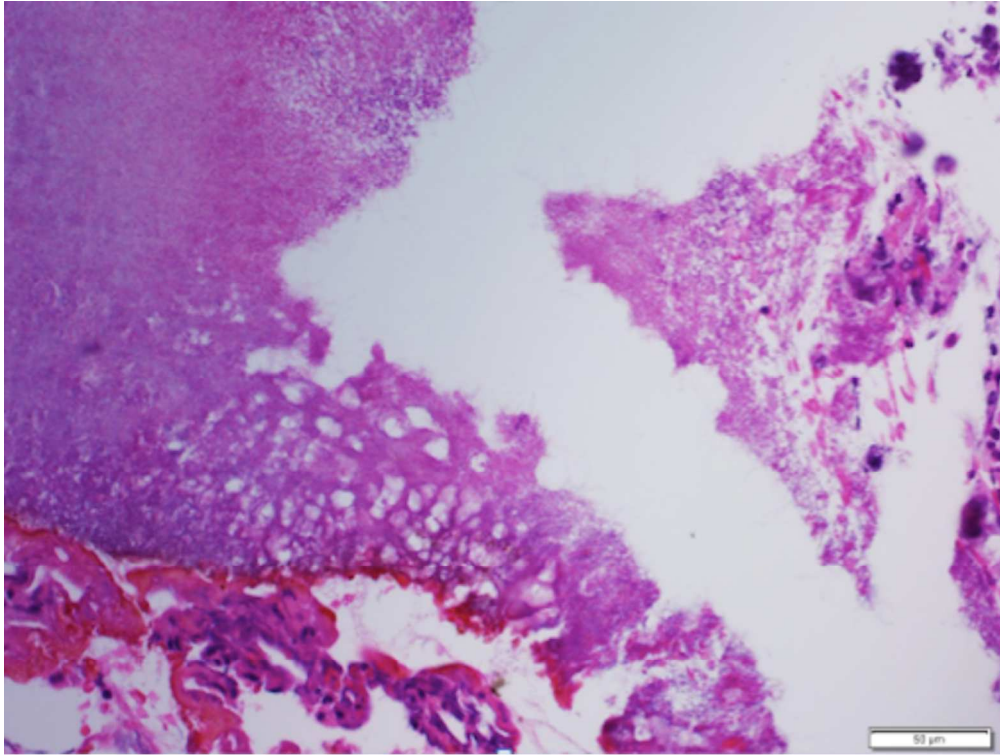


Figure 4. A hematoxylin-eosin stain of a broncholith revealing sulfur granules and branching rods consistent with Actinomyces.
179x135mm (300 x 300 DPI)

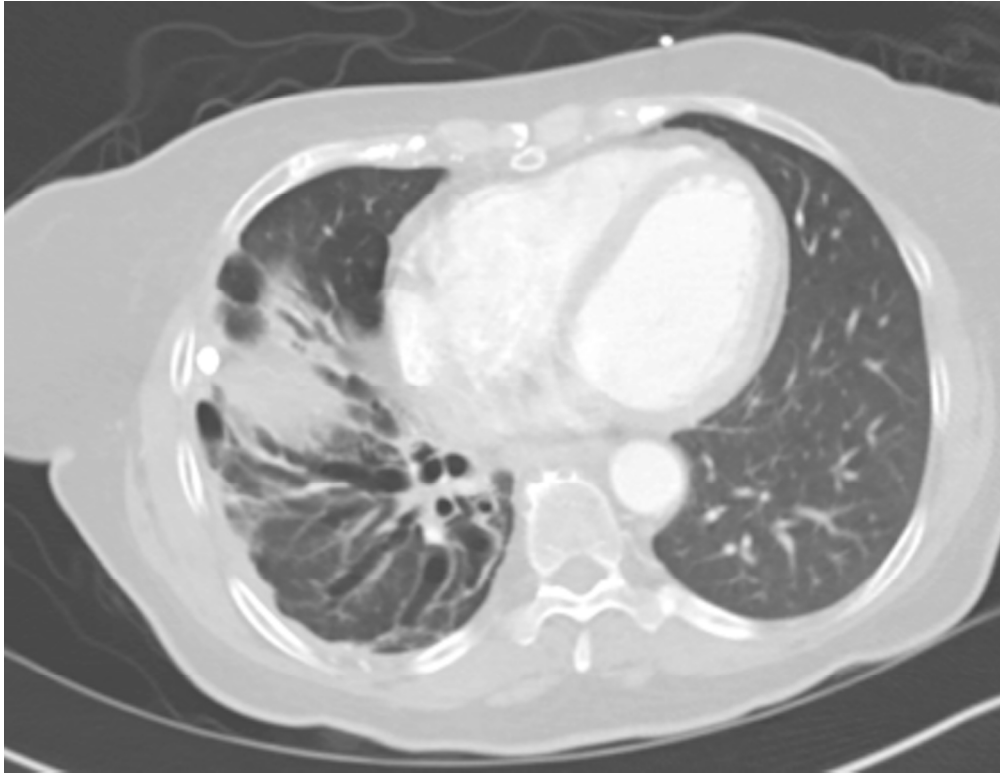


Figure 5. A contrast-enhanced computerized tomographic scan displaying bronchiectasis of the right lower lobe following antimicrobial treatment.
131x101mm (300 x 300 DPI)



Figure 6. A computerized tomographic coronal reconstruction of the chest following antimicrobial treatment displaying additional right calcified hilar lymph nodes that have not eroded into an airway.
199x137mm (300 x 300 DPI)