Pulmonary actinomycosis in a male patient with a tracheal bronchus

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BACKGROUND: Tracheal bronchus is a congenital malformation comprising an abnormal bronchus originating from the trachea or other bronchus. This malformation has been associated with recurrent pneumonia in children, but is rarely associated with infection in adults. Actinomyces species are rare causes of necrotizing pneumonias that often masquerade as malignancy, lung abscesses and tuberculosis.

METHODS AND RESULTS: A case involving a 46-year-old man with a tracheal bronchus and chronic pneumonia syndrome is presented. Bronchialveolar lavage and transbronchoscopic needle biopsy demonstrated the presence of Actinomyces meyeri and Fusobacterium species.

CONCLUSIONS: The present article reports the first documented case of actinomycosis occurring in a patient with a tracheal bronchus.

Key Words: Accessory tracheal bronchus; Actinomyces; A meyeri; Bronchus suis; Congenital lung abnormality; Pulmonary actinomycosis

Tracheal bronchus is a congenital condition characterized by an abnormal bronchus originating from the trachea or right main bronchus and directed to the right upper lobe (1,2). In some cases, the bronchus arises from the right side of the trachea, proximal to the carina, and is referred to as ‘bronchus suis’ (pig bronchus) because a tracheal bronchus is normally present in swine (1). In children, this condition has been associated with recurrent pneumonia. In intubated patients, this condition has been associated with atelectasis and postobstructive pneumonia due to obstruction of the tracheal bronchus by the endotracheal tube (1). Pulmonary actinomycosis is an anaerobic infection secondary to Actinomyces organisms, which are normal commensals of oral and intestinal flora. Actinomyces species are a rare cause of necrotizing pneumonia, often initially misdiagnosed as malignancy, lung abscesses or tuberculosis (3,4). We present a case involving a 46-year-old man with a tracheal bronchus (bronchus suis) infected with Actinomyces meyeri. To our knowledge, the present report documents the first reported case of pulmonary actinomycosis in a patient with this congenital abnormality. The pathophysiology, diagnosis and management of pulmonary actinomycosis are reviewed.

CASE PRESENTATION

A 46-year-old man was admitted to hospital for evaluation of a five-month history of progressive right-sided pleuritic chest pain and productive cough with purulent blood-tinged sputum. He also reported recent night sweats. Medical history included coronary artery disease, hypertension, type 2 diabetes mellitus, alcohol abuse and 30 pack-year smoking. He had no history of tuberculosis, and previous tests for HIV were negative. He was afebrile with a respiratory rate of 24 breaths/min and an oxygen saturation of 99% on room air. Crackles were present in the right upper lobe, and his dentition was poor.

Over the five months preceding his admission, the patient presented to the emergency room on two occasions. At the time of the first presentation, he was diagnosed with community-acquired pneumonia and subsequently prescribed clarithromycin, resulting in an equivocal response. Three months later, he returned to the emergency room and was prescribed 21 days of amoxicillin/clavulanic acid. With this second course of antibiotics, he reported a notable improvement in his chest pain and sputum production. Unfortunately, his improvement was transient; his symptoms returned shortly after his antibiotic regimen was completed.

During this admission, the patient was initially treated with intravenous levofloxacin for presumptive pneumonia. Chest x-rays demonstrated progressive focal airspace disease with cavitation in the right upper lobe (Figure 1). This was confirmed on contrast-enhanced computed tomography (CT), which also demonstrated ground-glass opacities in the adjacent lung with interlobular septal and bronchial wall thickening. Multiple enlarged mediastinal lymph nodes were present. There was no evidence of pleural disease or chest wall invasion.

Bronchoscopy revealed a small accessory bronchial orifice on the right lateral tracheal wall just proximal to the bifurcation of the trachea into the left and right bronchi. Copious purulent secretions were removed from this tracheal bronchus and bronchoalveolar lavage was performed. The bronchus was carefully inspected to confirm the presence of bronchial mucosa.

Following the bronchoscopy, coronal reformations of previous CT images (Figure 2) confirmed the presence of a tubular air-containing structure arising from the lateral wall of the lower trachea and extending to the right upper lobe lesion, consistent with a tracheal bronchus.

Cytological examination of transbronchial needle aspirate and bronchoalveolar lavage samples did not show any evidence of malignancy. Stains for microorganisms demonstrated filamentous bacteria. Subsequent culture of both aspirate and lavage samples demonstrated the presence of Actinomyces meyeri and Fusobacterium species. Stains for acid-fast bacilli and cultures for Mycobacterium tuberculosis were negative.

When the culture results became available, the patient’s antibiotic regimen was changed from levofloxacin to penicillin G 5 million units
Actinomyces organisms are filamentous Gram-positive microaerophilic bacteria that are normal commensals of the oropharynx and gastrointestinal tract. They typically reside in carious teeth, dental plaque, and gingival and tonsillar crypts (3-7). The cervicofacial area is most commonly affected, accounting for 50% to 60% of cases (4). The classic description is that of a middle-age man presenting with a large jaw mass (3). Pulmonary actinomycosis is the next most frequent infection, accounting for 15% to 20% of reported cases. The remaining infection sites include abdominopelvic (20%), central nervous system (2%) and, very rarely, cutaneous, ophthalmic, cardiac, genitourinary and disseminated disease (3,4).

Diagnosis is challenging because microbiological identification may be difficult. Actinomyces shares many clinical and imaging findings with chronic suppurative lung infections, and may be mistaken for tuberculosis, nocardiosis, histoplasmosis, blastomycosis or mixed anaerobic infections, in addition to noninfectious etiologies such as pulmonary infarction and malignancy (3-7). Plain x-rays are nonspecific, with a nonsegmental pneumonia usually in the right upper lobe, peripherally crossing fissures. CT of the chest typically demonstrates segmental airspace consolidation with adjacent pleural thickening and microabscesses or necrotic tissue. Invasion of the chest wall may occur and mimic malignancy. The presence of air bronchograms is suggestive of actinomycosis as opposed to malignancy, which often obliterates the bronchi (3,7). Up to one-third of cases of actinomycosis are initially misdiagnosed as malignancy (3,10).

Culture of bronchoalveolar lavage secretions alone is not reliable because positive results may represent contamination from oral flora. Culture samples must be obtained anaerobically with a protected specimen brush. Prolonged incubation for up to 25 days may be required (9). Often, a diagnosis of actinomycosis is made only after resection (3,4,7,8). In many series, six months was the average duration of illness before a definitive diagnosis was made (3).

Among the Actinomyces species, A meyeri has a greater tendency for hematogenous dissemination and lung infection. Infection is believed to result from aspiration of oral and gastric contents, which is supported by the higher prevalence of alcoholism and poor dentition in infected patients, and the bibasilar predominance of the disease radiographically (7-9). Neglecting anatomical boundaries and crossing interlobar fissures, A meyeri induces necrosis, fibrosis and cavitation (3,5). It may further invade the pleura, chest wall, soft tissues and bony structures, and produce sinus tracts (3). Other organisms, such as Fusobacterium species and Bacteroides species, are frequently found in association with the Actinomyces organisms and are believed to enhance its pathogenicity by creating an anaerobic environment in which phagocytosis is inhibited (3). Dyspnea, productive cough and pleuritic chest pain are the typical presenting symptoms (6,8).

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The patient was clinically assessed for the final time 24 weeks after initiation of penicillin. By then, he had received four weeks of intravenous penicillin followed by 20 weeks of oral penicillin, for a total duration of penicillin therapy of 24 weeks. The total duration of metronidazole therapy was six weeks. At this point, the patient was essentially asymptomatic, with a normal respiratory examination. A final chest x-ray demonstrated some persisting right upper lobe opacity, compatible with scarring.

**DISCUSSION**

Actinomyces species A meyeri has a greater tendency for hematogenous dissemination and lung infection. Infection is believed to result from aspiration of oral and gastric contents, which is supported by the higher prevalence of alcoholism and poor dentition in infected patients, and the bibasilar predominance of the disease radiographically (7-9). Neglecting anatomical boundaries and crossing interlobar fissures, A meyeri induces necrosis, fibrosis and cavitation (3,5). It may further invade the pleura, chest wall, soft tissues and bony structures, and produce sinus tracts (3). Other organisms, such as Fusobacterium species and Bacteroides species, are frequently found in association with the Actinomyces organisms and are believed to enhance its pathogenicity by creating an anaerobic environment in which phagocytosis is inhibited (3). Dyspnea, productive cough and pleuritic chest pain are the typical presenting symptoms (6,8).

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The pathophysiology of our patient’s disease was probably multifactorial. He had a number of classic risk factors for actinomycosis including alcohol use, tobacco smoking and poor dentition, which predisposed him to infection. It is unknown whether the presence of a tracheal bronchus was also a risk factor for *Actinomyces* infection in our patient, although it is notable that the infection involved the area of lung served by the tracheal bronchus. At the very least, the tracheal bronchus complicated assessment of this patient’s problem by raising the possibility of fistula, a well-described complication of actinomycosis. The second bronchoscopy was performed after four weeks of therapy to verify the presence of the tracheal bronchus and rule out a fistula. To our knowledge, actinomycosis in an adult with a tracheal bronchus has not been previously described.

Our patient’s indolent course over several months is typical of pulmonary actinomycosis. Standard treatment of deep-seated actinomyces consists of intravenous penicillin G 150,000 units/kg/day to 200,000 U/kg/day, or 10 million units/day to 20 million units/day, divided three times daily or four times daily, for four to six weeks. This is followed by oral penicillin V 2 g/day to 4 g/day, divided three or four times daily, for an additional six to 12 months (11). Metronidazole is frequently added to cover accompanying anaerobic oral flora such as *Fusobacterium* (11). In some cases, the duration of antimicrobials may be further prolonged if the infection is extensive and involves poorly vascularized or devitalized tissue (5). Surgery may be required for empyemas with discharging fistulae, and rarely to control life-threatening hemoptysis (8). Untreated, it may ultimately be fatal (5,6). However, prognosis is excellent, with cure rates of 90% with early diagnosis and appropriate antimicrobials (3,4,6,12).

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**REFERENCES**
