A Case of Primary Endobronchial Actinomycosis Presenting as a Complicated Parapneumonic Effusion

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Endobronchial actinomycosis is a rare but important and challenging diagnosis to make. We report a case of 57-year-old man who presented with a fever and a right-sided chest pain diagnosed as an endobronchial actinomycosis. Chest computed tomography showed a segmental obstruction and consolidations in right middle lobe combined with an ipsilateral multiloculated pleural effusion. Bronchoscopic biopsy of a mass obstructing the medial segmental bronchus of right middle lobe revealed actinomycosis. This is the first reported case of primary endobronchial actinomycosis that presented with a chest pain and a complicated parapneumonic effusion. Primary endobronchial actinomycosis can be a cause of complicated parapneumonic effusion.

Keywords: Actinomycosis; Bronchial disease; Pleural effusion

INTRODUCTION

Actinomycosis is an infectious disease due to an anaerobic gram-positive, non-spore-forming bacteria of the genus Actinomyces, belonging to the resident flora of the oropharynx, gastrointestinal tract, and woman genitalia [1]. It mainly involves cervicofacial and abdominopelvic regions. Thoracic actinomycosis is rare and may affect the respiratory tract and the pleura, sometimes extending to the chest wall. In the lungs, actinomycosis is generally due to Actinomyces israelii or Actinomyces meyeri.

It is often misdiagnosed as primary or metastatic lung cancer or as other lung infections such as tuberculosis [2,3]. Imaging features of pulmonary actinomycosis are nonspecific, and frequently accompany a parenchymal consolidation, lung atelectasis, mass, or rarely endobronchial obstructing lesion such as broncholiths and foreign bodies [4]. Cultures are usually negative and the correct diagnosis mostly relies on a histopathologic examination. It is rare for a pulmonary actinomycosis to be diagnosed with a pleural effusion simultaneously, and we could not find any reference or literature review about an endobronchial actinomycosis presenting with a complicated parapneumonic effusion. In this case, we focus on describing an unusual case of primary endobronchial actinomycosis associated with a complicated parapneumonic effusion.

CASE REPORT

A 57-year-old male was admitted to the hospital for evaluation of a right-sided chest wall pain and a fever. He had been taking oral third-generation cephalosporin at the local clinic for cough, sputum production and a febrile sense. He was transferred to Hallym University Kangdong Sacred Heart Hospital because a pleuritic chest wall pain newly occurred. Over 2 months preceding his admission, the patient was diagnosed as pneumonia at other local clinic and his symptoms improved after 2 weeks of treatment with antibiotics. Although he had a 20 pack-year history of smoking, he had not been smoking for the last 4 years. He did not have any history of cerebrovascular events, swallowing difficulties, diabetes mellitus, alcoholism, or dental problems. He was taking oral entecavir once a day due to a hepatitis B viral infection. He had no history of foreign body aspiration.

A chest radiograph showed a pulmonary consolidation of right middle lobe and blunting of the ipsilateral costophrenic angle (Fig.,...

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A thoracentesis and pleural fluid analysis was done, and neutrophil-dominant exudates were documented (pH 7.045, white blood cell 116,000/mm³, red blood cell 320/mm³, neutrophil 85%, lymphocyte 5%, macrophage 10%, glucose 79 mg/dL, protein 4.9 g/dL, lactate dehydrogenase 1,771 IU/L). He was diagnosed as a presumptive pneumonia with a complicated parapneumonic effusion. We treated the patient with intravenous piperacillin/tazobactam plus levofloxacin due to the history of taking oral third-generation cephalosporin for previous 2 weeks. A chest computed tomography (CT) scan revealed an obstruction of the medial segment of right middle lobe and a moderate amount of multiloculated pleural effusions (Fig. 2). The CT scan showed no evidence of previous tuberculosis infection such as a calcified lymph node, or bronchiectatic change. Tube thoracostomy was indicated because of the low pH and multiloculated nature of the effusion, so a small bore catheter was inserted in the posterior pleural space of right hemithorax. A fiberoptic bronchoscopy revealed an irregularly-surfaced mass which was obstructing the medial segmental bronchus of right middle lobe (Fig. 3A). Macroscopically bronchogenic carcinoma was suspected, so bronchoscopic biopsy was done. After small bore catheter insertion, the amount of loculated pleural effusion increased even though 150 cc of pleural fluid was drained (Fig. 1B). Intrapleural urokinase 100,000 IU was inserted through the catheter. Chest radiograph improved after drainage of 600 mL of pleural fluid. Gram stain, culture, AFB stain results from the pleural fluid and bronchial washing fluid were all negative. Urinary streptococcal antigen test result showed negative and polymerase chain reaction (PCR) assay results for atypical pathogen.
were also negative. On the other hand, bronchoscopic biopsy specimen showed sulfur granules on hematoxylin and eosin stain and Grocott’s methenamine silver stain, and was finally proven to be an endobronchial actinomycosis (Fig. 4). After 5 days of drainage with a small bore catheter, the catheter was removed because the patient was clinically better and the amount of the drained fluid decreased gradually. Ten days after admission, he was discharged with a prescription of oral amoxicillin/clavulanic acid 1,500/375 mg daily. At 3 months follow-up, he had no respiratory symptoms, and his chest radiograph normalized (Fig. 1C). A follow-up bronchoscopy demonstrated a patent medial segmental bronchus of right middle lobe with a small fibrotic tissue (Fig. 3B).
DISCUSSION

Actinomyces organisms are filamentous gram-positive microaerophilic bacteria that are normal residents of the oropharynx and gastrointestinal tract. The cervicofacial area is most commonly affected, 50% to 60% of cases [2]. Thoracic actinomycosis is the next most frequent infection, 15% to 20% of cases. The other infection sites include abdominopelvic (20%), central nervous system (2%) and very rarely, cutaneous, ophthalmic, cardiac, genitourinary tract and disseminated disease [2,3]. Cough (77.7%), hemoptysis (64.9%), and sputum production (61.7%) are common symptoms [5]. Predisposing factors for pulmonary actinomycosis are poor dental hygiene, dental problems and interventions, oral trauma and infections, alcoholism, and chronic diseases such as diabetes mellitus, neurologic and psychiatric disease, malnutrition, drug abuse, and immunosuppressions [2,3,6]. With these factors, secretions containing Actinomyces spp. can easily be aspirated into the respiratory tract.

In a study of the CT findings for thoracic actinomycosis by Cheon et al. [7], CT findings of pulmonary actinomycosis usually showed chronic segmental air-space consolidation containing low attenuation areas with peripheral enhancement or adjacent pleural thickening. Kim et al. [5] reported the retrospectively studied data of 94 subjects in Korea. The most common chest CT finding was consolidation (74.5%), followed by hilar lymph node enlargement (29.8%), atelectasis (28.7%), cavitation (23.4%), ground-glass opacity (14.9%), and pleural effusion (9.6%). According to Kim et al. [8], who presented nine cases of endobronchial actinomycosis associated with broncholithiasis, CT findings of pulmonary actinomycosis presented broncholiths associated with postobstructive pneumatic consolidation, bronchiectasis, enlarged hilar lymph nodes, and adjacent pleural thickening or minimal pleural effusion. Invasion of the chest wall may occur and mimic malignancy. Thoracic actinomycosis is initially often misdiagnosed as malignancy, lung abscess or tuberculosis [3,6].

Definitive diagnosis is usually made with a bronchoscopic biopsy and at surgery or segmental resection [2,3,8]. The hallmark of actinomycosis is the formation of sulfur granules in the biopsy specimen. So pathologists play a key role in diagnosis. In many cases, six months was the average duration of illness before a diagnosis became conclusive [3]. The treatment of choice for actinomycosis is penicillin. Some clinicians favor aminopenicillin plus clavulanic acid [6]. In general, optimal treatment course of 3 to 12 months is recommended for pulmonary actinomycosis. Recently, shorter courses of antibiotics have been suggested. According to the previous reports by several clinicians, relatively short courses of antibiotic therapy were successful in treating actinomycosis [2,9]. Maki et al. [10] used oral antibiotics for 40 days after removing a foreign body with a flexible bronchoscope, and the patient remained well on follow-up for 14 months.

In our case, the patient had no obvious predisposing factors of actinomycosis, no history of foreign body aspiration, and no evidence of immunodeficiency. He presented to the hospital with a chest wall pain. Chest CT findings and a pleural fluid analysis demonstrated a parenchymal consolidation of right middle lobe and a complicated parapneumonic effusion. Small bore catheter drainage was conducted and nevertheless, the loculated pleural effusion increased. Intrapleural thrombolysis was done with urokinase. Culture results of pleural fluid and bronchial washing fluid showed no growth. Urinary streptococcal antigen test and sputum PCR assay for atypical pneumonia pathogen were also negative.

As for the cause of the complicated pleural effusion, concomitant parenchymal form of right middle lobe actinomycosis or secondary obstructive pneumonia caused by other microorganism should be considered. Although it is not clear which one is associated with the parapneumonic effusion, we assume that endobronchial and parenchymal actinomycosis is a better explanation for the parapneumonic effusion since consolidations and effusions were normalized after actinomycosis-directed treatment.

Usually, pulmonary actinomycosis often arises in the setting of aspiration but in this case, no broncholith or foreign body was detected in bronchoscopic findings. Other than aspiration of oropharyngeal or gastrointestinal secretions, distant hematogenous dissemination of Actinomyces spp. from local sites can occur in apparently healthy individuals [11]. The patient denied any history of aspiration, or any condition that could possibly result in loss of mucosal integrity. Infection of Actinomyces spp. in this case could have been aroused by either of the two mechanisms, but it is unclear at this point in time.

As far as we reviewed the literature, there were a few cases of pulmonary actinomycosis with a mild or minimal pleural effusion [5,6], but no reported case of a primary endobronchial actinomycosis associated with a complicated pleural effusion. With this case, we can suggest when an endobronchial lesion with a parapneumonic effusion is observed, although very rare, clinicians should include endobronchial actinomycosis in the differential diagnosis.
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Proving the diagnosis with a bronchoscopic biopsy is mandatory.

REFERENCES