Endobronchial aspergillosis and actinomycosis associated with broncholithiasis

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Abstract

Endobronchial aspergillosis or actinomycosis associated with broncholithiasis is extremely rare. Here, we describe two cases of endobronchial aspergillosis and actinomycosis associated with broncholithiasis. The patients underwent pulmonary resection due to massive hemoptysis. These cases reveal that a bronchiolith can potentially induce endobronchial fungal or bacterial infection, even in immunocompetent patients.

Keywords: Broncholithiasis; Aspergillus; Actinomyces

1. Introduction

Broncholithiasis is defined as a condition in which a calcified mass is present within the bronchial lumen. The most common cause of broncholithiasis is erosion and extrusion due to a calcified adjacent lymph node following infectious etiology, such as tuberculosis or histoplasmosis [1]. Endobronchial aspergillosis or actinomycosis associated with broncholithiasis is extremely rare.

2. Case reports

2.1. Patient 1

A 67-year-old female presented with a 3-month history of hemoptysis. She had a history of tuberculosis 40 years previously. She was not immunocompromised and had no history of smoking. A CT scan revealed a calcified nodule in the mediobasal segment of the right lower lobe with atelectasis (Fig. 1). Paratracheal, subcarinal and hilar lymph nodes were calcified, suggesting previous tuberculous infection. Endoscopically, bleeding from the mediobasal segment of the right lower lobe was observed, although the broncholith was invisible. Bronchoalveolar lavage analysis did not reveal Aspergillus. A thoracotomy was performed and the right lower lobe was resected. Gross examination of the resected specimen revealed segmental consolidation associated with a proximal obstruction caused by a calcified nodule. Microscopically, Aspergillus aggregation was observed around the broncholith, but invasive findings were not noted. The distal consolidation seen on CT was composed of obstructive pneumonia with lymphoplasmacytic infiltrations. Antifungal agents were not administered before or after surgery. The postoperative course was uneventful, and the patient has continued to do well for more than 1 year without any further symptoms.

2.2. Patient 2

A 57-year-old female presented with fever, cough and hemoptysis. She had a past history of tuberculosis 50 years previously. She had suffered recurrent pneumonia over the last several years. She was not immunocompromised and had no history of smoking. A CT scan revealed a calcified nodule in the ventrobasal segment of the right lower lobe (Fig. 2). Bronchiectasis and peripheral consolidation were also noted. Subcarinal and hilar lymph nodes were calcified. Bronchoscopic examination did not show an endobronchial calcified nodule. Although bronchoalveolar lavage analysis did not reveal Actinomyces, the tissue around the broncholith obtained via bronchoscopic biopsy was found to include Actinomyces organisms by microscopy. A thoracotomy was carried out and a right lower lobectomy was performed. Penicillin was administered before and after surgery. Gross examination of the resected specimen revealed segmental consolidation and bronchiectatic changes associated with the obstructive calcified mass. Microscopically, purulent exudates accompanied by Actinomyces colonies were found within the bronchial lumen. The distal pneumonic consolidation seen on
CT was histologically composed of organized inflammatory tissue. The postoperative course was uneventful, and the patient has continued to do well for more than 1 year without any symptoms.

3. Discussion

Endobronchial aspergillosis associated with broncholithiasis is extremely rare. Regarding our Patient 1, *Aspergillus* hyphae were microscopically limited to the bronchial lumen, and no invasion of the airway basement membrane or necrotic tissue was present. We speculate that a pre-existing broncholith was secondarily infected by *Aspergillus*, and the subsequent inflammation proceeded to the peripheral bronchus and obstructed the airway, associated with distal obstructive pneumonia.

Compared with parenchymal actinomycosis, predominantly endobronchial actinomycosis is rare. Endobronchial actinomycosis has been reported to be associated with broncholithiasis or aspired foreign bodies [2,3]. Endobronchial actinomycosis associated with broncholithiasis is speculated to be caused by secondary colonization of a pre-existing broncholith by aspired *Actinomyces* organisms, and usually manifests as a proximal obstructive calcified nodule and distal postobstructive pneumonia on CT, as was observed in our Patient 2 [2].

Calcifications are sometimes observed in the fungus ball and primary endobronchial actinomycosis [1]. These lesions mimic broncholithiasis and it is sometimes difficult to distinguish a secondary infection associated with broncholithiasis from de novo calcification in these infectious lesions. In the present cases, calcified hilar and mediastinal lymph nodes were revealed by CT, which indicate previous tuberculosis infection. Therefore, it is more likely that a pre-existing broncholith was secondarily infected by *Aspergillus* or *Actinomyces* organisms.

The modalities for treating broncholithiasis include bronchoscopic removal and surgery. The decision regarding which option should be selected is sometimes controversial. Although it was reported that bronchoscopic extraction of partly eroded or free broncholiths was safe and effective [4], life-threatening complications, including massive bleeding, may occur. In general, bronchoscopic removal is considered in those cases where the broncholiths are loose, mobile and without complications, such as bronchiectasis, massive hemoptysis, bronchoesophageal fistula and so on [5]. When broncholithiasis presents with symptoms such as hemoptysis or recurrent pneumonia, surgery should be considered [5]. It has been reported that surgical resection for broncholithiasis can be carried out with low mortality and morbidity [6].

We conclude that endobronchial aspergillosis or actinomycosis may develop in association with broncholithiasis. Therefore, bronchoscopic removal should be considered for asymptomatic cases if the broncholith is movable. A rigid or invisible broncholith should be followed, even when it is asymptomatic, and surgical resection is recommended when symptoms appear.

References


