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Case Report Open Access

Primary Pulmonary Botryomycosis

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Abstract

Botryomycosis is a bacterial infection that presents with eosinophilic granules and colonies of either gram positive or gram negative organisms, although Staphylococcus aureus is most commonly identified. It has been confused with lung cancer or actinomycosis due to similar presentation and symptoms. Treatment of this infection consists of a combination of antibiotics and surgery. We present a case of pulmonary botryomycosis.

Keywords: Lung infection; Botryomycosis; Thoracotomy

Introduction

Botryomycosis is a chronic bacterial infection that is recognized by inflammatory granules that present due to multiple bacterial species with the most prevalent being *Staphylococcus aureus* [1]. It is more common for this infection to be cutaneous than visceral, however various organs can be involved. Symptoms of the infection may include cough, chest wall pain, hemoptysis, unexplained weight loss, weakness, and dyspnea [2,3]. Since malignancy may present with similar symptoms, pulmonary botryomycosis may initially be mistaken for lung cancer [4-6]. We present a case of botryomycosis in a patient with a preoperative diagnosis of aspergilloma.

Case Report

A 69-year-old Jordanian female with a remote 17 pack-year smoking history and a history of allergic bronchopulmonary aspergillosis requiring a previous left lower lobe segmentectomy presented with intermittent hemoptysis. The hemoptysis occurred three times annually for the past nine years and lasted for a few days with each episode. She had received itraconazole daily for nine years as well as prednisolone during the acute episodes of hemoptysis. Immediately prior to her presentation to thoracic surgery clinic, the hemoptysis was increasing in frequency and severity (7 episodes in 2 weeks producing up to 2 cups of blood per episode). She had no pulmonary symptoms other than orthopnea. Physical examination was notable for rhonchi bilaterally. She was then admitted to our hospital. Flexible bronchoscopy was significant for lack of active bleeding but signs of recent hemorrhage from the right upper lobe.

Preoperative serologic examination revealed a white blood cell count of 12.9 K/uL (neutrophils 10.38 K/uL) and immunoglobulin E 384 IU/mL.

Chest computed tomography (CT) revealed a 2.7 x 1.8 cm lesion in the posterior segment of the right upper lobe (RUL). This had the radiographic appearance of an aspergilloma (Figure 1). There was also a 1 cm nodule in the lateral RUL and evidence of bronchiectasis.

Pulmonary function testing showed a forced expiratory volume in 1 second (FEV1) of 1.78 liters which was 96% of predicted. Diffusion capacity for carbon monoxide (DLCO) was 83% of predicted at baseline and increased to 118% when adjusted for alveolar volume.

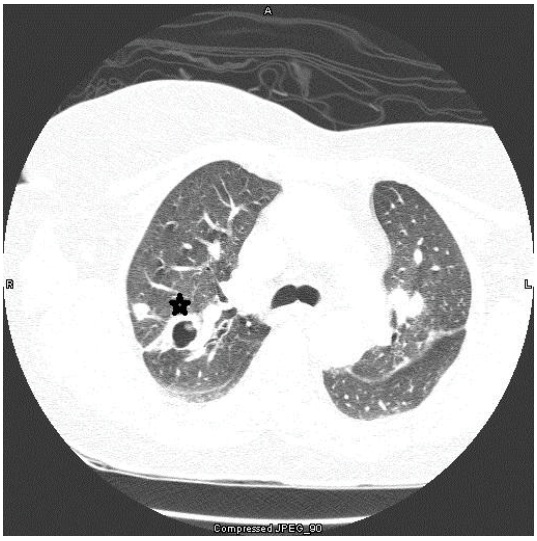

Figure 1: Computed tomography demonstrating right upper lobe cystic cavity (starred) and adjacent pulmonary nodule.

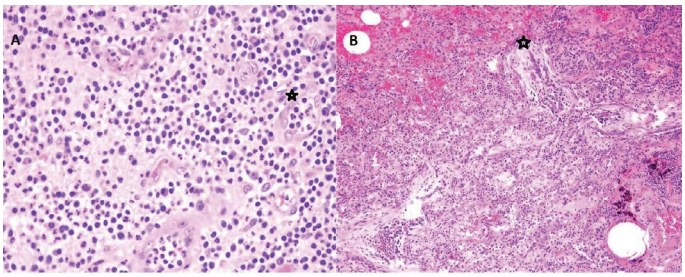

Figure 2: Panel A, Dense lymphoplasmacytic infiltrate with granulation tissue formation and focal neutrophilic infiltrate and abscess formation (starred, 400x). Panel B, Adjacent lung demonstrates mixed acute and chronic inflammatory infiltrate in association with fibroblastic proliferation in alveolar spaces (starred), consistent with organizing pneumonia (100x).

Given the patient's extensive history of hemoptysis and in anticipation of severe adhesions typically found with aspergillosis, a right posterolateral thoracotomy, right upper lobectomy, and thoracic lymphadenectomy were performed. Upon opening the specimen, gross pus was found. Frozen section showed no evidence of malignancy. Final pathology revealed abscess formations, granulomatous tissue, and lymphoplasmacytic infiltrate (Figure 2). Gram stain confirmed the presence of gram-positive cocci, and culture grew *Staphylococcus aureus*. Anaerobic culture was notable for few Actinomyces israelii. Expert consensus established the diagnosis of botryomycosis in the setting of bronchiectasis. Twenty resected mediastinal, hilar, and segmental lymph nodes only showed evidence of anthracosis. She did well postoperatively and was discharged from the hospital in routine fashion.

The patient remains stable with no further episodes of hemoptysis almost three years after surgery.

Comment

Botryomycosis is a chronic bacterial infection characterized by inflammatory granules. It more commonly affects the skin compared to visceral organs. However, when this rare infection is visceral, it most often appears in the lungs, as in this patient [2]. It may also be preceded by local trauma in the case of cutaneous involvement [7]. The most

prevalent bacterial species in patients diagnosed with botryomycosis is *Staphylococcus aureus*, however Escherichia coli and Pseudomonas aueruginosa have been reported as well [8,9]. Medical treatment relies on appropriate antibiotic coverage based on cultures and sensitivities. Immunosuppressed patients are more vulnerable to infection than those who are not. Botryomycosis is often diagnosed in patients with other illnesses such as human immunodeficiency virus, cystic fibrosis, and diabetes [1,3,4]. Our patient, however, did not have any significant comorbid conditions. There has also been a case report of pulmonary botryomycosis secondary to persistent vegetable matter in the tracheobronchial tree after a previous aspiration [10]. Symptoms of botryomycosis include dyspnea, unexplained weight loss, hemoptysis, chest wall pain, and chronic cough. These symptoms are also seen with malignancy. Thus, lung cancer is usually considered in the differential diagnosis.

The Splendore-Hoeppli phenomenon is the deposition of amorphous, eosinophilic, hyaline material around pathogenic organisms. This phenomenon is seen histologically with botryomycosis and is composed of an antigen-antibody complex along with tissue debris and fibrin [3,11]. For patients with cutaneous bortryomycosis, weeks to months of antimicrobial treatment may cure the infection. However, if the infection is visceral, such as in this case, a combination of surgery and antimicrobial treatment may be needed in order to alleviate the symptoms [2].

Botryomycosis is a rare bacterial infection that may mimic several different illnesses, including other infections and lung cancer, due to similar symptoms and its granulomatous appearance. Many patients in previously reported cases were misdiagnosed until final pathological examination and tissue culture. Greater awareness of this rare process is needed to ensure timely and effective treatment especially in the setting of visceral disease as antibiotics alone may not be effective.

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