Case Report

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TANAFFOS

Pulmonary Botryomycosis Mimicking Bronchogenic Carcinoma of the Lung

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Botryomycosis is a relatively rare disease found only in case reports. Most observed cases have been of cutaneous or visceral type. Given the prolonged duration and nature of symptoms, pulmonary botryomycosis may be mistaken for malignancy. We report the first case of pulmonary botryomycosis in Iran initially mimicking bronchogenic carcinoma.

Key words: Botryomycosis, Bronchogenic Carcinoma, Lung

INTRODUCTION

Botryomycosis is a chronic, suppurative infection due to a granulomatous response to bacterial infection .It usually presents as a cutaneous disease and visceral involvement has a lower prevalence (1). Botryomycosis is a rare disease, and our information about it comes from case reports in children and adults. It occurs more commonly among immunocompromised patients (2,3). Risk factors associated with botryomycosis include: alcoholism, diabetes mellitus, HIV infection, cystic fibrosis, chronic granulomatous disease, trauma and surgery (3-5). The most common microorganism isolated from botryomycosis lesions is *Staphylococcus aureus*. Other pathogens associated with this condition include *Pseudomonas aeruginosa*, *Escherichia coli*, Serratia, Proteus and coagulase-negative Staphylococci (2,6). Visceral botryomycosis most commonly occurs in the lungs, although involvement of other organs such as liver, spleen, kidneys, and brain has been described as well (7). Systemic symptoms such as fever, fatigue, or weight loss may or may not be present. Symptoms associated with pulmonary botryomycosis include chronic cough, dyspnea, hemoptysis, and chest wall pain (6,7). Clinical examination may be normal or demonstrate diminished breath sounds or rhonchi over the consolidated lung. Given the prolonged duration and nature of symptoms, pulmonary botryomycosis may be mistaken for malignancy (8). Histopathologically, botryomycosis is characterized by a central focus of necrosis surrounded by a chronic inflammatory reaction containing histiocytes, epithelioid cells, multi-nucleated giant cells, and fibrosis (9). We report a rare case of botryomycosis with symptoms and signs of bronchogenic carcinoma of the lung.

CASE SUMMARIES

A 56 year-old man was admitted with a massive hemoptysis .He had productive cough since 1 month ago, and productive cough with blood streaked sputum since 1 week before admission. During the past 6 months he had

fever, dry mouth, and significant weight loss (33kg). He was a heavy smoker (60 packs/year) suffering from coronary artery disease (CAD).

His medications were nitroglycerin, digoxin and triamterene-H. In the emergency department he was frightened of massive hemoptysis. On physical examination, chest auscultation revealed decreased pulmonary sounds in lower field of the right hemithorax and fine crackles over the base of the left hemithorax. After stabilization, chest radiography and spiral CT of thorax with intravenous contrast revealed a right hilar mass-like lesion and ground-glass opacity with air bronchogram in the apical segment of right lower lobe with extension to the posterior segment of right upper lobe. There were bilateral paratracheal and subcarinal adenopathies (Figure 1).

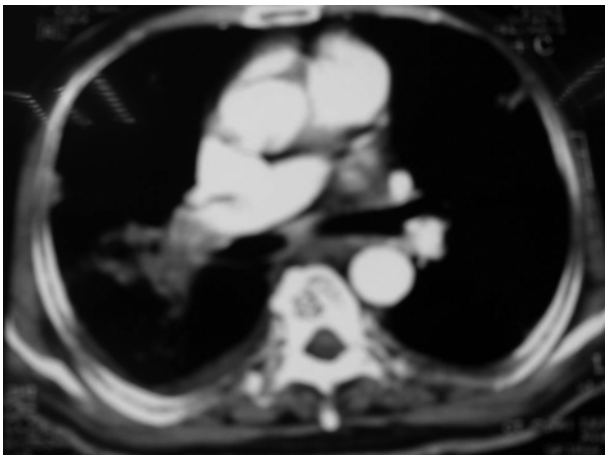

Figure 1. Chest CT-scan of patient.

Bronchoscopy was performed to rule out bronchogenic carcinoma; which revealed severe mucosal inflammation, congestion, bleeding and significant irregularity at the proximal of right main bronchus. The patient was given empiric antibiotic treatment with ceftriaxone and azithromycin for two weeks. In the follow-up CT scan no change from previous findings was noted.

Bronchogenic carcinoma was suspected and the patient underwent thoracotomy and lobar wedge resection of the involved lung. Microscopic examination revealed alveoli filled with pigment-laden histiocytes. The parenchyma was infiltrated by mixed inflammatory cells, fibrin exudates and vascular congestion. There were a few foci of abscess formation having central amorphous basophilic materials surrounded by pus discharges that were walled off by garanulomatous tissue and mild fibrosis. Some bronchioles were filled with fibrinoleukocytic exudates. The pleura was covered by fibrin exudates and showed marked vascular congestion, hyperemia and hemorrhage. Special stains were negative for acid-fast bacilli and fungi and granules typical for actinomycosis were not identified (Figure 2). There was no evidence of malignancy. The pathological diagnosis was pulmonary botryomycosis.

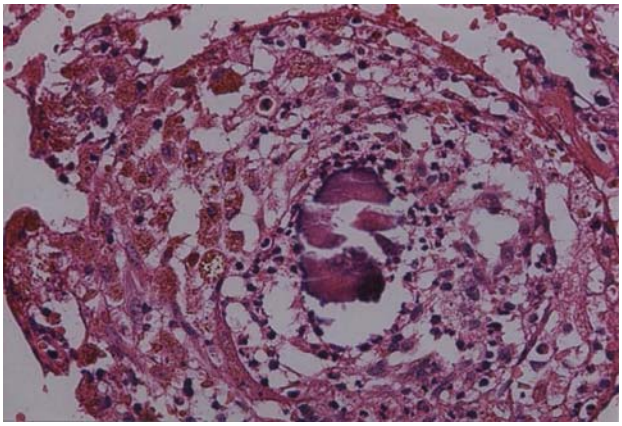

Figure 2 . The pathological diagnosis was pulmonary botryomycosis.

He was discharged 20 days after the operation in good condition. At six months follow up, the patient was in good condition and CT –scan of the chest was completely clear (Figure 3).

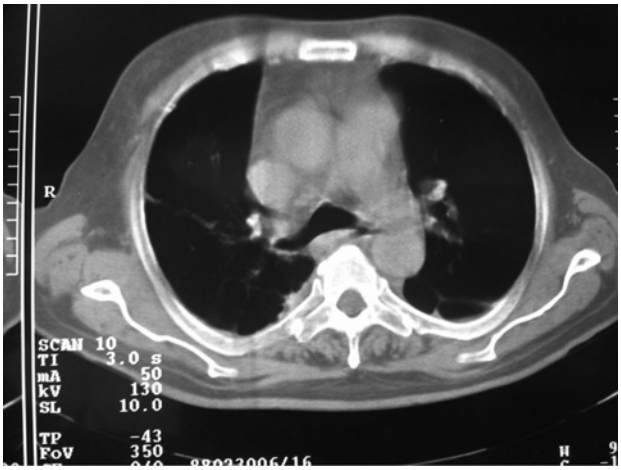

Figure 3. Chest CT-scan of patient at 6 months follow up.

DISCUSSION

Botryomycosis is a granulomatous inflammatory reaction to bacterial pathogens; it may present with cutaneous or, less commonly, visceral involvement (1). The terms used to describe botryomycosis include bacterial pseudomycosis, staphylococcal actinophytosis, granular bacteriosis, and actinobacillosis. Botryomycosis is a relatively rare disease that occurs more commonly among immunocompromised patients (2-4). However, our patient was not immunocompromised.

Botryomycosis usually occurs in patients with alcoholism, diabetes mellitus, HIV infection, cystic fibrosis, chronic granulomatous disease, trauma and surgery (3-5). None of the above-mentioned conditions were present in our patient. The most common microorganism isolated from botryomycosis lesions is *Staphylococcus aureus* (2,6). Botryomycosis may present as a cutaneous or visceral disease (8). Our patient suffered from visceral disease. Visceral botryomycosis most commonly occurs in the lungs (8). Systemic symptoms such as fever, fatigue, or weight loss may or may not be present. Symptoms associated with pulmonary botryomycosis include chronic cough, dyspnea, hemoptysis, and chest wall pain (6,8). Given the prolonged duration and nature of symptoms, pulmonary botryomycosis may be mistaken for malignancy (8). Our patient presented with fever, weight loss, chronic cough and hemoptysis. Botryomycosis can be diagnosed in one or more ways. Imaging may be useful to evaluate the size and extent of involvement (10). Pulmonary lesions may be present as a consolidation or mass lesion, while other forms of visceral botryomycosis usually present as a mass lesion. The histopathological pattern required for definite diagnosis of botryomycosis is characterized by a central focus of necrosis surrounded by a chronic inflammatory reaction containing histiocytes, epithelioid cells, multi-nucleated giant cells, and fibrosis (10). This histological appearance is commonly referred to as the Splendore-Hoeppli phenomenon, although it may not always be present. The diagnosis in our patient was established based on pathological findings from thoracotomy and lobar wedge resection of involved RUP. In general, patients should receive antibiotic therapy until signs and symptoms of infection resolve. A few weeks of therapy may be sufficient for those with superficial infection but patients with deep infections and/or those with underlying immunodeficiency may require months of therapy. In addition, prolonged antibiotic therapy may be necessary in cases with partial or no debridement. In such situations, antibiotic access to the microorganisms sequestered in the granules may be poor or limited (10). Thus, treatment of visceral disease requires a combination of surgical and antimicrobial therapy. Resection of the mass often occurs prior to diagnosis given concern for malignancy in most cases of visceral disease.

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