Case Report

Thoracic actinomycosis presented with tracheoesophageal fistula and fatal pulmonary infection

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ABSTRACT

We report a case of a 60-year-old Saudi woman who developed tracheoesophageal fistula and fatal pulmonary infection secondary to thoracic actinomycosis. The cause, clinical presentation, radiological features and treatment of thoracic actinomycosis are discussed.

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Actinomycosis is an uncommon infection caused by *Actinomyces israeli*, characterized by clinical and radiological findings that can resemble other inflammatory and neoplastic lesions. Thoracic involvement is rare and accounts for 15% of all cases. Moreover, tracheoesophageal fistula is a rare manifestation of thoracic actinomycosis with only 2 reported cases in the literature.

Case Report. A 60-year-old Saudi female presented with a history of dysphagia for liquid and solid food for 6 months and severe cough precipitated by drinking for 3 months. In addition, the patient also reported expectoration of large amounts of greenish sputum, dyspnea, weight loss and low-grade fever over a period of one month before seeking medical advice. She had no hemoptysis or night sweats. She was being treated medically for hypertension and diabetes mellitus. She has no history of tuberculosis (TB) exposure, and she was non-smoker. There was no significant family history of similar illness, TB or cancer. Physical examination was notable only for coarse breath sounds in the lower lung fields bilaterally and poor oral hygiene. Total leukocyte count was 21.9 x 10^5/dl, neutrophil count 88.5%, hemoglobin 11.5 g/dl, erythrocyte sedimentation rate 48 mm/hour, other investigations including platelet count, prothrombin time and partial thromboplastin time, urea, creatinine, electrolytes, and liver function test were normal. Serology for human immunodeficiency virus was negative. Plain chest films revealed diffuse bilateral reticular infiltrates. Upper gastrointestinal endoscopy was carried out and revealed a large deep esophageal ulcer at 20 cm from incisors, about 10 cm in length, broncho-esophageal fistula was noted with air bubbles and thick greenish sputum coming from the fistula (**Figure 1**). Multiple biopsies were taken which showed chronic inflammatory cells. A computerized tomography (CT) examination of the thorax with intravenous contrast carried out and revealed a hyperdense mass in the posterior mediastinum, containing air and central fluid density (**Figure 2**). Bronchoscopy revealed a large amount of greenish secretion, and a mucosal defect in the left main bronchus. Bronchial wash and biopsy were taken which showed bacterial colonial

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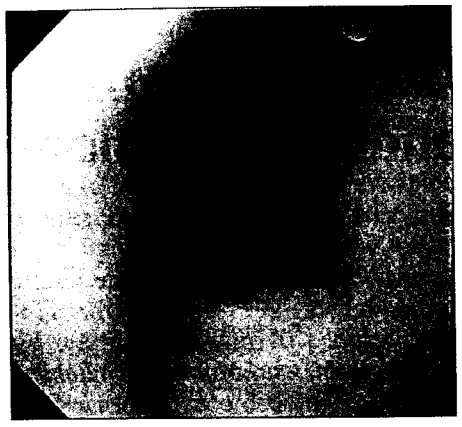

Figure 1 - Upper gastrointestinal endoscopy showing large esophageal ulcer with yellow base and the opening of the broncho-esophageal fistula.

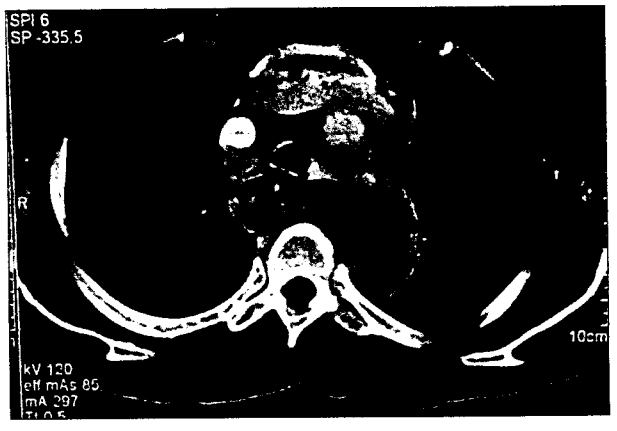

Figure 2 - A computerized tomography scan of the chest showing a hyperdense mass in the posterior mediastinum with air loculi and central fluid density.

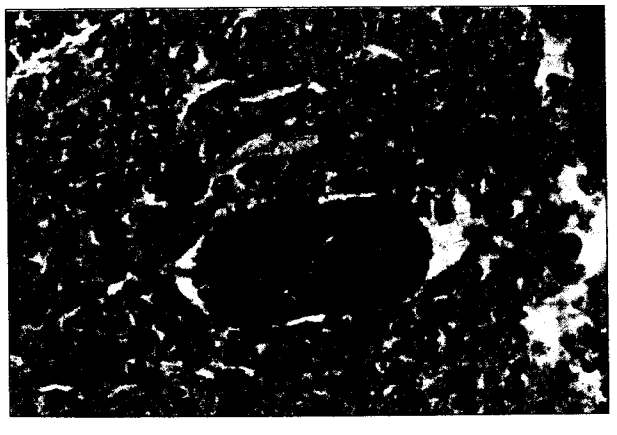

Figure 3 - Bronchial biopsy showing bacterial colonial granules with branching periphery consistent with actinomycosis.

granules with branching periphery most consistent with actinomycosis (**Figure 3**). The patient was started on intravenous penicillin. A self-expanded metallic stent was inserted in the esophagus to close the area of the fistula, but unfortunately the patient expired due to severe chest infection.

Discussion. Actinomycosis is a chronic, suppurative granulomatous infection, which is usually caused by *Actinomyces israeli*. The organism is a gram positive, obligate or facultative anaerobic, filamentous and branching bacterium of slow growth and is very difficult to culture.[1] It is a member of the normal flora of the oral cavity, intestinal tract and vagina.[2] They may form an abscess and sinus tract and cause purulent discharge with yellowish sulfur granules. It classically involves cervicofacial (55%), abdominopelvic (20%), thoracic (15%) and mixed organs (10%) including skin, brain, pericardium and extremities.[2,3]

Thoracic actinomycosis is uncommon which may present as chronic rehabilitating illness with clinical and radiographic manifestations simulating lung tumor or chronic suppuration.[3] It is usually present as bronchopulmonary disease and may extend to the pleura and the chest wall. It is believed that the organism enters the thorax by way of airway aspiration from the oral cavity, by hematogenous spread from distant foci, or by direct extension from cervicofacial or abdominopelvic disease.[2,3] Poor oral hygiene and immunocompromise are predisposing factors. The bacteria are always present synergistically with other opportunistic bacteria.[4,5] Clinically, the symptoms usually include fever, cough, hemoptysis, weight loss, chest pain, thoracic soft tissue swelling, and cutaneous fistula.[3-5] The disease may present with unusual manifestations, which include empyema thoracis, superior vena cava syndrome, pericardial effusion and tracheoesophageal fistula.[5] Diagnosis of thoracic actinomycosis is very difficult due to its clinical and radiological features usually mimic bronchogenic carcinoma or chronic suppurative lung disease such as TB. Radiographic examinations usually demonstrate pulmonary infiltration, cavitary lesion, and tumor-like mass.[2,3] Also, they may demonstrate bone changes in the ribs and pleural involvement. However, these findings are not specific.[2-4] Culture of the organism is difficult in the thoracic forms, and is obtained in less than 50% of the cases.[6] Sputum culture and bronchoalveolar lavage are inadequate for the diagnosis of pulmonary actinomycosis because Actinomyces may form part of the normal flora.[5] The reported accuracy of the biopsy methods, transbronchial and needle aspiration are low owing to inadequate aspiration. Therefore, it is very difficult to diagnose actinomycosis without histologic and microscopic examination of tissues taken via operation.[7]

Diagnosis of actinomycosis is achieved when histologic examination reveals sulfur granules containing filamentous organisms characteristic of actinomyces species.

Current literature recommended that using penicillin treatment for 2 months is adequate.[2,5,6] Also, it is necessary to treat any oropharyngeal or dental abscesses to avoid recurrences. Suspicion of malignancy is the most common indication for exploratory thoracotomy. Other indications for surgery may be drainage of an abscess or pleural empyema decortication, radical excision of sinus tracts, control of massive hemorrhage and closure of tracheoesophageal fistula.[3,6-9]

Literature review shows 3 reported cases of thoracic actinomycosis with tracheoesophageal fistula.[6,7] Dysphagia and cough when swallowing liquids were the presenting symptoms. In the first case, the cause of the fistula contributed to the necrotizing inflammatory process, while in the remaining 2 cases, was due to erosion of peribronchial lymph nodes into the bronchus and esophagus causing the fistula and broncholithiasis.[6,7,11] Treatment options for tracheoesophageal fistula include endoscopic or surgical closure of the fistula. Endoscopic closure of esophagobronchial fistulas of any etiology using a variety of techniques such as electrocautery, fibrin glue and stent have been reported. Tissue interposition during surgical closure prevents recurrence.[6]

Fatality is rare in thoracic actinomycosis, and usually results from delay in diagnosis and the development of complications. Severe hemoptysis, severe chest infection and dissemination are reported causes of death in this disease.[2,3,5,10]

In conclusion, thoracic actinomycosis is a rare chronic suppurative infection that is usually underestimated or misdiagnosed. The clinical and radiological features resemble those of bronchogenic carcinoma or chronic suppurative lung disease. It can present with tracheoesophageal fistula and fatal pulmonary infection, which are rare manifestations of this disease.

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