A RARE CASE OF THORACIC ACTINOMYCOSIS

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PRESENTATION OF CASE

Actinomycetes are branching gram-positive anaerobic bacteria belonging to Actinomycetaceae family and are commensals in human oropharynx, gastrointestinal tract and female genitalia. Thoracic or pulmonary actinomycosis is an uncommon bacterial infection. The diagnosis of pulmonary or thoracic actinomycosis is often confounding because of its shared clinical features with malignant lung diseases and chronic suppurative lung diseases. However, chest physicians should be aware of actinomycosis being a differential diagnosis in persistent shadows in lung as early diagnosis leads to good prognosis.[1]

DIFFERENTIAL DIAGNOSIS

Actinomycosis is a granulomatous infection of the cervicofacial, thoracic, abdominopelvic area and skin, brain, pericardium and extremities. The species mostly causing human disease is- A. Israeli. It has a predilection for the middle-aged males.[2] Although, cervicofacial type (lumpy jaw) is the most common human form of disease. No organ or body site is absolutely immune to the infection. The second most common type of presentation is the "pulmonary actinomycosis." Because of its simulating clinical features with malignant lung diseases, lung abscess and tuberculosis, it often poses a diagnostic challenge.[1] An early diagnosis is helpful in avoiding the dreaded and debilitating complications of the disease including unwarranted surgery.[3]

Pathogenesis is unclear. Actinomyces is not virulent and cause human illness when there is polymicrobial infection. Saliva laden with actinomyces is aspirated into bronchus leading to pneumonitis, which is later followed by the chronic features of necrosis, fibrosis and often cavitation.[1] The presence of companion bacteria further cause infection by production of a toxin or enzyme or reduced host defense leading to increase in invasiveness of actinomycetes. The nonresponsiveness to treatment is also due to copathogens.[4]

Subsequent to establishment of infection suppuration or granuloma develops followed by fibrosis. The anatomical boundaries are crossed and surrounding tissues or organs are invaded.

CLINICAL DIAGNOSIS

A 32 years old female presented with low-grade irregular fever. She had cough for 2 years, non-postural with scanty expectoration. She had two bouts of scanty haemoptysis with no loss of weight. She had taken several courses of antibiotics and antitubercular drugs with no improvement in the symptoms.

On clinical examination, multiple cervical glands with absent breath sounds in right infraclavicular and right infrascapular area was seen.

Chest x-ray PA view revealed a large heterogeneous mass in the right upper and mid zone with complete erosion of clavicle and loss of lung volume on the right side.

CECT thorax showed a lobulated mass 60 mm x 50 mm in right clavicular region extending to bilateral sternoclavicular joints along anterior and posterior surface of sternum from manubrium to level of sternocostal junction with central necrosis and lytic destruction of whole of right clavicle, anterior ends of bilateral first to fourth ribs and medial ends of left clavicle, also extending to right apical region and extra pleural space. Multiple necrotic lymph nodes were seen in right supraclavicular, retrosternal, paratracheal, aortopulmonary, subcarinal and anterior mediastinal stations. Left innominate vein was severely compressed. Right lung showed fibronodular opacities and traction bronchiectasis in right upper lobe and pleural thickening along right major fissure. Mild pleural effusion was seen.

A diagnosis of consolidation of right upper lobe due to tuberculosis/pneumonia was made. USG of abdomen and pelvis was normal except for mild splenomegaly and minimal ascites. Pleural fluid analysis showed protein 3.5

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gm/dL, ADA 17.5 U/L with lymphocytic predominance on cytology.

On FNAC of the mass, pus was aspirated. Cytology of the pus showed nonspecific inflammatory lesions, AFB-negative, Gram stain, gram-positive bacilli and culture was negative for aerobic organisms. Bronchoscopy was normal. A repeat FNAC of mass was performed and special emphasis on histopathology revealed sulphur granules against an inflammatory background confirming the diagnosis of thoracic actinomycosis.

DISCUSSION OF MANAGEMENT

Pulmonary actinomycosis is a rare, but important and challenging diagnosis to make. Even when the clinical suspicion is high, the disease is confused always with other chronic suppurative lung diseases or with malignancy. Actinomycetes belong to the family Actinomycetaceae. They are gram positive with a branching characteristic. As commensals, they reside in the oropharynx, gastrointestinal and female genitalia. Hence, cervicofacial and abdominopelvic are the most common and second most common type of actinomycosis infection respectively followed by thoracic actinomycosis accounting for 15% of total burden of the disease.[5] However, it can affect any organ.[1]

It is slightly more common in males than in females and children are rarely affected.[1,2]

The most common predisposing factors for thoracic actinomycosis are a preexisting respiratory illness such as emphysema, bronchiectasis, chronic bronchitis and other conditions in which there is destruction and loss of lung volume along with alcoholism, poor dental hygiene, facial or dental trauma and many chronic diseases such as diabetes mellitus, psychiatric and neurologic diseases, malnutrition, drug abuse and hiatal hernia that increase the risk for aspiration.[1,5,2] Actinomycotic infections are polymicrobial, acting synergistically with anaerobes, particularly Actinobacillus and Bacteroides spp. As the disease simulate many other pathologies, tissue diagnosis is mandatory.

A peripheral and/or lower lobe predominance reflects the role of aspiration in its pathogenesis.[5] Most lesions maybe unilateral.[6] The infection rate is not increased in immunocompromised patients as in other granulomatous diseases.[6]

Thoracic actinomycosis typically runs a slow course with fever, weight loss, cough and expectoration being the predominant symptoms simulating a slowly progressive pneumonia. Spread to the pleural chest wall may result in chest wall pain. The disease may present as a chronic debilitating illness.[2] The clinical and radiological signs mimic lung cancer or tuberculosis.[6] If the disease progresses to the proximal airway and vessels, life-threatening complications such as massive haemoptysis or bronchoesophageal fistula may occur. The course of the disease depends largely on early and adequate antibiotic therapy. The principal treatment of actinomycosis is long-term use of high-dose intravenous pencillin.[3] However, many studies have reported that the short-term treatment is successful and that the traditional, intensive and long-term regimen is not necessary.[3] In the setting of abscess, percutaneous drainage in combination with medical therapy maybe an option. Surgical resection maybe a valid option for patients who do not respond to antibiotics for up to 12 weeks according to a recent study.

According to Kim et al, at least four different forms of thoracic actinomycosis have been described.

Lung parenchymal actinomycosis is probably the most common bronchiectatic form of actinomycosis. This form usually affects isolated and previously damaged lobe or a segment by tuberculosis or secondary bacterial infections.

The other two forms are rare forms of endobronchial actinomycosis associated with broncholithiasis and foreign body.[7]

PATHOLOGICAL DISCUSSION

Imaging Modalities- Chest radiograph though nonspecific is often the initial and starting point of all radiological investigations. Similar chest radiographic findings may be seen in tuberculosis, lung abscess and lung cancer.[7] Radiograph may present any of a wide variety of changes such as diffuse consolidation, fibrosis, cavitation and emphysema and maybe bilateral or unilateral. Many radiological appearances have been described as typical of pulmonary actinomycosis. Bone change is the only characteristic radiological change produced by thoracic actinomycosis. Staub-Oetiker (1921) first documented the feature of several adjacent ribs showing periostitis in the presence of a pulmonary infection without empyema. This feature helped radiologist to arrive at the correct diagnosis.[3]

CECT (contrast-enhanced computerised tomography) is a much more rewarding imaging modality. Based on CECT findings, thoracic actinomycosis can be further classified into parenchymal, bronchiectatic, endobronchial and endotracheal.[8] A centrally located low density in the parenchymal consolidation with a halo of surrounding pleural thickening is the most typical finding in parenchymal pattern of thoracic actinomycosis. The most common radiological feature is nonspecific consolidation resembling pneumonia or lung malignancy. Actinomycosis may spread from the lungs to the pleura, mediastinum and chest wall with little regard for anatomic barriers as the organisms produce proteolytic enzymes. Thus, peripheral pneumonia leads to empyema and then invades the chest wall such as the ribs or vertebrae. Chest wall involvement may also occur due to direct extension from the neck, oesophagus, abdomen or retroperitoneum. The chest wall involvement is rare now due to antibiotics being administered early in the course of the disease.[8]

CT manifestations are also varied with each feature being nonspecific on its own. These include a patchy air space consolidation, a relatively common feature. Pleura and chest wall maybe involved leading to empyema necessitans. Dense consolidation can occur and may have

associated central areas of attenuation. Multifocal nodular appearances, cavitation and associated pleural thickening are some of the other relatively common feature. Pleural effusions is relatively common feature (50%). Associated hilar and/or mediastinal lymphadenopathy maybe seen.[8]

Detection of sulphur granules by Gram staining or histopathological staining is the gold standard and principal method of detection of actinomycetes.[2] The sulphur granules are yellow or white flecks, hard to the touch and about 2 mm in diameter consisting of masses of hyphae cemented by a polysaccharide-protein complex and calcium phosphate. Under the microscope, they appear light brown with a granular centre and radiating bands with oeosinophilic clubs at the periphery. They may sometimes be found in sputum if it is fixed directly in formalin rather than smeared for cytology. The diagnosis is made with FNAC, transbronchial biopsy, ultrasound or CT-guided biopsy.[4]

Histopathology confirms the diagnosis. It reveals an acute inflammation surrounded by fibrosing granulation tissue. However, as the organism is difficult to culture, the importance of its histological recognition in pus, sputum or tissue specimens is not much emphasised.

Culture and Staining Characteristics- Culturing of Actinomycosis is difficult as they are fastidious bacteria, hence, a bacterial proof in a clinical diagnosis of the disease is obtained in <50% of cases. Characteristically, colonies of actinomyces appear as "molar-tooth" or "bread-crumb" colonies in broth media after 3-7 days of incubation. A newer technique that aids species identification is the 'fluorescent conjugated antibody typing.' In a magnifying glass, sulphur granules can be seen as round or oval basophilic masses with oeosinophilic clubs surrounding them in a radiating fashion, which is diagnostic. The average duration of illness before definitive diagnosis is made, was more than 6 months, a consistent figure in most series.[1]

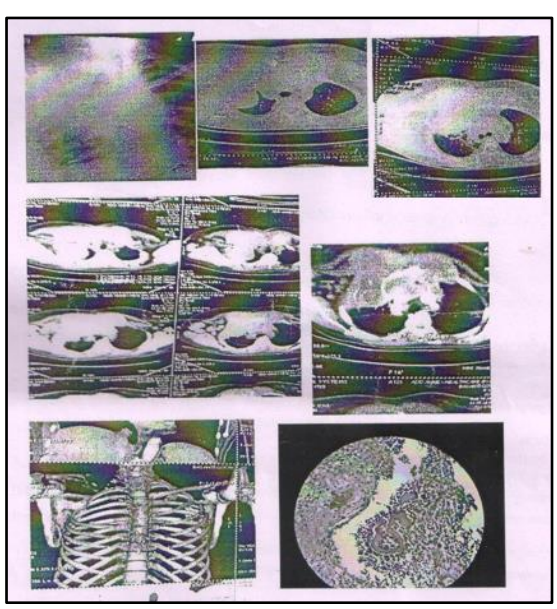

Figure 1. USG of Neck Revealed Bilateral Cervical and Supraclavicular Lymphadenopathy

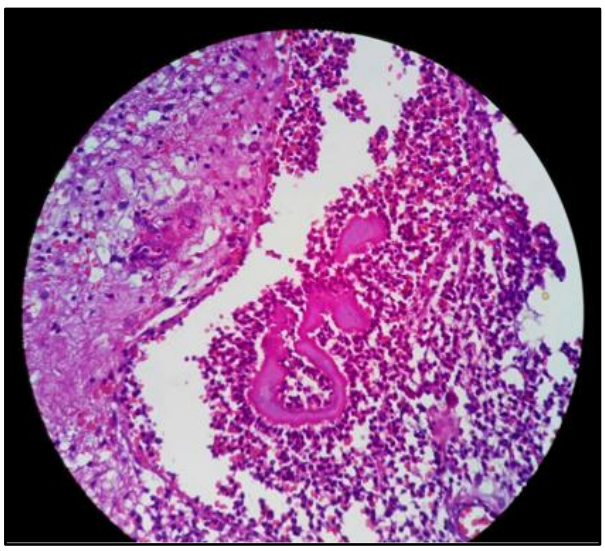

Figure 2. Histopathology of the Right Upper Lobe Mass

FINAL DIAGNOSIS

Thoracic actinomycosis is a rare disease. High index of clinical suspicion is required for its diagnosis. The typical imaging findings of thoracic actinomycosis is chronic refractory pneumonia that does not obey anatomic barriers or is predominantly an endobronchial lesion.[8,3,4] The clinical course runs an extremely diverse course, which maybe a mild form of subclinical illness to an end-stage fatal disease. However, an early diagnosis followed by adequate treatment improves the prognosis leading to minimising complications and avoiding debilitating surgery.

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