Actinomicose torácica - a propósito de 3 casos clínicos

Thoracic Actinomycosis - apropos of 3 clinical cases

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RESUMO

A actinomicose é uma infecção bacteriana, supurativa e crónica caracterizada por múltiplos abscessos, trajectos fistulosos, fibrose e tecido de granulação envolvendo a face, pescoço, tórax ou abdômen devido a uma bactéria anaeróbica, gram positiva, saprófita e que só vive em cavidades. A actinomicose primária pulmonar é uma doença rara e provavelmente resulta da aspiração de secreções da orofaringe. Sem sinais clínicos característicos, pode apresentar-se como uma doença respiratória crónica. Aproximadamente 90% dos doentes são submetidos a procedimentos de diagnóstico e terapêutica baseado numa hipótese errada. Os autores apresentam uma revisão da actinomicose torácica a

ABSTRACT

Actinomycosis is a chronic, suppurative bacterial infection characterized by multiple abscesses, sinus tracts, fibrosis, and granulation involving the face, neck, thorax and abdomen due to an anaerobic gram positive commensal bacteria, harbored in the healthy cavities. Primary actinomycosis of the lung is a rare disease and probably results from aspiration of oropharyngeal secretions. Without characteristic clinical signs, may present as chronic respiratory disease with abnormality of radiography. Approximately 90% of patients have undergone diagnostic and therapeutics procedures based on a wrong diagnostic hypothesis. The authors present a comprehensive review of thoracic actinomycosis

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INTRODUCTION

Actinomycosis is a rare infection disease, and the incidence of thoracic involvement has decreased significantly (1). Although fairly uncommon, represents an important entity because of its varied presentations that may mimic other more common disease, the difficulties involved in its diagnosis, ad the long course of treatment necessary to eradicate the disease. Thoracic actinomycosis is a chronic supplicative infection of the chest wall, lungs, pleural space, and mediastinum caused by filamentous bacterial species from the genus Actinomyces, characterized by the formation of multiple abscesses, sinus tract formation, and purulent discharge containing yellowish "sulfur granules". The onset of thoracic infection is frequently occult and usually proceeds indolently, producing nonspecific symptoms. Effective management of this disease requires a clear recognition of its varied clinical presentations.

Bacteriology

The first reported case of human infection with actinomycosis was made by Von Laneenbeck in 1845 and was attributed to a fungus. The word actinomycoses means "ray fungus", and reflects the general belief at the time that the organism was a fungus. The organism was first isolated from humans in 1891, when Wolff and Israel reported culturing it anaerobically growing only at body temperature. In the 1960, Waksman concluded that Actinomyces was a gram-positive bacteria.

The organisms that cause actinomycosis are members of the families Actinomycetaceae that comprise the five genera: actinomyces, arachnia, nocardia, actinomadura and streptomyces. The genus Actinomyces are prokaryotes with cell walls that contain both muramic acid and diaminopimelic acid. Unlike the cell walls of fungi, the cell walls of these organisms do not contain sterols and are insensitive to polyene antibiotics. The organisms are gram positive, branching, nonspore forming bacilli and they are anaerobic or microaerophilic (2). They are very difficult to grow in culture, usually 3-10 or more days are required before the organism can be macroscopically detected. The culture recovery rate from active infection is only approximately 30%. The organisms responsible for infection in man are thought to be actinomyces israelii, naeslundii, viscosus, and odontolyticus; of these israelii is most common (78%) (3).

Pathogenesis

Apparently, in the normal host, the organism can...
live as oral and gastrointestinal commensals in regions with anaerobic growth conditions such as peritonsillar crypts, gingivodental crevices, salivary calculus, and the lower gastrointestinal tract. The lower respiratory tract in-patients with underlying chronic airway diseases may also harbour Actinomyces as saprophyte (4). Pulmonary infection is more common in-patients with underlying respiratory disorders such as emphysema, chronic bronchitis, chronic pneumonia (foreign body), and bronchiectasis (4). Actinomycosis remains a rare infection not having emerged as an important opportunistic pathogen in immunocompromised hosts (4).

Common predisposing conditions that promote thoracic actinomycosis include: trauma, surgical manipulation, suppuration from other bacterial pathogens and anaerobic tissue conditions as occur after particulate aspiration with resultant pulmonary atelectasis. Pulmonary infection occurs when patients with orogingival infection aspirate particles laden with organisms. Subsequent occlusion of a bronchus promotes atelectasis or pneumonitis from accompanying pathogens. Anaerobic conditions and tissue necrosis ensure, creating a favorable environment for the development of actinomycosis. There is a basilar predominance of the disease, but some studies reported apical predominance. Less commonly thoracic infection may be introduced by extension into the mediastinum from the neck (5,6), by esophageal perforation (7), or by spread through the diaphragm from an abdominal site (7); hematogenous spread to the lung is rare.

Incidence

The incidence of symptomatic actinomycosis infection is quite low. Most reviews report approximately one case per year in major medical centers. The organisms have no definite infective sites. Classically it involves cervicofacial (55%), abdominalpelvic (20%), thoracic (15%) and mixed organs (10%), including skin, brain, pericardium, and extremities (8).

There are a slight male predominance and the age of patients centers around the 4th to 6th decade of life. There are no known predisposing racial, environment or geographic factors.

Clinical manifestations and diagnosis

During the early stages of pulmonary infection, patients may appear only mildly ill or remain asymptomatic without fever or leukocytosis. Typical symptoms include cough productive of purulent or blood-streaked sputum, fever, and night sweats. The infection progresses slowly over weeks to months as the patient experiences additional complaints of weight loss, anemia, and leukocytosis. In the absence of the chest wall invasion or sinus tract formation, clinical manifestations are nonspecific and mimic other chronic infections and thoracic neoplasm (2,9,10,11). Less commonly, the infection extend to pleura and chest wall, it will cause empyema, nodular thickening of the pleura, the abscess or sinus tract formation of the chest wall. When the infection spreads to the pleura the patient notices worsening fever and severe pleuritic chest pain. Further progression into the chest wall causes a more persistent and localized thoracic pain associated with tenderness. Signs of chest wall inflammation may be apparent at this time.

The chest radiographic features of actinomycosis are varied and depend on the stage of the disease and presence of pleural and chest involvement (12). The typical radiographic findings consist of acute non-segmental airspace disease, usually in the lung periphery and with a lower lobe predominance (13). Focal infiltrates occur early and progress slowly to consolidation and cavitation. The infiltrate does not respect segmental or lobar boundaries and extends across lobar fissures in contrast to other more common causes or bacterial pneumonia (12). Continued consolidation creates a dense, masslike infiltrate that simulates a pulmonary neoplasm (14) or a lung abscess when cavitation occurs. Multiple cavities
develop in more than 50% of patients. Fibrocavitary infiltrates with loss of volume may follow and mimic pulmonary tuberculosis (4). Actinomycotic intracavitary colonization with an air meniscus mimicking a "ball-in-hole" is a rare presentation of thoracic actinomycosis (15). Solitary infection of the bronchial mucosa without pulmonary parenchyma involvement has been observed with actinomycosis (16). The patients present with fever, cough, and hemoptysis with radiographic evidence of lobar or segmental bronchial obstruction. Bronchoscopy will detect the firm inflammatory mass.

The pleura is radiographically abnormal in 50 to 80% of patients with thoracic actinomycosis (7,12) with thickening, and small or moderate pleural effusion. Massive effusions seldom occur (2,5). Chest wall invasion and pleural transgression are quite suggestive of actinomycosis. Patterns of chest wall disease include focal mass, periosteal reaction along ribs, and vertebral body destruction (12). A wavy periosteal reaction involving ribs is said to be highly suggestive of pulmonary actinomycosis.

A computed tomographic (CT) scan of pulmonary actinomycosis usually reveals a soft tissue mass with varying degrees of infiltration, abscess formation, and pleural thickening adjacent to the airspace consolidation which is seldom diagnostic (15).

The diagnosis of thoracic actinomycosis is very difficult. Clinically, thoracic actinomycosis may stimulate a wide variety of pulmonary disorders. This includes poorly responding pneumonia, tuberculosis, lung tumor, nocardiosis, cryptococcosis, or histoplasmosis (2,14,17,18). Radiographic findings are also often nonspecific, leading to difficulty in differential diagnosis from other disease (16). Definite diagnosis of the pulmonary actinomycosis is usually based on microscopic examination or anaerobic culture of the material aspirated from lesion, anaerobic sputum culture on multiple samples, or histological examination of the resected specimen and from fiberoptic bronchoscope. The bronchoscope findings are usually not diagnostic and include yellowish, hard or friable endobronchial masses (19,20). Identification of typical yellow or white sulfur granules from clinical specimens (sputum, endobronchial biopsy, and pleural fluid) is strongly suggestive of the diagnosis (10,21).

Management

Once the diagnosis of thoracic actinomycosis was established, a prolonged course of high-dose antibiotics to penetrate fibrotic, suppured tissue is required (5). Penicillin remain the first-line drug intravenously in a dosage of 10-20 million units per day for 2-6 weeks (22), followed by an oral phenoxyethyl penicillin in a dosage of 2-4 grams per day for an additional 6 to 12 months. Alternate antibiotics include tetracycline (12), erythromycin, clindamycin (23), chloramphenicol (24), first-generation cephalosporin and imipenem (25). The surgery should be performed to rule out cancer if the diagnosis is not con firmed in some cases. However, surgery also required for complications such as empyema, chronic sinus traction formation or recurrent hemoptysis (25,26).

Thoracic actinomycosis previously had a mortality rate of 80% before the advent of effective antibiotic therapy. With appropriate antibiotic drug and surgical therapy when indicated, most patients survive (9).

CASES REPORTS

We now report 3 cases, which was considered as other diseases before the histological examination. The final diagnosis was confirmed by biopsy.

Case 1

A 62-year-old Chinese man, heavy smoking habit, with chronic bronchitis some years ago, who suffered from chronic cough with muco-purulent sputum and intermittent hemoptysis for 4 months. He denied any history of exposure to tuberculosis, fever or other constitutional symptoms. No special finding on
physical examination. Sputum culture for bacterium, acid fast smears and culture for mycobacterium tuberculosis were negative on three occasions. The hematological studying, liver biochemical tests, and renal function tests were all within normal limits. The Chest radiography revealed two round opacity in the right lung field (Fig 1). A computed tomographic of the chest (CT) scan revealed one nodule with calcification in the right pulmonary hilum (Fig 2) and another round basal lesion. Bronchoscope examination showed a white color, hard mass that occluded the right basal bronchus (Fig 3) and biopsy was done. Before the histology results, the clinical diagnosis is highly suspected the lung tumor. The biopsy samples result obtained by bronchoscope show extensive purulent inflammation with necrosis, and some granules composed of radiated filaments surrounded by neutrophils (sulfur granules) (Fig 4) the typical pathological finding of actinomycosis. So the diagnosis was primary bronchial actinomycosis and the patient was treated with intravenous penicillin G for two weeks followed by oral penicillin V for two months. The radiographic aspects do not disappear and we could not exclude definitively a lung malignant neoplasm.

Therefore the patient underwent a thoracotomy to exclude the diagnosis of lung cancer. The pathological examination of right lower lobectomy revealed in
one specimen the bronchi filled with polymorphonuclear neutrophils and some granules that show filament or granules in Gomori methenamine silver (GMS) (Fig. 5) and Gram stain similar to actinomycetes. On the other specimen show a tumor well circumscribed, lobulated and composed of benign maturation of cartilaginous tissue – a chondroma. The patient was treated again with penicillin for six
months and he was clinically stable during one year of follow up.

Case 2

A 42-year-old Chinese female, non-smoker, who had a 5 years ago history of right lung squamous carcinoma and underwent a pneumonectomy and chemotherapy in a China hospital. She suffered recurrent cough and expectoration after the operation but these symptoms worsen in recent 6 months. Sputum smears and culture for acid fast bacilli were negative. The hematological studies, liver biochemical tests, and renal function tests were all within normal limits. Chest film shows a homogeneous opacity of the all right hemithorax (pneumonectomy). Examination with fiberoptic bronchoscope showed irregular white mass in the stump of right main bronchus. These findings suggested the diagnosis of malignancy recurrence. The bronchial washing for cytological studies was negative. Histopathologic examination of a specimen, obtained by bronchial biopsy, revealed many sulfur granules with infiltration of neutrophils, which led to the diagnosis of endobronchial actinomycosis.

After six months of treatment, with intravenous penicillin G followed by amoxicillin, the symptoms improved but the comparison of bronchoscope findings before and after treatment was not possible, because the patient refused repeats this examination.

Case 3

A 62 year-old Chinese man, with alcoholic habits had a history of right middle and lower lobe obstructive pneumonia. Bronchoscope examination showed the mucous of the right intermedium bronchus to be erythematous, filled with purulent secretions and obstructed by a chicken bone that was take out of respiratory tract. A yellowish friable formation of the mucous was seen in the middle and lower lobe. Biopsy samples obtained by bronchoscope showed "sulfur granules" and hyphae-like components. Bronchial washing for acid fast bacilli were negative. The patient had received a long course of penicillin oral with an obvious clinical improvement but without resolution of the radiographic changing. Some months later the patient began with recurrent bacterial infections with necrosis of the lung that improve temporarily with large spectrum antibiotic. Thoracotomy with lobectomy was considered but the patient refused. The patient died 4 years later due to recurrent severe bacterial infection and respiratory failure.

CONCLUSION

Thoracic actinomycosis is a rare chronic infection disease caused by several species of the family Actinomycetaceae, gram-positive anaerobic bacteria. Usually is secondary to aspiration of colonized material from oropharynx in-patients with poor oral hygiene and without characteristic clinical manifestations.

In our hospital, we have found 3 cases with bronchial actinomycosis. All of these cases are no typical manifestation of actinomycotic infection. The first case was suspected as lung tumor, the second case was consider as post-operative recurrence of lung malignancy, and the third case present as obstructive pneumonia after foreign body (chicken bone) aspiration in airway. The bronchoscope findings are usually not diagnostic and include similar aspects of these three patients with white or yellowish, hard or friable endobronchial neoformation. The diagnosis was based on histological examination of the bronchoscope biopsy that revealed "sulphur granules", the typical pathological finding of actinomycosis.

Long term course of Penicillin is the first choice. Another antibiotics such as clindamycin, tetracycline, erythromycin and chloramphenicol are also effective in therapy. The medical therapy remains the first choice for actinomycosis and surgical procedures are performed when necessary. If the infection is not control well, the illness becomes chronic or a progressively fatal disease.
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