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ARTIGO DE REVISAOIREVIEW ARTICLE

Actinomicose toracica - a proposito de 3 casos clinicos

Thoracic Actinomycosis - apropos of 3 clinical cases

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RESUMO

A actinomicose e uma bacteriana,
supurativa e cronica caracterizada por multiplos
abcessos, trajectos fistulosos, fibrose e tecido de
envolvendo a face, torax ou
abdomen devido a uma bacteria anaerobica,gram positiva, saprofita e que so vive em cavidades. A actinomicose primaria pulmonar e uma rara
e provavelmente resulta da de
da orofaringe. Sem sinais clinicos caracteristicos,
pode apresentar-se como uma respiratoria
cronica. Aproximadamente 90% dos doentes sao submetidos a procedimentos de diagnostico e tera-peutica baseado numa hip6tese errada. Os autores apresentam uma revis&o da actinomicose toracica a

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ABSTRACT

Actinomycosis is a chronic, suppurative bacterial infectio" 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 aspirati-on of oropharyngeal secretions. Without characte-ristic clinical signs, may present as chronic respira-tory ,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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prop6sito de his casos dinicos. Um caso era suspei-to de ter um tumor pulmonar,o segundo caso foi considerado ser a recurrinda de uma mali2na e o terceiro caso apresentou-se com uma forma de pneumonia recidivante ap6s de um corpo estranbo. 0 diagn6stico foi confirmado atraves do n ame bistol6gico.

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Palavras-chave: Actinomicose; Radiografia de tQrax; Broncoscopia; Granulos sulfurosos; Penicilina G.

INTRODUCTION

Actinomycosis is a rare infection disease, and the incidence of thoracic involvement has decreased signifcantly (1). Although fairly uncommon, repre-sents an important entity because of its varied presen-tations that ll)ay 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 suppurative 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 formati-on, and purulent discharge containing yellowish "sulfur granules". The onset of thoracic infection is frequently occult and usually proceeds indolently, producing nonspecific symptoms. Effective manage-ment of this disease requires a clear recognition of its varied clinical presentations.

Bact enology

The first reported case of human infection with actinomycosis was made by Von Laneenbeck in 1845 and was attributed to a fungus. The word actinomy-ces means "ray fungus", and reflects the general belief at the time that the organism was a fungus. The

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apropos of three clinical cases. One case was suspec-ted as lung tumor, tbe second case was consider as recurrence of lung malignancy, and tbe thJrd case present as recurrent pneumonia after foreign body aspiration in airway. Final diagnosis was confirmed by histolocY examination.

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Key-words: Actinomycosis; Thoracic radiography; Bronchoscopy sulfur granules; Penicillin G

organism was first isolated from humans in 1891, when Wolff and Israel reported culturing it anaerobi-cally growing only at body temperature. In the 1960, Waksman concluded that Acrmomyces was a gram-positive bacteria.
The organisms that cause actinomycosis are members of the families Actinomycetaceae that comprise the five genera: actinomyces, arachma, nocardia, actmomadura 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 insen-sitive 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 macros-copically 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, naeslundu. viscosus. and odontolyticus; of these israelii is most common (78%) (3).

Pathogenesis

Apparently, in tbe normal host, the organism can

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live as oral and gastrointestinal commensuals 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 Actinomy-ces as saprophyte (4). Pulmonary infection is more common in-patients with underlying respiratory disorders such as emphysema, chronic bronchitis, chronic pneumonia (foreign body), and bronchiecta-sis (4). Actinomycosis remains a rare infection not having emerged as an important opportunistic patho-gen 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 organjsms. Subsequent occlusion of a bronchus promotes atelectasis or pneumonitis from accompan-ying 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 esopha-geal perforation (7), or by spread through the diaph-ragm 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 approxima-tely 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 (tO%), including skin, brain, pericardium, and extremities (8).

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There are a slight male predominance and the age of patients centers around the 41.h to 6'b decade of life. There are no known predispOsing racial, environment or geographic factors.

Clinical man({estations 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, anemJa, 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 feyer and severe pleuritic chest pain. Further progres-sion 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 involve.ment (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 com-mon 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

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develop in more than 50% of patients. Fibrocavitary infiltrates with loss of volume may follow and mimic pulmonary tuberculosis (4). Actinomycotic intracavi-tary colonization with an air meniscus mimicking a "bali-in-hole" is a rare presentation of thoracic acti-nomycosis (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. Bronchoscope will detect the finn inflammatory mass.
The pleura is radiographically abnormal in 50 to 80% of patients with thoracic actinomycosis (7.12) wtth 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 suggesttve of pulmonary actinomycosis.
A computed tomographic (CT) scan of pulmonary actinomycosis usually reveals a soft tissue mass with varying degrees of infiltration, abscess fonnation, and pleural thickening adjacent to the airspace consolida-tion 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 mcludes poorly responding pneumonia, tuberculosts, lung tumor, nocardiosis, cryptococcosis, or histoplas-mosis (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 examina-tion 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).1dentification of typical

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or white sulfur granules from clinical speci-mens (sputum, endobronchial biopsy, and pleural fluid) is strongly suggestive of the diagnosis (10.21).

Managemenl

Once the diagnosis of thoracic actinomycosis was established, a prolonged course of high-dose antibio-tics to penetrate fibrotic, suppurated tissue is required (5). Penicillin remain the first-line drug intravenously in a dosage of 10-20 million un its per day for 2-6 weeks (22), followed by an oral phenoxymethyl penicillin in a dosage of 2-4 grams per day for an additional6 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 finned in some cases. However, surgery also required for complt-cations 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 finaJ diagnosis was con finned 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 intermitted hemoptysis for 4 months. He denied any history of exposure to tuberculosis, fever or other constitutional symptoms. No special finding on

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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. I). A computed tomographic of the chest (CT) scan revealed one nod ule with calcifi-cation in the right puJmonary hilum (Fig.2) and another round basal lesion. Bronchoscope examinati-on showed a white color, hard mass that occluded the right basal bronchus (Fig.3) and biopsy was done. Before the histology resuJts, the clinical diagnosis is highly suspected the lung tumor. The biopsy samples result obtained by bronchoscope show extensive purulent innammation with necrosis, and some granules composed of radiated filaments surrounded by neutrophils (sulfur granules) (Fig.4) the typical pathological finding of actinomycosis. So the diagno-sis 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 malig-nant neoplasm.

Fig. I - Case I View of the right hemithorax from a posteroan-terior roentgenogram reveal a poorly defined round area
on the right hilar region, with calcifications and a infiltrate in the right lower lobe

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Fig. 2- Case I. Computed tomography scan of the chest revea-led a 30x40 mm. well defined, calcified nodule on the
right lower hilar region

Fig. 3- Case I Bronchoscope examination shows a white and smooth mass-like lesion in the posterior basal segment
of right lower lobe

Therefore the patient underwent a thoracotomy to exclude the diagnosis of lung cancer. The pathologi-cal examination of right lower lobectomy revealed in

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Fig. 4 - Case I Htstologic slide from a bronchial biopsy sho-wing a typical sulfur granules surrounded by numerous polymorphonuclear leukocytes

one spectmen the bronchi filled with polymorfhonu-clear neutrophils and some granules that show filament or granules in Gomori methenamine si lver (GMS) (Fig. 5) and Gram stain simi lar to actinomy-

ces. On the other specimen show a tumor well Cir-cumscribed, lobulated and composed of bemgn
maturation of cartilaginous tissue-a chondroma. The
patient was treated again with penici llin for six

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Fig. 5- Case I. Silver stain (Gomori methenamine-silver) of granule showing thin, branching filaments of actinomyces

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months and he was clinjcally stable during one year of follow up.

Case2

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 biochemi-cal 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 infiltrati-on 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.

Case3

A 62 year-old Chinese man, with alcoholic habits had a history of right middle and lower lobe obstruc-tive pneumonia. Bronchoscope examination showed the mucous of the right intermedius 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 "sulphur granules" and hyphae-like components.

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Bronchial washing for acid fast bacilli were negative. The patient had received a long course of penicillin oral with an obvious clinical improvement but wit-hout resolution of the radiographic changing. Some months later the patient began with recurrent bacteri-al Infections with necrosis of the lung that improve temporarily with large spectrum antibiotic. Thoraco-tomy with lobectomy was considered but the patient refused. The patient died 4 years later due to recur-rent 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 manifesta-tions.

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 obs-tructive 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 bron-choscope biopsy that revealed "sulphur granules", the· typical pa1hological finding of actinomycosis. Long term course of Penicillin is the first choice. Another antibiotics such as clindamycin, tetracycline, erythtromycin 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 becames chronic or a progres-sively fatal disease.

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