

Cervicofacial actinomycosis: still a difficult differential diagnosis

Actinomicosi cervicofacciale: diagnosi differenziale ancora difficile

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Key words

Head and Neck • Actinomycosis • Diagnosis

Parole chiave

Testa e collo • Actinomicosi • Diagnosi

Summary

Cervicofacial actinomycosis, a rare chronic infectious disease, is, however, an important clinical entity, due to the difficulties involved, still today, in its diagnosis. Following personal experience in a case referred to our Department, and in agreement with reports in the literature, attention is drawn to the presenting clinical manifestations, stressing that these are often confusing since they mimic those of other diseases. Moreover, many pre-operative investigations (radiological scans, incisional biopsy, fine-needle aspiration) are generally non-specific. Finally, surgical excision of the mass is now the last essential step to make a definitive diagnosis and define the appropriate antibiotic therapy.

Riassunto

L'actinomicosi cervicofacciale è una rara malattia infettiva cronica che rappresenta tuttavia un'importante entità clinica, a causa delle difficoltà che ancora pone in fase diagnostica. Basandoci sull'osservazione di un caso clinico occorso nel nostro Reparto, in accordo con la letteratura, abbiamo sottolineato come le manifestazioni cliniche d'esordio siano spesso confondenti, poiché comuni ad altri processi patologici, e come le indagini pre-operatorie (scansioni radiologiche, biopsie escissionali, agoaspirato) risultino in genere non specifiche. L'escissione chirurgica della massa è quindi attualmente ancora essenziale per porre diagnosi certa ed istituire l'appropriata terapia antibiotica.

Introduction

Actinomycosis is a suppurative and granulomatous chronic infectious disease, that usually spreads into adjacent soft tissues without regard for tissue planes or lymphatic drainage; it may also be associated with a draining sinus tract¹⁻³.

Actinomyces are Gram-positive, non-acid fast, anaerobic or microaerophilic filamentous branched bacteria, living as commensal organisms in the human oral cavity and respiratory and digestive tracts, becoming invasive when, through a mucosal lesion, they gain access to the subcutaneous tissue. Thus, dental caries, dental manipulations and oromaxillofacial traumas are the most common triggering events¹⁻⁴⁻⁹.

In 1938, Cope classified actinomycosis infection into three distinct clinical forms: cervicofacial (50%), pulmonothoracic (30%) and abdominopelvic (20%). The first of these manifestations is the most frequent, although fairly uncommon: a review of literature revealed 48 cases of cervicofacial actinomycosis reported over the last 25 years. The condition is considered an important clinical entity, on account not

only of the difficulties involved in the diagnosis but also the long-term treatment necessary to eradicate the disease¹⁰⁻¹³.

Case report

A 51-year-old Egyptian male was referred to our Department of Otolaryngology, with a two months' history of a slow-growing painless right submandibular mass, not initially associated with any discharge. Antibiotic therapy, previously prescribed by a physician, did not lead to a decrease in size of the mass. The patient denied any history of oromaxillofacial trauma or recent dental extraction.

Head and neck examination revealed a 4 x 4 cm mass in the right submandibular region, which was tender upon palpation and partially fixed on the deep tissue planes, covered by slightly erythematous skin, but without breakdown associated with the mass. Panendoscopy was normal.

Routine blood tests were normal and PPD was placed which was found to be nonreactive and there was no response to PPD.

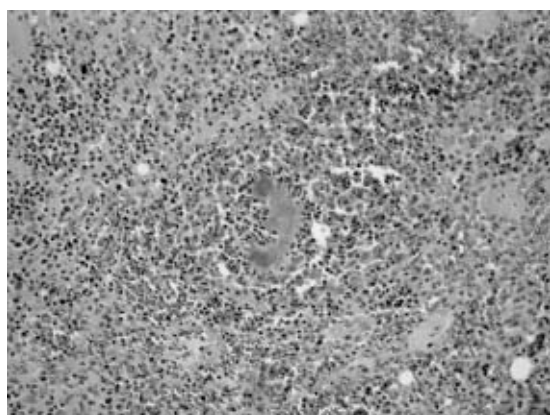


Fig. 1. Microscopic findings of chronic inflammation with the presence of multiple granules surrounded by polymorphocytes were consistent with diagnosis of actinomycosis (haematoxylin and eosin stain; magnification X 400).

A computed tomography (CT) scan of the neck revealed an expansive large mass (approximately 4.5 cm in size), located in front of the right sternocleidomastoid muscle.

A haematic-caseous discharge from the lower fluctuant portion of the mass was collected through a percutaneous incision. A specimen submitted to microbiologic culture revealed the presence of *Fresobacterium Nucleatum*, *Porphyromonas Asaccharolytica* and *Staphylococcus Aureus*.

The patient, therefore, underwent surgical excision of the mass, the histopathological examination of which showed chronic inflammation with the presence of multiple granules surrounded by polymorphocytes: this microscopic finding being consistent with diagnosis of actinomycosis (Fig. 1).

The patient was started on high doses of penicillin for 4 weeks by the specialist in infectious diseases. The patient made a complete recovery and, moreover, follow-up revealed no recurrence of the infection.

Discussion

Actinomyces are gram-positive, non-acid fast, anaerobic or microaerophilic filamentous branched bacteria which are very difficult to grow in culture, with < 30% of cultures being positive. In man, the pathogenic Actinomyces most frequently isolated is *A. Israelii*; less commonly, infection is caused by *A. Propionica*, *A. Naeslundii*, *A. Viscosus* and *A. Odontolyticus*. These bacteria are all normal commensals of the human oral cavity^{3 11 14-16}.

In cervicofacial actinomycosis, which is the most frequent manifestation, infection is frequently the result of oromaxillofacial trauma, dental manipulation or dental caries¹⁰. In the present case, the patient denied any clinical history of oromaxillofacial trauma and showed no sign of immunodeficiency: lack of these risk factors did not help us make the diagnosis.

The infection, most commonly, presents as a chronic, often fluctuant mass, frequently located at the border of the mandible, becoming progressively larger within weeks or months¹¹.

Symptoms are often non-specific: pain is rare, slight fever occurs in >50% of patients^{5 9}, associated with a sensation of superficial tension around the mass. Initially, the mass may be surrounded by induration or erythema; later, it may become tender to palpation, on account of a central necrosis process^{17 18}.

The classic formation of spontaneous sinus tracts draining purulent material is observed in approximately 40% of cases, and, when present, may be helpful in the differential diagnosis¹⁹. Since our patient presented a submandibular mass without external drainage, a glandular disease was initially suspected.

Although Actinomyces rarely involves the lymph nodes, regional lymphadenopathy is sometimes observed.

Furthermore, imaging techniques (computed tomography (CT) and magnetic resonance imaging (MRI) scan) usually yield non-specific findings, contributing only to define radiological features of the mass and its involvement in adjacent soft tissues. Also in the present case, CT was found to be useful in planning the surgical treatment^{20 21}.

On account of these non-specific manifestations and radiological aspect, the clinical differential diagnosis of actinomycosis still remains difficult.

Definitive diagnosis may be established only by a positive culture, however, Actinomyces growth is very difficult even on appropriate anaerobic media (recovery rates from culture are < 50%)²². Thus, microbiological identification of this organism is often impossible. The macroscopic presence of the classic sulfur granules in tissue specimens or drainage may be of some help when making diagnosis, even if these features are not pathognomic, since nocardiosis may also present with sulfur granules²³.

Several Authors agree that incisional biopsy can be of great help in the diagnosis of actinomycosis, since microscopic examination reveals a typical finding of an outer zone of granulation and a central zone of necrosis which contains multiple basophilic granules, that represent lobulated micro-colonies of Actinomyces⁵.

Over the last few years, as investigators have been searching for less invasive diagnostic techniques, fine-needle aspiration (FNA) has become more and

more important, since not only does it allow morphologic identification, comparable to that obtained from incisional biopsy, but is also an effective means of collecting material for microbiologic identification²⁴⁻²⁶. In the light of these important diagnostic difficulties, cervicofacial actinomycosis has been referred to as the great masquerader of head and neck disease²⁷; thus, fewer than 10% of infections are correctly diagnosed⁹, and surgical excision remains the only really resolutive approach to make definitive diagnosis, particularly in those cases presenting the formation of an abscess, unresponsive to antimicrobial therapy or when FNA is non-diagnostic. Even if surgery plays an important role both in the diagnosis and treatment of actinomycosis, recurrence following surgery alone is very common, and 2-4 weeks of high-dose intravenous antibiotics are a fundamental part of treatment, followed by 3-6 months of oral antibiotics. Penicillin is the drug of choice;

tetracycline and erythromycin are employed in patients allergic to penicillin^{9,28,29}. In the acute phase of treatment, penicillin can be replaced by cephalosporins which are also effective if a co-infection with other bacteria not responding to penicillin causes the persistence of symptoms due to Actinomyces³⁰⁻³².

Also in our case, in accordance with data reported in the literature, surgical excision was ultimately required for definitive diagnosis, and complete resolution of symptoms was achieved after adequate post-operative antibiotic treatment.

In conclusion, although it is a rare infectious cervicofacial disease, actinomycosis of the head and neck represents, among neck masses, an interesting disease, on account of the difficulties involved in the diagnosis. A comparison between clinical and microbiologic findings avoids serious errors in the differential diagnosis.

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■ Received: November 19, 2004
Accepted: January 3, 2005

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