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Maxillofacial Surgery

A Case Report of Mandibular Actinomycosis: Unusual Evolution

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Abstract Case Report

Actinomycosis of the jaws is an underdiagnosed disease, which has a large clinical polymorphism that can affect all organs. This bacterial condition is very variable, making its clinical diagnosis difficult. The histo-pathological examination remains the key to diagnosis due to the difficulties of bacteriological examination. Treatment is surgical debridement and prolonged antibiotic therapy. our study highlights the case of a patient treated at the MOHAMMED VI Department of Maxillofacial Surgery in Marrakech, followed for a period of 2 years between 2022 and 2024. Objective of this study is to report the possibility of occurrence of this pathology in an immunocompetent subject and the good clinical and radiological evolution under medical treatment alone.

Keywords: Actinomycosis, Infection, Jaw, Penicillin.

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INTRODUCTION

Actinomycosis is a specific, rare, non-contagious bacterial disease, sometimes acute, often chronic, preferentially affecting the cervico-facial sphere. It is a purulent, granulomatous infection. It is caused by Gram-positive anaerobic bacteria: Actinomycetes, which are generally saprophytes of natural human cavities, notably the oral cavity, but can become pathogenic under certain conditions.

Actinomyces israelii is often the cause in human pathology. Actinomycosis occurs primarily through contiguity from a dental focus [1]. It is a specific, primary infection of soft tissue and rarely of bone.

Actinomycosis osteitis mainly affects the mandible [2]. Histopathological examination remains the key to diagnosis, given the difficulties of bacteriological examination.

CASE REPORT

Our patient is 52 years old and underwent surgery for cholyestitis 10 years ago. She is in good general condition and wears a dental prosthesis. She presented with right lower jugal swelling and pain over the right mandibular horizontal branch which had been present for 6 years, associated with homolateral vestibular ulceration (Figure 1).

Maxillofacial examination revealed pain on palpation of the mandible in relation to the mandibular region.

The patient was edentulous, and intraoral examination revealed a vestibular ulceration of the right mandibular horizontal branch, with no pus emission, no limitation of mouth opening and no bone exposure.

An orthopantomogram (Fig 2) revealed multiple osteolytic lesions involving the right mandibular horizontal branch, prompting us to perform a facial CT scan which revealed the same lesional process of interest, classified as Lodwick stage 2, immediately suggesting a tumoral or infectious origin (Fig 3).

In view of this clinical and radiological presentation, a bone biopsy was performed under local anesthesia, with an anatomopathological study revealing actinomycetes grains (Figure 4).

We opted for medical treatment alone with amoxicillin 4 grams daily for 4 months, which showed a good clinical and radiological evolution. No signs of recurrence or local complications were detected after 2 years' follow-up.



Figure 1: A: Slight swelling in the cheek; B: Appearance of a right lower vestibular ulceration adjacent to the mandibular horizontal ramus

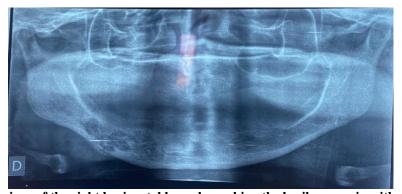


Figure 2: Osteolytic lesions of the right horizontal branch reaching the basilar margin without periosteal reaction or soft tissue infiltration

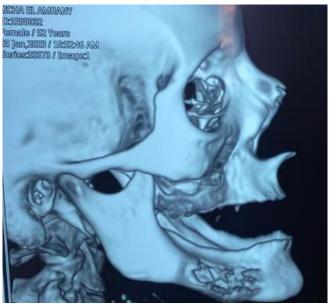


Figure 3: Facial CT scan: mandibular mandibular lesion process stage 2 of Lodwick

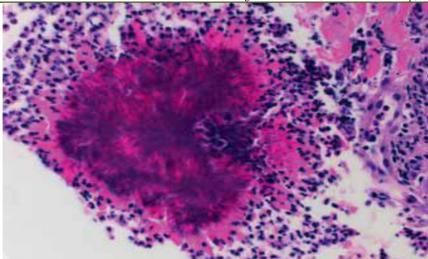


Figure 4: Histological section in Gram stain (magnification 200): actinomycotic grain or "sulphur granule"



Figure 5: 1-year follow-up orthopantomogram: total remission with complete osteogenesis of the right horizontal branch of the mandible

DISCUSSION

Actinomycosis is a rare disease (5/100,000), with cervicofacial localization the most common (50-75% of cases). It is mainly caused by Actinomyce israelii, a species of Gram-positive filamentous bacteria in the Actinomycetaceae family, of which it is the type type. Actinomycetes are saprophytes of the oral cavity, tonsil crypts and digestive tract [3].

Their pathogenicity manifests itself under certain conditions, such as poor oral hygiene, trauma such as tooth extraction, surgery or dental surgery [3].

They are also pathogenic in patients with a history of anesthesia, radiotherapy, bisphosphonate treatment, immunosuppression, diabetes or neoplastic pathology [4]. Cervico-facial actinomycosis presents as a picture of chronic suppuration, most often evolving at a low level with an extensive tendency [5].

More rarely, the affection may be localized under the oral mucosa. Lingual, jugal, pharyngeal and salivary forms have been reported. In the majority of cases, lesions remain localized to the soft tissues. In literature, only 15 cases show bone extension, mainly by intra-oral contiguity, more rarely by extra-oral contiguity, from a cutaneous focus [6].

The diagnosis of actinomycosis should be considered in all cases of chronic recurrent cellulitis and abscesses that fail to respond to short-term antibiotic therapy. Radiological signs are non-specific, with bone involvement manifested mainly in the form of osteolytic images, osteo-condensation or mixed images [7].

Diagnosis is based on anatomopathological study of biopsied fragments, showing typical radial bacterial filamentous structures; the classic yellow grains are colonies of actinomycetes [4].

From a pathophysiological standpoint, the hyper-reaction of the host's defenses leads to the formation of a sclerotic gangue that favors bacterial survival in an anaerobic environment and prevents the penetration of in vitro antibiotics. This explains not only the clinical appearance, but also recurrences and therapeutic failures.

Differential diagnoses include pyogenic abscess, tubercular suppuration, syphilis, deep mycosis and tumoral etiology.

The approach is primarily prophylactic, involving oral hygiene and treatment of periodontal infection and dental caries.

In the literature, treatment combines surgery and prolonged penicillin antibiotic therapy. The antibiotic treatment protocol for mandibular actinomycosis is two to four weeks of intravenous penicillin G, 12 to 20 million units per day, followed by penicillin V or oral amoxicillin 2 to 4 g/d; the duration of treatment is three to six months, depending on the clinical course. In case of allergy, macrolides, tetracyclines or third-generation cephalosporins (ceftriaxone) are possible alternatives to penicillin [8].

On the other hand, our case corresponds to a therapeutic success showing that only with Amoxcillin alone at a dose of 4g per day the evolution was marked by a total remission.

The surgical approach described essentially includes debridement, curettage, sequestrectomy and treatment of the portal of entry. In the case of bone loss, reconstruction may be necessary depending on the extent of the loss [9].

The prognosis of this disease depends on its extent, and any delay in diagnosis and treatment may be responsible for functional and aesthetic complications that can be disabling (loss of bone substance, permanent constriction of the jaws and retractive scars).

In severe forms, the disease can spread to the base of the skull, the cervical spine and the meninges.

CONCLUSION

Actinomycosis is a rare disease caused by the anaerobic bacterium Actinomyces israelii. It often affects the cervico-facial region and manifests itself as chronic suppuration. Several risk factors are implicated in its genesis. Diagnosis is based on histological analysis of the infected tissue. Treatment generally involves surgical intervention combined with long-term antibiotic

therapy, often based on penicillin, whereas our patient was only treated with prolonged antibiotic therapy.

The prognosis is generally favourable, so rapid diagnosis and appropriate treatment are essential to avoid complications

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