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# Cervicofacial Actinomycosis: A Case Report of a Mandibular Localization

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**Abstract** **Case Report**

Actinomycosis is a rare disease. The cervicofacial region is the most frequent localization. A chronic pus discharge, sometimes tumor-like, suggests the diagnosis, which is often confirmed by anatomopathological study. The treatment is surgical debridement and antibiotic therapy, frequently long-term. We report in the present work a case of a rare type of mandibular tumor about a 10-year-old girl who presented, to our maxillofacial surgery department of the university hospital center of Rabat, with a left fistulized mandibular swelling and normal mouth opening range. In her case, the diagnosis of actinomycosis was made by histology. The clinical outcome was good following an enucleation and a strong curettage and an antibiotic therapy. The lesions of this pathology, if untreated, may evolve to osteitis and sequestration which makes early diagnosis crucial.

**Keywords:** Actinomycosis, Mandible, Cervicofacial, Rare tumor.

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## INTRODUCTION

Actinomycosis is a rare and non-contagious bacterial disease, sometimes acute, often chronic, preferentially affecting the cervicofacial area. It is a purulent and granulomatous infection due to Gram-positive anaerobic bacteria: the Actinomycetes that usually saprophyte the natural cavities of the body, especially the oral cavity where it can become potentially pathogenic under certain conditions. Actinomyces Israeli is often the most implicated bacteria in human pathology. However, Actinomyces naeslundii, Actinomyces viscosus, and Actinomyces odontolyticus are occasionally identified. Actinomyces produces chronic, slowly developing infections, particularly when normal mucosal barriers are disrupted by trauma, surgery, or a preceding infection [1]. A break in the integrity of the mucous membranes and the presence of devitalized tissue can result in invasion of the deeper body structures and cause illness [2]. Cervicofacial actinomycosis occurs most of the time by contiguity from a dental focus.

It is a specific and primary infection of the soft tissues and rarely of the bone. Actinomycosic osteitis, which has become exceptional these days, often affects the mandible.

## CASE REPORT

We report a case of a 10-year-old Mauritanian girl, without any notable medical history who presented to our department with a 1-year history of swelling, pain, and purulent intra-oral discharge involving the region of the left mandible. The drainage was described as white, thick, and malodorous. The clinical examination found an afebrile child, in good general condition. Local examination reveals a painful, firm, fixed, medium-sized (nearly 7 centimeters in its largest diameter), left lower jaw swelling on palpation going from the 44th tooth to the left retromolar trigon. The examination of the oral cavity found a normal mouth opening range with damaged furcation and a bad oral state with very dilapidated left lower molars just next to the tumor. The tumor was fixed to the jaw bone with the presence of an intraoral fistula next to the 35th tooth with a purulent, white, thick, and malodorous discharge (Figures 1 & 2). In cervical exam no palpable cervical lymphadenopathy was found. An orthopantomogram and CT scan where done that showed a mandibular osteolytic tumor in the left horizontal branch of the mandible (Figures 3 & 4). The patient benefited from an enucleation of the tumor followed by a strong curettage. The follow-up after 1 month and 3 months was satisfying: Absence of inflammatory signs both on extraoral and intraoral level neither pus discharge was

present, with a post-operative CT scan that showed good follow-up results (Figure 5).

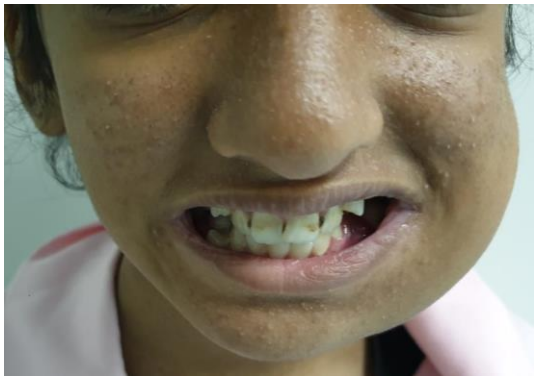

**Figure 1: Picture of the patient showing a left cheek swelling**

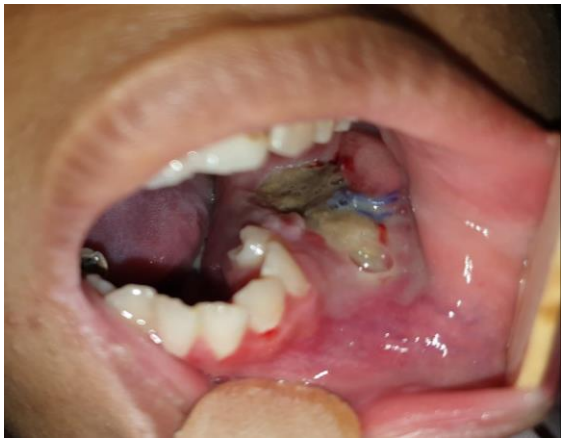

**Figure 2: An intraoral view of dilapidated left lower molars, the fistula and pus discharge**

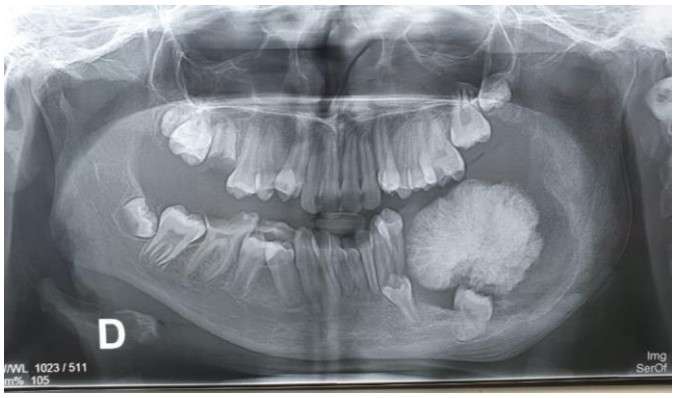


**Figure 3: An orthopantomogram showing an osteolytic tumor, as an opacity, in the left mandible**

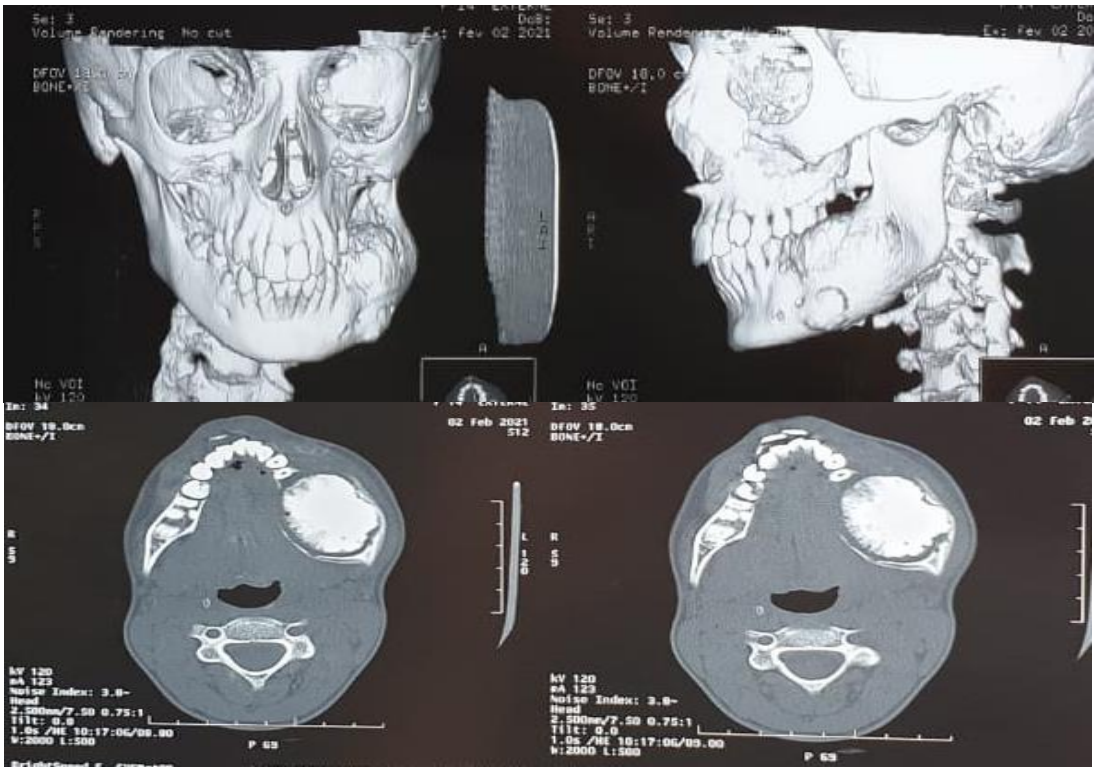

**Figure 4: CT scan 3D and axial images showing a mass of the left horizontal branch of the mandible**

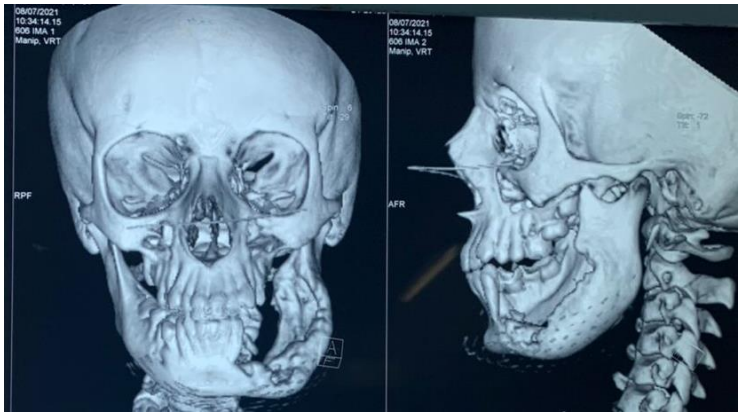


**Figure 5: Immediate Post-operative 3D CT scan showing the residual cavity**

## DISCUSSION

Actinomycosis was first described in the early 19th century as a disease of cattle [3]. The disease was recognized in 1854 by Graefe and in 1875 by Cohn, but the publication of the first description of actinomycosis in humans was in 1857 by Lebert [3]. In 1999 Schaal *et al.*, suggested several subgroups based on the composition of cell wall components and found that these groups correlate well with those seen in phylogenetic contemporaries [4].

Cervicofacial actinomycosis is a rare condition with a frequency of 5/100,000, this figure is probably underestimated due to the difficulty of diagnosis and the frequent association with other germs [5].

Actinomycosis usually occurs in adults between the ages of 20 and 60, but cases have been described in children [5, 6]. Women are less frequently affected than men with a sex ratio of 3/1 [5].

Bennhoff notes that there is no male predominance and that the result obtained is because of the frequency of maxillofacial trauma and alcoholism among men [3]. The prevalence of the disease in rural areas is classic [5].

In the literature, many localizations of actinomycosis have been reported. However, the cervicofacial location predominates with an average of 55 % [7, 8]. Bone involvement is a rare complication in the literature: between 1 and 15% of cases of cervicofacial actinomycosis. It most often affects the mandible [9, 10]. Actinomycosis of the upper jaws represents 5.7% of cervico-facial locations while actinomycosis of the mandibles represents 53.6 % [3]. Other sites that can be infected are thoracic (15٪), abdominal-perineal (20٪) and cerebral (2٪). Disseminated forms are exceptional [10].

Contributing factors to actinomycosis are:

- Poor oral hygiene,
- Traumas: Trauma creates a solution of continuity of the mucosa creating an anaerobic environment conducive to the multiplication of Actinomycetes [11].
- The local chronic infectious focus: Healing can only be obtained after the eradication of the infectious focus, especially dental [5, 6, 9]
- Chronic alcoholism [10].
- General factors include: diabetes, neoplasia, chemotherapy, radiotherapy, prolonged corticosteroid therapy, malnutrition, HIV infection, etc [5, 7, 9].
- Salivary stones: by promoting stasis and retrograde infection [5, 6].
- The low socio-economic level [12]: By poor hygiene and undernutrition.
- Iatrogenic factors: Cases of actinomycosis have been reported postoperatively in patients treated for head and neck cancer [13].

Cases of actinomycosis have been reported in patients undergoing mandibular reconstruction as well [14].

Actinomycosis evolves in a subacute or chronic mode and can affect all the viscera. The cervicofacial site is the most frequent localization. In the typical form, the starting point of the infection is most often tonsillar or dental [5].

The clinical aspects of cervicofacial actinomycosis are highly variable, depending on the evolution and the topography [5]. Most often the onset is insidious, in the peri maxillary region it presents in the form of a deep, indurated, bumpy swelling mass, infiltrating the superficial and deep teguments, more or less inflammatory, which can be accompanied by pain and trismus. The pain is more or less acute, occurring mainly in nocturnal crises or during inflammatory flare-ups. When it exists, trismus is early and may persist until healing [5].

The skin next to the swelling infiltrates and becomes purplish, it is covered by a series of papules separated by furrows [5].

In the absence of treatment, this swelling progresses to fistulization of the skin. At the top of a papule, the abscess opens which lets a serosanguinolent, lumpy liquid flow out, containing yellow grains [5]. This fistula evolves intermittently, so new inflammatory flare-ups occur, leading to new abscesses and new fistulas [5].

The general signs are moderate; the general condition is later altered. The bone remains intact for a long time [5]. The swelling progresses with impairment [5]:

- Bone: giving a picture of mandibular osteitis, which is rare.
- Muscular: damage to the masticatory muscles leading to painful trismus.
- Soft parts: with multiple fistulizations at distance.

Often, the lesions remain localized to the soft tissues. However, forms where the infection spreads to contiguous bone resulting in osteomyelitis have been reported [15]. A sudden onset, rapid progression of fever and fluctuating, painful, fistulizing swelling accompanied by trismus are signs of an acute form [7]. Fistulization to the skin or into the oral cavity, like in our case, gives rise to a thick pus containing actinomycotic seeds [7]. In the oral cavity, actinomycotic infection can lead to extensive tissue destruction that can progress to fistulization. However, there may be secondary damage to the sinuses, the orbit, then the bones of the skull. The infection can even spread to the chest [11].

Imaging during actinomycosis is nonspecific. The panoramic X-ray can show the mass and decay of the teeth with damage to the furcation. The CT scan and MRI can show the mass, bone sequestrations and a thickening of the soft tissues especially of the masseteric region as well as an osteoperiosteal peripheral reaction. Ultrasound can reveal cervical adenopathy. The positive diagnosis is based on the bacteriological examination of the pus taken from the lesion, which confirms the diagnosis by the demonstration of gram-positive filamentous bacilli. But the latter can be tricky because of the low rate of isolation of this germ [16]. It is the anatomopathological examination of the sampled material that most often makes it possible to make the diagnosis of this condition by identifying the actinomycotic grain [17, 18]. The treatment of actinomycosis is based on antibiotic therapy alone or combined in some cases with surgery [19, 20]. The duration of the antibiotic treatment is adapted according to the clinical evolution. Penicillin, cyclins, macrolides and synergistins are active on actinomyces [17]. Depending on the early diagnosis, the treatment can last from 1 month to several months. Surgery is indicated in the event of an abscessed, fistulized infection site like in our case [21]. Surgery will allow drainage of abscesses, flattening of fistulous tracts and excision of necrotic masses, thus avoiding recurrences [21]. Surgery can be for purely therapeutic purposes, or for diagnostic and therapeutic purposes.

The prognosis of the condition depends on early diagnosis and treatment, as it is a curable condition if recognized and treated well.

The histological differential diagnosis arises with mycetoma, nocardiosis, streptomyces, botryomycosis, leptothrix, aspergillosis. Mandibular actinomycosis can evolve over several years, accompanied by a progressive deterioration in general condition, and evolving towards death [18]. Oral hygiene is the key to prevention against cervicofacial actinomycosis, since it is the starting point of this condition [19].

Prevention involves the treatment and eradication of all oral infectious foci, which may constitute a shelter for actinomyces. Treatment of ENT infections is also necessary [20].

## CONCLUSION

Actinomycosis, particularly in its cervicofacial location, is an infection caused by Gram-positive anaerobic bacilli called Actinomyces that has become exceptional in industrialized countries. However, it is relatively common in developing countries. It often follows dental care or oral trauma. It most often affects the mandible. Its clinical diagnosis is quite difficult. Histopathological examination remains the key to diagnosis. Early, appropriate and prolonged intravenous and then oral antibiotic treatment is essential to prevent the occurrence of mutilating lesions. In some cases, a combination of drug treatment and extensive surgical debridement may be necessary.

Prophylaxis is based on good oral hygiene and the treatment of oral infectious foci. Our work aimed to discuss through a case report of actinomycosis of the mandibular bone and through bibliographical research, the clinical, diagnostic, therapeutic and evolutionary aspects of this affection.

**Conflicts of Interest:** The authors declare no competing interest.

**Authors' contributions:** All the authors participated in the treatment of this patient and in the redaction of this article.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due

efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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