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CASE REPORT

Actinomycotic osteomyelitis of the mandible: an unusual case

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Abstract
Background Actinomycotic osteomyelitis is an infection in soft tissues and/or bones, being associated with trauma or a previous nonspecific infection. This article presents an unusual case of mandibular osteomyelitis caused by *Actinomyces*.
Case report A 19-year-old male patient was referred for endodontic treatment of the lower right first molar about 16 months ago and removal of lower right third molar approximately 3 years before. The panoramic radiography showed change in bone density in the region of ill-defined mandibular angle boundaries, and the computed tomography (CT) showed mixed density image in the mandibular angle, with discreet expansion of cortical vestibular and lingual. Biopsy was performed, and content was aspirated in small quantity and purulent tissue fragments were sent to anatomical–pathological examination. The collected purulent secretion was colored for cytopathologic study, which showed infection by *Actinomyces*.
Discussion In this case, the causative agent was Actinomyces, which makes it even more unusual. The origin of the microorganism has not been clearly established; however, the diagnosis allowed long-term treatment with antibiotics, which has resulted in the resolution of the case.

Keywords *Actinomyces* · Actinomycotic osteomyelitis · Actinomyces-associated lesions · Jaw infection

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Background

Microorganisms classified as *Actinomyces* are common inhabitants of the oral cavity and human pharynx region. Actinomycotic infection in chronic inflammation has specific action on primary soft tissues, rarely affecting the bones [1–3].

Historically, Langenback may have been the first researcher to describe this disease in humans at 1848. In 1891, Israel and Wolf isolated an anaerobic filamentous organism in humans. In 1898, this microorganism received the name *Actinomyces israelii*. *A. israelli* is a gram-positive microorganism, not fixed by acid, and a producer of sulfur granules. Its morphological characteristics vary, being usually filamentous microaerophilic with isolated bacillus organism shapes and can be in several regions, such as the tonsillar region, carious cavity, salivary calculi, dentin, periodontal pockets, and in the oropharynx mucosa and gastrointestinal regions [4–6].

The pathological mechanism of actinomycotic osteomyelitis is unknown. All sites with microorganisms can be the path to infection in soft tissues and/or bones, associated with trauma or a previous nonspecific infection. *A. israelli* is most commonly involved in the pathogenesis of actinomycosis; however, other buccal species of *Actinomyces* may be related to human infection, such as *Actinomyces viscosus*, *Actinomyces naeslundii*, and *Actinomyces odontolyticus* [1, 4, 6]. This article presents an unusual case of mandibular osteomyelitis caused by *Actinomyces*.

Case report

A 19-year-old male patient, Caucasian, was admitted into oral and maxillofacial ambulatory, presenting with pulsating pain and swelling of large intensity in the right parotid-masseteric region, starting 3 days prior (Fig. 1). There was intense trismus and discreet erythema of the skin area. It had not presented with regional lymphadenopathy or febrile episodes. Cutaneous fistulas or intraoral was not present. The patient was referred to self-medication with paracetamol 500 mg+30 mg codeine without any remission of pain, but pain improved with diclofenac 50 mg. There was no history of trauma or recent surgery. The patient reported endodontic treatment on the lower right first molar about 16 months ago and removal of the lower right third molar approximately 3 years prior. The other teeth on that side were vital and without any associated periapical lesion or periodontal pocket.

The panoramic radiography showed change in bone density in the region of the ill-defined mandibular angle boundaries (Fig. 2). The computed tomography (CT) showed mixed density image in the mandibular angle, with discreet expansion of cortical vestibular and lingual (Fig. 3). A biopsy was performed where content was aspirated in a small quantity, and purulent tissue fragments were sent for anatomical–pathological examination. The purulent secretion was colored for cytopathologic study, which showed infection by *Actinomyces* (Fig. 4). The patient was treated with clindamycin 300 mg VO every 6 h, with good response. The drug treatment was extended for 6 months, but in the fourth month, it was stopped spontaneously by the patient who spoke later of epigastric pain. After 2 months of interruption, he presented again with pulsating pain, followed by pulp necrosis of the lower right second molar (Fig. 5). The patient was submitted to endodontic treatment of the tooth in question and restarted drug treatment with amoxicillin 1 g VO every 12 h for 6 months. After 5 years, the patient remained well and without signs of infectious relapse (Fig. 6).

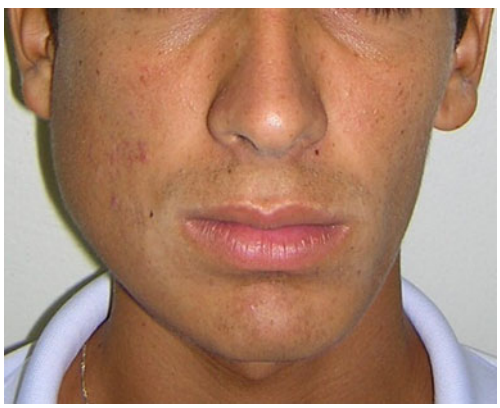

Fig. 1 Initial appearance of patient's infection

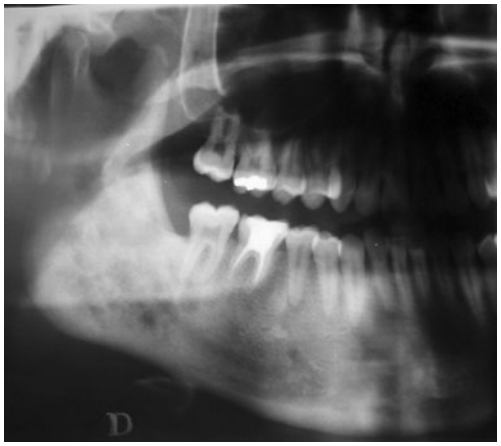

Fig. 2 Panoramic radiography showed changes in bone density in the region of the ill-defined mandibular angle boundaries

Discussion

Primary actinomycotic osteomyelitis is rare, corresponding to about 12 % of cases [6]. It affects the cervicofacial region, typically the body of the mandible, followed by the region of the chin, branch, and angle of the mandible, but rarely affecting the upper jaw or temporomandibular joint. It has prevalence in the mandible in relation to the maxilla of 4:1, as reported in the present case [1–7].

Actinomycotic osteomyelitis of the mandible is a result of the presence of the *Actinomyces* bacteria in the oral cavity, in regions such as the palatine tonsils, gingival fluid, mucosal surfaces, dentin cavities, and sites of post-extraction. The infection manifests especially when the normal composition of oral microbiota is disrupted. Its manifestation leads to a primary chronic infection and, consequently, pathological changes in bone [1, 3, 4].

Actinomycosis develops primarily in certain circumstances, such as injuries caused by dental extractions, jaw injury by trauma, diabetes, immunosuppression, corticoid treatment for extended periods, alcoholism, and smoking. None of these circumstances were observed in the case reported. The area most affected was the head and neck region, corresponding to about 55 % of cases, although there may be also the involvement of the eye, neurological, respiratory, urogenital, and abdominal regions. The age group most frequently affected is between 30 and 60 years; the present case is not included in the age variance described in the literature, with a 4:1 predilection for the masculine gender in relation to female, according to case reported [5–8].

Clinically, the infection can manifest itself as acute or chronic. The acute infection is less common and can manifest as a floating swelling, which resembles an acute odontogenic infection. It can be painful and is associated with temperature rise, having the possibility to spread rapidly in tissues, with reported compatible features in the acute case,

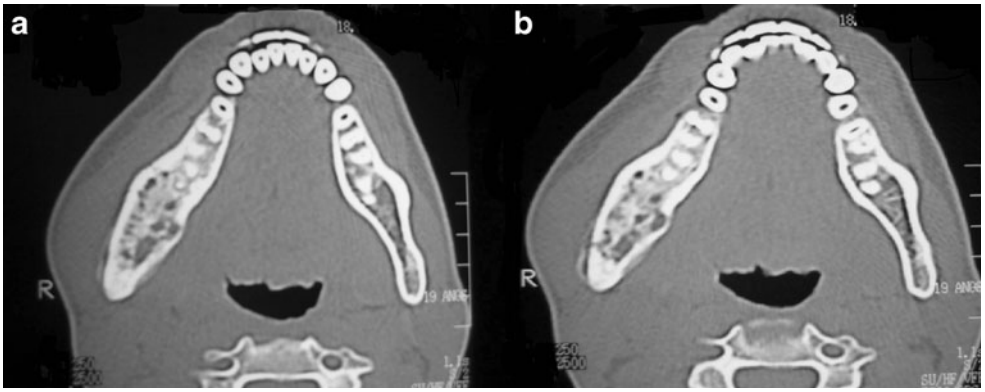

Fig. 3 CT showed mixed density image in the mandibular angle, with discreet expansion of vestibular and lingual cortical regions

except for the presence of febrile episodes [9]. A chronic injury is the more common, with slow and progressive increase in volume. It may or may not exist with the presence of painful symptoms. It is associated with a course or with minimum elevation of temperature, and its development can take weeks, months, or even years, with no changes in the rates of hematological examinations and without patient complaint of malaise [5, 6, 10–12].

Although history and physical examination are essential to the diagnosis of any disease, in the case of osteomyelitis actinomycotic, other diagnostic methods are used, such as image examinations and examination of bacterial culture and cytopathologic assessment of tissues and secretions collected at the infection site [1, 3, 4, 8]. Radiographs can be useful in the infectious process extension recognition in bone but are nonspecific for actinomycosis, even with the presence of edema in the region. The CT and scintigraphy with gallium can be useful in differentiating between inflammatory changes and neoplasms. A CT can be performed to verify the presence of osteolysis and formation of fibrous tissue in an infected region; scintigraphy can determine the effectiveness of therapy; however, no image mode can be used as the only form of diagnosis [1, 4–6].

Confirmation of the diagnosis depends on careful anaerobic culture of these bacteria that are sensitive to oxygen; it should be held preferably if the patient has not received antibiotics from 7 to 10 days in advance of the realization of culture. If the culture is poorly executed, delayed, or suffers interference by a concomitant or recent antibiotic therapy by the patient, diagnosis may remain obscure. Microscopic diagnosis in the specimen training presents actinomycosis referred as sulfur granules [12, 13]. In this case, exfoliative cytology allowed the identification of sulfur granules produced by *Actinomyces*, providing a fast diagnosis of actinomycosis. The cytological exam also detected the *Actinomyces*, surrounded by a great number of neutrophil granulocytes. The culture did not reveal the actual cause of the problem as it would be aerobic and, therefore, would not grow *Actinomyces*.

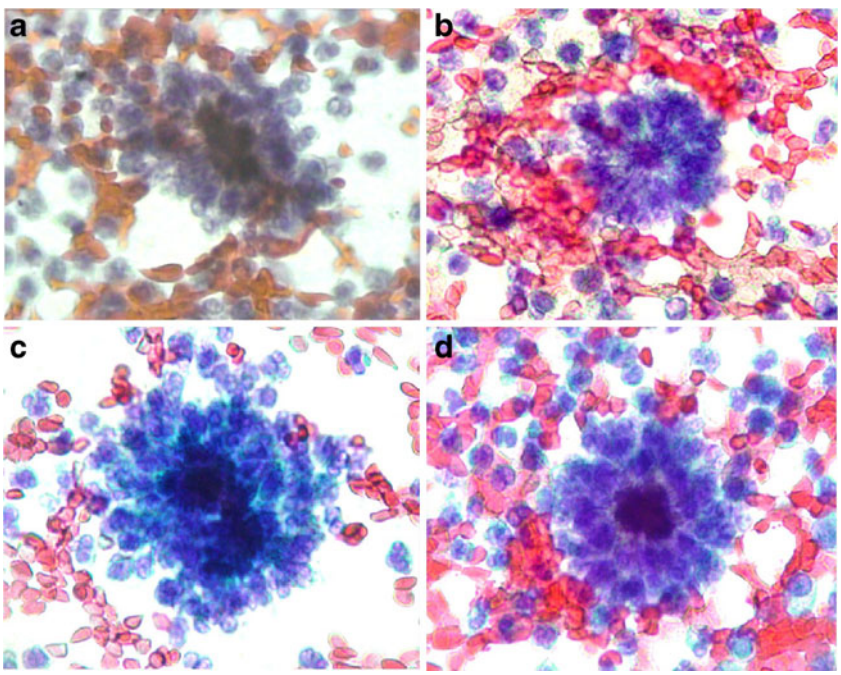

Fig. 4 Chronic inflammation with the presence of sulfur granules and actinomycotic colonies of gram-positive filaments, diagnosed by exfoliative cytology

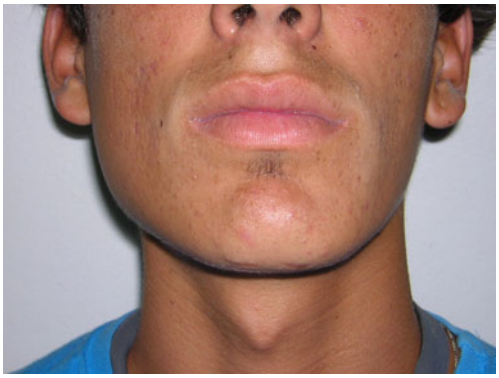

Fig. 5 Appearance of recurrent infection after 2 months of suspension of clindamycin by the patient

The initial treatment for actinomycotic osteomyelitis consists of high doses of penicillin that, depending on the severity of the case, can be administered by intravenous infusion in doses ranging from 3–12 million units daily or oral administration of 2–4 g per day for a period ranging from 3 to 12 months, depending on the response of the host to the infection. According to the described earlier treatment, the oral penicillin was the best choice for the present case [1, 3, 11–14]. Other antibiotics that are effective include clindamycin, erythromycin, chloramphenicol, cephaloridine, minocycline, and imipenem. Metronidazole and aminoglycosides are ineffective against *A. israelii* [5, 6, 14, 15]. Sometimes, it is necessary to curette the region, remove bone sequestration, and refer the patient to maxillofacial reconstruction in cases of substantial loss of bone and soft tissue [1, 3, 6]. In the case reported, there were no necessary surgeries for curettage or reconstruction. After the end of successful therapy, secondary surgical repair or reconstruction may be indicated when needed once the surgeon is confident that the infection is completely eliminated [1].

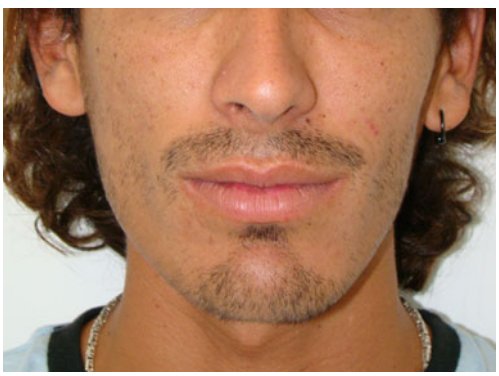

Fig. 6 Final appearance, after 5-year follow-up

Conclusion

Mandibular osteomyelitis is an infection that is challenging to manage due to the poor vascularization of bone that favors the proliferation of microorganisms. In this case, the causative agent was *Actinomyces*, which makes it even more unusual. The origin of the microorganism has not been clearly established; however, the diagnosis allows long-term treatment with antibiotics, which resulted in the resolution of the case.

References

1. Bartkowski SB, Zapala J, Heczko P et al (1998) Actinomycotic osteomyelitis of the mandible: review of 15 cases. J Craniomaxillofac Surg 26:63–67
2. Sharkawy AA (2007) Cervicofacial actinomycosis and mandibular osteomyelitis. Infect Dis Clin N Am 21:543–556
3. Rubin MM, Krost BS (1995) Actinomycosis presenting as a midline palatal defect. J Oral Maxillofac Surg 53:701–703
4. Gupta SD, Gupta MK, Naidu NG (1986) Mandibular osteomyelitis caused by actinomyces Israelii. J Max-fac Surg 14:291–293
5. Nagler R, Peled M, Laufer D (1997) Cervicofacial actinomycosis. A diagnostic challenge. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 83:652–656
6. Goldberg MH (2003) Diagnosis and treatment of cervicofacial actinomycosis. Oral Maxillofacial Surg Clin N Am 15:51–58
7. Finley AM, Beeson MS (2010) Actinomycosis osteomylelitis of the mandible. Am J Emerg Med 28:118.e1–118.e4
8. Bourée P, Bisaro F, Resende P (2009) Actinomycose : du saprophytisme à la pathogénicité. Antibiotiques 1–8
9. Nielsen PM, Novak A (1987) Acute cervicofacial actinomycosis. Int J Oral Maxillofac Surg 16:440–4
10. Miller M, Haddad AJ (1998) Cervicofacial actinomycosis. Oral Surg Oral Med Oral Pathol 85:496–508
11. Topazian RG (2006) Osteomyelitis of the jaws. In: Topazian RG, Goldberg MH, Hupp JR (eds) Oral and maxillofacial infections, 4th edn. WB Saunders Co, Philadelphia, pp 214–42
12. Rothschild B, Naples V, Barbian L (2006) Bone manifestations of actinomycosis. Ann Diagn Pathol 10:24–27
13. Das DK (1994) Actinomycosis in fine-needle aspiration cytology. Cytopathology 5:243–50
14. Flynn TR, Halpern LR (2003) Antibiotic selection in head and neck infections. Oral Maxillofacial Surg Clin N Am 15:17–38
15. Goldberg MH (2001) Antibiotics: old friends and new acquaintances. Oral Maxillofacial Surg Clin N Am 13:15–30