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Actinomycosis in the maxilla of a young asymptomatic patient: unusual case report

Actinomicose em maxila de um paciente jovem assintomático: relato de caso incomum

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ABSTRACT

The aim of this study is to report an unusual case of actinomycosis in the maxilla region of an asymptomatic patient. A 21-year-old white man was referred for the analysis of panoramic radiography and cone beam computed tomography, where it was observed the presence of a hypodense lesion on the left side of the maxilla, which extended from the maxillary left central incisor to the region of the maxillary left second premolar. During intraoral examination, a depression was observed in the hard palate mucosa, as well as a fistula in the alveolar mucosa close to maxillary left central incisor, which had pulp vitality confirmed by thermal tests. A fistulography was performed, with periapical radiography, where it was found that the fistulous path did not originate from the tooth mentioned above. An incisional biopsy was performed for diagnostic purposes. However, given the inconclusive microscopic findings, four months later, a new biopsy was performed. The histopathological examination revealed the presence of a colony of microoganisms with filamentous pattern of radiated rosette, surrounded by polymorphonuclear inflammatory cells. Based on the morphological characteristics, the diagnosis of actinomycosis was established. The treatment was based on antibiotic therapy. Six months after treatment of the infection, no signs of recurrence were observed, and the patient remains in follow-up. Actinomycosis in the maxilla is an uncommon infection with a predilection for males between the ages of 20 and 60, whose treatment is antibiotic therapy associated or not with surgical excision.

Indexing terms: Actinomycosis. Bacterial infections. Maxillary diseases. Pathology. Surgery, oral.

RESUMO

O objetivo deste estudo é relatar um caso incomum de actinomicose em região de maxila de um paciente assintomático. Um homem de 21 anos, branco, foi encaminhado para análise de radiografia panorâmica e tomografia computadorizada de feixe cônico, onde

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foi observada a presença de lesão hipodensa do lado esquerdo da maxila, que se estendia da região do incisivo central superior esquerdo ao segundo pré-molar superior esquerdo. Ao exame intrabucal, observou-se depressão na mucosa do palato duro, bem como fístula na mucosa alveolar próxima ao incisivo central superior esquerdo, no qual tinha vitalidade pulpar confirmada pelos testes térmicos. Uma fistulografia com radiografia periapical foi realizada, onde foi observado que o trajeto fistuloso não era originado do dente mencionado anteriormente. Foi realizada biópsia incisional para fins diagnósticos. Porém, diante dos achados microscópicos inconclusivos, quatro meses depois, uma nova biópsia foi realizada. O exame histopatológico revelou a presença de colônia de microrganismos com padrão filamentoso de roseta irradiada, circundado por células inflamatórias polimorfonucleares. Com base nas características morfológicas, foi estabelecido o diagnóstico de actinomicose. O tratamento foi baseado em antibioticoterapia. Seis meses após o tratamento da infecção, não foram observados sinais de recidiva e o paciente permanece em acompanhamento. A actinomicose da maxila é uma infecção incomum, com predileção por pacientes do sexo masculino com idade entre 20 e 60 anos, cujo tratamento é antibioticoterapia associada ou não à excisão cirúrgica.

Termos de indexação: *Actinomicose. Infecções bacterianas. Doenças maxilares. Patologia. Cirurgia bucal.*

INTRODUCTION

Actinomycosis is a subacute to chronic infection caused by filamentous Gram-positive bacteria, anaerobic [1,2], non-acidic [1,2] and without spores. This disease has, clinically, four types: thoracic, abdominopelvic, cerebral and cervicofacial [2]. The latter usually affects the body of the jaw, followed by the mental region and angle of the jaw, but rarely affects the maxilla or the temporomandibular joint [3].

Actinomycosis in the maxilla accounts for only 0.5% to 9% of all head and neck cases [4], with periapical actinomycosis being one of the rarest forms of actinomycosis occurring in the maxillofacial region [5]. The pathogenesis of cervicofacial actinomycosis is not exactly known, however, dental caries and trauma seem to be favorable factors to the development of the disease [4,6]. Loss of mucosal integrity caused by tooth extractions, periodontal disease [4,6], local anesthesia, bone or dental fractures, tooth eruption, pulp exposure or even endodontic treatment [6] and non-vital teeth can function as a gateway for these bacteria, and thus, start the infection [4].

The most common clinical presentation of actinomycosis is soft tissue abscess and drainage of cervical fistulas [4,7]. The skin that covers the abscess is red to purplish and hardened on palpation [7]. The discovery of "holes" or large defects of the jaw bone within the oral cavity is a relatively unusual presentation of cervicofacial actinomycosis, when it results in osteomyelitis [3,4].

Actinomycosis is termed "masker" in the head and neck region due to its unusual presentations [4], because it often mimics a malignant lesion or a granulomatous lesion [8]. Therefore, the final diagnosis is usually reached only after surgical removal of the lesion and the histopathological examination of the sample. Long-term high-dose penicillin is needed for treatment [4,6], which can vary from four weeks to one year, based on the severity of the disease.

Given the above, the objective of the present study is to report an unusual case of actinomycosis in the maxilla region of an asymptomatic young patient.

CASE REPORT

A 21-year-old white man was referred by an orthodontist for the analysis of cone-beam computed tomography, in which significant imaging alterations were observed. During the anamnesis, the patient reported that there had been no previous use of bone modifying agents. The extraoral physical examination did not reveal any changes. On intraoral physical examination, a soft and resilient depression was observed in the mucosa of the hard palate, in addition to a parulis in the alveolar mucosa at the height of the maxillary left central incisor, which had pulp vitality confirmed by thermal tests. A fistulography was performed, with periapical radiography, where it was found that the fistulous path did not originate from the tooth mentioned above.

In cone beam computed tomography, a hypodense image was observed, extending from the region of the maxillary left central incisor to the region of maxillary left second premolar, with regular margins and well-defined limits.

In addition, it was also possible to observe the withdrawal of the roots of maxillary left central incisor, maxillary left lateral incisor and maxillary left canine, in absence of root resorption (figure 1A); compression of the anterior wall of the maxillary sinus (figure 1B) and expansion and rupture of the vestibular and palatal cortex of the incisive canal (figure 1C).

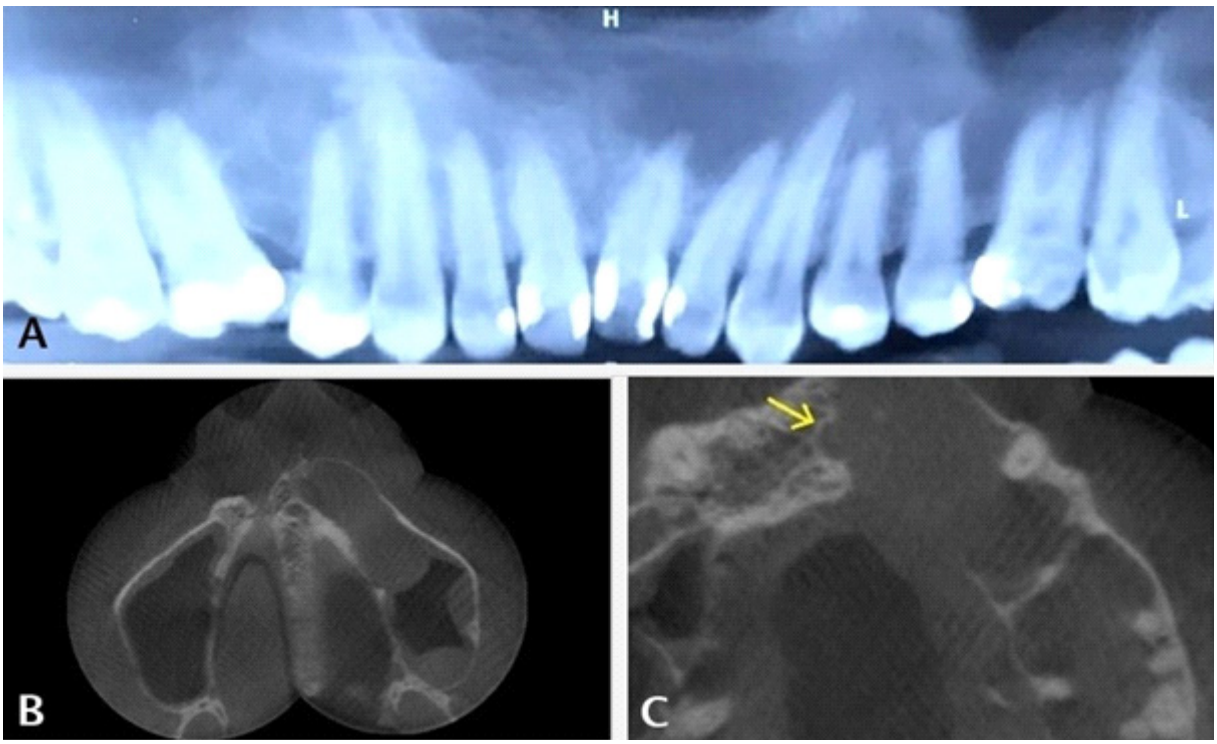

Figure 1. A: Cone beam computed tomography in panoramic reconstruction. To observe hypodense image in the maxilla, on the left side, of teeth 21 to 25. B: Cone beam computed tomography, in axial section, showing the extent of the lesion. C: Axial section on a cone beam computed tomography showing a rupture of the incisive canal cortex.

Under the diagnostic hypothesis of a cyst of odontogenic nature, an intraosseous incisional biopsy was chosen to be performed, with access through the palatal region. In view of the inconclusive microscopic findings, monitoring of the patient for four months was recommended, to then perform a new biopsy, which was performed after evaluation of a tomographic examination, which revealed the stagnation of the lesion, without increasing the extent of bone destruction. In the second biopsy, a partial resection of the lesion was performed. At this time, a Partsch type incision was made in the alveolar mucosa, vestibule background, followed by a bone window in the left hemimaxila, to access the lesion (figures 2A and 2B). Then, the bone cavity was curetted, and fragments of the lesion were removed followed by the overlying mucosa suture (figures 2C and 2D).

Histopathological analysis of soft tissue fragments showed the presence of connective tissue which varied in density, with collagen fibers randomly arranged, permeated by fusiform and ovoid fibroblasts, in addition to inflammatory infiltrate, predominantly lymphoplasmacytic, and blood vessels, of various calibers and congested by red blood cells, with hemorrhagic areas. In the focal area, there was a microorganism colony with a filamentous pattern of radiated rosette, whose central portions were basophilic and the peripheral ones, eosinophilic, surrounded predominantly by polymorphonuclear cells (figure 3A).

Regarding the analysis of the histological sections of the bone pieces, it was possible to verify, on the periphery of the specimen, trabecular bone with normal aspect. In addition, there was an intense presence of foamy macrophages,

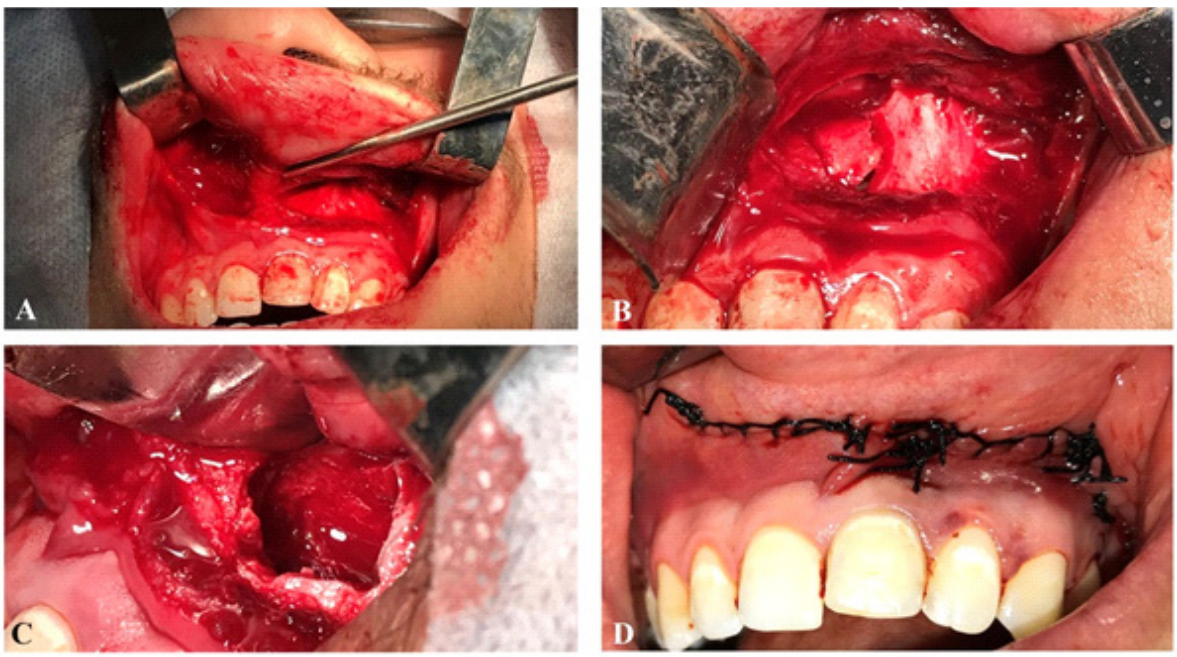

Figure 2. A: Incision in the mucosa. B: Opening the bone window. C: Access to the injury. D: Suture (3-0 silk thread).

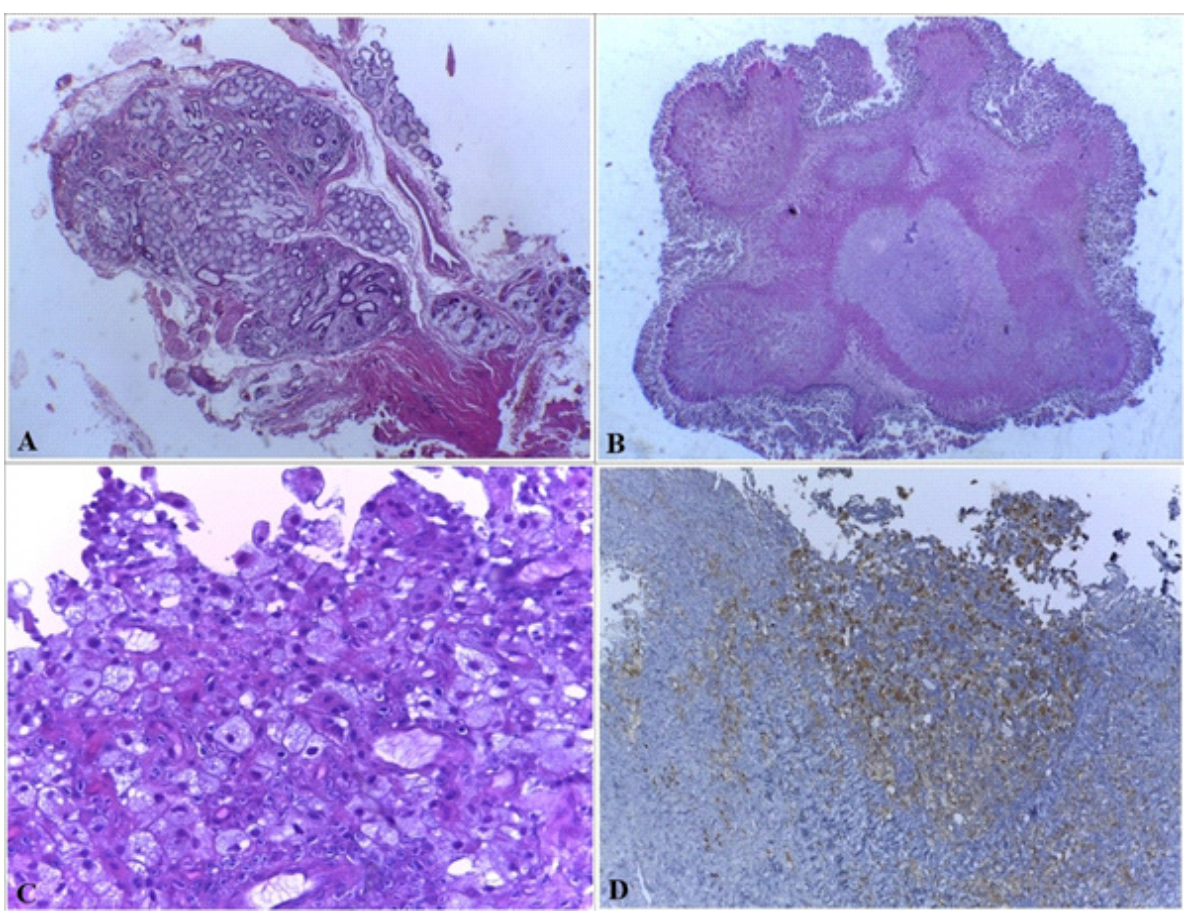

Figure 3. A: Photomicrograph showing normal salivary glandular parenchyma (Hematoxylin and eosin – H/ E, 40x). B: Colony of microorganisms with filamentous pattern of radiated rosette, with central basophilic and peripheral eosinophilic portions, predominantly surrounded by polymorphonuclear cells (H/ E, 40x). C: Intense presence of foamy macrophages, endothelial cells, and rare multinucleated giant cells (H/ E, 400x). D: Photomicrograph demonstrating macrophage immunopositivity for CD68 (100x).

endothelial cells, and few multinucleated giant cells. In these areas, cells with altered morphology and some mitosis figures were observed (figures 3B and 3C). In order to rule out the possibility of being a neoplastic lesion, the immunohistochemical marker CD68 was used, which demonstrated immunopositivity for foamy macrophages (figure 3D).

The diagnosis of actinomycosis was established, and the chosen treatment was antibiotic therapy with Amoxicillin (875mg) associated with Clavulanic Acid (125mg) (Generic Medication Law No. 9,787, 1999), for 30 days, one tablet every 12 hours. The antibiotic time was reduced as a result of the surgical intervention performed previously. After finishing the drug therapy, a panoramic radiography was performed, indicating slight regression of the lesion. Therefore, the patient was instructed to attend periodically.

Upon patient's return, four months later, it was observed that the maxillary left central incisor had extensive provisional restoration and compromised marginal sealing, as well as the maxillary left lateral incisor, which presented unsatisfactory restoration. Sensitivity tests were performed to evaluate the pulp vitality of both teeth, and both showed a negative response to the tests. Periapical radiography was performed, and the lesion was located at the root apex of these two teeth. In view of this, endodontic treatment of both teeth was requested, and, after the endodontic treatments were completed, total lesion regression was observed (figure 4). Six months after the infection treatment, no clinical or radiographic signs of recurrence were observed, and the patient remains in follow-up.

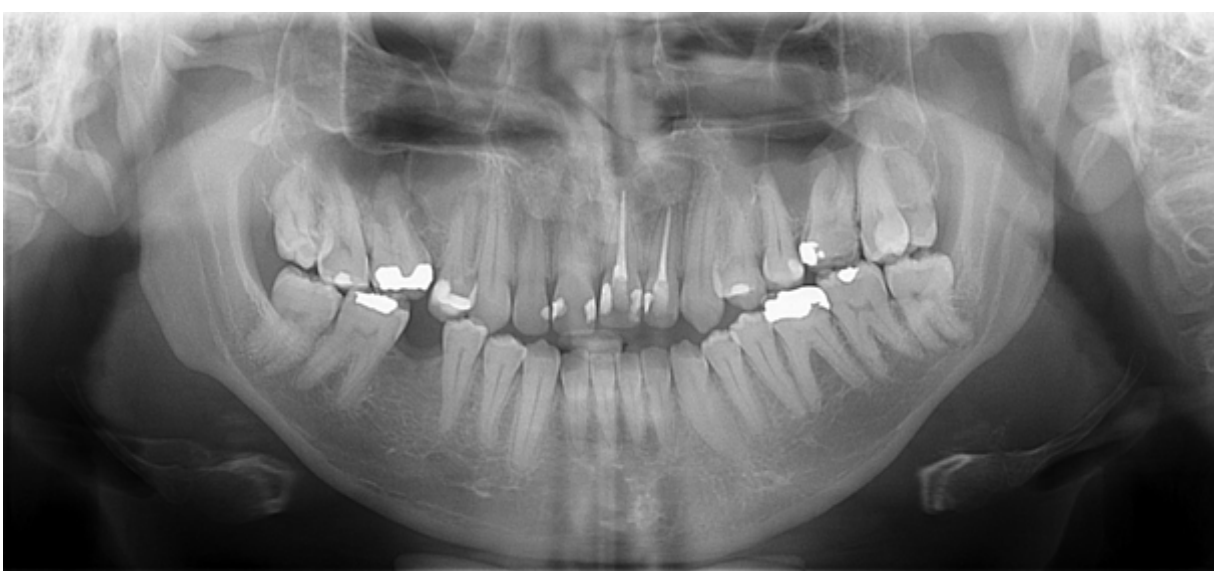

Figure 4. Panoramic radiograph showing total regression of the lesions that were at the apexes of teeth 21 and 22.

DISCUSSION

Actinomycosis is a rare granulomatous suppurative disease caused by Actinomyces spp [3,4], originated from the Greek word "Aktino", which means radiant appearance of sulfur granules, and "Mykos", which labels the condition as mycotic disease [1,3]. However, actinomycetes are now grouped as bacteria, due to their composition in the cell wall, lack of nuclear membrane and lack of growth inhibition by antifungal agents [4]. Like fungi, these bacteria form a mycelial network of branched filaments, but like bacteria, they are thin, have cell walls containing muramic acid and are susceptible to antibiotics [3].

Actinomyces can live in the oropharynx, gastrointestinal tract and urogenital tract as a normal microbiota [9], acting as commensals [1]. The most important species involved in clinical infection include Actinomyces viscosus, Actinomyces naeslundi, Actinomyces odontolyticus e Actinomyces israelii [4], the latter being the most prevalent [9].

Typically, these bacteria have low potential for pathogenicity or invasion [1], but they become pathological when gaining access to subcutaneous tissues. In addition, the infection is polymicrobial, with up to five to 10 other bacterial species present [10], like Staphylococcus and Streptococcus [4]. These associated bacteria seem to increase the low pathogenic potential of actinomycetes [4], working synergistically to form a specific ecosystem with low oxidation reduction potential favorable to anaerobic growth [11].

In cases of actinomycosis in the apical region, the source of infection can be intraradicular biofilm or extraradicular bacterial aggregations, such as sulfur granules [9]. In addition to local risk factors for the development of infections, diabetes mellitus, alcohol use disorders, malnutrition, malignancies, HIV+ patients, transplantation of solid organs such as lungs and kidneys, biological agents such as infliximab and acute lymphoblastic leukemia treated with chemotherapy are other known risk factors [10]. In the case reported here, the patient did not have any systemic factors that could trigger an infection, and one of the teeth associated with the lesion, confirmed with fistulography, showed positive response to pulp sensitivity tests. In addition, the patient had no history of trauma or soft tissue lesion.

Cervicofacial actinomycosis is a relatively rare condition worldwide, without any predilection for age, race [6] and sex. In the study by Pulverer et al. [12], aiming to evaluate microbiological and clinical data from 1997 cases of human cervicofacial actinomycosis, a predisposition of male patients was observed, which varied with age and it seemed to be especially pronounced in patients aged from 20 to 60 years, with a higher incidence found in female patients aged from 11 to 40 years and in male patients aged from 21 to 50 years old. This infection usually involves tissues around the maxilla or mandible, including the mandible itself in approximately 50% of cases, buccal mucosa (15%), mentum (15%) and branch and angle of the mandible (10%) [6]. Few cases in the literature report maxillary involvement [1,11,13-29] (table 1). In the present case, the patient was male, 21 years old and the lesion was present in the anterior region of the maxilla. Although the patient in the present case was quite young, two cases of 16-year-old patients have already been reported [13,14].

Table 1. Cases of actinomycosis involving the maxilla reported in the literature.

1 of 2

Authors/ year	Age (yr)/ sex	Duration	Condition	Signs and symptoms	Treatment
Main & Macphee [13]	16/ Male	3 weeks	Normal	Mass of hyperplastic tissue buccal to the upper second molar and detachable from the tooth. This tooth was slightly mobile but in full functional use.	Excisional biopsy and 600,000 units of penicillin, administered 1 hour prior to operation; phenoxymethylpenicillin, one 250 mg. tablet every 4 hours.
Musser et al. [14]	16/ Female	-	Normal	Clinical examination disclosed a nontender, spongy swelling in the anterior maxilla that appeared to involve soft, tissue. The area extended from the right lateral incisor to the left canine in the maxilla.	Surgery; tetracycline.
Fergus & Savord [15]	23/ Male	1 year	-	A raised yellow swelling in the anterior left maxilla, which was tender to palpation.	Surgery and prophylactic dose of 250 mg. phenoxymethyl penicillin four times a day for 1 week.
Rubin & Krost [16]	58/ Male	-	Cocaine snorting	Nasal congestion, yellow sputum, and purulence	Intravenous aqueous penicillin + oral clindamycin; intravenous penicillin
Herman et al. [17]	65/ Female	2 weeks	Chronic lymphocytic leukemia	Pain, malaise, generalized prostration, difficulty eating and swallowing	Intravenous penicillin
Tarner et al. [18]	49/ Male	-	Vasculitis involving the lungs and brain	Mucosal defect of the palate representing the orifice of a fistula between the oral and nasal cavities was found in the area of tooth 23 and 24, which had been extracted without antibiotic prophylaxis 6 months earlier.	-

Table 1. Cases of actinomycosis involving the maxilla reported in the literature.

Authors/ year	Age (yr)/ sex	Duration	Condition	Signs and symptoms	Treatment
Crossman & Herold [19]	85/ Male	-	Normal	Bony sequestra bilaterally affecting the upper premolar regions	Oral clindamycin
De et al. [20]	32/ Male	2 years	Normal	-	Intravenous crystalline penicillin G
Yadegarynia et al. [21]	48/ Female	8 months	Normal	An indurated area on the hard palate and the overlying skin became reddish. Abscess-like lesions eventually draining to the surface of her developed, which gradually led to a spontaneous discharging sinus. The patient also had post-nasal drip. A discharging sinus on the mid-line of the anterior hard palate with slight deviation to the right.	Biopsy and oral ampicillin 500 mg four times a day. It was planned to continue the treatment for 6 months.
de Andrade et al. [22]	46/ Female	4 days	Diabetes mellitus	Dysphagia and pain	penicillin
Jamshidi et al. [23]	40/ Female	More than a year	Normal	Persistent sinus tract discharge. In clinical examination, the cutaneous lesion expressed a purulent discharge	Osteotomy and apicoectomy of mesiobuccal, distobuccal, and palatal roots of maxillary right first molar were performed and retropreparations were made
Suhasini et al. [1]	19/ Female	2 years	History of trauma in the region	Oblique fracture involving incisal third toward distal side seen with slight discoloration in relation to maxillary right central incisor. Swelling measuring about 3 cm × 3 cm in the palatal aspect of maxillary right central incisor-maxillary right second premolar, extending anteroposterior and mediolaterally 0.5 cm crossing away the mid-palatine raphe and 1 cm away from the marginal gingiva of premolar (palatal) was seen.	Apical surgery and Systemic antibiotics (amoxicillin; Amox 500 mg 3 times daily 5 days) and analgesics (ibuprofen; brufen 400 mg twice daily for 3 days)
Klein et al. [24]	31/ Male	2 months	HIV-positive diagnosis with secondary syphilis, pneumocystis pneumonia, and retinitis caused	A far-reaching destructive lesion on the right maxilla. Necrotic gingiva and alveolar mucosa exposed bone and roots of maxillary right canine and maxillary right first molar.	5,000,000 IU of penicillin G crystalline IV for three weeks supplemented with amoxicillin 500 mg orally administered every 8 h for six months.
Dahiya et al. [25]	79/ Female	10 months	Diabetic and hypertensive; history of carcinoma of breast detected 1½ years back with postoperative renal failure needing dialysis.	A large 2 × 2 cm ulcer with bone erosion involving anterior half of midline of the hard palate more on the right side with oroantral and oronasal communications. It was covered with slough at the edges and blackish necrotic tissue was present in the surrounding areas	Crystalline penicillin and oral amoxicillin 500mg thrice daily for 12 months
Gurbanov & Torul [26]	30/ Female	1.5 months	Previous exodontics in the region	Painful, bleeding prone area with necrotic bone in the left side of the hard palate	Excision of the necrotic bone and surrounding soft tissues; Penicillin and analgesic.
Jangla et al. [27]	78/ Female	6 months	Trauma to left side tooth which was self-inflicted using a wooden stick.	A lump on the left side of the upper jaw and difficulty in opening the mouth; ulcer in the mouth at the same site of the trauma with whitish discharge	Biopsy and amoxycillin-clavulanic acid for eight days; ampicillin-sulbactam and doxycycline for four weeks
Ahmedbrahim et al. [28]	70/ Male	6 months	Smoker and had been diagnosed with epidermoid carcinoma of the lung 1 year previously, which was in remission after radio chemotherapy treatment.	Extensive ulcerative lesion on the hard palate that measured approximately 4 cm in diameter with yellow pseudomembrane deposit and posterior erythema on the soft palate.	Oral penicillin

Table 1. Cases of actinomycosis involving the maxilla reported in the literature.

Authors/ year	Age (yr)/ sex	Duration	Condition	Signs and symptoms	Treatment
Samanta & Paul [29]	36/ Male	3 years	-	Maxillary left first molar was having grade III with furcation involvement on the buccal aspect along with distobuccal root exposure.	Tooth extraction and histopathological analysis of the soft tissue involved the tooth; Amoxycillin (875 mg) plus clavulanic acid (125 mg) twice a day for a period of 6 weeks, Metronidazole (400 mg) thrice a day for 5 days along with analgesics for 5 days, which was further prolonged for another continuous five weeks.
Present case	21/ Male	-	Normal	Soft and resilient depression was observed in the mucosa of the hard palate, in addition to a parulis in the alveolar mucosa at the height of the maxillary left central incisor	Resection of the lesion and Amoxicillin (875mg) associated with Clavulanic Acid (125mg), for 30 days, one tablet every 12 hours.

Clinically, cervical actinomycosis presents as a significant increase in volume associated with external drainage ulcers, fistulas and, occasionally, sulfur granules [10]. In addition, it is asymptomatic, hardened on palpation and may present a reddish to purplish color, which evolves to multiple abscesses [10]. When located in the maxilla region, the authors report, in their cases, as ulcerated areas involving bone destruction [11,22], which may be associated with areas of multiple bone sequestrations exposed in the oral cavity [19]. In its occurrence, when symptomatic, in the periapical form, it presents as a persistent and recurrent drainage fistula in the periapical region [5]. In the present case, the patient did not present painful symptoms or any increase in volume, despite the extent of the lesion. On the other hand, a parulis was observed in the region above maxillary left central incisor, which was vital by pulp sensitivity tests.

The imaging findings are nonspecific and do not contribute to the diagnosis of the disease but will help to assess the degree of involvement of soft tissues and bones. Periapical radiographs are useful in assessing apical abscesses [10]. In the case reported here, panoramic radiography and cone beam computed tomography were performed for orthodontic purposes, being the lesion discovered accidentally. A fistulography was performed, with periapical radiography, where it was found that the fistulous path did not originate from the tooth mentioned above. The other imaging exams were essential for knowing the extent of the lesion and planning the surgery.

The diagnosis of infection is made based on clinical manifestations, associated with microbiological and histopathological findings [30]. In the case reported here, clinical, imaging, and histopathological findings were used to close the diagnosis due to the initial diagnostic hypotheses. A fact also to be considered is the difficulty of conducting the cultivation of bacterial species in an anaerobic environment for subsequent microbiological analysis.

Microscopically, it is possible to observe a zone of granulation tissue that consists of collagen fibers around central purulent loculations containing abundant neutrophils [4,22]. In the central part, bacterial colonies are observed, with small basophilic heads called conidiophores towards the center and thin eosinophilic hyphae radiating outwards towards the periphery [4]. In the present case, this characteristic pattern was observed and to discard the hypothesis of neoplastic lesion, the immunohistochemical marker CD68 was used, which was positive for foamy macrophages.

For the treatment of actinomycosis, antibiotic therapy with 6 to 12 million IU penicillin is recommended, which can vary from four weeks to one year, based on the severity of the disease [8]. In addition, drainage of abscesses, debridement and surgical excision of the sinus tract is recommended to increase the penetration of antibiotics [4].

Surgical treatment without antibiotic therapy is associated with recurrence. Although good responses have been reported with combined surgical treatment and short-term antibiotic therapy for cervicofacial actinomycosis, surgical

treatment is associated with morbidity, especially with extensive lesions [8], therefore, the need for surgical treatment associated with antibiotic therapy must be carefully considered. In the present case, curettage of the lesion associated with the prescription of 30 days of amoxicillin (875 mg) associated with clavulanic acid (125 mg) was performed. After this period, no signs of recurrence were observed, and the patient remains in follow-up. However, even though surgery was effective in the treatment and essential to reduce the time of antibiotic therapy, probably led to necrosis of the involved teeth.

It is concluded that the present case was of a young patient with actinomycosis in the maxilla without clear evidence of infection, whose treatment of choice was surgery associated with antibiotic therapy. Surgical treatment favored a reduction in the time of antibiotic use and this association promoted an excellent recovery. Despite this, the surgery led to necrosis of the involved teeth and endodontic treatments were performed.

Collaborators

TJ Silva Filho and DQC Gomes, conception and design of the case report. JLSH Pereira, RCV Souza, TJ Silva Filho and DQC Gomes, clinical management of the patient. DFB Silva and TJ Silva Filho, analysis and interpretation of radiographic and histopathological data. JLSH Pereira, DFB Silva and IJ Dias, article writing. TJ Silva Filho, RCV Souza and DQC Gomes, critical revision of the text.

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