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Actinomycotic Osteomyelitis and Myiasis in Oroantral Communication: Clinicopathological and Tomographic Findings

Osteomielitis Actinomicótica y Miasis en Comunicación Oroantral: Hallazgos Clinicopatológicos y Tomográficos

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ABSTRACT: Actinomycotic osteomyelitis of the maxilla presenting with oroantral communication is very rare, herein we report the first case of this condition in association with myiasis. A 50-year-old man reported chronic sinusopathy and a non-healing maxillary lesion, with 30 years of evolution, presenting occasional nasal and intraoral purulent discharge, with foul smell, and recurrent episodes of larvae presence. Cone beam computed tomography showed a large hyperdense image inside the left maxillary sinus, with focal areas with soft tissue density, and extensive discontinuity of the maxillary sinus floor, confirming the oroantral fistula. The necrotic tissue curetted during surgery presented hard consistency, and dark greenish color, and was submitted for histopathological analysis. Microscopically, necrotic bone, masses of filamentous bacteria colonies, compatible with actinomycosis, and large rhomboidal structures surrounded by eosinophilic capsule - suggestive of larvae, were observed. The diagnosis of actinomycotic osteomyelitis with presence of structures compatible with larvae was established.

KEY WORDS: actinomyces, maxilla, myiasis, oroantral fistula, osteomyelitis.

INTRODUCTION

Rupture of the mucous membrane of the maxillary sinus by dental surgery, oral trauma, or a preceding infection favors the access of commensal and anaerobic microorganisms to the antral regions. Actinomyces, commonly A. israelli, can produce chronic, resistant and slowly progressing infections, particularly in soft tissues (Kaplan *et al*., 2009; Sezer *et al*., 2017). Bone involvement by Actinomyces is rare, usually representing a secondary infection. Primary bone infection by actinomyces, actinomycotic osteomyelitis, has been rarely reported in jaws and can occur in immunocompromised patients (Gannepalli *et al*., 2015; Sezer *et al*., 2017; Agarwal *et al*., 2019).

Actinomycotic osteomyelitis is more common in the mandible than in the maxilla, possibly due the higher vascularization of the last (Crossman & Herold, 2009; Gannepalli *et al*., 2015). Despite being a curable infectious process, if not treated early, can cause extensive destruction of the affected tissues. Clinically, it is characterized by a prolonged chronic inflammatory process, presenting as a tumor-like mass or as suppurative areas with continuous destruction of the affected tissues (Kaplan *et al*., 2009; Gannepalli *et al*., 2015).

Myiasis is uncommon in the nasal and oral regions, but it is occurrence may be favored by

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preexisting tissue exposure or destruction (Manjunath & Pinto, 2018) as dental extraction wounds, and ulcerative lesions as carcinoma, especially in patients with psychiatric or disabling conditions, associated with lip incompetence, and poor oral hygiene (Aggarwal *et al*., 2014).

We herein present a rare case of actinomycotic osteomyelitis of the maxilla associated with myiasis, causing oroantral communication in a middle-aged male. To date, there are few cases reported of actinomycosis of the maxilla presenting with oroantral communication, and to the best of our knowledge, its association with myiasis, has not been reported in the jawbones.

CASE REPORT

A 50-year-old man attended the Oral and Maxillofacial Surgery and Traumatology service of the Governador Valadares Hospital, Minas Gerais, Brazil, with the main complaint of "foul smell in the mouth and nose". He reported a non-healing maxillary lesion, with 30 years of evolution, presenting occasional nasal and intraoral purulent discharge. Besides chronic sinusopathy, the patient informed recurrent episodes of larvae presence in the region, which were not treated. History included multiple tooth extractions, low socioeconomic status, and social isolation due to the smell. During anamnesis, hypernasal speech was noted. He was in good general health, and did not use medications, except amoxicillin, whenever he noted pus discharge and odor.

Extraoral examination revealed a discrete proptosis, and greenish, necrotic and purulent tissue was visualized in the nostrils. Intraoral examination showed a painless and extensive defect affecting the left maxilla, involving the alveolar ridge and palate with evident oroantral communication. Necrosis and purulent discharge were observed (Fig. 1A). The patient presented an edentulous maxilla. Based on the history and clinical findings, a provisional diagnosis of chronic inflammatory/infectious process was suggested.

Panoramic radiograph showed a mixed lesion with radiopaque predominance in the left maxillary sinus region, as well as discontinuity of the left upper alveolar ridge (Fig. 2A). Cone beam computed tomography confirmed a large hyperdense image inside the left maxillary sinus, compatible with calcified material, presenting areas of soft tissue density permeating and around the hyperdense mass. Extensive discontinuity of the maxillary sinus floor was observed, indicating an oroantral fistula. Additional

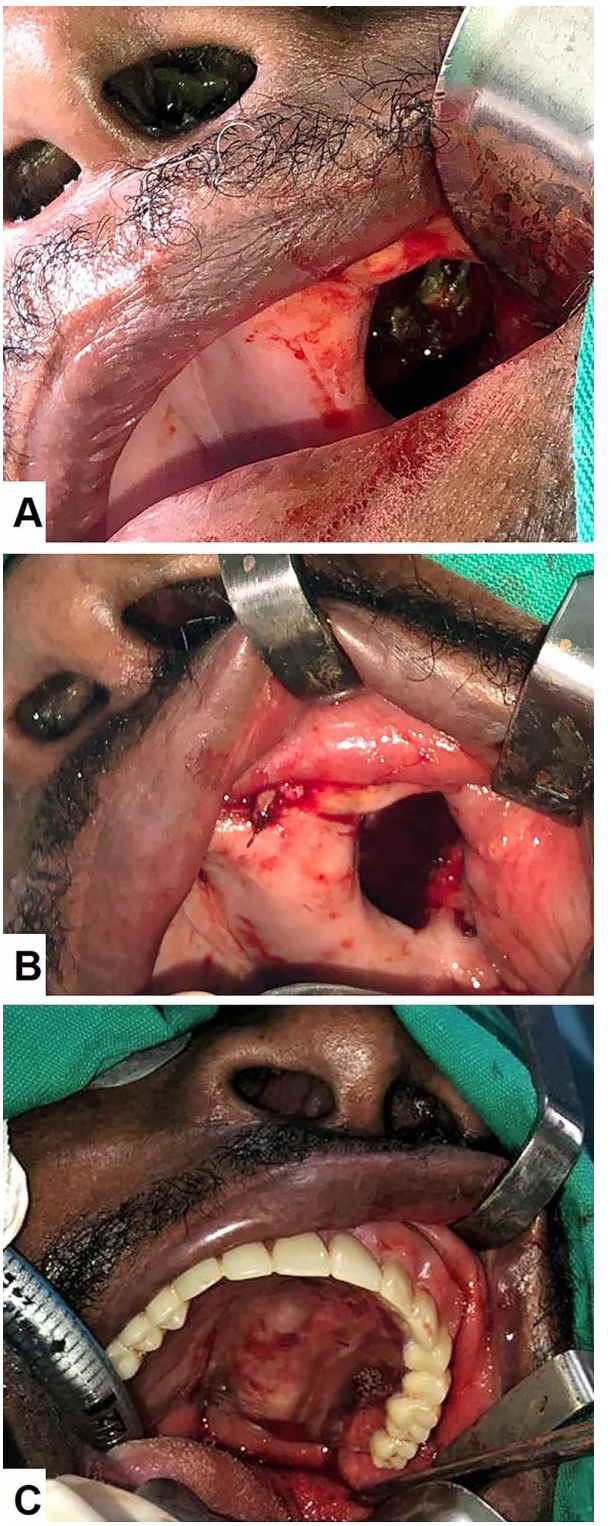

Fig. 1. Clinical presentation before and after surgery. A: Greenish necrotic tissue was visualized in the nostrils and maxillary defect. B: Intraoral extension of the painless defect affecting the left maxilla (alveolar ridge and palate), presenting oroantral communication. C: The prosthesis was installed immediately after surgery.

findings were expansion of the left maxillary sinus walls, with discontinuity, thickening and sclerosis areas, compatible with an intraosseous chronic inflammatory process (osteomyelitis). The lesion extended medially with displacement of the nasal septum to the right, and invasion of the lower and left posterior nasal turbinates, through the soft and hard palate. Additionally, it was possible to observe impairment of ethmoid air cells and discontinuity of the left orbit floor, with involvement of the superior orbital fissure, optical foramen and left infraorbital foramen, as well as displacement of the orbital structures to the left. There was also bone thickening and sclerosis with circumferential involvement of the left carotid foramen, smaller sphenoid wing, base and lateral walls of the sella turcica and anterior portion of the clivus and left zygomatic bone (Figs. 2B and 2C).

The patient was instructed to wash the region with 0.12 % chlorhexidine and to use oral amoxicillin for 3 days. Surgery was planned followed by the installation of a prosthesis to prevent displacement of water, food and saliva to the maxillary sinus that also would facilitate cleaning.

At time of surgery, all lab tests were normal, including blood count, platelets, glucose (93.9 mg/dl), creatinine (0.89), urea (28 mg/dl), leukogram (5.300 p/mm3). Intraoral evaluation showed a necrotic area without pus (Figs. 1A and 1B). Under general anesthesia and orotracheal intubation, the patient received 2g of intravenous cephalothin as prophylaxis. Total flap incision and full-thickness flap with total exposure of the maxilla and zygomatic complex was realized. Curettage and sinusectomy with abundant irrigation with chlorhexidine (0.12 %) was performed to remove all calcified and necrotic tissues as well as the epithelium covering the antral communication. During surgery, it was observed that orbital musculature was preserved with a well-conditioned fat pad, without the need of infraorbital support. After fixation and sutures, the prosthesis was installed immediately (Fig. 1C). Cephalothin 1g was maintained during hospitalization.

The necrotic tissue curetted during surgery was submitted for histopathological analysis, comprising several irregular necrotic tissue fragments, of hard consistency, and dark greenish color. The decalcified

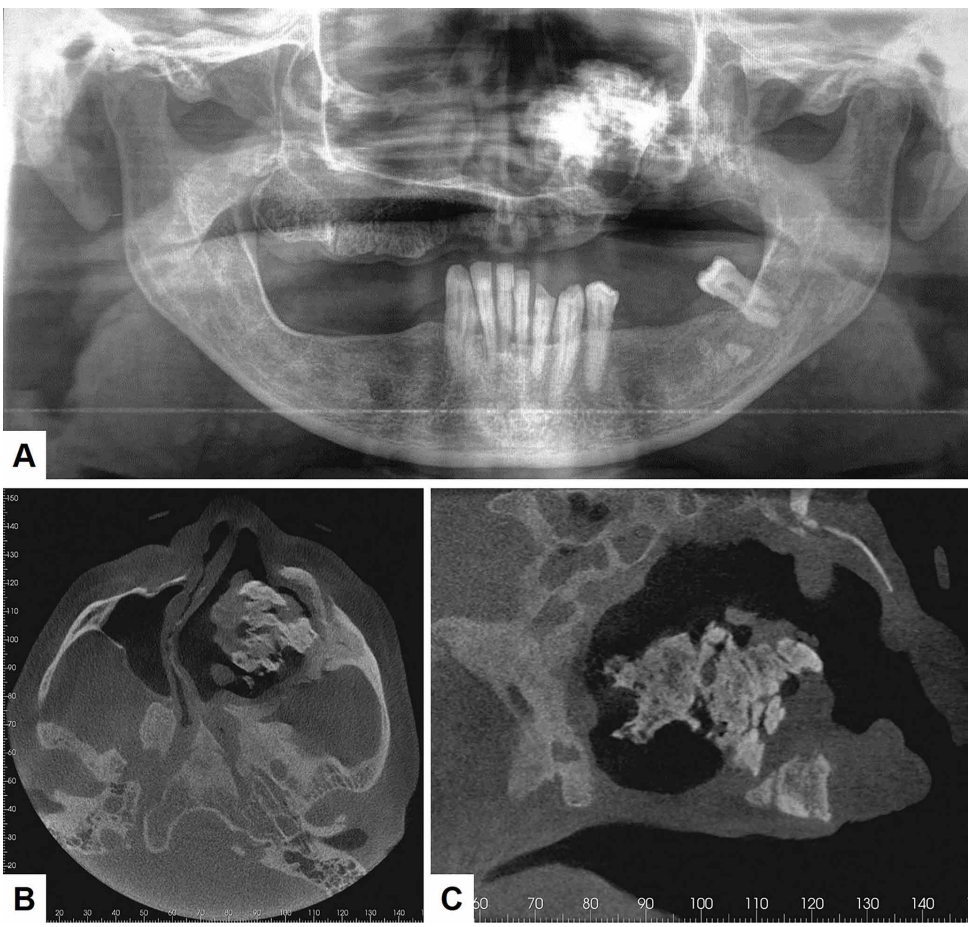

Fig. 2. Imaging. A: Panoramic radiograph showed a mixed lesion with radiopaque predominance in the left maxillary sinus region, as well as discontinuity of the left upper alveolar ridge. B, C: Cone beam computed tomography showed a large hyperdense image inside the left maxillary sinus, with density of calcified material, presenting areas of soft tissue density within and around the hyperdense mass. The lesion extends medially with displacement of the nasal septum to the right. Extensive discontinuity of the maxillary sinus floor was observed.

material microscopically showed trabeculae of necrotic bone and granulation tissue, with mixed inflammatory infiltrate and bacteria colonies (Fig. 3A). Areas of round-to-ovoid masses of filamentous bacteria colonies with a peripheral eosinophilic radiating rim, compatible with actinomycosis, were noted (Fig. 3B). Within necrotic bone large rhomboidal-to-ovoid structures surrounded by eosinophilic capsule, suggestive of larvae were found (Fig. 3E). A diagnosis of actinomycotic osteomyelitis with presence of structures compatible with larvae was established. Periodic acid-Schiff (PAS) stain was performed which highlighted the characteristic filamentous radiating aspect of actinomyces colonies (Figs. 3C and 3D).

The patient was evaluated during the 15th, 21st and 30th postsurgical days to assess the stability and sealing of the prosthesis. The foul smell disappeared and there was a significant improvement in speech, without signs of infection, and the patient was in good general condition.

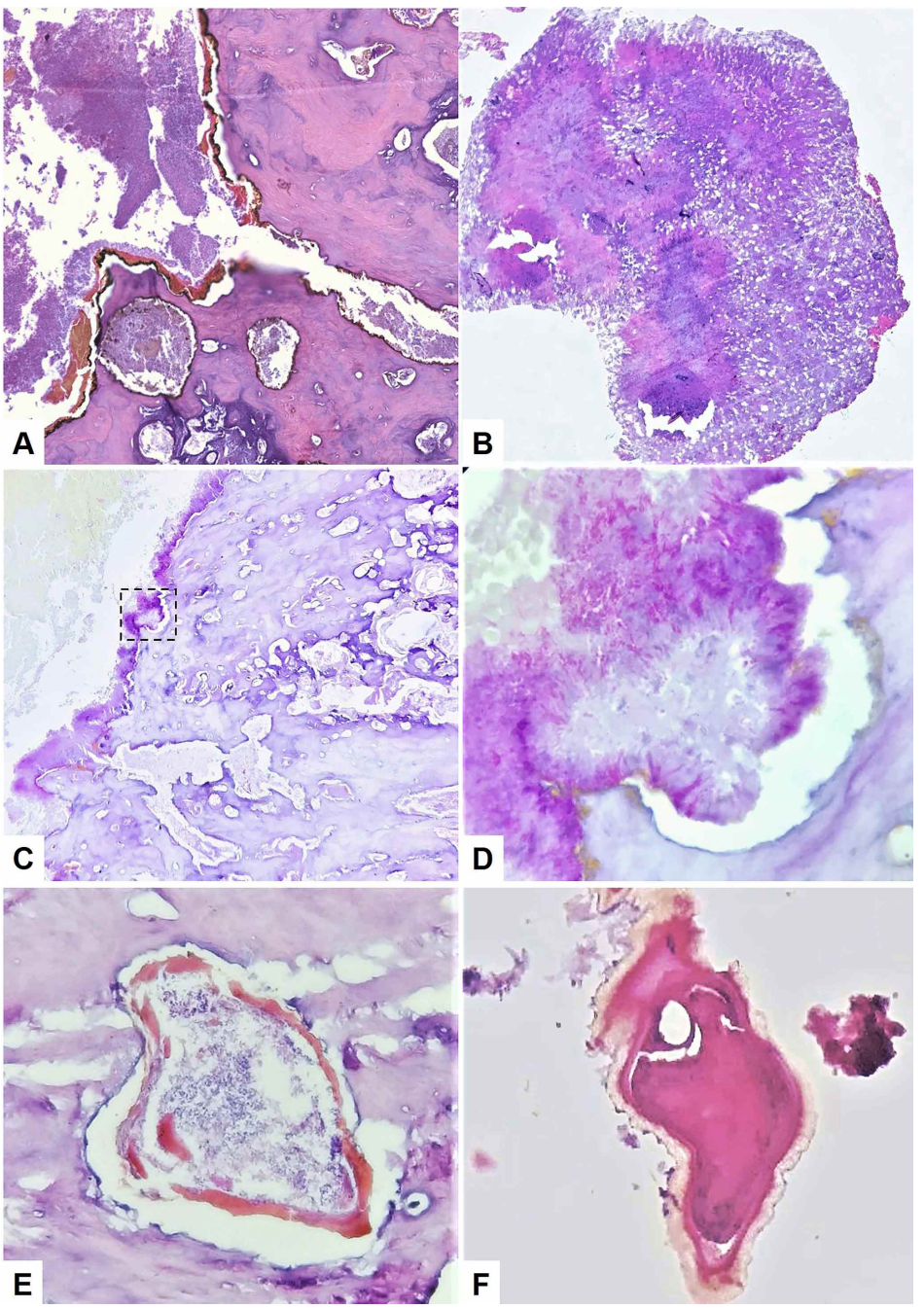

Fig. 3. Microscopical features. A: Trabeculae of necrotic bone admixed with abundant bacterial colonies (200X). B: High power view of the filamentous bacteria colonies, showing peripheral eosinophilic radiating rims (100X). C, D: Periodic acid-Schiff (PAS) stain highlights the characteristic filamentous radiating aspect of actinomyces colonies adjacent and within the necrotic bone. The area inside the dashed line square in C, corresponds to figure D (C: 50X, D:100X). E: One of the several structures suggestive of larvae observed in the specimen (400X). F: Histological section of a larva (grown in food) for morphological comparison (400X). (A,B,E,F: H&E staining; C, D: PAS staining).

DISCUSSION

In the present case, it is challenging to determine the primary cause of the osteomyelitis, however, considering the long-time evolution, and absence of systemic disease, we suggest that exposure of alveolar bone and an eventual small oroantral fistula as a result of teeth extractions, evolved to non-specific osteomyelitis with secondary actinomyces and larvae colonization, resulting in a progressive tissue destruction and large oroantral communication.

Oroantral communication can occur as a result of diverse conditions causing destruction of maxillary and sinonasal structures such as tumors, infections (e.g. deep mycoses, syphilis), Wegener granulomatosis, cocaine or intranasal narcotic abuse, or surgical procedures (Jewers *et al*., 2005; Murthy *et al*., 2014). Oroantral communication as a surgical complication, occurs more commonly after extraction of posterior maxillary teeth, usually molars and premolars. Most maxillary sinus perforations smaller than 5 mm close spontaneously, however larger communications require surgical procedures to avoid secondary infections (Dyn & Wolf, 2012).

Osteomyelitis is a heterogenous disease characterized by progressive bony destruction and formation of sequestrum (Singh *et al*., 2010). For development of the disease, the transport of pathogens into deeper tissues with an anaerobic environment is necessary, which may occur through spread of preexisting infection, hematogenous seeding, or direct inoculation of microorganisms into the bone (Singh *et al*., 2010; Gannepalli *et al*., 2015). Despite their low virulence and invasiveness, these bacteria act synergistically, creating a favorable environment for the growth of anaerobic bacteria, through the destruction of the vascularized aerobic environment and its substitution by poorly irrigated granulated tissue (Gannepalli *et al*., 2015). The clinical results of this infection are necrotic bone, pus discharge and foul odor (Agarwal *et al*., 2019) as observed in this case. Osteomyelitis is more frequent in mandible, presenting as a chronic soft tissue swelling (Garg *et al*., 2011). However, it can affect the maxilla with eventual involvement of the maxillary sinus (Agarwal *et al*., 2019).

Actinomycosis is a relatively uncommon infection, characterized by granulomatous and suppurative tissue destruction caused by commensal oral microorganisms (*Actinomyces israelli, A.viscosus*, *A. naeslundii*, etc.) (Lo Muzio *et al*., 2014; Gannepalli *et al*., 2015) that usually act synergistically with coparticipant bacteria principally streptococci and staphylococci. Chronic, persistent, purulent and slowly developing infections that typically spreads contiguously, not following anatomical planes, characterizes actinomycosis (Gannepalli *et al*., 2015; Sezer *et al*., 2017). In this case, the tomographic analysis revealed a large bone destruction, including orbit floor involvement. Thus, chronic destructive and slow-growing lesions should alert clinician to the possibility of actinomycosis (Agarwal *et al*., 2019).

Diagnosis of osseous actinomycosis may be confirmed microscopically, as in the present case, by presence of conglomerates of eosinophilic filamentous bacteria with radiating rim at the periphery (Splendore-Hoeppli phenomenon), surrounded by neutrophils. This pattern of actinomyces colony is known as sulfur granules (Lo Muzio *et al*., 2014; Gannepalli *et al*., 2015). A diagnostic criterion is the presence of an inflammatory response and/or fibrosis in proximity with these colonies (Kaplan *et al*., 2009). Additional histochemistry such as PAS, Gram, and silver stainings highlight this typical pattern of actinomyces colonies as seen in this case by PAS. The treatment of actinomycosis should be vigorous, with removal of the granulation tissue and necrotic bone until exposure of the healthy tissue, accompanied by high doses of antibiotic, generally penicillin, during prolonged time. If proper treatment is instituted, recurrence is rare (Sezer *et al*., 2017).

There are few reports of actinomycotic osteomyelitis with palate destruction, involvement of the maxillary sinus and oroantral communication (Crossman & Herold, 2009; Garg *et al*., 2011; Gannepalli *et al*., 2015; Sezer *et al*., 2017). Progression of actinomycotic osteomyelitis in the maxilla, if not treated, can bring serious consequences, including brain infection, being potentially life threatening (Singh *et al*., 2010; Agarwal *et al*., 2019).

The present case has a history of repeated myiasis in the affected area, and interestingly, in the microscopical analysis we found structures suggestiveof rests of maggots. Myiasis is characterized by the presence of larvae due deposition of eggs by flies, in accessible tissue (Ahmed *et al*., 2011). Intraorally it is described mainly in patients with psychiatric disorders, alcoholics, mouth-breathing, poor oral hygiene, facial trauma, malignances, and suppurative lesions (Ahmed *et al*., 2011; Manjunath & Pinto, 2018).

In the present case, bad odor was the main concern of the patient. He lived in social isolation, in a rural area with a low socioeconomic background, and had a long-term necrotic sinonasal/intraoral lesion, favoring the occurrence of myiasis. Treatment of oral myiasis includes mechanical removal, application of asphyxiating agents and pharmacological treatment with systemic ivermectin (Ahmed *et al*., 2011). Although, in this case at the time of surgery no live larvae were observed, microscopically structures compatible with larvae were found, coinciding with the recurrent myasis history. The calcification of larvae within affected tissue after occlusive management of myasis have been previously described (Francesconi & Lupi, 2012).

Complete removal of all necrotic tissue until exposure of healthy tissue, followed by obturator prosthesis and antibioticotherapy was performed in this case. The use of oral antibiotics alone is usually ineffective and should be combined with surgical removal. In chronic long-standing cases, the poor vascularization of the affected tissues due to fibrosis and necrosis, as well as the presence of bacteria within the sulfur granules can inhibit the direct effect of the antibiotics, justifying the high doses usually applied (Gannepalli *et al*., 2015).

The prothesis aimed to establish an oronasal seal and help to reestablish masticatory function. In this case the large defect was associated with reduced masticatory function, hypernasal speech and fluid in the nasal cavity. An earlier approach could facilitate closing the communication and a better outcome. The patient is followed up without recurrence and in good health with adequate masticatory functions.

In summary, we presented a unique case of an adult male with chronic actinomycotic osteomyelitis with history of recurrent episodes of myasis causing extensive destruction of left maxilla and sinonasal structures, producing oroantral communication and foul smell. These infections may result of wounds of multiple tooth extractions without proper care and follow-up, especially in patients presenting other risks factors such as low socioeconomic status and living in rural areas, as our patient. In conclusion diagnosis of actinomycotic osteomyelitis should be considered in long-term destructive lesions of the jawbones.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has/have given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name and initial will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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RESUMEN: La osteomielitis actinomicótica del maxilar que se presenta con comunicación oroantral es poco frecuente. En este trabajo reportamos el primer caso de esta condición en asociación con miasis. Un hombre de 50 años que refiere sinusopatía crónica y lesión maxilar que no cicatriza, de 30 años de evolución, presenta secreción ocasional purulenta nasal e intraoral, con mal olor y episodios recurrentes de presencia de larvas. La tomografía computarizada de haz cónico mostró una gran imagen hiperdensa en el interior del seno maxilar izquierdo, con áreas focales con densidad de partes blandas y una extensa discontinuidad del piso del seno maxilar, lo que confirma la fístula oroantral. El tejido necrótico legrado durante la cirugía presentó consistencia dura, coloración verdosa oscura, y fue remitido para análisis histopatológico. Microscópicamente se observó hueso necrótico, masas de colonias de bacterias filamentosas compatibles con actinomicosis y grandes estructuras romboidales rodeadas de cápsula eosinofílica sugestiva de larvas. Se estableció el diagnóstico de osteomielitis actinomicótica con presencia de estructuras compatibles con larvas.

PALABRAS CLAVE: actinomyces, maxilar superior, miasis, fístula oroantral, osteomielitis.

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