

CASE REPORT

ORAL CAVITY EUMYCETOMA

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SUMMARY

Mycetoma is a pathological process in which eumycotic (fungal) or actinomycotic causative agents from exogenous source produce grains. It is a localized chronic and deforming infectious disease of subcutaneous tissue, skin and bones. We report the first case of eumycetoma of the oral cavity in world literature. **Case report:** A 43-year-old male patient, complaining of swelling and fistula in the hard palate. On examination, swelling of the anterior and middle hard palate, with fistula draining a dark liquid was observed. The panoramic radiograph showed extensive radiolucent area involving the region of teeth 21-26 and the computerized tomography showed communication with the nasal cavity, suggesting the diagnosis of periapical cyst. Surgery was performed to remove the lesion. Histopathological examination revealed purulent material with characteristic grain. Gram staining for bacteria was negative and Grocott-Gomori staining for the detection of fungi was positive, concluding the diagnosis of eumycetoma. The patient was treated with ketoconazole for nine months, and was considered cured at the end of treatment. **Conclusion:** Histopathological examination, using histochemical staining, and direct microscopic grains examination can provide the distinction between eumycetoma and actinomycetoma accurately.

KEYWORDS: Mycetoma; Oral cavity; Fungi.

INTRODUCTION

Mycetoma is a chronic granulomatous infection of skin and subcutaneous tissue, which may involve bone²⁻⁴. Clinically it presents with swelling, nodules, discharging pus, and grain³. The grain or granules are clusters of filaments of bacteria or fungi³. There are two forms of mycetoma: 1. actinomycetoma, caused by a family of filamentous, Gram positive branching bacteria belonging to the order *Actinomycetales*; and 2. eumycetoma caused by several genera of true fungi²⁻⁴.

Epidemiologically, the disease is more common in tropical or subtropical countries, occurring in populations of low socioeconomic level, principally in rural areas, usually in men who work barefoot²⁻⁴.

Mycetoma usually involves the extremities, particularly the lower limb, hence the name "Madura foot", first recorded in the Madura District in India in 1861³.

We report the first case of oral cavity eumycetoma of all literature and discuss the possible origin of the fungal contamination of this area.

CASE REPORT

A 43-year-old male patient, complaining of swelling in the palate over

the course of two years, from which would often drain a liquid content, leading to a reduction of the edema.

On examination, swelling of the anterior and middle hard palate, with soft consistency, with a fistula draining a dark fluid content was observed. The panoramic radiographic showed a large radiolucent area involving the region of teeth 21-26 (Fig. 1A) and the cone beam computed tomography (CT) confirmed the lesion and showed communication with the nasal cavity, suggesting the diagnosis of periapical cyst (Fig. 1B).

Surgery was performed to enucleate the entire lesion (Fig. 2). The cyst contained a large quantity of yellow granules inside.

Histological examination of surgical specimen revealed granulomatous inflammation with pus containing characteristic grains in the center (Fig. 3A). Gram staining for bacteria was negative. The Grocott-Gomori staining, used to detect fungi, showed that the grains were composed of tangled hyphae (Fig. 3B and 3C), concluding the diagnosis of eumycetoma.

The patient had no history of sinusitis or tonsillitis or bacterial or fungal skin lesions suggestive of mycetoma of the face. However, he was submitted to an endodontic treatment of the teeth 21-22 before. The patient was treated with ketoconazole (200 mg/day) for nine months. Five

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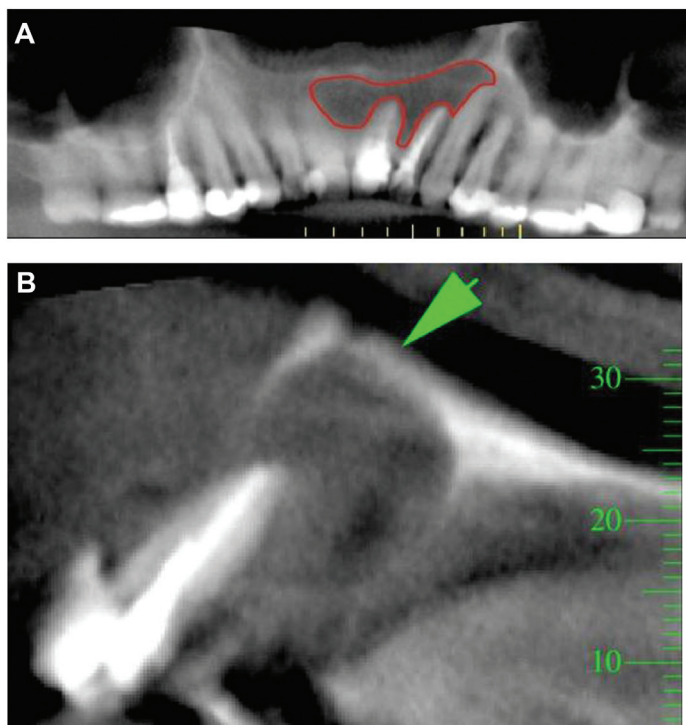


Fig. 1 - A. Panoramic radiographic showing demarcated radiolucent area. B. CT scan in sagittal view showing the cyst and the nasal cavity floor just above (arrow), with a small fistula.



Fig. 2 - Surgery showing the extent of injury.

months after surgery, a new cone beam CT was performed, which found a marked lesion repair. Fifteen months after the initial diagnosis, the patient has complete repair of the region affected by the injury, evidenced by clinical and radiographic examination and was considered cured (Fig. 4).

Informed consent was obtained from the patient. This study was approved by the Research Ethics Committee of University of Western São Paulo, UNOESTE (Process no. RC01/2010).

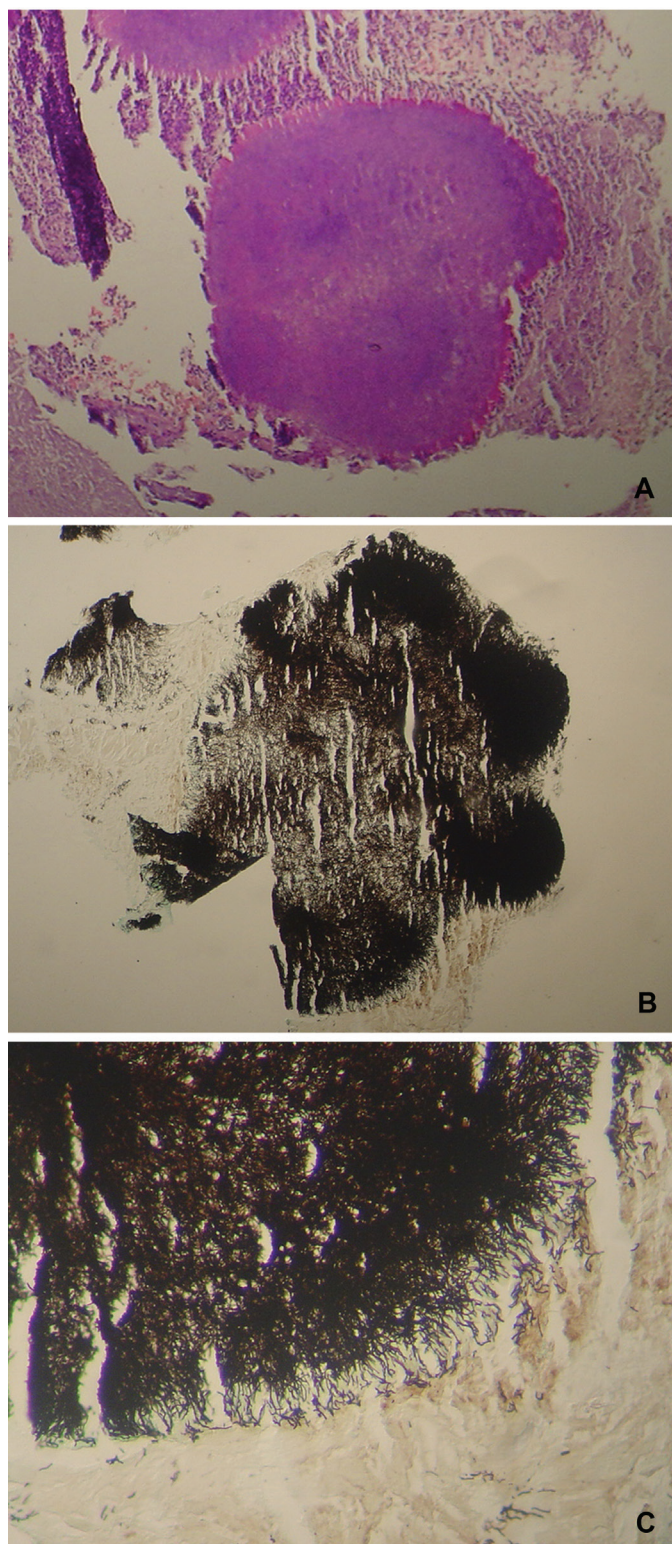


Fig. 3 - Light microscopy of the content of the lesion. A. Nodular structure (grain) within the inflammatory exudate (Hematoxylin and eosin, x250). B. Cluster of fungi (grain) stained in black (Grocott-Gomori, x250). C. Hyphae on the periphery of grain (Grocott-Gomori, x 400).

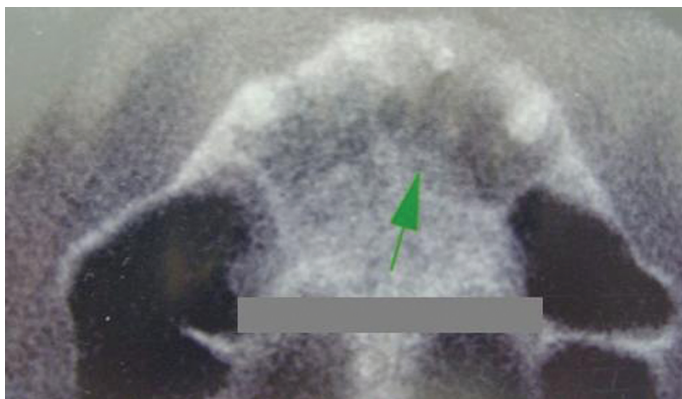


Fig. 4 - CT scan in axial view showing complete repair of the lesion (arrow).

DISCUSSION

Mycetoma is an infectious disease that progresses slowly, most commonly with little pain and can manifest itself many years after infection. This usually occurs by traumatic inoculation of the agent in tissue^{2,3}. Pain and tumor are the two main symptoms that bring patients to the medical service⁴. In this case, the patient went to the dental service with complaint of swelling and fistula in the hard palate over the course of two years.

Clinical findings of inflammation are more commonly associated with actinomycetoma, while tumoral and cystic features are more associated with fungal (eumycetoma) mycetoma⁴, as observed in our case, in which the lesion was cystic, and the main differential diagnosis is periapical cyst due to its location.

Imaging may help to elucidate the differential diagnosis of bone lesions and soft tissue. The X-ray may show signs of chronic osteomyelitis. CT can best detail the bone lesion, showing destruction, periosteal reaction, erosion and also tissue involvement. Magnetic resonance imaging (MRI) is superior to other radiological imaging in the evaluation of mycetoma, because it can reveal small lesions of low signal intensity on T1- and T2-weighted images, which correspond to the grain of mycetoma³. The ultrasound can also differentiate mycetoma from other infections or tumor lesions, due to the appearance of grain, characterized by the presence of numerous sharp, hyper-reflective echoes within multiples cavities³. In our case, there was only the cystic cavity without echoes inside, which contributed to the diagnosis of periapical cyst.

An X-ray or ultrasound cannot differentiate actinomycetoma from eumycetoma³. Confirmation of the diagnosis and the distinction between the two are based on histopathological and mycological examination^{4,9}. Histologically, a typical mycetoma consists of an area of granulomatous inflammation with a purulent center, surrounded by a thick, fibrous capsule and containing characteristic grains³. The grains can also be observed in the direct examination⁹. The distinction between actinomycetoma and eumycetoma can be made in histological sections of biopsy material using histochemical staining. The Gram staining is used for detection of actinomycetomas, because these are caused by Gram-positive bacteria. The Gomori-Grocott staining is used to identify fungi, marking fungi in black without marking bacteria from the order of *Actinomycetales*⁷. In this case, the grain was identified histologically,

and it was positive for Grocott-Gomori and negative for Gram staining, determining the diagnosis of eumycetoma.

Histopathology does not allow the identification of species of bacteria and fungi that cause mycetoma, nor determine their sensitivity to chemotherapeutic agents. This requires the complementation of bacteriological or mycological examination⁷. *Actinomadura pelletieri* and *Actinomadura madurae* can cause actinomycetoma. Eumycetoma are caused mainly by *Madurella mycetomatis* but also by *Pseudoallescheria boydii* and *Rhinoctadiella atrovirens*⁴. In this case, mycological examination was not performed because the diagnosis of eumycetoma was not suspected initially.

The treatment of mycetoma is difficult and failure is not uncommon. Generally, the response is better in cases of actinomycetoma than eumycetoma^{1,3}. Bone involvement may complicate the clinical management¹. Surgery in combination with azole treatment is the recommended regimen for small eumycetoma lesions in the extremities^{1,9}.

Ketoconazole, an orally active antimycotic imidazole, is inhibitory *in vitro* against a variety of pathogenic fungi, and has been recommended for therapy of eumycetoma at a dosage of 400 mg daily for 8-24 months, associated with limited surgical excision⁹. Patients with eumycetomas were considered cured when all the sinuses closed, swelling disappeared and no cavities or bone destruction was seen in roentgenogram; greatly improved when the pain and tenderness had completely disappeared, the existing sinuses closed but with one or two fresh nodules still developing, discharging pus with or without granules which were not viable; and slightly improved when the pain and tenderness disappeared, but the sinuses remained active, often with viable granules⁹. Our patient used ketoconazole (200 mg/day), displaying proof of cure after nine months of treatment, showing that this is also an adequate therapeutic scheme for these lesions in the oral cavity. Although considered cured by the criteria described above, the patient remained under follow-up dental care for one year more.

GUMAA *et al.*⁵ described a case of eumycetoma of jaw caused by *Madurella mycetomatis*, but there were no cases of eumycetoma involving the oral cavity reported worldwide.

Some authors have described the presence of actinomyces and fungi in the amygdale, especially in adults, associated with a minimal tissue response, indicating that these organisms are saprophytic and not primary pathogens in these sites^{6,8}. Faced with a traumatic injury, this may be a source of contamination to other parts of the oral cavity, with inoculation of the agent in tissue, leading to mycetoma formation. In this case, two hypotheses can be suggested to the contamination of the palate: 1. The patient had a periapical cyst that became infected secondarily, because of the fistula with the nasal cavity, or 2. The patient had fungi in the amygdale and a local trauma (e.g. endodontic treatment) resulting in the inoculation of periapical region and the formation of eumycetoma, which subsequently drained into the nasal cavity.

Dental treatment must take into account not only the possibility of contamination of the periapical region or bone by bacteria but also fungi.

The distinction between eumycetoma and actinomycetoma is very important for treatment, and histopathological examination, using

histochemical staining, and direct microscopic grain examination can provide this distinction accurately.

RESUMO

Eumicetoma da cavidade bucal

Micetoma é um processo patológico no qual agentes eumicóticos (fungos) ou actinomicóticos de origem exógena podem causar formação de grãos. É uma doença infecciosa localizada, crônica e deformante do tecido subcutâneo, pele e ossos. Relatamos o primeiro caso de eumicetoma da cavidade bucal da literatura mundial. **Relato do caso:** Paciente masculino, de 43 anos, com queixa de edema e fístula no palato duro. Ao exame clínico, observava-se edema da região anterior e média de palato duro, com fístula drenando líquido escuro. A radiografia panorâmica mostrou área radiolúcida extensa, envolvendo a região dos dentes 21 ao 26 e a Tomografia computadorizada evidenciou comunicação com a cavidade nasal, sugerindo o diagnóstico de cisto periapical. Foi realizada cirurgia para remoção da lesão. O exame histopatológico revelou material purulento com grãos característicos. A coloração de Gram para pesquisa de bactérias foi negativa e a coloração de Gomori-Grocott para pesquisa de fungos foi positiva, concluindo o diagnóstico de eumicetoma. O paciente foi tratado com Cetoconazol durante nove meses, obtendo cura ao final do tratamento. **Conclusão:** O exame histopatológico, usando colorações histoquímicas, e o exame dos grãos por microscopia direta podem proporcionar adequada distinção entre eumicetoma e actinomycetoma.

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Received: 4 November 2010

Accepted: 3 March 2011