

Tongue Actinomycetoma Due to *Actinomadura madurae*: A Rare Clinical Presentation

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A 60-year-old male farmer presented with tongue swelling of 1-month duration. Local oral clinical examinations showed a painless firm mass in the anterolateral aspect of the anterior third of the tongue. Fine needle aspiration for cytology confirmed the diagnosis of tongue actinomycetoma due to *Actinomadura madurae*. The patient underwent wide local excision under general anesthesia and had an uneventful postoperative recovery. He was started on amikacin sulfate 15 mg/kg daily and cotrimoxazole 15 mg/kg twice per day for 6 months. The lesion healed completely, with no evidence of recurrence at 6-month follow-up. The route of infection in this patient is unclear; however, direct traumatic inoculation is the most likely route. To the authors' knowledge, this is the first report of tongue mycetoma in the medical literature.

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Mycetoma is a chronic, specific, granulomatous, progressive, destructive inflammatory disease that involves the subcutaneous tissues and spreads to the skin and deeper structures, leading to massive destruction, deformity, disability, and possibly death. The authors present a case of tongue mycetoma in a 60-year-old man.

Report of Case

A 60-year-old male farmer from the White Nile State in central Sudan reported to the Mycetoma Research Center in

Khartoum, Sudan in 2011 with a painless swelling in the left anterolateral aspect of the tongue. Four weeks before presentation, he noticed a painless swelling in his tongue that was of gradual onset and course. The swelling gradually increased then 2 sinuses developed, which started to discharge an odorless, purulent liquid. No grains were noticed in the discharge. He had no constitutional symptoms. On direct questioning, he recalled biting his tongue during chewing. The patient regularly brushed his teeth with a locally produced Acacia tree stick, a common habit in Sudan. He was of low socioeconomic status and had no family history of mycetoma. His medical, drug, or geographic history was not contributory to his presenting condition.

General clinical examination disclosed a fit, well-oriented, cooperative man. He was afebrile, with a pulse rate of 80 beats/min, a respiratory rate of 18 breaths/min, and a blood pressure of 130/85 mm Hg. Examinations of his cardiovascular, respiratory, and central nervous systems, abdomen, limbs, and back were unremarkable.

On oral examination, there was a swelling in the left anterolateral aspect of the anterior third of the tongue. It was oval in shape with a smooth surface measuring 1 × 0.5 cm in diameter. The swelling was nontender, hard in consistency, and fixed to the deep structures. There were 2 healed sinuses. The regional lymph nodes were not enlarged (Fig 1). The working differential diagnosis included squamous cell carcinoma, traumatic ulcer, and inflammatory granulomas caused by tuberculosis, syphilis, or leishmaniasis.

His renal and hepatic profiles and full blood cell count were within normal limits. A swab culture taken from the

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FIGURE 1. Photograph showing the tongue swelling.

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sinuses showed no growth. Multiple fine needle aspirations for cytology yielded bloody material. The smears and cell blocks disclosed *Actinomadura madurae* grains with massive inflammatory cells, including neutrophils, giant cells, lymphocytes, and plasma cells (type II tissue reaction; Fig 2).

The patient was hospitalized and a wide local excision was performed under general anesthesia, with an uneventful recovery. He was discharged on amikacin sulfate 15 mg/kg daily and cotrimoxazole 15 mg/kg twice per day for 6 months. He was seen regularly every 5 weeks in the outpatient department for follow-up, with no evidence of drug side effects or recurrence at 6-month follow-up.

Discussion

Mycetoma is a chronic, specific, granulomatous, progressive, destructive inflammatory disease that involves the subcutaneous tissues and spreads to the skin and deeper structures, leading to massive destruction, deformity, disability, and possibly death.¹ It is caused by a wide range of true fungi (Eumycetes) or higher-order bacteria (Actinomycetes) and thus classified as eumycetoma or actinomycetoma, respectively.^{2,3}

Mycetoma has a worldwide distribution that is extremely uneven. It is endemic in many tropical and subtropical regions, where it is considered a serious medical and health problem.⁴ It prevails in the mycetoma belt, which stretches in a band from the latitudes of 15°S to 30°N, including the countries of Sudan, Somalia, Senegal, India, Yemen, Mexico, Venezuela, Columbia, Argentina, and a few others. The African continent seems to be the area of the highest prevalence, although mycetoma is occasionally reported in the Western world.⁴

Although Sudan is considered the home of mycetoma, this is the first reported case of tongue mycetoma. In Sudan, actinomycetoma is less common than eumycetoma and it accounts for only 30% of clinical cases. *Actinomadura madurae* is an even rarer

cause. These factors make the reported presentation a rare event.^{3,4}

The susceptibility and resistance to mycetoma remain unclear, as does the route of infection. Direct traumatic inoculation of the causative organism is the most acceptable theory for the entry route in mycetoma. The present patient had a history of local trauma at the mycetoma site.

In mycetoma-endemic areas, Acacia trees are plentiful and they are armed with strong thorns. These thorns are considered important facilitators of the subcutaneous inoculation of infection.⁴ The present patient used a locally produced Acacia tree stick to brush his teeth, which may be in accord with the inoculation theory. However, the habit of using locally produced Acacia tree sticks is very common in the region, so if the traumatic inoculation hypothesis is true, then the incidence of oral mycetoma should be higher than it actually is.

The use of wood sticks for brushing teeth is a common habit in many Afro-Asian communities.⁵ Usually these sticks are used for a longer period than a modern toothbrush and each cleaning is usually implemented for 5 to 10 minutes.⁶ Many of these plant species have related medicinal properties, one of which is antibacterial.^{5,7} This may partly explain the rarity of oral mycetoma despite the habit of using wood sticks for tooth brushing in endemic areas.

Mycetoma is characterized by the presence of a painless subcutaneous mass, multiple sinuses, and discharge that contains grains. This triad is pathognomonic of mycetoma. The infection usually gradually spreads to involve the skin, deep structures, and bone, leading to massive destruction, deformities, and

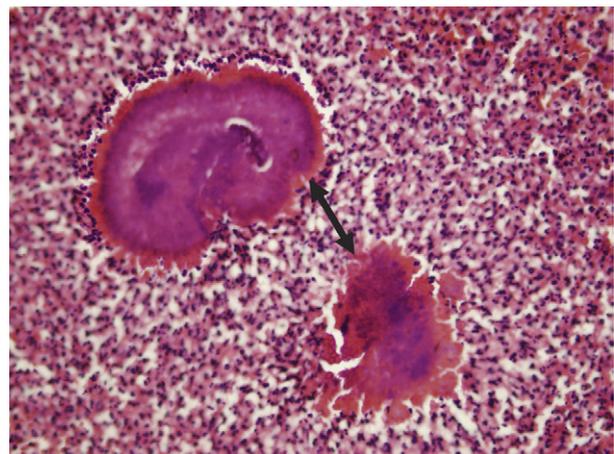


FIGURE 2. Photomicrograph showing *Actinomadura madurae* grains (2-headed arrow) with massive inflammatory cells that include neutrophils, giant cells, lymphocytes, and plasma cells (type II tissue reaction; magnification, 200×).

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disabilities. The foot is affected most, accounting for 70%, followed by the hand, seen in 12% of patients. Less common sites are the head and neck, leg, thigh, and back.^{1,2,8}

The oral cavity is a rare site for mycetoma; there are only 2 reported cases of mycetoma at this site.^{9,10} Both cases had eumycetoma caused by *Madurella mycetomatis*. The eumycetoma was located in the hard palate in 1 case and in the buccal floor in the other. The first case presented with a typical mycetoma triad, whereas the second presented with a buccal floor mass and the diagnosis of mycetoma was an operative surprise. These patients underwent wide surgical excisions and received long-term antifungal treatment.

The temporomandibular joint was the site of mycetoma in another 2 cases; in 1 case *M mycetomatis* was the causative organism, whereas in the other case mycetoma was caused by *Streptomyces somaliensis*. Temporomandibular joint masses and ankylosis were the common presentations. These patients were treated with a wide local excision and antifungal treatment for the eumycetoma and antibiotic and antimicrobial treatments for the actinomycetoma.^{11,12}

The diagnosis of mycetoma requires proper identification of the causative organisms by culture and by histopathologic and molecular techniques. This can be performed by fine needle aspirates taken directly from the lesion, as in the present patient, or by surgical excision and biopsies. The extent of disease can be ascertained by different imaging techniques, including radiography, ultrasound, computed tomography, and magnetic resonance imaging.¹⁻³ Intraoral mycetoma is difficult to image by the available imaging techniques, but the most appropriate technique is magnetic resonance imaging.

The treatment of mycetoma depends on the causative organism and the duration, extent, and site of the disease. The common treatment regimen for actinomycetoma is amikacin sulfate 15 mg/kg given in 2 divided doses daily and cotrimoxazole 14 mg/kg twice daily. They are given in the form of cycles, and each cycle consists of 3 weeks of amikacin sulfate and

5 weeks of cotrimoxazole until cure is achieved and the response to medical treatment is satisfactory.¹⁻³

For eumycetoma, the current treatment of choice is ketoconazole 400 to 800 mg/day or itraconazole or voriconazole 200 to 400 mg/day. In general, the response to medical treatment is not satisfactory, and these drugs have many serious side effects. They require a long duration to effect a cure, and the recurrence rate after adequate treatment is high.¹⁻³

In mycetoma, surgical interventions range from a wide local excision, to multiple debridements, to amputation of the affected part in advanced disease. In the present patient, a wide local excision combined with medical treatment was considered optimal treatment because the disease was early and localized.¹⁻³

In conclusion, mycetoma should be considered in the differential diagnosis of any intraoral mass in tropical and subtropical regions, although it is a rare event. With massive international travel and population movements and migrations, it seems worthwhile to bear mycetoma in mind in the differential diagnosis of intraoral masses and pathology.

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