

Case Report

Mycetoma caused by *Streptomyces sudanensis*

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Abstract

Introduction: Mycetomas are deep, chronic skin infections caused by bacteria or fungi.

Case Presentation: We report the case of an adult male patient with mycetoma caused by *Streptomyces sudanensis* located on the foot. The patient was treated with i.m. gentamicin (80 mg/day) for six weeks, followed by three-week stop period. A total of eight courses six-week courses of gentamicin therapy, each followed by a three-week stop period, were completed. The patient is still undergoing treatment with the same regimen.

Conclusions: Pulsed therapy may be considered in the management of mycetoma in order to prevent the development of resistance.

Key words: Mycetoma; *Streptomyces sudanensis*; gentamicin; pulsed therapy.

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Introduction

Mycetomas are deep, chronic skin infections caused by bacteria (actinomycotic mycetomas or actinomycetomas) or fungi (eumycotic mycetomas or eumycetomas). Actinomycetomas occur mainly in Central and South America, while eumycetomas are more prevalent in Sub-Saharan Africa.

Etiological agents penetrate the skin through breaks caused by thorns, shrubs, and wood splinters: mycetomas are therefore considered occupational diseases among farmers and lumberjacks.

Adult males are much more affected than females.

The Latency period is often very long ranging from months to years.

The clinical picture is characterized by the gradual appearance of swelling and inflammatory papules, nodules, and plaques. Over time, abscesses, fistulae, and ulcers develop, with seropurulent discharge containing grains: the latter may be white, yellow, red, or brown. Involvement of the legs, ankles, and feet is typical, although the buttocks and thighs may also be affected. Extension to muscles, aponeuroses, ligaments, tendons, joints, and bones is relatively common. Pain frequently causes difficulty walking.

Bacterial superinfections by *Staphylococcus aureus*, Gram-negative bacteria, and anaerobes are common.

Systemic symptoms are uncommon and generally

mild. The clinical course is chronic and relapsing [1,2].

Histopathology reveals a dermal infiltrate composed of lymphocytes, histiocytes, neutrophils, and giant cells, with suppurative granulomas containing grains in the deep dermis and subcutaneous tissue. In actinomycetomas, thin filamentous bacteria are observed; in eumycetomas, eosinophilic structures containing hyphae and spores are seen [3].

We report a case of mycetoma caused by *Streptomyces sudanensis*.

Case Report

A 37-year-old man from North Macedonia, living and working in Siena, Italy, as a farmer and lumberjack, was admitted due to a chronic dermatosis located on his right foot. The patient reported that his condition had first appeared approximately 20 years earlier and had been diagnosed, based on microbiological examination, as actinomycetoma caused by *Streptomyces sudanensis*. In accordance with literature data [4,5], the patient had previously been treated - albeit unsuccessfully- over long periods with numerous antibiotics (amoxicillin, amoxicillin/clavulanic acid, doxycycline, levofloxacin, minocycline, trimethoprim/sulfamethoxazole) and antifungals (itraconazole). Amikacin had not been used due to a suspected drug allergy. Two surgical interventions had also proven unsuccessful. The patient was referred to one of us (S.V.) in July 2020, at which

time he was on a regimen of trimethoprim/sulfamethoxazole (160 + 800 mg/day), rifampicin (750 mg/day), and i.m. gentamicin (80 mg/day).

The patient reported being in good general health and denied the use of any other other systemic medications.

Dermatological examination revealed swelling and multiple papules, nodules, and plaques on a mildly erythematous base, located on the plantar surface of the right foot (Figure 1). The patient complained of pain and difficulty walking.

General physical examination was unremarkable: in particular, no inguinal lymphadenopathy was detected.

Laboratory tests were within normal ranges or negative.

We decided to treat the patient with i.m. gentamicin (80 mg/day) for six weeks, followed by a three-week stop period. A total of eight six-week courses of gentamicin therapy, each followed by a three-week stop period, were completed. The patient was reviewed every six weeks. Marked clinical improvement of the lesions (Figure 2) and near-complete resolution of pain were observed. No side effects were reported or detected, and no significant laboratory changes were noted. The patient is still undergoing treatment with the same regimen.

Discussion

Streptomyces sudanensis was first described in 2008 by Quintana *et al.* [6]. It is a Gram-positive, aerobic, non-acid-fast, non-motile actinomycetes, that forms a branched, light yellow substrate mycelium on several agar media [6]. *S. sudanensis* is closely related to *S. somaliensis* [6]. *S. sudanensis* is a soil bacterium [7]; however, it can live in symbiosis with fungi, plants, and animals [7]. Rare cases of *S. sudanensis* mycetomas

Figure 2. A and B are clinical pictures from January 2023.



Figure 1. A and B are clinical pictures of the time patient was first seen (July 2020)



have been reported from Sudan [5-8].

The case described here is, in our opinion, noteworthy for several reasons: a) to our knowledge, it represents the first case of mycetoma caused by *S. sudanensis* observed outside Sudan; b) as in many cases of actinomycetomas, our patient was found to be resistant to multiple drugs despite good adherence and compliance with therapy: these had been administered at correct daily dosages and for prolonged periods (as previously mentioned, amikacin was not used due to a suspected allergy). Gentamicin, administered as pulsed therapy (six weeks on, three weeks off, to prevent the development of bacterial resistance), proved to be effective: to our knowledge, it has been used to treat actinomycetomas only in India [9-11] and never according to a pulsed regimen.

Conclusions

This case highlights the potential effectiveness of pulsed gentamicin therapy in the treatment of *Streptomyces sudanensis* mycetoma. The regimen may help reduce the development of bacterial resistance and improve patient outcomes, particularly in resource-limited settings. Further research is needed to validate this approach.

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Conflict of interest

No conflict of interest is declared.

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