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# Resistant Actinomycotic Mycetoma: A Case Report

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***Authors' contributions***

*This work was carried out in collaboration among all authors. All authors contributed equally in treatment, management and follow-up of the patient and in compilation of the case report. All authors read and approved the final manuscript.*

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***Case Report***

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## ABSTRACT

**Introduction:** Mycetoma Pedis is the most common form of Eumycetoma known widely as the Madura Foot. Favourable respnoses have been reported with drug therapy alone even in long-standing lessions with involvement of bone and lymphatics. The case report aims to present a case of Madura Foot in an adult male residing in a geographically non-prevalent region for the disease; who required amputation because of failure of resolution after medical therapy alone.
**Case Presentation:** A 50-year-old male presented with complaints of Swelling and Discharge from Multiple Sites over the Right Leg x 3 years.
**Discussion:** The disease is usually seen in field workers like farmers of tropical and sub-tropical regions and generally affects males between 20 and 40 years.
Ideally, specific therapy depends on the identification of the causative agent and determination of its drug sensitivity. Only a few cases have been reported where amputation was required after medical therapy failure.
**Conclusion:** Favourable respnoses have been reported with drug therapy alone even in long-standing lessions with involvement of bone and lymphatics. Exploration and drainage of sinus tracts, debridement of diseased tissue, and removal of bone cysts assist greatly in healing especially in resistant cases.

*Keywords: Actinomycetoma; madura foot; therapy failure; amputation.*

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## 1. INTRODUCTION

Mycetoma is a chronic, suppurative, granulomatous disease of the subcutaneous tissues and bones characterized by localized swellings with multiple sinuses discharging granules that are the microcolonies of the causative agent [1]. In view of severity it is a progressive and destructive disease.

The etiologic agents range from bacteria to fungi. they exist freely in nature as soil or plant saprophytes and gain entrance into the human body via abrasion or implantation [2,3]. Actinomycotic Mycetoma is caused by aerobic species of actinomycetes belonging to the genera Nocardia, Streptomyces and Actinomadura. Eumycotic Mycetoma is associated with a variety of fungi, the most common being Madurella Mycetomatis, Pseudallescheria Boydii, and Acremonium species.

Mycetoma has been known for a long time in India. John Gill in 1842 described the clinical features of the disease 'Madura Foot' for the first time. Dr. Henry Vandke Carter of Grant Medical College, Mumbai first coined the term Mycetoma meaning Fungal tumor.

In India Actinomycotic Mycetoma is more common than Eumycotic Mycetoma, and males are affected more than females.

Mycetoma essentially requires a predisposing trauma to create a point of entry for the causal saprophytic micro-organisms and it often begins as a small, painless, subcutaneous nodule at the ste of injury [3]. Gradually, multiple nodules develop, ulcerate, and drain through sinus tracts. The discharge may be serosanguineous, seropurulent, or purulent and often contains its characteristic granules. With further progress of the disease, the surrounding tissues become swollen, indurated, and deformed by fibrous tissue reaction and multiple sinus formation. Pain is not a serious complaint when only the soft tissue is invaded; the condition becomes very painful with the involvement of bones or with the secondary bacterial infection. Mycetoma usually remains localized, extending slowly by direct contiguity along fascial planes and invading the subcutaneous tissue, fat, ligaments, muscles, and bones but sparing the tendons and nerves until very late. The degree and extent of bone involvement vary with the species of the infecting agent. In Eumycotic mycetoma, the lessions takes the form of single or multiple, punched-out lytic areas with well-defined walls and little sign of bone reaction.

In actinomycotic mycetoma, both osteolytic and osteporotic changes are present at the same time. Destruction of the ligaments and articular surfaces results in ankylosis of the joints. When the infection is localized in the head, neck, chest, or buttocks region, visceral invasion by contiguity may occur. Lymphadenopathy is common, more common in actinomycetoma.

Mycetoma should be differentiated from actiomycocsis, botromycosis, and osteomyelitis. A number of diseases such as neoplasms, Kaposi's sarcoma, syphilis, yaws, leprosy, tuberculosis, cutaneous leishmaniasis, and other mycosis that may mimic mycetoma at some stage of their evolution. Other uncommon areas involved are hand, knee, head and neck.

## 2. PATIENT PRESENTATION

A 50-year-old married male presented with complaint of of Swelling and Discharge from Multiple Sites over the Right Leg x 3 years. Patient had history of right sided hemiplegia in 2007. The patient had no complaint of fever / local site pain / history of trauma. The patient was not a not a known case of DM, HTN and was non-alcoholic and a non-smoker.

### 2.1 Clinical Findings

Her vital signs were blood pressure 100/60, heart rate 120 beats per minute, respiratory rate 25 breaths per minute and temperature 37 °C. Local examination: 10cm x 12cm sized swelling present over the right lower limb beginning from foot above. Multiple discharging sinuses present with purulent granular discharge. Surrounding skin was reddened and temperature was rasied as compared to the contralateral limb. Pitting edema present. No crepitations present.

### 2.2 Investigations

Basic routine blood investigations including the total WBC count, Hemoglobin, serum creatinine and bilirubin were done and found to be normal.
X-ray chest was Normal.

X-ray of the local part suggested generalised osteopenia, excessive soft tissue prominence.

Ultrasonography of the local region was done ruling out underlying DVT.

PUS from the sinuses yielded Gram-positive filamentous branched bacilli S/O Actinomycetes seen.

Wedge-Biopsy yielded findings S/O Actinomycetoma.

### 2.3 Therapeutic Intervention

Initially a trial of medical management was given since favourable responses have been reported in this disease [4,5]. Following that;

Inj.Gentamycin (80mg) iv 12 hourly x 4 weeks,

Tab.Co-Trimoxazole (480mg) DS 2 BD x 4 weeks,

AFTER 4 WEEKS; Start,

Tab.Doxycycline (100mg) 1 BD x 4 weeks,

Tab.Co-Trimoxazole (480mg) DS 1 BD x 4 weeks,
The patient however showed no clinical improvement and hence was considered failure of medical therapy and hence a Below-Knee amputation under spinal anaesthesia was then planned [6].

Ultrasonography of the local region was done ruling out underlying DVT.

PUS from the sinuses yielded Gram-positive filamentous branched bacilli S/O Actinomycetes seen.

Wedge-Biopsy yielded findings S/O Actinomycetoma.

### 2.4 Follow–Up and Outcome

The patient was discharged on Tenth post-operative day after alternate suture removal. Follow up was then done on every fifth day, and complete suture removal done on the 15$^{th}$ day.

The patient was given oral antibiotics for 7 days post discharge along with analgesics.

No wound discharge/dehiscence/seroma formation or any other immediate post-operative complications were noted.

After suture removal, the patient was kept on monthly follow up for six months.

## 3. DISCUSSION

Mycetoma is a chronic granulomatous inflammatory response involving bacteria or fungi that triggers the formation of grains containing aggregates of the causative organisms that may be discharged onto the skin surface through multiple sinuses, causing the progressive development of granulation and scar tissue that can cause deformity.

**Fig. 1.**

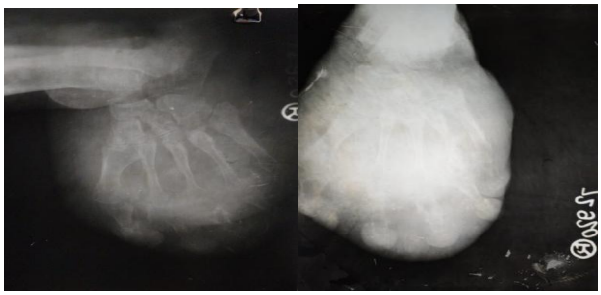

**Fig. 2.**

The type of mycetoma is often suggested by the color of the grains. Red grains are indicative of an actinomycotic mycetoma. Black grains are consistent with a eumycetoma, a white and yellow colored grains are indicative of either actinomycetoma or eumycetoma [7].

The differentiation between eumycetoma and actinomycetoma is an important one with regard to therapy. The combination of the clinical specific lesions, typical grains and the histopathological appearance (presence of a granulomatous inflammatory reaction with abscesses containing granules of the infecting organism) is characteristic of the diagnosis.

X-rays, tomography, and magnetic resonance imaging are all useful to determine the extension of the lesions in bone and other tissues.

Surgery is indicated in mycetoma for resistance to medical treatment, better response to medical treatment in patients with massive disease or for localized lesions. The surgical options range from local excisions to amputations. Amputation is indicated in advanced mycetoma not responding to medical treatment with severe secondary bacterial infection.

## 4. CONCLUSION

Surgery is indicated in mycetoma resistant to medical treatment, or better the response to medical treatment in patients with massive disease or for localized lesions. The surgical options range from local excisions to amputations. Amputation is indicated in advanced mycetoma not responding to medical treatment with severe secondary bacterial infection hence the importance of earlier diagnosis and treatment [5,6].

## CONSENT

Consent has been taken from the patient.

## ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

## COMPETING INTERESTS

Authors have declared that no competing interests exist.

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