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Recurrent actinomycotic mycetoma masquerading as eumycetoma of the right foot- a case report

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Abstract

Mycetoma is a persistent granulomatous infection produced by filamentous fungi (eumycetoma) or aerobic bacteria (actinomycetoma), and effective treatment depends on differentiation between the two. Here we report a case of a male farmer from rural South India who developed ulceration, swelling, and multiple discharging sinuses from his right foot. After an initial diagnosis of eumycetoma based on the potassium hydroxide (KOH) mount, itraconazole treatment was initiated. However, recurrence within a month required additional evaluation. Actinomycetoma was confirmed by repeat histopathology, which showed basophilic filamentous sulphur granules with PAS-positive organisms. Significant clinical improvement was achieved when the patient received treatment with amikacin, cotrimoxazole, and rifampicin. This case emphasizes the challenge in diagnosing mycetoma and the importance of demonstrating a correlation between histopathological, microbiological, and clinical findings. To prevent misdiagnosis, repeated biopsy, culture and if feasible molecular testing should be carried out in situations that are recurring or non-responding.

Keywords: Mycetoma, Actinomycetoma, Eumycetoma, Recurrent mycetoma, Neglected tropical disease

Introduction

Mycetoma is defined as a chronic, progressive, granulomatous infectious disease affecting cutaneous and subcutaneous tissues, especially in the foot. It is majorly seen in the skin and soft tissues, rarely in bones which is caused by two types of organisms, either fungi (eumycetoma) or bacteria (actinomycetoma) [1, 2]. The infection typically occurs post traumatic implantation of the causative organisms found in soil, water and other species. These invade into the subcutaneous tissue via minor trauma or injury in the form of thorn pricks or cuts, particularly among individuals walking barefoot in rural, agricultural environments [3, 4]. It is clinically characterized by swelling, multiple draining sinuses containing blood, pus, grainy discharge of causative organisms that are eliminated through sinus tract [4, 5]. Common organisms causing Mycetoma includes *Madurella grisea*, *Madurella mycetomatis*, *Leptosphaeria senegalensis* and *Pseudoallescheria boydii* (Eumycetoma Species) While Actinomycetoma species includes *Actinomadura madurae*, *Nocardia brasiliensis*, *Actinomadura pelletieri*, *Streptomyces somaliensis*, and *Nocardia asteroides* [6].

This condition is included under neglected tropical diseases (NTD) by the World Health Organisation (WHO) that mainly affects economically weaker section in rural areas with lack of knowledge, diagnosis and treatment facilities due to limited research funding [7]. Recurrent mycetoma is uncommon and rarely documented in the regions of South India. Eumycotic and actinomycotic differentiation is necessary, as their treatments are variable, despite overlapping clinical symptoms that can lead to errors in diagnosis, especially in rural areas. [8]. This case highlights about recurrent actinomycotic mycetoma in right foot, initially identified as eumycetoma, later diagnosed to be actinomycetoma, underscoring the diagnostic and therapeutic challenges.

Case report

A 62-year-old male who was a farmer by occupation presented to the General Surgery outpatient department with complaints of pain, swelling, and ulceration over the right foot

with multiple discharging sinuses on the dorsal surface associated with blackish discoloration of the skin for 10 days (Fig 1a). He denied fever or systemic symptoms. The following day, he was referred to Dermatology, where he additionally reported foul-smelling discharge from the right foot for 15 days and was admitted in the department of dermatology. The patient reported that he often walked barefoot but did not note any past foot injuries. He has no relevant history of diabetes, HIV, immunosuppressive therapies or any other chronic illness. He had previously taken ayurvedic treatment for the same through different clinics with no improvement. Clinical examination revealed multiple draining sinus tracts over the plantar aspect of the right foot, discharging serosanguinous fluid with a foul odour. No regional lymphadenopathy was noted.

The initial biopsy specimen, which was mostly stratum corneum, was insufficient for diagnosis. Hence, repeat biopsy was recommended.

Gram stain showed occasional pus cells and occasional Gram-positive cocci in pairs, while pus culture grew only normal skin flora. Direct KOH (potassium hydroxide) examination showed black brittle grains with broad septate hyphae.

Based on the clinical suspicion of eumycetoma, the patient was started empirically on itraconazole 200 mg twice daily (BD), analgesics, proton pump inhibitors, antihistamines, and vitamin supplementation. After 22 days of hospitalization, he was clinically stable and discharged with itraconazole 130 mg twice daily (BD) along with supportive medications (Fig 1b).

However, after one month of continuous treatment with itraconazole, the patient returned with recurrent swelling of the right foot, worsening pain on walking, and persistent sinus discharge (Fig 2a). A detailed history revealed that the persistent pain and swelling had actually been present for the past seven years, indicating a chronic mycetoma that was previously under-recognized, with laboratory tests showing altered inflammatory markers and all the other baseline renal and liver function tests were within the normal limits (Table 1). Histopathological examination of the lesion revealed degenerated basophilic filamentous organisms (Sulphur granules) morphologically favouring Actinomycosis (Fig 3a, b). Periodic acid Schiff stain showed PAS positive organisms. No culture of grains or biopsy tissue was performed. X-ray examination showed no bony involvement.

The lack of clinical response to antifungal therapy and the recurrence of symptoms prompted reconsideration of the diagnosis. Due to the absence of culture confirmation, treatment was initiated based on histopathology findings consistent with actinomycetoma (bacterial mycetoma).

So, the patient was started on Inj Amikacin 500 mg twice daily (BD), T. Rifampicin 600 mg once weekly in the morning, T. Cotrimoxazole (Trimethoprim 160 mg + Sulfamethoxazole 800 mg) – 1 tablet once daily (OD) along with supportive therapy, as one 21 days cycle and two cycles planned with 15-day interval and the treatment was planned for total of 6-12 months.

The regimen was chosen considering the patient's age (renal function, hepatic function), duration of treatment and the toxicity profile of the antibiotics chosen and the dose was adjusted accordingly. Baseline and routine renal function, liver function tests and patient's hearing were checked for drug toxicities. This regimen led to a notable improvement

with reduction in discharge, pain and swelling showing early symptomatic response highlighting sensitivity of organism to the chosen antibiotics. Hence, after 10 days of the treatment the regimen was altered to Inj. Amikacin 500 mg OD, T. Rifampicin 600 mg OD and T. Cotrimoxazole (Trimethoprim 160 mg + Sulfamethoxazole 800 mg) – 1 tablet twice daily (BD) to enhance long term efficacy and minimise toxicity risks aligning with standard protocols for actinomycetoma. Therefore after 21 days of continuous treatment, patient showed symptomatic improvement (Fig 2b) and was discharged with T. Rifampicin 600 mg OD, T. Cotrimoxazole (Trimethoprim 160 mg + Sulfamethoxazole 800 mg) – 1 tablet twice daily (BD), T. Itraconazole 130 mg at bedtime (HS) as treatment for suspected mixed infection.

The patient showed clinical improvement after two cycles and sinuses were healing at 3-month follow up.

Discussion

Mycetoma is a persistent granulomatous infection produced by filamentous fungi (eumycetoma) or aerobic bacteria (actinomycetoma), with mixed cases being extremely rare [9, 10]. Tumefaction, draining sinuses, and the presence of granules can make it difficult to distinguish between the two [8, 9]. However, accurate diagnosis is essential, as the treatment approach differs significantly, including antifungal drugs for eumycetoma and prolonged antibacterial therapy for actinomycetoma [6].

Here, based on the presence of broad septate hyphae and black grains on the KOH mount, eumycetoma was initially diagnosed, for which the patient was prescribed itraconazole, a standard azole therapy for fungal mycetoma [9], but lack of improvement after several weeks led to re-examination. Histopathological report revealed basophilic filamentous sulphur granules along with PAS-positive organisms and good clinical response to a regimen including amikacin, cotrimoxazole, and rifampicin supported the final diagnosis of actinomycetoma.

A major barrier in the treatment of mycetoma is the potential for mixed infections, and here it can be seen by this diagnostic change from eumycetoma to actinomycetoma. Although mixed mycetoma is rare, several case reports including those by Bonifaz *et al.* [10], Fahal *et al.* [3], and Mallick *et al.* [9] have shown the chance of bacterial and fungal coexistence in endemic regions causing mixed infections. Failure to properly diagnose and treat such infections may result in partial treatment responses and frequent recurrence, as fungal components dominate the superficial layers while actinomycetes invade deeper tissue [9, 10].

Recurrent mycetoma, in general, is caused by inadequate medication penetration leading to incomplete organism eradication from the fibrotic tissue [2, 7]. Culture is the most reliable method for distinguishing between eumycetoma and actinomycetoma and determining the underlying species [11]. In this particular case, definitive etiological evidence is limited because culture could not be carried out.

The condition's chronicity was likely contributed to by a delayed diagnosis, empirical antifungal treatment without culture confirmation, and the patient's long-term usage of alternative medications. Imaging excluded bone involvement, which has prevented the need for surgical debridement. Amikacin plus cotrimoxazole with rifampicin often added in extended protocols (welsh regimen)

continues to be the standard treatment for actinomycetoma, and it usually takes several months to years for it to completely resolve [2]. The standard Welsh regimen was modified based on patient-specific factors including age, renal tolerance, and early therapeutic response, making the regimen more feasible and clinically promising for long-term management.

The present case had only three months of follow-up, which limits assessment of long-term response. Early symptom improvement in this case shows the importance of proper diagnosis and regimen modifications based on antibiotic sensitivity.

The patient was scheduled for serial reviews every 4–6 weeks with documentation of sinus closure, reduction in discharge, and repeated clinical imaging. The use of adjunct itraconazole has been justified with regard to the initial diagnosis of fungal mycetoma, which suggests a possible mixed mycetoma rather than a fully actinomycotic condition.

From the perspective of public health, mycetoma is still a neglected tropical disease (NTD) that mostly affects low-income agricultural workers and is often given less importance because of limited access to diagnosis and treatment [6, 7]. For early detection and improved outcomes, healthcare professionals must be aware of mixed infections, histopathological evaluation, and the need for recurrent biopsy or culture in non-responding or persistent cases. Definitive and affordable diagnostic tests are still not available in rural areas which can differentiate mycetoma from other subcutaneous infections and prevent long term morbidity. Unlike many chronic conditions, vaccinations can be made available for mycetoma with the use of modern scientific advancements.



Fig 1: 1A Initial clinical presentation showing swelling, ulceration, and discharging sinuses with blackish grains on the right foot. 1B Partial improvement after initial itraconazole therapy.



Fig 2: 2A Recurrence of swelling and persistent sinus discharge with yellowish grains after one month of itraconazole. 2B Significant clinical improvement after antibacterial therapy.

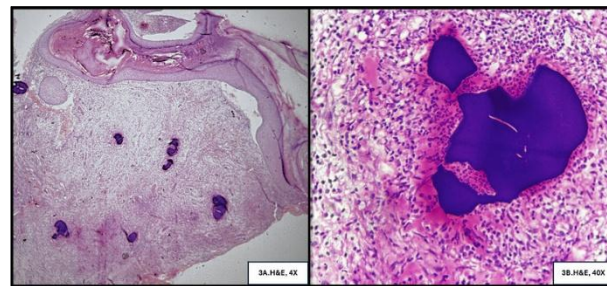


Fig 3: 3A Skin with subcutaneous tissue showing abscess cavity with multiple foci of degenerated filamentous organisms. 3B Higher magnification showing degenerated filamentous organisms morphologically favoring actinomycosis surrounded by mixed inflammatory cells.

Table 1: Laboratory results showing abnormal inflammatory markers

Sl. No	Laboratory values	Values	Interpretation
1	Haemoglobin	11.6 g/dL	reduced
2	PCV	34.3%	reduced
3	RBC	3.65 M/Cumm	reduced
4	Lymphocytes	51.4%	elevated
5	ESR	42 mm/hr	elevated
6	CRP	15 mg/L	elevated

Table 1: Laboratory findings during presentation. PCV- Packed cell volume, RBC- Red blood cell, ESR- Erythrocyte sedimentation rate, CRP- C reactive protein

Conclusion

This case serves as a reminder that, despite being uncommon, mycetoma still affects South Indian rural communities where barefoot farming is prevalent. Early recognition, repeated diagnostic evaluation, and access to basic microbiological and histopathological testing in non-responding cases can greatly improve outcomes. Improving awareness within the community and early intervention at the primary-care level can help prevent chronicity, disability, and the social burden associated with this neglected tropical disease.

Consent: Patient consent was obtained for the use of data and clinical images in the case report

Conflicts of interest: none

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