International Surgery Journal
Pandey VK et al. Int Surg J. 2026 Aug;13(8):1513-1516
https://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902

DOI: https://dx.doi.org/10.18203/2349-2902.isj20262481

Case Report

Recurrent plantar eumycetoma in a young soldier managed with wide local excision and vacuum assisted closure: a rare case report

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Received: 08 May 2026
Revised: 16 June 2026
Accepted: 17 July 2026

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ABSTRACT

Mycetoma was first reported in the year 1843 in Madurai, India by Dr. John Gill, and was then called "Madura foot". The disease is caused by true fungi (Eumycetoma) or bacteria (Actinomycetoma). Commonly, affecting farmers in the tropical and sub-tropical regions, in contact with contaminated soil. The organism gets inoculated into the skin and soft tissue after minor trauma. It causes chronic progressive granulomatous inflammation, leading to destruction of skin and soft tissue characterised by classical triad of painless soft tissue swelling, multiple sinuses and discharge of grains. The treatment consists of early recognition, histopathological confirmation of causative organism, systemic antifungal therapy and surgical excision if not responding to medication. In this article, we present a rare case of recurrent Eumycetoma in a 34-year-old male. He had a history of Eumycetoma in 2018 affecting right foot, treated with systemic antifungals. A recurrent lesion occurred after approximately five years, affecting deeper muscle layers. MRI revealed no bony involvement. He was treated with oral Voriconazole, surgical excision and Vacuum-assisted closure of defect, with a favourable outcome. No recurrence noted upto one year of follow-up. Aim was to emphasize on diagnosis and management of Mycetoma, in cases presenting with classical triad of painless soft tissue swelling, multiple sinuses and discharge of grains, especially in young adults with history of minor trauma while in contact with soil. Early recognition, systemic antifungals with wide local excision leaving behind a healthy margin, followed by vacuum-assisted closure, a high protein diet and physiotherapy, can provide favourable outcome in treatment of Eumycetoma. Reconstructive surgical options can be explored for larger defects. Case report providing level IV evidence.

Keywords: Mycetoma, Eumycetoma, Madura foot, Painless swelling, Discharging sinus, Antifungal, Surgical excision, Wide local excision, Reconstructive surgery, Vacuum-assisted closure

INTRODUCTION

Mycetoma is a rare progressive chronic granulomatous infection involving the skin and soft tissue. It was first reported in 19th century from Madurai, India and hence famously known as "Madura foot". The disease can occur due to true fungi (eumycetoma), or by bacteria (actinomycetoma). Therefore, Eumycetoma is a deep fungal infection of the skin and soft tissue caused by filamentous fungi. The most common organism causing a eumycetoma is *Madurella mycetomatis*.[1] These organisms are present in soil and are implanted in the skin after trauma. It primarily affects young men between 20 to 40 years of age, and is the prerogative of farmers who are exposed to contaminated soil during minor injuries in most cases.[2] The most common site of infection is the foot, followed by hands. Less frequently, other areas may be involved.[3] Slow progressive painless subcutaneous swelling develops, followed by multiple nodules that evolve into suppurative lesions with multiple draining sinus tracts. The sinuses discharge colonies of causative organisms.[4] Secondary infection can lead to

pain and redness. Eumycetoma is morphologically and histologically, characterized by deep granulomatous inflammation and the formation of grains, leading to the destruction of deep tissue, muscle, bone and tendons in neglected cases. Although the true incidence of mycetoma remains uncertain, 60% of mycetoma are bacterial and 40% are fungal.[5] Most of the cases fall between latitude 15°S and 30°N, the so-called "mycetoma belt" characterized by warm, dry, semi-desert regions with low rainfall.[6,7] The treatment is often challenging and requires systemic antifungal therapy (preferably triazoles) and wide local excision (WLE) if not responding to medication. In cases where a large defect occurs after excision of affected soft tissue, reconstruction surgery is required. In endemic areas, it affects the socio-economic status of the patient and their family.

CASE REPORT

This case is a 34 years old soldier, from a city in Madhya Pradesh, India. He presented with a swelling over the sole of right foot since December 2022, insidious onset, gradually progressive in size. He noticed multiple sinuses over the swelling, approximately 15 days since onset, intermittently discharging blackish grains mixed with pus. Associated with mild pain on palpation or weight bearing, which was relieved on rest. He had few episodes of intermittent low-grade fever. No history of any bleed from the lesion.

He had an episode of similar swelling in 2018, which was diagnosed as a fungal infection. He underwent a course of anti-fungal (no supportive documents available). The lesion subsided with medical therapy alone. He had no known co-morbidities, allergies, family history of such disease. No contact history of tuberculosis. He had the habit of walking bare feet in the muddy lawn of his house. No known history of any thorn prick or trauma. No history of significant unintentional weight loss or anorexia. He reported to a local medical facility after approximately three weeks from onset. On evaluation, core biopsy from the lesion revealed features suggestive of lichenoid granulomatous inflammatory lesion, likely fungal.

He was initiated on tablet Itraconazole 200mg twice daily for several months, but showed no significant improvement. He reported to our reconstructive surgery centre at Command hospital, Eastern Command, Kolkata for review.

Clinically, a 06×06 cm, foul smelling, firm, mildly tender swelling was noted at the plantar aspect of right mid-foot with scaly irregular surface and multiple discharging sinuses (Figure 1).

Figure 1: Swelling on plantar aspect of right foot, with multiple sinuses discharging black grains.

The discharge consisted of black grains mixed with purulent material. Skin around the swelling was indurated. Range of movement of the foot was full and free. No evidence of any focal neuro-vascular deficit noted. On evaluation, magnetic resonance imaging (MRI) right foot revealed a heterogeneously enhancing soft tissue lesion in plantar aspect of right, involving muscles adjacent to second, third and fourth metatarsals with peri-osteal extension (Figure 2).

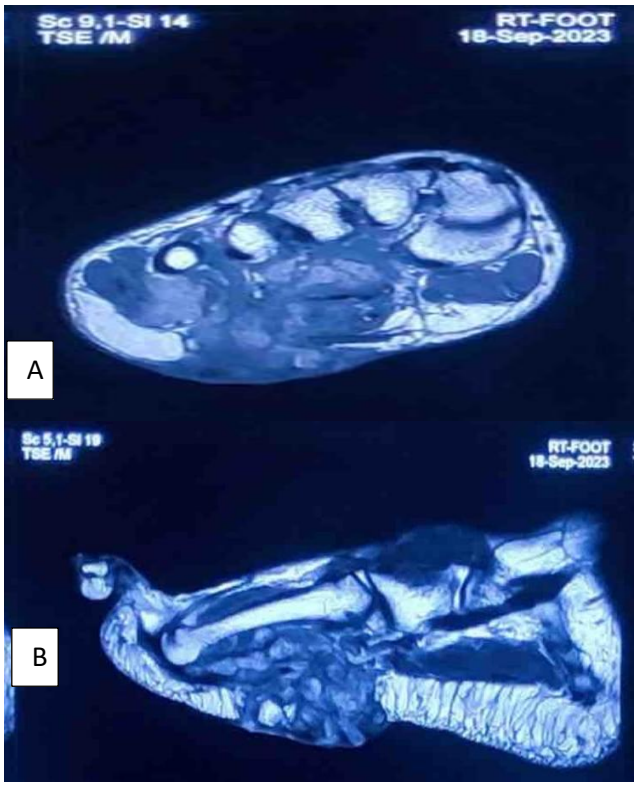

Figure 2: MRI scan of right foot revealing a soft tissue lesion infiltrating deeper structures.

Opinion of dermatologist was taken and started on tablet voriconazole 200 mg twice daily.

He was planned for excision of the lesion and reconstruction, with an impression of Eumycetoma of right foot. After pre-anaesthetic workup, he underwent excision under spinal anaesthesia on 09 November 2023. Intra-operative findings-swelling over plantar aspect of right mid-foot with multiple discharging sinuses containing black grains mixed with purulent material. The soft tissue lesion was infiltrating the muscular layer upto inter-metatarsal space (Figure 3 and 4). The bones were spared.

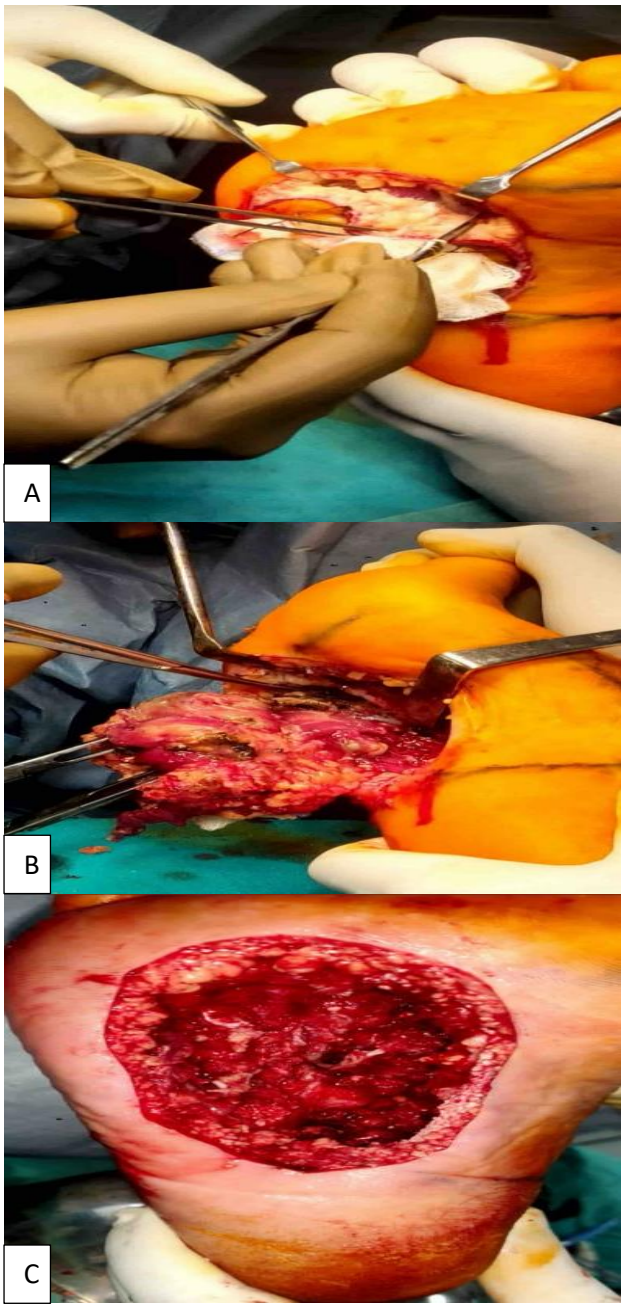

Figure 3 (A-C) Intra-operative images-note the deep tissue involvement by the presence of black grains in the inter-metatarsal space.

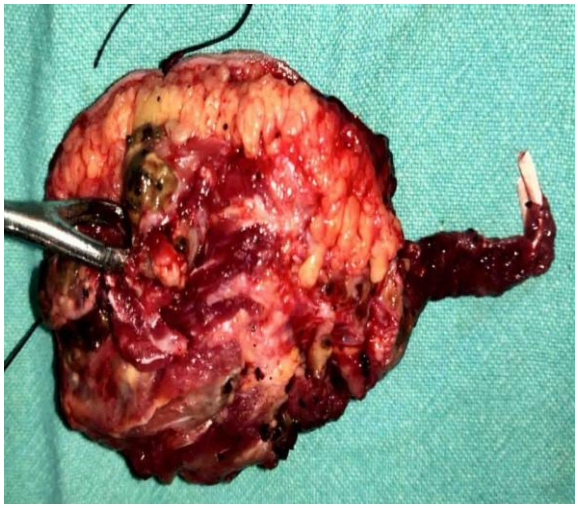

Figure 4: The excised specimen-note the black grains mixed with purulent material.

In view of active infection, the defect was thoroughly washed with normal saline and packed with betadine-soaked gauze and dressed. VAC therapy was initiated on post-operative day one after confirming haemostasis and healthy bed. After five days of VAC therapy, the wound was healthy with red granulation tissue bed and no visible black grains or purulent material. Histopathological examination of excised soft tissue revealed multiple scattered grains characterised by presence of branching filamentous organism in the core with homogenous eosinophilic material surrounding it. These fungal septate were surrounded by macrophages, giant cells and inflammatory infiltrate suggestive of Eumycetoma (Figure 5).

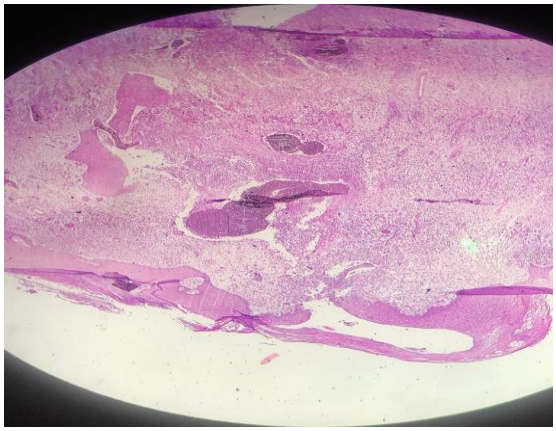

Figure 5: Slide showing microscopic view of the excised specimen.

Regular dressing of the surgical wound was done. He was discharged with medication (tablet Voriconazole) on post-operative day 12. He underwent wound care and physiotherapy at home. The defect was completely healed by secondary intention in approximately three months (Figure 6). The antifungal therapy was stopped.

Figure 6 (A and B): Healing of surgical defect by secondary intention, (A) POD-45 (B) POD-90.

He was followed up for approximately a year. He was ambulant with normal gait. Range of movements of right foot was full and free. No evidence of any neurovascular deficit noted. No evidence of any recurrence noted.

DISCUSSION

Mycetoma remains a neglected tropical disease with significant morbidity. The 30-50% of patients presenting late have functional disability and deformity, with 10-15% of advanced cases undergoing amputation.[8] Eumycetoma is commonly caused by *Madurella mycetomatis*, typically presenting with multiple sinus tracts discharging black grains. The classical triad includes tumefaction, multiple sinus tracts, discharging grains.[9] MRI is essential for evaluating extent of involvement of soft tissue and bone. The characteristic "dot-in-circle" sign is highly specific, helping in differentiating it from chronic osteomyelitis or neoplastic lesions.[10] Histopathological examination helps in confirmation of diagnosis. Medical therapy alone is mostly insuffecient in Eumycetoma, with recurrence rate of 25-50%, due to poor penetration into dense fibrotic tissue.[9] In contrast, Wide local excision with prolonged systemic anti-fungal therapy offers best outcome with reduced recurrence rate of 10-15%.[11] VAC therapy promotes faster wound closure, making it a valuable adjunct in helping early return to normal activity. Larger defects may require other reconstructive surgical options. In our case, aggressive surgical clearance of infected tissue with healthy margin, VAC of surgical site and anti-fungal therapy with Voriconazole for three months, ensured favourable outcome with no recurrence till approximately one year post-operatively.

CONCLUSION

Eumycetoma should be strongly suspected in patients presenting with chronic foot swelling with sinuses discharging black grains, in endemic areas. A history of walking barefoot or thorn prick makes it even more evident. Clinical examination and histopathological examination help in confirmation of diagnosis. MRI is done to determine the extent of involvement, thus helpful in surgical planning. Medical therapy with systemic anti-fungal and Wide local excision of the lesion is done to terminate the disease. Regular dressings of the surgical bed are done to facilitate wound healing. Once the surgical bed is healthy, VAC is an effective adjunct to facilitate early wound closure. In case of large defects post-excision of lesion, reconstructive surgical options should be considered. Followed by healthy wound closure, physiotherapy and rehabilitation helps early return to normal activity. This has proved to provide reduced risk of recurrence and better patient satisfaction.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Pandey VK, Jaiswal K, Pothina S. Recurrent plantar eumycetoma in a young soldier managed with wide local excision and vacuum assisted closure: a rare case report. Int Surg J 2026;13:1513-6.