A Unique Presentation of Eumycotic Mycetoma Involving the Left Medial Cuneiform: A Case Report

Atanu Mohanty[1], Anuraag Mohanty[2]

Abstract

Eumycotic mycetoma is a chronic, destructive fungal infection predominantly affecting the subcutaneous tissues and bones, commonly seen in tropical and subtropical regions. We report a rare case of eumycotic mycetoma involving the left medial cuneiform in a 42-year-old male factory worker from India. The patient presented with a two-year history of a painful ulcer on the sole of his foot following a thorn prick. Clinical examination revealed tumefaction, multiple discharging sinuses, and purulent exudate containing black granules, characteristic of eumycotic mycetoma. Radiological evaluation showed an osteolytic lesion localized to the medial cuneiform bone, suggestive of chronic osteomyelitis. Surgical intervention comprising debridement and saucerization of the affected bone and soft tissues was performed. Tissue biopsy confirmed the diagnosis of eumycotic mycetoma, demonstrating granulomas with fungal hyphae. The patient was treated postoperatively with oral itraconazole for one year, resulting in complete resolution of symptoms and radiological healing. This case highlights the diagnostic challenges posed by eumycotic mycetoma, the importance of integrating clinical, radiological, and histopathological findings, and the need for prolonged antifungal therapy in combination with surgical management for optimal outcomes.

Keywords: Eumycotic mycetoma, Medial cuneiform, Chronic osteomyelitis, Fungal infection, Black granules, Itraconazole therapy

Introduction

Mycetoma, a chronic and progressive infectious disease, remains a significant clinical challenge due to its insidious nature, diagnostic complexities, and limited therapeutic options. First reported in 1843 by Dr. John Gill in Madurai, India, and colloquially known as "Madura foot," this condition is now recognized as a neglected tropical disease [1]. Characterized by a granulomatous response to infection, mycetoma is caused by either fungi (Eumycetoma) or filamentous bacteria (Actinomycetoma). The etiological agents are introduced into subcutaneous tissues via traumatic inoculation, such as thorn pricks or minor abrasions, frequently occurring in individuals engaged in agricultural or manual labor.

Geographically, mycetoma is endemic in the so-called "Mycetoma belt," which includes countries such as India, Sudan, Chad, and Yemen [2]. Within this belt, environmental and socioeconomic factors contribute to the high prevalence of the disease. India, as part of this endemic region, sees a disproportionate impact among rural populations, particularly in young men who perform labor-intensive occupations [3]. The disease predominantly affects the extremities, with the foot being the most common site of involvement. However, rare presentations involving other anatomical locations, such as the medial cuneiform, pose unique challenges in diagnosis and management.

The clinical hallmark of mycetoma is a triad of painless subcutaneous swelling, multiple discharging sinuses, and purulent discharge containing characteristic granules [4]. These granules, which may be black, white, or yellow depending on the causative organism, are pathognomonic but often overlooked in initial evaluations. If left untreated, mycetoma progresses to involve deep structures, including fasciae, tendons, muscles, and bones, with resulting osteomyelitis and significant morbidity.

Case Report

A 42-year-old male factory worker presented with a persistent, painful ulcer on the sole of his left foot, which had remained unresolved for two years despite multiple courses of oral antibiotics prescribed at various healthcare facilities. The patient reported a history of a thorn prick at the site of the ulcer, which had marked the onset of his symptoms. On examination, the affected area exhibited significant tumefaction with multiple discharging sinuses exuding purulent material. The exudate contained black granules of varying shapes and sizes, a distinctive feature of Eumycetoma [5] (Fig. 1, 2, 3).

Radiological evaluation of the left foot revealed an osteolytic lesion localized to the medial cuneiform bone, characterized by cortical erosion and loss of trabecular pattern. These findings were consistent with chronic osteomyelitis, indicative of the deep-seated nature of the infection. While advanced imaging modalities such as magnetic resonance imaging (MRI) were not feasible due to resource limitations, the clinical and radiological correlation supported the provisional diagnosis.

[1]Department of Orthopaedics, Jajati Kesari Medical College and Hospital, Jajpur, Odisha, India.
[2]Department of Orthopaedics, Kalinga Institute of Medical Sciences, Bhubaneswar, Odisha, India.

Address of Correspondence
Dr. Anuraag Mohanty,
Department of Orthopaedics, Kalinga Institute of Medical Sciences, Bhubaneswar, Odisha, India.
E-mail: anuraag98@gmail.com

Received 14/07/2023; Reviewed 11/08/2023; Accepted 26/10/2023; Published 10/01/2024

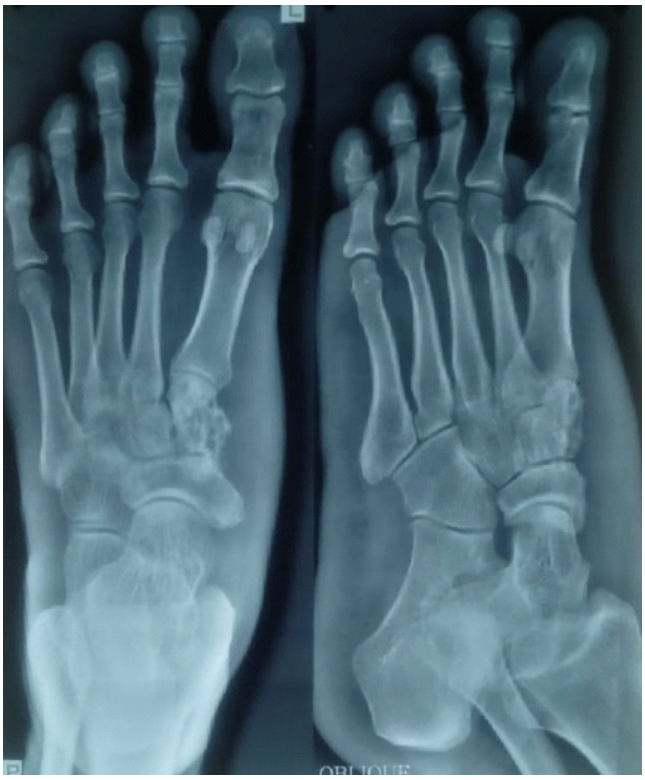

Figure 1: Radiograph of the left foot showing an osteolytic lesion localized to the medial cuneiform bone with cortical erosion and loss of trabecular pattern, consistent with chronic osteomyelitis.

Surgical intervention was deemed necessary to control the disease and prevent further progression. The patient underwent extensive debridement and saucerization of the medial cuneiform bone, along with excision of necrotic soft tissues surrounding the lesion. Tissue samples obtained during the procedure were sent for histopathological analysis. Examination revealed granulomas composed of neutrophils and histiocytes surrounding characteristic fungal grains. These grains, observed under high magnification, displayed aggregates of broad, septated, and radially arranged hyphae, confirming the diagnosis of Eumycetoma [6].

Postoperatively, the patient was started on a prolonged course of oral itraconazole. During follow-up, the patient exhibited significant clinical improvement, with resolution of the discharging sinuses and complete healing of the ulcer. At the end of one year, there was no radiological or clinical evidence of disease recurrence, underscoring the efficacy of the combined medical and surgical approach in managing this complex case.

Discussion

Eumycetoma, while rare, is a condition with profound implications for affected individuals due to its chronicity, potential for severe disability, and diagnostic complexity. The disease's pathogenesis begins with the traumatic introduction of fungal spores into subcutaneous tissues, where they elicit a granulomatous inflammatory response [4]. The prolonged incubation period and slow progression often delay diagnosis, particularly in resource-constrained settings like India, where healthcare access and awareness may be limited. In endemic regions, mycetoma primarily affects individuals with occupational exposure to soil, particularly those engaged in farming, labor, or other manual trades.

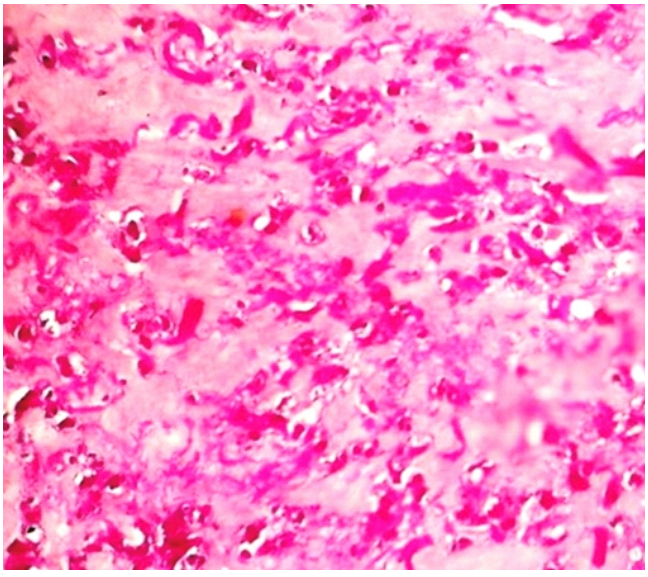

Figure 2: Histopathological section showing granulomas composed of neutrophils and histiocytes surrounding fungal grains. The grains display aggregates of broad, septated, and radially arranged hyphae.

Figure 3: Purulent discharge containing characteristic black granules of varying shapes and sizes, a pathognomonic feature of Eumycetoma.

Advanced diagnostic modalities, such as dermoscopy, MALDI-TOF, and PCR, offer precise species identification and aid in guiding antifungal therapy. However, these tools are often inaccessible in resource-limited settings, necessitating reliance on traditional methods like histopathology and culture [6].

The treatment of Eumycetoma poses significant challenges due to its chronic and refractory nature. Antifungal therapy, using agents like itraconazole and ketoconazole, forms the backbone of medical

management. Surgery, as in this case, plays a critical role in reducing the fungal burden, preventing further tissue destruction, and enhancing the efficacy of antifungal therapy [7].

Conclusion

Eumycetoma is a rare but challenging infectious disease requiring a multidisciplinary approach for effective management. This case illustrates the diagnostic hurdles and therapeutic strategies involved in addressing a rare presentation of Eumycetoma in the medial cuneiform bone. Early recognition, integration of clinical and histopathological findings, and a combination of medical and surgical interventions are critical to achieving favorable outcomes. In resource-limited settings, increasing access to diagnostic modalities and fostering awareness among healthcare providers can significantly improve the management and prognosis of this neglected tropical disease. The successful resolution of this case highlights the importance of tailored, patient-specific approaches in combating this complex condition.

References

1. Dubey N, Capoor MR, Hasan AS, Gupta A, Ramesh V. Epidemiological profile and spectrum of neglected tropical disease eumycetoma from Delhi, North India. Epidemiol Infect. 2019;147:e294. doi:10.1017/S0950268819001766.
2. Singh G, Roy P, Grover S. Madura foot: An interesting case of mycetoma reoccurring fifteen years after initial treatment. Bull Am Coll Surg. 2020;105(2):36-38.
3. van de Sande WWJ. Global burden of human mycetoma: A systematic review and meta-analysis. PLoS Negl Trop Dis. 2013;7(11):e2550. doi:10.1371/journal.pntd.0002550.
4. Krishnamoorthy A, Dinesh P, Kumaran S, et al. An overview of mycetoma and its diagnostic dilemma: Time to move on to advanced techniques. Indian J Dermatol Venereol Leprol. 2020;86(3):251-259. doi:10.25259/IJDVL_107_20.
5. Yadav A, Singh J, Rani S, et al. Eumycetoma infection of foot: A rare case report. J Orthop Surg. 2021;29(3):563-566. doi:10.1177/23094990211019123.
6. Verma P, Jha A. Madura foot: A comprehensive exploration of diagnosis and treatment. World J Pharm Med Res. 2018;4(1):201-204.
7. Priyanka AN. Mysterious mycetoma foot – A case series. IP Indian J Clin Exp Dermatol. 2023;9(1):58-60. doi:10.18231/j.ijced.2023.010.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his/her consent for his/her images and other clinical information to be reported in the Journal. The patient understands that his/her name and initials will not be published, and due efforts will be made to conceal his/her identity, but anonymity cannot be guaranteed.

Conflict of Interest: NIL
Source of Support: NIL

How to Cite this Article

Mohanty A, Mohanty A | A Unique Presentation of Eumycotic Mycetoma Involving the Left Medial Cuneiform: A Case Report | The Odisha Journal of Orthopaedics and Trauma | January 2024; 05;01:24-26 | https://doi.org/10.13107/ojot.2024.v05i01.054