

A classic case of Madura foot in South Rajasthan

Paashupat Bhanuda¹, Parthasarathi Hota², Gunjan Sharma³

From ¹Assistant Professor; ²Associate Professor; ³Surgical Resident, Department of General Surgery, Ananta Institute of Medical Sciences Udaipur, Rajasthan, India

ABSTRACT

Madura foot, known for its rarity and tendency to recur, is a chronic granulomatous disease of fungal and bacterial origins. Eumycetoma, the fungal variant, involves deeper tissues at a late stage and it is function-sparing even during the advanced stage of the disease. Clinically, diagnostic black grains, imaging, and histopathology play a critical role in clinching the diagnosis of this deep infestation. Meticulous wide surgical excision, serial debridements, and long-term antifungals provide the mainstay for the treatment of eumycetoma. We report an under-treated case of recurrence of eumycetoma in a young rural farmer who was suffering from eumycetoma for about a year. Initial sub-optimal management resulted in the disease festering over a long period of time. Most of the left midfoot was involved on the presentation without any bony involvement. Even after exhibiting all classical signs of eumycetoma at various points in time, our patient was under-treated highlighting the need for increased awareness, early diagnosis, and better long-term management of this disease.

Key words: Black grains, Debridement, Eumycetoma, Fungal, Madura foot, Recurrence

Eumycetoma, a tumor caused by fungi residing in the soil of semi-arid regions, is a destructive chronic granulomatous inflammatory disease that involves the skin, subcutaneous tissues, and sometimes even deeper structures. The so-called “Mycetoma belt” (15 South to 30 North) mostly comprising of low-income agrarian societies is the breeding ground for this tropical disease [1]. With the advent of modern medicine and increased vigilance in the mycetoma belt, the disease is becoming rarer and rarer to encounter. Historically, it has been described in ancient Indian writings as “Padavalmika” which is translated to foot anthill [2]. Madura’s foot was coined in 1842 after its modern description by Gill and the fungal origin of the disease was established in 1860 by Carter [2]. It still remains a cumbersome load on public health facilities in the aforementioned tropical regions leading to the World Health Organization classifying “Eumycetoma” as a neglected tropical disease in 2016 [3]. Highlighting the same is the fact that it can take between 3 months and 50 years from the time of infection for the patient to first seek healthcare advice [4]. The inoculation of the organism usually occurs through a recognized, or mostly unrecognized, trivial trauma in individuals who walk bare-footed in a terrain full of thorns [1]. Farmers and shepherds, who are often men between the ages of 20 and 40, are at a particularly

high risk for developing the granuloma [4]. Most of the patients belong to low-income groups resulting in delayed diagnosis, under-treatment, and incomplete long-term management of this neglected disease leading to its recurrence.

Here, we discuss a case of eumycetoma in semi-desert south Rajasthan, who was living with the disease for almost a year, and was undertreated due to a mixture of patient apathy and diagnostic dilemmas.


CASE REPORT

A 33-year-old male farmer by occupation presented with a painless ulcerative lesion over the planter aspect of the left foot, sparing the anterior and posterior foot pads, with multiple discharging sinuses and exuding sub-centimetric black granules (Fig. 1). The patient also complained of an associated swelling over the left inguinal region. On further evaluation, the patient reported a history of getting hit by a stone while working barefoot 1 year back and subsequently developing a small firm, and rounded swelling which gradually increased in size over a period of 6 months. This eventually ruptured forming a sinus with pus discharge from the left foot after which the patient sought medical attention.

On general examination, the patient was anemic, well-built, and under-nourished with a body mass index of 18. Routine investigations were within normal limits with the exception of a low hemoglobin of 10.5.

Correspondence to: Paashupat Bhanuda, J B Hospital, 22 Hiran Magri, Sector 6, Udaipur-313002, Rajasthan, India. E-mail: paashuv@gmail.com

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The patient was then managed conservatively, but due to under treatment with antifungals, the wound over the left foot failed to heal and gradually increased in size in the form of an ulcer. The patient also developed multiple discharging sinuses and started exuding black granules from the lesion. Eventually, a swelling also developed in the left inguinal region that gradually progressed to the current size.

After presentation, the lesion was examined and it showed a 7 × 6 cm ulcerative growth over the planter aspect of the midfoot sparing most of the anterior and posterior heel pad and associated with scant seropurulent discharge and multiple black granules. The non-healing ulcer (due to past instrumentation) was surrounded by thickened edges and hyperpigmented margins suggestive of the chronic nature of the lesion. In congruence with the growth, was the swelling in the left inguinal region of size 4 × 3 cm, which was firm in consistency and appeared to be clustered (not matted), suggestive of lymph nodal origin.

On further evaluation, X-ray of the left foot showed signs of tissue calcification inferior to the 5th metatarsal with normal bony architecture and no signs of scalloping (Fig. 2). Pus culture showed superinfection with *Klebsiella* species. A magnetic resonance imaging (MRI) of the left foot showed an ill-defined large ulcer proliferative lesion in the planter aspect of midfoot predominantly in the central and medial compartments associated with multiple rounded lesions seen in the intertarsal and intermetatarsal soft tissue and subcutaneous planes and in the lateral compartment of the midfoot. An irregular skin defect was also seen in the planter aspect of the midfoot measuring approx. 4 cm and showing exophytic component. No bony involvement was observed (Fig. 3). The patient underwent incision and drainage for exploration and a biopsy specimen was sent for histopathological examination. A potassium hydroxide mount showed no fungal elements.

With the imaging findings suggestive of eumycetoma, the patient was initiated on itraconazole and supportive antibiotics. Fine needle aspiration cytology (FNAC) from the left inguinal swelling showed chronic granulomatous lymphadenitis. A deep biopsy was obtained containing grains that confirmed the

granulomatous nature of the disease with septate fungal hyphae. The patient was planned for wide local excision after explaining the possibility of future recurrence and the lesion was excised with all possible attempts made to clear the intertarsal and intermetatarsal region of the fungal infestation (Fig. 4a).

The histopathological report showed acceptable free margins with fungal colonies and other features consistent with eumycetoma (Fig. 5). The patient reported back on post-operative day 15 for



Figure 2: X-ray left foot (anteroposterior/oblique) showing soft-tissue calcification without bony involvement

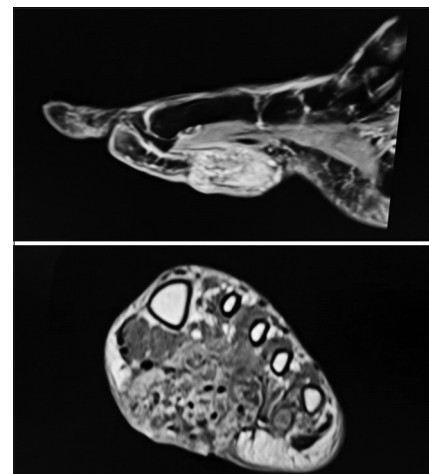


Figure 3: Magnetic resonance imaging left foot in sagittal and coronal sections showing eumycetoma without bony involvement



Figure 1: Eumycetoma on presentation

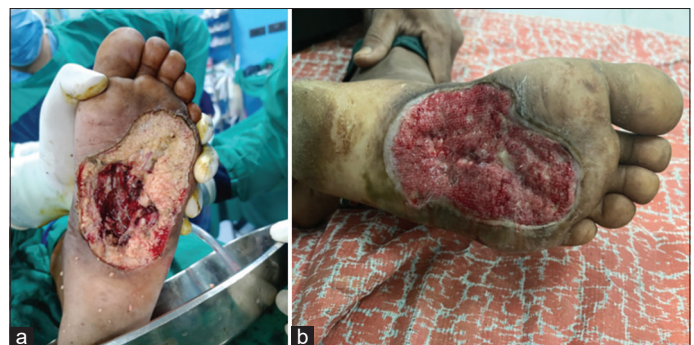


Figure 4: (a) Intraoperative picture of the wound; (b) Post-operative (2 weeks follow-up)

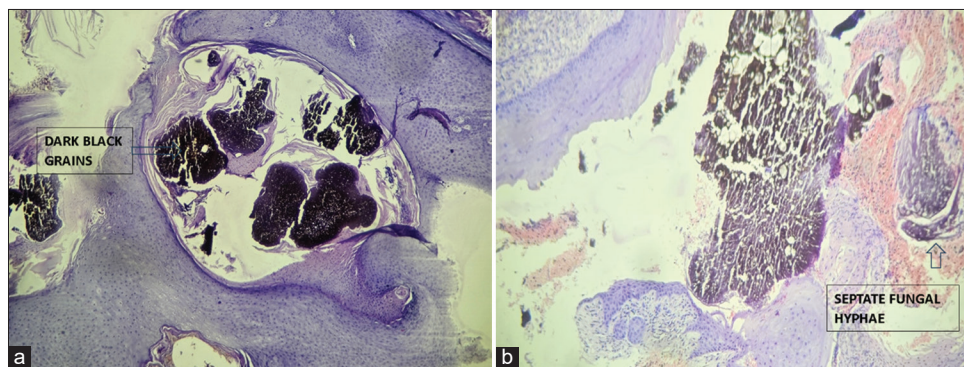


Figure 5: (a) H and E stain in $\times 10$ magnification showing dark black grains of eumycetoma in stratum corneum; (b) H and E stain in $\times 10$ magnification showing septate fungal hyphae with surrounding black grains

follow-up and the wound showed healthy red granulation tissue with no macroscopic evidence of black fungal granules (Fig. 4b). The patient was supposed to visit the hospital regularly for wound evaluation but even after multiple efforts were made to make him report to the hospital, the patient was lost to follow-up.

DISCUSSION

Eumycetoma, with its debilitating effects on patients' life, remains a challenging diagnostic "anthill" to encounter. The commonly affected areas are those that come in contact with soil during daily activities such as foot and hand, though it has also been described on the knee, arm, head, neck, and even perineum [1]. It must be distinguished from malignant melanoma, Kaposi sarcoma, fibroma, and even foreign body granuloma and cutaneous tuberculosis, endemic in most "mycetoma" countries, which also adds to the confusion surrounding the disease [5]. Most patients, especially chronic cases, also show regional lymphadenopathy which may be due to lymphatic spread of eumycetoma and secondary bacterial infection [1].

Although it remains localized, cachexia and anemia from malnutrition are common [6]. In contrast to actinomycetoma, eumycetoma involves the deeper structures at a later stage [7]. This was highlighted in the present case where even with the long-standing nature of the disease, all movements of the foot were preserved alluding to the fact that tendons and nerves are spared until late in the eumycetoma disease process [7]. This propensity of the disease leads to further complexity with patients delaying treatment. Nonetheless, our patient followed a preset course of the disease evolving to multiple secondary nodules discharging through multiple sinus tracts and the presence of quintessential black granules while the disease was running through its chronic course.

MRI is an important modality to rule out bony involvement and indicates granulomatous lesions interspersed within the fibrous tissue. Some publications have claimed a "dot in circle" sign as a characteristic feature of this condition [8]. Biopsy plays a pivotal role in assessing the host tissue reaction and is diagnostic though FNAC is also useful in being rapid and sensitive to distinguish between eumycetoma and actinomycetoma [1]. The grains exuded through the sinuses are contaminated and usually do not yield

growth on pure culture. Hence, a deep biopsy is usually what aids in the identification of the causative organism for eumycetoma [9]. Fungal culture can also be ascertained from the abundant black grains from deep tissue biopsy. Other serodiagnostic tests such as enzyme-linked immunosorbent assay and immunoelectrophoresis could also help where there is a diagnostic dilemma and fungal culture is not conclusive [1].

Eumycetoma responds only partially to antifungal therapy with itraconazole, voriconazole, and ketoconazole [10]. Oral ravuconazole has been shown to be an equally effective and much cheaper option for the management of this disease [11]. Long-term treatment with antifungals, though effective, can have serious side effects. While not curative, antifungals localize the disease by forming thickly encapsulated lesions that are responsive to surgical excision [1]. Surgery is indicated for a better response in patients with massive disease and needs to be much more extensive than initially clinically apparent as the disease usually extends into much deeper planes [12].

Many cases with eumycetoma are under-treated with excision under local anesthesia, though contraindicated, resulting in significant morbidity to the patient. The recurrence rate varies from 25% to as high as 50% and can be local or distant to regional lymph nodes [1]. In our case, the recurrence of presentation occurred due to non-compliance with drugs due to financial reasons and lack of health education.

CONCLUSION

The "classical" nature of the case is attributed to the presence of the classic pathognomonic triad of eumycetoma in the present case: Painless subcutaneous mass, multiple sinuses, and seropurulent discharge. The incidence and prevalence of eumycetoma remains enigmatic and it seems to be commonly misdiagnosed at least during the initial stages of the disease. The authors would also like to focus on the fact that due to impoverished conditions in low-income countries, a lot of sufferers with this "neglected" disease tend to be under-treated and ill-informed about the disease they are suffering from. Increased awareness among healthcare professionals can go a long way toward early diagnosis and management of this obstinate recurrent disorder and can be limb-salvaging. Simple hygienic measures like wearing appropriate

footwear while working in fields and washing hands and feet at regular intervals may help prevent the disease.

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