CASE REPORT

Madura Foot: A Rare Case of Eumycetoma and Proposed Classification and Treatment Guidelines

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ABSTRACT

Madura foot or mycetoma pedis is a chronic granulomatous disease of the foot caused by fungus or gram-positive aerobic bacteria. It is commonly encountered in tropical and subtropical countries but is rare in Malaysia. We report a 40-year-old Rohingya refugee, a resident in Malaysia for the past 11 years, who presented with a long-standing left foot mass that was proven to be a eumycetoma. Wide local excision of the mass and full-thickness skin graft were done, followed by a 6 -month course of postoperative itraconazole. He subsequently recovered well without any sequela. A high index of suspicion should be raised in a patient presenting with a chronic mass and discharging sinus in order to diagnose and treat mycetoma as early initiation of therapy leads to better outcomes. Based on literature review, we propose a new classification of eumycetoma and its treatment guidelines.

INTRODUCTION

Madura foot or mycetoma pedis is a localized chronic granulomatous disease caused by fungus (eumycetoma) or gram-positive aerobic bacteria (actinomycetoma).[1-3] First reported in 1832 by Gill the atypical multinodular appearance of legs among field workers in Madurai district of Tamil Nadu. It was termed the 'Madura foot' in 1846 by Colebrook.[4] In 1860, the term mycetoma was used by Carter to describe the fungal aetiology of this condition.[4] In 1913, Pinoy elucidated aerobic bacteria belonging to the actinomycete group as a causative agent of mycetoma.[4] Madura foot is a disease of tropical and subtropical countries such as India, South Africa and Sudan where barefoot walking is common.[4] Hitherto, there are only three reported cases of mycetoma in Malaysia in years 1968, 1969 and 1982 respectively.[1] We report a rare case of eumycetoma of the foot as it has not been described in Malaysia for the past 35 years. Based on the literature review, we propose a new classification of eumycetoma and its treatment guidelines.

CASE REPORT

Mr. R, a 40-year-old Rohingya refugee, presented with a progressively enlarging left foot mass for 4 years. Generally, the mass was painless, but there was intermittent left foot discomfort while walking for the past 6 months. There was no night or resting pain and he denied having any constitutional symptom such as fever, malaise, loss of appetite or loss of weight. There was no history of trauma or insect bite to the affected foot. No known family history of malignancy. He has been living in Malaysia for the past 11 years, working as an odd-job worker who infrequently required walking barefooted. He started seeking medical attention in the past 1 year but unfortunately, he was unable to afford the cost of treatment and hence he defaulted follow-ups.

On examination, he was a thin-built gentleman with normal gait. A multilobulated mass was seen at the dorsum of his left forefoot, measuring 6 x 10 cm (Figure 1A and 1B). There were multiple superficial sinuses at the

overlying skin of the mass with hyperpigmentation. The mass was firm in consistency, non-tender, non-mobile vertically and horizontally and the left ankle range of movement was full. No palpable regional lymph nodes. Neurovascular examination of the foot was normal. Plain radiographs of the left foot and ankle showed thickened soft tissue shadow with no calcification or adjacent bony involvement. Biochemical investigations were unremarkable. Magnetic resonance imaging of the left foot revealed soft tissue inflammation with no bony involvement. There were conglomerate of small spherical T2 hyperintense lesions with peripheral hypointense rim. Few of these lesions showed the classical "dot in circle" sign on the T2-weighted images, suggestive of a mycetoma (Figure 1C, 1D and 1E).

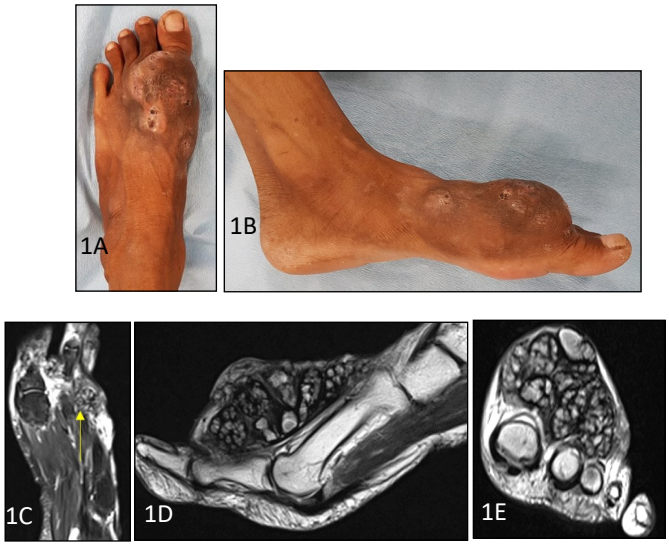

Figure 1: Figures 1A and 1B show the left foot tumour with multiple sinuses on the overlying skin. Figures 1C, 1D and 1E show MR T2-weighted images of the left foot in axial, sagittal and coronal views respectively. Multiple hypointense foci within high-intensity spherical lesions representing the surrounding granulomas can be seen, giving the typical "dot-in-circle" appearance (yellow arrow).

Wide local excision of the left foot mass and full-thickness skin graft for soft tissue closure were performed (Figure 2A and 2B). Post-operatively, he was discharged home without immediate complication. The histopathological examination of the mass showed fungal organism within a suppurative granuloma on Haematoxylin and Eosin (H&E) stain and fungal hyphae was positive on Grocott's Methenamine Silver stain, hence confirming the diagnosis of eumycetoma (Figure 2D, 2E and 2F). Thereupon, he was commenced on oral antifungal (itraconazole 200mg twice daily) for 6 months. During follow-up at 6 months, his wound at the left foot had healed well with normal range of movement of his left ankle and all toes (Figure 2C). No recurrence was noted, and he was discharged well.

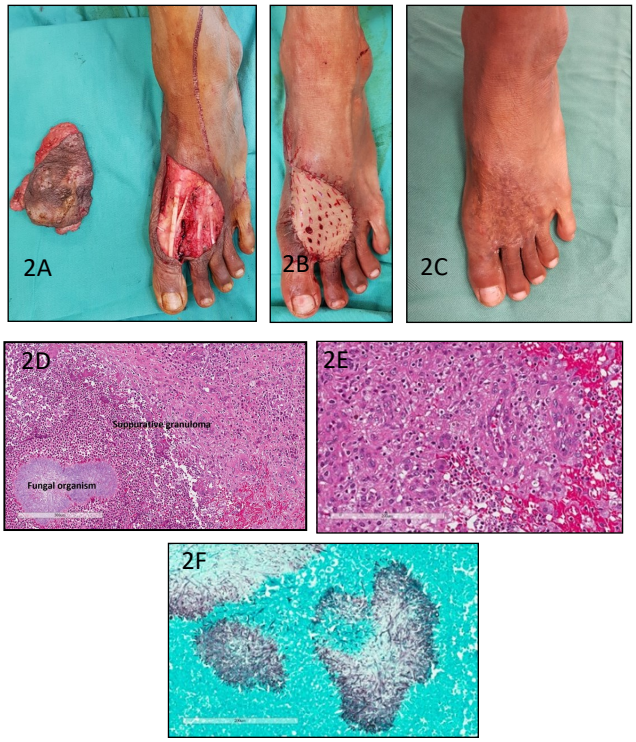

Figure 2: Figure 2A shows the intraoperative figure after wide local excision of the left foot mycetoma. Figure 2B shows the initial wound picture post full-thickness skin graft closure of the wound. Figure 2C shows the well-healed wound at 9 months after surgery. The patient has good range of movement of the left ankle and all toes with no recurrence. Figure 2D shows the fungal organism (yellow arrow) surrounded by suppurative granuloma (H&E stain, 100x magnification). Figure 2E shows the granuloma is composed of epitheloid cells, lymphocytes and multinucleated giant cells at 200x magnification. Figure 2F shows positive fungal hyphae on Grocott's Methenamine Silver stain at 20x magnification.

DISCUSSION

Madura foot is often a result following traumatic inoculation of subcutaneous tissue with contaminated soil through thorn pricks, pre-existing abrasions or trauma. The infection subsequently advances to the surrounding soft tissues and bones.[3,4] Symptoms may take up to 12 years to develop as this disease progresses slowly following the traumatic inoculation.[3] Our patient has the typical presentation of mycetoma as affected patients often present with chronic indurated swelling with draining sinuses and discharging fistulas.[3]

In order to distinguish fungal or bacterial aetiology of mycetoma, a deep biopsy is mandatory as it is more superior compared to culture in identification of the causative organism.[3] Identification of the causative agent is paramount in treating this disease as patients with actinomycetomas are treated with antibiotics while patients with eumycetomas are treated with antifungal.

Although clinical and histopathological studies are adequate in confirming the diagnosis of mycetoma, radiological studies have an indispensable role in the diagnosis and management of mycetoma. Magnetic resonance imaging (MRI) is often helpful in determining the level of proximal extension as well as bone and joint involvement of the infection.[5] Besides that, fungal grains are often depicted on MRI as hypointense foci within high-intensity spherical lesions representing the surrounding granulomas on T2-weighted images.[4,5] This is termed as "dot-in-circle" appearance which is highly specific for mycetoma.[4,5]

Management of mycetoma is tailored to the aetiology of the mycetoma. Medical therapy, either antibacterial or antifungal agent, is the mainstay of treatment for mycetoma. In some cases of mycetomas, surgical interventions are indicated and their indications are highlighted in Table 1.[3]

A review by Suleiman et al stratifies mycetoma into 3 different categories: small lesions (<5cm) without bone involvement; moderate lesions (5-10cm) with bone involvement; and massive lesions (>10cm) with bone involvement and secondary bacterial infection. We propose a modified classification, adding a new category viz lesions 5-10 cm without bone involvement (Table 2). This is because treatment varies between eumycetomas with bone involvement and those without bone involvement. In patients with large lesions, antifungal therapy is initiated pre-operatively for 6 months to promote the formation of pericapsular fibrous tissue surrounding the lesion to facilitate the delineation of normal and pathological tissue intra-operatively.[3] Local excision with wide margin is advisable for patients with moderate or massive lesions in view of their high recurrence rate.[3] In patients with atypical presentation, wide local excision is beneficial to prevent a suboptimal surgical resection of a locally aggressive or malignant tumour. Our patient presented with a moderate lesion (5-10 cm) without bony involvement, hence we proceeded with a single-stage wide local excision with wound coverage and 6-month postoperative itraconazole.

Table I: Indications of surgical treatment of mycetoma

1.	Small, well-localized lesions
2.	Patients not responding to medical therapies
3.	To reduce disease burden in large lesions for better response to medical therapy
4.	To control secondary bacterial infection
5.	Medical therapies are contraindicated, eg: pregnancy or lactation
6.	Bone and joint involvement
7.	As life-saving procedure in advance disease complicated by sepsis or poor general condition.

In this case, single-stage surgery is the best option. In order to minimize the risk of local recurrence, we opted for wide excision which had left a wound at the dorsum of his foot. We decided for split-skin grafting to facilitate wound closure to prevent the exposure of extensor tendons that will lead to drying of tendons. A full-thickness skin graft or flap will be less optimum as the dorsum of the foot will be bulky after healing, potentially leading to ulcer formation in a poorly fitted shoe. Six months of antifungal should be prescribed to patients with moderate or massive lesions while small lesions warrant an antifungal therapy of 3 months duration.[3] He subsequently recovered well with no sequela or recurrence.

Eumycetoma is extremely rare, as evidenced by only three cases reported in Malaysia, with no cases reported for the past 35 years. Owing to the rarity of this disease, treatment of eumycetoma is individualised based on patient's condition. Nevertheless, Suleiman et al had proposed a classification system that may guide the management of this rare disease based on his case series and literature review.[3] Our case did not fit in any of the 3 categories that he had described, hence we thought that the modified classification with 4 categories will be a

Table II: The proposed eumycetoma classification and treatment guidelines

Category	Descriptions	Proposed Treatment	Note
1	Lesions <5cm without bone involvement	Wide local excision, followed by antifungal for 3 months	Follow up for recurrence
2	Lesions 5-10cm without bone involvement	Wide local excision, followed by antifungal for 6 months	Follow up for recurrence
3	Lesions 5-10cm with bone involvement	Antifungal for 6 months, followed by wide local excision and another 6 months of antifungal	Follow up for recurrence
4	Lesions >10cm with bone involvement with/without secondary bacterial infection	Antifungal for 6 months (with debridement(s) in case of secondary bacterial infection), followed by wide local excision and subsequent antifungal for 6 months	Consider amputation in cases where: Patient presented with life threatening sepsis Multiple recurrences after both surgical and medical therapies Function of the affected lower limb is expected to be worse after limb-salvaging therapies compared to amputation.

better guide to treat this disease. In certain cases, pre-operative antifungal is prescribed to promote formation of pericapsular fibrous tissue surrounding the lesion to facilitate the delineation of normal and pathological tissue intra-operatively.[3] In our case, wide excision was performed as we were able to delineate the tumour and the reactive zone through the magnetic resonance images. Besides that, even though the tumour was big, there was no bony involvement based on MR images, thus we did not think pre-operative antifungal therapy would offer any benefit in terms of optimising the treatment outcome of this patient. Nevertheless, we prescribed 6-month post-operative antifungal instead of 3 months as proposed by Suleiman et al in category 1 (lesion < 5 cm with no bone involvement) owing to the large tumour size (5-10 cm with no bone involvement) and presence of soft tissue inflammation detected on MR images. Even though a wide excision was performed, it was impossible to remove the whole reactive zone as we needed to preserve extensor tendons for proper function of the foot. Removing the extensor tendons would lead to clawing of toes due to unopposed action of flexor tendons.

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Due to the rarity of this disease, we were not able to provide any literature to support this modified classification. Nevertheless, this case report and modified classification could provide a framework for the development of future studies.

In a nutshell, mycetoma is a rare disease in Malaysia. Treating physicians should have a high index of suspicion to identify and treat this disease as early diagnosis and initiation of appropriate therapy would lead to a better outcome. We propose a new classification of eumycetoma and its treatment guidelines.

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