

## Mistaken identity: eumycetoma masquerading as squamous cell carcinoma

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We report a case of eumycetoma in a Filipino patient who presented with a solitary reddish brown, moist, multinodular tumor on the dorsum of the left foot of 2 years duration. Biopsy with Periodic acid Schiff (PAS) & Gomori methenamine silver (GMS) staining, fungal culture, ultrasound and X-ray of the foot were done in our institution which confirmed the diagnosis of eumycetoma. The patient was successfully treated with itraconazole 400/day for 3 months, followed by 200mg/day for the succeeding 9 months, leading to complete resolution of the lesion leaving an atrophic hypopigmented scar. A high index of suspicion supported by diagnostic tests aided in the early detection of the disease which also resulted to complete resolution of the disease.

*Keywords: Mycetoma, fungal infections, itraconazole*

### INTRODUCTION

Eumycetoma is a rare deep fungal infection characterized by the triad of draining sinuses, tumefaction and presence of grains. The disease spreads worldwide in the mycetoma belt which stretches between latitude 15° south and 30° north equators, and is found in countries like Sudan, Somalia, Senegal, India, Yemen, Mexico, and Argentina. It is also endemic in tropical and subtropical agricultural countries like the Philippines. Due to the poor socio-economic condition and low living standards of people living in these areas, the disease is often neglected in the initial stage.

The foot is the most common site of infection. 1,2 Only 4 cases have been reported in our institution from 2004 to 2014. Eumycetoma may appear clinically as a tumor and maybe misdiagnosed as squamous cell carcinoma or other chronic granulomatous infections caused by bacteria. Therefore, thorough investigation is required to establish the diagnosis and provide the appropriate management.

### CASE REPORT

A 37-year-old Filipino female presented with a tumor on the dorsum of the left foot of 2 years duration, which started from a small punctured wound. She consulted at an orthopedic facility, where the lesion was diagnosed as squamous cell carcinoma. Incisional biopsy was then performed which suggested a possibility of fungal infection hence the patient was referred to our institution.



**Figure 1.** Solitary reddish, brown, moist, multi-nodular tumor on the distal third of the left foot

Physical examination showed a 7 x 9 cm reddish brown, moist, multi-nodular tumor on the distal third of the left foot extending from 1st to 4th digit (Figure 1). Black granules were expressed from the lesion (Figure 2).

A repeat incisional wedge biopsy on the lesion was done which revealed granulomas surrounding sulfur granules, and in which the fungal elements were highlighted by PAS and GMS stains (Figure 3). Ultrasound of the left foot showed “circle-in-dot” sign while X-ray revealed a periosteal reaction (Figure 4). Fungal culture revealed *Madurella mycetomatis*, which confirmed the etiologic agent. Clinical, histopathological, radiological, and fungal culture findings confirmed the diagnosis of eumycetoma.

The patient was started on itraconazole 400mg/day for 3 months and 200 mg/day for the succeeding 9 months leading to complete resolution of the lesion leaving residual atrophic hypopigmented scar (Figure 5). The liver function tests at baseline, every 2 months and after completion of treatment were normal.

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**Figure 2.** Black Eumycetoma grain

## CASE DISCUSSION

Mycetoma was first described in the mid-19th century where it was then known as “Madura foot”, after the province from where it was first identified.<sup>1,3,4</sup> It can be caused by either aerobic bacteria (actinomycetoma) or fungi (eumycetoma).<sup>4,5</sup> Several fungal species cause eumycetoma, with *Madurella mycetomatis*, being the most common causative agent.<sup>5</sup> Eumycetoma is listed in the World Health Organization’s list of neglected diseases and deserves attention as it can be prevented.<sup>2</sup> Moreover, it is frequently misdiagnosed as a neoplasm, a chronic bacterial disease, or a tuberculous infection.<sup>2</sup> In our case, it masqueraded as squamous cell carcinoma.

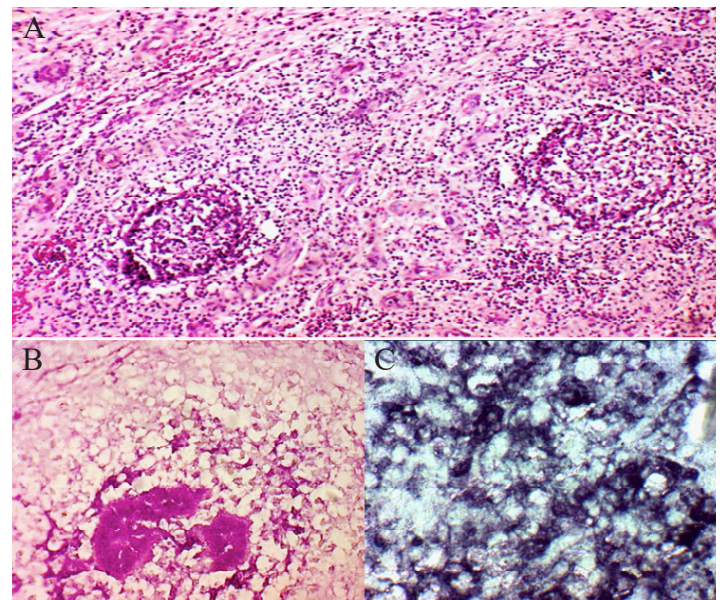
Eumycetoma presents as a gradually enlarging painless tumor at a site of previous trauma.<sup>5</sup> It commonly affects the lower extremities and is defined by the clinical triad of draining sinuses, tumefaction, and the presence of grains.<sup>5,6</sup> Our patient fulfilled the criteria for the diagnosis for eumycetoma. While the condition may be limited to the epidermis, dermis and subcutaneous

tissue, it may also lead to progressive destruction of the bone and cause significant deformity.<sup>2</sup>

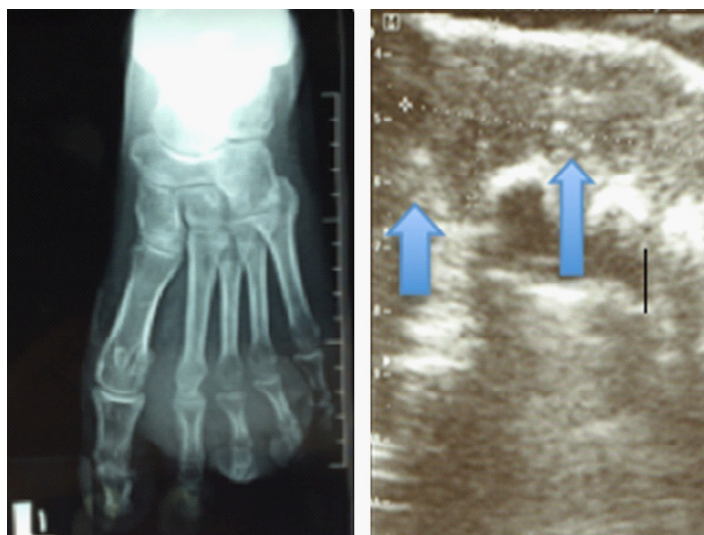
Biopsy, microbiological cultures, and radiological imaging provide a definitive diagnosis for eumycetoma.<sup>2</sup> Hematoxylin and Eosin stain would reveal sulfur granules with surrounding granulomatous reaction, which was seen in our patient.<sup>4</sup> PAS and GMS highlight the fungal elements.<sup>4</sup> Culture on Sabourauds dextrose agar will demonstrate the etiologic agent.<sup>4</sup> In our case, it revealed *Madurella mycetomatis*.

Radiological imaging is useful in determining the extent of the disease, including soft tissue and bone involvement. Eumycetoma grains produce hyperechoic echoes on ultrasound, presenting as a “dot in circle” sign, which is highly characteristic. Radiographs may be normal, demonstrate soft tissue enlargement, bone sclerosis, bone cavities, periosteal reaction, bone expansion, extrinsic cortical scalloping, fanning of the rays or osteoporosis. Ultrasound and X-ray of the left foot of our patient showed the “dot-in-circle” sign and periosteal reaction, respectively.<sup>3</sup>

Some cases of Eumycetoma have been treated with



**Figure 3.** (A) Sulfur granules with surrounding neutrophils (Haematoxylin and Eosin stain, x 100) (B) Periodic acid-schiff stain highlighted the fungal elements (x 400) (C) Gomori methenamine silver stain of tissue specimen outlined the septated hyphae (x 1000)



**Figure 3.** Radiologic findings of the left foot. (A) X-ray revealing periosteal reaction (B) Ultrasound showing “dot-in-circle” sign

surgical debulking or amputation of the affected part. However, we opted a non-invasive approach to save the affected limb. Our patient was treated with itraconazole 400 mg/day for 3 months and 200 mg/day for the succeeding 9 months leading to complete resolution of the lesion. We followed the regimen suggested by Fahal and colleagues in which itraconazole 200-400mg/day was given for an extended period of time with a mean duration of 6 months to 3 years.<sup>2</sup>

Mahgoub reported a successful outcome among 8 out of 12 patients with eumycetoma who were treated with itraconazole 200 mg daily.<sup>2</sup> Resnik et al also reported that Itraconazole proved to be effective, safe and well-tolerated therapy to be given to eumycetoma patients. The patient should have a regular check-up for at least 2 years after the end of treatment.<sup>7</sup> The prognosis of eumycetoma depends on how early the diagnosis is established and when appropriate management is initiated.<sup>8</sup> Furthermore,





**Figure 4.** (A) Marked improvement noted after 2 months of treatment with Itraconazole (B) Complete resolution of the lesion noted after 12 months of treatment

## CONCLUSION

the degree of fibrosis and underlying deep tissue damage might delay functional recovery despite aggressive management.<sup>9</sup> Patient was initially advised to have an amputation, however, we achieved success with conservative management and was able to preserve the affected limb.

In endemic areas, mycetoma should always be included as a differential diagnosis of tumor on the foot. The case highlights the importance of thorough investigation to establish the diagnosis of eumycetoma. Early diagnosis and proper management are important to prevent massive tissue damage, deformity and disability. ■

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