Stage III Eumycetoma successfully treated with Ketoconazole and Surgical Debulking

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Abstract

Mycetoma is a chronic, debilitating, granulomatous disease affecting the subcutaneous tissue, fascia, muscle, bone and adjacent organs characterised by triad of tumefaction, draining sinus, and grains. Ten-year incidence at our institution is 3/81,015. We present a 33-year old male with a 9-year history of painless nodules with draining sinuses on the left foot unresponsive to oral antibiotics and topical antifungals. Biopsy of the nodule was consistent with mycetoma. Fungal culture revealed Madurella mycetomatis growth. Xray of the left foot showed poorly marginated lucencies on the calcaneus. Ultrasound of the left foot revealed mixed hyper reflective echoes and multiple small cavities. Diagnosis was Stage III Eumycetoma. Ketoconazole 200mg twice daily was given for 9 months achieving 50-60% decrease in lesion size. Surgical debunking was done and Ketoconazole continued for 9 months. There was good granulation tissue formation and no appearance of new lesions.

Research Institute for Tropical Medicine

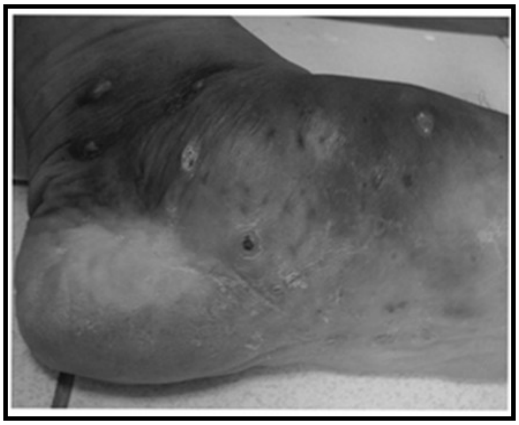
INTRODUCTION

Eumycetoma is a chronic, granulomatous disease involving the skin, subcutaneous tissue, fascia, muscle, bone and adjacent organs. Lesions evolve over months to years and may spread to involve deeper structures resulting in destruction, deformity, loss of function and even amputation.

CASE REPORT

We present a 33-year old male with a 9-year history of painless nodules with draining sinuses on the left foot. Lesion started as a solitary erythematous, pruritic nodules on the sole of the left foot after walking barefoot in the rice fields. It developed draining sinus tracts with black grains and purulent discharge. Patient applied topical anti fungal and took Cloxacillin 500mg four times for 2 weeks with no improvement. The lesions continued to spread to the dorsum and lateral aspect of the left foot, hence consult. On dermatologic examination, there were multiple ill-defined hyper pigmented and skin-colorer nodules and sinuses with erosions and scales on the medial and plantar area of the left foot. Some areas of scarring and localised edema surrounded the nodules. On the dorsal aspect, there was a well-defined hyper pigmented, hypertrophic plaque with few skin-colorer nodules with sinuses. On the lateral aspect, there were some erosions, scarring and scales.

Biopsy showed sulfur granules demonstrating Splendore-Hoeppli phenomenon with hyphae-like structures positive with Periodic Acid Schiff stain. Fungal culture revealed Madurella mycetomatis growth. Xray showed poorly marginated licences on the calcaneus and navicular bones. Ultrasound revealed mixed hyper reflective echoes and multiple small cavities. Diagnosis is Stage III Eumycetoma.



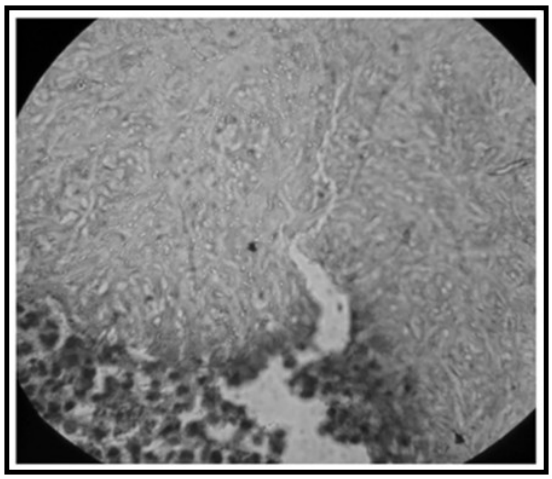


Figure 2: Hyphae-like structures in the granules (H&E, HPO x 40)

Ketoconazole 200mg twice daily was given for 9 months achieving 50-60% decrease in lesion size after which surgical debulking by wide excision of the nodules was done. Ketoconazole was continued for another 9 months. There was good granulation tissue formation and no appearance of new lesions.

DISCUSSION

Eumycetoma occurs in the tropics with low annual rainfall and relative humidity. It is common in males during the 2nd-4th decades, agricultural workers, or those who walk barefoot in dry, dusty condition. Minor trauma allows pathogens to enter the skin, which are then implanted into the hist tissue. Most common site is the dorsal aspect of the foot (79.2%) similar to that of our patient.[2]

Early stages are manifested by firm, painless nodules that spread slowly with development of draining sinus tracts over the surface. Grains discharged from sinuses vary in size, color and consistency. Lesions painlessly burrow deeply until it reaches the bone.[4]

New Radiographic classification of bone involvement in pedal mycetoma

PATTERN OF SPREAD	STAGE	EFFECT	FINDINGS
Limited to entry site	0	Soft-tissue swelling	No bone involvement
Expanding granuloma	I	Extrinsic pressure	Displacement or scalloping
Impending bone invasion	II	Bone irritation	Periosteal reaction or reactive sclerosis
Localized bone invasion	III	Erosion or cavitation	Solitary bone involvement
Longitudinal spread	IV	Joint involvement	Localized along single ray
Horizontal spread	V	Invasion of adjacent structures	Localized to forefoot, midfoot or hindfoot
Multidirectional spread	VI	Total disruption	Multiple rays and multiple rows involved

Figure 3: New radiographic Classification in bone involvement in pedal mycetoma.

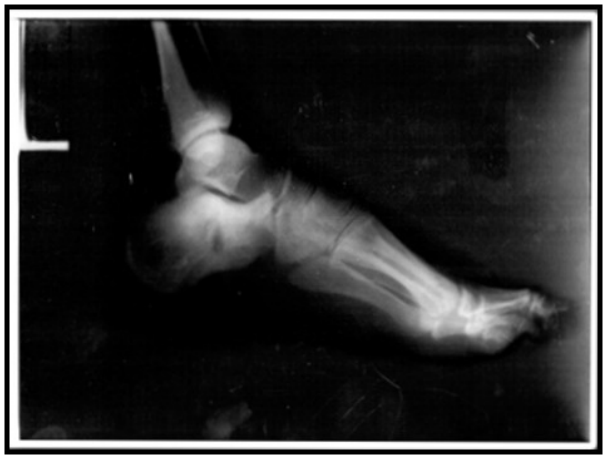


Figure 4: Radiographic imaging.

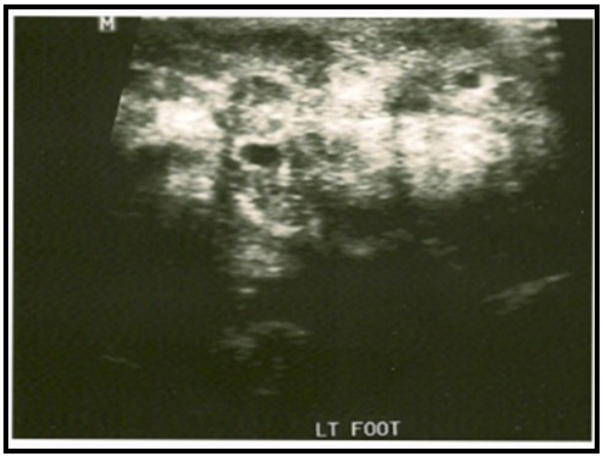


Figure 5: Ultrasonic imaging.

Clinical features do not always provide reliable measure of the extent and spread of disease. Diagnosis of Eumycetoma may be made through clinical findings, histology, mycology, radiology and ultrasonic imaging.[5]

It is necessary to determine the extent and severity of bone involvement through radiography since all mycetoma agents are osteophilic. This can aid the clinician decide the proper treatment in order to halt the progression of the disease and prevent recurrence. The radiographic parameters include the type of bone involvement, direction and pattern of spread and location of bone lesion in the foot. Based on the radiographic classification of bone involvement in pedal mycetoma, our patient belongs to Stage III (localized bone involvement) with note of cavitation on the calcaneus and navicular bones.[1]

Ultrasound imaging can differentiate between eumycetoma and actinomycetoma and between mycetoma and other non-mycetoma lesions. In eumycetoma, there are numerous, isolated sharp bright hyperreflective echoes corresponding to the grains in the lesion which was the finding in our case. Single or multiple thick-walled cavities with no acoustic enhancement are commonly identified and the cavities may contain debris and filaments. In actinomycetoma, the findings are similar but the grains are less distinct.[1]

The goals of treatment are to eradicate infection, to slow the course of the disease thereby halting the progression of deformity and preventing untoward amputation. Ketoconazole is given at a dose of 400 to 800mg daily. It has better coverage against organisms producing black grains than white or pale grains. Treatment may continue for periods ranging from months to years. It is usually stopped with clinical, mycological, radiological and ultrasonic cure.[3]

Eumycetoma are only partially responsive to anti-fungal therapy but can be reared by surgical debunking due to their normally well-circumscribed nature.[5]

Our patient successfully responded to oral Ketoconazole at 200mg twice daily for 18 months and

surgical debulking. There was no appearance of new lesions. Repeat culture was negative. Repeat X-ray of the left foot showed on the calcaneus and navicular bones and ultrasound revealed absence of hyperreflective echoes and cavities.

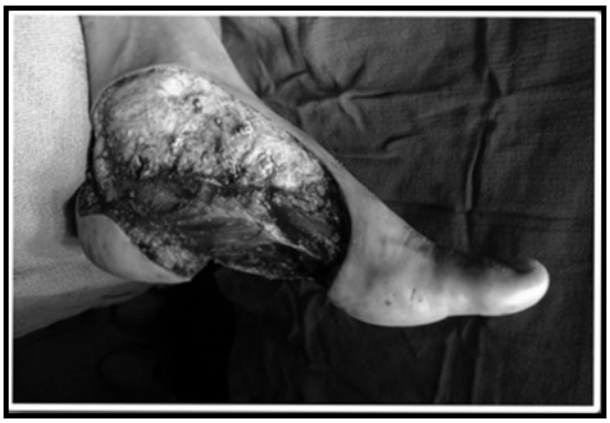


Figure 6: Immediately after surgical debulking.

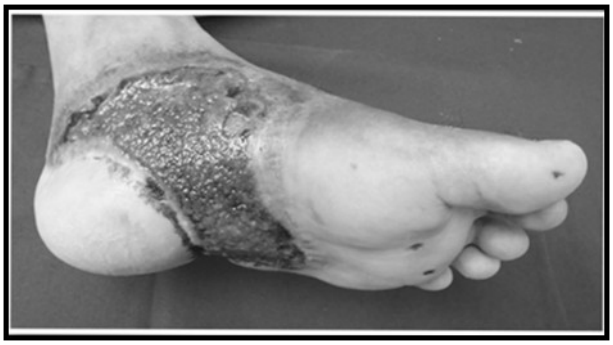


Figure 7: Two months after surgical debulking.

CONCLUSION

Our patient is a confirmed case of Stage III Eumycetoma through clinical findings, histology, mycology, radiology and ultrasonic imaging. Eumycetoma must be considered when confronted with persistent nodules on the foot and if not treated appropriately can lead to unwanted deformities and even amputation. Institution of medical treatment and surgical debunking in this case was curative and has improved our patient's quality of life.

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