

Case Report

A chronic case of *Madurella mycetoma* involving left foot

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ABSTRACT

Mycetoma is a localized chronic, suppurative, and deforming granulomatous disorder of subcutaneous tissue, skin and bones. Mycetoma is endemic in tropical and subtropical areas, where it is a real public health issue. It is a rare and neglected disease in our country. Etiological classification divides it into eumycetoma caused by fungus, and actinomycetoma caused by bacteria. Since the treatment of these two etiologies is completely different, a definite diagnosis after microbiological and histopathological examination, though difficult, is important. The disease is notoriously difficult to treat. Eumycetoma may be unresponsive to standard antifungal therapy. Authors report a case of eumycetoma in a 38 year-old male from Bihar, with history of thorn prick 30 yrs back. *Madurella mycetomatis* was isolated from the granular discharge from sinuses and itraconazole was given followed by surgical debridement of the lesion. The patient recovered well and is still on follow up.

Keywords: Diabetes, Eumycetoma, Sinuses, Surgery

INTRODUCTION

Mycetoma is a chronic suppurative infection of the skin and subcutaneous tissues that affects the lower limbs in more than 80% of cases.¹ It is characterized by a symptomatic triad: swelling of the affected area, multiple sinus formation, and a purulent discharge containing grains.²⁻⁴ The grains represent microcolonies of the causative agent. The progression of the disease is slow and painless, but may affect deep structures such as muscles, tendons, joints, fascia and bones.³ Patients are otherwise in a good health condition with no satellite adenopathy's.^{2,5}

The etiologic agents of mycetoma are aerobic and anaerobic bacteria (actinomycetoma) and fungi (eumycetoma). In the tissue, these organisms are found in compact aggregates of filamentous bacterial colonies or fungal hyphae, which will characterize the grains. These structures are the fundamental elements of the disease.

The bacteria are represented by delicate filaments of about 1µm or less, which are known as actinomycotic grains, while fungi are formed by thicker and coarser filaments, measuring 2µm or more in diameter, known as eumycotic grains.⁴⁻⁶

Managing such infections is still challenging and treatment should involve early diagnosis, the use of antibiotics or systemic antifungals, and surgical removal of the lesions.^{4,7} Mycetoma remains a real cause of disability among population living in rural area because this infection is often neglected in the initial stage. Authors present here a case of madura foot in a 38 yrs. old patient from Bihar, diagnosed and treated accordingly with good clinical response.

CASE REPORT

A 38 year-old man, a rural worker, with no records of traveling to tropical areas, comes to the consultation, with

history of multiple discharging sinuses over left dorsum and sole of foot with appearance of black granules, for 20 yrs. He also complained of swelling over the left foot since 4 yrs associated with few ulcers, over past 1 yr (Figures 1 and 2).



Figure 1: Multiple discharging sinuses with ulcers on the left foot.



Figure 2: Multiple discharging sinuses oozing black grains at the sole of left foot.

The pt. gives history of thorn prick, 23 yrs back. Few years back he had gone to the local practitioner where excision of the lesion with wound closure was performed. He was put on analgesic and antibiotic without any improvement. After one year of surgery, small swelling of 0.5 cm appeared over dorsum of left foot which ruptured to discharge black granules.

Then multiple such lesion appeared at an interval of 6 months over dorsum of the foot. Lesion ruptured to leave ulcer covered with necrotic slough. Examination revealed swelling over left foot, surface shiny and hyperpigmented with multiple sinuses (active and inactive) discharging purulent exudate which contained black granules and

presence of 3-4 ulcers on the surface. The black grains were hard in consistency with different shapes and sizes. Black grains suggested a fungal etiology. There was no regional lymphadenopathy and no other abnormalities were found.

Radiological findings showed erosive arthritis of tarso-metatarsal joints with secondary demineralization. (Figure 3).



Figure 3: X-Ray of left foot showing erosive arthritis of tarso-metatarsal joints with secondary demineralization.

The grains were collected by pressing sinus from periphery thereby enhancing discharge mixed with granules, which was collected on a sterile gauze. The grains were crushed between two slides and were examined under 10% KOH which revealed wide interwoven hyphae with large, globose swollen cells (chlamydospores) (Figure 4 and 5).

The discharge was subjected to Gram stain that showed no microorganism. Few grains were washed several times in sterile saline, crushed and cultured on Sabouraud dextrose agar (SDA) and were incubated both at 25°C and 37°C. Culture at 25°C showed scanty growth after 3 weeks but culture at 37°C showed yellowish-brown color colonies which were smooth, folded, glabrous in consistency giving brown diffusible pigment in agar making reverse of the tube dark brown (Figure 6).

On PAS staining of the crushed grains showed septate hyphae with large swollen cells interspersed between cementing substances were seen (Figure 7). So based on clinical examination, color and appearance of grains, microbiology, histopathology and radiological findings, a final diagnosis of Eumycetoma caused by *Madurella mycetomatis* was made.

The patient was put on Itraconazole 400mg for 3 months following which surgical debridement of lesion was done. The patient condition improved with no active sinus at the time of discharge. He is still on antifungals and is being followed up.

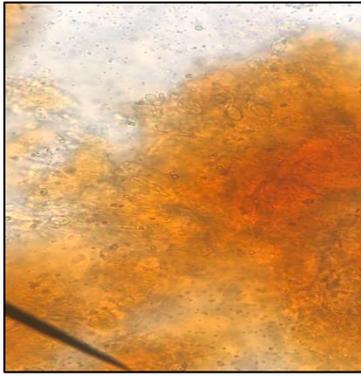


Figure 4: KOH mount of the crushed grains shows wide interwoven hyphae with large, globose swollen cells (chlamydospores).

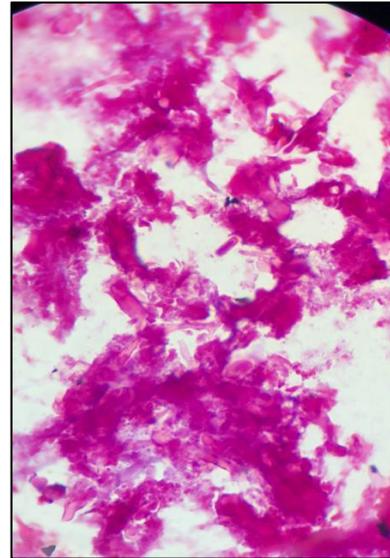


Figure 7: PAS staining of the crushed grains- showing septate hyphae with large swollen cells interspersed between cementing substances.

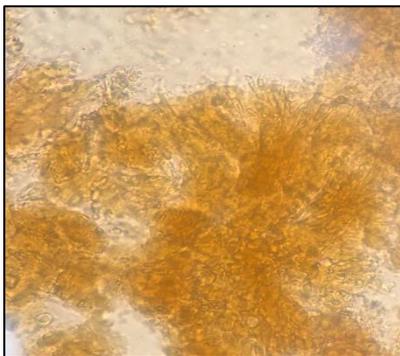


Figure 5: KOH mount of the crushed grains: showing palisade arrangement of the hyphae.



Figure 6: On SDA Culture- yellowish-brown color colonies which is smooth, folded and glabrous in consistency giving brown diffusible pigment in agar making reverse of the tube dark brown.

DISCUSSION

Mycetoma is a slowly progressive, chronic granulomatous inflammatory response involving bacteria or fungi that triggers the formation of grains which contains aggregates of the causative organisms. The triad of 1) tumefaction of affected tissue 2) formation of multiple draining sinuses and 3) presence of granules in discharge, clearly defines the disease.⁸ Mycetoma is more prevalent in developing countries as compared to the developed countries and the incidence is more in rural areas. In India, mycetoma is widely distributed in different geographical areas with larger number of cases in Tamil Nadu, the moist southern part and in Rajasthan, the dry western part of India.

Actinomycetoma is more common in India especially in Southern and eastern India, with predominance of eumycetoma mainly in Rajasthan. However, sporadic cases of either type are reported from various parts of the country and epidemiology is fast changing due to change in climate and human activities. It is more commonly seen in young adults of 20-40yrs and is more common in male than female with ratio of 3.5:1 respectively. This could be explained by the fact that young male are more often involved in field work and hence more prone to trauma and thorn prick injury.⁹

The type of mycetoma can be suggested by the color of the grains. Red grains indicates actinomycotic mycetoma whereas black grains are consistent with a eumycetoma. A white and yellow colored grain are indicative of either actinomycetoma or eumycetoma.¹⁰ The differentiation between eumycetoma and actinomycetoma is important as the treatment in both cases differ.¹¹

The combination of the clinical specific lesions, type of granules, microbiological and histopathological appearance (presence of a granulomatous inflammatory reaction with abscesses containing granules of the infecting organism) is characteristic of the diagnosis.

X-rays, tomography, and magnetic resonance imaging are all useful to look for the extension of the lesions in bone and other tissues.¹² Combined medical and surgical treatment is the standard approach for mycetoma. The medical treatment for actinomycetes consists of antibiotic therapy (cotrimoxazole, amikacin or minocycline) and for eumycetoma it is antifungal therapy (ketoconazole or itraconazole).¹³ Various antifungals are recommended in resistant cases of eumycetoma such as terbinafine, voriconazole, posaconazole, caspofungin and andulafungin.¹⁴

A prospective study showed that itraconazole at a starting dose of 400 mg and then tapering it down to 200 mg for the treatment of patients with mycetoma due to *Madurella mycetomatis* is safe, well tolerated with minimal side effects. It is also recommended to give itraconazole at a higher dose (400 mg) preoperatively to facilitate localization of lesion by fibrosis.¹⁵ Surgery is indicated in mycetoma for better response to medical treatment in patients with massive disease or for localized lesions. It is also indicated for cases that are resistant to medical treatment. The surgical options range from local excisions to amputations. Amputation is recommended in advanced cases of mycetoma not responding to medical treatment and one with severe secondary bacterial infection.¹⁶ Therefore, it is important to diagnose and treat mycetoma at the earliest.

CONCLUSION

Mycetoma's involves prolonged treatment and consequences of mycetomas are devastating. As it requires aggressive treatment and mutilating surgeries, more attention should be given to improve the quality of life for those affected. Prompt diagnosis is required to initiate early management of the disease and to reduce morbidity. Knowledge gaps must be bridged and awareness on mycetoma must reach health professionals working in primary health care, so that they can diagnose and refer patients to specialist at the earliest.

Those working in rural areas must be given proper guidance and preventive measures to promote the use of adequate shoes and should be offered proper clothing. Eumycetoma is a fungal disease that requires an early and accurate diagnosis. Medical and surgical treatment could be of great benefit for these patients but a delay in diagnosis can lead to functional and esthetical impairments.

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