Case Report

A case report on eumycetoma: An uncommon disease in the suburban areas of Maharashtra

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ABSTRACT

Mycetoma is an uncommon chronic granulomatous infection of cutaneous and subcutaneous tissues. The causal agents are soil saprophytes with around 33 species identified, namely bacteria or fungi. The World Health Organization categorized mycetoma as a neglected tropical disease. In India, the southern states have a high prevalence. Our case report presents the case of a 37-year-old male patient who presented to the hospital for diagnosis and treatment of foot swelling in his right foot region. Initially, the cause of the lesion was unknown. After radiology and laboratory investigations, the case was diagnosed with Mycotic Mycetoma or Eumycetoma. Although this study reports a single case, the findings might expand our understanding of mycetoma and its prevalence, even in the uncommon suburban regions of Maharashtra. Reporting neglected tropical diseases can alert healthcare about their spread and global burden, even in uncommon areas.

Key words: Eumycetoma, Foot swelling, Mycotic mycetoma, Neglected disease, Tropical disease

Mycetoma is a chronic granulomatous infection of the skin and subcutaneous tissue from which grains of causative organisms are eliminated through the sinus tract. This progressive, destructive disease can be caused by either filamentous bacteria known as Actinomycetoma or true fungi called Eumycetoma, in other words, Mycotic Mycetoma infection [1]. The causal agents are soil saprophytes; at least 23 species of true fungi and 10 species of actinomycetes have been identified [2]. The passage of fungal or bacterial agents is mainly through cuts or wounds that encounter the soil. In the tissue, the granules are arranged as grains. A small nodule will arise which gradually grows into a large subcutaneous mass with sinuses that secrete pus and grains [3]. Typically, mycetoma affects the upper and lower limbs but can also manifest in other body parts [3]. The first documented clinical case of mycetoma was described in 1842 by Gill [4]. This condition is frequently observed in Asian and African countries, with a limited disease burden in the United States of America. The World Health Organization has classified this disease as the most neglected tropical disease in medicine. Mycotic mycetoma is commonly found in tropical regions and is also referred to as Madura's foot, named after the initial reported case in the township of Madurai [5].

Access this article online

Received - 22 December 2023
Initial Review - 06 January 2024
Accepted - 26 February 2024

DOI: 10.32677/ijcr.v10i3.4405

Quick Response code

Although the prevalence rate of eumycetoma in the central areas of Maharashtra is less, the occurrence is still not negligible and cannot be ignored. Our case report highlights a case of eumycetoma seen in the suburban land of Mumbai, Maharashtra.

CASE REPORT

A 37-year-old male patient reported to the outpatient department complaining of swelling in the right foot region for the past 3 months. The discomfort was mostly seen in the foot sole area which caused a slight difficulty while walking. On inquiry, the patient described that he used to jog daily barefoot for 30 Min-1 h in his next-door park but did not give any history of interstate travel. The patient did not have a loss of appetite or loss of weight and denied previous traumatic injury to the affected site.

The patient has been on a combination of metformin 500 mg and vildagliptin 50 mg twice a day for diabetes mellitus for the past year and has no other co-morbidities. No significant surgical history was found. The patient was afebrile with a pulse of 78 bpm, a blood pressure of 150/90 mmHg, and a SpO_2 of 98%. During the physical examination, the patient was found to have regular heart sounds and clear lung sounds on auscultation. It was noticed that the swelling was on the sole of the right foot region,

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diffused, palpable, and mildly tender. No signs of inflammation were seen.

To gauge the extent of the lesion, magnetic resonance imaging (MRI) of the right foot was done. Radiographically, MRI reports confirmed the lesion size of 5.2×3.4×4.0 cm large lobulated well-defined collection in the plantar muscles, predominantly quadratus plantae at the level of second to fourth metatarsal bones with two focal extensions into the subcutaneous region and superiorly extending into the first and third intermetatarsal regions (Fig.1a and 1b). Mild-to-moderate edema in the adjoining region was observed. These findings suggested of infective abscess. No bone biopsy was obtained due to the negative reports of osseous involvement from the MRI.

Laboratory values showed hemoglobin 15.3 g/dL, white blood cell count 8230/cumm, and platelet count 252×10^3/cumm. The liver function test and serum creatinine were found to be normal. The erythrocyte sedimentation rate was 20 mm/h. Blood glucose level was elevated with fasting blood sugar (FBS) of 259 mg/dL and post-prandial (PP) of 386 mg/dL. Viral hepatitis markers such as hepatitis B surface antigen and hepatitis C virus were negative.

The treatment plan included complete excision and drainage under spinal anesthesia, and the wound was kept open. A third-generation cephalosporin, namely cefpodoxime along with antifungal was given as medication, and hyperglycemic medications were continued. The blood sugar level at the time of discharge was FBS 150 mg/dL and PP 186 mg/dL.

Post-operatively, intraoperative deep tissue specimens were sent to microbiology and histopathology labs. The histopathological report revealed that the biopsy specimen of the foot sole region consisted of gray, brown, soft-to-firm tissue balls aggregating 5.0×4.0×0.8 cm, confirming the diagnosis as Mycetoma (Fig. 2a). Long chains of spores with the same characteristics were observed in the histopathology sample of the patient (Fig. 2b). Special stains such as Grocott-Gömöri's methenamine silver stain and Periodic acid-Schiff stain, which are used to identify fungal elements, were positive and suggestive of Eumycetoma with foreign body reaction (Fig. 3a and 3b). Culture and sensitivity testing were not done as a fungal infection was not suspected in the very first place.

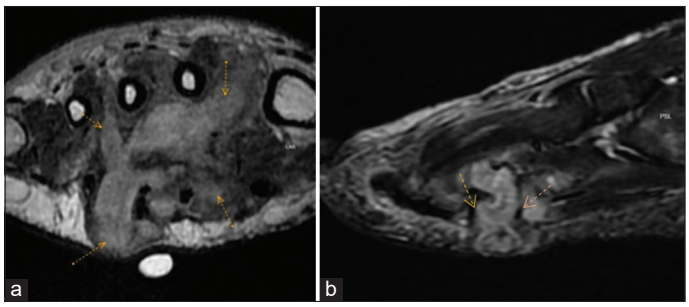

Figure 1: (a) MRI of the right foot- cross-sectional view: Radiograph showing lobulated well-defined collection in the plantar muscles. (b) MRI of the right foot- lateral view: Focal extensions into the subcutaneous region and superiorly extending into the 1st and 3rd intermetatarsal region.

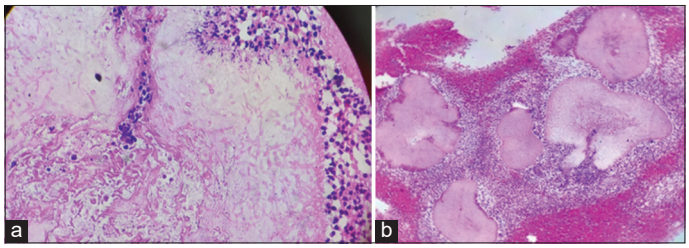

Figure 2: (a) *Madurella mycetomatis* grains showing its hyphal structure histopathology (Hematoxylin and Eosin, ×40); (b) Photomicrograph showing *Madurella mycetomatis* and inflammatory infiltrate in a cytologic smear. ×40)

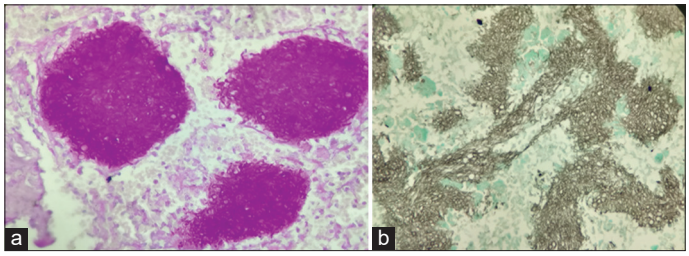

Figure 3: (a) On periodic acid–Schiff stain, positive result of eumycetoma; (b) On Grocott–Gömöri's methenamine silver stain, positive results of eumycetoma

DISCUSSION

Mycetomas can occur in any part of the body with the foot being the most common site. It is typically found in tropical or hot temperate zones. Our patient gave a history of jogging barefoot in the soil areas where any small cut paves the way for microbes to enter the body. Mycetoma is categorized as a neglected tropical disease, and the global reported number of cases is therefore 127 cases/year [3].

The term mycetoma was originally referred to as a lesion produced by fungi but was later used to describe cases involving *Actinomyces* or *Nocardia* [6]. Amid these, *Madurella Mycetomatis* is the most common causative agent for eumycetoma, while *Streptomyces somaliensis* and *Nocardia* spp. are the common actinomycetes [6-8]. Mycetomas, irrespective of the etiologic agent (actinomycetoma or eumycetoma), present similarly but require distinct treatments, underscoring the importance of accurate diagnosis [9]. The course of the disease is progressive and chronic, with old sinuses healing and new adjacent sinuses forming. Constitutional symptoms are rare, and the lesion is usually painless. The typical clinical appearance of mycetoma with indurated swelling, draining sinuses, and the presence of granules allows a tentative diagnosis. The initial lesion is usually a small, indolent, painless papule, or nodule. Subcutaneous swelling occurs, and the lesion gradually enlarges, ruptures, and forms sinus tracks. The process may involve connective tissue and bone, but tendons, muscles, and nerves are usually spared. Our case exhibits a similar pattern to that of a diffused lesion, characteristic of bacterial origin (actinomycetoma). However, in our case, the origin is fungal (eumycetoma) [9,10].

Treatment of eumycetoma mainly includes antifungals like ketoconazole with a dose of 300–400 mg and surgery of

the entire lesion; often, amputation is also considered when bone and other adjoining areas are involved. Patients have also shown a good clinical response to itraconazoles [10]. In the case of Actinomycetomas, antibiotics are the first line of therapy. Several antibiotics such as cotrimoxazole, dapsone, streptomycin, trimethoprim, rifampicin, and the amoxicillin-clavulanic acid combination have been used and found to be effective [9,10].

Differential diagnoses of eumycetoma are malignant neoplasia, tuberculosis, or nocardiosis because it spreads continuously and progressively [11]. The lesions are often histologically similar, regardless of the infectious agent. With an immunocompromised condition like diabetes, the microbial bed multiplies in number. Although the patient did not have any pus discharge, the lesion gradually increased in size, alarming the patient about the early treatment. It is observed that the recurrence rate of Eumycetoma cases is high; therefore, regular check-ups and care must be prioritized by the patient [10]. The occurrence of Eumycetoma in the urban areas of Maharashtra is infrequent compared to Actinomycetoma. Moreover, the prevalence rate is notably higher in the southern belt of India [12]. A collaborative team approach is essential, involving detailed discussions among the surgeon, infectious disease physician, pathologist, and microbiologist.

CONCLUSION

Furthermore, the extent of the global burden is unknown, as cases get missed from being reported. Early diagnosis and timely intervention can yield the best clinical outcome for such tropical diseases.

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Funding: Nil; Conflicts of interest: Nil.

How to cite this article: Shah BC, Puthenpurayil SJ, Singh DA. A case report on eumycetoma: An uncommon disease in the suburban areas of Maharashtra. Indian J Case Reports. 2024; 10(3):92-94.