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REPORT OF TWO CASES OF EUMYCETOMA IN A TERTIARY CARE HOSPITAL OF EASTERN INDIA

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ABSTRACT

Mycetoma is a chronic granulomatous infection. The causative agents are fungi and bacteria. It is commonly seen in tropical and subtropical countries in India. Two cases of eumycetoma presented here were all agricultural workers from rural south Bengal. Diagnoses were made by both histopathological and KOH mount preparation examination. Culture was negative in both of the cases. First patient was treated by amputation and second one by oral voriconazole therapy without relapse in subsequent follow-up.

INTRODUCTION

Eumycetoma is a chronic granulomatous infection of the subcutaneous tissue caused by traumatic inoculation through the skin of some types of filamentous aerobic bacteria (actinomycetoma) and some fungi (eumycetoma) [1]. It may affect muscles, bones, cartilage, and joints, mostly affecting the lower extremities, particularly the foot and characterized by the formation of sinuses fistulous tracts that discharge seropurulent exudates [2]. It is distributed worldwide, but largely confined to tropical climates [3]. Here we report two cases of eumycetoma in a tertiary care hospital of eastern India.

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CASE REPORTS

Case 1

A 43 year old woman presented with a three year history of a slowly growing mass on calcaneous and plantar region of her left foot, which exhibited erythematous nodules, cutaneous sinuses with no pain or constitutional symptoms (Fig-1). There was no history of trauma to the foot, or fever, cough, loss of weight and past history and family history were unremarkable. The patient had taken treatment from a local hospital for some time without any improvement. On general examination pallor was present, systemic examination was within normal limits. Local examination of foot showed swelling with indurations, scarring, hyperpigmentation and sinus tract on lateral side.

Lesions in right side showed puckered sinuses over malleoli and heel with discharge of minimal amount of dark colored pus from some of the sinuses. The local temperature was not raised. Routine investigations

(complete blood count, liver and renal function tests, urine examination, chest X-ray) were within normal limits. Her Hemoglobin level was 9gm/dl, erythrocytic sedimentation rate (ESR) 42mm in 1st hour and peripheral blood film showed microcytic hypochromic anemia. Rheumatoid factor (RA factor) and C reactive protein levels were within normal limit. X-ray of left foot showed cortical irregularity with lytic bone lesions over tarsal and metatarsal bones.

A surgical biopsy was taken from a sinus from which a dark colored grain was obtained after curettage. The material obtained from biopsy was sent for culture and sensitivity (C/S) for aerobic and anaerobic bacteria, acid-fast bacilli and fungi and for histopathology. All cultures for bacteria, AFB, and fungi were negative. Histopathology showed keratinized stratified squamous epithelium with formation of granulation tissue comprising of new blood vessels, lymphoplasmacytic infiltrate and young fibroblasts. (Fig-3) Dark brown material showed entangled mass of broad septate hyphae. On basis of clinical presentation and H/P, a provisional diagnosis of dark grain eumycetoma was made and the patient was started on tab itraconazole (200mg) twice a day. At the patients first follow up at one month there was no improvement. Therefore a surgical debridement was performed and itraconazole was continued in the same dose for a further of four months and patients counseled about chronic nature of disease and importance of medication compliance. However the patient showed no sign of any improvement in spite of continued antifungal therapy. At this point, the patient was counseled about treatment options and possibility of cure by an amputation followed by prosthesis was offered. Patient gave informed consent for surgery and an above ankle amputation was done. Itraconazole was continued for a period of 6 more months, thus constituting a total antimicrobial therapy for 11 months. The patient attended follow up regularly and till last follow-up examination after 9 days of stopping itraconazole, there was no recurrence in this stump clinically.

Case-2

A 58 year old woman from rural south Bengal presented with a one year history of progressive pain and swelling of left foot.(Fig-4)

There was history of walking bare foot. Physical examination revealed swelled left with several draining sinus tracts on medial aspect. Foot plain radiography showed widening of joint spaces, periosteal reaction, bone destruction and erosive changes and demineralization.The sinuses were with flat opening and were discharging serous fluid with black-colored granules. Expressed blackish discharge was collected aseptically from sinus tract. One portion of this collected material was send for histopathology and from other portion 10% KOH mount was done. In KOH preparation the mycelia clump with septate pheoid hyphae was seen.(Fig-5) Haematoxilin and eosin stain demonstrated a granulamatous response on the dermis containing grains of 0,2-0.5mm. in diameter. She failed to respond to itraconazole (200mg/day) therapy in two years of regular use of the drug and the disease showed clinical evidence of progression. The patient refused surgical resection of the limb. Therapy with oral voriconazole at a dose of 200mg twice daily was initiated, showing clinical improvement and good tolerance. At follow-up, three years later, her clinical signs had completely resolved and foot pain radiography demonstrated partial regression of periosteal reaction and bone sclerosis that suggested response to treatment.

Figure 1. Eumycotic lesions involving foot Case-1

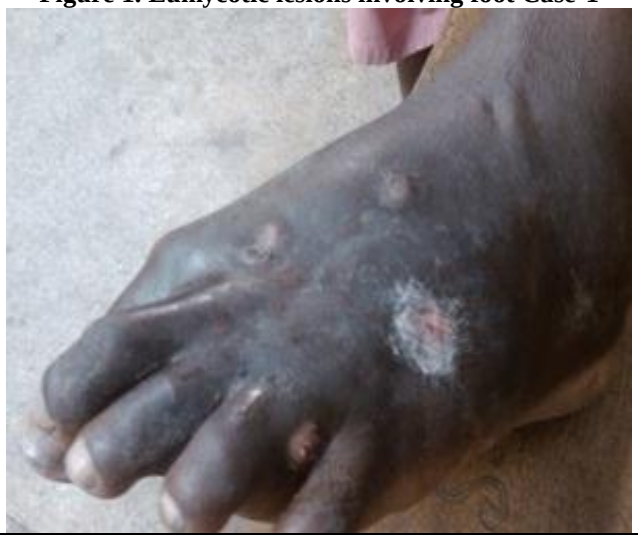


Figure 2. X-ray appearance of involved foot

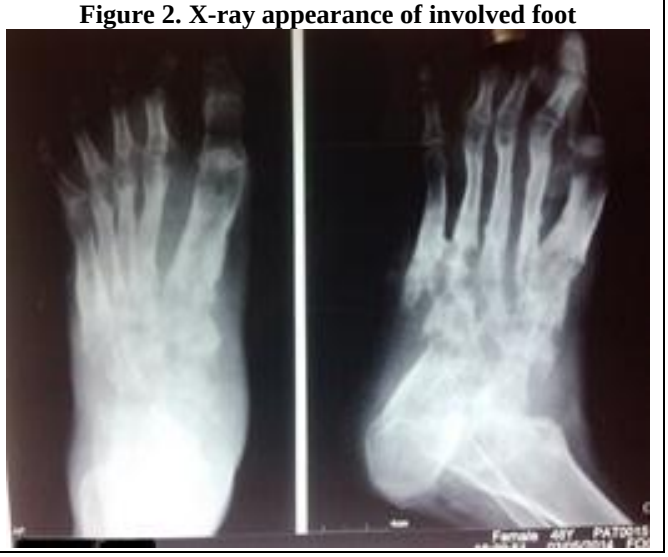


Figure 3. Histopathological appearance of grain

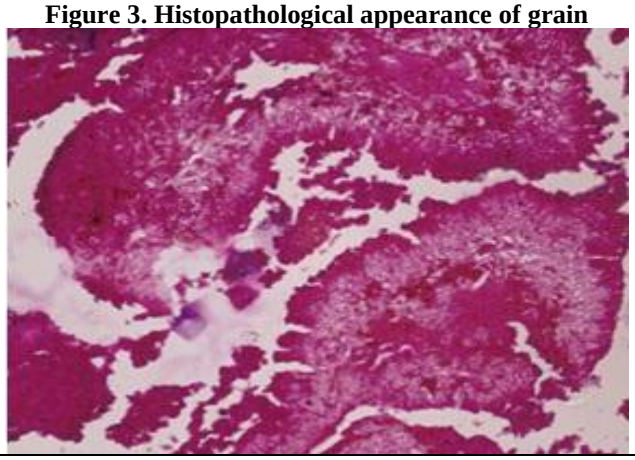
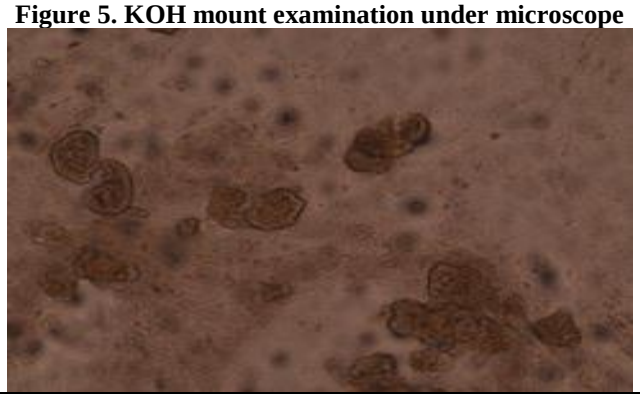


Figure 4. Mycetoma involving left foot case-2

Figure 5. KOH mount examination under microscope



DISCUSSION

Distribution of mycetoma is worldwide, but it is largely confined to tropical climates. In India, there are reports of mycetoma both from eastern and northern part [4,5]. As in our case, more than 75% of cases of mycetoma affect lower extremities, most commonly the feet, hands are affected only in about 15% cases, and lesions affecting more than one anatomical site are very uncommon [6]. The disease process can affect the skin, subcutaneous tissue, muscle and bone, spreading along the fascial planes [5]. Overlaying skin is usually smooth and shiny and may be fixed to the underlying tissue [1]. There Skin may be hypo and hyperpigmented, with tall-tale features of both old healed and active sinuses, showing the cycle of spontaneous healing of older sinus tract and continuing spread of infection to new adjacent areas [7]. In spite of extensive local involvement, the condition is relatively painless and patient's general health is usually not affected. Left untreated, the disease continues to progress and bacterial superinfection may cause morbidity from localized abscess formation, cellulitis, bacterial osteomyelitis and very rarely septicemia.

Clinical diagnosis of mycetoma is relatively easy to make given the classical triad, common differential diagnosis include bacterial osteomyelitis, botromycosis, tubercular osteomyelitis, deep fungal infections e.g. chromoblastomycosis, phaeohyphomycosis and soft tissue or bone tumors. Diagnosis of the causative etiological organism can be made by microscopic study of grains (granules) and culture; however the later may turn out to be negative even in specialized reference laboratories. On microscopic examination, the actinomycetes are recognized by filaments of .5 to 1 mm width and fungi by .2 to 5 mm broad hyphae. Various imaging modalities e.g. X-ray, USG, CT scanning and MRI are helpful for detecting the extent and spread of the disease process.

Treatment of mycetoma is challenging and depends primarily on the causative agents (bacterial and fungal) as well as severity of illness at the time of presentation. For eumycotic mycetoma, a combination of medical and aggressive surgical therapy has been usually recommended. Recently some workers have reported improved outcome as well as cure with itraconazole therapy of mycetoma caused by Madurella mycetomatis, which is the most common cause of mycetoma worldwide. Still in our case surgery was to be combined with prolonged antifungal therapy. Recent data however suggest that newer antifungals (voriconazole, posaconazole, ivavuconazole, terbinafine) may decrease

the need of aggressive surgical interventions due to their higher efficiency, better tissue penetration and bioavailability [8,9]. There use in developing countries at present is greatly limited by their availability and high cost associated prolonged course of treatment.

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