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Case Series

A Case Series of Mycetoma

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Abstract

Nocardia is a gram-positive, aerobic, acid-fast bacteria presenting a range of clinical symptoms. They arise as a result of local trauma and contamination of the site, although invasive disseminated infections are more common in immunocompromised or less robust hosts.[1] Examples of clinical manifestations include abscesses, cellulitis, and mycetoma infections. The presence of the causal organism in tissue samples, cultures, or exudates is required to support the diagnosis, which is made mostly by clinical presentation (as granules). A diagnosis may take longer than expected due to the wide range of clinical symptoms, but it is crucial for effective chemotherapy and surgery, which can help the majority of patients. The five case reports presented in this research illustrate the need for a high index of clinical suspicion, particularly in costalbelt.

Key words: Actinomycosis, Mycetoma, Mucormycois

INTRODUCTION

Tumefaction, draining sinuses, and granules in the discharging pus make up the distinctive clinical triad of mycetoma, which is simple to diagnose clinically. [1,2] Confirmatory steps include the isolation of the microorganism in culture and species identification using different biochemical reactions or molecular approaches. To prevent harsh surgical procedures like deep tissue debridement or amputation, an early diagnosis of mycetoma cases and early treatment are crucial. Mycetoma infection in non-endemic locations, a lack of clinical suspicion, a variety of clinical manifestations, and resemblances to deep mycoses are the causes of the diagnostic challenges. This article shows five clinical instances of the rare disease mycetoma while highlighting the value of a high index of clinical suspicion in terms of diagnosis and care.

Differential diagnoses include chromomycosis, blastomycosis, coccidioidomycosis, sporotrichosis, TB, botryomycosis, syphilis, yaws, and neoplasia.

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CASE SUMMARY

Case 1

A 38-year-old farmer man presented with swelling and several sinuses that had been draining for the past 4 years on his left foot, which appeared a few days after the injury [Figure 1]. Upon examination, the dorsum of the foot and the ankle were swollen and dotted with many discharge sinuses. Systemic evaluation and routine tests such as complete blood counts, serum biochemistry, urinalyses, and chest X-ray films did not reveal anything. We took a biopsy and sent it off for histological analysis. It showed focal epidermal hyperplasia, a chronic granulomatous inflammatory infiltration with neutrophils, lymphocytes, histiocytes, and a small number of plasma cells, as well as fibroblastic and vascular proliferation. Special stains like Periodic Acid Schiff (PAS) and Ziehl-Neelsen (Z-N) staining were done out for additional confirmation. Purulent material in potassium hydrogenide (KOH) mounts revealed multilobulated, vermiform grains. Based on a histological examination and the results of special stains, the case was ultimately identified as having Actinomycosis. The patient received a modified Welsh regimen of antibiotic injections Amikacin and Clotrimazole BD for three weeks.

Case 2

A 42-year-old male presented with nasal obstruction and sinusitis for the past six months. One year prior, the patient had been infected with COVID-19. On examination,

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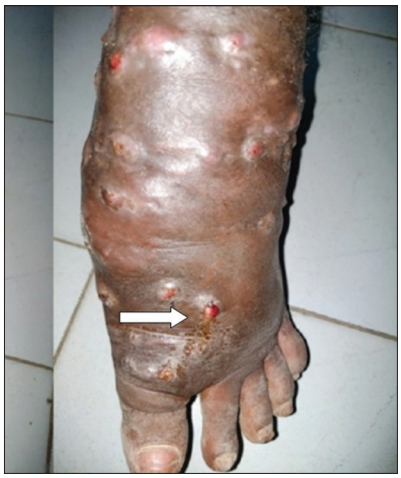

Figure 1: Severe edema, multiple sinuses, and yellowish-white discharge affecting the left foot and lower leg

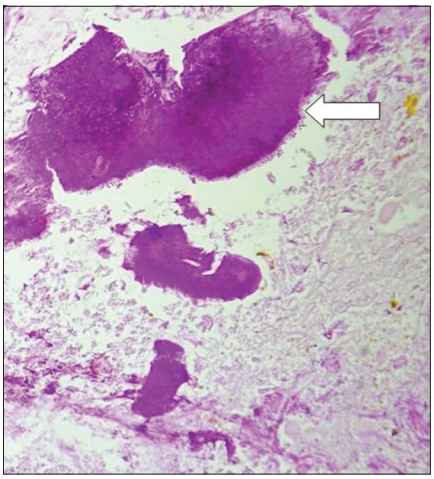

Figure 2: Actinomycosiss, filamentous bacteria on histopathology surrounded by suppurative inflammation, H&E 100X

the patient had tenderness over the bilateral maxillary sinuses. CT scan revealed necrtotic masses involved in both maxillary sinuses. Surgery was performed, and the curettage was done [Figure 2]. The tissue was sent for histopathological analysis, and revealed a suppurative and granulomatous inflammation which supported the actinomycosis diagnosis. In addition to this, there are aseptate, wide, right-angled branching hyphae exhibiting Mucor mycosis-specific morphological characteristics. Finally, a diagnosis of fungal sinusitis caused by an infection with Mucor Mycosis and Actinomycosis was made.

Case 3

A 40-year-old male was presented with left foot swelling that had been going on for 4 years. He had type 2 diabetes mellitus and was taking oral medication. The swelling has gotten worse over the last four years, and there are several dischraging drains. Upon inspection, a huge hyperpigmented swelling with many sinuses stretching

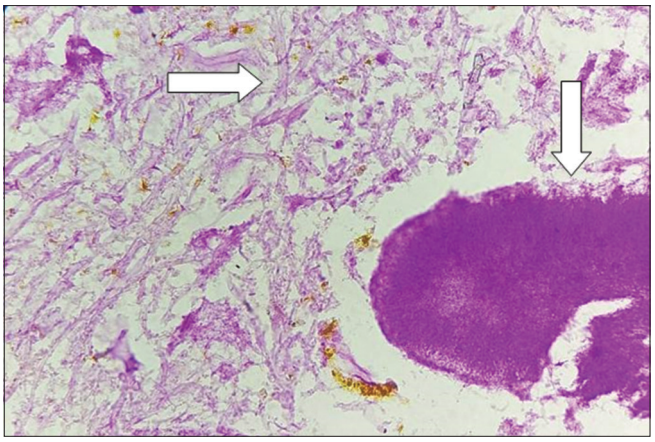

Figure 3: Mixed infection of Actinomycosis and Mucor mycosis, H&E 100X

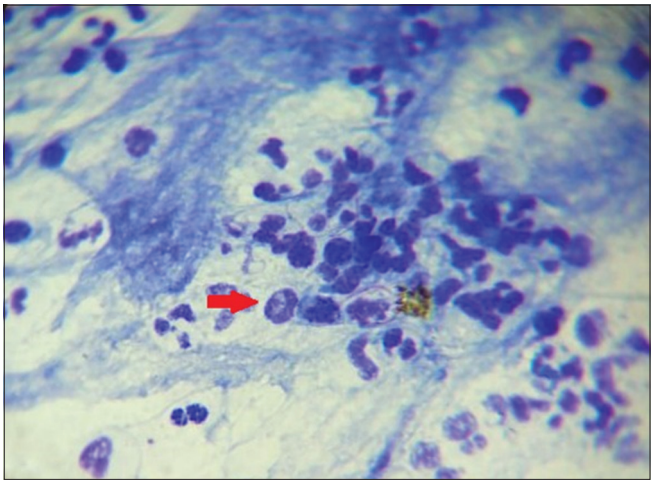

Figure 4: Long filamentous, branching bacilli on modified Acid fast staining

from the dorsum of the foot to the lower third of the left leg was discovered. The discharge was yellowish-white in colour and contained yellowish-white granules.

On histology, it was shown that the underlying tissue had a slight inflammatory infiltration and haemorrhage as well as a hyperplastic stratified squamous epithelium. Additionally, there are fungi that may indicate actinomycosis. Microbiological staining revealed the presence of Gram-positive, periodic acid-Schiff-negative, and acid-fast-positive organisms [Figure 3]. The sample was not cultured on Sabouraud dextrose agar, and there was no growth. The tarsals and metatarsals of the left foot have osteolysis, as seen on the X-ray. A computed tomography scan of the distal leg region also revealed soft tissue edoema with thickening in addition to the ongoing infection and inflammatory illness in the foot.

Clinical examinations revealed that the patient had mycetoma, and subsequent microbiological tests led to the ultimate diagnosis of Actinomycetoma caused by Nocardia.

He started taking antibiotics after being hospitalised. Along with exploration, sinus tract drainage, and debridement of sick tissue, parenteral antibiotics were initiated.

Case 4

A 45-year-old male has had swelling on his left flank for three months. He was on ART for ten years and tested positive for HIV. Upon examination, the patient had a 10x7 cm enlarged mass with several leaking sinuses. The biopsies were taken and sent for histopathological analysis. He has several erythematous, painful, nodular lesions in her right flank and lumboscral areas. He had recieved routine antibiotics and anti-tuberculosis medication at remote facilities. In addition to a few sinuses with serosanguineous discharge and creamy white granules, cutaneous examination revealed sensitive, erythematous nodules overlying the right flank.

Regular haematology, serum biochemistry, chest, and lumbar spine X-rays as well as the thorough inspection did not find any anomalies. Gram-positive, branching filamentous bacilli were found as granules in KOH mounts and Gram-stained smears [Figure 4]. Chronic inflammation and several foci of PAS-positive organisms were seen on the histology. After 3 days of incubation, an aerobic culture of the biopsy material on blood agar at 35 °C produced minute, pale-white colonies that were recognised as belonging to the Nocardia species. The final diagnosis in this case was Actinomycosis. He was prescribed co-trimoxazole, which contains trimethoprim (160 mg bid) and sulfamethoxazole (800 mg), but he failed to show up for the follow-up appointment.

Case 5

A 68-year-old woman came with a persistent, non-healing ulcer and underlying right foot edema that had been present for six months. She was diabetic in the past. During a clinical examination, the patient had a 4x3 cm enlargement, an ulcer with granulation tissue, several discharging sinuses, and oozing black granules. According to her medical history, she had initially developed several erythematous, barely itchy, papular lesions, some of which had minor purulent discharge, over her right calf. She was unable to recall any prior wounds. She was treated by general practitioners with intramuscular penicillin and anti-tuberculosis medications without experiencing a noticeable benefit. Her lesions had grown during that interval. The right calf showed extensive swelling, along with a number of sinuses, some of which were puckered and covered in adherent black-gray crusts. There was no regional lymphadenopathy.

Her systemic checkup, and regular diagnostics, including a chest X-ray, turned up no abnormalities. On blood agar at 35 °C, chalky white colonies were visible that were later determined to be Nocardia spp., branching, gram-positive, filamentous bacilli. X-ray films of the injured leg also showed soft tissue edema that suggested thickened skin beneath. The case was finally diagnosed as Madhura mycetoma.

DISCUSSION

Mycetoma, chromomycosis, blastomycosis, coccidioidomycosis, sporotrichosis, tuberculosis, botryomycosis, syphilis, yaws, and neoplasia are all differential diagnoses for chronic discharging localized disease in an extremity.[3,4] Mycetoma (Madura Foot, maduromycosis) is characterized by indolent swelling and discharge from sinuses containing granules, which are aggregates of microcolonies of the organism. It is a chronic, localized infection of the dermis and subcutaneous tissue. Approximately 40% of cases are due to true fungi (eumycetoma), and 60% are caused by aerobic actinomycetes (actinomycetoma). In most cases, the infection is caused by Nocardia brasiliensis or N. asteroids and develops after a mostly forgotten, traumatic implantation or contamination of a wound involving a limb.

The involvement of the flank, leg (as in our case), arm or thigh is unusual but frequently documented in the literature because the lesions are generally brought on by injury.[5,6] Particularly at risk are 20 to 50-year-old men and women who labor outside barefoot in rural areas. All of our patients shared, in large part, the same clinical characteristics. Our first instance experienced a left foot mycetoma as a result of a work-related accident. Since even long-standing nocardial mycetomas respond well to co-trimoxazole therapy, a high index of clinical suspicion would have averted surgery in him.

In actinomycotic and nocardial mycetoma instances, fibrosis, mutilation, and ultimately loss of function can advance quickly. Involvement of the bones happens occasionally. Without requiring surgery, two cases were responded to co-trimoxazole in an appropriate manner.

All of our patients went for a long time without receiving a diagnosis despite distinct clinical symptoms, probably as a result of a lack of clinical suspicion. The significance of an investigative work-up guided by clinical correlation, a good biopsy, and repeated microscopy for granules in pus specimens needs to be stressed to combat the issue of delayed or no diagnosis. For a successful outcome, prompt treatment commencement is essential, but the length of therapy is unpredictable.[7,8] Given that many patients experience relapses following shorter courses of medication, it must last for an extended period. The preferred first-line treatment for Nocardia infections is co-trimoxazole. The

therapeutic regimen introduced by Welsh *et al.* [9,10] consists of intravenous amikacin 500 mg, and co-trimoxazole (both in b.i.d. doses) for 3 weeks, followed by the continuation of co-trimoxazole alone for another 2 weeks.

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