

# Eumycetoma Osteomyelitis Calcaneus in Adolescent; Report of Case and Review in Literature.

**Ammar Alwad**

University of Kordofan

**Adnan Alnaser**

Omdurman Teaching Hospital

**Hozifa Abdelmaged**

Omdurman Teaching Hospital

**Reyad Abdallah**

Khartoum North Hospital

**Hussam S Khougali** (✉ [husamseddig12@gmail.com](mailto:husamseddig12@gmail.com))

Wad Madani Teaching Hospital <https://orcid.org/0000-0002-2728-9030>

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## Case report

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# Abstract

**Background:** Mycetoma is the most common deep mycosis in humans. It is a chronic, progressive, and destructive disease primarily caused by actinomycetes (98%). It involves the skin, soft tissues and occasionally bone, central nervous system as well as internal organs, and characterized by formation of black grains and poor response to treatment.

**Case presentation:** We present a case report about 19 year old male patient with annoying Right ankle pain and swelling for more than 1 year. Diagnosed initially as abscess and drainage was done 8 months ago without any improvement. No X-ray, biopsy or swab for culture and sensitivity done at time of first presentation. On his current presentation at Omdurman hospital mycetoma foot with osteomyelitis was diagnosed based on radiological and pathological assessment. Patient treated with aggressive debridement and bone curettage plus postoperative Itraconazole for 1 year.

**Conclusion:** Surgical Debridement with aggressive bone curettage followed by regular follow up and X-ray can improve the prognosis and achieve good surgical outcome with very low risk of recurrence.

## Background

Mycetoma defined as a chronic cutaneous and subcutaneous swelling caused by substantial numbers of micro-organisms, primary caused by actinomycetes (98%) [1]. It considers the most common deep mycosis in human [1, 2]. It involves the skin, soft tissues and occasionally bone, central nervous system as well as internal organs, and characterized by formation of black grains with poor response to treatment and recurrence rate [2]. Among the all mycetoma belt countries, Sudan considers to be the highest area of endemicity [3–4]. The disease is characterized by formation of chronic inflammatory granuloma, numerous deformations, disabilities and high morbidity rate, and in its late stage it is potentially fatal [5–6]. Clinically, mycetoma starts as a small painless subcutaneous mass that gradually increases in size, then multiple sinuses with seropurulent discharge that contained grains of different color and sizes develop [7]. Clinical, radiological and histopathological assessment of such lesion is deemed important for accurate diagnosis and management of osteo-articular infection. Treatment of mycetoma depends mainly on the etiological agent, site of infection, and extent of the disease [8]. This review is to highlight and demonstrate the common problems usually associate with incomplete clinical and pathological assessment of this lesion and the impact of delayed presentation or late diagnosis to the overall outcome.

## Case Report

A 19 years old male farmer from rural area in Sudan presented to our clinic complaining of hind foot swelling and pain for 1 year, started with small painless swelling that increased in size gradually and very slowly, over period of time his life style got affected since the pain started and increased with walking and activity, also limited restriction of movement occur mainly with inversion and eversion of the subtalar

joint. Sinus formation with purulent pale discharge and black grains were noted. Without proper assessment he was diagnosed as simple abscess which treated with surgical drainage under local anesthesia. No biopsies or swabs have been taken for culture and sensitivity. Post-operative oral antibiotic was given to him for 7 days. Since then deterioration of the condition was obviously seen, painful limp, swelling and limited movement on the affected side worsen dramatically. Furthermore he ends with using crutches. On clinical examination there was obvious ankle swelling and tenderness mainly at the lateral side of the hind foot, single sinus with active greenish discharge was identified [figure 1]. Hematological investigations were unremarkable, X-ray revealed calcaneus scalloping lesion forming 2 cavities posterior to the posterior facet of the calcaneus consistent with chronic osteomyelitis [Figure 2, 3]. Surgical debridement under spinal anesthesia and tourniquet was employed in lateral decubitus position with lateral extensile calcaneus approach. Debridement was done and the 2 cavities were cleaned with aggressive curettage, the black grains consist of eumycetoma confirmed by histopathology [figure 4]. Hence, Itraconazole 400 mg daily for at least 1 year was prescribed. At 2 weeks follow up visit the wound was healed nicely with no early complications. At 4 weeks follow up visit the patient came to the clinic very satisfied with post-operative results. Pain free and walking without crutches at the 2 months follow up visit. X-Ray was performed after 2 months showed no features of recurrence. Patient scheduled for monthly follow up visit and hematological investigations for the long term use of anti-fungal drug side effect.

## Discussion

Late presentation of the majority of patients is commonly seen in Sudanese patients, the justification of this delay are multifactorial, with reasons including the painless clinical nature of the lesion, poor health centers in remote areas, patients' low socioeconomic status and lack of health education.

As mentioned above the treatment of mycetoma depends mainly on the etiological agent, site of infection, and extent of the disease [8]. Until recently in Sudan, the only available treatment for mycetoma was amputation or multiple surgical excisions, as no therapeutic consensus has been reached.

Actinomycetoma is usually treated with medication only. For eumycetoma, a combination of medical treatment in the form of anti-fungal and various surgical excisions is the gold standard [9].

According to the New Radiographic Classification of Bone Involvement in Pedal mycetoma by Mohamed E. Abd El Bagi, our patient radiographs shows soft tissue involvement, cortical erosion, and central cavitation of solitary bone (calcaneus) its classified as class 3 [9]. Cortical erosion and central cavitation are commonly seen in patients with Eumycetoma Osteomyelitis, revision of X-ray by orthopedic surgeon or radiologist is always recommended in such cases to minimize the rate of the misdiagnosis.

Eumycetoma causative agents is difficult to ascertain. Hence, assessment should include full pathological analysis of the affect area such fine needle aspiration cytology and histopathology to build solid diagnosis. Fine-needle aspirations under aseptic conditions is required to identify the causative agent of mycetoma and the tissue reaction against it. Wide local excision or deep incisional biopsy taken

under local anesthesia are usually inadequate specimens, avoidance of such sample are now recommended as a Tru-Cut needle biopsy in use and above all immunohistochemistry is needed [10].

The postoperative recurrence rate varies from 25–50%, the Predictors of Post-operative mycetoma Recurrence depends on age, duration, site of involvement and no previous history of mycetoma surgical operation considers the lowest risk of recurrence [11]. Therefore, our patient was classified accordingly as low risk of recurrence.

Unfortunately since eumycetoma has a poor response to medical therapy, surgical approaches are all that is available. Many Sudanese patients undergo many operations with several regimens of ketoconazole and itraconazole to enable better response to good outcome. Surgical options for mycetoma treatment range from a wide local surgical excision to repetitive debridement excisions to amputation of the affected part. Adequate anesthesia, a bloodless field, wide local excision with adequate safety margins are mandatory for good prognosis and surgical outcome [12]. Surgical intervention usually associated with high rate of morbidity and disability among mycetoma patients in Sudan. In order to reduce rate of complication we do need to raise the awareness among the patients and families about the importance of early medical advice especially in endemic areas.

Post-operative wound care, physiotherapy and adherence of antifungal agent are mandatory for better surgical outcomes and to avoid the joint stiffness and reduce deformities and disabilities. Peri-operative and post-operative antibiotics with good dressing techniques are needed to improve the overall surgical outcome.

## **Conclusion**

Eumycetoma Osteomyelitis Calcaneus in Adolescent is extremely rare condition. Bone can be involve with mycetoma in adult as well as in children. Surgical Debridement with aggressive bone curettage followed by regular follow up and X-ray can improve the prognosis and achieve good surgical outcome with very low risk of recurrence. Generally the treatment of mycetoma osteomyelitis is case - by - case according to the predictors of post-operative recurrence.

## **Declarations**

### **Compliance with Ethical Standards**

### **Availability of data and material**

The data used in this report is available to readers.

### **Ethics approval and consent to participate:**

Authors declare that this article doesn't not involve human participant and/or animal.

### **Acknowledgement**

Not applicable

### **Consent for publication:**

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal

### **Authors contributions:**

All authors have read and approved the manuscript.

### **Corresponding author**

Correspondence to Hussam S. Khougali.

### **Competing interests**

The authors declare that they have no competing interests.

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## Figures



**Figure 1**

Sinus formation with purulent pale discharge and black grains.



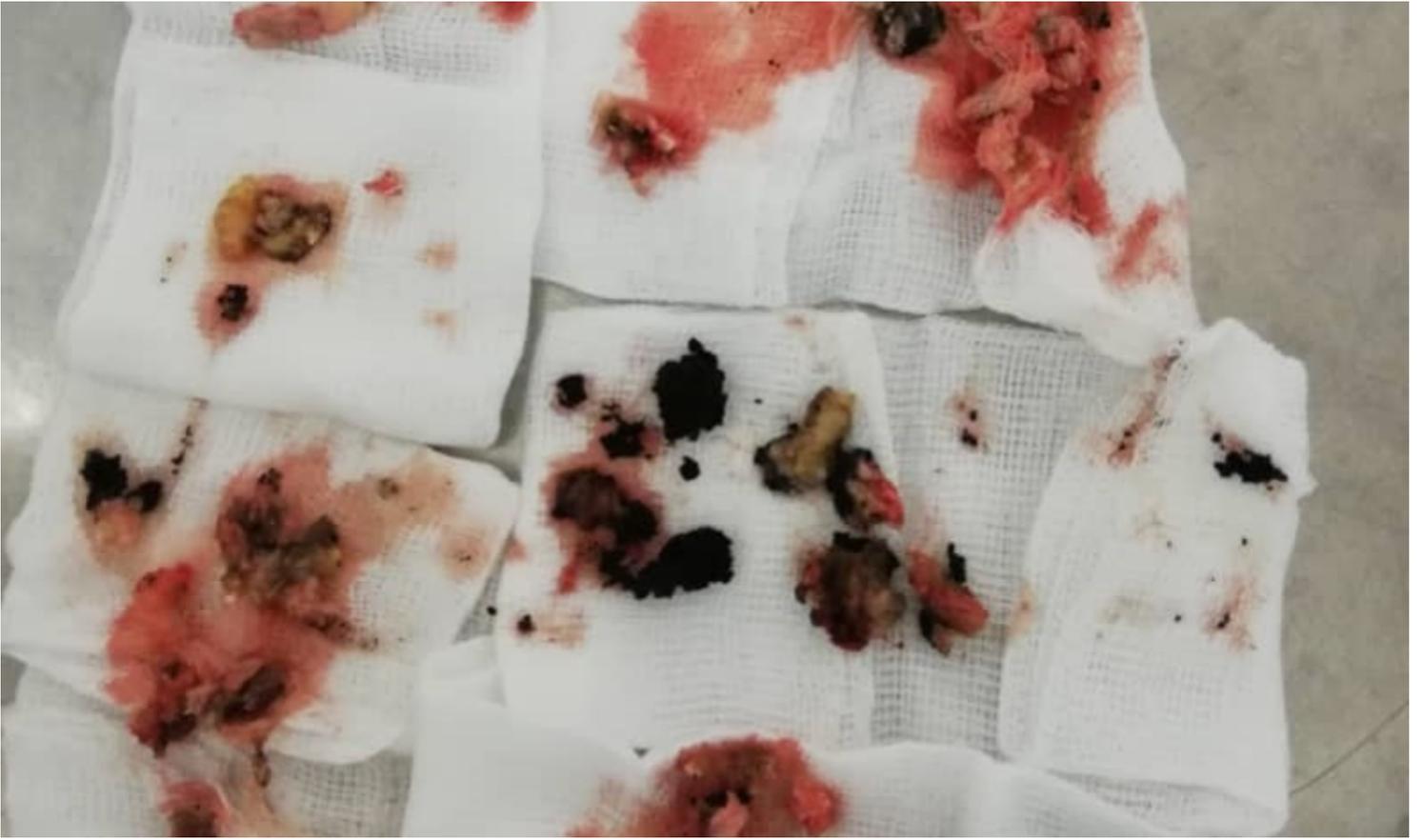
**Figure 2**

lateral view X-ray Right foot, shows features of cortical erosion and central cavitation consistent with chronic osteomyelitis in the calcaneum bone.



**Figure 3**

AP view X-ray revealed calcaneus scalloping lesion forming 2 cavities posterior to the posterior facet of the calcaneus consistent with chronic osteomyelitis.



**Figure 4**

image shows intraoperative finding and some black grains excised from the bone during curettage.