

## The Ethiopian SORT IT Course 2022

### Mycetoma patients in Ethiopia: case series from Boru Meda Hospital

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#### Abstract

**Introduction:** Mycetoma is a chronic infection that can affect the skin, subcutaneous tissue, and bone. Although Ethiopia is in the so-called mycetoma belt, very little has been published about the disease in Ethiopia. There are no data about mycetoma in Ethiopia yet. Here, we present the first detailed description of mycetoma patients in Ethiopia.

**Cases presentation:** Seven cases of clinically diagnosed mycetoma from Boru Meda Hospital are described. All patients presented with swelling of the foot, although sinuses and grains were identified for only one patient. Patients presented late with a median lesion duration of five years, and most had previously tried modern or traditional treatment. Differentiation between lesions of bacterial or fungal origin was not possible in our hospital, and therefore all patients were started on combined treatments of antifungals and antibiotics.

**Conclusions:** We confirm that mycetoma is present in Ethiopia, although there is no formal reporting system. Well-designed systematic studies are warranted to determine the exact burden of mycetoma in Ethiopia. A national strategy for mycetoma disease control should be designed with a focus on reporting, diagnosis, and management.

**Key words:** Mycetoma; case series; deep fungal infections; Skin NTDs; SORT-IT.

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#### Introduction

Mycetoma is a neglected subcutaneous infection that is caused by diverse microorganisms, both of bacterial (actinomycetoma) and fungal (eumycetoma) origin [1,2]. Its classical cases present with large tumor-like swellings, characterized by tumefaction, draining sinuses, and the presence of grains [3]. The disease mostly affects the lower extremities [2,3].

Soil is believed to be the source of infection, after traumatic inoculation of pathogens (like *Madurella mycetomatis*, and *Actinomadura madurai*) into the subcutaneous tissue when walking barefoot [4]. Further spread to the skin and deeper structures may cause progressive destruction, deformity, loss of function; and even death in rare cases [1]. Because patients typically live in rural areas far away from referral centers, health-seeking behavior is poor and many mycetoma patients present late. This can lead to massive lesions with extensive tissue destruction and complications including chronic osteomyelitis, resulting in severe

physical disabilities, economical loss, and social stigma [1,4].

Mycetoma was added to the World Health Organization list of Neglected Tropical Diseases (NTDs) in 2016 [5], but getting basic data on its epidemiology is still a big challenge. Sudan is the only country to have a national mycetoma control program in the world [5,6], while for many other countries no strategies can be devised because of a dearth of information. Part of the problem is that no surveillance and routine data registration systems exist for mycetoma [7].

The Ethiopian NTD strategic plan currently does not include mycetoma, as data is needed to formulate a mycetoma control strategy. Even though Ethiopian mycetoma cases were mentioned in a few studies [8–10] a detailed description of mycetoma cases has never been reported from Ethiopia. Here, we report for the first time a detailed description of seven mycetoma cases from Boru Meda Hospital, North East Ethiopia.

**Ethical statement**

Ethics approval was obtained from the Union Ethics Advisory Group of the Center for Operational Research at the International Union against Tuberculosis and Lung Disease, Paris, France, and Wollo University, Dessie, Ethiopia. Consent was obtained for photographs.

**Cases presentation**

Here we present seven patients who were clinically identified as having mycetoma in Boru Meda general Hospital, Amhara Region, Ethiopia in 2019 - 2020. Three dermatology professionals independently agreed on the clinical diagnosis of all cases based on photographs.

The cases are described in Table 1, and examples of cases are shown in Figures 1-3. Most patients were

**Figure 2.** Swollen, nodular, ulcerated, hyperpigmented, scaly plaque over the dorsum to the medial aspect of the left foot.



**Figure 1.** Firm, swollen, nodular, ulcerated hypo pigmented scaly plaque lesions with multiple sinuses and scarring over the dorsum of the right foot.



male, farmers, adults, and from rural areas. The median duration of the lesion was five years. Only one patient reported having trauma (with a thorn) before lesion onset, and all patients mentioned that they wore shoes, although only three wore fully protective shoes. Lesions were only located on the foot. All cases presented with

**Figure 3.** Swollen, infiltrative, nodular lesions with multiple crusted plaques over the dorsum of the left foot.



**Table 1.** Characteristics of mycetoma cases in Boru Meda general hospital, Ethiopia 2021.

Case	Age	Sex	Occupation	Duration (Years)	Location	History of trauma	Footwear	Swelling	Sinus	Grain	Previous treatment	Xray result
1	55	Male	Farmer	9	Foot	Yes	Unprotective	Yes	No	No	Yes	Normal
2	50	Male	Farmer	1	Foot	No	Unprotective	Yes	No	No	Yes	Not done
3	23	Male	Farmer	2	Foot	No	Protective	Yes	No	No	Yes	COM
4	21	Male	Farmer	5	Foot	No	Unprotective	Yes	Yes	Yes	No	COM
5	65	Female	Farmer	2	Foot	No	Unprotective	Yes	No	No	Yes	Normal
6	45	Female	Farmer	10	Foot	No	Protective	Yes	Yes	No	Yes	Normal
7	14	Male	Farmer	5	Foot	No	Protective	Yes	No	No	Yes	Not done

COM: Chronic osteomyelitis.

swelling, two had sinuses, and only a single case presented with grains, which were black in color. Six patients had previously received treatment from traditional healers and/or health institutions.

Only one case of mycetoma had a comorbidity (diabetes mellitus). Diagnostic tests like histopathologic examinations including potassium hydroxide (KOH) and gram stain and culture were not done for any patient. X-ray was performed for five patients and it revealed chronic osteomyelitis as a complication in two cases. All patients were prescribed a combination treatment of cotrimoxazole and ketoconazole.

## Discussion

This is the first clinical description of mycetoma cases from Ethiopia. Cases were characterized by swelling of the foot, although sinus formation and grains were uncommon. Most patients presented late with a lesion of at least one year duration, and six of seven patients had already looked for treatment elsewhere before coming to the hospital. Etiological identification was not possible, hence cases were diagnosed clinically and empirically treated with a combination of antifungal and antibacterial medication.

These cases of mycetoma are likely to present the tip of the iceberg as they were collected in a single-health center within a limited period of two years. Antidotal evidence suggest that dermatologists have encountered infections comparable to those described in this report from their respective centers throughout the country. Inclusion of mycetoma in the national health management information system would likely shed more light on the exact magnitude, trend and distribution of the diseases and associated causative agents in the country.

The clinical and socio-demographic characterization of these patients is similar to patients described in Sudan [3,6]. The fact that lesions are affecting the foot, and that most patients are male and farmers is most likely related to occupational exposure to the soil as a main source of infection. Five (71.43%) of the patients described here did not have sinus formation and visible grains. A large report on over 6000 mycetoma patients from Sudan [11] showed that 41.7% of mycetoma patients had grains, while 79% had sinuses, which suggests that the presentation in Ethiopia could be slightly different, though the caseload is not comparable.

Although its typical symptoms may allow for a clinical diagnosis of mycetoma, differentiating actinomycetoma from eumycetoma is difficult [12,13].

The available diagnostic tools include cytology, histopathological examination using potassium hydroxide (KOH) or periodic acid shift (PAS) stain, and culturing of grains to identify the causative agent [13]. Imaging can be helpful to determine the extent of the lesion, and local complications like osteomyelitis, which may lead to a change in antibiotic regimen to cover both mycetoma and bacterial bone infection. For the reported cases, it was not possible to identify the specific etiology because diagnostic modalities were not available. We recommend that diagnostic tools to differentiate between eumycetoma and actinomycetoma should be made available at referral centers.

Ideally, the management of mycetoma should be based on the causative organism. For eumycetoma, recommended treatment is itraconazole or ketoconazole for more than 12 months with or without surgery, although outcomes are poor [14,15]. Recommended treatment for actinomycetoma is a combination of sulfonamide with amikacin, dapsone, or streptomycin, and outcomes are generally better [15]. Most of these drugs are not routinely available at Boru Meda Hospital, and therefore patients received ketoconazole and cotrimoxazole to cover both fungal and bacterial pathogens. Such empirical combination treatment may unnecessarily expose the patients to extended treatments with significant side effects and risk of promoting antimicrobial resistance.

We did not collect data on follow-up visits, outcomes, and side effects as this data is often missed in routine practice. Although mycetoma can be diagnosed clinically and three dermatology professionals agreed on a mycetoma diagnosis for these patients, we unfortunately could not confirm the diagnosis with laboratory tests as they were not available. Since this study was embedded in routine care, we also cannot describe detailed information on risk factors and behaviors that could be related to the development of mycetoma and we also did not investigate the impact of mycetoma on patients. Future formal studies on diagnosis, treatment outcomes, risk factors, and qualitative studies on impact could provide more detailed local information on mycetoma in Ethiopia.

## Conclusions

Mycetoma is present in Ethiopia, although it is likely to be underreported. Comprehensive studies on mycetoma are needed in other regions of Ethiopia involving larger numbers of patients, and a specific focus on diagnosis, treatment, and psychosocial impact.



A national strategy for the disease should be designed aimed at addressing identified issues in reporting, diagnosis, and management in line with the WHO NTD road map 2021-2030 for “Ending the Neglect to Attain the Sustainable Development Goals” [7].

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