

Cystic mycetoma of hand- An unusual presentation of Mycotic Mycetoma

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Abstract

Mycetoma, an uncommon chronic infection of skin and subcutaneous tissues; commonly seen in tropical and subtropical countries.

It is caused by true mycetes named eumycetoma and filamentous bacteria belonging to group actinomycetes. The incidence is more common in agricultural workers walking in bare foot. Foot is most commonly affected. Other less commonly affected sites includes hand, knee, chest and head and neck region. Both forms of mycetoma present as a progressive, subcutaneous multiple nodular swellings discharging sinuses containing black colour granules.

The treatment of these two etiologies is entirely different, a definite diagnosis after histopathological examination is mandatory. We here by present a case of eumycetoma affecting elbow.

Keywords: Eumycetoma, Discharging Sinus, elbow.

Introduction

- Mycetoma is a chronic progressive infectious disease involving skin and subcutaneous tissue. Mycetoma has worldwide distribution and endemic in tropical and subtropical regions [1]. The organisms are usually present in the soil as saprophytes in different forms. They are implanted into the host tissue after traumatic inoculations of causative organism. (2)
- Mycetoma can be caused by true fungi or by filamentous bacteria and hence it is classified as eumycetoma and actinomycetoma respectively. (3,4)
- Mycetoma in general involves those parts of the body that come in contact with soil during daily activities, commonly seen in agricultural workers and in barefoot walkers in dry and dusty areas.
- The foot is the most affected site and this is seen in 70% of patients. The hand is the next commonest

site which occurs in 12% of patient. Less frequently knee, arm, head and neck, thigh, chest and the perineum are involved [5].

- It is a slow growing infection presenting with characteristic symptom of swelling with ulcer, draining sinuses and extrusion of characteristic coloured grains in the exudates [6].
- The grains discharged from the sinuses vary in size, colour and consistency.
- Dark grains are characteristic of eumycotic mycetoma. It may spread to involve the deep structures and bone resulting in destruction, deformity and loss of function.
- Since the treatment of these two etiologies is entirely different, a definite diagnosis after histopathological and microbiological examination is mandatory [7,8,9]. Here, we reported a case of cystic mycetoma of the elbow. The disease was identified only after surgical excision.

Case Report

- A 27/M presented in surgery OPD with a swelling over right elbow since 3 months.
- **On Local examination** : A swelling of 10cm x5cm was present over the dorsal aspect of the elbow. It was cystic, non-mobile and non tender.
- On clinical examination the diagnosis was Sebaceous cyst.
- FNAC of the lesion was advised.
- It was suggestive of cystic lesion with dense mixed inflammation.

Histopathological examination

- The lesion was excised and sent for histopathological examination.
- Revealed a thick cyst wall covered at places by neutrophils and foamy cells.

- The wall was composed of hyalinised fibrovascular connective tissue and showed dense lymphoplasmacytic inflammatory infiltrate and few congested capillaries.
- Pigmented granules were seen showing septate fungal hyphae and chlamydo spores.
- Fungal colonies were surrounded by dense neutrophilic infiltrates and few multinucleate giant cells.

Discussion

- Cystic change in mycetoma is rare. Infection usually presents as firm nodular swelling but it may be soft, lobulated and rarely cystic which grows in skin and subcutaneous tissue.
- Mycetoma is endemic in dry tropics and sub-tropic region.
- It generally affects agricultural workers and people who walk Barefooted in dry and dusty environment. [10,11]. Pathogens are found in the soil and are introduced through skin wounds during minor trauma.
- Infection presents as firm nodular swelling but it may be soft, lobulated and rarely cystic which grows in skin and subcutaneous tissue.
- The swelling can rupture forming discharging sinus tracts exuding characteristic coloured grains [6]. The granules vary in size, colour and consistency depending on the etiological species. These grains are the hallmark of mycetoma [10]
- The two main etiological groups of mycetoma - actinomycetic mycetoma and eumycetic mycetoma are caused by a number of species.
- Actinomycotic mycetoma is caused by aerobic species of actinomycetes belonging to the genera *Nocardia*, *Streptomyces* and *Actinomadura*.

Eumycotic mycetoma is caused by a group of fungi with thick, septate hyphae, including *Madurella mycetomi*, *Madurella griesia*, *Allescheria boydii* and *Acremonium* species [4,12].

- Dark (black) grains are found only among the eumycotic mycetoma. The pigment is a melanoprotein or related substance.
- In eumycotic mycetoma, there may be multiple punched out lytic lesions in bones. Actinomycotic mycetoma is characterized by both osteolytic and osteosclerotic lesions (6,11)
- Actinomycetic mycetomas expand faster, are more invasive and have more sinuses than eumycotic variants. Histopathological examination proves useful in differentiating actinomycetoma from eumycetoma.
- The granules of actinomycetoma consists of fine branching filaments, only about 1um thick are gram positive, whereas the grains of eumycetoma are gram negative [14]
- Eumycotic grains are composed of 4-5µm thick septate hyphae and are demonstrated by PAS (periodic acid-Schiff) and GMS (Gomori methenamine silver) stains [14].
- Confirmation of diagnosis and exact identification of the species requires culture. Malt extract, Sabouraud's and Glucose nutrient agars are the commonest types of media used in cultures of Mycetoma organisms. [8, 15)
- Thus histology along with special stain has a beneficial role and remains the only option in culture negative cases. [11].
- Differentiation between actinomycetoma and eumycetoma is important because of the different responses to treatment. Surgery is indicated in mycetoma for small localized lesions, resistance to

medical treatment or for better response to medical treatment in patients with massive disease.

- Surgical debridement, followed by appropriate combination of antibiotic therapy Amikacin Sulfate and Co-trimoxazole for several months is required for action mycetoma, where as Many other drugs such as Amoxicillin-Clavulanic Acid, Rifampicin, dapsone, Sulphonamides, Gentamicin, and Kanamycin were tried as a second line of treatment for action mycetoma in
- patients with resistant cases or who developed serious drug side effects [15].
- Eumycetomas are only partially responsive to anti-fungal therapy but can be managed by Surgery in combination with azole group drugs (Ketoconazole/ Itraconazole). The duration of nine to twelve months is the recommended treatment.

Conclusion

- In mycetoma, the foot being the most common site we report an unusual case of cystic mycetoma over elbow.
- Eumycotic mycetoma should always be considered in differential diagnosis of subcutaneous swellings with discharging sinuses containing black granules. The morbidity caused by mycetoma is massive and enormous resulting into deformities and septicemia. As treatment of the eumycotic mycetoma is primarily limited to surgical methods ,increased awareness and emphasis on early and correct diagnosis after clinical assessment and histological study with use of special stain is required.

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Legends Figure



Figure1: Gross picture showing cut surface of cystic mass with a cystic and a necrotic area. Wall is thickened.

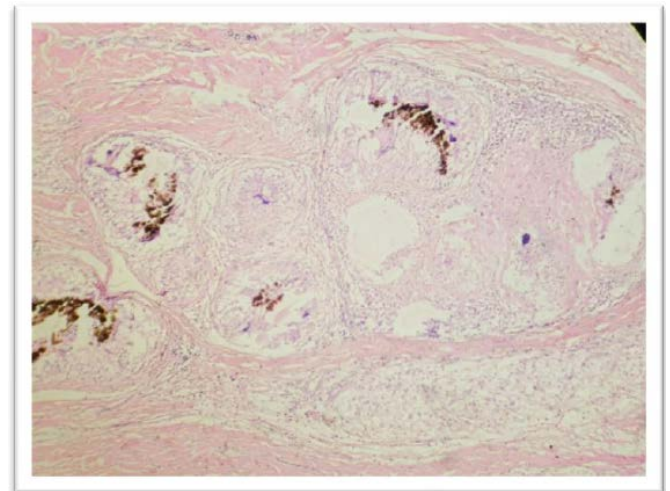


Figure 2: Photomicrograph showing (H&E X100) fibrocollagenous wall with fungal colonies.

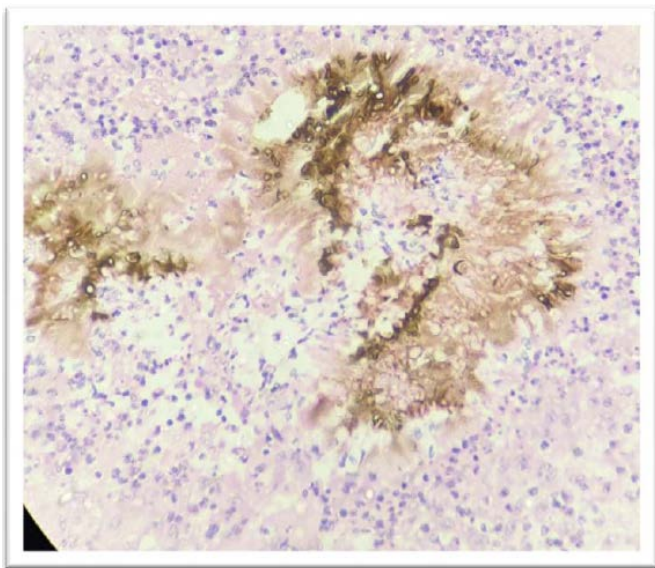


Figure 3: Photomicrograph showing (H&E X 400) fungal dematiaceous hyphae & chlamydospores with surrounding inflammatory infiltrate.

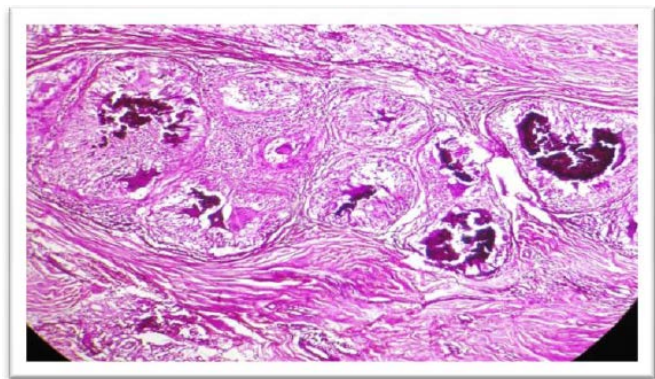


Figure 4: PAS stain (X 100) to highlight fungal filaments.