

Eumycotic Mycetoma Masquerading As Malignant Neoplasm: A Case Report

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ABSTRACT

Eumycetoma is a chronic and progressive subcutaneous infection caused by various genera of fungi. It is characterized by a triad of painless swelling and tumefaction, draining sinus tracts and purulent discharge consisting of granules. Approximately 40% of mycetomas worldwide are eumycotic (infection caused by fungi) in comparison to actinomycotic (caused by filamentous bacteria actinomycetes). Hallmark of the disease is progressive destruction of soft tissues and nearby anatomic structures. Eumycetoma are primarily saprophytic microorganisms that reside in soil or plant matter. Healthy individuals acquire the infection as a result of traumatic implantation of thorns, splinters or any plant matter. Here we discuss about 45 year old farmer presented with a knee swelling and diagnosed as Eumycotic mycetoma who was initially misdiagnosed to have a malignant neoplasm.

Key words: Eumycetoma, Malignant, Neoplasm.

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CASE REPORT

45 years old farmer presented with a diffuse soft swelling and pain around Rt knee joint since 2 months (Fig 1). Pain was gradual in onset, dull aching in character, progressive, with no diurnal variation & radiation. Swelling was initially small and gradually progressed to a globular size of 7x5cm on the medial side. There was no scar, sinus or venous engorgement around the swelling. On palpation there was tenderness and no local rise of temperature. Xray revealed eccentric lytic lesion on lower end of right femur involving both epiphysis and metaphysis, with expansion of bone, compressing trabeculae, with no involvement of articular cartilage (Fig 2).

Radiological and clinical diagnosis narrowed to differential diagnosis of GCT, osteosarcoma. Systemic examination was within normal limits. Laboratory investigations revealed a normal haemoglobi (15.1 g/dl), total WBC count, ESR and CRP. There was no regional lymphadenopathy. FNAC of the swelling revealed presence of lymphocytes, plasma cells, abundant histiocytes and few giant cells –reported as inflammatory lesion.

Past history of the patient revealed a trauma history on the same

site 5 months back while working on fields which subsided with treatment of antibiotics and dressing of the wound. There was no history of any granules or discharge from the wound.

The intra-operative picture mimicked a malignant mass with high vascularity firm to friable areas. The mass deeply infiltrated surrounding structures including extensor tendons. A subtotal excision was done preserving adjacent tendons and skin as far as possible with Plastic surgery assistance. A primary closure was achieved. The enbloc specimen was sent for histopathological study. Grossly it was skin lined partially globular soft tissue mass measuring 6x5 cm. C/S was soft to friable (Fig 3).

On microscopy it showed keratinized stratified squamous lining epithelium, dermis showing plenty of mixed inflammatory infiltrate with good number of histiocytes and giant cells. Focal areas showing collection of pigmented chlamydoconidophores arranged peripherally with central areas containing polymorphs Fig 4(a,b,c,d).

To our histological surprise the diagnosis of Eumycotic mycetoma was signed out following Gram's stain and PAS stain.



Fig 1: Showing a diffuse swelling around Right knee joint.

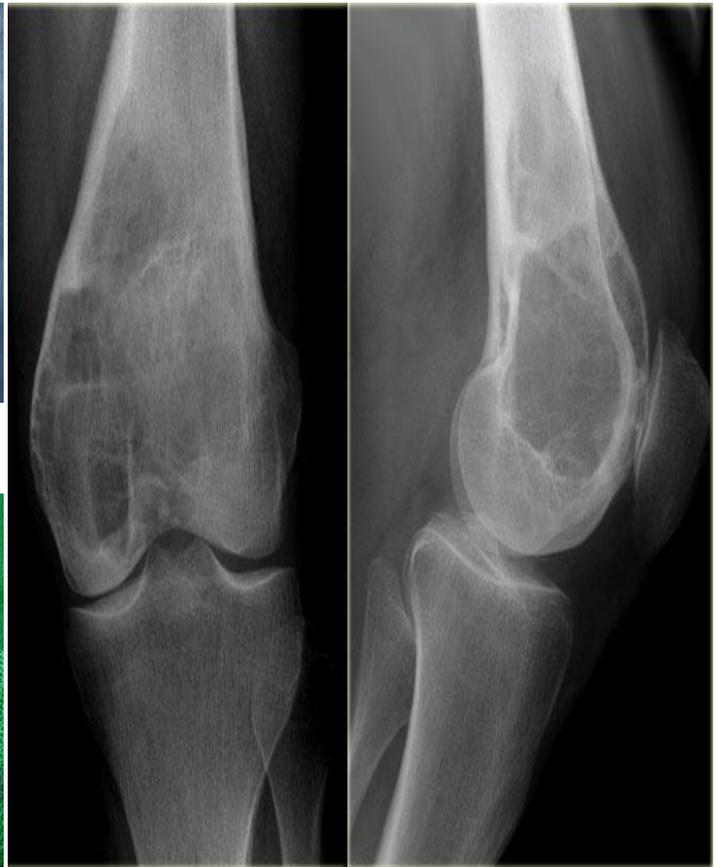


Fig 2: Eccentric lytic lesion involving lower end of femur.



Fig 3: Gross pic-skin lined friable, haemorrhagic soft tissue mass mimicking a malignant neoplasm.

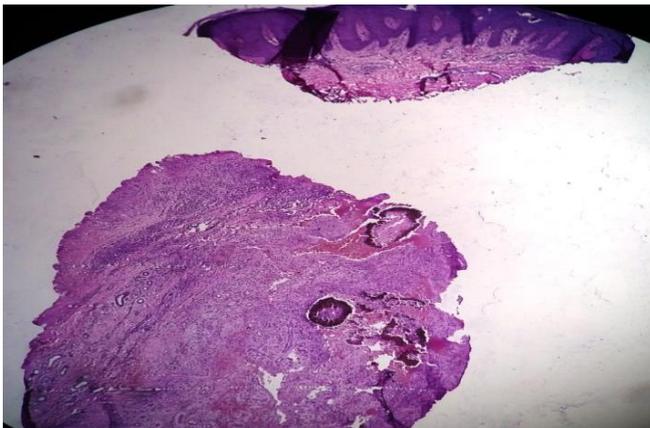


Fig 4(a): Scanner view 40x-Showing skin lining with pigmented lesions in the dermis.

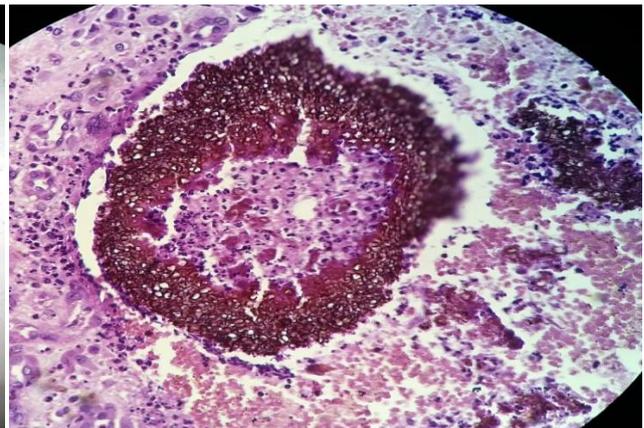


Fig 4(b): HP- 400x-Showing fungal colonies of Eumycotic mycetoma.

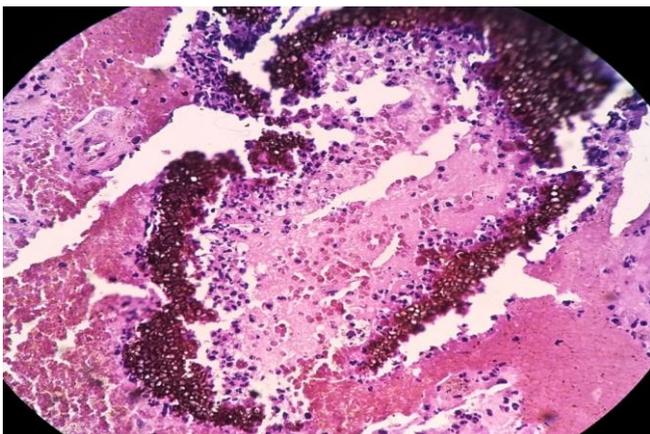


Fig 4(c): HP 400x-Showing fungal colonies of Eumycotic mycetoma.

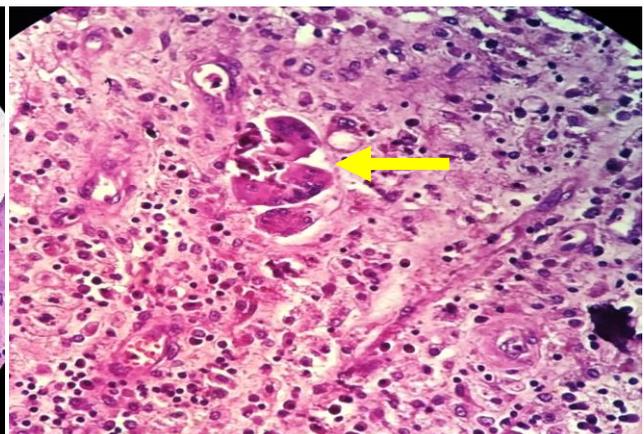


Fig 4(d): LP 100X-Showing presence of giant cells (arrow)

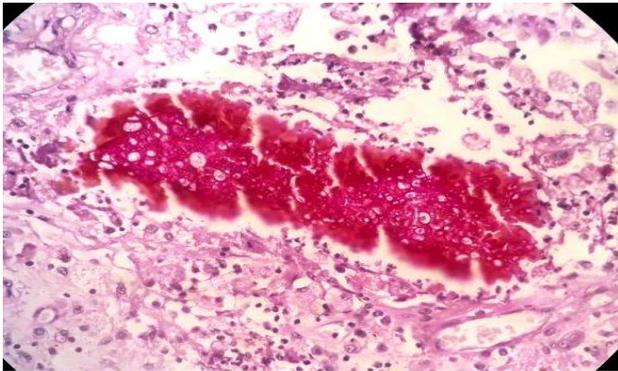


Fig 5: Showing fungal granules bordered by eosinophilic Splendore-Hoeppli material composed of vesicular chlamydoconidia. [PAS Stain]

DISCUSSION

Mycetoma was first described in the mid-19th century and initially named "Madura foot", after Madurai in Tamil Nadu, India, where the disease was first identified.¹ Extra pedal involvement also occurs and has been detected in hand, leg, head and neck, abdominal wall, buttock and perineum. It is initially limited to skin and subcutaneous tissue and may eventually spread to fascial planes and to contiguous structures such as muscle, bone, lymphatics and nerves. It is mainly a disease of the tropical and subtropical zones, especially between the Tropic of Cancer and the Tropic of Capricorn, which is between latitudes 15° S and 30° N especially in arid zones interspersed with short bouts of heavy rainfall and higher relative humidity. The disease is usually seen in field workers like farmers affects men between 20 to 40 years. Initial lesion is a small subcutaneous swelling following minor trauma, later sinuses are formed that discharge purulent and seropurulent exudates containing grains which are the fungal colonies and are infective.² Spread of infection via lymph or blood is unlikely. Mycetoma may be caused by bacteria from the phylum Actinomycetes or by fungi (Eumycetes) where it is called Eumycetoma. Bacterial and fungal species that cause mycetoma are classified on basis of characteristic colours of discharge from the infected wounds. Red discharge (*Actinomadura pelletieri*), white or yellow discharge (*Actinomadura madurae*, *Pseudallescheria boydii*, *Acremonium strictum*, *Aspergillus nidulans*) and black discharge (*Madurella grisea*, *Madurella mycetomatis*, *curvularia* species, *Cladophilaphora bantiana*). Infections that produce a black discharge mainly spread subcutaneously. In the red and yellow varieties deep spread occurs early, infiltrating muscles and bones but sparing nerves and tendons. It is important to differentiate between an actinomycetoma, caused by aerobic bacteria, and a eumycetoma, caused by true fungi, because the treatment is markedly different.³ Identifying the exact type of fungi by fungal culture or other advanced techniques will aid in selecting the appropriate treatment protocol.^{4,5} In developing countries like ours, facilities for fungal cultures and other advanced investigations are not easily available.^{5,6} More over culture requires prolonged incubation in specialised media for close to a month which is cumbersome and even then one may fail to identify the organism. So it is unwise to withhold patient's treatment this long, as recurrence rates are significantly high even with prolonged treatment.⁷ Histopathology being easy available modality plays a vital role in planning treatment strategy of these patients.

Although clinically and radiologically it mimicked a malignant lesion the past history of trauma, a farmer by occupation along with characteristic histopathological finding helped in arriving at a correct diagnosis. It was supplemented by grams stain which showed gram negative compact mycelial aggregates of Eumycotic mycetoma and PAS stain showing granules bordered by eosinophilic Splendore-Hoeppli material composed of broad septate hyphae and vesicular chlamydoconidia. (Fig 5)

Ketoconazole or Itraconazole combined with aggressive surgical debridement is recommended for patients with eumycetoma. As recurrences of eumycetoma are more likely than those in actinomycosis, prolonged treatment up to 36 months with Ketoconazole is often advocated to prevent recurrences.⁸

CONCLUSION

Eumycetoma often mimicks a malignant soft tissue mass and can be associated with significant morbidity in terms of gradual enlargement and deformity of the infected site. Severe involvement of lower extremity may impair ambulation. So early diagnosis is essential to prevent the adverse complications. In conclusion, it is important to maintain a high index of suspicion for an infectious agent causing a focal mass.

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