

Case Report

A case report of eumycetoma in the thigh: an unusual presentation as a lump

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ABSTRACT

Mycetoma is a chronic disease, which is endemic in tropical and subtropical countries. Mycetoma is a chronic subcutaneous infection caused by Actinomycetes or fungi. This infection results in a granulomatous inflammatory response in the deep dermis and subcutaneous tissue, which can extend to the underlying bone. Mycetoma is characterized by the formation of grains containing aggregates of the causative organisms that may be discharged onto the skin surface through multiple sinuses. Here we report an unusual presentation of eumycetoma in the thigh of a 41 year old male rural industrial worker hailing from South India.

Keywords: Mycetoma, Eumcetoma, Actinomycetoma, Madura foot, Tropical

INTRODUCTION

In tropical countries, mycetoma is a real public health issue. Mycetoma is a chronic granulomatous inflammatory response involving bacteria or fungi that triggers the formation of grains containing aggregates of the causative organisms that may be discharged onto the skin surface through multiple sinuses. It is an infection of the deep subcutaneous soft tissues and bones that can develop over several years. The prevalence of this disease is 1.81 cases per 1,00,000 inhabitants in Sudan, whereas it is less than 0.01 cases per 1,00,000 inhabitants in India. The incubation period for this disease is highly variable and ranges from 3 months to 10 years.¹ It commonly affects young adults aged 20 to 40 years, predominantly males, living in rural areas. It is more frequently reported in farmers, shepherds and workers of low socio-economic status. The body parts commonly affected by mycetoma are the feet or lower legs with infection of the dorsal aspect of the forefoot being typical.² Untreated, the disease progresses to destruction

of soft tissue and adjacent bone structures with deformation of the limb.³

CASE REPORT

A 41 year old male rural industrial worker with no comorbidities reported to the OPD with a painless right thigh swelling of 6 months duration which was gradually increasing in size. Owing to continuous increase in size he had come for further management. No history of any recent or past traumas, prick injuries or fever in the last 6 months. Local examination of the region revealed a 4×3cm firm swelling in the lateral aspect of left thigh, mobile and not fixed to underlying structures. Skin over the swelling was normal and pinchable. Ultrasonogram of the local region showed a cystic lesion of size 4.2×2.8 cm with mixed echogenicity suggestive of infected cystic lesion. Exploration was done under local anesthesia and a subcutaneous thick walled cyst was excised in toto. No deep extension was noted and skin closed with sutures. To our surprise, on exploring the specimen a thick walled

fluid filled cyst with multiple blackish granules within it was seen as shown in Figure 1.

Patient was treated with broad spectrum oral antibiotics and wound healed without any complications. HPE of the specimen showed abscess cavity lined by sheets of chronic inflammatory cells, foamy macrophages and foreign body type giant cells enclosing brownish fungal elements composed of hyphae consistent with eumycetoma. MRI was done subsequently which showed no deep muscle or bony involvement. Patient was started on anti-fungal therapy following HPE report -Tab. Itraconazole 200 mg once daily for 6 months. Patient improved well with no signs of recurrence on a follow up period of 2 years as shown in Figure 2.



Figure 1: Multiple black granules with turbid fluid was seen on opening the specimen after excision.



Figure 2: Excision site scar at 1st year follow up.

DISCUSSION

The triad of a painless subcutaneous mass, multiple sinuses and purulent or seropurulent discharge that contains grains is pathognomonic of mycetoma.⁴ Usually the infection is contracted through a puncture wound in

the feet, especially in persons walking barefoot (Madura foot account for 70% of all mycetoma cases). Involvement of the foot is described in almost 80% of cases of mycetoma other sites being the thigh and knee (3%), trunk (4%), upper limb (9%) and head and neck (4%).⁵ This chronic subcutaneous infection can be caused by bacteria or fungi. Mycetoma caused by microaerophilic *Actinomycetes* is termed actinomycetoma, and mycetoma caused by true fungi is called eumycetoma. The next step is to differentiate them through histological skin biopsies to identify the causal agents, if the grains are black or white it is a fungal infection- eumycetoma and if the grains are white, yellow or red it is a bacterial infection-actinomycetoma.

Once the mycetoma is diagnosed, it is essential to look for bone lesions by additional imaging. Radiography shows infiltration of soft tissue, associated more or less with bone resorption. On X-rays images, bone destruction is visible through multiple and non-specific osteolytic lesions (geodes, cavities, reactive sclerosis). In 2003, Bagi devised a 7-stage classification based on the extent of bone damage on the radiographs, ranging from stage 0 (no bone damage) to stage VI (multi-directional bone damage). On ultrasound imaging single or multiple thick-walled cavities with hyper-reflective echoes and no acoustic enhancement are always observed with mycetoma often aggregated at the lower part of the swollen mass. MRI is the most helpful investigation for a positive diagnosis and staging the disease. The characteristic appearance is that of an infiltrating mass made up of small cavities, hyperintense on T2 weighting, and circumscribed by hypointense fine partitions containing central dots, hypointense on all sequences and creating a nearly pathognomonic sign, called the “dot in-circle”, especially useful when clinical, microbiological and histological findings are not determinative. This dot in-circle sign is correlated with the histology: the primary hypointense point corresponds to the mycelial granule, the surrounding hyperintense signal to the inflammatory granuloma, and the hypointense partitions to the fibrous matrix.⁶ Few radiographic bone changes have been described to distinguish between actinomycetoma and eumycetoma. Eumycotic lesions tend to form a few cavities in bone ≥ 1 cm in diameter, while actinomycetes often form smaller, but more numerous cavities. In a study by Lewall et al, a moth-eaten appearance caused by a combination of irregular periosteal reaction, periosteal erosion, and small cavities within bone were seen in 25% of cases of actinomycetoma, but in none of the patients with eumycetoma. The distinction between the 2 forms of soft tissue mycetoma was not possible with MRI.⁷

In the treatment of mycetoma medicines and surgery should be combined. The medical treatment consists of antibiotic therapy (cotrimoxazole, amikacin or minocycline) or antifungal therapy (ketoconazole or itraconazole) and should be combined with surgery that should be as thorough as possible. In spite of these treatments, disease recurrence is quite common.

Amputation may be required in selective cases of extensive bone involvement.

CONCLUSION

Mycetoma is an infection of tropical and subtropical regions that classically involves the dorsum of foot with multiple discharging sinuses with or without an underlying soft tissue mass. Our case report is a rare presentation of a mycetoma as an uncomplicated lump and a good reminder that this disease can also affect areas other than the foot. Imaging, in particular MRI is the most reliable investigation in a difficult clinical presentation/suspicious cases for a positive diagnosis and also aids in staging the disease to plan appropriate management.

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REFERENCES

1. Neyaz Z, Mohindra N, Bhatnagar A, Marak RSK. Mycetoma CI. Case 249: Intramuscular Mycetoma 1. 2018;286(1):353-9.
2. Asly M, Rafaoui A, Bouyermane H, Hakam K, Moustamsik B, Lmidmani F, et al. Le pied de Madura. À propos d'un cas. Ann Phys Rehabil Med. 2010;53(10):650-4.
3. In vivo measurement of compression bandage interface pressures: Evaluation of different bandages, application methods and positions The effects of different sudden ankle inversion degrees on ankle brace efficacy. 2018;(1):20100.
4. Fahal A, Mahgoub ES, Hassan AMEL, Abdel-Rahman ME. Mycetoma in the Sudan: An update from the Mycetoma Research Centre, University of Khartoum, Sudan. PLoS Negl Trop Dis. 2015;9(3):1-19.
5. Roberts IF, Karim QN, Rosin RD. Case reports actinomycotic mycetoma of the thigh. J R Soc Med. 1989;82(9):552-3.
6. Bouziane M, Amriss O, Kadiri R, Adil A. The role of computed tomography in the exploration of Madura foot (pedal mycetoma). Diagn Interv Imaging. 2012;93(11):884-6.
7. Kumar J, Kumar A, Sethy P, Gupta S. The dot-in-circle sign of mycetoma on MRI. Diagnostic Interv Radiol. 2007;13(4):193-5.

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