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Case Report

The missing grains in Madura foot: imaging to the rescue

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ABSTRACT

Mycetoma or Madura foot is a chronic localized granulomatous infection caused by varied species of fungi or actinomycetes clinically diagnosed by active discharging sinuses containing 'grains'. In atypical presentations or patients unwilling for invasive investigations, imaging essentially plays a key role in diagnosis and helps to differentiate actinomycetoma from eumycetoma. Here we present such an atypical case of unilateral foot swelling without sinuses–cryptic mycetoma. This 69 year old male from Kerala had a swelling over the left foot following trauma initially painless. An x-ray taken showed normal underlying bones with mild soft tissue swelling. To evaluate further USG was taken which showed fine hyperechoiec foci settled at the bottom of cavities highly suggestive of mycetoma. For confirmation USG guided biopsy was taken but was inconclusive. As patient was not willing for repeat biopsy, we advised non invasive MRI that revealed classical 'Dot In Circle' sign specific for mycetoma. The distinction that makes the case stand out is its absence of classical picture of discharging sinuses and the fact that imaging rather than histopathology helped confirm the diagnosis in mycetoma.

Keywords: Mycetoma, Atypical, Dot In Circle, Imaging

INTRODUCTION

Mycetoma is characterized by the presence of the classical triad of swelling, sinuses and granules. In atypical presentation there can be absence of sinuses – cryptic mycetoma.[1] It is a chronic granulomatous infection of dermal and subcutaneous tissues that may extend to underlying muscles and even bones caused by fungi or actinomycetes.[2] The disease is characterized by aggregates of causative organisms known as 'grains' within abscesses that slowly form sinuses discharging them.[3] Diagnosis is primarily by the presence and histopathological or microbiological assessment of grains.[4]

Here we present a case report of mycetoma with atypical presentation that was diagnosed with the help of imaging by the presence of classical 'Dot In Circle' sign specific for Mycetoma.[5]

CASE REPORT

A 69-year-old male agricultural worker from Kerala presented with swelling over left foot (Figure 1) following trauma that occurred 20 years back. A year back, when admitted for a hernioplasty the surgeon noticed the swelling and biopsy done was inconclusive. As it was asymptomatic patient opted for no further management. Subsequently from prior to 2 months he has difficulty in walking due to pain in the swelling and adjacent area in the left foot. X-ray (Figure 2) of the left foot was done which showed normal underlying bones with mild soft tissue swelling. To evaluate soft tissue swelling USG (Figure 3) was taken wherein cavities were noted with fine hyperechoiec foci settled at the bottom that was highly suggestive of mycetoma and for confirmation biopsy was taken and histopathologically (Figure 4) assessed that was reported as inflammatory infiltrates in dermis. Patient was advised for deep biopsy

under regional anaesthesia but refused. Following which we suggested a noninvasive imaging to evaluate the swelling. MRI (Figure 5) was done that revealed classical 'Dot In Circle' sign diagnostic of mycetoma. Patient was treated by modified Welsh regimen.[6]

Figure 1: A large swelling of dorsum of left foot with multiple nodules.

Figure 2: Normal underlying bones with mild soft tissue swelling.

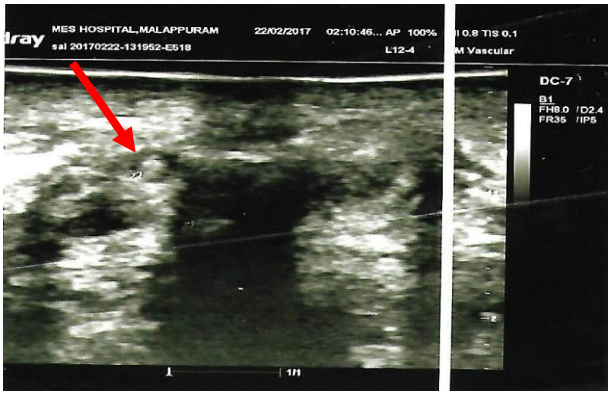

Figure 3: Cavities with fine hyperechoiec foci settled at the bottom.

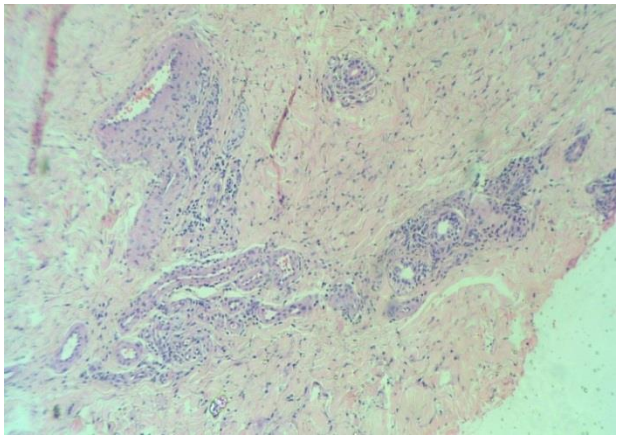

Figure 4: HP slide showing inflammatory infiltrates in dermis.

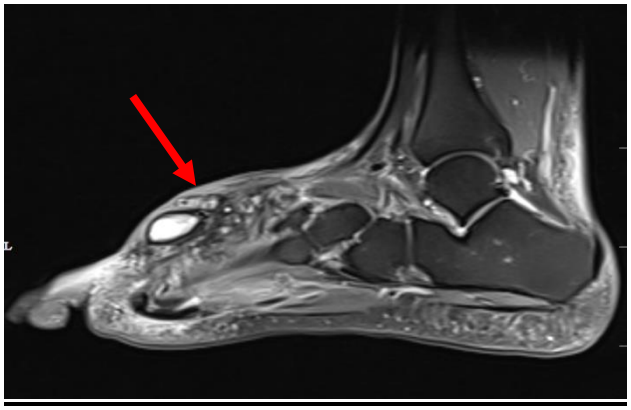

Figure 5: Classical 'Dot In Circle' sign diagnostic of mycetoma.

Table 1: Causal agents of actinomycotic mycetoma and colour of their grains.

Agent	Colour of grains
Nocardia asteroids	White
Nocardia brasiliensis	White
Nocardia otitidiscaviarum	White
Actinomaduramadurae	White
Actinomadurapelletieri	Red/Pink
Streptomyces somaliensis	White – Yellow

Table 2: Causal agents of eumycotic mycetoma and colour of their grains.

Agent	Colour of grains
Madurellamycetomatis	Black – Brown
Madurellagrisea	Black – Brown
Leptosphaeria senegalensis	Black
Pseudallescheriaromeroi	Black
Exophialajeanselmei	Brown – Black
Pseudallescheria boydii	White – pale Yellow
Acremonium spp.	White – pale Yellow
Neotestudinarosatii	Yellow

Table 3: Modified Welsh Regimen.[28]

Intensive phase Amikacin 15 mg/kg/d iv every 12 h for 21 days [1 cycle]; 3-4 cycles with an interval of 15 d	Maintenance phase Cotrimoxazole 35 mg/kg/d and Rifampicin 10 mg/kg/d continuously [from the first day of therapy] for 3 months after the last cycle of Amikacin

DISCUSSION

Madura mycosis or mycetoma is a chronic infection caused by *Actinomycetes* or true fungi endemic to tropical areas, seen often in extreme climatic conditions and what is referred to as the "mycetoma belt".[7,8] This infection results in a granulomatous inflammatory response in the deep dermis and subcutaneous tissue with fibrosis, which may extend to the underlying bone. The eponym Madura foot was attached owing to the first case reported by Gill in Madura, a place in Tamil Nadu in India, in 1842.[9] Mycetoma is broadly categorized into two subtypes – eumycetoma and actinomycetoma, based on the causative organisms.[10] Eumycetoma is more common in Northern India where rainfall is scarce and actinomycetoma is implicated as common cause in Southern India that receives heavy rainfall and correlates with our case.[11]

Commonly affected sites are the feet more than lower legs or hands, with infection of the dorsal aspect of the forefoot being typical.[12] A male preponderance within the ages of 30 and 40 are seen infected.[13] Usually the infection is preceded by a trauma in the feet, especially in people walking barefoot, those engaged in farming or maybe nomads.[14]

Lesion initiates as small painless subcutaneous nodule that becomes multiple, ulcerate and drain via sinus tracts. The disease is characterized by organisms aggregating in abscesses forming grains or granules. Discharge from these tracts often contains the characteristic granules. Skin biopsies and microbiological culture identify the causal agents. White, yellow or red grains are found in bacterial (actinomycetoma) (Table 1) and Black or white grains in fungal (eumycetoma) (Table 2) infections.[15] Bacterial infections are vastly progressive and invade at a faster rate as compared to the slowly advancing Eumycetoma.

Two among the classical triad of localized swelling, sinus tracts and grains were absent in this patient.[16] Generally sinuses discharging grains are formed within a year of infection.[17]

Laboratory diagnosis, before the appearance of the sinuses and grains, is difficult as observed in this case.[18] Time-consuming procedures like biopsy, staining and microbiological culture of the discharge can derive definitive diagnosis but still may not be possible with fastidious organisms.[19]

Mycetoma can progress slowly becoming painful with involvement of bone and also lead to secondary bacterial infection.[20]

Early diagnosis is essential considering the therapeutic implications as delay may lead to serious deformity and restriction to mobility by extensive destruction of bone underneath.[21] Once diagnosed, the presence and extent of bone and soft tissue lesions are investigated by imaging.[22] X-rays can portrait soft tissue swellings or multiple osteolytic lesions (reactive sclerosis, invasion).[23] Imaging techniques assuredly helps to evaluate the extension of destruction. Ultrasonography helps to differentiate eumycetoma from actinomycotic mycetoma-eumycetoma grains produce sharp hyperechoiec foci, actinomycetoma produce fine hyperechoiec foci that commonly settle at the bottom of the rounded lesions.[24] MRI shows 'Dot In Circle' sign specific for the disease.[25] It has been demonstrated that MRI supersedes other imaging techniques to diagnose mycetoma accurately.[26]

In our case X-ray foot showed only soft tissue swelling and biopsy revealed no relevant findings. USG findings were suggestive of actinomycotic mycetoma. MRI confirmed the diagnosis by the presence of classical "Dot In Circle" sign. The patient was successfully treated by the modified Welsh regimen.[27]

CONCLUSION

Mycetoma is still considered endemic in low socioeconomic background and therefore it is vital to insist upon hygiene and use of protective footwear to prevent such diseases. Imaging can aid in timely diagnosis when the expected clinical picture is absent and the usual investigations do not contribute. Imaging techniques like magnetic resonance imaging (MRI) are less time consuming and noninvasive revealing diagnosis particularly in atypical presentations for suitable interventions to avoid complications that can lead to functional and aesthetical impairments. This case is a good reminder of the importance of imaging in dermatology especially in atypical clinical presentations.

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REFERENCES

1. Bravo FG, Arenas R, Sigall DA. Actinomycosis Nocardiosis and Actinomycetoma. In: Wolff K, editor. Fitzpatrick's dermatology in general medicine, 7th ed. New York: The McGraw-Hill Companies; 2008: 1784-1786.
2. Relhan V, Mahajan K, Agrawal P, Garg VK. Mycetoma: An Update. Indian J Dermatol. 2017;62:332-40.

3. Ahmed AA, Sande WVD, Fahal AH. Mycetoma Laboratory Diagnosis: Review Article. PlosNegl Trop Dis. 2017;11(8):E0005638.
4. Kumar MMS, Kumar VS, Kumar P, Anandan V. Mycetoma- A resurgence. IP Indian J Clin Exp Dermatol. 2018;4:44-7.
5. Laohawiriyakamol T, Tanutit P, Kanjanapradit K, Hongsakul K, Ehara S. The "dot-in-circle" sign in musculoskeletal mycetoma on magnetic resonance imaging and ultrasonography. Springer Plus. 2014;3:671.
6. Welsh O, Al-Abdely HM, Salinas-Carmona MC, Fahal AH. Mycetoma Medical Therapy. PLoS Negl Trop Dis. 2014;8(10):e3218.
7. Reis CMS, Reis-Filho EGM. Mycetomas: an epidemiological, etiological, clinical, laboratory and therapeutic review. An Bras Dermatol. 2018;93:8-18.
8. Hay RJ, Mahgoub ES, Leon G, Al-Sogair S, Welsh O. Mycetoma. J Med Vet Mycol. 1992;30:41-9.
9. McGinnis, Michael R. Mycetoma. Dermatologic Clin. 1996;14:97-104
10. Van de Sande WWJ. Global Burden of Human Mycetoma: A Systematic Review and Meta-analysis. PLoS Negl Trop Dis. 2013;7(11):1-11.
11. Gooptu S, Ali I, Singh G, Mishra RN. Mycetoma foot. J Family Community Med. 2013;20:136-8.
12. Lynch JB. Mycetoma in the Sudan: Hunterian Lecture delivered at the Royal College of Surgeons of England on 12th March 1964. Ann R Coll Surg Eng. 1964;35:319-40.
13. Fahal AH, Hassan MA. Mycetoma. Br J Surg. 1992;79:1138-41.
14. Tilak R, Singh S, Garg A, Bassi J, Tilak V, Gulati A. A case of Actinomycotic mycetoma involving the right foot. J Infect Dev Ctries. 2009;3:71-3.
15. Iffat H, Abid K. Mycetoma Revisited. N Dermatol Online. 2011;2:147-50.
16. Palestine RF, Rogers RS. Diagnosis and treatment of mycetoma. J Am Acad Dermatol. 1982;6:107-11.
17. Saha S, Dhar A, Karak AK. Mycetoma Sans Sinuses. Indian J Dermatol Venereol Leprol. 2006;72:143-4.
18. Jain V, Makwana GE, Bahri N, Mathur MK. The "Dot in Circle" Sign on MRI in Maduramycosis: A Characteristic Finding. J Clin Imaging Sci. 2012;2:66.
19. Sen A, Pillay RS. Case Report: Dot-In-Circle Sign – An MRI And USG Sign For "Madura Foot". Indian J Radiol Imaging. 2011;21:264-6.
20. Abdalla OA, Ahmed. Unexpected High Prevalence of Secondary Bacterial Infection in Patients with Mycetoma. J Clin Microbiol. 1998;36:850–1.
21. Fahal A, Mahgoub ES, Hassan AME, 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):e0003679.
22. Develoux M, Dieng MT, Kane A, .Management of mycetoma in West-Africa. Bull Soc Path Exo. 2003;96(5):376-82.
23. Abd El-Bagi ME1, Fahal AH. Mycetoma revisited. Incidence of various radiographic signs. Saudi Med J. 2009;30(4):529-33.
24. Fahal AH, Sheik HE, Homeida MM, Arabi YE, Mahgoub ES. Ultrasonographic Imaging Of Mycetoma. Br J Surg. 1997;84:1120-2.
25. Cherian RS, Betty M, Manipadam MT, Cherian VM, Poonnoose PM, Oommen AT, Cherian RA. The "Dot-In-Circle" Sign- A Characteristic MRI Finding In Mycetoma Foot: A Report Of Three Cases. Br J Radiol. 2009;82:662-5.
26. Czechowski J. MR And Other Imaging Methods In The Investigation Of Mycetomas. Acta Radiologica. 2014;42:24-6.
27. Damle DK, Mahajan PM, Pradhan SN, Belgaumkar VA, Gosavi AP, Tolat SN, Gokhale NR, Mhaske CB. Modified Welsh regimen: a promising therapy for actinomycetoma. J Drugs Dermatol. 2008;7(9):853-6.
28. Palit, Aparna, Ragunatha S, Inamadar AC. Actinomycetoma: dramatic response to modified two-step regimen. Int J Dermatol. 2011;50:446-9.

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