CASE REPORT

THE CASE SERIES OF MUSCULOKELETAL MADURA ON MAGNETIC RESONANCE IMAGING

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PJR July - September 2019; 29(3): 207-210

ABSTRACT

Mycetoma is a rare chronic cutaneous and subcutaneous infection caused by various genera of fungi. Approximately 40% of mycetomas worldwide are eumycotic as opposed to actinomycotic (i.e, caused by bacterial actinomycetes). The disease is marked by progressive destruction of soft tissue and nearby anatomic structures. In this paper, the characteristic features of mycetoma of the foot on magnetic resonance imaging (MRI) in three of Dow University Hospital patients are presented.

MeSH terms: Madura foot, Mycetoma, MRI, Dot-in-circle sign, Musculoskeletal system.

Introduction

Mycetoma is a debilitating chronic granulomatous infection that affects the subcutaneous tissues and may extend to involve deeper structures. It is an extremely rare condition in temperate zones, though prevalent in the tropical and subtropical regions. The infection painlessly burrows deeply until it reaches the bone.[1,2]

Mycetoma commonly affects adults in the age group of 20-40 years, with men being more commonly affected than women with a ratio 3.5:1.3. The disease is caused due to direct transcutaneous implantation of the causative organism Actinomycetes (bacteria) or Eumyces (fungus) which are normal soil inhabitants, secondary to a penetrating wound like a thorn prick. Histologically, mycetoma is characterized by aggregates of the organism, known as grains within micro abscesses surrounded by abundant granulation tissue.[3]

Clinical Presentation

Case 1

21 yrs. old female presented with status post excision of the right calcaneum lesion in last year. There is history of swelling of the right foot. No discharge. It's a case of biopsy proven mycetoma.

Complementary radiograph shows there is marked circumferential resorption of the shaft of the right 3rd metatarsal and expansile osteolytic lesion is seen in the posterior aspect of the calcaneum with reactive sclerosis (Fig. 1). On MR, it shows multiple hypointense lesions surrounded by diffuse edema with subtle enhancement in the calcaneum- which appears to be fungal balls. There is multilocular focal osteolytic lesion seen in the distal shaft of the right 4th metatarsal with reactive sclerosis. There is secondary pathology fracture with heterogeneous enhancement. There are multiple intercommunicating peripherally enhancing collections are seen in the flexor compartment of the foot involving the abductor hallucis, quadratus plantae, flexor digitorum brevis, interossei muscles, encasing the tendons of the flexor digitorum longus tendons. One of them measures 5.2 x 2.7 cm. It is also involving the subcutaneous fat and skin measuring 2.1 x 2.3 cm and also extending into the extensor compartment to the secondary intermetatarsal space. The collection shows hyperintense content on T1WI, T2WI and STIR sequence and

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Submitted 9 August 2019, Accepted 6 September 2019

showing small rounded areas with faint iso to hyper-intense signal with hypointense rim & central tiny dot like hypointense focus suggesting dot-in-circle sign.

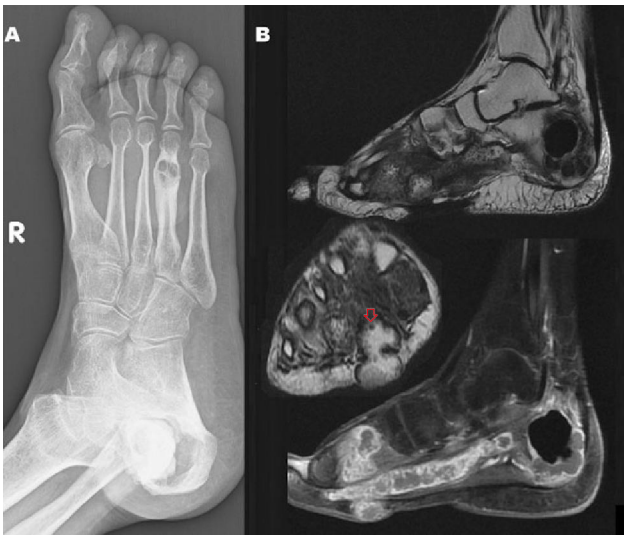

Figure 1: Radiograph **(A)** and multiplanar plain and contrast MR of foot **(B)** demonstrating the osseous and soft tissue lesions of Mycetoma. Subcutaneous and deep multiple granulomas, abscesses with fungal granules in the dorsal planar and inter meta-tarsal web spaces.
(Red arrow - Dot in Circle Sign in each figure)

There is a peripherally enhancing tract is seen extending upto the subcutaneous fat and the skin along the lateral surface of side of the plantar surface of the skin measuring upto 1.8 cm. It represents chronic suppurative infection of the right foot with sinus; multifocal mycetoma of the tarsus and metatarsus with soft tissue abscesses.

Case 2

45 yrs. old male, known diabetic and chronic liver disease presented with foot swelling. The right foot abscess was suspected. On MR, there are multiple lobulated heterogenous abnormal signal intensity areas of variable sizes are seen along the plantar and the dorsal musculature of the foot. These lobulated areas with irregular enhancing thickening in the soft tissues of the foot encasing the flexor and extensor tendons, shows centre in dot sign, however, no intra tendinous abnormal signals. Another focal enhancing area of soft tissue thickening noted in the intertarsal space of the 1st and 2nd toe is measuring 1.8 x 3.2 cm. The planter aponeurosis shows reactive thickening with subcutaneous fat stranding. No abscess cavity /collection and no bone involvement noted. The MR appearances suggest multifocal atypical infection with finger in dot sign (Fig. 2). The Biopsy of tissue reveals fungal hyphae in tissue.

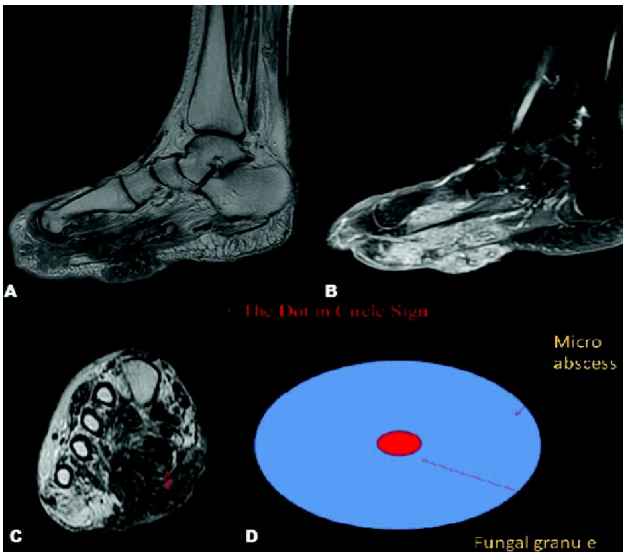

Figure 2: Gallery of Rad **(A-C)** and Path **(D)** correlation.

Case 3

55 yrs old female presented to the hospital with repeated episodes of foot swelling and multiple discharging sinuses on and off for the last 30 yrs. Initially, she has history of trauma. The sinus discharge is of yellow and black color. She underwent surgical debridement 5-6 times after which the swelling subsides and develops again after an interval.
Radiograph shows periosteal reaction along the lateral aspect of 3rd metatarsal. There is remodelling abnormality of 2nd metatarsal bone. (Fig. 3)

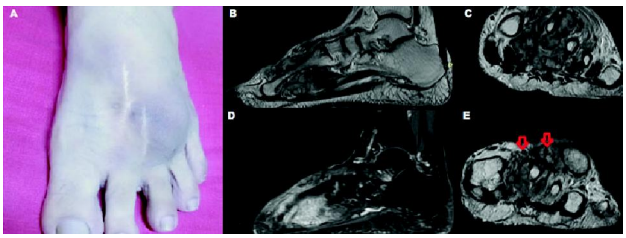

Figure 3: Photomontage of clinico-radiologic spectrum. Picture A shows grayish tan swelling on dorsum of foot and **(B- E)** MR of foot shows multi focal soft tissue and bone involvement.

On MR, cortical erosions are seen in talus along posterior subtalar joint with enhancement along the articular surface of calcaneum. Significant skin and subcutaneous soft tissue swelling and edema is identified involving dorsal and plantar aspects of left mid foot represents cellulitis.

Muscles at flexor and extensor compartment of foot including interossei, flexor digitorum longus and brevis, adductor hallucis, flexor hallucis brevis and extensor digitorum muscles, inter tarsal ligaments appear diffusely thickened and show post contrast enhancement. There are multiple well defined collections, encasing flexor and extensor tendons, second, third and fourth metatarsals, extending into sub cutaneous space of extensor compartment, largest measure 2 x 1cm in axial dimension.

These shows hyperintense content on T1WI, T2WI and STIR sequence shows significant post contrast enhancement, and contains multiple small rounded areas with faint iso to hyperintense signal with hypointense rim & central tiny dot like hypointense focus suggesting dot-in-circle sign. This spectrum represents musculoskeletal maduromycosis with cellulitis and focal osseous involvement along subtalar joint.

Discussion

Mycetoma is a unique neglected disease, characterized by devastating deformities, disability, high morbidity and serious negative socioeconomic impacts on patients, families and community. Many cases are painless, although painful lesions may prompt the individual to seek medical attention.[4] Predisposing factors include the following:

History of trauma
Walking barefoot
Agricultural work
Poor personal hygiene
Poor nutrition
Wounds or multiple infections

There is a proposed classification system for Madura.

1. Stage I refers to the extrinsic pressure effects on the intact bones in the vicinity of an expanding granuloma.
2. Stage II results from irritation of the bone surface without actual intraosseous invasion.
3. Cortical erosion and central cavitation occur in stage III.
4. If the disease spreads longitudinally along a single ray, stage IV is established.
5. Horizontal spread along a single row represents stage V.
6. Multidirectional spread due to uncontrolled infection is classified as stage VI.[5]

The Dot in Circle Sign describes the pathology of fungal infection in imaging. The central dot represents the fungal granule which is surrounded by T2 and STIR hyperintense microabscess surrounded by fibrous capsule. The tiny fungal granule returns hypointense signals on all MR sequences due to susceptibility. This sign on MRI reflects the unique pathological features of mycetoma and is likely to be a highly specific sign for this lesion as detailed in the international literature as case reports from many parts of the world.[6]

In 2018, a unique atypical case of unilateral foot swelling without draining sinuses - cryptic mycetoma in 69 year old male from Kerala, India.[7] The swelling was over the left foot following trauma and initially painless. He underwent MRI that revealed classical 'Dot In Circle' sign specific for mycetoma.

A total of 8 cases between 2004 and 2013 from Songklanagarind Hospital, Southern Thailand with histopathological proof of mycetoma affecting the musculoskeletal system, with MR specific sign of mycetoma are presented in the literature.[8]

In 2003, radiological and histological findings of two patients with fungal mycetoma of the foot are presented by Sarris et al.[9] MRI revealed multiple 2-5 mm lesions of high signal intensity interspersed within a low-intensity matrix.

Sen Anitha[10] in 2011 presented histologically proven eumycetoma in 50-year-old male from Tamil Nadu. MRI showed extensive soft tissue edema and center in dot sign in the foot and ankle region with tibial and talar involvement. CT scan showed bony osteolytic areas and showed periosteal reaction.

Like in a case of Sen Anitha, the initial differentials we proposed on MR appearances are soft tissue hemangioma, tuberculosis and atypical fungal infection. The "dots" were mistaken for phleboliths of hemangioma and rice bodies - hypointense foci seen in the synovial fluid of tendons or joints.

In this study, all are biopsy proven cases of fungal mycetoma and all are middle aged patients who presented initially with history of a indolent lump in the region of foot and all cases demonstrate the specific dot-in-circle sign.

After exhuberant search, the authors pledge that such research from radiology perspective has not been conducted in Pakistan apart from a case report.[11] The benefit of MRI in the diagnosis of madura is three fold, it is accurate in localizing the focus (es) and depth of soft tissue Infection and whether there is bone involvement or not. Second, presence of center in dot sign has near 100 percent accuracy for madura on MRI, patient can reach early diagnosis and treatment, reducing morbidity.

Acknowledgements: The authors thank Dr. Imran Soomro, Resident- III, Dow Institute of Radiology for assistance in access to histopathological notes and clinical data of patients.

Competing interests: The authors declare that they have no competing interests.

Funding disclosure: Nil

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