

Dot-in-circle Sign of Mycetoma on Magnetic Resonance Imaging

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Abstract

Mycetoma is a chronic granulomatous disease prevalent in tropical countries, but it also occurs in Europe and the United States. Early diagnosis is important as it has therapeutic implications. Although biopsy and microbiological culture provide the definitive diagnosis, these are difficult to achieve in many instances. The dot-in-circle sign is a recently proposed magnetic resonance imaging (MRI) sign of mycetoma, which is likely to be highly specific. We present a case of mycetoma of the left calcaneum with characteristic MRI features.

Key words: Disease, Mycetoma, MRI

INTRODUCTION

Mycetoma is a debilitating chronic granulomatous disease prevalent in tropical and subtropical regions, but it also occurs in Europe and the United States. It was first described in Madura, India in 1846; hence, the eponym Madura foot.^[1] The disease can be caused by two groups of organisms, the Eumyces or true fungi (eumycetoma) and the Actinomyces, which are filamentous bacteria of the order *Actinomycetales* (actinomycetoma).^[1] The evolution of the disease is slow and mostly painless. Patients present many years after the onset of infection, often with extensive soft tissue and bone involvement.^[2] The organism first lodges in the soft tissues. Bones are almost always attacked from outside, in contrast to bacterial osteomyelitis which occurs through the hematogenous spread, and periosteal reaction and cortical erosion may then be seen. Early diagnosis, before the appearance of sinuses and grains, is difficult. If left untreated, it may result in severe disability, often necessitating amputation. Although biopsy or microbiological culture of the discharge will yield the definitive diagnosis, both may be difficult to achieve with

fastidious organisms. Imaging can aid in the early diagnosis of the disease.

CASE REPORT

We present the magnetic resonance imaging (MRI) characteristics of a patient with mycetoma that demonstrated the recently described dot-in-circle sign, suggesting the possible diagnosis before the histological diagnosis. A 25-year-old male came to the emergency department with complain of swelling and pain in the left ankle and foot for 6 months. Progressive in nature, pain increases during walking. No discharging sinus or ulcer is seen. General examination was unremarkable. Blood and serum chemistry were also unremarkable. Plain radiograph of the left ankle with foot showed soft tissue swelling. No calcification or bone destruction was seen.

MRI was performed to characterize and evaluate the extent of the disease. T2-weighted and T2-weighted fat-saturated MRI revealed diffuse hyperintensity involving subcutaneous tissue, muscles with multiple focal fluid collections. In addition, multiple small discrete spherical hyperintense lesions were noted. In the center of some of these lesions, there was a tiny hypointense focus, resulting in the dot-in-circle sign. Small conglomerated low-intensity foci and microabscesses were also seen. The diagnosis of mycetoma was made based on these findings.

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DISCUSSION

The term mycetoma is a clinical entity, which applies to a chronic inflammatory process of soft tissue, usually of the foot, resulting from the implantation of one or various fungi or actinomycetes. Initially, there is the formation of soft tissue swelling with induration due to the underlying granulation tissue. It usually progresses to the formation of sinuses and extrusion of grains. The lesion may be confined to the soft tissue for years before bone involvement occurs. The diagnosis of mycetoma should not be considered by physicians when presentation is limited to soft tissues, without sinus or bone involvement. Although mainly a disease of the tropics, patients living in temperate regions may also be affected by this entity,^[3,4] they are often misdiagnosed as soft tissue tumors in the early stage.

Histopathologically, the inflammatory reaction of mycetoma is non-specific and in the absence of isolation of fungal grains, it is difficult to differentiate from other inflammatory soft tissue processes and cold abscesses,

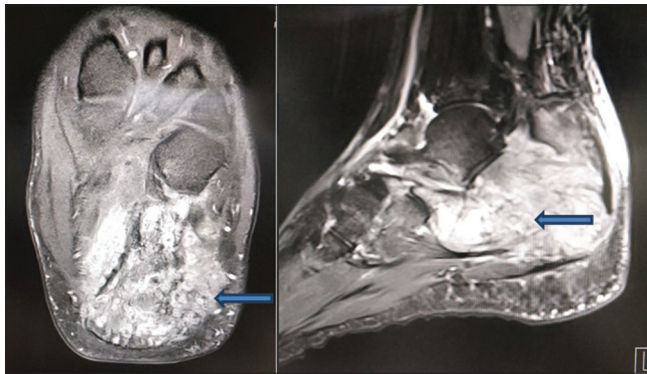


Figure 1: Magnetic resonance imaging of left ankle with foot. The dot-in-circle sign is a recently proposed magnetic resonance imaging sign of mycetoma, which is likely to be highly specific. Central dot sign (arrow) axial image

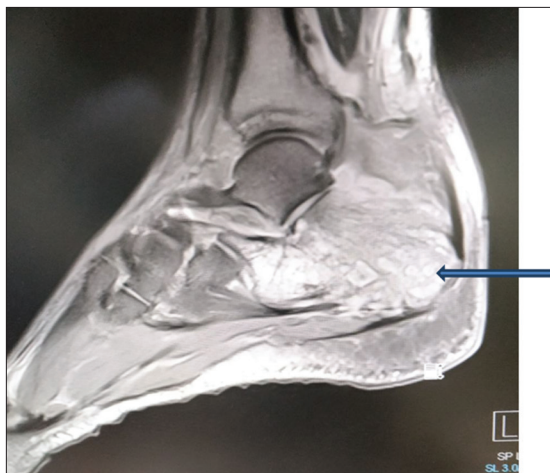


Figure 2: Sagittal T2-WI shows central dot sign (arrow) in the left calcaneum

which is not an uncommon occurrence. Although various radiographic bone changes have been described in cases of mycetoma, bone involvement occurs late in the course of the disease, when non-surgical cure is unlikely. Non-invasive imaging with MRI can characterize the soft tissue masses of mycetoma and aid in early diagnosis. Czechowski *et al.*^[5] described the magnetic resonance (MR) appearance of mycetomas and found small low-signal intensity lesions on T1-weighted and T2-weighted MR images in 16 of 20 patients. They suggested that these appearances were due to susceptibility from the metabolic products within the grains. They observed lesions showing a conglomerate of low-intensity foci, as were seen in the presented case.

The dot-in-circle sign [Figures 1 and 2] is a recently described sign reflecting the unique pathological feature of mycetoma. It is seen as a tiny hypointense focus within high-intensity spherical lesions. This sign was proposed by Sarris *et al.*^[6] in 2003 on T2-weighted, short inversion time inversion-recovery, and T1-weighted fat-saturated gadolinium-enhanced images. They correlated the MRI and histological findings in 2 cases of mycetoma and concluded

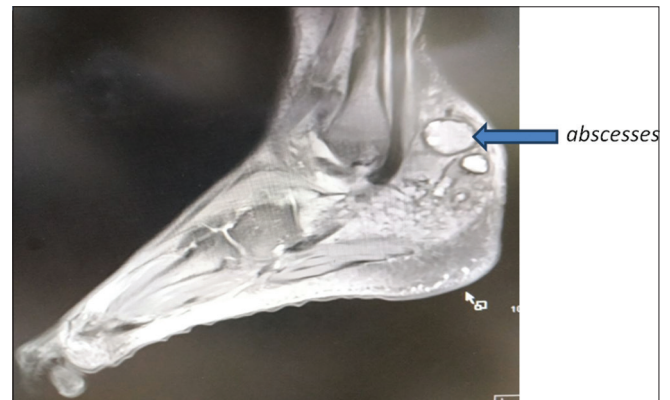


Figure 3: Magnetic resonance imaging of the Madura foot. A soft tissue ill-defined swelling involved the posterior aspect of the left ankle and foot likely microabscesses with the preserved architecture of calcaneum. In this image: Hyperintense conglomerate areas of nodules which were separated by a low signal intensity rim. In the center of some of these nodules, there is a tiny hypointense focus, resulting in the characteristic dot-in-circle sign, which indicates mycetoma grains

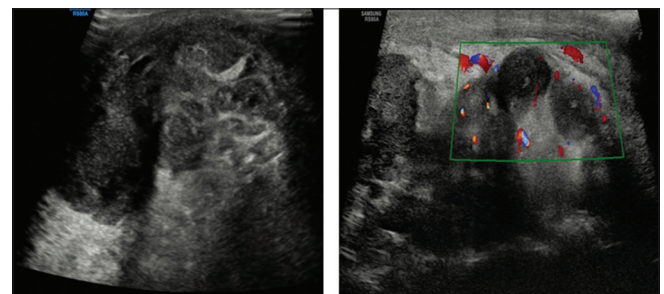


Figure 4: Ultrasonography of left ankle showing heterogenous hypoechoic lesion likely abscess formation

that the small central hypointense foci represented the fungal balls or grains [Figure 3], while the surrounding high-signal intensity foci represented the inflammatory granulomata.^[6] The low-intensity tissue seen surrounding these lesions represented the fibrous matrix. They proposed that it is likely to be a highly specific sign for mycetoma.^[6] We were able to demonstrate similar MRI findings in the presented case.

Few radiographic bone changes have been described to distinguish between actinomycetoma and eumycetoma.^[1] 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.*,^[1] a moth-eaten appearance caused by a combination of irregular periosteal reaction, periosteal erosion, and small cavities within bone was seen in 25% of cases of actinomycetoma, but in none of the patients with eumycetoma. The distinction between the two forms of soft tissue mycetoma was not possible with MRI.

To conclude, we stress the importance of MRI in the early diagnosis of mycetoma, even before the development of sinuses and/or extrusion of grains. Furthermore, as

these fastidious organisms may be difficult to demonstrate either on biopsy or microbiological culture, the clinical picture often necessitates multiple surgical biopsies, thus exacerbating morbidity due to delays in diagnosis and therapeutic intervention. MRI can be useful in such situations. It can strongly suggest the diagnosis of mycetoma when it demonstrates the dot-in-circle sign, conglomerated low signal intensity foci or microabscesses [Figure 4] in the background of a hypointense matrix as described above.

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