Case report: Dot-in-circle sign - An MRI and USG sign for “Madura foot”

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Abstract

Mycetoma is a chronic granulomatous disease that is more common in tropical than in temperate regions. Early diagnosis is important due to the therapeutic implications. Although biopsy and microbiological culture provide definitive diagnosis, they are time-consuming procedures and may not be able to provide a definite diagnosis in cases of fastidious organisms. The “dot-in-circle” sign has recently been proposed as a highly specific magnetic resonance imaging (MRI) and ultrasonography (USG) sign of mycetoma, which may allow a noninvasive as well as early diagnosis. We present a case of histologically proven mycetoma that demonstrated this sign.

Key words: Dot-in-circle; magnetic resonance imaging; mycetoma; ultrasonography

Introduction

Mycetoma is a chronic granulomatous disease affecting mainly the feet, which are more prone to trauma, and hence more likely to get infected, as compared to other organs in the body such as the lower legs, hands, head, neck, chest, shoulders and arms. It is more common in males between the ages of 20 and 50 years.[1] Although mainly a disease of the tropics, patients residing in temperate regions may also be affected. A noninvasive and early diagnosis may be possible with USG[3,4] and MRI.[3,5-8]

Case Report

A 50-year-old male from Tamil Nadu presented with a soft tissue swelling of his ankle and foot. Workup included punch biopsy, from which a diagnosis of eumycetoma was made.

MRI showed extensive soft tissue edema in the foot and ankle region with tibial and talar involvement [Figure 1A] and multiple small cystic areas with central hypointensity, suggestive of the “dot-in-circle sign” [Figure 1A, C, D]. CT scan confirmed bony osteolytic areas and showed periosteal reaction [Figure 1B]. USG demonstrated hypoechoic areas with central hyperechoic foci [Figure 2].

Discussion

Mycetoma or Madura foot is a chronic granulomatous infection of the dermis and epidermis caused by the bacteria Actinomyces (Actinomycetoma) or by true fungi (eumycetoma).[9] It was first described in the Indian district of Madura in 1846, hence the eponym Madura foot.[10] Endemic in Africa, Mexico and India, it is also found in Central and South America and the Middle and the Far East.[11] Eumycetoma is more common in areas with scarce rainfall and actinomycetoma in areas of abundant rainfall.[12]

The infecting organism is presumed to be directly inoculated after penetration of the skin with a sharp object,[13] e.g., a thorn. Patients present with painless subcutaneous nodules and fistulae, from which a purulent exudate may be discharged. The process is usually indolent but with a potential for abscess formation, draining sinus tracts, osteomyelitis, and fistula formation,[14] with severe deformity and disability ensuing if treatment is not provided. Although antifungal medication is successful in almost 90% of cases, lesions not arising in the foot or
due to fungus tend to have a worse prognosis and require surgery.\[13\]

Histologically, the lesion consists of “grains” of fungal hyphae or bacteria in microabscesses within a granulomatous fibrous-tissue reaction.\[14\] Gram stain, Gomori methenamine silver, periodic acid-Schiff and lactophenol blue stains are useful to differentiate actinomycetoma and eumycetoma.\[15\] Early laboratory diagnosis, before the appearance of the sinuses and grains, is difficult. Though biopsy (with demonstration of the characteristic features) or staining and microbiological culture of the discharge from the lesion usually gives the definitive diagnosis, both are time-consuming procedures and diagnosis may be difficult to achieve, especially with fastidious organisms.

Radiographs may be normal, demonstrate soft tissue enlargement, bone sclerosis, bone cavities, periosteal reaction, bone expansion, extrinsic cortical scalloping, fanning of the rays or osteoporosis.\[17\] The bones are almost always attacked from the outside, in contrast to bacterial osteomyelitis.\[18\] Radiographic classification of bone involvement (stages 0–6) has been suggested.\[18\] A few radiographic bone changes have been described that help distinguish between actinomycetoma and eumycetoma.\[14\] Eumycotic lesions tend to form a few cavities in bone that are ≥1 cm in diameter, while actinomycetes often form smaller but more numerous cavities, leading to a moth-eaten appearance.\[14\] CT scan provides better delineation of the bone changes than radiographs.\[19\]

Initial reports of the MRI findings of mycetoma described lesions with low signal on T1W and T2W images, which were assumed to be due to susceptibility from the metabolic products of the ‘grains’.\[20\] The “dot-in-circle” sign, seen as tiny hypointense foci within the hyperintense spherical lesions, was initially described by Sarris et al.,\[21\] in 2003 on T2W, STIR, and T1W fat-saturated gadolinium-enhanced images. Correlating the MRI and histological findings, they suggested that the high-signal areas seen on MRI represented inflammatory granulomata, the low-intensity tissue seen surrounding these lesions represented the fibrous matrix, and the small central hypointense foci within the granulomata represented the fungal balls or grains. They proposed that it is likely to be a highly specific sign for mycetoma. It was also later reported in 2007,\[22\] 2009,\[23,24\] and 2010.\[25\] The last (2010) case was misdiagnosed as a soft tissue hemangioma on MRI due to the presence of serpiginous enhancing masses with the “dot-in-circle” sign (the “dots” were mistaken for phleboliths). Another differential for the “dots” is rice bodies – hypointense foci seen in the synovial fluid of patients with articular or tendon tuberculosis.\[21\]

The USG appearances were initially described by Fahal et al.,\[4\] who demonstrated on in vitro imaging of the mycetoma lesions that the hyper-reflective echoes corresponded to the grains; eumycetoma grains produce sharp hyperechoic foci, while actinomycetomas produce fine hyperechoic foci that commonly settle at the bottom of the rounded lesions. The USG “dot-in-circle” sign is similar to the MRI sign, with multiple round hypoechoic lesions containing hyperechoic foci.\[3,4]\n
Sarris et al. had predicted that with the increasing availability of MRI in the metropolitan centers of countries where mycetoma is endemic, the sensitivity and specificity of this sign could be determined. This case from Tamil Nadu, the land of the Madura foot (Madura being a district in the Indian state of Tamil Nadu), reiterates the specificity of the “dot-in-circle” sign.

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