INTRODUCTION

Mycetoma is a slowly progressive localised, granulomatous, subcutaneous tissue infection common among adult males aged 20 - 50 years. The causing agents include both bacteria (actinomyctoma) and fungi (eumycetoma). This chronic infection was described in literature as Madura foot and eventually mycetoma, due to its etiology. Early clinical diagnosis before the appearance of sinuses and grains (aggregates of organism surrounded by granulation tissue, which are discharged from the draining sinuses) is difficult. Early diagnosis is important due to the therapeutic implications. Although biopsy and microbiological cultures provide definitive diagnosis. Delay in diagnosis may lead to amputation of the affected part. The recently described "dot-in-circle" sign on magnetic resonance imaging (MRI) is easy to recognise and highly specific. We present a case of histologically proven mycetoma with characteristic MRI features.

CASE REPORT

A 58-year male of Nagozi village presented to our hospital with progressively increasing diffuse soft tissue swelling of the left ankle and feet for the past 10 years. On examination, discharging sinuses were observed. Plain radiograph of the foot showed marked soft tissue swelling in foot region, extending to the lower leg. Multiple well defined punched out lytic defects were seen involving the tarsal and metatarsal bones as well as distal tibia. There was marked joint space reduction with cortical irregularity, periosteal reaction, and patchy areas of increased sclerosis involving the tarsal and metatarsal bones (Figure 1A).

MRI foot with T1, T2 weighted and STIR images was also performed to characterise and evaluate the exact extent of disease process. There was an infiltrating low T1, heterogenous high STIR (short-tau inversion recovery) signal abnormality diffusely involving the soft tissues of the left foot extending up to the ankle with complete obliteration of the normal anatomical planes. There were multiple rounded abnormal T2 and STIR bright signal areas with a central hypointense dot and peripheral hypointense rim in bones and soft tissues, suggesting granuloma formation. The metatarsals also showed cortical thickening and sclerosis with lamellated periosteal reaction, more pronounced in the fifth metatarsal bone. To lesser degree, bony changes in the lower tibia were also noted. The disease was more pronounced at the second-fourth tarsometatarsal joints (Figures 1B, 1C and 1D).

On the basis of MRI findings, the diagnosis of mycetoma was made. Biopsy was performed, which showed granulomatous inflammation. Microbiological tests revealed the presence of actinomycetes. There was no evidence of fungal infection or malignancy. Below knee
amputation of foot was performed and he was prescribed antimicrobial therapy. Follow up after one month showed complete wound healing.

**DISCUSSION**

Mycetoma or Madura foot is a clinical entity which applies to chronic granulomatous infection of the soft tissues. After penetrating skin injury, such as a thorn prick, these normal inhabitants of the soil are introduced into the skin. There is development of soft tissue swelling with induration due to underlying granulation tissue. Patients usually present with painless subcutaneous nodules, fistulae and discharging sinuses, from which a purulent exudate may discharge. This is an indolent process but there is a potential for abscess formation, draining sinus tracts, osteomyelitis, and fistulas, with resultant severe deformity and disability if treatment is not provided. Antifungal medication is successful in about 90% of cases; but the lesions not arising in the foot or due to fungus, tend to have a worse prognosis and require surgery.

Histologically, these lesions comprise of grains of fungal hyphae or bacteria in microabscesses within a granulomatous fibrous-tissue reaction. Techniques available for differentiating actinomyctoma and eumycetoma include gram stain, gomorimethenamine silver, periodic acid-schiff and lactophenol blue. But early laboratory diagnosis, before the appearance of the sinuses and grains, is difficult. Definitive diagnosis can be made through biopsy (with demonstration of the characteristic features) or staining and microbiological culture of the discharge from the lesion; but these are time-consuming procedures, and diagnosis may be difficult to achieve, particularly with fastidious organisms.

Radiographic bone changes may include soft tissue swelling, bone sclerosis, bone cavities, bone expansion, extrinsic cortical scalloping, periosteal reaction, fanning of the rays or osteoporosis. Radiographic classification of bone involvement (stages 0-6) has been described in literature. Contrary to the bacterial osteomyelitis, bones are almost always attacked from the outside.

Few radiographic osseous changes can differentiate between actinomycetoma and eumycetoma. Eumycotic lesions tend to form few cavities in the bone ≥1 cm in diameter. However, actinomycetes often form numerous but smaller cavities. Lewall et al. found that 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. Bone changes are better delineated on CT scan as compared to radiographs.

On ultrasound, dot-in-circle sign is similar to the MRI sign, with multiple round hypoechoic lesions containing hyperechoic foci. It was first coined by Fahal et al. 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.

Mycetoma is characterised by the formation of aggregates of the organism, known as grains, which are present within abscesses surrounded by granulation tissue. On MRI, these are depicted as conglomerates of small (2 - 5 mm) round hyperintense lesions, representing the inflammatory granuloma, surrounded by low-signal intensity rim, representing intervening fibrous septa. The central low-signal intensity focus is the result of susceptibility effect caused by the presence of fungal grains. This unique appearance, known as the dot-in-circle sign, seems to be highly suggestive of mycetoma, and was first proposed by Sarris et al. in 2003 on T2-weighted short-tau inversion recovery (STIR), and T1-weighted fast spin gadolinium-enhanced images in two cases. They also correlated the MRI findings with histological findings. Cherian et al. described similar appearance in three cases. The distinction between the two forms of mycetoma is not possible with MRI.
To conclude, the dot-in-circle sign on MRI could diagnose the disease process in initial stages, even before the development of sinuses or extrusion of grains; so appropriate treatment strategy can be implemented. Antimicrobial therapy is mostly curative in cases of mycetoma involving the soft tissues; but if there is involvement of bones, partial resection or amputation of the foot could be the final available resort.

REFERENCES