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Case Report

Maduramycosis masquerading malignancy: A case report from rural oncology center

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ABSTRACT

Mycetoma is a chronic granulomatous inflammation of the dermis and epidermis caused by filamentous bacteria in 60% of cases (actinomycetoma) or fungi (eumycetoma) in 40% of cases. A 45-year-old female presented with a left foot ulcer and gradually increasing swelling for 13 years in our department. The radiograph revealed multiple punch-out lytic areas in the tarsals with sclerosis and destruction of the calcaneum. The characteristic "dot-in-circle" sign was appreciated on both ultrasound and magnetic resonance imaging. The causative organism and the radiological evaluation, especially bone involvement, are the most important factors that determine the choice of treatment. In the initial stages, chemotherapeutic drugs along with surgery are effective. The current concept is with treatment with a combination of drugs under the name of Welsh regimen.

Keywords: Maduramycosis, Eumycetoma, Dot-in-circle, Welsh regimen

INTRODUCTION

Mycetoma, often known as "Madura foot," is a persistent granulomatous infection of the dermis and epidermis that was first described in 1843 in Madurai, India, by Dr. John Gill. In 60% of instances, the causative organisms are filamentous bacteria (actinomycetoma), which are more widespread in locations with ample rainfall, or fungi (eumycetoma), which are more common in areas with insufficient rainfall.[1]

Young adults, particularly men in their third and fourth decades, are frequently affected; males are 4 times more likely to experience it than females. According to legend, the disease is spread through direct injection following skin puncture by a sharp object or other trauma. It is not a contagious illness, though. Due to this, it is more common in rural areas and affects manual workers more such as farmers and laborers, those who walk barefoot.[2] It is a slowly progressive disease with incubation periods ranging from 3 months to 10 years.

CASE PRESENTATION

A 45-year-old female presented with left foot ulcer and gradually increasing swelling for 13 years and presented to the civil hospital and she was misdiagnosed as soft-tissue sarcoma and she was referred to our tertiary care oncology center.

It all started from a small swelling in the sole which later ruptured and the similar process repeated in adjacent soft tissue until the entire foot was affected.

She sought medical advice and was diagnosed with a foot ulcer and was prescribed antibiotics which brought no improvement. The patient is a homemaker and denies any history of any trauma or penetration.

There was no history of diabetes mellitus, tuberculosis, hypertension, or any immunosuppression.

On physical examination [Figure 1], marked swelling and deformity with innumerable nodules and a few draining sinuses were noted on the left foot.

There was tenderness on deep palpation with mild limitation of range of movement.

Her renal function test and urine examination was within normal limits. Complete blood count and blood sugar level were also normal.

She was negative for hepatitis C virus, hepatitis B virus, and human immunodeficiency virus. The acid-fast bacilli smear was also negative. She was rheumatoid factor negative however her C-reactive protein was positive.

The radiograph [Figure 2] revealed multiple punch-out lytic areas in the tarsals with sclerosis and destruction of the calcaneum. Narrowing of joint space and soft-tissue swelling was also noted.

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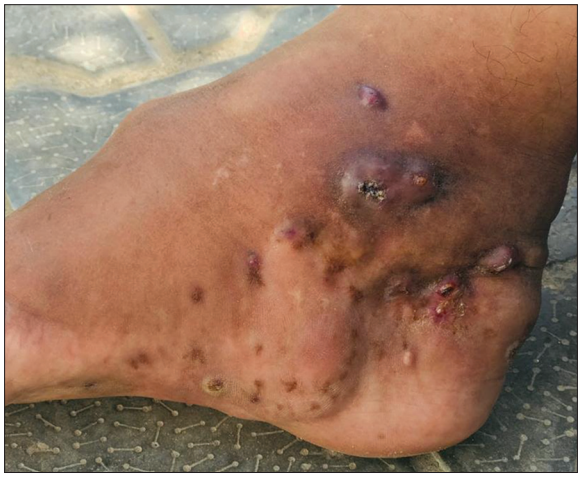

Figure 1: Marked swelling, inflammation, and deformity with numerous nodules and few draining sinuses were found. There was mucopurulent discharge from the lesion.

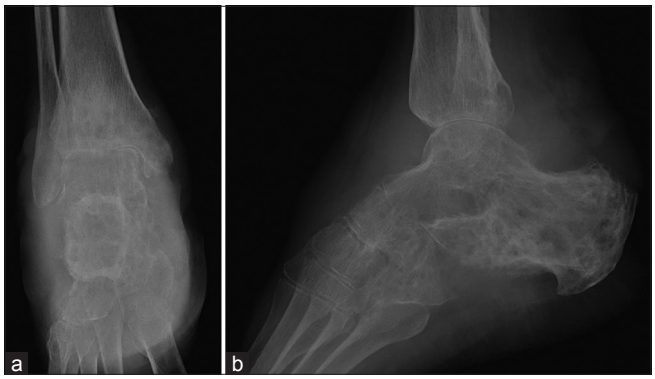

Figure 2: (a) Frontal radiograph and (b) lateral radiograph revealed multiple punch-out lytic areas in the tarsals with sclerosis and destruction of calcaneum. Narrowing of joint space and soft-tissue swelling was also noted.

Contrast-enhanced computerized tomography (CT) was performed which better denoted the multiple small punched-out lesions in the calcaneus with associated sclerosis. CECT also demonstrated mild synovial effusion in the ankle joint associated with osteoarthritic changes in the tibiotalar, tibionavicular, and talocalcaneal joints, demonstrating features of chronic osteomyelitis.

On magnetic resonance imaging (MRI) [Figure 3], the outstanding abnormality was innumerable sub cm sized soft tissue and osseous T1 hypointense and T2 hyperintense lesions with peripheral hypointense rim and adjacent inflammatory changes, studded in the soft tissue as well as bones such as the lower end of tibia and fibula, calcaneum, talus, navicular, all cuneiforms, cuboid, and metatarsals.

Nearly, all tendons were also involved, predominantly the tendon Achilles.

Ultrasound (USG) [Figure 4] was performed which supported the diagnosis and showed multiple clustered lesions, which have a hypoechoic periphery and hyperechoic center. No internal movement or significant vascularity is seen.

DISCUSSION

Clinically, the patient presents with a painless hard soft-tissue swelling, multiple sinus tracts, and characteristic macroscopic grains. The foot is the most common site as they are prone to trauma nonetheless all parts of the body can be affected. To begin with, soft-tissue swelling occurs due to the genesis of granulation tissue. In addition, this granulation tissue causes the development of numerous discharge sinuses, resulting in grains whose color points in the direction of the causal organism. In addition, over many years, bone involvement happens gradually. Last but not least, if the patient is not treated, persistent osteomyelitis may manifest.[3]

In the early stages of the disease, radiographs only show soft-tissue swelling, which might be mistaken for a soft-tissue tumor or a persistent, inactive bacterial or tubercular infection. Multiple soft-tissue nodules may also be mistaken for cysticercosis, which exhibits oblong calcific specks parallel to muscle fibers in the skeletal muscles, giving the illness a notorious "rice grain appearance." Cysticercosis also seldom affects the bones.

Davies from Uganda provides a thorough account of bone involvement, including the early stages of soft-tissue invasion, the stage of external cortical invasion, and the last stage of spread across the entire bone. Bone irritation starts with an increase in extrinsic pressure and progresses to bone degradation or cavitation. Invasion of nearby structures and mutual involvement are also recorded. Ultimately, total disruption occurs.[4]

Bony changes on radiographs are occasionally helpful in the differentiation of eumycetoma (tendency to form few and bigger cavities) from actinomycetoma (more and smaller cavities giving a moth-eaten appearance).[5]

Furthermore, high-resolution USG may be used to distinguish between eumycetoma and actinomycetoma. Eumycetoma grains are numerous, sharp, and exhibit highly reflective echoes in single or multiple cavities with thick walls, while actinomycetoma grains are small, closely aggregated, and settle at the bottom of cavities. In contrast to eumycetoma, actinomycetoma is more frequently associated with muscle involvement.

CT scan is helpful in depicting the extent of bony changes and is better than radiographs.

Due to its superior soft-tissue contrast, multiparametric evaluation capabilities, and quantitative sequences like diffusion-weighted imaging, MRI is the preferred radiological investigation. To diagnose and assess the severity of the condition, an MRI of the left ankle and foot is done.

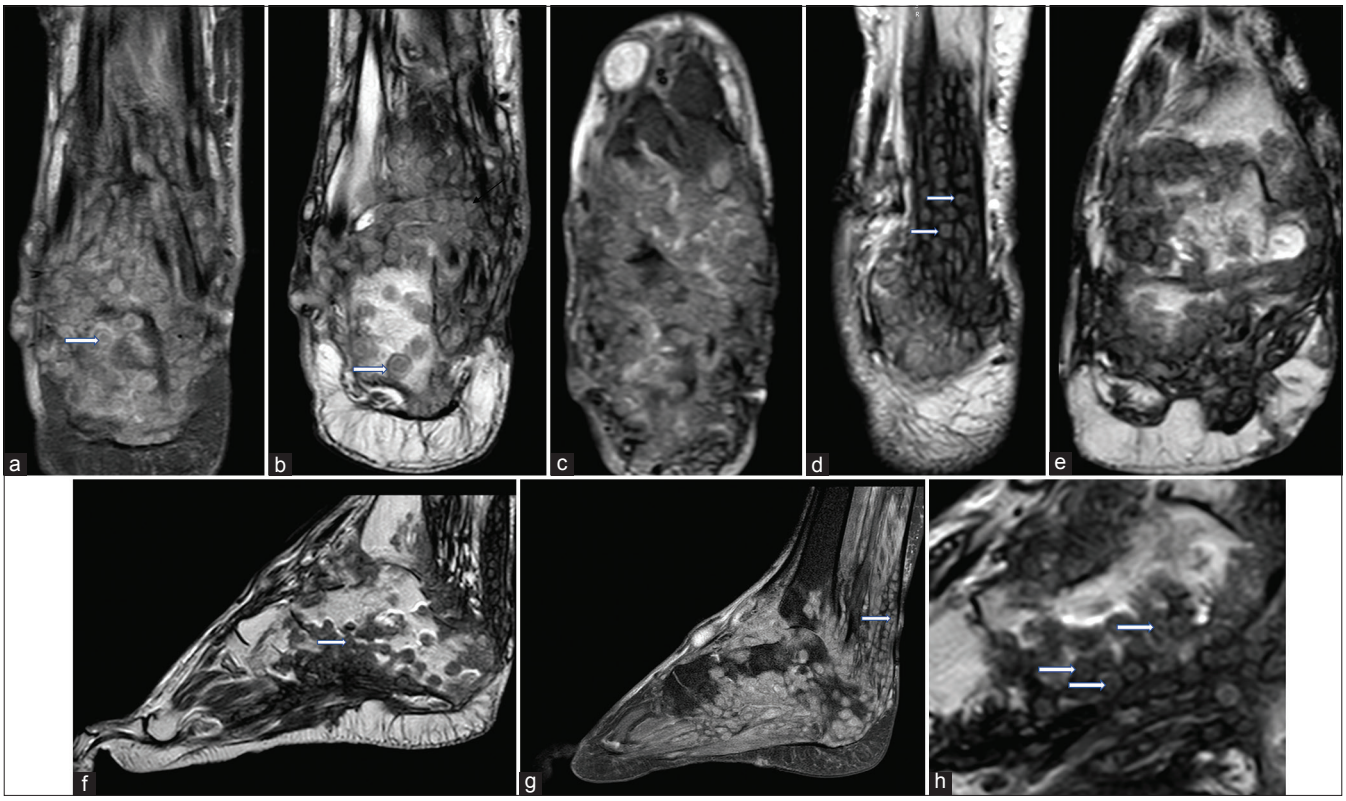

Figure 3: Magnetic resonance imaging (MRI) findings of foot with mycetoma (a) Short-tau inversion recovery (STIR) (b) Cor T2 (c) Cor STIR showing bone and joint involvement by mycetoma with multiple small, round to spherical hyperintense lesions (arrows). MRI findings: (d and e) different sections of Cor T2 showing achilles tendon studded with multiple small, round to spherical hyperintense lesions. Arrows showing "dot-in-circle" sign. MRI findings of left foot with mycetoma: (f) Sagittal T1 without contrast agent shows multiple isointense lesions containing some central hypointense dots and surrounded by hypointense tissue (arrow), and (g) short-tau inversion recovery images showing central hypointense dot, resulting in the "dot-in-circle" sign (arrows). The surrounding hyperintense inflammatory soft-tissue changes are also seen. (h) Magnified view of "dot-in-circle" sign. Some of the lesions are showing central hypointense dot surrounded by hyperintense inflammatory tissue.

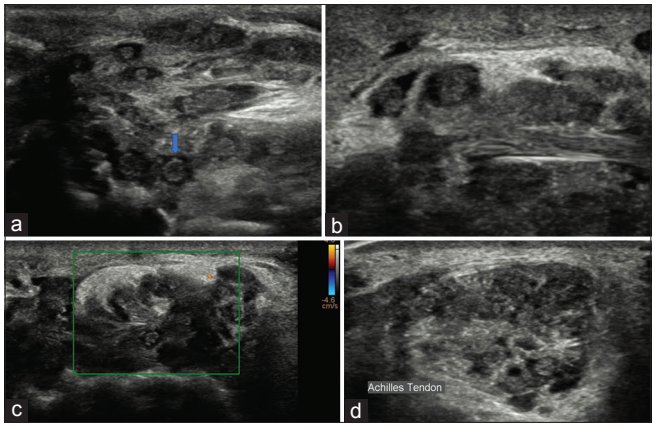

Figure 4: (a) High-resolution sonography image and (b) magnified image showing multiple clustered lesions, showing hypoechoic thick-walled lesions containing small, hyperechoic foci. No internal movement was seen. (c) On Doppler, there was no significant vascularity. (d) Multiple nodules were also seen in Achilles tendon.

The distinctive "Dot-in-Circle" indication on MRI, first identified by Sarris *et al.* in 2003, is characterized by minute hypointense foci within of hyperintense spherical or circular lesions on T2w and short-tau inversion recovery sequences, which are thought to be caused by susceptibility from the metabolic products of the "grains."[6]

Microbiological culture is the gold standard for diagnosis; however, because the organism is challenging to cultivate, histopathology is another reliable test. Tender filamentous filaments (hyphae) are seen in tissue samples mixed with histiocytes, inflammatory cells, connective tissue, and neutrophils. Fine-needle aspiration cytology frequently fails to provide a diagnosis, necessitating a biopsy.

The most crucial elements that affect treatment selection are the causal agent and the radiological assessment, particularly bone involvement. Chemotherapeutic medications and surgery are beneficial in the early stages. Recurrence rates are extremely high, ranging from 20% to 90%, and inadequate resection is their main cause.[7] An early diagnosis, determining the full scope of the disease, and keeping track of recurrence are all made possible by MRI.

When the patient has bone involvement, modern chemotherapy becomes ineffective, and surgery becomes mandatory.

Welsh regimen is used for treatment that involves a pharmacological combination.[8]

It includes Injection Amikacin 15 mg/kg intravenous divided unto two doses for 21 days followed by Tab Trimethoprim-sulfamethoxazole (7 and 35 mg/kg/day) for 6 months.

CONCLUSION

We conclude that imaging is crucial for diagnosis and for planning the treatment of maduromycosis. "Dot-in-circle" sign is very specific for mycetomas and is seen in both USG and MRI and treatment can be initiated even if the histopathology is non-conclusive. A good prognosis can be ascertained with accurate diagnosis combined with aggressive surgical approach and patient's adherence to prolonged adjuvant medication.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The author(s) confirms that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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