Radiology Case Reports 19 (2024) 3354–3357

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journal homepage: www.elsevier.com/locate/radcr

Case Report

Early presentation of mycetoma (Madura Foot): A case report ☆

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ARTICLE INFO

Article history:
Received 24 March 2024
Revised 25 April 2024
Accepted 1 May 2024

Keywords:
Madura foot
Mycetoma
Eumycetoma
Actinomycosis
Dot-in-circle

ABSTRACT

Mycetoma, commonly known as Madura foot, is a chronic and progressively destructive granulomatous disease caused by a fungus or anaerobic filamentous bacteria that affects the skin, subcutaneous tissue, and bones primarily in tropical and subtropical regions, with males between the ages of 20-40 having occupational exposure to outdoor environments, such as farming, predominantly affected. It is one of the World Health Organization's 17 "neglected tropical diseases," characterized by a clinical trial of localized mass-like soft tissue injury with draining sinuses that discharge grains of infectious material. Here, we present a case report of a 40-year-old male with type 2 diabetes mellitus and a history of fieldwork, who exhibited early manifestations of mycetoma. Unlike the typical diffuse presentation seen in advanced cases, this patient's early presentation prompted diagnostic challenges due to its atypical nature. We highlight the importance of recognizing the early signs of mycetoma, particularly in individuals with predisposing factors such as diabetes and occupational exposure. Diagnostic dilemmas may arise, leading to potential misdiagnosis. Additionally, we emphasize the crucial role of biopsy in confirming the diagnosis, alongside imaging techniques, to facilitate timely intervention and management, thereby significantly impacting patient outcomes.

Background

The history of mycetoma dates as far back as the Byzantine period (AD 300-600) [1]. However, it was first reported by Gill in 1832 for the unique nodular pattern of the leg among the field laborers in the Madurai district of Tamil Nadu, India. Colebrook gave it the name Madura Foot in 1846. Mycetomas that are a result of fungal infection are referred to as eumycetoma, whereas those caused by aerobic actinomycetes are known as actinomycosis [2]. They occur following the inoculation of organisms, which can happen through thorn pricks or preexisting abrasions or trauma that go unnoticed. Cells in the innate immune system try to engulf and disable these organisms. The disease starts as a minor, painless nodule under the skin. This nodule slowly gets larger, adheres to the tissue un-

☆ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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https://doi.org/10.1016/j.radcr.2024.05.007

derneath, and finally develops sinus tracts that leak pus with granules of various colors [3].

Case presentation

A 40-year-old male type 2 diabetic patient, who has been on metformin for the last five years and works as a field worker, noticed a small swelling on the medial surface of his ankle two years ago. The swelling slowly increased in size, causing minor discomfort when wearing shoes. Four months before his presentation, he began to notice dark granular discharge from the swelling. He was given unspecified topical ointment, however, there was no improvement. Apart from this swelling, the patient is relatively healthy with no fever, night sweats, weight loss, and no difficulty walking.

A 2 × 2 cm swelling was observed in the right medial calcaneal region during the physical examination. The overlying skin appeared normal, with no redness, warmth, or tenderness. A 3 × 3 mm opening was noted in the center of the swelling, through which black dot-like granules discharged upon pressure. The range of motion of the ankle joint was maintained. Laboratory investigations were within the normal range, including complete blood count, erythrocyte sedimentation rate, liver function tests, and renal function tests. HbA1c was controlled (6.2%).

Initial investigation with an X-ray revealed multiple calcaneal lytic lesions with wide zones of transition and diffuse sclerotic changes. A focused ultrasound scan showed multiple hyperechoic foci surrounded by hypoechoic lesions resembling "dot in circle." An ankle MRI with MR PD fat-sat sequence revealed multiple well-defined small high signal intensities with central hypointensity, also resembling "dot in circle." Fine needle aspiration from the lesion suggested fungal elements.

Imaging findings

The patient then was subsequently investigated by X-ray (Fig. 1), ultrasound (Fig. 2), and MRI (Fig. 3). The X-ray findings were multiple lytic-permeative lesions having wide zones of transition in the calcaneal neck with associated diffuse sclerotic changes of the surrounding bone. A focused ultrasound of the swelling showed multiple soft tissue hypoechoic relatively well-defined rounded nodules with central echogenic foci. The foci did not show any posterior acoustic changes. Sagittal and axial MR images with fat-saturated proton density sequence revealed multiple small, well-defined round high-signal lesions with a central low signal intensity on the calcaneal neck and the adjacent soft tissue. Additional features suggestive of diffuse bone marrow edema of the calcaneus were also noted.

Histopathology

Fine needle aspiration performed from the soft tissue lesion yielded findings consistent with fungal elements upon microscopic examination using potassium hydroxide (KOH) staining.

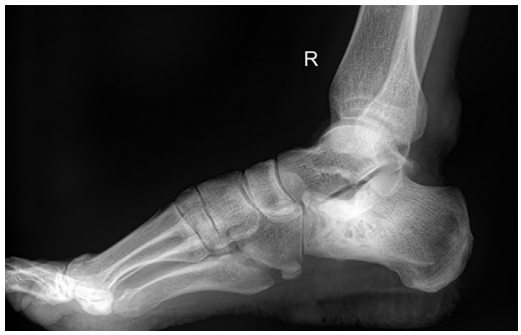

Fig. 1 – Lateral X-ray of the ankle showing nodular soft tissue opacity with small rounded lucencies and multiple lytic permeative lesions calcaneal lesion with diffuse sclerosis.

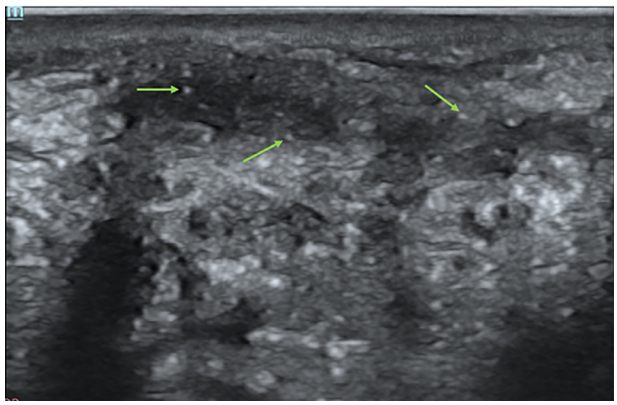

Fig. 2 – Focused high-frequency ultrasound over the soft tissue in the medial calcaneus showed thickening and heterogeneity with multiple hypoechoic lesions showing central echogenic foci (green arrows).

Discussion

Madura foot is frequently observed in young men aged 15 to 30, predominantly in developing nations with limited economic resources. Those who frequently walk barefoot, including individuals involved in manual jobs such as agriculture, labor-intensive, and herding, are at a higher risk of contracting this condition [4].

In many instances, patients affected by this condition often encounter delays in seeking medical attention, primarily due to limited access to advanced care facilities. Consequently, they typically present at later stages of the disease, characterized by notable local swelling, sinus formation, discharge of distinctive grains, and varying degrees of pain [3]. However, when patients from atypical demographics seek medical evaluation, diagnosis can prove challenging. In this case report, a patient initially diagnosed with acute osteomyelitis prompted

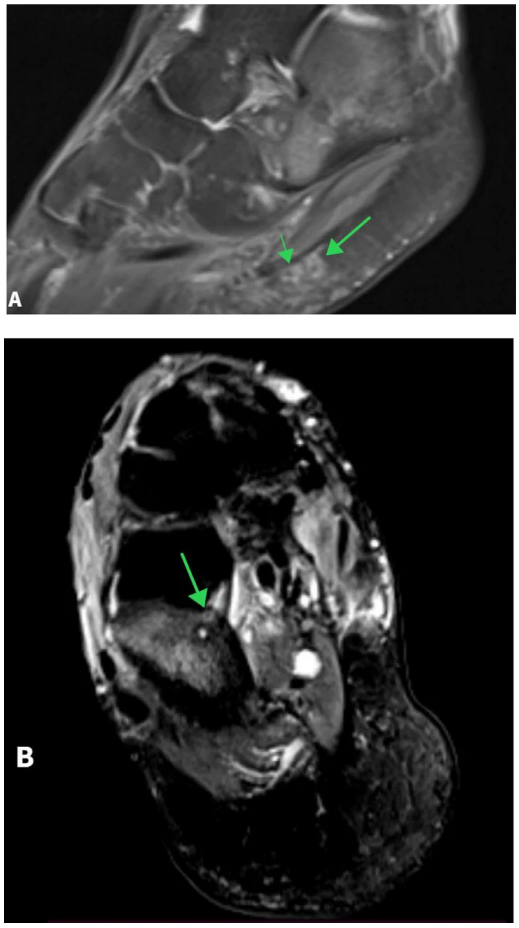

Fig. 3 – (A and B) MR PD fat-sat sequence showing well-defined round high signal intensities with central hypointense dots. (Dot-in-circle) with diffuse high marrow signal suggested associated marrow oedema.

clinicians to seek a second opinion due to observed clinical and radiological inconsistencies. Thorough clinical history is crucial for accurate diagnosis, especially in urban populations where radiologists might overlook certain conditions.

In most advanced cases, X-ray findings typically show lobulated soft tissue swelling, moth-eaten lytic changes, periosteal reaction, cortical erosion, rounded lucencies, joint destruction, osteopenia, and bone lysis [4,5]. In our case report even though most of these features are not present there are multiple small, rounded lucency and nodular soft tissue swelling that could suggest the diagnosis. The differential solely in X-ray could be vast including ranging from pyogenic and tuberculosis osteomyelitis to aggressive bone tumors. However, the findings observed on ultrasound and MRI can help to narrow the diagnosis to Madura foot. Ultrasound can show a “dot-in-circle” sign. This is characterized by a hyperechoic central area (the dot) surrounded by hypoechoic tissue (the circle) [6]. This "dot-in-circle" sign has been described more often on MR images and is considered to be highly specific. The typical feature is small round high-signal intensity lesions that depict granulation tissue and are surrounded by a low-signal intensity rim representing fibrous septa. The central low signal intensity dot is created due to the susceptibility of fungal grains [2,7-9]. This sign is considered to be specific for the diagnosis of soft tissue mycetoma, seen in up to 80% of individuals with the disease [7]. The main differential diagnosis for the dot in circle sign on MRI is phleboliths in hemangiomas and rice bodies in synovial fluid in rheumatoid arthritis or tuberculosis [10,11].

Differentiating Madura foot from other differentials should be based on clinical, laboratory, and imaging assessments. Chronic pyogenic osteomyelitis of the calcaneus ranges from 3%-10% of all osteomyelitis cases and is mostly associated with diabetes which would put our patient at risk [12]. However, pyogenic osteomyelitis usually has a relatively acute clinical presentation and associated symptoms such as pain, erythema, and tenderness [13]. X-ray could be a challenging modality to solely differentiate between chronic pyogenic osteomyelitis and Madura foot because both might show thick, irregular, sclerotic bone interspersed with radiolucencies. However, in cases of osteomyelitis, additional signs of periosteal reaction and sequestra formation might be a clue to suggest the diagnosis [14]. When incorporating ultrasound and MRI differentiating is easier. Osteomyelitis is usually characterized by diffuse marrow edema on MRI [15]. If an intraosseous abscess occurs/Brodies abscess is usually a well-defined solitary and in an oval configuration the pathognomic feature is a penumbra sign which is characterized by T1 hyper-intense outer ring enhanced on contrast administration with central hypointensity. The T1 hyperintense outer ring has T2 hypointensity [16]. In addition to the disparity in signal intensity on MRI between the Madura Dot in Circle sign and the penumbra sign of Brodie’s abscess, the latter typically appears as a relatively larger solitary lesion and exhibits enhanced contrast uptake following administration. Tuberculous osteomyelitis clinically may present with localized swelling with or without warmth or erythema. However, associated systemic symptoms such as low-grade fever, night sweats, and weight loss are expected with tuberculosis. Furthermore, having a normal ESR value makes TB less likely [17,18]. On X-ray tuberculous osteomyelitis shows osteopenia and poorly defined lytic lesions with minimal surrounding sclerosis [19]. However, our patient shows diffuse sclerotic changes which make it less likely. On MRI tuberculous osteomyelitis has similar features to pyogenic osteomyelitis and similar distinguishing features could be used to exclude it from the diagnosis.

Conclusion

In conclusion, the diagnosis of Madura foot can be challenging, particularly when patients are presented with atypical demographics or clinical features. However, this case highlights the importance of obtaining comprehensive clinical data alongside typical radiological findings for accurate diagnosis. While X-ray findings may not always be conclusive, ultrasound and MRI can provide crucial diagnostic clues, such as the "dot-in-circle" sign which is considered highly specific. Therefore, a multidisciplinary approach integrating clinical history, imaging modalities, and typical features remains paramount in effectively diagnosing Madura foot.

Patient consent

Written informed consent was obtained from the patient's parents for anonymized patient information to be published in this article.

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