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TEST YOURSELF: ANSWER

Chronic swelling of the foot

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Discussion

In the case demonstrated, radiography of the great toe (Fig. 1) shows sclerosis and periostitis on either side of the proximal phalanx of the first digit, and flowing sclerosis in the medial aspect of the proximal phalanx of the second digit. Axial and coronal T2-weighted fat-suppressed images (Fig. 2) show ovoid, hyperintense lesions with low signal rims and faint, tiny central dots (dot-in-circle sign), particularly in the dorsal and medial aspects of the forefoot. Coronal (Fig. 3) and axial (Fig. 4) T1-weighted and T1-weighted fat-suppressed postcontrast images demonstrate enhancement of these lesions separated by internal, non-enhancing, low-intensity tissues. Histopathology confirms the presence of filamentous colonies of *Actinomyces* (Fig. 5).

Mycetoma, first described in the Madura district of India in 1842 [1–3], is a clinical entity referring to a chronic, debilitating, granulomatous disease of soft tissues, usually of the foot, resulting from infection either with filamentous bacteria (actinomycetoma) or true fungi (eumycetoma) [1, 2, 4–6]. It is endemic in equatorial, tropical, and subtropical regions, including the Middle East [1, 4, 5], but is not well known in the USA and Europe [5], except in migrant populations [7]. Data on its prevalence have been few and far between. A meta-analysis by van de Sande et al. in 2013 reviewed several single-center studies from various countries and estimated a prevalence between <0.01 to 3.49 cases per 100,000 inhabitants, with Mexico, Sudan, and India being the most commonly affected countries [3]. Patients are typically male [1, 3, 6] and under 30 years old [3, 6]. An update from the Mycetoma Research Center in Sudan released in 2015 predicted higher numbers in the future, with possibly more accurate estimates of its exact global burden as more research and surveillance programs are implemented [6].

The disease is usually slow and painless [5], with the organism first lodging in the soft tissues typically as an aftermath of skin trauma [1, 4–6], before multiplying and forming colonies that spread along fascial planes to the deeper soft tissue structures [1, 6]. It may be confined to the soft tissues for years [4–6], but has the potential for imminent osseous changes such as periosteal reaction and cortical erosion that almost always commences from the outside, in contrast to the centrifugal spread typical of bacterial osteomyelitis [1].

The clinical picture of mycetoma is uniform, regardless of the causative species [1]. The characteristic triad includes a painless subcutaneous mass, draining sinuses, and exuding "grains" representing either a conglomerate of fungal hyphae [1, 6, 7], or bacteria in microabscesses [8], although these findings may not be present until the advanced stages, when extensive bone and soft tissue damage has already occurred [4]. Moreover, constitutional symptoms and signs are rare and may occur only after secondary bacterial infection of open sinuses [1].

The available diagnostic tests for mycetoma are few and not without limitations; early laboratory diagnosis before the appearance of sinuses and grains is particularly challenging [8]. Isolation of the fungal grains may be performed either via a deep-seated biopsy or aspiration cytology [2], necessitating the use of special stains for thorough identification of the causative organism [2]. Microbiological culture provides definitive diagnosis, but may take several weeks to months [2, 6, 9], with the potential for inadvertent contamination, and

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additional, more extensive biopsies [8, 10]. Moreover, the causative organisms may be fastidious and difficult to demonstrate [2], leading to further delays in diagnosis and intervention. Imaging may therefore be crucial in its early detection.

Radiologic findings in mycetoma usually begin with a soft tissue mass in its early stages [1], which may eventually involve the bony cortex from the outside, producing scalloping and variable periosteal reaction [1]. Other radiographic features in the early stages include lytic lesions, sclerosis, and bone remodeling that may be difficult to differentiate from chronic osteomyelitis [5]. Computed tomography (CT) and magnetic resonance imaging (MRI) may be more helpful in early detection, even before the development of sinuses and/or extrusion of grains [4], and are also the ideal modalities for the assessment of bone destruction, soft tissue and periosteal involvement [5]. CT mirrors the radiographic findings [4], and is able to demonstrate additional abnormalities such as soft tissue infiltration of the plantar and dorsal surfaces of the foot, hyperdense subcutaneous fat and soft tissue nodules, and muscle thickening [4]. Its main advantage compared to MRI is its superior sensitivity in showing osteolysis, early osteoarticular damage and subtle cortical erosions [4]. Sarris and colleagues described the pathognomonic MR appearance of Madura foot lesions as hyperintense foci with hypointense rims, and small central dots representing fungal grains; these foci exhibit enhancement on postcontrast images, and are separated by internal, non-enhancing, low-intensity tissues [10]. This dot-in-circle sign is composed of granulation tissue surrounded by low signal intensity fibrous septa, with the "dot" caused by the susceptibility effect of the fungal grains [10]. More recent studies have also demonstrated a similar sign on ultrasound with hyper-reflective echoes corresponding to the grains (dot) surrounded by hypoechoic tissue (circle) [4]. My review of existing literature has not revealed any other condition associated with this sign, concurring with other authors [5, 8, 10] that this finding is highly specific for mycetoma foot.

Treatment can be challenging, and initially consists of a combination of antimicrobial agents [4, 5] with extensive surgical debridement and limb amputation being potentially employed in unresponsive or poorly controlled cases [4, 5, 7, 9]. The particular regimen is dictated by the causative organism, with eumycetoma reportedly requiring a longer period of treatment [1], exhibiting more resistance, a higher recurrence rate and overall poorer prognosis compared with actinomycetoma [1, 7].

Mycetoma should be part of the differential diagnosis in any case of a chronically swollen foot, particularly one with discharging sinuses. MRI may be important in the early recognition of the disease to prevent progressive bone destruction requiring surgery. The dot-in-circle sign that can be seen both in MRI and ultrasound reflects the unique pathologic features of the disease, is easy to recognize, is virtually pathognomonic of mycetoma, and may suggest the possible diagnosis without the need to wait for the histologic outcome.

Compliance with ethical standards

Grants received None.

Disclosures None.

Conflicts of interest None.

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