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CASE REPORT

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Eumycotic mycetoma involving the right foot: A new Tunisian case

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Abstract

A Tunisian patient with painless swelling and discharging sinuses of the right foot was diagnosed with Eumycetoma after 1 year of evolution. Mycetoma is a neglected disease relatively rare in our country. An early and accurate diagnosis, based on histopathological and microbiological examinations, limits functional and esthetical damage.

KEYWORDS

chronic infection, foot, mycetoma, mycotic

1 | INTRODUCTION

Mycetoma is a chronic infection affecting the skin and the subcutaneous soft tissues.[1]

It is caused by aerobic filamentous bacteria (actinomycetoma) or fungi (eumycetoma).[2] The disease commonly occurs in men aged between 20 and 40 years, especially those working in agricultural sector.[2,3] Mycetoma is characterized by a clinical presentation including swollen tissue in the affected area, sinuses formation, and grains identification from purulent discharge.[4] The progression of the disease may affect bone causing a large mutilating lesion and leading to foot amputation.[5] Mycetoma often occurs in tropical and subtropical regions, extending between 15° South and 30° North of the equator. The most affected countries are Mexico, Senegal, India, and Sudan.[2,3]

Environmental factors such as rain and nature of the soil play an important role in the distribution of this affection by influencing the prevalence of the causative agent.[6] Unfortunately, the diagnosis of this affection and the identification of the causative agent are challenging, especially in non-endemic areas.[2] In Tunisia, mycetoma is uncommon and only observed in sporadic cases.[7]

We are describing here a new Tunisian case of mycetoma and the particularities of this affection in Tunisia.

2 | CASE PRESENTATION

A 36-year-old woman with a rural background was referred to our hospital for a 1-year progressive painless swelling of the right foot, without history of foot injury. Physical examination found four subcutaneous lumps on the dorsal, medial, and plantar surface of his left foot, which were painless and soft in consistency (Figure 1). Skin ulcers with purulent discharge were noticed in the medial surface of the foot. The temperature was normal.

Routine blood tests were performed with the hemoglobin (Hgb), white blood cells (WBC) total count, and majority of chemistry within normal ranges. There was, however, elevated erythrocyte sedimentation rate (ESR) at 52 mm/h. X-ray of the left foot was performed and showed thickening of the soft parts opposite the lesions (Figure 2).

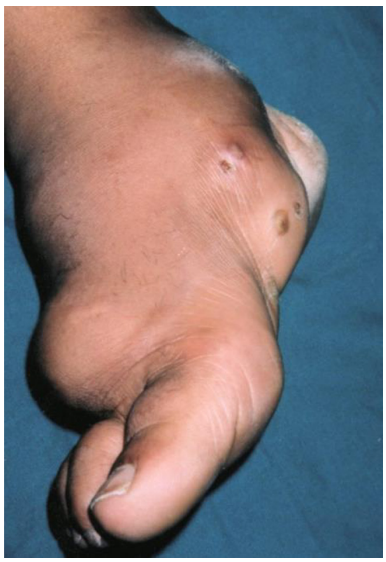

FIGURE 1 Madura foot with lumps and skin ulcers of the medial foot surface

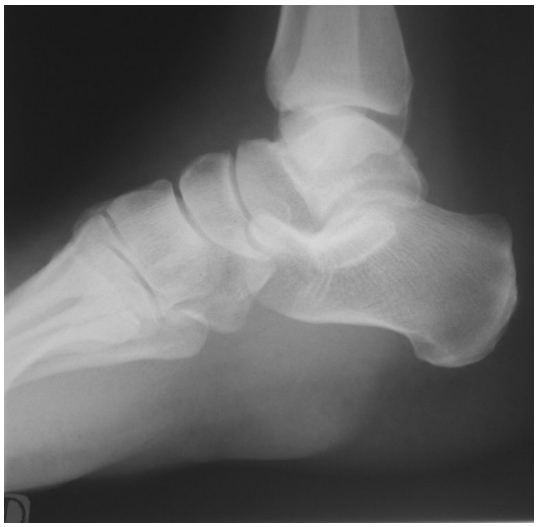

FIGURE 2 X-ray of the left foot showing thickening of the soft parts opposite the lesions.

A computed tomography (CT scan) was performed showing a mass with tissue density involving the plantar aponeurosis, which is moderately enhanced after injection of contrast agents. Besides, there was a second mass of the same density opposite to the second intermetatarsal space (Figure 3).

A complement by magnetic resonance imaging (MRI) revealed two nodular tissue masses of the dorsum and two other masses of the medial foot, of variable size, hypointense on T1-weighted and hyperintense on T2-weighted, which are heterogeneously enhanced after gadolinium (Figure 4). These masses respect bone, tendons, and muscles.

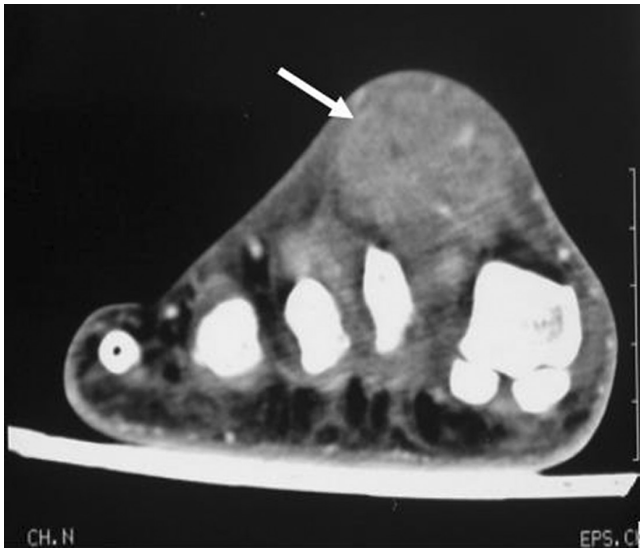

FIGURE 3 CT-scan coronal image of the rightfoot showing a mass with tissue density opposite to the second intermetatarsal space.

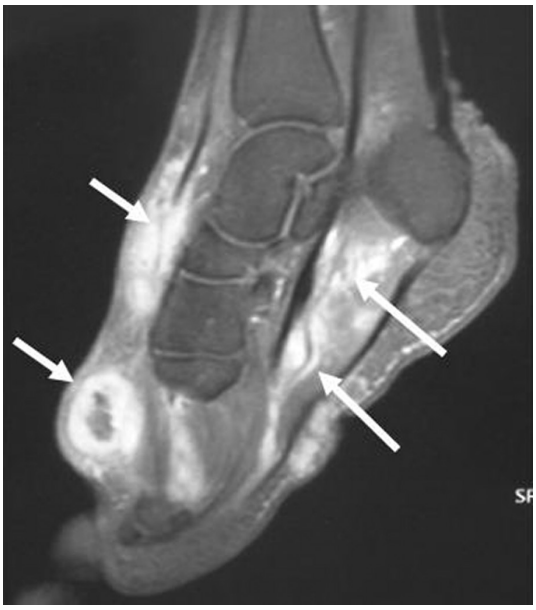

FIGURE 4 MRI T2-weighted sagittal plane image shows multiple nodular hyperintense masses of the dorsum and medial foot.

Bacterial and Mycological cultures were negative. A surgical biopsy of one of the lumps was performed, and the anatomopathological examination showed a richly vascularized fibro-inflammatory tissue with an infiltrate consisting of polynuclear neutrophils and eosinophils, as well as lymphocytes, plasmocytes, histiocytes, and foam cells, realizing micro-abscess within which granulomas are observed consisting with periodic acid Schiff (PAS)-positive spores and filaments. These histological findings

were consistent with eumycotic mycetoma. Treatment with cotrimoxazole was initiated, and additional surgery is planned for refractory lesions.

3 | DISCUSSION

Mycetoma is a unique and troubling neglected infectious disease that is endemic in tropical and subtropical areas of the world.[2,8] It was first described in Madurai in South India, by Gill in 1842 and was initially called "Madura foot".[7,9]

Mycetoma is a granulomatous inflammatory disease affecting the deep subcutaneous soft tissues and bones.[10] This is a severe condition that can develop over several years and has a profound and negative impact on medical and socioeconomic aspects of the lives of patients in the endemic areas.[8,10]

The global incidence of mycetoma varies from country to country and from region to region.[1] Diagnosis of this disease in such uncommon areas requires a high degree of suspicion.[6] Health professionals in these locations are not familiar with the disease; therefore, cases were usually misdiagnosed and mistreated leading to serious consequences for patients.[2] Differential diagnoses are malignant neoplasia, tuberculosis, or nocardiosis because mycetoma spreads progressively and continuously.[11] In fact, our case is from Tunisia, a country that is out of the "mycetoma belt," where no more than 100 cases have been reported.[12]

This condition affects more often men than women with the male to female ratio ranging between 3:1 and 5:1. However, the disease in Tunisia is relatively more frequent in women.[13,14]

Mycetoma can affect all age groups, but it commonly affects young adults aged 20–40 years.[1] The disease affects typically poor people living in rural areas and usually working in farms such as farmers and shepherds or those engaged in activities related to the environment.[1,10]

The suspicion of mycetoma is based clinically on the triad that includes progressive painless subcutaneous swelling, sinus tract formation, and granular discharge.[2]

Lower limbs are mostly affected in more than 80% of cases especially foot and leg, but it can affect any part of the body.[1,2] The foot involvement was predominant in the Tunisian cases reported in the literature.[12,13]

This infection may be caused by fungi and termed "eumycotic mycetoma," or by gram-positive aerobic filamentous bacteria termed "actinomycotic mycetoma" or "actinomycetoma."

Eumycetoma is more prevalent in humid regions and actinomycetoma in drier regions.[1] Common actinomycotic species include *Nocardia Brasiliensis*, *Actinomadura Madurae*, *Actinomadura Pelletieri*, and *Streptomyces Somaliensis*, and common eumycotic agents are *Madurella mycetomatis*, *Pseudoallescheria boydii*, *Madurella grisea*, and *Leptosphaeria senegalensis*.[3,15-17] The actinomycotic agents, particularly *A. madurae*, seem to be the most involved in Tunisian cases.[7,13]

The causative organisms trigger the formation of grains that may be discharged into the skin surface through multiple sinuses.[7,10] Some sinuses heal with scarring while new sinuses emerge elsewhere, leading to gruesome deformity of the affected limb.[16,18-21] Despite its several worrisome impacts, simple mentioning of mycetoma still leads astonishment. This is probably owing to poor awareness of its existence as a consequence of low identification and reporting as it is not a reportable disease.[3]

Diagnosis in early stage of the disease is the most challenging issue with Mycetoma. The issue becomes more challenging if it concerns the identification of the causative agent. Cultures are the gold standard in the diagnosis confirmation and the identification of wide species involved in Mycetoma.[2,16]

When the cultures are falsely negative, as in our case, the diagnosis of mycetoma is based on clinical and histopathological findings.[6]

The histopathological study is useful to confirm the clinical diagnosis and distinguish between eumycetoma and actinomycetoma, but it does not allow the identification of the causative agent. The histological sections show a granuloma containing grains situated in micro-abscesses.[1]

These grains may not be observed on histological sections, but when present, it is difficult to miss it due to its large size varying between 0.2 and 5 mm and surrounding neutrophils cluster. The distinction between actinomycetes and fungi is histologically possible.[7]

Actinomycetoma's colonies have different sizes, often round or multilobulate, with thin filaments; their thickness does not exceed1 μm. The actinomycotic colonies are typically gram-positive, but negative for PAS stain. Eumycetoma's colonies have many histological aspects that can have confusing appearance with actinomycotic colonies, but their filaments are thicker, varying between 2 and 6 μm. These colonies are positive for PAS stain and gram-negative.[2] The histological study of our case stained positive to PAS, allowing us to rule out actinomycetoma.

Molecular biology methods have become recently important in the identification of the causative agent by direct sequencing of biopsy specimens. However, the lack of accessibility and the high cost limit the use of molecular diagnosis to a complementary diagnostic tool and not a replacement for the conventional microbiology.[1,7]

The imaging technique such as X-rays, ultrasound, CT scan, and MRI allow to define the extension of mycetoma and the involvement of bone.[2]MRI can be useful also for the early diagnosis of dot-in-circle sign, high-intensity

lesion on T2 images with a tiny central low-signal focus reflecting fungal grains within inflammatory granuloma. This aspect is highly specific for mycetoma.[6,7]

The treatment of mycetoma depends on the causative agent (bacterial or fungal) and the extent of the lesion. It includes typically antimicrobial agents, sometimes combined with surgery.[1,2]

Eumycetoma infections require long-duration therapy with antifungal drugs belonging to the azole class such as itraconazole, voriconazole, or posaconazole. The most commonly used drug for the treatment of eumycetoma is itraconazole. Its prolonged use is associated with a favorable clinical response, especially if followed by surgical excision.[1,11] Eumycetoma treatment usually extends over many years.[6]

The treatment of actinomycetoma is based on systemic antibiotics. Combination of more than one drug is preferred to increase the response and prevent the development of resistance.[15] Actinomycetoma is commonly treated with an association of trimethoprim and sulfamethoxazole with aminosids (amikacin or netilmicin), for weeks.[2] Some existing reports indicate to prolong antibiotics until 5–6 months after complete healing of all sinuses.[3] Surgical excision is required for extensive lesions or abscess in order to reduce the necessity of prolonged anti-infective treatments.[11] Actinomycetoma prognosis seems to be better than eumycetoma.[2] Amputation is generally reserved to very advanced diseases that spread to deeper structures of the body.[2,7]

4 | CONCLUSION

We have reported in this article a new case of eumycetoma diagnosed in a Tunisian woman. Mycetoma is uncommon in Tunisia, and it is characterized by the relative female predominance, the frequency of foot involvement, and the predominance of actinomycosic origin. Our case illustrates the diagnostic challenge of mycetoma in non-endemic area. However, we were not able to specify the causative agent. Therefore, health practitioners should be aware of this condition and evoke it in front of any chronic painful suppuration of the foot, in order to avoid the delaying diagnosis and the functional and aesthetic prejudice.

AUTHOR CONTRIBUTIONS

Ben Tekaya A, Gzam Yosra, Saidane O, Rouached L, Bouden S: writing the article and collection of figures. Tekaya R, Mahmoud I, Abdelmoula L: final evaluation and correction.

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CONFLICT OF INTEREST

None.

DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no datasets were generated or analysed during the current study.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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