

## An Unusual Development of a Madura Foot: A Case Report

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### **Authors' contributions**

This work was carried out in collaboration between all authors. All authors read and approved the final manuscript.

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### **Case Study**

## **ABSTRACT**

Mycetomas are chronic inflammatory pseudo-tumoral lesions caused by fungal or bacterial agents developing in subcutaneous soft tissues and bone. Mycetoma is frequently located in the foot, realizing the Madura foot.

We report the case of a 42-year-old male farmer, who presented two years following an injury to the plant of the right foot. He already had two excisions elsewhere with recurrence and aggravation of the swelling.

He had a large painless subcutaneous swelling of his right foot with plantar sinuses discharging pale white and yellow grains. A surgical biopsy with histological study confirmed the diagnosis of mycetoma.

He received an antibiotic therapy consisting in penicillin G followed by amoxicillin then an association of penicillin G and co-trimoxazole during almost three years without any improvement. A below-knee amputation was performed complicated with necrosis and infection of the stump, which was resumed twice followed by an above-knee amputation.

Through this case of unusual development of a Madura foot we discuss the diagnostic and especially therapeutic difficulties of this chronic infection.

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## 1. INTRODUCTION

Mycetomas are chronic inflammatory pseudo-tumoral lesions of stereotyped slow development. They are often multi-fistulized containing grains. These may be caused by fungal agents (Eumycetoma) or filamentous bacteria (Actinomycetoma).

They develop in subcutaneous soft tissues and bone and can metastasize, which makes the severity of the condition. The localization in the foot is by far the most common found in 80% of cases, realizing the Madura foot. Mycetomas are frequent in agricultural areas in tropical countries but very rare in north Africa [1,2].

We report the case of an unusual development of a madura foot illustrating the diagnostic and especially therapeutic difficulties of this chronic infection.

## 2. CASE REPORT

We report the case of a 42-year-old north-african male farmer, without any significant medical history and no records of travelling to tropical areas, who presented to our outpatient department two years ago with a large painless subcutaneous swelling of his right foot. The symptomatology began a year before,

following an injury to the plant of the right foot and he already had two excisions elsewhere with recurrence and aggravation of the swelling.

The clinical study showed large painless subcutaneous swelling of his right foot with plantar sinuses discharging pale white and yellow grains (Fig. 1).

Plain radiography of the right foot showed thickening of the soft tissue without bone involvement (Fig. 2).

A surgical biopsy was performed with histological study showing actinomadura madurae grain with a type I tissue reaction consisting in grains surrounded by a layer of polymorphonuclear leukocytes, with neutrophils closely attached to the surface of the grain or invading the substance of the grain, confirming the diagnosis of a madura foot.

A below-knee amputation was proposed but the patient refused and continued his treatment elsewhere.

He received an antibiotic therapy consisting in 24 Mui of Penicillin G three times a week followed by six g of Amoxicillin every day during 12 months. No improvement was observed.



**Fig. 1. Clinical aspect of the right foot showing a large subcutaneous swelling with plantar discharging sinuses**



**Fig. 2. Plain radiography of the right foot showing thickening of the soft tissue without bone involvement**

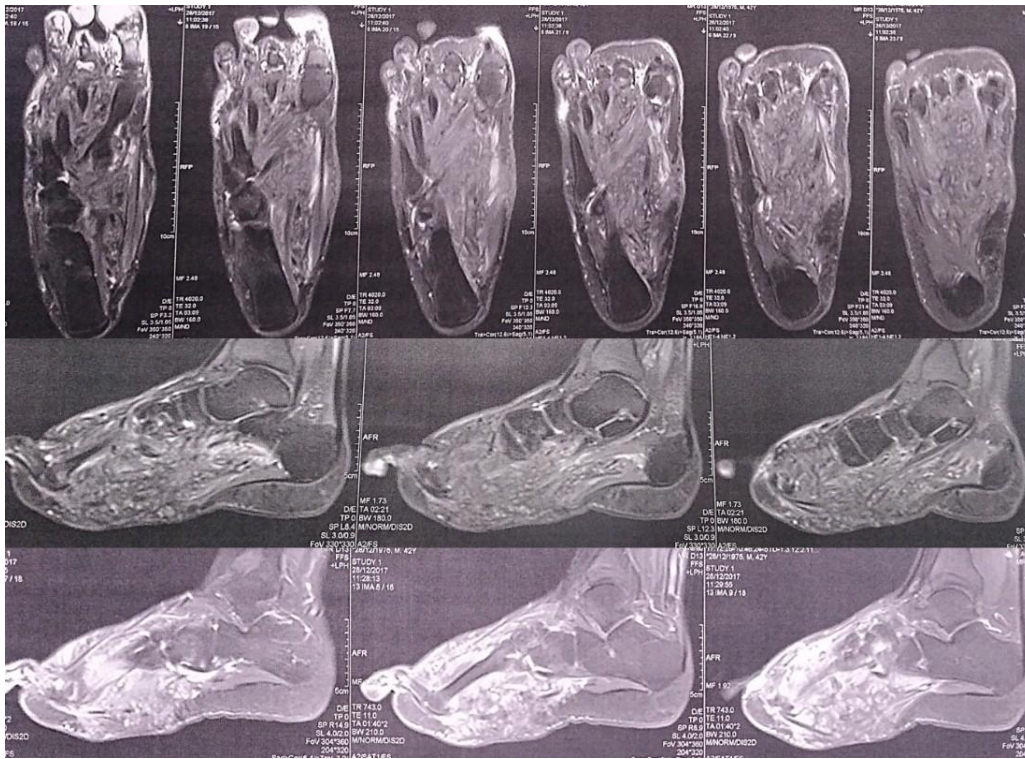
An MRI (Magnetic Resonance Imaging) was performed showing small lesions within mycetoma on the medial and plantar aspect of the foot due to grain contents, which are characteristic for mycetoma, with low signals characteristic ovoid shape in the T2-weighted image (Fig. 3).

The patient received another antibiotic therapy consisting in 24 Mui of Penicillin G in association with six g of co-trimoxazole per 24 h followed by six g of co-trimoxazole every day during six months.

The patient returned after almost two years of antibiotic therapy without any clinical improvement.

A below-the-knee amputation was performed complicated with necrosis and infection of the stump, which was resumed twice (Fig. 4), followed by an above-the-knee amputation (Fig. 5).

At the last follow-up of two months post-operatively the patient had a good local state without any signs of recurrence.



**Fig. 3. MRI of the right foot showing small lesions within mycetoma on the medial and plantar aspect of the foot due to grain contents, which are characteristic for mycetoma, with low signals characteristic ovoid shape in the T2-weighted image**

### 3. DISCUSSION

The Madura foot corresponds to a chronic subcutaneous inflammatory pseudo-tumoral lesion predominant in agricultural areas in tropical countries but very rare in north Africa [1,2]. It was first described in 1874 by Carter. It is characterized by the presence of fungal (*eumycetoma*) or bacterial (*actinomycetoma*) aggregates in the form of filaments or grains [1].

Its pathogenesis remains unexplained: the immune status of the patient seems to play an important part in explaining the discordance between the banality of infectious agents in the environment and the frequency of cutaneous injuries in the genesis of mycetoma. Contamination occurs through subcutaneous penetration of thorns, splinters or contaminated instruments that live saprophytically in soil and on plants [2,3].

Mycetoma is frequently located in the foot and hand; however, no body part is exempted and visceral involvement could be fatal [4].

Time to diagnosis is frequently delayed. Early clinical diagnosis of mycetoma is usually difficult but it is obvious at an advanced stage with the typical mycetoma triad of mass, multiple discharging sinuses, and grains [5]. The pseudo-tumor is emitting grains of different sizes and colors that invade the tissues step by step: the fasciae, the muscles and then the bone. Local sur-infection is common, leading to gangrene and amputation [6].

Grains are capital for diagnosing the causative organism. Although the morphological characteristics of the grains can provide a rapid provisional identification of the etiological agent. To avoid contamination, grains should be obtained by deep surgical biopsies under aseptic conditions. Grains obtained from open sinuses are usually not viable and often contaminated [7].

Fine needle aspirations cytology in three different directions under aseptic conditions is a fast and simple tool that could lead to the identification of the mycetoma's causative agent and the tissue reaction against it. If negative, a surgical biopsy should be performed [4].

The histopathological and histochemical techniques can provide accurate identification of the causative organism only if the grains are available in the tissue section. Grain culture in sterile conditions is very useful in case of bacterial mycetoma. Molecular based identification techniques are also effective but only available in well-equipped institutions [7,8].

CT-scan (Computed Tomography Scan) allows the early detection of bone lesions [9]. MRI (Magnetic Resonance Imaging) has made it possible to distinguish mycetoma from other similar lesions. However, the differentiation between actinomycetoma and eumycetoma infection is not possible [10].

The distinction between actinomycetoma and eumycetoma is of great therapeutic interest. The current recommended management of actinomycetoma is a combination therapy in 5-

week cycles. Patients received 3 weeks of amikacin sulphate intra-muscularly combined with 5 weeks of oral trimethoprim-sulfamethoxazole. For eumycetoma, currently, an itraconazole therapy associated with excision surgery is recommended [4].

The treatment of mycetoma is still disappointing and the need for surgery is often required. Leg amputation is frequently inevitable and represents the best option for acquiring complete healing. post-surgical rehabilitation care, providing the patient with a proper prosthesis and socio-economic integration program is necessary [1].

There are no clear criteria to affirm complete healing, the risk of post-operative recurrence is still present. Only prolonged surveillance over several years can affirm the cure. In this case, treatment should be continued for several months at a reduced dose [1].



**Fig. 4. Clinical image showing the twice resumed below-knee amputation**



**Fig. 5. Post-operative images of the right thigh after above-the-knee amputation**

The prevention is of great interest and should be educated to all farmers, it is based on the wearing of protective shoes, a thorough and systematic disinfection of wounds and the prevention of trauma especially with thorns [2].

#### **4. CONCLUSION**

Mycetoma of the foot is a rare and chronic infection. The painlessness, the slow development and the absence of spontaneous healing make the diagnosis particularly difficult.

The available treatments are not very effective and the prognosis remains reserved.

#### **CONSENT**

As per international standard or university standard, the patient's written consent has been collected and preserved by the author.

#### **ETHICAL APPROVAL**

As per international standard or university standard was written ethical approval has been collected and preserved by the author.

#### **COMPETING INTERESTS**

Authors have declared that no competing interests exist.

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