



Case Report

Severe and Late Diagnostic Form of Eumycetoma of the Foot with a Secondary Inguinal Location

Prise en charge d'une forme négligée sévère de mycétome du pied avec localisation inguinale secondaire

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Keywords : Eumycetoma, Madura foot, Secondary location, Amputation, Management

Mots-clés : Mycétome, Pied de Madura, Localisation secondaire, Amputation, Maladie tropicale

ABSTRACT

Eumycetoma is a chronic granulomatous inflammatory tropical disease, often localised and neglected. Endemic in the tropical and subtropical regions, its delayed diagnosis and lack of appropriate treatment can cause massive tissue destruction that can reach deep structures, deformities and disabilities. The authors report the case of chronic eumycetoma of the foot with severe bone lysis and unusual secondary localization to the inguinal region. In these complicated forms with irreversible bone damage, amputation appears as a definitive therapeutic alternative. Early management of this disease is essential to reduce morbidity.

RÉSUMÉ

Le mycétome est une maladie tropicale inflammatoire granulomateuse chronique, le plus souvent localisée et négligée. Endémique dans les régions tropicales et subtropicales, son retard diagnostique et l'absence de traitement adapté peuvent être à l'origine de destructions tissulaires pouvant atteindre les structures profondes, de déformations et de handicap. Les auteurs rapportent le cas d'un mycétome chronique du pied avec une lyse osseuse sévère et une localisation secondaire inhabituelle au pli inguinal. Dans ces formes compliquées avec atteinte osseuse irréversible, l'amputation apparaît comme une alternative thérapeutique définitive. La prise en charge précoce de cette maladie est essentielle pour réduire la morbidité.

INTRODUCTION

Mycetoma also called Madura foot is a neglected localised chronic granulomatous inflammatory tropical disease, often unique [1]. It is an endemic disease of the tropical and subtropical regions [2,3]. Eumycetoma is caused by a fungi infection with opposite to actinomycetoma caused by filamentous bacteria [4]. The causative organisms for eumycetoma are *Madurella* spp., *Leptosphaeria* spp., *Curvularia* spp., *Exophiala* spp., *Phaeoacremonium* spp., *Phialophora verrucosa*, *pyrenochaeta mackinnonii*, *P. romeroi*, *Pseudallescheria boydii*, *Acremonium* spp. and *Aspergillus* spp. Failure to early diagnosis and treatment leads to massive damage of deep structures and bones, deformity and disabilities [4]. The most frequent body parts affected are the limbs extremities particularly foot and hand [5]. It is characterised by painless subcutaneous mass, multiple sinuses that produce purulent discharges and grains [4]. The causative agent can sometimes be identified by the colour and size of various grains. Different laboratory techniques were developed over the years to determine and identify the causative agents; these

include direct microscopy, cytological, histological and immunohistochemical techniques in addition to the classical grain culture [1,4]. The natural course of the Eumycetoma is punctuated by complications but the diagnostic difficulty, inappropriate decisions and lack of consensus in its management would also be pejorative factors in the evolution of this disease

CASE REPORT

We report the case of a 30 years old male, farmer, residing in Douala, Cameroon. The past history was relevant for recurrent poor resections of multiple nodules and whitish grains discharging from sinuses for over 10 years. He presented at the National Rehabilitation centre for Person with Disabilities (NRCPD) with an asthenia, a painless left foot deformity and inability to walk. There was no symptom suggestive of a malignancy. On examination, he was afebrile but presented a deterioration of general condition with asthenia and pallor. On his left leg, we noted an ankle ankylosis in equinus with nodules and several whitish grains discharging from sinuses all over the foot up to the middle third of the left leg (Fig. 1).



Figure 1: Distal left leg in the neglected Madura foot.
 Figure 1: Lésions étendues de nodules ulcérés et laissant échapper des grains blancs évoquant un pied de Madura

The left foot was non tender. There was equally a localised left inguinal secondary location of several nodules and sinuses (Fig. 2).



Figure 2: secondary inguinal location
 Figure 2: Mycétome inguinal secondaire

Numerous scars of previous surgical resections could be seen at the left foot. A diagnosis of eumycetoma was made. Differential diagnosis retain were actinomycetoma and foreign body granuloma. The following investigations were done : X-ray of the left leg and foot revealed a massive soft tissue shadow, periosteal reaction and multiple bone cavities over the entire left distal tibia and foot (Fig. 3).



Fig 3: Severe bone destruction of the left foot on plain Xray
 Fig 3 : Ostéolyse sévère du pied à la radiographie standard

Full blood count was normal (with a white cell count of 4230/ul; Haemoglobin of 12 g/dl and platelet count of 224,000/ul), Erythrocyte sedimentation rate was normal and C-Reactive Protein slightly elevated at 6 mg/l ; Urea and creatininemia as well as coagulation profile was normal. The diagnosis retained was eumycetoma of the left distal leg and foot with a bony involvement and a secondary inguinal location. Surgical management, under general anaesthesia, consisted of complete resection of the secondary inguinal location and mid-leg amputation for this severe irreversible destruction of the foot (Fig. 4,5).



Figure 4: Surgical resection of secondary inguinal location of mycetoma
 Figure 4 : Exérèse large du mycétome inguinal



Figure 5: Mid leg amputation
 Figure 5: Moignon d'amputation trans-tibiale



The mass was sent after surgery for histopathology and culture which were not contributive.

The patient after surgery was given Ketoconazole 400mg/day and Terbinafine 500g/day and during one year associated with amphotericin B 1mg/kg/day the first month. The renal and hepatic markers were regularly controlled.

The evolution was marked by good wound healing (Fig. 6) and ambulation with crutches while awaiting leg prosthesis. The clinical exam and biologic check-up at the follow-up at 18 months were normal.



Figure 6: Evolution of wound healing at the inguinal region and the leg storm

Figure 6 : État des cicatrices opératoires à 3 mois post-opératoires

DISCUSSION

The particularities of this presentation are the severe bone lesions of the foot and the coexisting homolateral inguinal localization of mycetoma. During ten years, the patient lived with it, going from town to town. This errance could explain the severity of this evolution and the bone affect [6].

The mycetoma is a frequent fungal infection in with an average incidence was estimated at 0.32 per hundred thousand persons and a prevalence of 8.32 per hundred thousand persons per decade [1] ; the subtropical and tropical countries are the most affected [7]. The climate and the rural activities should be favorable conditions for the development of this pathogen [6].

The diagnosis is based on clinical presentation with histologic findings to confirm the diagnosis. The confirmatory diagnostic algorithm starts with a fine needle aspiration cytology (FNAC). If the FNAC is negative, we proceed by doing biopsy surgery before doing grain culture, histopathological culture and molecular techniques. If the FNAC is positive, we proceed directly by doing grain culture, histopathological culture and molecular techniques [2]. It is generally of late diagnosis in Africa because the histologic expertise is unavailable or not contributive [5, 8]. The grains produce here were white grains much suggestive of *A. madurea*. However black fungi may also produce grains of pale color. Eumycetoma frequently has lesions which grows slowly with clearly defined margins and remains encapsulated for a long time but Actinomycetoma has a rapid progression and is more inflammatory [5].

Eumycetoma treatment usually poses a challenge; antifungals with or without surgery can be use [7]. Treatment modalities include Itraconazole 200-400 mg/day, Ketoconazole 400 mg/day, Voriconazole 400-600 mg/day, Posaconazole (200 mg, four times daily), Terbinafine (500-1000 mg/day), and Amphotericin B (0.5-1.25 mg/kg/day) alone or with any combination [7]. The antifungal are rarely efficient if used alone and the

amputation could be done for the irreversible destruction [9].

In this case, in front of the unhelpful histopathology, we associated some antifungal medicine to treat the inguinal mycetoma and prevent a recurrence associated with the amputation of the leg.

At eighteen months term, there is no new lesion of mycetoma.

CONCLUSION

The authors describe their experience in the diagnosis and management of this rarely presentation of mycetoma which associated two different localizations and a very important destruction of bone tissue of the foot which indicated a radical amputation. Creation of registry, data bases and a holistic approach in the management of this disease in Africa are important starting points to improve the management and curb the morbidity and mortality of this infection.

DECLARATIONS

Conflict interest: The authors declare that they have no conflict interests in this section.

Fundings : none

Authors' contribution: All authors approved final manuscript and are responsible for it.

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