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Fungal and bacterial mycetoma in migrants from Haiti: A case series

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Dear Editor,

Mycetoma or "Madura foot" is a chronic granulomatous cutaneous and subcutaneous infection caused by bacteria or fungi. It is acknowledged as a Neglected Tropical Disease by the World Health Organization (WHO) [1]. Mycetoma is associated with an important functional impairment in case of treatment delay and/or misdiagnosis. More than 70 different species of fungi and bacteria have been incriminated as responsible pathogens agents [1,2]. Bacterial mycetoma, also called actinomycetoma, is often caused by *Actinomadura madurae*. Fungal mycetoma is also called eumycetoma and is often caused by *Madurella mycetomatis* [3]. The pathogens involved are found in the environment (soil, water and plants) and become pathogenic after skin injury. Clinically, mycetoma presents as a chronic tumoral swelling with little pain, multiple draining sinuses and fistulas and grains visible to the naked eye. The evolution is unfavourable with a risk of important local deformities, amputation or even death [4]. Mycetoma is endemic in several regions of Africa, Asia and South America, mainly in the "mycetoma belt" (between latitudes 15°S and 30°N).

Haiti is considered as non-endemic for mycetoma and is not mentioned in the WHO mycetoma chart. However, Bessis et al. [5] published the first case of actinomycetoma in a migrant recently arrived from Haiti (2020). This 38-year-old woman was infected with *Actinomadura mexicana*. However, it is unclear whether this case report was exceptional or whether the disease is actually present in the country. Besides, fungal cases have never been reported in Haiti.

Here, we describe one case of each type of mycetoma in Haitian patients.

Patient 1 was a 53-year-old woman. She had migrated from Haiti to French Guiana a few years prior to consultation. She used to work barefoot as a farmer. For several years prior to her arrival in French Guiana, she had presented an oedema of the left foot. She had no medical history except a mild hypertension. Multiple skin fistulisations and a discharge of grains progressively appeared. A foot deformity complicated with limping and disability was observed (Fig. 1a and b). She was seen in the Cayenne Hospital in 2020. An X-ray showed multiple lacunae in every bone of the left foot. CT scan showed a significant destruction of all tarsal and metatarsal bones and a significant infiltration of the adjacent soft tissues (Fig. 1c). C-reactive protein was high (116 mg/l) but there was no fever. The presence of macroscopic black grains suggested the diagnosis of fungal mycetoma, confirmed in histopathology (Fig. 1d). Methylene blue staining on skin biopsy showed mycelial filaments. Morphological identification was not possible, but mass spectrometry on fungal culture identified *Madurella mycetomatis*. The patient required a *trans*-tibial amputation due to the advanced nature of the lesions. There was no relapse at one year follow-up.

Patient 2 was a 35-year-old woman, 6 months pregnant. She consulted a few months after her arrival in French Guiana. She had suffered a skin injury on the arm when she lived in Haiti. She did not report any contact with soil or water. The injury evolved into a painless swelling more than 5cm wide, with multiple fistulae. The MRI showed an aspect of mycetoma with a subcutaneous infiltration of the forearm and T1-hypointense, t2-hyperintense cystic pockets. Histology on skin biopsy revealed clusters of actinomyces filamentous branching bacilli (Fig. 1e). However, the bacterial culture did not grow and species identification was not possible. She was treated with cotrimoxazole during 3 months which allowed an important decrease of the swelling and a closure of numerous fistulae. Then she was lost to follow-up.

We report here the first eumycetoma and the second actinomycetoma in migrants originating from Haiti. These data are in line with the conclusions of Bessis et al. as it appears that Haiti is endemic for both types of mycetoma. Patient 1 was probably contaminated while walking barefoot. While this information was not recorded for Patient 2, the same risk factor was probably present. In terms of latitude, Haiti is located on the "mycetoma belt". Another case of eumycetoma in the same island of Hispaniola has been reported in the Dominican Republic [6].

Dermatologists should be aware of the possibility of encountering this NTD in Haiti and probably in other insular countries in the Caribbean. Further studies are needed to evaluate the prevalence of bacterial and fungal mycetoma in Haiti and the subregion. This disease is probably under-diagnosed and under-reported by local health workers. The fact that several cases were reported in such a small period in migrants from Haiti implies that the actual number of mycetomas currently present in the country is probably far higher than expected.

Patient 1 is an example of the functional impairment that can be caused by a diagnosis delay leading to amputation. A targeted public health policy is necessary to detect and diagnose early cases of mycetoma and avoid such delays. Therefore, it is important to precisely

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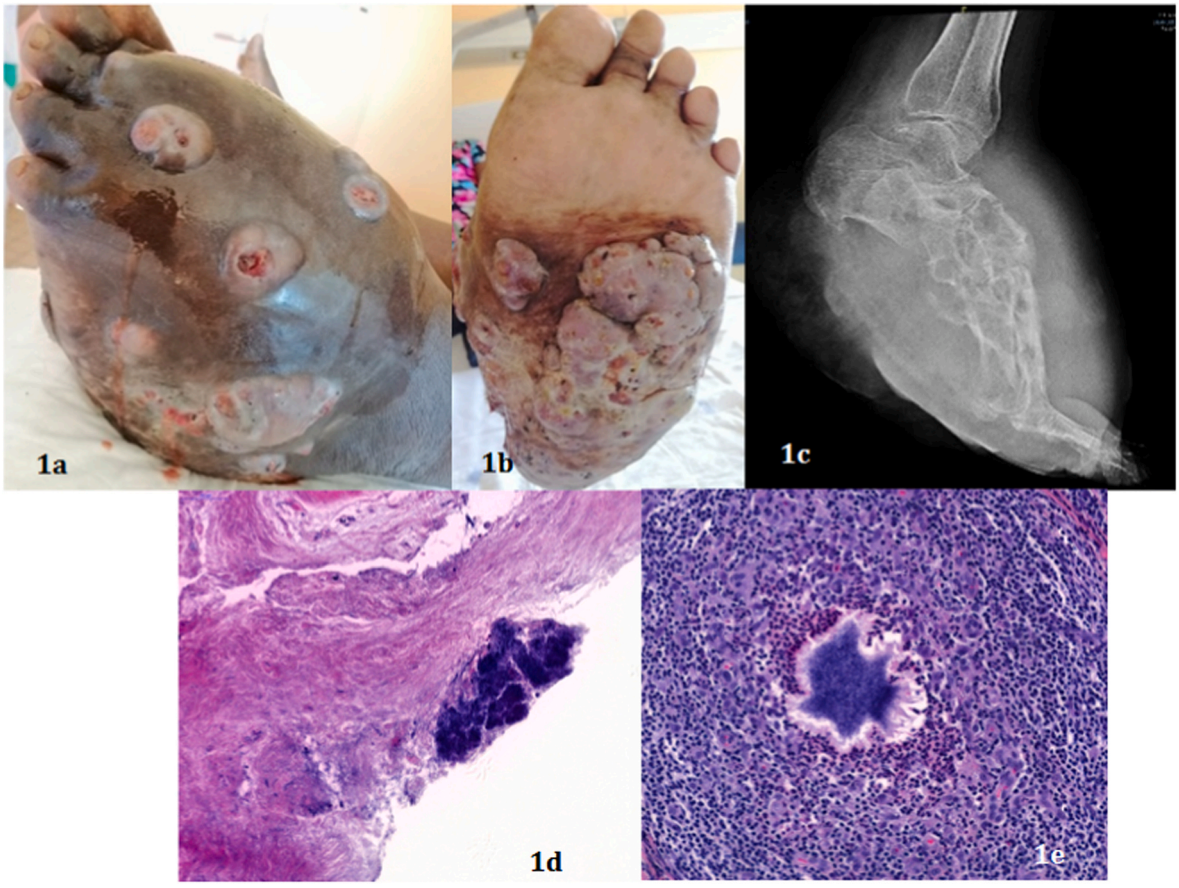


Fig. 1. Clinical, radiological and histopathological aspects of mycetoma. Eumycetoma with tumoral swelling and multiple draining sinuses and fistulas, excretion of black grains (A,B). CT scan of the left foot showing tarsal and metatarsal bone destruction (C); Histological examination of the left foot lesion showing basophilic clusters of very fine branching mycelia filaments in contact with necrotic bone tissue (hematoxylin and eosin stain, 60X) (D). Histological examination of the arm lesion shows a granulomatous inflammatory nodule in the dermis with a central basophilic cluster of actinomyces filamentous branching bacilli (hematoxylin and eosin stain, 60X) (E).

describe the endemicity of each type of mycetoma in the Caribbean and to start training of local health workers. The absence of precise data is a major barrier to the instauration of a roadmap for the control of this NTD. Though most mycetomas reported in South America are bacterial [3], fungal mycetomas can also be found and the identification of the involved pathogen should be performed, when possible.

Autochthonous cases of both kinds of mycetoma can be found in Haiti. The prevalence of this NTD is probably under-estimated and should be assessed by active detection throughout the country.

References

[1] Organisation Mondiale de la Santé O, World Health Organization. Results of the 2017 global WHO survey on mycetoma–Résultats de l'enquête mondiale de l'OMS surle mycétome, 2017. Week Epidemiol Rec= Relevé épidémiologique hebdomadaire 2018;93(33):423–8.
[2] Abbas M, Scolding PS, Yosif AA, EL Rahman RF, EL-Amin MO, Elbashir MK, et al. The disabling consequences of Mycetoma. Vinetz JM, éditeur. 10 déc PLoS Negl Trop Dis 2018;12(12). e0007019.
[3] Hao X, Cognetti M, Burch-Smith R, Mejia EO, Mirkin G. Mycetoma: development of diagnosis and treatment. J Fungi 2022;8(7):743. 19 juill.
[4] Agarwal P, Jagati A, Rathod SP, Kalra K, Patel S, Chaudhari M. Clinical features of mycetoma and the appropriate treatment options. Res Rep Trop Med 2021;12:173–9. juill.
[5] Bessis S, Noussair L, Rodriguez-Nava V, Jousset C, Duran C, Beresteanu A, et al. Actinomycetoma caused by *Actinomadura mexicana* , A neglected entity in the caribbean. Emerg Infect Dis 2020;26(2):379–80. févr.
[6] Gugnani HC, Denning DW. Burden of serious fungal infections in the Dominican Republic. J Infect Public Health 2016;9(1):7–12. janv.

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