

Case Report

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Eumycetoma: An unexpected diagnosis in Central Europe**Daniel Gödde^{1,3}; Holger Schubert²; Stephan Störkel^{1,3}**¹Department of Pathology and Molecular Pathology, Helios University Hospital Wuppertal, 42283 Wuppertal, Germany.²Centre for Orthopaedics and Rheumatology, Sankt Josef Hospital, 42105 Wuppertal, Germany.³Department of Medicine, Faculty of Health, Witten/Herdecke University, 58448 Witten, Germany.***Corresponding Author: Daniel Godde**

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Abstract

Mycetomas are slow growing pseudotumors, which develop after trauma with an infection caused by bacteria or fungi. This localized infection in the cutis and subcutis is identified as eumycetoma or “Madura foot” when caused by fungi. Without proper treatment, bulky and obstructive tumors develop. These tumors form fistulas from which pus oozes, sometimes granular. Eumycetomas may also lead to bone destruction, but usually do not show dissemination. In temperate zones, the diagnosis of eumycetoma is rare.

A female Korean patient, 62 years old, presented to the orthopedic department with a subcutaneous, slow-growing tumor on her ankle. After surgical removal of the tumor, an eumycetoma caused by *Fusarium solani* was diagnosed by histopathological and molecular methods. Two years after the diagnosis, no complications have occurred. Correct identification of the pathogen is essential for treatment. In addition to a culture of the pathogen, a molecular approach is possible. While patients with bacterial infections and actinomycotic mycetomas require systemic antibiotic treatment, the treatment of eumycetomas depends on the extent of the infection and the pathogen. In this case, systemic antifungal treatment, with or without additional surgery, is advisable.

Keywords: Deep Mycosis; Eumycetoma; Madura foot; *Fusarium solani*.

Introduction

Mycetomas of the leg, also known as “Madura’s foot”, is typically seen as slowly growing, painless inflammatory pseudotumors. They develop after trauma, with the infection of bacteria (actinomycetomas) or fungi (eumycetomas), and occur most often in adults between the ages of twenty and forty. In 60-80% of cases, they occur on the leg, but can also affect any other part of the body. Geographically, the disease is most common in areas between latitudes 30 degree north and 15 degree south in tropical or subtropical climate. Risk factors include walking barefoot and agricultural work [1-6].

In moderate climates, such as in Germany, these lesions are extremely rare. However, the disease should be considered in patients from tropical areas that exhibit slow growing subcutaneous masses with fistulas, or in patients who exhibit the same after visiting tropical areas, especially if there has been an injury. Due to the slow progression of the lesion, there can be years, even decades between infection of the bacteria and diagnosis [1-3]. Initially, clinical findings are non-specific and there is no systemic reaction. In progressive mycetomas, the lesions can ulcerate and necrotize, and fistulas can develop with secretion of pus and granules, important clues for diagnosis. Even though

mycetomas can lead to a destruction of adjacent bone, haematogenous dissemination usually does not occur [1,2].

Case presentation

A 62-year-old nurse of Korean descent living in Germany presented with a subcutaneous painless nodule, which had been present for about six months. The lesion was located between the lateral malleolus and the Achilles tendon of her left leg. The patient felt that the nodule was slowly growing and was now bothering her when wearing shoes. Her medical history was unremarkable, except for hyperthyroidism. Inspection of the patient's left leg revealed no abnormalities of skin, bone, or muscle. No discoloration or signs of inflammation were noted on the skin over the nodule. On palpation, the nodule remained as a firm, subcutaneous, well-demarcated lesion. It was not fused to the skin or skeletal structures. No bone involvement or destruction was noted on radiology (Figure 1). The lesion was resected and referred to pathology.



Figure 1: Radiography showing no bone destruction of the left foot.

Histology revealed confluent foci of inflammation in the subcutis with reticulo-histiocytic inflammatory infiltrate and abscess formation, necrosis, and calcification. PAS-stain showed multiple septate and branched hyphae consistent with deep mycosis. Based on the histologic findings, infection with domestic fungal species was considered unlikely, and the possibility of tropical fungal infection was considered (Figure 2). Histology did not reveal the fungal species; therefore, molecular tests were performed on the paraffin-embedded tissue. After DNA extraction, amplification, and sequencing of the ITS2-region of its rDNA, *Fusarium solani* was identified, leading to the diagnosis of eumycetoma.

After surgery, no further pathological findings were observed from examination and imaging. Since the lesion had been completely resected, no antifungal medication was given. Over the course of two years, the patient underwent regular clinical examinations with no evidence of disease recurrence.

Conclusions

Our diagnosis of mycetoma is unusual, since the patient had no history of traveling to either Korea or tropical areas in recent years. Most likely, she contracted *Fusarium solani* in Germany through an injury while working in her garden. No fistula was present at the time of diagnosis. The patient had visited her physician in the early stages of the disease, which is not always possible for patients in rural tropical areas.

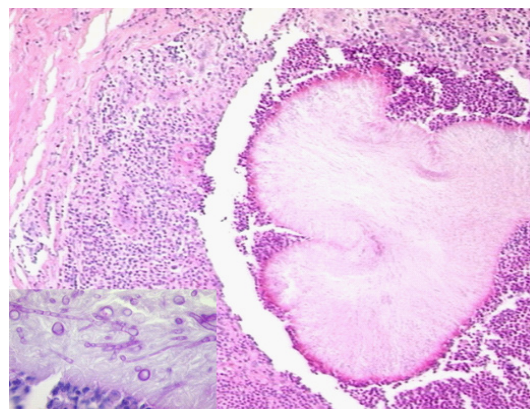


Figure 2: Fungal grain surrounded by reticulo-histiocytic inflammatory infiltrate with abscess formation and collagen capsule (HE, 100x). Inlay: Periodic acid-Schiff stain demonstrating fungal elements (PAS, 400x).

Fusarium species occur worldwide in nature and cause diseases like fungal keratitis, secondary infections of the skin, eumycetomas and in a setting of neutropenia, systemic mycosis. They can produce mycotoxins [7,8]. On histology, the hyphae of *Fusarium solani* resemble those of *aspergillus*. It is usually difficult to identify the fungal species by histology because morphological characteristics are nonspecific. Retrospective studies over ten years revealed a misinterpretation by histology in 21% of the cases [9]. A broad spectrum of fungal organisms can cause subcutaneous mycosis [10]. However, correct identification of the causing organism is necessary for adequate anti-mycotic treatment. Identification of the organism is possible through cultures as well as cytologic or histologic diagnosis with molecular studies, such as DNA-sequencing [11].

Unlike bacterial infections and actinomycotic mycetoma, which can be treated with systemic antibiotics, treatment of eumycetoma depends upon the progression of the disease and the infecting organism. Antibiotic or surgical treatment is possible, as well as a combination of both. In literature, treatment with trimethoprim-sulfamethoxazole is discussed for actinomycotic mycetoma, for eumycetomas, itraconazol and ketoconazol are treatment options, depending on the organism [6,13]. Small eumycetomas, like in our case, can be treated surgically (Welsh-method, [14]). When considering therapeutic options, superinfection or involvement of bone should be excluded.

Declarations

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