Painless swelling and recurrent subcutaneous abscesses of the lower leg - two distinct presentations of eumycetoma

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Background: Eumycetoma is a neglected tropical disease that is characterized by the triad of painless swelling, sinus formation and purulent discharge. This chronic disease causes significant disability if diagnosed late. We report two cases of eumycetoma, one early and one late presentation.

Material/methods: Diagnosis was established by culture in case 1 and by histopathology after surgical excision in case 2, followed by identification of Madurella mycetomatis as the causative organisms in both cases by panfungal polymerase chain reaction (PCR) targeting the internal transcribed spacer region.

Results: The first case was a 21-year-old Eritrean migrant who presented to our emergency department with progressive pain and swelling of his left ankle. He had emigrated from Eritrea about four months ago with stopovers in Sudan, Libya and Italy. He reported recurrent local infections on his left foot since the age of 10. On examination, multiple scars were noted on his left lower leg in addition to a subcutaneous abscess, which was drained. Abscess cultures grew a mold, which was identified as Madurella mycetomatis using panfungal PCR and sequence analysis (100 percent match with
Magnetic resonance imaging (MRI) demonstrated residual abscesses extending into the left ankle joint. Treatment with itraconazol was initiated which resulted in marked local improvement. Unfortunately, the patient was transferred back to Italy and was lost to follow up. The second case was a previously healthy 41-year-old Indian man who presented with a painless swelling of the left forefoot, which had increased considerably in size over three months. He was living in Switzerland since 1996, and his last visit to the Indian subcontinent (Pakistan) was five years ago. MRI imaging demonstrated an interdigital mass with marked capsular contrast enhancement (Figure 1A). The tumor was resected *in toto* and sent for histopathology only, which demonstrated a granulomatous necrotizing inflammatory reaction surrounding fungal hyphae (Figure 1B-E). Subsequently, panfungal PCR of the formalin-fixed paraffin-embedded tissue was positive for *Madurella mycetomatis*. Despite complete resection we opted for additional treatment with itraconazole for six months due to high relapse rates after surgical monotherapy. After a follow-up of 30 months there is still no evidence of relapse.

**Conclusions:** The present eumycetoma cases underscore the importance of considering epidemiological clues in the differential diagnosis of soft tissue swelling or recurrent abscesses on the lower leg. Physicians in countries currently hosting refugees from East and Central Africa need to consider eumycetoma in patients with soft tissue infections or painless soft tissue masses, in particular of the lower extremity. Panfungal PCR may aid in the identification of the causative agents of eumycetoma.

![Figure A](image1.png) ![Figure B](image2.png) ![Figure C](image3.png) ![Figure D](image4.png) ![Figure E](image5.png)