

Madura Foot: An Interesting Case of Mycetoma Reoccurring Fifteen Years after Surgical Excision

AUTHORS:

Richard Francesco^a, MS; Lauren M. DeStefano, MD^b; Guanjun Xia, MD, PhD^c; Gul Madison, MD^d; and Leon Clarke, MD^b

CORRESPONDENCE AUTHOR:

Dr. Lauren M. DeStefano
1500 Lansdowne Avenue
Darby, PA 19023
Email: LaurenDeStefanoMD@gmail.com
Phone: 973.865.7547

AUTHOR AFFILIATIONS:

a. Philadelphia College of Osteopathic Medicine. Philadelphia, PA
b. Department of Surgery. Mercy Catholic Medical Center. Darby, PA
c. Department of Pathology. Hahnemann University Hospital. Philadelphia, PA
d. Department of Infectious Disease. Mercy Catholic Medical Center. Darby, PA

Background	<i>Madura foot</i> , or mycetoma, is a chronic subcutaneous granulomatous soft-tissue infection. The etiological agent may be fungal (eumycetoma) or bacterial (actinomycetoma), and appears under microscopic examination as hyphae or thin filaments, respectively. The cardinal features of the disease are tumefaction, sinus formation, and presentation of grains or granules in draining exudate. Cases are prevalent in equatorial, tropic, and sub-tropic climates, but are not exclusive to these regions. Early treatment via surgical excision in combination with pharmacotherapy yields the best outcomes; however, reoccurrence is common and long-term patient observation is recommended.
Summary	A 50-year-old North African man from Mauritania, with a self-reported history of right foot surgical debridement of unknown pathology fifteen years prior, presented with a several month history of pain and swelling of the right foot. Physical exam revealed an erythematous, tender, palpable mass measuring 3.5 x 1 x 0.5 cm on the plantar aspect of the right foot. The patient underwent elective excision of the soft-tissue mass. Histopathological investigation of the excised mass revealed suppurative granulomas (consisting of neutrophils) with surrounding black grains in the subcutaneous tissue. The black granules showed thin, filamentous bacteria suggestive of actinomycetoma. Ideally, the tissue samples would have been Gram stained and cultured, but the tissue was set in formalin-fixed, paraffin-embedded blocks, thereby rendering additional tests impossible. Since a definitive diagnosis between actinomycetoma (bacterial) and eumycetoma (fungal) could not be obtained, the patient was placed on fluconazole and amoxicillin for four months. The patient's surgical wounds healed uneventfully, and the infectious disease department is currently following him over the course of his pharmacotherapy.
Conclusion	A high index of suspicion for Madura foot should be considered in patients endemic to India, Mexico, and Africa who present with subcutaneous masses of the foot and ankle. Treatment is best accomplished with combined surgical excision and prolonged antimicrobial therapy. Early diagnosis and a thorough workup, including both bacterial and fungal culture, are paramount to prevent further dissemination of subcutaneous granulomas, the formation of open lesions, secondary bacterial infection, and fatal septicemia.
Keywords	Madura foot, Mycetoma, Eumycetoma, Actinomycetoma

DISCLOSURE:

The authors have no conflicts of interest to disclose.

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Case Description

Madura foot, or mycetoma, is a chronic granulomatous infection characterized by subcutaneous masses, draining sinuses, and the presence of grains or granules in exudates. This condition is caused by either fungi (eumycetoma) or aerobic filamentous bacteria (actinomycetoma). The symptoms of Madura foot generally present several weeks to months, and at times even years, after inoculation.¹ Once diagnosed, the treatment modality is based on fungal or bacterial etiology.² Surgical excision of the mass is necessary in conjunction with pharmacotherapy in order to prevent further tumefaction; nonetheless even with proper treatment, reoccurrences of infection may still arise.³ Delayed diagnosis of this inflammatory disease may result in serious, devastating complications involving bones and ligaments, thereby leading to further complications. Bone involvement developing into osteomyelitis progresses to gross deformity and may necessitate amputation.³ As a local, chronic progressive infection, it is imperative that the treatment of Madura foot be started promptly to best prevent systemic spread or further bone deformation.

Madura foot may present on any area of the body; however, 70 percent of all cases involve the foot, with the primary site of affliction being the forefoot.⁴ It is endemic to equatorial, tropical, and subtropical regions within an expanse known as the “mycetoma belt,” where the regions of heat and humidity provide a suitable climate for the pathogens responsible to thrive.⁵ Although not endemic to the West, case presentations of Madura Foot are likely to continue to rise in accordance with immigration and continued international travel.⁵ In this case, we report a potential reoccurrence of Madura foot in an adult male of North African descent after fifteen years of the original onset.

A 50-year-old North African man from Mauritania with a self-reported history of right foot surgical debridement of unknown pathology fifteen years prior presented with a several-month history of pain and swelling of the right foot. Physical exam revealed an erythematous, tender, palpable mass on the plantar aspect of the right foot, measuring 3.5 x 1 x 0.5 cm; the patient underwent elective excision of this soft-tissue mass. Histopathological investigation (Figure 1, Figure 2, and Figure 3) of the excised mass revealed suppurative granulomas (consisting of neutrophils) with surrounding black grains in the subcutaneous tissue. The

black granules showed thin, filamentous bacteria suggestive of actinomycetoma. Ideally, the tissue samples would have been Gram stained and cultured, but the tissue was set in formalin-fixed paraffin-embedded blocks, thereby rendering additional tests impossible. Since a definitive diagnosis between actinomycetoma (bacterial) and eumycetoma (fungal) could not be obtained, the patient was placed on fluconazole and amoxicillin for four months. The patient’s surgical wounds healed uneventfully, and the infectious disease department is currently following him over the course of his pharmacotherapy.

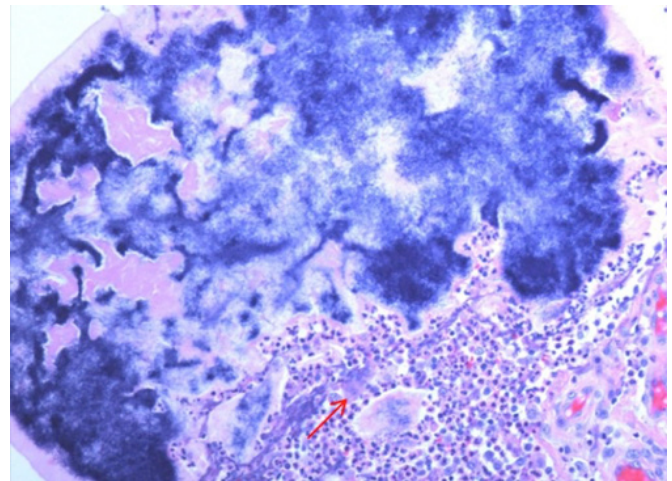


Figure 1. Thin filamentous bacteria

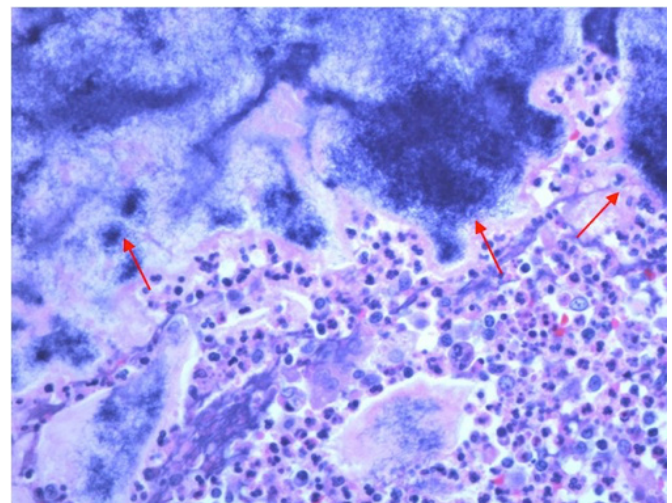


Figure 2. Giant cells

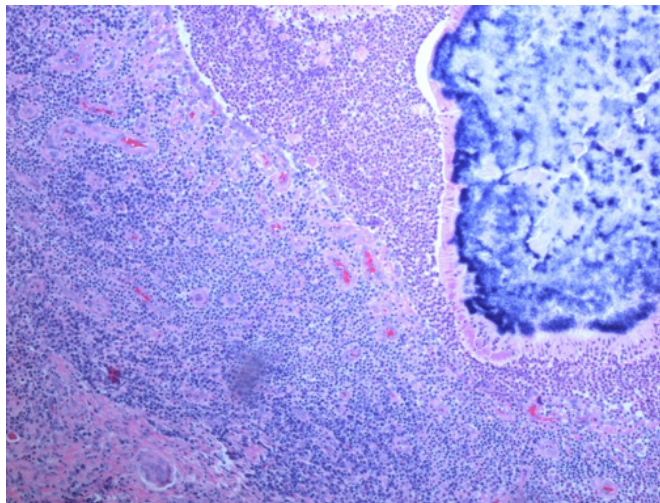


Figure 3. Granulation Tissue

Discussion

Mycetoma was first described in India in 1842, and this condition was designated Madura foot, named after the regional district of its discovery, Madurai.^{6,7} Madura foot is described as having three cardinal features: inflammatory subcutaneous masses, draining sinuses, and the presence of grains or granules within draining exudate. Acting as the etiological agent, grains spread throughout the original site of infection by newly formed sinus tracts that penetrate bone, muscle, and fascia.^{1,8} As tumefaction progresses, physical distortion of normal anatomy, impaired mobility, and pain ensue.^{5,8}

Both gross and histological analysis of grains must be employed to quickly identify the source of infection as being either bacterial or fungal.^{6,9,10} Although used in the past, the culturing of samples is generally futile due to the slow growth cycle of mycetoma and their high rate of contamination.¹ Respective of the etiological agent, grains within exudate represent collections of bacterial filaments or fungal hyphae; while the grains of actinomycetoma are generally lighter in color, eumycetoma may present with either a dark or light pigmentation.⁶ However, overlapping morphological characteristics often occur in both bacterial and fungal grains; therefore, Gram staining is invaluable to the confirmation of the causative agent in order to allow for proper treatment.⁹ Due to our patient's tissue sample being placed in formalin-fixed paraffin-embedded blocks, we were unable to Gram stain the tissue to confirm whether the causative agent was fungal or bacterial in origin, thus

the patient was treated broadly with both antifungal and antibiotic pharmacotherapies.

The most successful treatment outcomes for mycetoma are those begun early and consist of both surgical excision of the mass and prolonged pharmacotherapy.^{1,3} Surgical debridement alone has up to a 90 percent rate of recurrence, making antimicrobial therapy a necessity once tumor volume has been reduced.⁷ Due to its pain-free progression, and an often unnoticed time of infection, Madura foot is generally at an advanced stage upon presentation and diagnosis.⁶ Subcutaneous masses may range in size from 1 cm to 20 cm or more in diameter and present as an open wound, thereby increasing the patient's susceptibility to secondary bacterial infection.¹¹ From these open lesions, fatal septicemia may follow.^{3,9} Although used as a last resort, if clinicians are unable to control infection early on with the compromise of fascial planes, amputation may be necessary to halt the spread of infection and thus prevent further dissemination of muscle tissue.⁷

Conclusion

Endemic to equatorial, tropical, and sub-tropical regions, Madura foot, or mycetoma, is an uncommon chronic granulomatous soft-tissue infection. Although a rare disease in the Western Hemisphere, clinical prevalence is increasing due to the nature of immigration and international travel. Its etiological agent may be either fungal (eumycetoma) or bacterial (actinomycetoma) in nature, and the condition presents microscopically with hyphae or thin branching filaments. The three cardinal signs of infection are tumefaction, draining sinuses, and colonial grains in the exudate. Often infected by foot via soil saprophyte, mycetoma is a progressive infection that may remain asymptomatic for months to years after inoculation. Dissemination to bone, fascia, and muscle occurs via invading sinus tracts, causing anatomical deformity, pain, and immobility. Combination treatment modalities of surgical debridement and prolonged pharmaceutical therapy provide the best patient outcomes. The specimen should be sent to pathology, in addition to undergoing Gram staining, bacterial culture, and fungal culture to tailor treatment. Therapy must be initiated quickly to ensure complete tumor resolution and to prevent the formation of open lesions, secondary bacterial infections, and fatal septicemia.

Lessons Learned

A high index of suspicion for mycetoma should be considered in patients endemic to the tropics who present with masses on the foot and/or ankle. Early diagnosis and thorough workup are paramount in preventing further dissemination of subcutaneous granulomas, the formation of open lesions, secondary bacterial infection, and fatal septicemia.

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