Journal of Interdisciplinary Histopathology
www.scopmed.org
DOI: 10.5455/jihp.20141118013119

Case Report

Cytodiagnosis of primary actinomycotic mycetoma of the foot

Sneha S Desale, Dhiraj B Nikumbh, S N Kanthikar, Akshay Surana

Department of Pathology, ACPM Medical College, Dhule, Maharashtra, India

Address for correspondence:
Dhiraj B Nikumbh, Department of Pathology, ACPM Medical College, Dhule, Maharashtra, India.
E-mail: drdhirajnikumbh@rediffmail.com

Received: November 02, 2014
Accepted: November 18, 2014
Published: December 05, 2014

ABSTRACT

Mycetoma is chronic suppurative granulomatous disorder of subcutaneous tissue characterized by localized swelling with multiple discharging sinus tracts of granules that are micro colonies of the causative agent. Madura foot/mycetomas are the infection caused by true fungi (eumycetoma) in 40% or filamentous bacteria (actinomycetoma) in 60% of the cases. Painless subcutaneous mass, multiple sinuses and purulent or seropurulent discharge that may contain grains is characteristic of mycetoma. Mycetomas are usually diagnosed on histopathology. There are very limited data on role of fine-needle aspiration cytology (FNAC) in diagnosing these lesions. The distinction between eumycetoma and actinomycetoma in FNAC is as accurate as histopathology. Herein, we report a case of primary actinomycotic mycetoma of right foot in 65-year-old male patient. FNAC of soft tissue mass was advised, which revealed mixed inflammatory infiltrate with clumps of fibrillar organisms and occasional foreign body giant cells. Cytodiagnosis of actinomycetoma was rendered and Gram-stain was done later demonstrated Gram-positive thin branching filaments. Mycetoma can be accurately diagnosed by FNAC in outpatient department, which is simple, inexpensive, routine procedure for rapid diagnosis, and differentiation of etiology as the treatment differs.

KEY WORDS: Actinomycetoma, madura foot, mycetoma

INTRODUCTION

Mycetoma/madura foot is a chronic suppurative granulomatous disease characterized by localized subcutaneous swelling with multiple sinus tracts discharging granules. The infection caused by true fungi (eumycetoma) in 40% or filamentous bacteria (actinomycetoma) in 60% of the cases [1]. Actinomycetoma may be due to *Actinomadura madurae*, *Actinomadura peliietieri*, *Streptomyces somaliensis*, *Nocardia* species [1].

John Gill in 1842 described its clinical features for the first time and named it "madura foot" after the region of Madurai in India, where it was first identified [2]. Mycetoma occurs most often in people who work in rural areas, usually in farmers, field workers where they are exposed to cactus thorns or acacia trees that contain etiologic agents that normally live as saprophytes [1]. The organism enters through local trauma in the foot, hand or eyes from saprophytic soil. After entry into the body they form subcutaneous nodules, containing suppurative granulomas, multiple cavities, and sinus tracts. The sinus tract discharges exudates with fine grains. These grains are colonies of causal organisms [3,4] Usually, the diagnosis is made at an advanced stage due to ignorance and low socioeconomic status [5].

In India, actinomycotic mycetoma is more commonly encountered than eumycotic mycetoma. In Northern India, eumycotic mycetoma may be more common [5]. Surgical debridement followed by prolonged antibiotic therapy for several months is required for actinomycetoma. Eumycetoma are only partially responsive to antifungal therapy but can be treated by surgery due to their well-circumscribed nature [5].

Herein, we report a case of primary actinomycotic mycetoma of the right foot in a 65-year-old male patient who presented with 5 cm × 4 cm soft tissue mass with discharging sinus since 4 years. The diagnosis was made on fine-needle aspiration cytology (FNAC) and presented here in view of limited data in the literature on role of FNAC in diagnosing such lesions.

CASE REPORT

A 65-year-old male farmer patient presented to the Surgical Outpatient Department of our hospital with swelling and multiple nodular lesions over right foot since 4 years duration. The lesion initially was single painless nodule over dorso-lateral aspect of right foot and within a period of 4 years progress to form multiple small nodules with seropurulent discharge. There was history of trauma to foot in the past. There was no history of diabetes, hypertension or tuberculosis to the patient and in the family.

Physical examination revealed multiple nodules with discharging sinuses over dorsal and lateral aspect of right foot measuring 6 cm × 5 cm [Figure 1]. No difficulty in walking was noted. Hematological, biochemical, serological, and urine analysis were within normal limits. X-ray of the right foot showed soft tissue lesion with no underlying bony involvements [Figure 2]. Clinical differential diagnosis was made as soft tissue tumor or madura foot and FNAC of the lesion was advised. FNAC of right foot – dorsolateral aspect was performed under all aseptic precautions revealed granular whitish pus like material. Microscopic picture of FNAC showed predominantly neutrophilic exudates with lymphocyte and histiocytes along with many multinucleated foreign body type giant cells. Many colonies of "brownish black" filamentous bacilli suggestive of actinomycosis was noted in between inflammatory infiltrate and many multinucleated giant cells are noted [Figure 3]. The Pap stain demonstrate colonies of filamentous bacilli with some projections [Figure 4a and b], but characteristic "Splendore-Hoeppli" phenomenon was not seen. Later on Gram-stain was on unstained slides found positive Gram-positive bacilli and acid fast stain turned out to be negative. Hence, final diagnosis of primary actinomycotic mycetoma of right foot was rendered in view of no such lesions in other sites. Culture and histological study was advised to patient.

Figure 1: Gross appearance of swelling over the right foot with multiple nodules with discharging sinuses

Figure 2: Radiograph of the right foot showed soft tissue lesion with no underlying bony involvement

DISCUSSION

Actinomycetomas are caused by aerobic actinomycets belonging to the genera *Nocardia*, *Streptomyces* and *Actinomadura*. Although reported from all over world, they are common in tropical and subtropical regions where people walk barefoot [6]. The most common resulting skin lesions are on the lower extremities. Cutaneous manifestations include mycetoma, lymphocutaneous infections, superficial skin infestation/disseminated infection with cutaneous involvement [6].

"Dot in circle sign:" very rarely described in literature, is a unique appearance that is highly suggestive of mycetoma [7]. This sign is seen on MRI as hypo intense (fibrous tissue) shadow at periphery with grains in the center with hyper intense shadow of granulation tissue at lower parts of grains. The main differential diagnoses are chronic bacterial osteomyelitis, tuberculosis or early phase of buruli ulcer. Other rare possibilities as blastomycosis, coccidiomycosis and leishmaniasis, syphilis should be considered [5]. However, the main differentiation between actinomycetoma and eumycetoma is always warranted because of the different response to treatment.

The distinction between eumycetoma and actinomycetoma in FNAC is as accurate as histopathology [8]. However, the data are limited on the role of FNAC in diagnosing these lesions [9]. The cytological features were similar to histopatholoical features and differentiated eumycetoma and actinomycosis easily on FNAC [9]. On cytology, in actinomycetoma showed inflammatory exudates along with large colonies of filamentous bacilli (Gram-positive) with foreign body giant cells, as in our case. In the FNAC of eumycotic mycetoma, presence of septate, branching hyphae (fungal) with black granules against inflammatory background was evident [10].

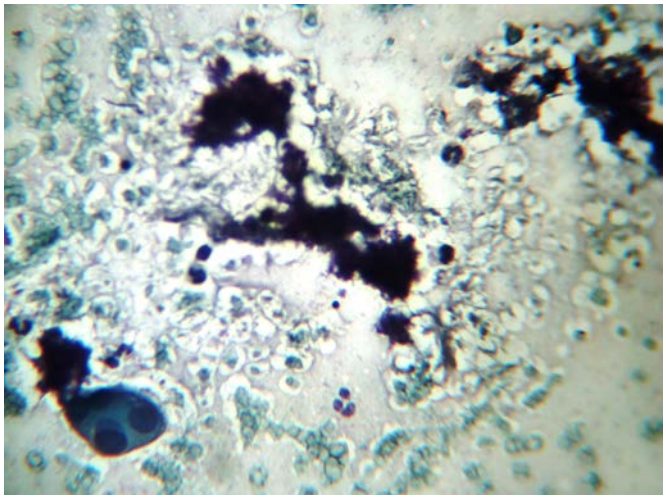

Figure 3: Cytology showed infl ammatory exudates and giant cells with many colonies of "brownish black" filamentous bacilli (PAP stain, ×100)

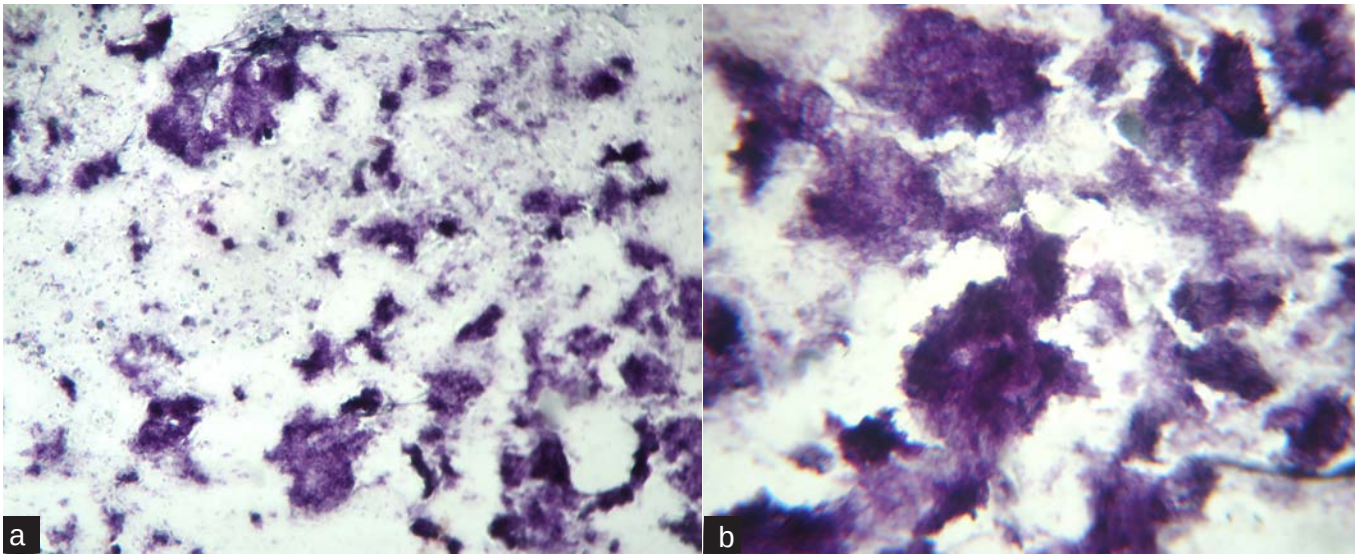

Figure 4: (a and b) Microphotograph showing colonies of filamentous bacilli with some projections (PAP stain, a: x200; b: ×400)

Mycetoma (often regarded to as "madura foot") is a Greek term for fungal tumor. It is granulomatous infection of dermal and subcutaneous tissue that may extend to muscle or even bone [10]. The literature mainly discussed the histopathological characteristics of mycetoma and very few studies have described their cytological features [10].

In our case, the diagnosis was made on FNAC and supported by Gram-stain. Unfortunately, the histological study and culture was not done in view of lost the follow-up of the patient.

Regarding treatment modalities, combined antibiotic therapy is preferable to monotherapy to avoid drug resistance and to eradicate residual infection [7]. Surgical debridement, followed by prolonged appropriate antibiotic therapy for several months is required for actinomycetoma, wherein combination therapy with sulpfa drugs, dapsone, and streptomycin has been used along with rifampicin for resistant cases [5]. Eumycetomas are partially responsive to antifungal therapy and can be treated by surgery due to their well-circumscribed nature [5].

CONCLUSION

Mycetoma can be accurately diagnosed by FNAC. It allows morphological identification of mycetoma and its classification into actinomycetoma and eumycetoma. We emphasized the diagnosis of actinomycetoma by FNAC in our article. Although it is a rare disease, it might be encountered in our regular practicing life especially in country where more than 75% of people are working barefooted into fields like in India. Hence, FNAC is simple, inexpensive, outpatient department, rapid procedure for morphological identification of mycetoma when there is a high index of suspicion.

REFERENCES

1. Mohammad N, Chaudhary A, Ruksara P, Rokon U, Abdul R, Moydul H. The madura foot- a case report. N Dermatol Online 2011:2;70-3.
2. Hugade S, Swoorooparani MS. Actinomycets mycetoma. J Lab Physicians 2011;3:126.
3. Mahgurb E, Murray IG. Mycetoma. London: Heinmann; 1973. p. 1-97.
4. McGinnis MR, Fader RC. Mycetoma: a contemporary concept. Infect Dis Clin North Am 1988;2:939-54.
5. Balkrishnan M, Nalliah R, Sridevi V, Vijaylakshmi CS. Primary actinomycotic mycetoma of the hand and wrist- A case report. J Med Sci Clin Res 2014;21:2266-9.
6. Miller SD. Madura foot: treatment of *Nocardia nova* infection with antibiotics alone. Am J Orthop (Belle Mead NJ) 2001;30:495-8.
7. Sharath Kumar BC, Shridevi CM, Gopal MG, Ramesh M, Gopi A, Shukla. A case report of actinomycotic mycetoma with osteomylitis of right foot treated with Welsh regimen. J Evol Med Dent Sci 2013;2:7968-71.
8. Fernandes H, D'souza CR, Shekar JC, Marla NJ, Swethadri GK, Naik R. Cytodiagnosis of actinomycetoma. Diagn Cytopathol 2009;37:506-8.
9. Hemalata M, Prasad S, Venkatesh K, Niveditha SR, Kumar SA. Cytological diagnosis of actinomycosis and eumycetoma: a report of two cases. Diagn Cytopathol 2010;38:918-20.
10. Gabhane SK, Gangane N, Anshu. Cytodiagnosis of eumycotic mycetoma: a case report. Acta Cytol 2008;52:354-6.

Source of Support: Nil, Conflict of Interest: None declared.