MYCETOMA IN AN UNUSUAL SITE

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Summary

A case of actinomycotic mycetoma is reported from Madras, India. Clinical and laboratory studies established the diagnosis. Gross microscopic features of the lesion and causative organism are described. The classification and geographic distribution of Nocardia species are discussed.

Mycetomas are characterized by localized swellings with multiple sinuses discharging granules or grains. The granules are composed of mycelial filaments. The dimensions of the filaments in the granule, the colour, shape, size, texture, internal morphology and staining properties of the granules are important in the identification of the causative agents.

Mycetomas are broadly classified into :

1. Actinomycotic and
2. Eumycotic categories.

The granules are composed of branching filamentous elements that are 1 μ or less in diameter in the actinomycotic mycetoma. In the eumycotic mycetomas, the granules contain broad, septate hyphae, 2 μ to 4 μ or more in diameter and chlamydospores are found at the periphery of the granule. The gross and microscopic appearance of the granules gives insight into the identity of the organism.

Although the earliest reports of mycetoma were made from India, only limited etiologic studies were undertaken here until recently. Mycetoma caused by *Nocardia* species, thought to be rare in Asia[1] was reported from South India[2], Andhra Pradesh[3] and Madhya Pradesh[4] based on histopathological evidence alone, where species differentiation was not possible. Since reports on isolation of *N. asteroides* were not many[5,6,7], it seemed worthwhile to present this case where *N. asteroides* was isolated in pure culture from mycetoma of an unusual site.

Case Report

A 30-year-old farmer injured his right thigh while cycling. Minimal local swelling developed at the site of injury. Within the course of six months, multiple dermal nodules developed in the region of the right thigh. The nodules later developed sinus tracts that discharged a sero sanguinous, occasionally purulent material. The lesion, 25 cm x 15 cm occurred over the upper/1/3rd of the anterior and lateral

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surface of the right thigh. (Fig. 1 page No. 343). Surface was nodular with keloidal scarring over the upper medial margin. Vitiligo was present in the centre of the scar. Right inguinal glands were enlarged but discrete.

Roentgenogram of chest was normal and that of the right thigh showed soft tissue swelling in the upper 3rd of the thigh on oblique view. Underlying bones were normal. The haemogram was within normal limits excepting for the erythrocyte sedimentation rate which was 120 mm/hr.

Biopsy of the lesion showed on haematoxylin eosin staining, hyperkeratosis and chronic granulomatous changes with both Langhans cells (Fig. 2 page No. 343) and foreign body type of giant cells. No granule was detected in the tissue submitted.

The pus and a portion of the biopsied tissue were sent for mycological investigations.

Mycological Studies

Direct examination

The purulent material, stained by Gram's method revealed numerous *Borrelia vincentii* with large fusiform bacilli and gram-positive bacillary and coccoid forms. No granules were seen in the pus.

Culture

The materials (pus and biopsied tissue) were inoculated on Sabouraud's dextrose agar slants with and without chloramphenicol and incubated at 26°C and 37°C respectively. Growth was visible in a few days on the plain Sabouraud's dextrose agar slants. Glabrous, mealy, irregularly folded orange coloured colonies were obtained in 5 to 10 days. No chalky aerial mycelium was seen (Fig. 3 page No. 343). Gram staining revealed the presence of gram-positive, delicate, branching hyphae breaking up into bacillary and coccoid forms. The organism was partially acid-fast. No growth was obtained in the media containing chloramphenicol.

Biochemical reactions

The organism failed to hydrolyse casein, decompose crystals of tyrosine or xanthine, showed very poor growth in 0.4% liquid gelatin. It utilised paraffin, and urease was positive. Acid was produced in glucose, sucrose and maltose, lactose and mannitol were not fermented.

Animal Pathogenicity

A saline suspension of the organism isolated, was inoculated intraperitoneally into a pair of mice. The animals died in 10 to 15 days and delicate, gram-positive, partially acid-fast, filaments fragmenting into bacillary and coccoid forms were demonstrated on direct examination of the peritoneal exudate and recovered in pure culture. Histopathological examination of the liver, spleen, lungs and kidney showed evidence of acute inflammatory reaction. Delicate gram-positive filaments were seen in sections stained by Brown and Brenn's modification of Gram's stain.

The isolate was sent to Prof. Vanbreuseghem, Institut de Medicine Tropicale Prince Leopold, Antwerp, Belgium, for confirmation and he identified it as *N. asteroides.*

Therapy

Good response was obtained with streptomycin sulphate 1 gm. and Dapsone, 100 mg. daily for three months. Iron and vitamin B6 were given as supportive therapy. The lesion had completely healed at the time of discharge and he was asked to continue dapsone for one more year.

Discussion

Mycetoma is caused by a variety of actinomycetes and eumycetes. Actinomycotic mycetomas are more common

in tropical and subtropical zones than in temperate zones. The species responsible for mycetoma varies from country to country.

Small grain mycetoma due to Nocardia species, namely *N. asteroides*, *N. brasiliensis* and *N. caviae* are indistinguishable on histopathological examination. So, for species identification, cultivation method of study is imperative. The small grains are usually not visible microscopically and therefore the purulent material itself, should be streaked on suitable media without any antibiotics.

Mycetoma due to *N. brasiliensis* is prevalent in Mexico, Central and South America and Africa. From India it has been reported from Bombay[6] and North India[5]. Only very few cases of *N. caviae* infections causing actinomycetoma pedis were on record[9,10,11,7]. The incidence of *N. asteroides* producing white to yellowish grains is rare, but it has been reported from United States and Lebanon. However, systemic nocardiosis has a worldwide distribution. From India few cases of mycetoma due to *N. asteroides* have been reported by isolation and the organism had also been isolated from the Indian soil[12,13,14,15].

Though the commonest site of mycetoma is the foot, extra pedal lesions can also occur. 3 cases of mycetoma have been reported from the gluteal region by Desai et al[6] out of which one was due to *N. asteroides*. Reddy et al[3] report involvement of the thigh from Vishakapatnam in one case and the causative agent was *Streptomyces pelletierii*. So this is the first case report of mycetoma of thigh from Madras where the causative agent *N. asteroides* has been isolated by cultural methods.

The tissue reaction is also unique here by the presence of prominent epitheliod reaction and foreign body giant cells which otherwise is characteristic of eumycotic mycetoma.

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