EUMYCETOMA PEDIS DUE TO EXOPHIALA JEANSELMEI

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Abstract

A case of eumycetoma of foot in an 8-year old male child was clinically diagnosed as chronic osteomyelitis and was microbiologically confirmed as eumycetoma. The case is being reported for its uncommon clinical presentation and etiological agent, Exophiala jeanselmei. The patient recovered completely after treatment with ketoconazole.

Key words: Black-grain, eumycetoma, Exophiala jeanselmei

Localized, slowly progressive, subcutaneous granulomatous infections caused by fungi or actinomycetes are known as eumycetoma or actinomycetoma respectively.1 The organisms are traumatically implanted in subcutaneous tissue from senescent plant material and form micro abscesses discharging aggregates of fungal or actinomycetes filaments known as grains. The sinuses drain on to the skin surface and may involve contiguous structures such as bone, causing osteomyelitis.2

Mycetoma is endemic around the Tropic of Cancer, including India, Pakistan, Sudan, Senegal, Somalia and Mexico.3 In India, its high prevalence is seen in Rajasthan, Tamil Nadu and Andhra Pradesh. Actinomycotic mycetoma is prevalent in south India, southeast Rajasthan and Chandigarh; while eumycetoma, which constitutes one-third of total cases is mainly reported from north India and central Rajasthan.4 It is important to distinguish between eumycetoma and actinomycetoma for treatment and prognosis of the disease.3

The common etiological agents of eumycetoma reported from different centers are Madurella mycetomatis, M. grisea, Acremonium spp, Aspergillus spp, Fusarium spp.4-8 We report here a case of black grain eumycetoma in a malnourished child.

Case Report

An 8-year-old boy, resident of Mathura (Uttar Pradesh), presented with swelling over dorsum of left foot of one-year duration at the outpatient Department of Orthopedics in September 2005. The patient also complained of purulent discharge from the site, sometimes associated with black granular deposits of three months' duration. There was past history of injury with wooden splinters a year and a half back. The patient had received various forms of treatment from village doctors (quacks) for the ailment but without any relief.

Examination revealed a malnourished child. A painless indurated swelling on left foot measuring 3 × 2.5 inches with multiple sinuses discharging purulent fluid with black granules was seen on local examination (Fig. 1).

There was no significant regional lymphadenopathy. Systemic examination was within normal limits. Laboratory investigations revealed low hemoglobin (8 mg%) and normocytic, normochromic anemia. Rest of the hematological investigations, including CD4+ and CD8+ counts, were normal. A radiograph of the left heel showed extensive loculated lytic areas of destruction within the body of the calcaneum with a typical “honeycomb” appearance. Magnetic resonance imaging of the foot revealed diffuse alteration in the marrow signal intensity of the calcaneum, appearing hypointense on T1- and hyperintense on T2-weighted images with expansile erosion of the bone. Multiple foci appearing hypointense on all pulse sequences were seen interspersed within the bone.

The patient was later referred to the Department of Microbiology for further investigations. Sample was collected by introducing a Pasteur pipette after thorough cleaning of the lesions. The aspirated material was rinsed in sterile saline and allowed to settle. A few black, irregular granules of variable size measuring 0.5-2 mm were observed in the aspirate. These were crushed, separated and subjected to microscopy and culture for mycological and bacteriological identification.1,3

Gram stain, 10% potassium hydroxide (KOH) mount, modified Ziehl-Neelsen stain (1% H2SO4) and periodic acid-Schiff (PAS) staining were done. The granules were inoculated onto two sets of Sabouraud dextrose agar with chloramphenicol (SDA-C) and were incubated at 30°C and 37°C respectively. These were also observed on discharge collected on the gauze of the dressing. These were crushed, separated and subjected to microscopy and culture for mycological and bacteriological identification.1,3

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incubated at 37°C for 14 days under aerobic and anaerobic conditions. The aspirate was also processed to exclude tubercular etiology by standard methods.

The microscopic examination of 10% KOH mount revealed irregular swollen cells, few septate hyphae and chlamydospores. Gram stain showed gram-variable irregular swollen cells and few septate hyphae. Periodic acid-Sciff revealed PAS-positive compact interwoven hyphae (Fig. 2). Oliveaceous black velvety colony with aerial mycelium appeared on SDA-C slants after 28 days of incubation (Fig. 3).

Microscopically, lactophenol cotton blue wet mount of the colony showed pale brown, branched, slender septate hyphae, giving rise to conidiophores from lateral and terminal position, bearing tapering annellids with spores at the tapered tips (Fig. 4). The microscopic morphology and conidiogenesis seen were compatible with that of *Exophiala jeanselmei*. The identification was reconfirmed at the All India Institute of Medical Sciences, New Delhi. Bacteriological cultures were sterile.

Antifungal treatment with tablet ketoconazole (100 mg × bid) was initiated on the basis of microscopic findings in the second week of September 2005. Surgical curettage and local debridement were also carried out once a month, followed by a five-day course of co-trimoxazole. After two months of therapy, the swelling regressed and the discharge of granules disappeared. No adverse effects were reported.

**Discussion**

Although eumycetoma was first reported from India in the middle of the 19th century, reliable epidemiological information remains scarce. The disease is characterized by a prolonged incubation period, slow and unremarkable clinical course and multiple causal agents. It goes undiagnosed perhaps due to lack of diagnostic facilities in mycology. Nevertheless, some information is available from different centers of India. *Exophiala jeanselmei* and *Curvularia lunata* are uncommon causes of black grain eumycetoma. There are few reports of occasional isolation of *E. jeanselmei* in Indian literature. Its presence in soil, senescent plant material and environment is well known. However, this fungus has

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**Figure 1:** Left foot showing multiple sinuses discharging black granules

**Figure 2:** Photomicrograph of the granule showing PAS-positive septate fungal hyphae (PAS, ×200; inset, ×450)

**Figure 3:** *Exophiala jeanselmei* showing oliveaceous black velvety colony with aerial mycelium on SDA-C plate after 28 days of incubation.

**Figure 4:** Lactophenol cotton blue mount of the colony showing pale branched, septate hyphae, giving rise to conidiophores bearing tapering annellids, with spores at the tapered tips (LCB, ×400)
not yet been reported from environmental sources in India. Epidemiologic data from other countries indicate that E. jeanselmei proliferates and survives better in relatively arid areas with short rainy season.\textsuperscript{2,3} It seems pertinent to add that the climate of Mathura is similar to that of other endemic areas, with average rainfall around 500 mm\textsuperscript{3} and climate with extremes of temperature.

The various predisposing factors cited are defect in T-cell function, diabetes mellitus and malnutrition.\textsuperscript{9,10} The malnourished status of the child could be an attributing factor. The localized form of eumycetoma is in accordance with studies reported earlier.\textsuperscript{1-4}

In this patient, direct microscopy of the granule revealed fungal elements, which were PAS positive. On this basis, diagnosis of eumycotic mycetoma was made and antifungal therapy was started within a week. Isolation of fungus in culture took 28 days, as E. jeanselmei is a slow-growing fungus. Thus, direct microscopy proved to be a useful rapid diagnostic tool.

The different modalities of treatment include trial of chemotherapy with ketoconazole or itraconazole. In case of failure, radical treatment by amputation surgery may be required. Currently, ketoconazole is the drug of choice. The drug is cost-effective but has hepatic and endocrinal side effects.\textsuperscript{10} In the present study, the patient was successfully treated with ketoconazole.

In conclusion, a greater awareness of the disease on the part of the clinician is warranted to suspect and investigate for mycotic etiology, especially in the absence of response to antibacterial therapy.

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References


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