A RARE CASE OF PHAEOHYPHOMYCOSIS CAUSED BY EXOPHIALA JEANSELMEI

A Case Report

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Summary

A case of phaeohyphomycosis in a three year old female child born to consanguineous parents is reported. The causative agent was *Exophiala jeanselmei*. The lesions were found in skin, subcutaneous tissue, lymph nodes and limb bones, which makes it a rare case.

KEY WORDS : Phaeohyphomycosis, *Exophiala jeanselmei*.

The term "Phaeohyphomycosis" was proposed by Ajello[1] for cutaneous, subcutaneous and systemic disease of man and animals caused by a variety of closely related dematiaceous fungi. Cystic subcutaneous lesions were caused by many species of fungi belonging to *Exophiala*, *Phialophora* and *Wangiella* whereas systemic infection often with cerebral lesions was reported to be caused often by *Cladosporium bantianum*[2]. This communication deals with a rare case of systemic phaeohyphomycosis without cerebral involvement caused by *Exophiala jeanselmei*.

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Case Report

A three year old female child born to consanguineous parents was brought with swellings all over the body in October, 1979 (Fig 1). Ten months earlier her mother noticed a small swelling in the left nostril and the child had some breathing difficulty. Within three days small raised itchy papular lesions appeared all over the face and these ultimately became verrucous plaques. One month later, child developed multiple swellings all over the extremities and subsequently spindle shaped swellings over the dorsum of the fingers and feet were found. This was followed by similar swellings over the wrist and elbows. The child had fever on and off during this period. On examination, the child was pale. Axillary and inguinal lymphnodes were enlarged on both sides, varying in size, firm, discrete and not tender. Other systems were normal except for hepatosplenomegaly. The verrucous plaques were dry and granulomatous in nature and were of varying sizes. Some were

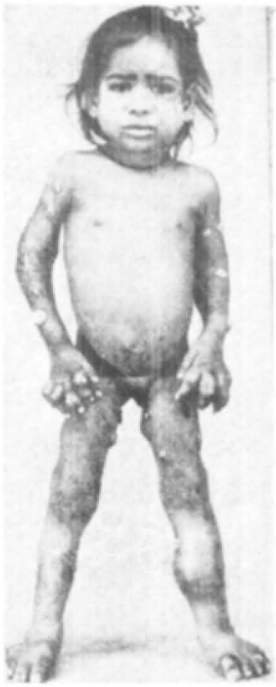

Fig 1
Photograph of the patient showing multiple verrucose plaques, subcutaneous swellings and spindle shaped swelling of the fingers.

grey in colour and were raised 3–5 mm above the skin. Some of the cutaneous lesions were firm and some were cystic. No signs of local inflammation were noted. Swellings over the fingers were cystic and some of these showed discharging sinuses but without granules. The finger movements were near normal. The swellings over the joints were also cystic and the ends of the long bones were tender and thickened. The joint movements were near normal.

Investigations

Hb. 5 gm%, total leucocytic count, 48000 per cu. mm. E.S.R. 35 mm/hour, Urine-normal; serum albumin 3.35 g%; globulins 2.3 g%; serum calcium 9.6 mg.%; serum phosphorus 4 mg.%, Mantoux – negative; V.D.R.L. – negative; C.S.F. (A) biochemical tests, normal (B) Culture – no fungus was grown; Bone marrow – normal picture. Roentgenograms showed evidence of chronic osteomyelitis in the long and short bones of upper and lower extremities (Fig. 2.) X-ray chest showed calcified spots in the left lung.

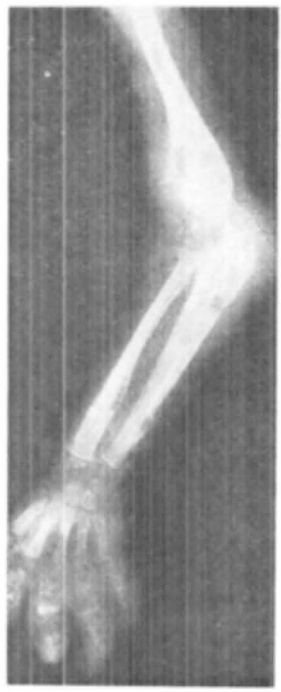

Fig. 2
Photograph of the X-ray film-left upper extremity showing chronic osteomyelitis.

Histopathology

Sections taken from the inguinal lymphnodes, skin and bone biopsies were stained with haematoxylin and eosin.

Skin biopsy showed moderate acanthosis with chronic inflammatory and granulomatous reaction around the dark walled fungii in the dermis (Fig. 3). Bone curettings showed fibrous tissue along with many histiocytes. A few tiny fragments of bone were seen being destroyed by the granulomatous tissue (Fig. 4). Lymphnode showed

Fig. 3
Photomicrograph of the skin biopsy showing chronic granulomatous reaction and fungal cells in the dermis (×400).

large foci of granulomatous reaction, containing many histiocytes and a few foreign body type of giant cells. The fungal filaments were seen within the histiocytes and giant cells as well as lying freely. None of the sections showed 'sclerotic bodies'.

Mycology

The aspirated pus from cystic lesions and biopsy material from skin, bone and lymph node were inoculated each into two sets of Sabouraud's dextrose agar slopes (SDA) with and without chloramphenicol and was incubated at room temperature and 37°C. Growth appeared within 4 to 7 days at room temperature and there was very scanty growth at 37°C. The colony which was moist and round initially turned into floccose with a raised centre and radial furrows and was olive grey in colour (Fig. 5). Microscopic examination of slide cultures and slant culture mounts under lactophenol cotton blue stain revealed dark pigmented septate mycelium. The conidia were found at the tips in small clusters and along the sides of the annellides (Fig. 6). On the basis of the above macroscopic and microscopic findings, the fungus was identified as *Exophiala jeanselmei*.

Discussion

A number of saprophytic dimorphic dematiaceous fungi cause subcutaneous mycotic infections such as chromomycosis and phaeohyphomycosis. The absence of diagnostic and pathognomonic thick walled, dark sclerotic bodies in tissues is the only difference between the above two diseases[2]. The present case did not show sclerotic bodies in any of

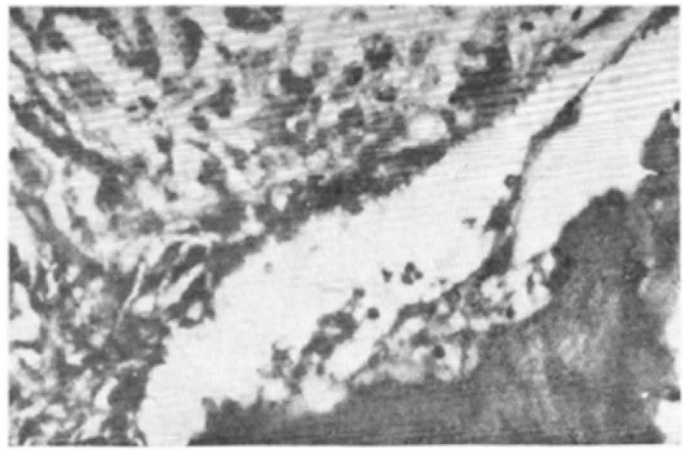

Fig. 4
Photomicrograph of the bone curettings showing the fungal chains at the upper left part of the field and a fragment of necrotic bone at the right lower part (×400).

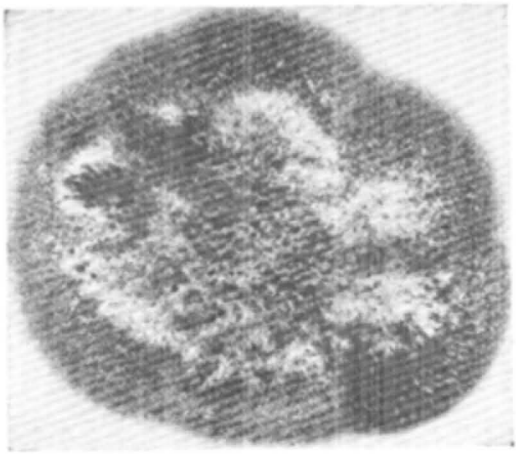

Fig. 5
Photograph showing the growth of *E. jeanselmei* on SAD at 30°C.

Fig. 6
Slide culture mount under lactophenol cotton blue stain showing pigmented septate mycelium with clusters of conidia at the tips and along the sides of the annellides.

the tissue sections studied and therefore is a case of phaeohyphomycosis and not chromomycoses. The presence of pigmented mycelial filaments in the tissues is confirmatory (Fig.3 & 4). To the best of our information so far, only one case of phaeohyphomycosis involving foot caused by *Cladosporium bantianum* is reported from India[2].

The aetiologic agents of phaeohyphomycosis are varied and numerous. At present 16 fungi belonging to genera are recognised as causative agents. In the present case *Exophiala jeanselmei* has been isolated from all the lesions (confirmed by Prof. L. N. Mohapatra, New Delhi) *E. jeanselmei* is known to cause mycetoma and also reported from India[3,4]. It is also reported to cause chromomycosis[5]. Thus the present case is to the best of our knowledge the first case of *E. Jeanselmei* causing phaeohyphomycosis in India.

This case is peculiar in its clinical presentation too. The patient was a 3 year old child and showed systemic involvement. The spread of the disease to distant parts including bones took place within a short span of 10 months. The involvement of lymph nodes and bones in this case makes it one of the rare presentations of infection by dematiaceous fungii. Rajam *et al*[6] reported a case of chromomycosis in a two year old male child with scaly nodules on the face and destructive

lesions in bones of limbs. The fungus implicated in that case was *Wangiella dermatitidis*. In the present case the occurrence of lesions in four different tissues namely skin, subcutaneous tissue, lymph nodes and bones, in a single individual is a unique feature and to the best of our knowledge not described earlier. It is also noteworthy that destructive lesions were seen only in bones of extremities.

This rare condition could be differentiated from mycetoma as there were no granules; from tuberculosis verrucosa cutis as the histopathological proof was not available. On the same grounds, it could be differentiated from candidiasis, leishmaniasis and tertiary syphilis.

Since the first lesion in this case was in the nose, it is possible that the infection occurred through inhalation of spores and that dissemination occurred through haematogenous route. It is also possible that the patient had some genetic abnormality that lead to immune deficiency which in turn was responsible for extensive systemic lesions. However, this hypothesis could not be verified in this case.

The patient was treated with thiabendazole 100 mg. tds for a period of 1 month. Afterwards, she was started on 5-Flurocystosine (150 mg/kg. body weight). The lesions started subsiding 1 month after initiation of treatment. Lopes *et al* reported 41.6% cure in chromoblastomycosis treated with 5-flurocystosine for a period of 3–10 months as stated by Marison *et al*[7]. Bayles[8] has shown a cure rate of 36.4% with thiabendazole. The child was taken home against medical advice. A follow up revealed that the child expired in August 1980. The cause of death was not known.

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