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Case report

Subcutaneous basidiobolomycosis, an unusual case

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ABSTRACT

Basidiobolus ranarum, belonging to the order Entomophthorales, is known to cause subcutaneous infection in healthy individuals. It has been isolated from extremities, trunk, intestinal tract, etc. Basidiobolomycosis usually occurs in children, less often in adolescents and adults. We report a case of subcutaneous basidiobolomycosis in a middle aged immunocompetent man, a farmer. He had presented with a painless swelling of three years duration at his right thigh. Initially the lesion was mistakenly treated as tuberculosis. Later, culture of biopsy specimen from the lesion yielded *Basidiobolus ranarum* showing characteristic conidia with beak-like papilla. The lesion completely subsided with oral potassium iodide treatment. Unlike mucorales, entomophthorales cause chronic, less aggressive infections and respond well to potassium iodide. This case emphsises the need for a high index of suspicion for fungal aetiology, including *Basidiobolus ranarum*, in chronic subcutaneous lesions. Fungal culture from tissue specimens helps in identifying the pathogenic organisms and choosing the appropriate treatment. Oral Potassium iodide can be used as an effective and cheap first line drug for basidiobolomycosis, especially in resource constrained settings.

1. Introduction

Fungal infections can present in a variety of forms. Fungi are often not considered in the differential diagnosis of unusual infections. *Basidiobolus ranarum*, belonging to the order Entomophthorales is known to cause subcutaneous infection in healthy individuals [1, 2]. It has been isolated from extremities, trunk, intestinal tract, etc. Basidiobolomycosis usually occurs in children, less often in adolescents and adults [3]. Here is a case report of subcutaneous basidiobolomycosis in an immunocompetent adult man which was first mistakenly treated as tuberculosis, later cured completely with oral potassium iodide (KI) therapy.

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2. Case Report

A 46-years-old, immunocompetent man, farmer by occupation presented with painless swelling at his right thigh since three years. The swelling was incidental in onset and there was no antecedent trauma to the site. It gradually extended to upper and lateral parts of the right thigh and also to upper part of the right leg. It was associated with darkening of the overlying skin and multiple small excoriations with minimal serous discharge. Around a year ago, histopathological study of the biopsy of the lesion was done in another centre, which showed non-caseating granulomatous tissue reaction, micro-abscesses and no acid-fast bacilli or fungal elements. Details of other investigations done at that time were not available. Patient was put on anti-tubercular treatment, which he had discontinued after 1 month.

On clinical examination after the patient presented in our hospital, an indurated non-tender flat subcutaneous swelling was seen at medial and anterior aspect of the right thigh extending to upper 1/3 of the right leg [Fig 1]. Margins were well defined. It was adherent to the overlying skin but not fixed to the underlying tissues. Overlying skin was hyperpigmented and excoriated at many places. Inguinal and popliteal lymph nodes were not

enlarged and movements at the right hip and knee joints were normal. Systemic examination revealed no other abnormalities. Laboratory investigations showed raised ESR [60 mm after 1 hr] and positive Mantoux test; Haemoglobin, random blood glucose, urine routine, peripheral smear study were within normal limits. X-ray of the right thigh showed swelling to be in the subcutaneous plane and no bone involvement.

Haematoxylin-Eosin stained smears of excision biopsy specimen of the lesion showed chronic granulomatous changes, Splendor-Hoepli bodies and no fungal elements. Microscopic examination of KOH mount of the biopsy tissue showed ribbon-like broad aseptate hyphae. The specimen was cut into smaller pieces and inoculated onto a pair of Sabouraud's dextrose agar and incubated at 25° C and 37° C. Both the tubes showed growth around 5th day. The colony was flat, waxy and buff coloured initially. It later became heaped up brownish coloured and was covered by fine, white surface mycelium. The reverse was not pigmented. Lactophenol cotton blue tease mount from the colony showed hyaline, broad pauciseptate hyphae (Fig 2) and many round sporangiospores with knob-like projection (Fig 3). Zygospores were not seen. Based on the clinical presentation, culture characters and microscopic morphology the organism was identified as Basidiobolus ranarum [3, 4].

The patient was started on oral saturated solution of potassium iodide (SSKI) 5 drops tid, gradually increased to 30 drops tid over 2 months, and continued for another 3 months. The swelling completely subsided after completion of the treatment.

Fig1. Clinical photograph showing subcutaneous lesion extending from thigh to upper 1/3 of the leg. Hyperpigmentation of the overlying skin and excoriations are seen.

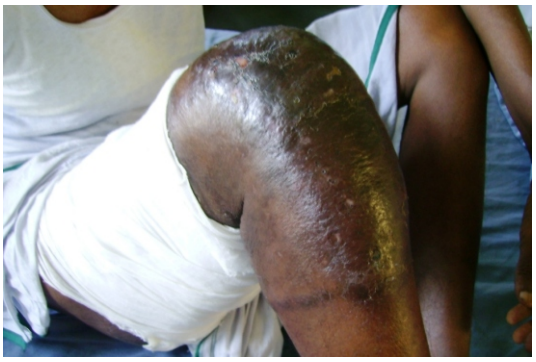


Fig 2. Lactophenol cotton blue preparation from colony showing hyaline pauciseptate hypahe and sporangiospores (100X)

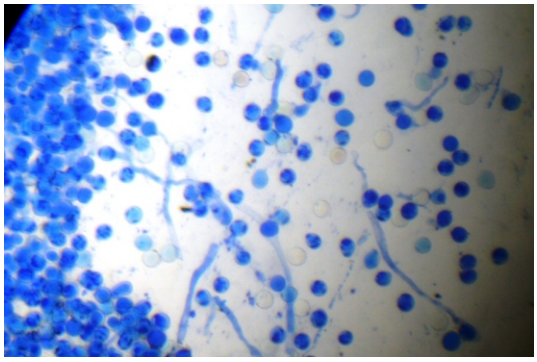
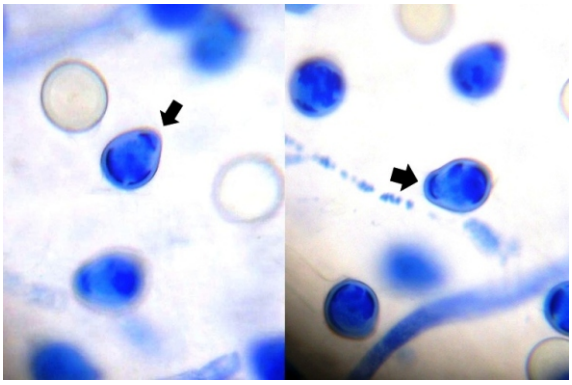


Fig 3. Lactophenol cotton blue preparation from colony showing sporangiospores with beak-like projections (Arrows) (400X)



3. Discussion

Basidiobolomycosis is a rare fungal infection due to *B. ranarum*, an environmental saprophyte found world-wide. It is a member of the order Entomophthorales, of the class zygomycetes, which has been isolated from decaying vegetation, foodstuffs, fruits, and soil and from the gastrointestinal tracts of reptiles, amphibians, fish, and insectivorous bats [2, 3]. Many cases have been reported from Africa and Asia, especially from southern parts of India [1-5]. Our patient was a native of south India.

There was no history of antecedent trauma to the site in the present case. The mode of acquisition of the disease remains poorly understood [3, 4]. The portal of entry is believed to be the skin, after insect bites, scratches, and minor cuts [3]. Ingestion of contaminated food and use of contaminated toilet leaves for cleaning of the skin after defecation have been considered the likely possibilities for intestinal basidiobolomycosis [4].

Basidiobolus and Conidiobolus species (Entomophthorales) cause chronic inflammatory, granulomatous disease collectively called entomophthoromycosis [6]. Although seldom life-threatening, these infections cause disfigurement and morbidity and are clinically significant where endemic [5, 6]. Subcutaneous zygomycosis is the commonest clinical form of basidiobolomycosis [2]. It is most common in male young children as a disease of skin and subcutaneous tissues, involving thighs and buttocks [1, 3]. It usually affects immunocompetent individuals with no predisposing factors [2, 3]. Rarely *B. ranarum* has been isolated from from intestine, paranasal sinuses and disseminated infections [1, 2, 3, 4]. The present patient was an immunocompetent adult with no apparent risk factors and presented with involvement of subcutaneous tissue of right thigh. Regional lymph nodes are usually not enlarged, as in this case [1].

Basidiobolus can be isolated from surgical specimens, and it should be inoculated soon after resection because it does not survive at 4° C [3]. Definitive diagnosis is by culture and Sabouraud agar is an adequate medium on which it forms characteristic waxy colonies in 2-3 days [3, 6]. The organism is microscopically identified by broad pauciseptate hyphae, sporangiospores and zygospores, both showing characteristic knob/beak-like projections [4]. In the present case typical hyphae and sporangiospores were seen and no special culture technique was done to demonstrate zygospores.

Basidiobolomycosis being a chronic granulomatous infection, histological study shows eosinophilic infiltration, granuloma and sometimes Splendore-Hoepli phenomenon, as in this case [1, 7]. Immunodiffusion test using mycelial antigen is specific, rapid and inexpensive alternative means of diagnosis [3, 4, 8]. Patients with subcutaneous entomophthoromycosis are immunoresponsive and hence serodiagnosis is possible with immunodiffusion and ELISA; Immunodiffusion can also easily differentiate histologically similar Basidiobolus and Conidiobolus spp. [4, 8]. As in this case, nonspecific clinical features, raised ESR and histological features such as granulomatous inflammation, Splendore-Hoepli phenomenon may mislead towards a far more common disease, tuberculosis [4]. Lack of culture facilities and molecular diagnostic techniques for confirming tuberculosis at all the centres in developing tropical countries like India and attaching undue significance to Mantoux test positivity in adults in TB endemic areas might also contribute to such errors. General awareness of fungal infections, high index of suspicion, routine use of simple fungal culture methods and use of fungus-specific stains in histopathology can help in correct diagnosis. Though serological tests can be done to detect basidiobolomycosis, they may not be routinely available.

Many isolates of *B.ranarum* have been reported to be resistant to Amphotericin B based on broth dilution studies [4, 8]. KI and Azoles (Ketoconazole, Fluconazole) have been proven to be very effective for entomophtoromycoses [1, 4, 9]. For convenience, KI is administered in the form of saturated solution (SSKI) at a dose of 47 mg/drop. KI is not effective for other group of zygomycoses, the mucormycoses [9]. KI does not have a direct antifungal action as demonstrated by in vitro studies; its effect in vivo is believed to be non-specific [4, 6, 9]. Despite some side effects KI continues to be used as the first line drug for basidiobolomycosis because of its effectiveness and low cost, especially in the tropical world [9]. Our patient's lesion disappeared completely with SSKI therapy.

4. Conclusion

Subcutaneous basidiobolomycosis though seen more commonly in children, it can affect adults also. Unlike mucorales, entomophthorales cause chronic, less aggressive infections and respond well to potassium iodide. The present case emphasises the need for a high index of suspicion for fungal aetiology, including *B.ranarum,* in chronic subcutaneous lesions. Fungal culture from tissue specimens helps in identifying the pathogenic organisms and choosing the appropriate treatment. Oral Potassium iodide can be used as an effective and cheap first line drug for basidiobolomycosis, especially in resource constrained settings.

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