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## Case Report

## Subcutaneous entomophthoromycosis in a child presenting as panniculitis: a case report from Bihar, India

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## A B S T R A C T

A 5-year-boy from Bihar, India was admitted to a tertiary care hospital with painful swelling over both lower limbs and buttocks, which had been increasing progressively for the past 1 year. The condition was initially undiagnosed and was later misdiagnosed as non-infective panniculitis, delaying treatment. Subsequently, the patient was diagnosed with subcutaneous entomophthoromycosis caused by *Basidiobolus* spp. A preliminary diagnosis was made by considering the history, clinical features, radiological findings and histopathological examination of the biopsied tissue. The confirmatory diagnosis was made using conventional techniques on aspirated pus, which included KOH wet mount and fungal culture on Sabouraud dextrose agar tubes incubated at 28°C and 37°C, respectively. Lactophenol cotton blue mount and slide culture were performed for identification of the fungal isolate. The patient responded well to oral itraconazole and oral potassium iodide.

Delayed diagnosis and extensive involvement in a rare case of subcutaneous entomophthoromycosis causing panniculitis emphasizes the importance of correct diagnosis and appropriate, effective treatment.

## Introduction

Entomophthoromycosis is a rare mycosis which causes chronic progressive soft tissue swelling and visceral infections, mainly in immunocompetent hosts. It includes basidiobolomycosis and conidiobolomycosis, with the former being more common and occurring predominantly in tropical and subtropical regions (Anand et al., 2010; Kumaravel et al., 2016). Entomophthoromycosis is often misdiagnosed as it can mimic cutaneous malignancy, tuberculosis or panniculitis clinically, leading to a delay in appropriate management (Takia et al., 2020). This article reports a case of subcutaneous entomophthoromycosis in a child presenting with panniculitis.

## Case report

A 5-year-old boy from Punpun, Bihar, India was admitted to a tertiary care hospital in Patna with multiple, painful nodules over both buttocks, back and thighs of 1 year's duration, and progressive swelling over both lower limbs of 4 months' duration. He was asymptomatic 1 year previously, when a small raised lesion was first noticed on his right buttock following a minor trauma. Within 2 months, he had developed multiple firm nodules over both buttocks and thighs. Later, he complained of a discharging sinus over his left knee, secondary to a surgical intervention. Although he visited several physicians and received various treatments, the condition remained undiagnosed (Fig. 1).

At the time of admission to the authors' hospital, the child was febrile with pitting oedema of the lower limbs and scrotum, and mild abdominal distension. Local examination revealed discrete, firm-to-hard, mildly tender nodules and indurated subcutaneous plaques over both buttocks, lower back, groin and thighs. The overlying skin was hyperpigmented.

On investigation, the child was found to have severe anaemia and hypoalbuminaemia, and raised erythrocyte sedimentation rate and C-reactive protein. Differential diagnoses of cutaneous tuberculosis, malignancy, infective panniculitis and lobular panniculitis were considered. Mantoux test and alpha-1 antitrypsin level were normal. Viral markers for human immunodeficiency virus, hepatitis B and hepatitis C were negative. Computed tomography scan of the pelvis and thighs revealed multiple, peripherally enhancing, loculated collections with inflammation in subcutaneous fat and tissues. Biopsy and histopathological examination of the tissue revealed predominantly lobular panniculitis, with the lower dermis showing histiocytes. A fair number of giant cells were seen with engulfed periodic acid–Schiff-positive foreign bodies suggestive of fungal aetiology. They were accompanied by an inflammatory reaction with numerous eosinophils, histiocytes and epithelioid cells. A typical Splendore–Hoepli phenomenon was not seen on histopathology.

Thick purulent material aspirated from the swelling over the left thigh was sent for bacterial, fungal and mycobacterial culture. A KOH wet mount of the aspirated pus showed wide (10–20 μm), pauci-septate,

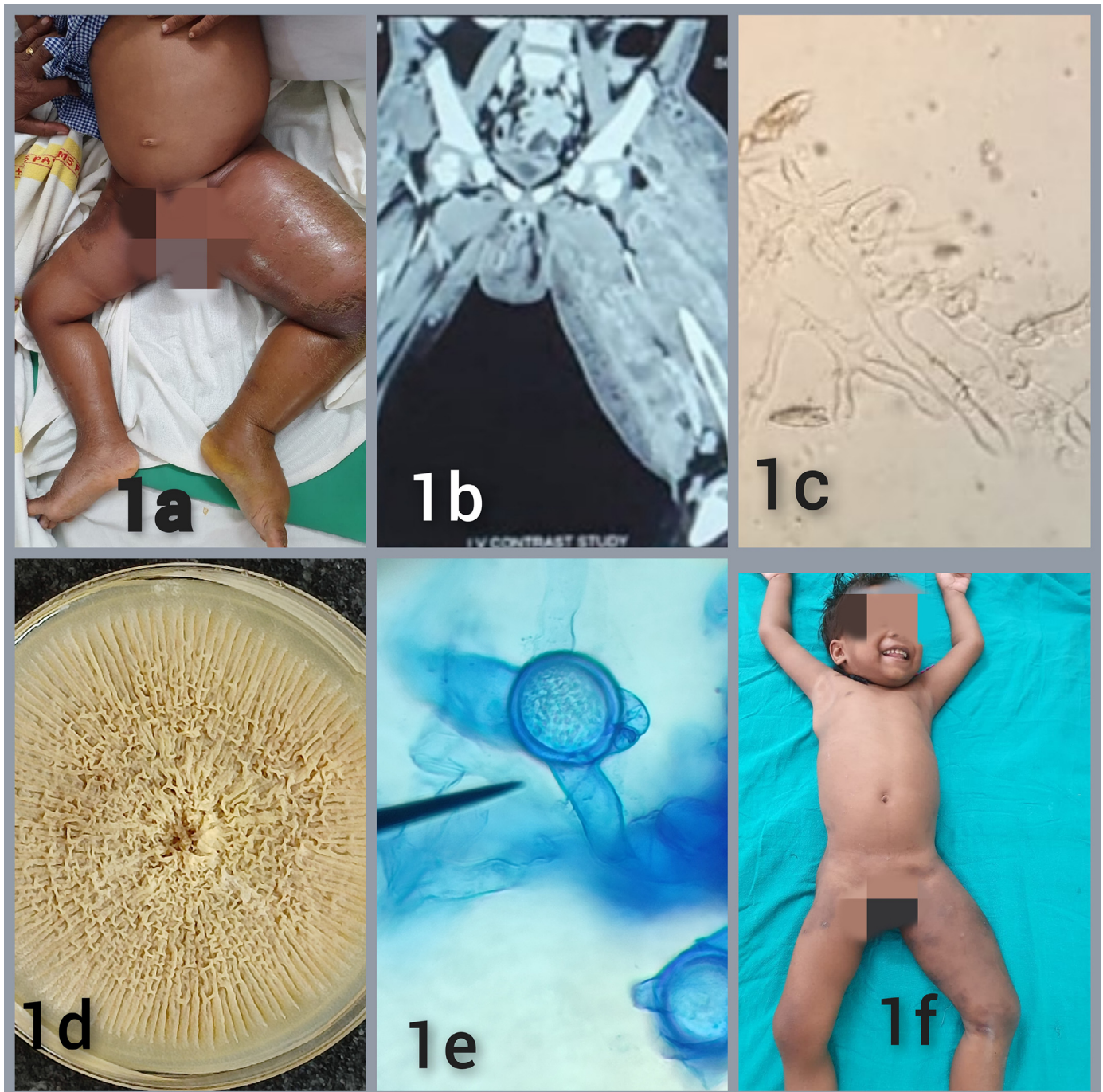
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**Figure 1.** (a). At initiation of treatment. (b) Computed tomography scan findings. (c) KOH mount (original magnification 40X): pauciseptate hyaline hyphae (width 10–20  $\mu\text{m}$ ). (d) Colony morphology on Sabouraud dextrose agar: flat, waxy, buff-coloured colony with 'heaped-up' centre and furrowed periphery. (e) Lactophenol cotton blue mount (original magnification 40X): wide hyaline hyphae with round, smooth, thick-walled intercalary zygospores (20–50  $\mu\text{m}$  in diameter) and prominent beak-like appendage. (f) At end of treatment with itraconazole.

wide-angle branched hyaline hyphae, suggestive of a fungal infection. Sabouraud dextrose agar (SDA) yielded rapid growth of flat, waxy, buff-coloured colonies with a pale reverse. Older cultures showed a 'heaped-up' centre with a furrowed periphery, and had a musty odour. Lactophenol cotton blue mount of growth revealed wide hyaline hyphae with round, smooth, thick-walled intercalary zygospores (20–50  $\mu\text{m}$  in diameter), with a prominent beak-like appendage on one side. The isolate was identified as *Basidiobolus* spp. Repeated sampling from various sites

of infection from the patient yielded the same growth, proving its role as the causative agent.

Oral itraconazole, 5 mg/kg/day, and supersaturated potassium iodide solution, 4 drops t.i.d., were started and the patient responded to treatment. After 3 weeks of treatment, the child became afebrile and his nodules started softening and reducing in size. At the time of writing, the patient remains on treatment and is clinically improving. His biochemical parameters are monitored frequently.

## Discussion

Entomophthoromycoses are a group of infections caused by fungi belonging to the order Entomophthorales, which includes two genera: *Conidiobolus* and *Basidiobolus* (Shaikh et al., 2016).

*Basidiobolus* spp. were first isolated in 1886 from frogs. They are present on decaying vegetation, insects, and faeces of amphibians and reptiles (Rajan et al., 2017). The first human infection by *Basidiobolus ranarum* was described in 1956 in Indonesia (Mondal et al., 2015). *B. ranarum* is endemic in tropical and subtropical regions such as India, Pakistan, Myanmar, Uganda, Kenya, Ghana, Ivory Coast and South America (Prabhu and Patel, 2004; Takia et al., 2020). In India, most of the initial reports were from South India (Jayanth et al., 2013; Mondal et al., 2015). Infection may be acquired by traumatic implantation following an insect bite, minor trauma from vegetation, or intramuscular injections (Diwakar et al., 2007; Anand et al., 2010). Basidiobolomycosis usually manifests as gradually progressive, firm-to-hard, painless, disciform nodules affecting the thighs, buttocks, perineum and gastrointestinal tract (Anaparthi and Deepika, 2014; Shaikh et al., 2016).

The case patient was a 5-year-old boy from Punpun, Bihar, India with a history of traumatic implantation. According to the literature, infections occur predominantly in male children with a history of trauma (Karuna et al., 2015; Mondal et al., 2015). The patient presented with chronic, slowly progressive, multiple, painful nodules over his buttocks, back and both lower limb with extensive swelling. Most reported cases have a similar clinical presentation, but without pain (Anaparthi and Deepika, 2014). The anatomical sites are similar to those reported previously. The patient had microcytic hypochromic anaemia, which has been well documented in the literature (Rajan et al., 2017). Subcutaneous entomophthoromycosis may mimic cutaneous tuberculosis, sporotrichosis, filarial elephantiasis or malignancy, and is often misdiagnosed (Anaparthi and Deepika, 2014; Raveenthiran et al., 2015). The study case was initially misdiagnosed as lobular panniculitis of non-infectious origin.

The aetiological agent can sometimes be confused with mucormycetes due to overlapping anatomical sites and similar histopathological findings. Also, during the coronavirus disease 2019 pandemic, when there was a peak in the number of cases of mucormycosis, all broad, pauci-septate and hyaline hyphae were not found to yield mucormycetes. Thus, culture remains the 'gold standard' for diagnosis (Anaparthi and Deepika, 2014). Subcutaneous *Basidiobolus* spp. infection is a potentially curable disease. Longstanding, grossly deformed lesions can be treated safely and completely with itraconazole and potassium iodide (Patro et al., 2019; Takia et al., 2020). Surgical debridement should be discouraged because of the potential of *Basidiobolus* spp. lesion to spread to other anatomical sites (Prabhu and Patel, 2004). This clinical condition has been reported previously in natives of Bihar, but was diagnosed at hospitals outside the state. To the best of the authors' knowledge, this is the first indigenously reported case from Bihar. As the region has subtropical conditions, it is considered that other cases from this region may have gone undiagnosed in the past.

## Conclusion

This case report highlights the importance of clinical suspicion and awareness of fungi as a probable cause of chronic subcutaneous disease, especially in children. Early and correct diagnosis can prevent morbidity and disfigurement caused by misdiagnosed advanced disease. This case report also emphasizes the role of microbiological procedures, including culture, in establishing the aetiological diagnosis for timely and appropriate management. Children with longstanding basidiobolomycosis can be treated safely and completely with itraconazole. Surgical intervention may not be necessary.

## Conflict of interest statement

None declared.

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## Ethical approval

The case patient gave consent to publish his story and picture anonymously.

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