Subcutaneous zygomycosis presenting as soft-tissue 'tumour' over scapula – case report and review of literature

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ABSTRACT

Subcutaneous zygomycosis due to Basidiobolus ranarum is endemic in India, especially in Southern India. This is a case report of animmunocompetent manual labourer who presented with a painless swelling overlying the left scapula. Preoperative imaging studies suggested the possibility of cavernous haemangioma. Excision biopsy was performed and gross as well as histopathological assessment showed it to be a fungal lesion (entomophthoromycosis). Culture of the affected tissue yielded B. ranarum. The patient was treated with potassium iodide and itraconazole and the lesion healed completely in 2 months.

Key words: Subcutaneous zygomycosis, Basidiobolus ranarum, entomophthoromycosis, itraconazole, phycomycosis

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INTRODUCTION

Basidiobolus ranarum is a saprophytic fungus that is present in soil, decaying organic matter and in the gastrointestinal tract of several species of amphibians and reptiles.(1) It may cause subcutaneous zygomycosis (most common), gastrointestinal zygomycosis or acute systemic infection. B. ranarum tends to cause lesions in immunocompetent persons (unlike mucorales, which proliferate in immunodeficient individuals). Children under the age of 10 years are most often affected, although few reports of adult cases exist.(2)Subcutaneous zygomycosis in a manual labourer is discussed in the present case report. It is of interest to the orthopaedic surgeon since it mimics soft tissue neoplasms and causes diagnostic dilemmas and needless surgical interventions if the surgeon is unaware of this curable condition.(3, 4)

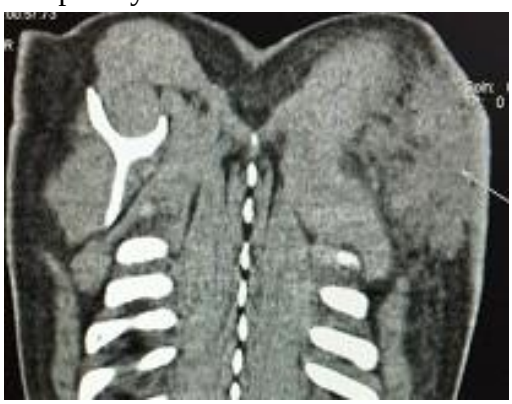
CASE REPORT

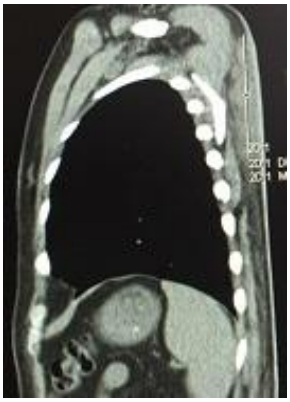
A 51-year-old male labourer presented to the out-patient department with a painless swelling in the left scapular region of 12-months duration. There was no history of previous injury. There was no history of fever and constitutional symptoms. There was no history of diabetes mellitus, chronic alcoholism and other diseases associated with immunocompromised status such as HIV infection. On examination, there was a 10 cm x 6 cm subcutaneous swelling overlying the left scapula which was variegated in consistency (firm and soft areas) and non-tender. The skin over the swelling was normal without any ulcers or sinuses.The edges were indistinct and merged into the surrounding tissues. The swelling was tethered to the underlying muscles but was not adherent to the scapula itself. There was no distal neurovascular deficit in the involved upper limb and no lymphadenopathy in the left axilla. There were no similar swellings elsewhere and systemic examination was normal.

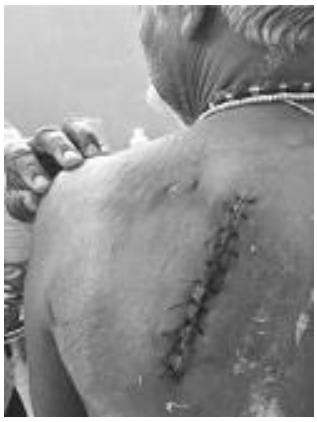
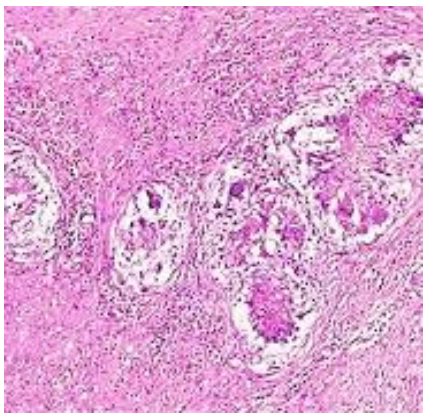
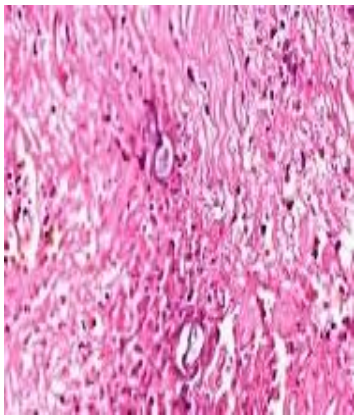
Blood investigations were within normal limits. Chest radiograph showed no bony abnormalities in the scapula or ribs underlying the swelling. CT scan report suggested the possibility of soft tissue neoplasm, possibly cavernous haemangioma (Fig. 1a and 1b). Since the investigations were inconclusive, it was decided to perform excision biopsy of the swelling (Fig 2). Histopathological examination of the excised mass showed fibrocollagenous and fibroadipose tissue exhibiting areas of necrosis surrounded by epithelioid cell granulomas. Stroma showed dense mixed inflammatory cell infiltrate rich in neutrophils along with few proliferating blood vessels and thin-walled, broad, often aseptate hyphae and hyphal fragments (fig 3a and 3b). The hyphal fragments had a thick eosinophilic sheath and a few hyphal-fragments were phagocytosed within giant cells. Splendore-Hoeppli phenomena (6) was noted (presence of eosinophilic material around the hyphae). Sections were negative for malignancy. The histopathological report was that of granulomatous inflammation secondary to entomophthoromycosis.

A 10% potassium hydroxide wet mount of the tissue showed broad, irregular, and aseptate hyphae. Culture on Sabouraud's dextrose agar showed a creamy white fungal colony andLactophenol cotton blue wet mount of the fungus showed it to be B.ranarum.

The patient was treated with a combination of oral potassium iodide (1.5g/day) and oral itraconazole (400mg/day) for 2 months following the biopsy report. The lesion healed completely with no recurrence at 12 months following the completion of treatment.





DISCUSSION

Subcutaneous lesions are the most common clinical presentation of basidiobolomycosis and is usually thought to occur via traumatic implantation of the fungus. The causative agent is Basidiobolus ranarum.It is endemic in India and the prevalence is probably higher in South India.(5)Histologically, eosinophilic infiltration is common in Basidiobolus infections and the Splendore-Hoeppli phenomenon is often present. The clinical importance of these subcutaneous lesions is that they occur in immunocompetent patients and are likely to be diagnosed as neoplasms.(2-4)Patient may undergo repeated and unnecessary surgical interventions for a pathology that is easily cured with medical management. It is reported to simulate soft tissue tumors, synovial sarcoma, Burkitt's lymphoma, Wegener's granulomatosis and infections such as tuberculosis, sporotrichosis and parasitic infestations such as onchocerciasis.

Simple excision of these 'tumors' without concomitant anti-fungal medical therapy is likely to result in recurrence of the lesion which causes leads to further diagnostic confusion. Verma et al have reported a patient with B. ranarum infection presenting as a subcutaneous swelling in the neck that had been managed with excision biopsy 3 times earlier.(3) No clear histopathological diagnosis had been established following the first excision biopsy. The swelling recurred and the second excision biopsy was reported as parasitic infestation and the patient was treated with anti-helminthic treatment. The swelling recurred again and the third excision biopsy was reported as tubercular infection and the patient received anti-tuberculous chemotherapy for a few months without improvement. Only in the fourth biopsy, the fungal nature of the infection was established and the patient was started on anti-fungal therapy. This was followed by complete resolution of the lesion without recurrence. Thus, a high index of suspicion is essential and in doubtful situations, a second review of the slides by an experienced pathologist is essential. This helps to avoid recurrences and unnecessary treatment with anti-tubercular or anti-helminthic medications. In our patient, the preoperative diagnosis was incorrect, but the histopathological report alerted us to the possibility of zygomycosis and anti-fungal therapy was instituted postoperatively.

Basidiobolomycosis is most often seen in children below the age of 10 years but our report as well as the few earlier reports shows that no age group is exempt.(1,2,5) The organism has

been treated with oral potassium iodide, azoles (such as itraconazole and fluconazole), trimethoprim-sulfamethoxazole and amphotericin B.(2-6) Some studies have reported that amphotericin B is ineffective against B.ranarum, we chose a combination of KI and azole in our patient. The learning point from this patient is that orthopaedic surgeons should consider the possibility of subcutaneous zygomycosis when dealing with soft tissue neoplasms and seek the assistance of expert microbiologists in confirmation of the diagnosis.

CONCLUSION

The learning point from this patient is that orthopaedic surgeons should consider the possibility of subcutaneous zygomycosis when dealing with soft tissue neoplasms and seek the assistance of expert microbiologists in confirmation of the diagnosis. These lesions may resolve with medical anti-fungal therapy without requiring surgical intervention.

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