

## CASE REPORT

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### A RARE CASE OF SUBCUTANEOUS ENTOMOPHTHROMYCOSIS IN A 11 YEARS OLD CHILD MIMICKING SOFT TISSUE TUMOR TREATED WITH ITRACONAZOLE: CASE REPORT

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**ABSTRACT:** Entomophthoromycosis (Subcutaneous zygomycosis) is a subcutaneous fungal infection that is mostly restricted to tropical areas of Asia, Africa, South America characterized by formation of firm, non-tender swellings, generally on the extremities, trunk. This fungal infection presentation may mimic soft tissue tumor in its presentation and there is a need to keep in mind this diagnosis in endemic areas so that early recognition can be done and disfigurement resulting from the advanced disease can be prevented. The disease occurs in age group of 20-60 years but rarely reported in children below 15 years. We present a 11 year old child who presented with multiple subcutaneous nodules in left forearm whose histopathology was showing features of entomophthoromycosis and successfully treated with itraconazole.

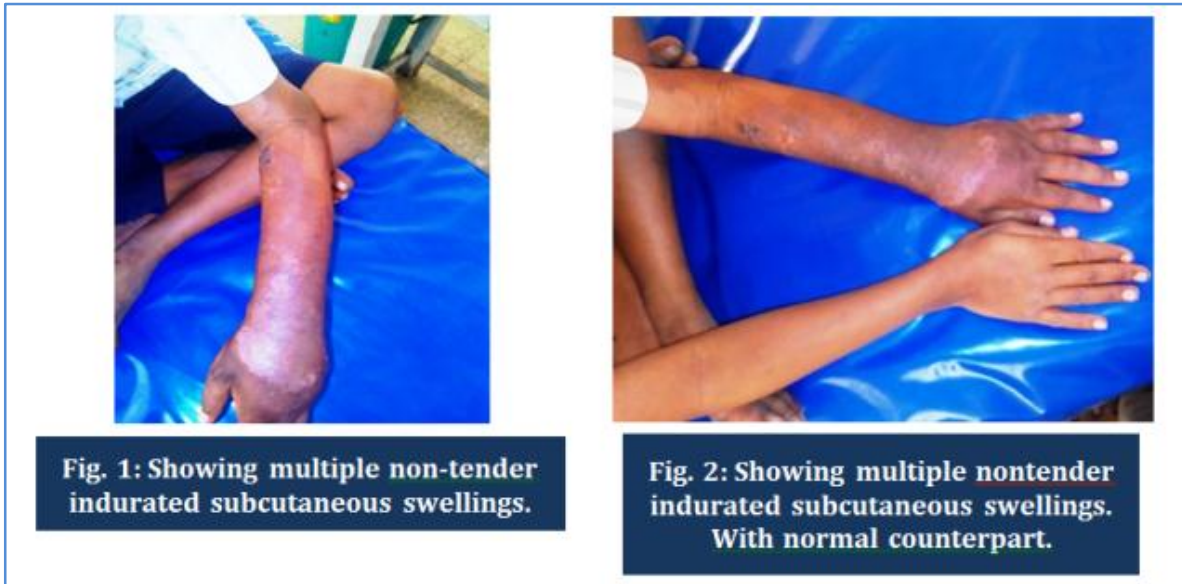
**KEYWORDS:** Entomophthoromycosis (subcutaneous zygomycosis), Itraconazole.

**INTRODUCTION:** Entomophthoromycosis is a rare entity caused by fungal infection. It occurs in two clinical forms, subcutaneous zygomycosis caused by *Basidiobolus ranarum* and rhinofacial zygomycosis caused by *Conidiobolus coronatus*.<sup>1</sup> The infection involves subcutaneous tissue affecting immunocompetent individuals.<sup>1</sup> The disease occurs in age group of 20-60 years but rarely reported in children below 15 years of age.<sup>2</sup> We present a 11 year old child who presented with multiple subcutaneous nodules in left forearm whose histopathology was showing features of entomophthoromycosis and successfully treated with itraconazole.

**CASE REPORT:** A 11-year-old previously healthy boy was admitted with complaints of multiple gradually increasing painless swellings over the left forearm and hand since 6 months with a significant past history of excision of a soft tissue swelling 6 months back details of the procedure done and histopathology not available.

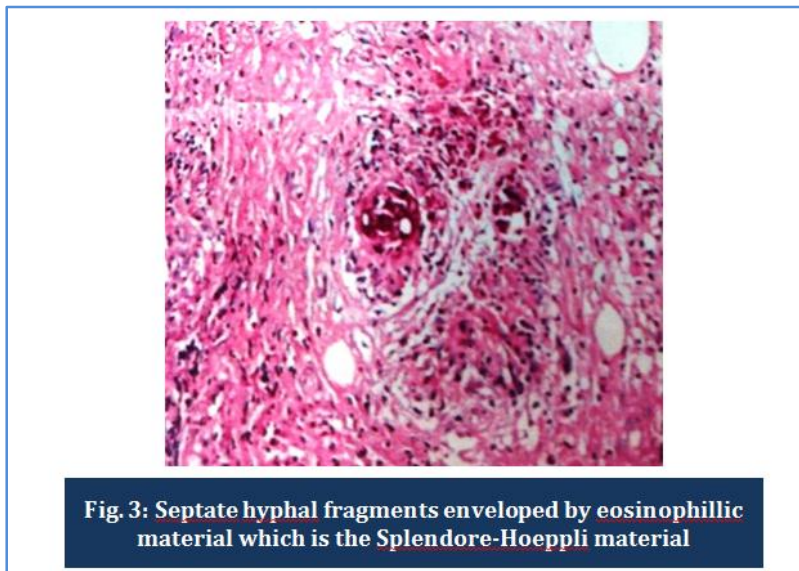
Local examination showed multiple nontender indurated subcutaneous swellings 1-2cm size concentrated over dorsal surface of left forearm and left hand with incision scar over ventral aspect. The swellings were firm in consistency and clinical diagnosis of soft tissue tumor recurrence was made. 2x1cm epitrochlear lymph node is palpable.

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Routine laboratory investigations revealed total count of 12,200/cmm. Hiv status was negative.

Excision biopsy of the epitrochlear lymph node done and incisional biopsy of one of the forearm lesion done. Histopathology of the incisional biopsy showed dense mixed inflammation rich in eosinophils and foreign body giant cell reaction seen around fungal elements that are very broad based, pauciseptate with paucibranching and show Splendore – Hoeffli reaction. Lymph node histopathology showed reactive follicular hyperplasia. Sections are negative for malignancy.



The patient was started on antifungal treatment with tablet itraconazole 100mg per day. And patient was followed up after 15 days which showed decrease in the size of lesions and limb swelling also decreased. Patient continued on itraconazole for 6 months. Complete recovery noted.

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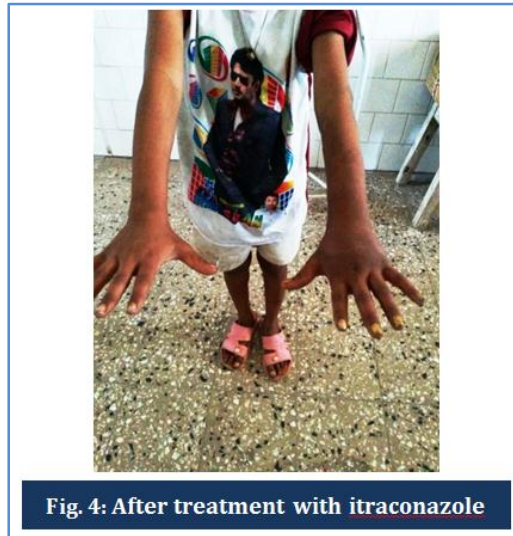


Fig. 4: After treatment with itraconazole

**DISCUSSION:** Lei-Kian Joe described the first case of entomophthoromycosis in humans in 1956 in Indonesia whose infective agent was *Basidiobolus ranarum*.<sup>3</sup> There are approximately 250 cases caused by *Basidiobolus* have been reported.<sup>3</sup> Zygomycosis is a class of fungi which causes infections of respiratory tract and subcutaneous tissues.<sup>2</sup> This class contains two orders of organisms that cause disease in humans, i.e., Mucorales and Entomophthorales.<sup>4</sup> Entomophthoromycosis cases have been reported from tropical countries like Nigeria, Ghana, India, Sudan, Uganda and Indonesia.<sup>5</sup> The infection usually occurs as small papules following a scratch or puncture by an insect, plant, or fungus-bearing object.<sup>6</sup> Possibly due to the production of protease enzyme by the fungus, it is able to invade subcutaneous tissues and often mimick soft tissue tumor.<sup>7</sup> The disease primarily involves subcutaneous tissue, but occasionally deeper structures are invaded.<sup>3</sup>

Entomophthoromycosis is a curable condition which can mimick a neoplasm like soft tissue tumor, synovial sarcoma, and Burkitt's lymphoma.<sup>8</sup> The disease occurs in age group of 20-60 years but rarely reported in children below 15 years of age.<sup>2</sup>

Histologically, entomophthoromycosis is associated with eosinophilic infiltration due to Th1 and Th2 immune response which causes release of IL-4 and IL-10.<sup>9</sup> The other important feature the "Splendore-Hoeppli" reaction characterized by presence of broad thin walled infrequently septate hyphal fragments enveloped by eosinophilic material which is the Splendore-Hoeppli material.<sup>1</sup>

Patients with this fungal infection respond well with oral potassium iodide as well as azoles especially itraconazole.<sup>10</sup>

Thus to summarize, entomophthoromycosis presents as subcutaneous swellings, painless, and usually observed in tropical areas which often mimicks soft tissue tumor. High index of suspicion in endemic areas and demonstration of aseptate fungal hyphae on histopathology clinches the diagnosis. Fungus is difficult to grow on culture. Treatment is with potassium iodide and itraconazole.

**CONCLUSION:** Entomophthoromycosis should be in the differential diagnosis of subcutaneous swellings in tropical areas because the condition is potentially curable. Treatment should be started based on the histopathology report with potassium iodide and azoles especially itraconazole.

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