

Subcutaneous zygomycosis - a case report

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Abstract

A case of subcutaneous zygomycosis caused by *Conidiobolus* species occurring in a 16 year old boy from Ingiriya, a rural area of Sri Lanka is reported. This is the first published report of this rare disease from Sri Lanka. The patient presented with chronic swelling of the upper lip. Fungal spores were detected in the histology sections. Patient was treated with saturated potassium iodide (KI) solution and itraconazole.

Introduction

Subcutaneous zygomycosis is caused by the fungal class zygomycetes¹. This consists of primitive, fast growing, widely distributed, terrestrial group of fungi. They are largely saprophytic on plant debris and are found in the soil. Mucorales and Entemophthorales are the two subclasses¹.

Mucorales grow rapidly and is commonly seen in the immuno-compromised. The disease is acute, rapidly spreading and fulminant. The fungi can invade the blood vessels. It can affect the skin, lungs, gastro-intestinal tract, and can disseminate^{1,2}.

Entemophthorales cause a slowly progressive disease with subcutaneous nodules. These are uniform, hard, movable, disc shaped masses. The surface skin may be edematous and tense but does not ulcerate. Lymph nodes are not involved. Usual transmission is by traumatic inoculation. There are two clinically important species^{1,2}.

Basidiobolus (*B.ranarum*), which causes lesions in limbs, chest, back and buttocks has been reported from South East Asia and Africa⁴. *Conidiobolus* (*C.coronatus/incongruous*) enters via nasal mucosa and gives rise to nasal obstruction, sinus pain and nasal discharge. Perinasal nodules can occur. This infection has been reported from, West Indies, South America, Africa and India⁴.

Case report

A 16 years old boy was referred from an ENT ward to the dermatology unit at National Hospital of Sri Lanka in October 2002, with a history of progressive painless swelling of upper lip for one year. He

had knocked against a lamp post causing trauma to his nose, while playing. Traumatized nasal septum was surgically corrected. Swelling of upper lip gradually started few weeks later. There were no constitutional symptoms.

It was a uniform, hard, non tender subcutaneous swelling of central part of the upper lip (Figure 1). Margins could be defined by palpation. Skin and mucosa were normal. There was no regional lymphadenopathy. There were a few lesions of acne. Other systems were clinically normal.

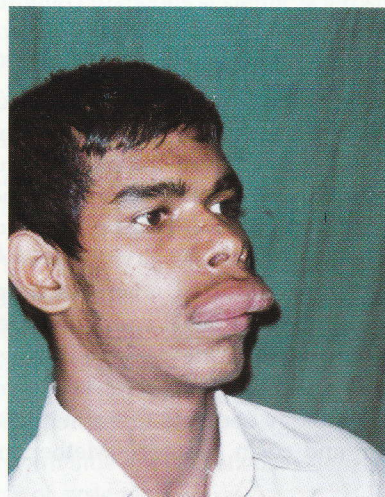


Figure 1. Grossly swollen lip of the patient before treatment.

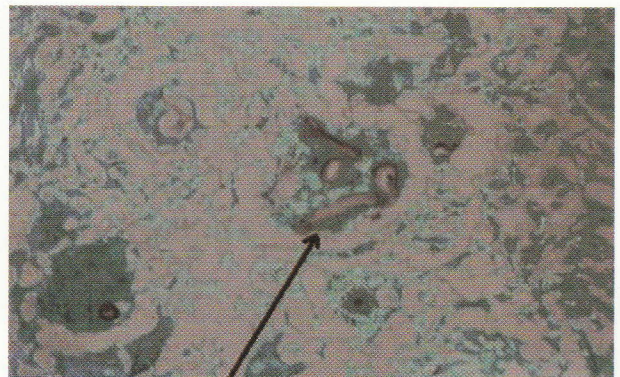


Figure 2. Fungal hyphae in tissue section stained with Grocott's stain (Arrow points to the hyphae).

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Fungal smears and culture were negative. Histopathology revealed a mixed cellular infiltrate with foreign-body type giant cells. Fungal hyphae and spores were detected which were identified as conidiobolus species by the mycologist.

The basic blood investigations, liver and renal function tests, serum electrolytes and thyroid status were normal.

The boy was started on saturated Potassium Iodide (KI) solution with 10 drops (500 mg) three times a day. It was gradually increased to 15 drops (750 mg) three times a day. After three months oral Itraconazole 200 mg twice daily was added. Serum electrolytes, liver functions and thyroid functions were periodically assessed. Exacerbation of acne was treated with local applications.

After 12 months of treatment, there was a considerable clinical improvement.

Discussion

Subcutaneous zygomycosis usually occurs following traumatic inoculation^{1,2}. Our patient gave a typical history; surgical correction of the nasal septum after contaminated injury. He did not develop much nasal symptoms, since the lesion had spread out to the upper lip. His fungal culture gave negative results. There are some cases reported, where the fungal studies were negative but the fungi were identified in tissue sections. Excessive tissue damage during sample collection and processing can render false negative results. The histology sections often help diagnosis. Haematoxylin and eosin, Gomori's Methenamine Sil-

ver (GMS / Grocott) and P.A.S. can be used for staining. The hyphae are broad (10-15 μ m), infrequently septate, and thin walled^{1,2}. There are focal bulbous dilatations and irregular branching at right angles, which help differentiate from other fungi¹.

Subcutaneous zygomycosis due to basidiobolus or conidiobolus can be treated with saturated KI solution (30 mg/kg/day) for 6-12 months^{3,4}. (Maximum up to 6 g/day) Oral ketoconazole, itraconazole and fluconazole can be helpful^{4,5}. Our patient was treated with combination of KI and itraconazole since there was no satisfactory improvement with KI alone. Cotrimoxazole has shown some success⁴. Sometimes, spontaneous recovery occurs. Surgical debulking was not attempted for our patient since it has been found to be non curative and may even enhance the spread¹.

References

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