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Rhinoentomophthoromycosis- a case Report

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ABSTRACT: Rhinoentomophthoromycosis is a chronic, localized, subcutaneous disease of the genere conidiobolus or baccidiobolus which are members of of the zygomycetes. Infection in humans is very rare. It is characterized by painless, woody swelling of the rhinofacial.region. Here we present a rare case of zygomycosis caused by an entomopthorale called conidiobolus coronatus.
KEYWORDS: Entomopthorale, Zygomycosis, Rhinoentomophthoromycosis

I. INTRODUCTION

Zygomycosis is any infection due to any member of the Zygomycetes group which include Mucorales and Entomoptharales. The zygomycosis caused by the later is called Entomophtoromycosis, which causes subcutaneous infection.Conidiobolus and Basidiobolus are the genera causing disease.Infected tissues are invaded by broad non-septate hyphae that become surrounded by eosinophilic material in the form of zygomycosis. Conidiobolus genera causing Entomophtoromycosis, is a chronic granulomatous disease ,restricted to rhinofacial region.This is characterised by formation of polyps or palpable restricted subcutaneous or submucosal masses. Systemic involvement also rarely seen. Such type of infection is also seen in horses, termites, insects and spiders

II. CASE REPORT

Here we present the case of a 57 year old male patient. He presented with bilateral nasal obstruction for 1 year. It was initially a small swelling of right nasal cavity ,which gradually progressed to a huge swelling ,causing bilateral nasal obstruction. He had similar symptoms 6 years back, got operated and symptom-free for last 1 year. No history of fever, headache, nasal bleeding, vertigo or loss of consciousness

On examination, there is widening of the nasal dorsum. Anterior Rhinoscopy revealed a firm mass occupying the right nasal cavity being continuous with the inferior turbinate. Nasal septum was grossly deviated to left completely obscuring the left nasal cavity. Post-nasal discharge was there.

There was no systemic involvement.

Routine investigations including haemogram, blood sugar, VDRL, hepatic and renal functions were within normal limits.

X-ray of paranasal sinuses showed features of frontal and maxillary sinusitis without any bony erosion. There was a mass filling the right nasal cavity with septal deviation to left side.

Plain CT scan of the paranasal sinuses showed soft tissue swelling of nose on the right side, a retention cyst in left maxillary sinus, and hypertrophy of left inferior turbinate.

The mass was excised under general anaesthesia per nasally and sent for histopathological studies. Biopsy showed granulpmatous features with collection of Multi-nucleated giant cells in the center of which fungakl hyphae were seen showing Splendoure-Hoeppli phenomenon around the hyphae. These histological findings were consistent with Entomopthoromycosis conidiobolae.

In Fungal Culture in the Sabouraud's dextrose agar medium, rapidly growing flat cream-colored and glabrous colonies were grown.The sides of the culture tube soon became covered with conidia. Lactophenol cotton blue (LPCB) mount showed severalconidiophores, and terminal spherical conidia with villi .Based on these features, the fungus was identified as Conidiobolus coronatus. A final diagnosis of Rhinoentomophthoromycosis was made.

Post-operatively, the patient was treated orally with freshly prepared saturated solution of potassium iodidein a concentration of 1 gm/ml (i.e., 1 drop = 67 mg). The dose was 30 mg/Kg of body weight in three daily divided doses for 6 months.Started with 5 drops t.i.d., and increased by 3 drops every 3 days, upto a maximum of 15 drops t.i.d. After the patient became symptomless, this was gradually tapered to a maintenance dose of 5 drops t.i.d. Before and during treatment, the patient's thyroid function test, SGPT, and serum

potassium were monitored and were found to be unaltered. At 6 months of treatment, nasal swelling and nasal block disappeared, and the patient regained his normal facial appearance.

Repeat CT scan of para nasal sinuses (PNS) done at 2 months and 5 months of treatment showed no regrowth. A nasal endoscopy at 5 months was normal. A repeat tissue fungal culture from the left inferior turbinate and adjacent nasal mucosa, did not show any growth. Hence, oral potassium iodide was stopped (total treatment duration-6 months).

III. DISCUSSION

Rhinoentomophthoromycosis (conidiobolomycosis) is a rare, chronic, localized, subcutaneous zygomycosis, characterized by painless, woody swelling of the rhinofacial region. It is chronic,slowly progressive. It causes severe facial disfigurement (like that of a hippopotamus). It occurs mainly in the tropical rain forests of Africa, South and Central America, and South-East Asia. A few cases have been reported from India. Adult males are more affected. It usually begins in the inferior turbinate, and spreads in the submucosa through the natural ostia to the paranasal sinus, and to the subcutaneous tissue of the face (forehead, periorbital region and upper lip). Nasal polyposis and nasal granulomas can occur. As a rule, the lesions are firmly attached to the underlying tissue, although the bone is spared. Overlying skin remains intact.One characteristic is lack of vascular invasion or infarction. The smooth rounded edge of the swelling can be demarcated by insinuating a finger underneath it. The condition is slowly progressive, but seldom life- threatening. The most common symptom is a unilateral nasal obstruction. This fungal infection is caused by Conidiobolus coronatus (Entomophthora coronata), a mould belonging to the order Entomophthorales of the class Zygomycetes.It was first isolated in 1897, and the first human case with substantiative mycologic evidence was reported by Bras et al in 1965.The fungus lives as a saprophyte in soil humus and on decomposing plant matter in moist, warm climates.It can also parasitize certain insects and frogs. Infection is acquired through inhalation of spores, or their introduction into the nasal cavities by soiled hands. Most cases affect men with agricultural or outdoor occupations.Even if the diagnosis is obvious from the clinical appearance, mycological and histological examinations are essential for confirmation. Potassium hydroxide preparation of the nasal smear, or biopsy tissue from the lesion reveals broad, nonseptate, thin-walled mycelial filaments. In Sabouraud's dextrose agar (SDA) medium, colonies of Conidiobolus coronatus grow rapidly. Histopathological features of the biopsy specimen include fibroblastic proliferation, chronic granulomatous inflammatory reaction, and broad thin walled hyphae.The Splendore-Hoeppli phenomenon (hyphal elements, in the tissue being surrounded by an eosinophilic sleeve) may be seen. Periodic acidschiff 's (PAS) stain is useful to demonstrate the fungal hyphae. Treatment of rhinoentomophthoromycosis is difficult because the diagnosis is usually established late, but patients often respond to oral itraconazole (200 to 400 mg/day), ketoconazole (200 to 400 mg/day), fluconazole (100-200 mg/day), amphotericin-B, and cotrimoxazole. Of these, itraconazole and fluconazole are both effective and relatively safe. Treatment should be continued for at least 1 month after the lesions have cleared. Saturated potassium iodide solution (1 gm/ml) is useful for patients in developing countries, because of its ease of administration and low cost. It is initiated in a dose of 5 drops/day (diluted in water, milk or fruit juice), and gradually increased upto a maximum of 40-50 drops per day, as tolerated.The exact mechanism of its action is not known. Iododerma, acneiform eruption, gastric intolerance, increased salivation and lacrimation, unpleasant brassy taste, hypothyroidism etc. are the usual side effects. Combination therapy with oral potassium iodide and oral azoles, give rapid and lasting results.Surgical resection is often needed.Cryotherapy has been tried with little success. Relapse is common, even after successful treatment. Differential diagnosis of Rhinoentomophthoromycosis includes cellulitis, rhinoscleroma, lymphoma, lymphoedema, and sarcoma.

REFERENCES

[1]. Ochoa LF, Duque CS, Velez A. RhinoentomophthoromycosisReport of two cases. J Laryngol Otol 1996;110:1154-6
[2]. Thomas MM, Bai SM, Jayaprakash C, Jose P, Ebenezer R. Rhinoentomophthoromycosis. Indian J Dermatol Venereol Leprol 2006;72:296-9.
[3]. Kwpon-Churg KJ and JE Bennett,1992 Medical Mycology Lea and Fabiger
[4]. Ellis,DH. 1997,Zygomycetes,chapter 16 in Topley and Wilsons Microbiology Microbial Infections.9th Edition Edward Arnold London.247-277
[5]. Rippon JW.1998. Medical Mycology WB Saunders and Co.

[6]. Restrepo A. Treatment of tropical Mycoses. J Am Acad Dermatol 1994;31:91-102.
[7]. Hay RJ. Moore M. Mycology. In: Champion RH, Burton JI, Burns DA, Breathnach SM, editors. Rook/Wilkinson/Ebling Textbook of Dermatology. 6th ed. London: Blackwell Science Limited: 1998. p. 1277-376.