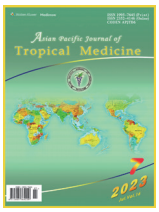

Case Report

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Rhinofacial conidiobolomycosis in an immunocompetent 30-year-old male: A case report

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ABSTRACT

Rationale: Fungal rhinosinusitis is a rare entity in immunocompetent patients and is a diagnostic challenge. Conidiobolomycosis is a rare cause of fungal rhinosinusitis which happens to affect immunocompetent patients.

Patient concerns: A 30-year-old male patient complained of painless progressive swelling of nose for 5 years and painless progressive swelling of upper lip for 4 years associated with nasal obstruction for 5 years.

Diagnosis: Rhinofacial conidiobolomycosis.

Interventions: Systemic anti-fungals and saturated solution of potassium iodide.

Outcomes: Swelling initially reduced but again increased eventually as he discontinued treatment.

Lessons: Proper adherence to drugs and need for facial reconstructive surgery may need to be considered in such cases of conidiobolomycosis.

KEYWORDS: Conidiobolomycosis; Fungus; Rhinosinusitis; Tropical; Rhino facial; Entomophthoramycosis

1. Introduction

Infections caused by fungi in the subphylum Entomophthoramycotina, called entomophthoramycosis, include both conidiobolomycosis and basidiobolomycosis. These are rare infections of the paranasal sinus and subcutaneous tissues, principally encountered in the tropics, that rarely affect other tissues[1]. Although rare, case clusters of invasive disease have been reported in both immunocompromised and immunocompetent patients[2,3]. Conidiobolomycosis affects primarily the head and face, whereas basidiobolomycosis is often localized to the subcutaneous tissues of the trunk and arms or the Gastro Intestinal tract[4,5]. The infections are characterized by slow-growing, tumor-like masses in infected tissues that can remain indolent for years. The infection is mainly prevalent in adult males predominantly among those living or working in tropical rain forests of Western Africa, South and Central America as well as Southeast Asia[4]. The disease occurs in tropical Africa, India, Puerto Rico, Columbia, and Brazil. Entomophthoramycosis also demonstrates some age specificity: conidiobolomycosis is uncommon in children, but 88% of basidiobolomycosis cases occur in patients younger than 20 years[6]. Subcutaneous rhinofacial conidiobolomycosis is the most common manifestation of infection caused by *Conidiobolus coronatus*. Symptoms typically begin with nasal discharge, epistaxis, unilateral nasal obstruction, sinus tenderness, and extensive and persistent facial swelling that may result in disfiguration. The infection slowly progresses with granulomatous inflammation in the subcutaneous tissue without bone involvement or ulceration of the skin[7]. Systemic symptoms are rare, but disseminated conidiobolomycosis has been observed[4]. We here present a case of conidiobolomycosis forming centrofacial deformity in a 30-year-old male farmer after taking proper consent from the patient and approval from ethical committee.

2. Case report

A 30-year-old male farmer from West Bengal presented with

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painless progressive swelling of nose for 5 years and painless progressive swelling of upper lip for 4 years associated with nasal obstruction for 5 years. He had no history of human immunodeficiency virus infection, renal or any chronic disease, immunosuppressive drug intake, or trauma (Supplementary Figure 1).

The patient initially had a left sided swelling in nasolabial area since February 2017. He was initially diagnosed as nasolabial cyst which was excised for histopathological examination (HPE). HPE showed features suggestive of chronic granulomatous inflammation favouring fungal infection. He was provisionally diagnosed as reparative granuloma of nose and discharged with daily oral itraconazole 200 mg twice daily. He came to us as he developed swelling of left nasolabial area as well as entire nose for 2 months. He was having nasolabial mass. HPE results of the excised mass suggested epithelioid granuloma as well as Splendor Hoeppli phenomena. He was provisionally diagnosed as recurrence of reparative granuloma with most probably *Conidiobolus* infection. He was treated with amphotericin B deoxycholate 50 mg daily for 15 days, followed by 150 mg of liposomal amphotericin B for 5 days. His swelling over nose reduced and he was discharged with tab itraconazole 100 mg twice daily and fluconazole 200 mg twice daily. He was left to follow up since 2018 January and did not take any medications for almost 1 year.

He presented to Department of Dermatology in All India Institute of Medical Sciences Bhubaneshwar in October 2019 and complained gradually progressive hard swelling over the nose and upper lip area. On examination, a 3 cm woody hard nasal mass was found fixed to surface, showing vessel prominences obliterating left nasal cavity. Firm to hard mass involving upper lip and central face around upper lip was found which was non tender. A skin biopsy was done from the nose. The HPE showed broad aseptate fungal hyphae surrounded by eosinophilic material, indicating Splender Hopple phenomenon. He was advised to continue itraconazole 200 mg twice daily along with oral saturated solution of potassium iodide (SSKI) 23 drops thrice daily to be increased by 1 drop/day till 30 drops thrice daily.

He was reviewed at All India Institute of Medical Sciences, Bhubaneshwar, on 31 January 2020. The mass size again started increasing. He was advised oral itraconazole 200 mg twice daily for 2 months and SSKI 40 drops thrice daily for 2 months. Then he again reviewed after 2 months on March 2020. He was advised to continue itraconazole 200 mg twice daily. He was also advised to increase SSKI drops to 43 drops thrice daily. He was asked to review after 3 weeks but he was left to follow up for 2 years due to COVID-19 pandemic. He did not take medications regularly during March 2020 to March 2022.

When reviewed to us on March 2022, his nose swelling was huge and upper lip swelling was also increased. Non contrast CT of Para nasal sinuses done on March 2022 showed soft tissue mass over nose along with maxillary and ethmoidal sinusitis with hypertrophy of inferior nasal concha. A biopsy from upper lip was done and sent for HPE and fungal cultures. HPE showed epithelioid granuloma and Grocott's methenamine silver stain and Periodic Acid Schiff stain showed occasional broad fungal hyphal elements within granuloma surrounded by eosinophilic material (Figure 1). Fungal stain by 10% KOH showed occasional aseptate hyaline fungal hyphae. However, the morphological identification of the fungus could not be definitely confirmed as molecular identification methods were not available in our study setup. Fungal culture was attempted by Sabouraud dextrose agar and potato dextrose agar without cycloheximide at 25-35 ℃ but it showed no growth. He was started on amphotericin B deoxycholate 1 mg/kg/day for 23 days and then as his creatinine was increasing and hypokalaemia was developing, he was given liposomal amphotericin B 5 mg/kg/day for 14 days. He also received itraconazole 200 mg 1 tab twice daily and SSKI 15 drops thrice daily. Still his swelling did not reduce and he was referred for reconstructive facial surgery.

3. Discussion

Conidiobolus spp. are generally more resistant to systemic antifungals than *Basidiobolus* spp[8]. In the older literature agents used to treat entomophthoramycosis included potassium iodide, trimethoprim and sulfamethoxazole, ketoconazole, itraconazole, and amphotericin B with varying success and clinical outcome[5,9]. At present, the preferred drugs for rhinofacial conidiobolomycosis

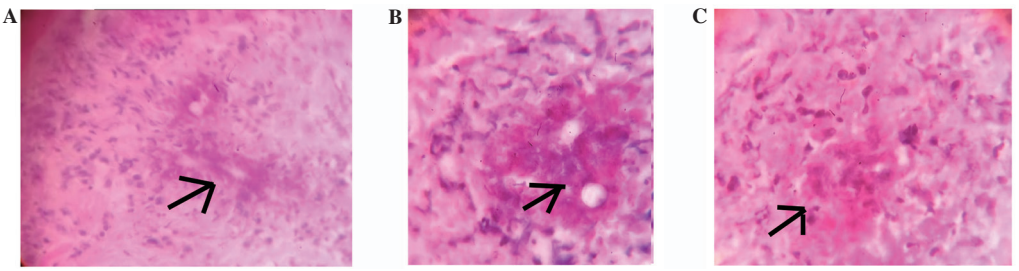

Figure 1. Hematoxylin-eosin staining showing epithelioid granuloma. A. Fungal hypahe surrounded by eosinophilic material (10×), arrow showing Splender Hoeppli phenomenon; B. Epitheloid granulpma (10×), arrow showing area of necrosis surrounded by lymphocytes and macrophages; C. Epitheliod granuloma (10×), arrow showing area of necrosis surrounded by lymphocytes and macrophages.

appear to be a combination of a saturated solution of potassium iodide and itraconazole[9]. Surgical removal and reconstructive surgery for grossly swollen or disfigured tissues, combined with medical therapy, often provides the best chance for complete recovery. However, case series from previous literature showed that surgery alone without antifungals offered no cure. Even some literature reported that surgical intervention may help in spread of the disease. However, atypical cases such as those having highly elevated infectious parameters (*e.g*., leukocytes, C-reactive protein, and erythrocyte sedimentation rate) which is uncommon otherwise in this disease have shown to benefit from surgery like orbital decompression to avoid possible complications. Adjuvant surgery after starting antifungal treatment could be an option in early disease as resection of infected tissue might contribute to improved outcome[10].

In this case, we have treated the patient with oral itraconazole and SSKI as well as systemic amphotericin B but still the swelling has not shown much regression. This might be due to inadequate response due to poor adherence or resistance to itraconazole, further studies are needed in this aspect. Given the controversy regarding definite role of surgical intervention in cure of this disease, and the disease not showing atypical features may be the possible reason of deferring the decision of surgical intervention in this case. However, the role of reconstructive surgery needs to be highlighted more in future as in this case.

Conflict of interest statement

The authors declare that there is no conflict of interest.

Declaration of patient's consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patients has given hisconsent for his images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Authors' contributions

SC took the case history, developed theoretical formalism and edited the manuscript. Both SK and SC finalised the manuscript. SK supervised the case.

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