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Case Report

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A facial deformity treated, non-surgically - A case report

Abstract

Facial deformities are a major cause of loss of self-esteem in patients. Treatment usually involves surgical correction. Medical treatment is rarely possible if the cause is infective and curable with oral medication. We report a case of Conidiobolomycosis in a 43 years old agriculturist from rural part of Eastern India who presented with a significant facial deformity and incessant sneezing and nasal discharge. His biopsy revealed the fungus which resolved with oral medications.

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Introduction

Facial deformities always take a toll on the self- esteem and confidence of a person, whichever strata of society he/she may belong to. These could be congenital or acquired. There are three major categories of acquired facial deformities. Firstly, deformity due to trauma, such as road traffic accidents, sports injuries, falls, facial burns, gunshots, attacks by animals or another human being etc. Secondly, deformity caused by post-surgical excision of a benign or malignant tumor of the face and facial organs. Thirdly, there could be deformity due to diseases like Duchenne's muscle dystrophy, rheumatoid arthritis of the mandible, idiopathic resorption of facial bones, facial hemi-atrophy etc. We report a case of facial deformity due to a fungal infection, in a male patient from the rural set-up in Eastern India.

Infections caused by fungi in the phylum Zygomycota called entomophthoramycosis, include both Conidiobolomycois and Basidiobolomycosis.[1]

Conidiobolomycosis is a rare, chronic, localized infection involving the subcutaneous tissues of the nose and paranasal structures of patients residing in tropical and sub-tropical regions. *Conidiobolus coronatus* is the most important species that causes infections in humans.[2]

In Immunocompetent patients, the disease frequently presents as a hard, painless swelling with features of localized obstruction/compression. In immunocompromised as well as some immunocompetent patients, disseminated form of infection has rarely been reported.[2] The mode of transmission is believed to be inhalation of spores, or inoculation of wounds.[3] The clinical features often mimic malignancy or subcutaneous granulomatous disease. The lesions cause localized mucosal inflammation, which in turn causes pressure symptoms like nasal obstruction, discharge, epistaxis. If the mass is large enough it leads to facial deformity.[4] The infection has a predilection to spread to surrounding structures such as paranasal sinuses, facial soft tissues, intracranial region, mediastinum or lungs. An early diagnosis and accurate treatment is therefore mandatory to prevent complications due to contiguous spread.[5]

The definitive diagnosis is based on evidence of fungus by culture of tissue biopsy or by histopathological examination (HPE) from the lesion.[6,7,8] Treatment involves antifungal therapy. Surgical debridement is required only when antifungal therapy fails or there's a recurrence of infection.[5]

Case presentation

A 43 year old male patient came with chief complaints of swelling over the nose for 4 months associated with sneezing and nasal discharge intermittently not associated with fever, or any history of nasal trauma. He was a known diabetic and hypertensive on regular treatment. The patient's routine investigations were normal. Hence, we proceeded with CT PNS imaging which revealed involvement of the maxillary sinus. Maxillary exploration was done and material sent for histopathological examination and fungal microscopy and culture. HPE showed broad, pauciseptate, hyaline fungal hyphae surrounded by Splendore-Hoeppli phenomenon with features suggestive of Zygomycotic infection. 10% KOH mount revealed broad pauciseptate fungal hyphae with large spores. Culture revealed the growth of *Conidiobolus coronatus*. The patient was started with syrup SSKI, initially 5 drops three times a day then escalated to 30 drops, and combined with Tab. Itraconazole 200 mg/day, for 6 months. Serial routine investigations including thyroid profile were done. The patient was followed up. After completion of treatment, he showed complete resolution of symptoms (sneezing, and nasal discharge) with significant decrease in nasal swelling. The patient is still under follow up and there's no recurrence of the swelling six months after completion of treatment (Figure 1-4).

Figure 1 The paranasal deformity of patient involving the cheeks, forehead and nose leading to periorbital swelling- image before treatment.

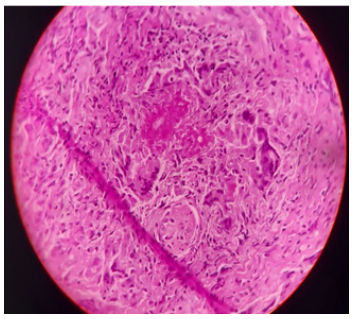

Figure 2 H&E stain on HPE examination showing Splendore-Hoeppli phenomenon.

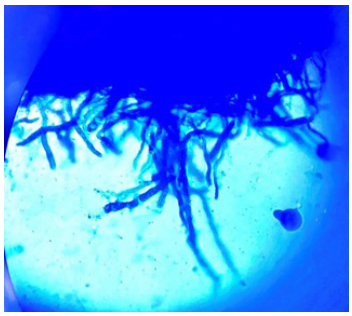

Figure 3 LPCB mount SHOWING GROWTH OF C.coronatus with beak like projection on macroconidia.

Figure 4 The swelling reduced considerably with oral therapy. Patient on follow-up, after 6 months.

Discussion

Conidiobolus spp. are saprophytic fungi found ubiquitously in soil, and decomposing plant and animal matter in warm tropical and subtropical climatic regions of west Africa, South America and southern parts of India. *C.coronatus* happens to be an insect pathogen. Out of the 27 identified species, only three (*C. coronatus, C. incongruous, C. iampranges)* are known to infect humans, the commonest being *C. coronatus* infecting mainly immunocompetent hosts. The other two species have been reported as disseminated infections amongst immunocompromised hosts such as HIV patients or renal transplant recipients.[7,9,10]

As the head and neck are the commonest sites of disease, it's hypothesized that inhalation of spores or surface inoculation of minor injuries or nasal mucosa could be the mode of transmission of this saprobe occurring via soiled hands, fomites or even insect bites.[9]

This infection has a male predilection, with a male to female ratio of 10:1. Reason being occupational exposure in men during outdoor activities such as farming, gardening or soil tilling.[10]

The differential diagnosis includes conditions like rhinoscleroma, lymphoma, sarcoma, lymphedema, angiolymphoid hyperplasia with eosinophilia and kimura disease.[11]

On histological examination, by Haematoxylin-eosin staining, the fungal elements are seen surrounded by an amorphous eosinophilic material, with an adjacent stroma consisting of inflammatory cells, mainly eosinophils. This is called the Splendore-Hoeppli phenomenon and is believed to be antigen- antibody precipitate formed around the fungal nidus.[2]

Majority of conidiobolomycosis cases are negative on fungal cultures as the aseptate/pauciseptate hyphae get damaged easily and become non-viable during the biopsy collection procedures or in the laboratory while processing for inoculation.[12] Culture media such as Sabouraud's Dextrose Agar (SDA), potato dextrose agar (PDA) and corn meal agar are used for culture of *Conidiobolus* spp. that are rapidly growing fungi at 37°C. Our case was positive on culture with typical beak-like macroconidia and pauciseptate hyphae. Colonies are initially white and later change into beige or brown with waxy appearance. With further aging, they become folded and powdery.[13]

The Lactophenol cotton blue (LPCB) mount of the colony demonstrates pauci-septate, broad, thin-walled hyaline fungal hyphae, with unbranched conidiophores that produce large, round to pyriform, single-celled macroconidia, with a protruding papillae on their wall, giving the typical appearance of a bird's beak.[8]

The treatment regimens that have been tried with varying results include SSKI (Potassium Iodide Oral solution) along with oral Azoles like Ketoconazole (200-400mg/d), Itraconazole (200-400mg/d), Fluconazole (100-200mg/d); Miconazole, and Voriconazole. Terbinafine, Amphotericin B, Cotrimoxazole and Hyperbaric oxygen have also been tried with varying efficacy.[14]

Itraconazole or fluconazole along with SSKI (1gm/ml) is useful for patients in developing countries, because of ease of administration, low cost, high clinical efficacy and fewer side effects. SSKI is initiated as 5 drops/day (diluted in water/milk/fruit juice) and gradually increased up to a maximum of 40-50 drops/day or the maximum tolerated dose. The probable mechanism of action of SSKI could be its role in phagocytic killing promotion by helping in the respiratory burst within phagolysosomes. Its side effects include acneiform eruption, nausea, vomiting, increased lacrimation/ salivation, hypothyroidism and unpleasant metallic taste.[15]

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Combination therapy with oral azoles and oral Potassium Iodide solution (SSKI) gives early and satisfactory remission.[16] Treatment should be extended for three months beyond lesion clearance. Relapse isn't uncommon after complete treatment. In such cases surgical excision along with oral steroids may have some role.[17]

Conclusion

Conidiobolomycosis is a treatable cause of facial deformity, provided an early diagnosis is made. This is possible only when an infectious cause is on the list of differential diagnosis. Occupational history should provide an adequate clue. A histopathological examination is the backbone of definitive diagnosis. Treatment should be started as soon as an HPE report is available as cultures may take time and are most often negative. A prompt treatment goes a long way in preventing complications of contiguous spread. By increasing disease awareness, we hope there would be frequent reporting of this medically treatable cause of facial deformity.

Acknowledgments

None.

Conflicts of interest

The authors declare there is no conflicts interest.

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