

CASE REPORT

Hyalohyphomycosis of maxillary antrum

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ABSTRACT

Fungal infection of the paranasal sinuses is an increasingly recognized entity, both in normal and immunocompromised individuals. The recent increase in mycotic nasal and paranasal infections is due to both improved diagnostic research and an increase in the conditions that favor fungal infection. Although fungal infections of the paranasal sinus are uncommon, 3–5% of incidence is reported. *Aspergillus*, *Candida*, and *Mucor* species are the most common causative agents of fungal sinusitis, but infection with lesser known species have been reported across the world infrequently. This article reviews and presents a case report of chronic fungal sinusitis in an immunocompetent adult male infected with two species of Hyalohyphomycosis group namely, *Paecilomyces* and *Scopulariopsis* which are opportunistic soil saprophytes, uncommon to humans.

Key words: Fungal sinusitis, hyalohyphomycosis, mycosis, opportunistic infection

INTRODUCTION

Fungal infection of the paranasal sinuses with uncommon species is an increasingly recognized entity, both in normal and immunocompromised individuals.^[1,2] The pattern of fungal infection of the sinuses can be divided into invasive, noninvasive, and semi-invasive based on the presence of fungal hyphae in the tissue with associated granulomatous reaction or tissue necrosis [Table 1].^[3] Although fungal infections of the paranasal sinus are uncommon, 3–5% of cultured sinus samples are positive for fungi.^[4] *Aspergillus* species are the most common causative agents of fungal sinusitis followed by *Candida* and *Mucor*.^[5] Hyalohyphomycosis (Gk. Hylos—glass) is an infection caused by a number of species in which the basic tissue form of fungi is hyaline without any pigment in their cell wall. Important human pathogens of this clinical entity are *Pseudallescheria*, *Fusarium*, and *Penicillium* species. The disease may present as cutaneous, subcutaneous or systemic infection in the immunocompetent or immunocompromised patients.^[6] This article presents a case report of maxillary sinus hyalohyphomycosis caused by two opportunistic soil saprophytes of *Paecilomyces* and *Scopulariopsis* species.

CASE REPORT

A 40-year-old adult male reported to the outpatient department with the chief complaint of a firm mass in the left cheek region. The duration of the swelling was approximately 6 months and the growth was continuous, slow, and painless. The patient was a farmer by profession and had no history of illness. On further probing, the patient gave the history of decay and infection in the left maxillary second molar a year ago. He underwent an unwarranted and irregular course of antibiotics and other drugs for almost 2–3 months prescribed by traditional practitioners in his village followed by extraction. The swelling appeared on the left cheek after approximately 2 months of extraction.

Clinical examination

On examination, the swelling was firm, nontender, sessile, mobile and oval in shape. It measured 4 × 3 cm and was not fixed to the surrounding soft tissue. There was no paraesthesia over the distribution of infraorbital nerve and the overlying

Table 1: Classification of fungal sinusitis

Invasive	Noninvasive	Semi-invasive
1. Granulomatous	1. Allergic fungal sinusitis (AFS)	Noninvasive disease with bone destruction
2. Acute fulminant	2. Fungus ball	
3. Chronic invasive: Can also be seen in immunocompetent patients		

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skin was unaffected. Intraorally, the left maxillary teeth were vital on pulp testing with no periapical radiolucency in dental radiographs.

Radiology and clinical diagnosis

An occipito-mental view (OMV) of skull showed generalized opacification of the left maxillary sinus. The computerized tomography (CT) scan showed evidence of well-defined, homogeneously enhancing soft tissue mass lesion seen in the left maxillary sinus [Figures 1 and 2]. No evidence of any necrosis or calcification was seen. The lesion was causing lytic destruction of anterior wall of the sinus and also the erosion of left floor of orbit. However, no intraorbital/intracranial extension was noted. Nasal septum was deviated mildly toward the right with relative inferior turbinate hypertrophy. Routine hematological evaluation showed mild leucocytosis with eosinophilia and an increased erythrocyte sedimentation rate (ESR) [Table 2]. The chest x-ray was normal. A differential diagnosis of a chronic sinusitis, benign cyst or tumor, and maxillary antrum mycosis was made, and an incisional biopsy was performed under local anesthesia. The pathological tissue was creamish in color, firm in consistency, and hyalinized in texture [Figure 3].

Histopathological and microbiological findings

The histopathological picture showed diffuse areas of fibrosis and granulation tissue with patchy infiltrate of acute and chronic inflammatory cells. There were few septate organisms, giant cells, macrophages, epitheloid cells, and foci of hemorrhage. The report was suggestive of a fungal granuloma. Microbiological studies included KOH (potassium hydroxide) wet mount, LCB (Lactophenol Cotton Blue) preparation, and culture on Sabouraud’s dextrose agar (SDA). Growth on SDA showed two types of colonies. One thin and white at first and then became gray tan in color and second golden colored, fast growing and powdery. Microscopic finding in the first culture showed chains of single-celled conidia produced in basipetal

succession from annellide (basocatenate). Annellide were solitary with globose, truncate, smooth, and brown colored conidia [Figure 4]. Microscopic finding of second colony showed conidiophores bearing dense, verticillately arranged branches bearing phialides. Phialides were ellipsoidal tapering abruptly into long cylindrical neck. Conidia were spherical, smooth walled, yellowish, and in long divergent chains. Chlamydospores were present in short chains, brown, sub spherical and thick walled [Figure 5]. Based on all these findings, the isolates were identified as *Paecilomyces* and *Scopulariopsis* species.

Further course of treatment

The patient was put on oral itraconazole 200 mg twice daily for a week which resulted in a significant reduction of

Table 2: Hematological values of the patient

Test	Values	Unit	Reference value
Hemoglobin	10.2	g/dl	12.5–17 (male) 12–16 (female)
BT	2–40	Mins secs	1–3 (Ivy method)
CT	7–20	Mins secs	2–8
TLC	11400	/cumm	4000–11000
DLC			
Neutrophils	54	%	40–70%
Lymphocytes	34	%	20–40%
Monocytes	04	%	02–08%
Eosinophils	08	%	01–05%
Basophils	00	%	00–02%
ESR	20	mm 1 st hr	<10 (Westergren’s method)
Fasting blood sugar	98	mg/dl	75-110
Random blood sugar	137	mg/dl	90-145
HIV Abs 1 and 2	Nonreactive	Reactive/ nonreactive	
HCV Abs	Nonreactive	Reactive/ nonreactive	
Hbs-Ag	Nonreactive	Reactive/ nonreactive	

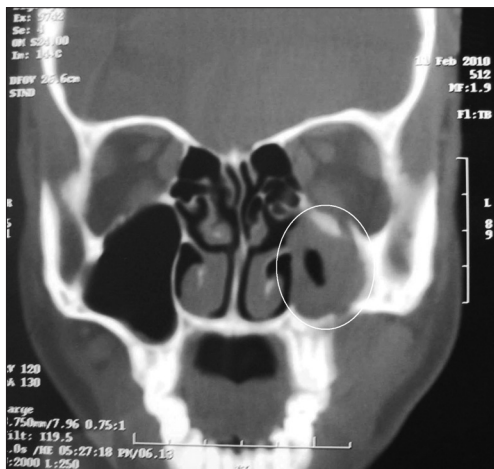


Figure 1: Coronal CT scan

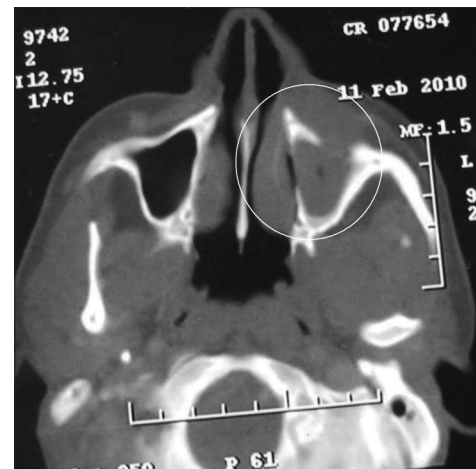


Figure 2: Axial CT scan

the extraoral swelling. A thorough sinus debridement and lavage was planned under general anesthesia. The sinus was approached by the standard Caldwell-Luc method because of the evident bony erosion of the anterior wall which had to be removed simultaneously [Figure 6]. A plastic drain was fixed orotranchally for irrigation after sinus clearance. In the postoperative phase, the patient was again put on oral itraconazole 200 mg twice daily and the sinus was irrigated with 1% acetic acid once daily for 7 days. The patient is been followed regularly for the last 1 year and a check OMV x-ray of skull confirmed repneumatization of the left maxillary sinus with complete resolution of clinical sign and symptoms.

DISCUSSION

Chronic indolent fungal sinusitis can occur in both immunocompetent and immunocompromised individual and was first described by Hora in 1965.^[7] It is mostly invasive and is characterized histologically by a chronic granulomatous inflammation surrounding broken fungal hyphae. This type of fungal sinusitis may extend beyond the bony confines of the sinuses to the orbit or even to the anterior cranial fossa.



Figure 3: Pathological specimen



Figure 5: Pictomicrograph showing *Scopulariopsis species* (KOH $\times 10 \times 40$ magnification) having basipetal globose to pyriform conidia on annellides (solitary and in groups)

Therefore, clinically it may mimic a malignant neoplasm, Wegener's granulomatosis, osteomyelitis, tuberculosis, and rhinoscleroma. Paranasal sinus mycoses are common in north India, northern Sudan, and south-western states of the North America which have warm and humid climate.^[8]

Saprophytic fungi are ubiquitous and omnipresent in nature, mostly surviving on dead organic plant matter. Some species survive as commensals within the body cavities of humans, including the nose and paranasal sinuses.^[9] When a favorable situation (immunocompromised status) arises as in prolonged steroid or antibiotic therapy or postsurgical exposure of raw areas, these dimorphic fungi metamorphose into invasive forms and present themselves in a panoramic spectrum of clinical manifestations [Table 3].^[10] An unwarranted and uncontrolled misuse of drugs, especially antibiotics, steroids, and antihistamines, in semi-urban and rural population is an important factor to contribute to this disease in developing countries such as India. The maxillary and ethmoid sinuses are more commonly involved because drainage depends on mucocilliary propagation in these sites.



Figure 4: Pictomicrograph showing *Paecilomyces species* (KOH $\times 10 \times 40$ magnification) having elongated and tapering phialides with ovoid conidia in basipetal succession

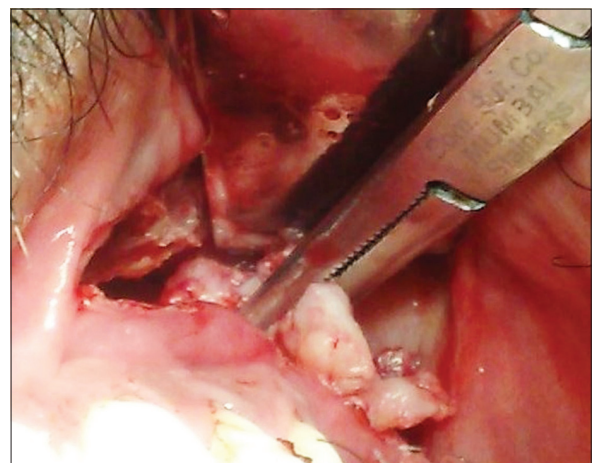


Figure 6: Pathological tissue removed from the sinus

Table 3: Criteria for sinonasal fungal infection

Major	Minor
1. Suggestive radiological evidence of invasive infection in the sinuses (i.e., erosion of sinus walls or extension of infection to neighboring structures, extensive skull base destruction)	1. Upper respiratory symptoms (nasal discharge, stuffiness, etc.) 2. Nose ulceration or eschar of nasal mucosa or epistaxis 3. Periorbital swelling 4. Maxillary tenderness 5. Black necrotic lesions or perforation of the hard palate

During the recent decades, paranasal sinus mycosis has been recognized more frequently in different parts of the world mainly because of the following reasons: (1) increased awareness among clinicians; (2) improved diagnostic tools; and (3) increased host susceptibility.^[11,12] Successful treatment of such indolent mycotic infections largely depends on the accurate identification of the pathogen and early, appropriate intervention by surgical debridement and sinus ventilation, supported with antifungal medications as per standardized regimen.

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