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 sinuses 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 | | |
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               |
| Neutrophils       | 54     | %        | 40–70%                   |
| Lymphocytes       | 34     | %        | 20–40%                   |
| Monocytes         | 04     | %        | 02–08%                   |
| Eosinophils       | 08     | %        | 01–05%                   |
| Basophils         | 00     | %        | 00–02%                   |
| ESR               | 20     | mm 1st 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    |
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 oroantrally 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 has 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. 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. 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.

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. When a favorable situation (immunocompromised status) arises as in prolonged steroid or antibiotic therapy or postsurgical exposure of raw areas, these dimorphic fungi metamorphosize into invasive forms and present themselves in a panoramic spectrum of clinical manifestations [Table 3]. 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 mucociliary propagation in these sites.

Figure 3: Pathological specimen

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

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

Figure 6: Pathological tissue removed from the sinus
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. 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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