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Research Paper

A Case Report of Chronic Invasive Fungal Rhinosinusitis

Medical Science

KEYWORDS : *Mucormycosis,* Chronic invasive fungal rhinosinusitis, Diabetes mellitus

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ABSTRACT

Chronic invasive fungal rhinosinusitis continues to present major diagnostic & therapeutic challenges to the clinician. Although its incidence has increased over the years, still most of the cases remain undiagnosed. A high index of suspicion and good diagnostic tools are required for its early diagnosis so as to provide the clinician ample opportunity to intervene earlier in the course of disease. Delay in the diagnosis leads to severe complications which are difficult to treat. The most important point to consider for the management is to establish the role of surgery and anti fungal agents. Here we report a case of chronic invasive fungal rhinosinusitis caused by Mucor.

INTRODUCTION

Fungal rhinosinusitis once considered as a rare disorder, is being recognized and reported with increasing frequency over the last two decades. Amongst all sinuses most commonly involved sinus is ethmoidal sinus. The disease is broadly categorized into invasive & noninvasive forms. Invasive fungal rhinosinusitis can be categorized as acute, chronic invasive and chronic granulomatous forms. Noninvasive form includes allergic fungal sinusitis and fungus ball. Fungal sinusitis has emerged in modern times because of increased travel into and out of endemic areas, immunodeficient states such as AIDS, immunosuppression from transplantation, chemotherapy and use of long term broad spectrum antibiotic therapy. Poorly controlled diabetes is another common predisposing factor. Chronic rhinosinusitis in immunocompetent individuals is being recognized with increasing frequency due to increased awareness and better diagnostic techniques.

Chronic rhinosinusitis accounts for more than 90% of all cases of rhinosinusitis, has a slow protracted course, and has different aetiologies, fungal infections being a major cause[1]. The most common fungal pathogens are *Aspergillus* & *Mucor* species. Apart from these dematiaceous hyphomycetes, *Pseudallescheria boydii*, *Candida spp*., *Fusarium spp*., hyalohyphomycetes and other members of *Zygomycetes* are also reported from some cases. Histopathology is important to distinguish the invasive from the non-invasive type and classify the disease. Direct microscopy and culture helps in establishing the aetiology[2]. In India, the disease once considered prevalent only in North India is now reported from other parts of the country as well. Thus, early diagnosis and accurate classification of fungal rhinosinusitis may help in deciding the treatment protocol and preventing multiple surgical procedures and can lead to effective treatment.In this paper we report a diabetic patient who was diagnosed with chronic ethmoidal fungal sinusitis due to Mucor.

CASE REPORT

A 52yr old diabetic female presented in the OPD with headache, fever, facial numbness, serosanguinous nasal discharge, postnasal drip and decrease in visual acuity. On history the patient revealed similar episodes in the past for which she was given amoxyclav to which she had responded. However in the current episode there was no response even after two weeks of treatment with antibiotics. On Physical examination the temperature was 101°F, blood pressure 100/70 mm Hg, pulse 92/min and respiration rate 14/min. There was marked tenderness just below the eyes and the extraocular muscular movements were limited. There was no proptosis. Visual acuity was decreased. Examination of the heart, abdomen, and extremities revealed no abnormalities. Patient was immediately sent for CT scan for sinuses which revealed hypoattenuating mucosal thickening or an area of soft-tissue attenuation within the lumen of the ethmoid sinus and nasal cavity. There was prominent, slightly hyperdense soft tissue shadow in the paranasal sinus with associated sinus wall erosion. CT findings were suggestive of chronic invasive sinusitis. Endoscopic sinus surgery was performed for removal of necrotic areas and ethmoidectomy. The necrotic material was sent for fungus culture and histopathological examination. Patient was put on amoxyclav and amphotericin B.

KOH mount of the necrosed material revealed presence of broad, aseptate and ribbon like hyphae. On Gomori's methenamine silver staining broad aseptate hyphae with wide angle branching were seen. Examination of hematoxylin and eosin stained sections revealed pseudostratified ciliated columnar epithelium about 3–4 cell layer in thickness. Underlying connective tissue was fibrocellular with chronic inflammatory cell infiltrate & broad aseptate hyphae were seen. These histopathological features were suggestive of fungal infection with tissue necrosis.

Fungal culture was done on Sabourads dextrose agar. Dense cottony fluffy growth was observed after three days of incubation. The growth was initially white and then became grey. Lactophenol cotton blue mount of the fungus showed the presence of aseptate hyphae, rhizoids and sporangia containing spores. The isolate was identified as Mucor. With all the above findings diagnosis of chronic invasive fungal sinusitis due to mucormycosis was made. Anti fungal susceptibility was done as per standard guidelines and mucor was found to be sensitive to itraconazole. Patient was put on itraconazole to which she responded.

DISCUSSION

The classification of fungal sinusitis is ever changing, but under the most recent and widely accepted classification, fungal rhinosinusitis is broadly categorized as either invasive or noninvasive. Invasive form is defined by the presence of fungal hyphae with in the mucosa, submucosa, bone or blood vessels of the paranasal sinuses. Noninvasive form is subdivided into allergic fungal sinusitis and fungus ball (fungal mycetoma). Invasive fungal rhinosinusitis is further classified into three clinical forms: acute invasive fungalrhino sinusitis, chronic invasive fungal rhinosinusitis and granulomatous invasive fungal rhinosinusitis. Its course may be progressive & destructive (non fulminant) or highly aggressive & lethal (fulminant) [3, 4]. The invasive

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nature of the infection can be compared with a locally aggressive neoplasm[5]. Histologically, there may be evidence of bone necrosis along with an infiltrate of lymphocytes, plasma cells, neutrophils, eosinophils, and Langhans giant cells[6, 7]. Panda et al., based on histopathological and mycological investigations categorized 178 patients diagnosed as having paranasal sinus mycoses into three disease groups- Allergic (8), non-invasive (92) and invasive (78)[8].

Once the diagnosis of invasive fungal rhinosinusitis is made, prompt intervention is warranted. Debridement of all involved tissue and bone should be performed. Adjuvant pharmacologic treatment should include amphotericin B[6]. Once fungal culture and sensitivity is available, ketoconazole or itraconazole may be substituted as per sensitivity report.

Poorly controlled diabetes mellitus is a major predisposing factor of invasive fungal sinusitis due to mucormycosis. Neutropenia is another predisposing factor, especially when neutrophil count is below 500/µl. Other main predisposing factors are long-term use of glucocorticosteroids, broad spectrum antibiotics, AIDS, leukemia, chemotherapy and graft-versus-host disease.

The prognosis is good, with prompt control of the predisposing factor and early management by radical debridement and antifungal therapy as seen in our case.

Overall about 50% of patients with invasive fungal sinusitis survive. The highest survival rates are seen in diabetic patients, the lowest being in uncontrolled leukemia, late diagnosis and extensive disease with extra-sinus tissue involvement & / or bony destruction. Brain abscess and cavernous sinus thrombosis if occurs are difficult to treat. Selecting the correct antifungal agent initially is very important, as is adequate surgical debridement. Granulocyte transfusions have occasionally been used successfully, if neutropenia is prolonged.

The therapeutic strategy must be defined according to a multidisciplinary approach (ENT, hematologist, mycologist, pathologist, ophthalmologist, neurosurgeon and anesthetist). Control of fulminant fungal sinusitis requires early recognition, aggressive surgery, systemic antifungal therapy and correction of the immunological deficits.

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