

Mucormycosis of the Frontal Sinus: A Rare Case Report and Review

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Abstract

Mucormycosis of the frontal sinus are rarely seen in day to day clinical practice. Although this fungus is commonly found in the environment, the disease is usually prevented by the immune system and is hence rare. Well-recognized risk factors for the disease include diabetes mellitus, leukemia, aplastic anemia, myelodysplastic syndrome, blood dyscrasias, and immunosuppressive therapy in organ transplantation, renal disease, sepsis, and severe burns. The disease is primarily found in those who are immunocompromised, but it may also manifest in immuno competent persons. Current therapy for the invasive disease includes early surgical debridement, antifungal therapy management of underlying predisposing factors. Early recognition of the disease and treating the underlying cause of mucormycosis, such as diabetes, are key to improving outcomes. The antifungal treatment of choice for mucormycosis is amphotericin B, although very high doses are required because of the relative resistance of the fungus to the drug. Here, we present a case of rhinocerebral mucormycosis of frontal sinus in a diabetic patient, who was managed by systemic antifungals, surgical debridement, and obliteration procedures.

Keywords: Diabetes mellitus, frontal sinus, mucormycosis

INTRODUCTION

Fungal infection of the paranasal air sinuses commonly affects the maxillary antrum, and the frontal sinus is seldom involved. The fungi however carry high mortality rate, which either may be mucormycosis (zygomycosis) or *Aspergillus* infections. Mucormycosis is ubiquitous in nature; found in soil and on decaying vegetation. It has the ability to rapidly grow and release large numbers of spores that become airborne and gain entrance to the human body through inhalation or ingestion. Given its ubiquitous nature, individuals are exposed on a regular basis; however, it rarely causes an infection in one with an intact immune system which can phagocytize the spores.^[1] In the immunocompromised patients germination and hyphae formation occurs which can spread and cause a variety of infections such as: orbitorhinocerebral infection, pulmonary, gastrointestinal, cutaneous, renal, and isolated central nervous system infections.^[2] The disease may be associated with one of the following risk factors: Diabetes mellitus, metabolic acidosis, hematological malignancy, solid organ transplant, use of chronic immunosuppressant and HIV/AIDS. Common symptoms include sinusitis, nasal stuffiness and purulent

discharge, headache, and fever. Pain may be present or absent at the time of presentation.^[3] The infection can spread to adjacent structures and cause more widespread and devastating disease. Imaging with a computed tomography (CT) or the use of magnetic resonance imaging is quiet helpful in the diagnosis of such disease. Infections carry a high mortality rate and prognosis depends on the underlying condition. Mortality rates quoted in literature range from 20% to 70%.^[4] Managing this infection is primarily surgical but requires close medical management and often a collaboration of multiple specialties.^[5]

Definitive treatment for invasive fungal disease is surgical debridement because systemic medications cannot reach the infected tissue due to vaso-occlusion. Surgical management includes aggressive debridement of all necrotic tissue, sometimes requiring multiple debridement to reduce the microbial load.

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The involved tissue rarely bleeds, so debridement until normal, well-perfused, bleeding tissue is encountered is the ideal endpoint. Reconstructive surgery should only be considered after complete recovery from infection.^[6]

Obliteration of the frontal sinus involves use of coronal flap and inferiorly based osteoplastic flap if needed.^[7] The original description consisted of meticulous debridement of the frontal sinus lining and obliterating the space with a compatible material. The procedure is mainly indicated for extensive frontal sinus infections including mucormycosis, mucocele, arteriovenous malformation, and frontal bone osteomyelitis.^[8]

It is advised that following debridement, obliteration of the frontal sinus is required to eliminate the dead space. More recently, reports advocating the use of the pericranial (PC) flap for advanced frontal sinus operations have dwelt almost exclusively on the use of the PC flap in obliteration of the frontal sinus. These flaps are basically pedicled flaps based on the supraorbital and supratrochlear vasculature.^[8,9]

We, report to a rare case of mucormycosis of the frontal sinus in a known case of type II diabetes mellitus and chronic renal disease, managed in our institution.

CASE REPORT

A 57-year-old male was referred from the Department of Internal Medicine. He reported with the chief complaint of nasal stuffiness and occasional watery discharge, pain in the forehead and pain around the right eye for 2 weeks. The history revealed that he was a known case of type II diabetes mellitus and chronic renal disease for the past 5 years. He had undergone renal dialysis three times. Maxillofacial examination revealed periorbital edema, mild proptosis, darkening of nasal mucosa, and tenderness supero medial region of the right orbit [Figure 1]. Nasal examination revealed continuous seropurulent discharge from the right nostril and sloughing of the mucosa at the region of superior meatus. CT revealed irregular hyperdense mass in the region of the right frontal sinus, without any breach of the anterior and posterior table [Figure 2]. Other paranasal sinuses were not involved. Routine hemogram revealed increased creatinine and hyperkalemia and urine examination showed proteinuria along with severe hyperglycemia (556 g/dL). Biopsy of the sloughed area over the right nostril was carried out and the specimen was subjected to histopathological examination which revealed dense aggregates to hyphae suggestive of mucormycosis. A decision for debridement and obliteration of the frontal sinus under general anesthesia was made. Preoperatively, the individual was administered intravenous anti-fungal amphotericin B (5 mg/kg/day) and other antibiotics such as ceftriaxone (50 mg/kg/day) and metronidazole (30 mg/kg/day). The frontal sinus was accessed with coronal approach and dissection was done along the loose areolar tissue, sparing the pericranium. A pericranial flap was increased separately till the approximate limit of the frontal sinus, with the help of micro-motor, bur holes were made through the outer wall of the frontal sinus and were connected with osteotomy [Figure 3],

thereby a pedicled osteoplastic flap was raised exposing the frontal sinus [Figure 4]. The sinus was multi septate and the lining was thick and infected. Dirty white patches of fungal growth were clearly visible. Thorough debridement of the frontal sinus was carried out, bony irregularities and septae were removed under copious irrigation. The opening of the fronto-nasal duct was identified, everted, and plugged. Inner walls of the frontal sinus were intact with no cranial exposure. The free-end of the pericranial tissue was inserted into the frontal sinus and osteoplastic flap was repositioned. In this case, the outer table was not fixed. Suction drain was placed and wound closure was done. Nasal irrigation and debridement was carried out. Pressure dressing was applied and postoperatively the antifungal and antibiotics were continued for next 7 days as per the institutional protocol. Renal functional tests were carried out at regular intervals. The sinus lavage was subjected for histopathological evaluation, which revealed multiple tissue bits composed of abundant noninflammatory tissue. Most of the bits showed dense mixed inflammatory infiltrate in the form of eosinophils, plasma cells, and lymphocytes. Large areas of necrosis as well as hemorrhage were also seen. Admixed with these were collections of basophilic hyphae which were aseptate and broad based along with irregular borders. A few of the bits showed fragments of pseudo stratified ciliated columnar epithelium with no signs of granuloma or angio-invasion. The histopathological examination did not reveal any signs of atypia or malignancy [Figure 5]. Periodic Acid-Schiff stain confirmed fungal hyphae of mucormycosis [Figure 6]. Hence, it was opined histopathologically as specimen from frontal sinus showing the evidence of mucormycosis. There was a dramatic improvement in the general condition of the individual. Contour of the upper third of the face was maintained postoperatively [Figure 7]. He was discharged after 15 days of observation to an outpatient rehabilitation program, where he was advised oral posaconazole (400 mg stat followed by 200 mg once daily for next 2 weeks). The glycemic control was emphasized throughout the course of the treatment and was administered Insulin lispro (0.5 unit/kg/day) once daily and Insulin glargine (1/kg/day). Regular follow up every week for 1st month and every 15 days for next 3 months and every month for a year was carried out. The individual showed no signs of recurrence [Figure 8] and did not require any further renal dialysis. However, the individual lost to follow-up after 1 year.

DISCUSSION

Management of frontal sinus involves the elimination of dead space, either by obliteration or cranialization. A wide variety of materials have been used to obliterate the sinus such as: hydroxyapatite, oxidized cellulose, absorbable gelatin sponge, acrylic, along with autogenous materials such as: Bone, cartilage, muscle, temporalis fascia, or fat (usually abdominal). Obliteration was chosen in this case as it was a viable method to enhance sealing and thus eliminating the dead space occupied by the frontal sinus.^[10]

PC flap was used in our case because of its rich vascularity and versatility, along with no morbidity of the donor site, thus eliminating the chances of infection.^[11] Furthermore, due to the



Figure 1: Preoperative frontal view



Figure 2: Axial computed tomographic image showing hyperdense area occupying frontal sinus

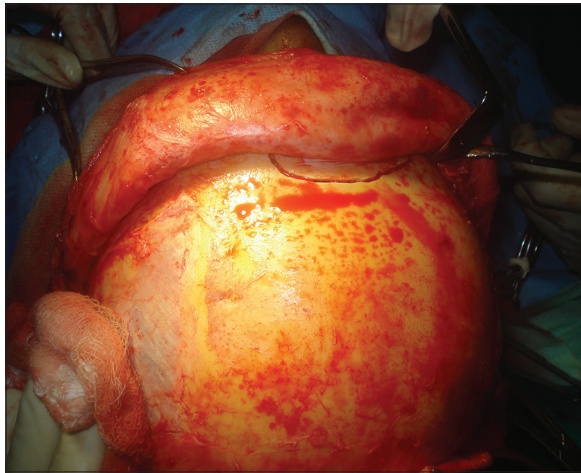


Figure 3: Coronal approach to access the frontal sinus and osteotomy cut

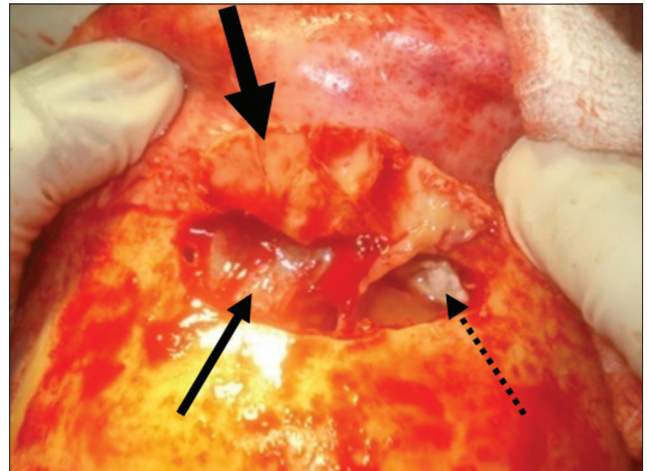


Figure 4: Frontal sinus exposure (thick arrow: Osteoplastic flap, thin arrow: Mucosal lining, dotted arrow: mucor growth)

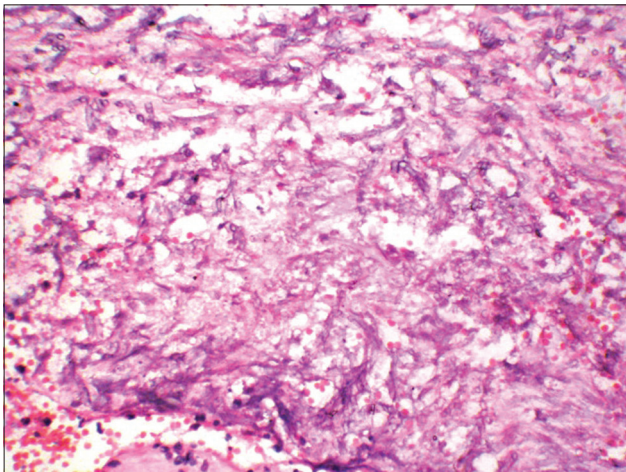


Figure 5: H and E, ×40 section showing aseptate basophilic hyphae

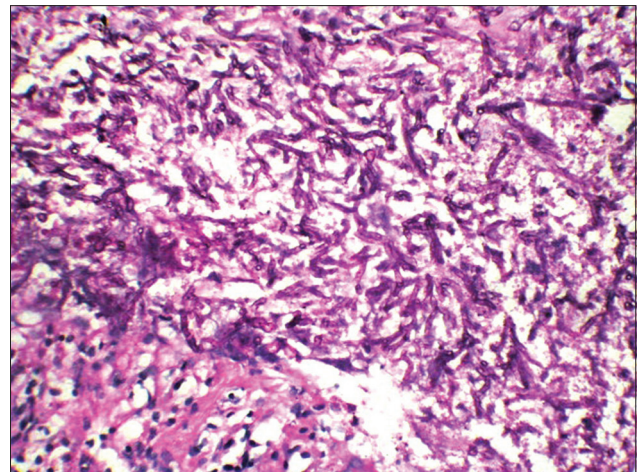


Figure 6: Periodic acid-Schiff ×40, stained section showing fungal hyphae of mucormycosis



Figure 7: Postoperative profile view

fact that we dealt with a serious fungal infection and the presence of underlying systemic condition of the case, the use of alloplastic

material was not considered as viable option.^[12] The method of harvesting the flap also varies as per clinician's need either by

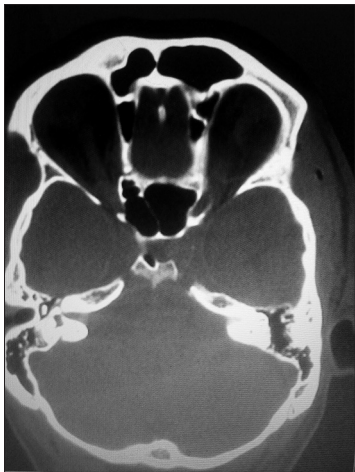


Figure 8: Postoperative axial computed tomographic image showing no recurrence

initial elevation subperiosteally, following which the pericranium is dissected from the scalp, or by elevation from the subgaleal plane, to be followed by elevation of the pericranium from the cranial bone. We preferred to elevate the flap in the subgaleal plane through a coronal incision because the flap is more easily designed and dissected from the stable platform of the calvarium as opposed to dissection from a mobile, previously elevated scalp.^[13]

Current therapy for cases of rhinocerebral mucormycosis involves treatment of underlying pathologies, antifungal therapy and surgical debridement of the affected tissues. The antifungal medication of choice is amphotericin B; although, high doses may be needed, owing to the relative resistance of the fungus to the drug and we chose to administer posaconazole postoperatively as a salvage drug.^[14,15] The individual's glycemic control was emphasized throughout the course of the treatment and continued to be managed for his systemic condition at the Department of Internal Medicine in the same institution.

The reduction of postoperative infection and early recovery may be attributed to the use of pedicled PC flap for the obliteration of the frontal sinus and our findings are coincident with those of the findings seen by Gerbino *et al.*^[16] These cases are required to be followed up for a longer duration to rule out recurrence and any associated adverse sequel.

CONCLUSION

The PC flap is easily harvested and versatile. Using this vascularized tissue during obliteration affords added protection by providing an extra barrier between the intracranial cavity and the frontal bone and sino-nasal tract. This technique is inexpensive, safe, and effective and should be considered when obliteration of the infected frontal sinus is performed.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their 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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Conflicts of interest

There are no conflicts of interest.

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