Acute Fulminant Invasive Fungal Sinusitis with Cavernous Sinus Syndrome
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ABSTRACT
Acute fulminant invasive fungal sinusitis is most commonly found in immunocompromised patients with conditions such as diabetes mellitus, malignancies and acquired immune deficiency syndrome. The most common pathogens are Aspergillus and Mucoraceae and the sinus most frequently involved is the maxillary sinus. Fever, rhinorrhea, facial pain, headache, and diplopia are common presenting symptoms. Complications of this infection include intracranial and/or intraorbital spread of the infection; the prognosis is poor. Here, a rare case of acute fulminant invasive fungal sinusitis with cavernous sinus syndrome is reported.


INTRODUCTION
Fungal infections of the nose and paranasal sinuses include infections ranging from colonization to invasive rhinosinusitis. The clinical presentation of the patients depends on the immune status of the host.1 Fungal sinusitis can be classified as invasive and non-invasive fungal sinusitis on the basis of the extent of the infection as determined by histopathology. Among the invasive fungal sinusitis infections, acute fulminant invasive fungal sinusitis most commonly affects the immunocompromised hosts. Fever, cough, crusting of the nasal mucosa, epistaxis, headache are the most common presenting symptoms. If the diagnosis is initially overlooked, intracranial or intraorbital infection may occur. The prognosis may be poor with a high mortality rate.

A 55 years old man with an acute fulminant invasive fungal sinusitis leading to cavernous sinus syndrome is reported.

CASE REPORT
A 55 years old man had a history of type-2 diabetes controlled without regular oral hypoglycemic agents for 5 years and no known family history of disease presented to the otolaryngology emergency department to seek treatment for the presence of nasal obstruction, cough with yellowish sputum, swelling of the right periorbital region, right facial pain and a tingling sensation over the right frontal and temporal region for 3 days. The patient had no history of habitual cigarette smoking and alcohol consumption.

The initial physical examination showed redness, swelling, focal tenderness of right periorbital region with proptosis and paresthesia over the right frontal and temporal regions. Fiberoptic endoscopy revealed a large blackish eschar with filamentous material over the middle turbinate of right nasal cavity (Figure 1). In addition, the ears, oral cavity, pharynx, larynx, and neck were negative for any significant findings on the physical examination. The laboratory findings showed a white blood cell count of 16.8 x 10^3 /µL, neutrophil count of 81.8%, lymphocyte count of 11.6%, a C-reactive protein of 198.7 mg/L, and sugar of 563 mg/dl. The computed tomography of the sinuses showed partial opacification over the right maxillary and ethmoid sinuses. The Magnetic Resonance Imaging (MRI) of the brain showed hypersignal intensity over the right cavernous sinus in T2-weighted images (Figure 2).

The patient underwent functional endoscopic sinus surgery with right uncinectomy, ethmoidectomy and removal of necrotic soft tissue followed by intravenous liposomal Amphotericin B, 350 mg per day (5 mg/kg/day) treatment for 2 weeks. The pathology findings of right nasal lesion revealed septate hyphae, branching with acute angles, features compatible with Aspergillosis (Figure 3). The patient was followed in the otolaryngology outpatient department for 6 months. The right ostiomeatal complex lesion healed well without obstruction or discharge. The symptoms improved without evidence of recurrence or neurological sequela.

DISCUSSION
Fungal sinusitis is an inflammation of the sinuses that is caused by many different fungi. Fungal sinusitis can be divided into invasive and non-invasive types based on the extent of the sinusitis as defined by histopathology.
Invasive fungal sinusitis can be further categorized as acute fulminant, chronic and granulomatous invasive fungal sinusitis based on the clinical manifestation of the infection. The difference between acute fulminant and chronic invasive fungal sinusitis is the duration of clinical symptoms; that is, less or more than four weeks.

Acute fulminant invasive fungal sinusitis occurs most frequently in immunocompromised patients; such as those with diabetes mellitus, malignancies, neutropenia, and acquired immune deficiency syndrome. Mucoraceae and Aspergillus are the most common organisms found in acute fulminant invasive fungal sinusitis. The early clinical symptoms of acute fulminant invasive fungal sinusitis include: nasal obstruction, rhinorrhea, facial pain, headache, proptosis, and diplopia. However, these symptoms are not specific and are consistent with acute bacterial sinusitis. This may lead to misdiagnosis or delayed treatment. Radiography can be used to help with the differential diagnosis. A unilateral nidus, severe nasal mucosal edema, or extrasinus invasion such as bony erosion may be noted on the computed tomography of the sinuses. In the pre-antibiotic era, the mortality rate of acute fulminant invasive fungal sinusitis was 50 - 80%. However, the prognosis has improved because of greater recognition and improved treatment.

Sinusitis resulting in orbital, bony, or intracranial complications is uncommon. Orbital complications such as orbital cellulitis and orbital abscess formation usually occur with ethmoid sinusitis. Bony complications such as frontal and maxillary osteomyelitis are common with frontal and maxillary sinus involvement. Intracranial complications such as meningitis, intracranial abscess formation, and cavernous sinus thrombosis usually are associated with frontal, ethmoid and sphenoid sinusitis. Most of these complications are a result of acute exacerbation of chronic sinusitis. However, fungal sinusitis complicated by inflammation of the cavernous sinus is unusual.

The cavernous sinuses are complex structure of the venous plexus that are located on either side of the sphenoid bone, posterior to the superior orbital fissure and anterior to the apex of petrous temporal bone. The internal carotid artery with its periarterial sympathetic plexus passes through the sinus. The oculomotor nerve, trochlear nerve, ophthalmic division, and maxillary division of the trigeminal nerve are located in the lateral wall of the sinus from above to below. The abducens nerve is located inferolateral to the internal carotid artery. When the cavernous sinus is involved in a neoplasm, vascular lesion, non-infectious inflammation, or infection, multiple neuropathies occur that are referred to as the cavernous sinus syndrome. The clinical symptoms include eye pain, proptosis, ophthalmoplegia, chemosis, and/or sensory impairment of regions including the forehead, eyelid, cheek, nose, and upper lip.

If fungal sinusitis results in intracranial extension, the mortality rate increases. Magnetic resonance imaging of the brain is an important instrument used to evaluate intracranial lesions. In this patient, hyper-signal intensity over the right cavernous sinus on T2-weighted images of the brain on MRI was observed. This allowed for early detection of the intracranial lesion caused by fungal sinusitis. A rapid diagnosis and the appropriate treatment aided in the good outcome of this patient.

The treatment of acute fulminant invasive fungal sinusitis with cavernous sinus syndrome should include antifungal medications and drainage of the infected sinus by surgery. Surgical management of the cavernous sinus may not be necessary. The principal antifungal medication used is Amphotericin B. In addition, debridement of the infected sinus by functional endoscopic sinus surgery is also essential. Necrotic soft tissue should be removed until a bleeding border is noted. Control of the underlying condition is important in the immunocompromised patient.

REFERENCES

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