CASE REPORT

Invasive Fungal Sinusitis with Intracranial Extension and Fatal Outcome: A Case Report

Syeda Areeba Tabassum,[1] Saba Zaidi[1*] and Rajesh Kumar[1]

[1]*Department of Neurology, Liaquat National Hospital and Medical College, Karachi, Pakistan*

Abstract

Invasive fungal sinusitis is a morbid condition usually encountered in patients with poor immunity, for instance, diabetics or those with lymphoproliferative disease. We report invasive fungal sinusitis in a young immunocompetent host. This case presents a 21-year-old man with an invasive fungal infection and intracranial extension. The patient initially complained of right-sided weakness, headache, vomiting, and diplopia and was subsequently found to have a significant post-contrast enhancement along the left temporal region, encasing the internal carotid artery (ICA). The patient was started on antifungal therapy and underwent functional endoscopic sinus surgery (FESS) which revealed the growth of Aspergillus on fungal culture. He later developed severe cardiac dysfunction and neurological deterioration, ultimately leading to death. This case highlights the significance of taking into account the importance of fungal sinusitis as a potential cause of neurological symptoms, particularly in younger age groups. Early diagnosis and appropriate management are crucial in preventing potentially fatal complications.

Keywords: *Young adult, sinusitis, internal carotid artery, mycoses, headache.*

INTRODUCTION

Invasive fungal sinusitis (IFS) is a rare and potentially fatal condition, characterized by the invasion of fungi into the paranasal sinuses, orbit, and cranial cavity. The incidence of IFS varies between different geographic regions and patient populations, with higher rates reported in immunocompromised individuals, such as those with diabetes, haematological malignancies, and organ transplants [1]. However, IFS can also occur in immunocompetent patients, especially in the presence of predisposing factors such as sinus surgery, trauma, or exposure to contaminated soil or water [2]. In this report, we present a young immunocompetent male with an acute history of invasive fungal sinusitis that resulted in fatal outcomes regardless of aggressive management and taking a multidisciplinary approach to treatment [3, 4]. The patient's initial presentation was suggestive of stroke; however, further investigations and imaging scans raised suspicion for intracranial fungal infection.

CASE REPORT

We presented a 21-year-old boy, with no known co-morbidities, who came to the outpatient neurology department complaining of right-sided weakness for one week. The patient further reported that he had headache and nasal congestion for 3 months being managed as sinusitis on nasal sprays and antibiotics. But now his condition worsened for a week. He was a non-smoker, with no history of drug abuse. The patient had gone to his primary care physician from where he was referred to a tertiary care hospital. On physical examination, he was a normal-looking boy with a normal build, vitally stable. His higher mental functions were intact, with no cranial nerve palsies. He had reduced power in his right upper and lower limb around 3/5 with left upper motor neuron facial nerve palsy. The Plantar was extensor on the right side. He was admitted to the ICU for the management of his symptoms. His haemoglobin was found to be 9.2 mg/dL on a complete blood picture, and his white blood cell count was 330,000/cu mm^3. The basic metabolic panel was unremarkable.

MRI Brain with contrast showed that the intracranial part for enhancement measured 3.5 * 2.0 * 2.5 (AP * TS * CC) with cavity compression over the temporal lobe. It also encased the left cavernous sinus and the cavernous part of the left Internal Carotid Artery. Along with these findings, a left temporal arachnoid cyst and several ischemic infarcts in the left frontoparietal region were seen. A diagnosis of fungal sinusitis was made, with intracranial extension. While MRA showed a blockade of the left MCA from its stem (**Fig. 1 a-c**).

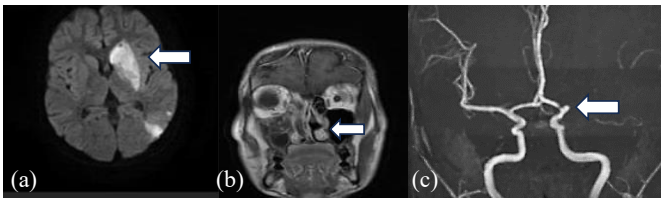

Fig. (1): (**a**) Diffusion weighted MRI sequence showing restriction in left Basal ganglia, (**b**) FLAIR Sequence showing filled maxillary and ethmoid sinuses and (**c**) MRA brain complete left MCA obstruction.

He was immediately started on Amphotericin B 10 mg/kg/day and was being continued. Infectious disease consultation was taken. The patient's chronic headaches persisted while he was hospitalized. Growth was not detected in interim blood cultures. Fungal cultures which were sent simultaneously, came out as negative.

**Corresponding author: Saba Zaidi, Department of Neurology, Liaquat National Hospital and Medical College, Karachi, Pakistan, Email: drsabazaidi@gmail.com*

Received: November 02, 2023; Revised: January 30, 2024; Accepted: February 09, 2024

DOI: https://doi.org/10.37184/jlnh.2959-1805.2.9

To further help in reaching a definite diagnosis, the otorhinolaryngology team was taken on board. Subsequently, he was advised for functional endoscopic sinus surgery, which removed a copious amount of mucopurulent fluid. The histopathology analysis of the retrieved specimen showed septate hyphae, suggesting Aspergillosis infection. Infectious disease switched him to Voriconazole 400 mg Twice daily dose from Amphotericin after five days. After FESS and sinus wash, the patient was found to be short of breath for which his cardiac enzymes were checked they were raised. His echocardiography showed an ejection fraction of 30% and severe hypokinesia. Cardiology gave the impression of fungal myocarditis. His clinical condition started deteriorating. He was taken on ionotropic support due to persistent hypotension. Later he dropped his conscious level and when assessed corneal reflexes were absent, no reaction to noxious stimuli, was fixed, his pupils on both sides were fixed and dilated, and did not react to light with the absent doll's eye movements were found. MRI brain was repeated which showed diffusion restriction in the left parieto-occipital region with a hemorrhagic component in the left Basal Ganglia and intraventricular extension. Her MRI brain also shows a flow void in the left ICA. Extension of the disease process both clinically and radiologically (**Fig. 2 a-c).** Cardiac resuscitation was started immediately but failed to achieve ROSC (return of spontaneous circulation) so death was declared.

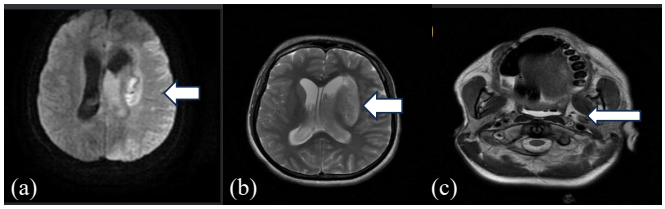

Fig. 2: (**a**) Diffusion weighted sequence showing left basal ganglia and parietal cortex heterogenous area of diffusion restriction, (**b**) T2 showing mixed signal intensity in Left basal ganglia with intraventricular extension of hemorrhage and (**c**) Left ICA flow void.

DISCUSSION

The diagnosis of invasive fungal sinusitis (IFS) with intracranial extension requires a high index of suspicion, as the initial symptoms may be nonspecific and overlap with other conditions such as bacterial sinusitis, meningitis, or stroke [1, 2]. Imaging studies, such as computed tomography (CT) or magnetic resonance imaging (MRI) provide significant information regarding the extent and severity of the disease, as well as potential complications for instance cavernous sinus thrombosis or cerebral infarction. However, definitive diagnosis often requires histopathological examination of tissue specimens, which can reveal the presence of fungal elements and guide the choice of antifungal therapy. Our case elaborates on the importance of recognizing this life-threatening fungal infection in an immunocompetent host. The literature review revealed a limited number of such cases [5, 6].

The optimal management of IFS involves a multidisciplinary approach, including infectious disease specialists, otorhinolaryngologists, neurosurgeons, and critical care physicians. The goals of treatment are to eradicate the fungal infection, control the inflammatory response, and prevent or treat associated complications such as abscess formation or cranial nerve palsies. Antifungal therapy with agents such as Amphotericin B or Voriconazole [7] is typically the mainstay of treatment, but surgical interventions such as FESS or debridement may also be necessary in cases of extensive or refractory disease. However, despite aggressive management, the prognosis of IFS with intracranial extension remains poor, with mortality rates ranging from 30% to 80%, depending on the underlying host factors and extent of the disease [8, 9].

Our case is the representation of one of the lethal complications of invasive fungal infection which is vasculitis. Aspergillus is also known for its angioinvasion and causing vasculitis. It is evident from the first MRI brain that his left MCA was completely blocked and later it extended to involve the whole left ICA. Despite proper and timely management, we were not able to prevent this drastic progression of disease. We believed that vessel invasion in our case might be due to the near location of the cavernous part of ICA from where the hyphal element travelled and caused blockade initially of left MCA and later the whole ICA or it might be fungal element got embolized in the circulation. Another phenomenon which was highlighted in the literature [10] about the Aspergillus invasion was it destroys the integrity of the vessel by digesting the elastin and it grows out of the vessel wall leading to the formation of mycotic aneurysms of large arteries leading to rupture and massive hemorrhage. In our case, the MRI brain which was repeated showed a massive hemorrhagic component in basal ganglia and intraventricular extension raising the possibility of mycotic aneurysm.

Invasive fungal sinusitis may affect patients with an intact host defense system and its worrisome complications include intracranial involvement. Our patient did not respond to timely medical management.

CONCLUTION

In conclusion, IFS with intracranial extension is a rare but serious condition that requires immediate recognition and aggressive management to improve outcomes. This

case highlights the significance of keeping a vigilant eye for IFS while assessing the patients who presented with nonspecific clinical signs and symptoms and the need for multidisciplinary collaboration while managing this complex disease.

CONSENT FOR PUBLICATION

Written informed consent was taken from the participants.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

ACKNOWLEDGEMENTS

Declared none.

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