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Case Study

Right lateral rectus palsy as a presenting feature of rhinocerebral mucormycosis infection in an immunocompetent teenager

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This study reports a case of an immunocompetent patient with right lateral rectus palsy secondary to mucormycosis infection in the sphenoid and ethmoid sinuses extending to the carvenous sinus. The patient used for this study is a healthy, well built and well nourished 14 year old girl presented with symptom of headache of 2 weeks duration. Complaint of diplopia was made on day one of admission. The patient's case is a known case of asthma, but her last attack was a year ago. Metered dose inhalers (MDI) was hardly used due to the fact that the patient is an active girl with no history of any other illnesses or hospital admissions. Clinical examination and investigations revealed that the patient had right lateral rectus palsy secondary to mucormycosis infection of the ethmoid, sphenoid sinuses which extended intracranially (cavernous sinus). The investigations carried out were done through Computed Tomography, Magnetic Imaging Resonance and biopsy of a polyp from the nasal cavity. The patient underwent endoscopic transeptal/transphenoidal approach and excision of fungal sac with a 30 day duration of intravenous amphotericin B. Resolution of the right lateral rectus palsy was noted immediately on post operative day one. Even a healthy, active and an immunocompetent young person can develop a severe fungal infection of the sinuses which extends intracranially. However, this is a very rare occurrence and it shows that a high index of suspicion is warranted in the diagnosis of an intracranial fungal infection in a person who only presented with a common headache.

Key words: Immunocompetent, lateral rectus palsy, mucormycosis infection, antifungal treatment.

CASE REPORT

This study reports the case of a healthy 14 year old girl presented with diplopia with prior history of headache of two weeks duration. Snellen visual acuity of 6/9 OU was observed in the girl, though the anterior and posterior segment findings were unremarkable. Ocular motility examination showed abduction, elevation and depression deficit of the right eye. It was observed that there were no classical clinical signs of rhino-orbital-cerebral mucormycosis such as proptosis or chemosis, in that the patient did not have preceding rhinocerebral trauma, local surgeries or chronic sinusitis. Figure 1 shows the Hess chart used for observation of diplopia presented by the patient. Routine blood tests noted an elevated white cell count of 13.4×10^9/L. A Magnetic Resonance Imaging (MRI) scan was ordered and it revealed a mass involving all the sinuses with erosion into the anterior cranial fossa. Features of mucormycosis are suggested based on findings of mucosal thickening with enhancement and aggressive bony erosions (Figure 2). An ENT referral was made and the study proceeded with an endoscopic transeptal and transphenoidal excision of fungal sac. Intraoperatively, fungal materials and sinusitis were noted in the sphenoid, ethmoid sinuses with carvenous sinus

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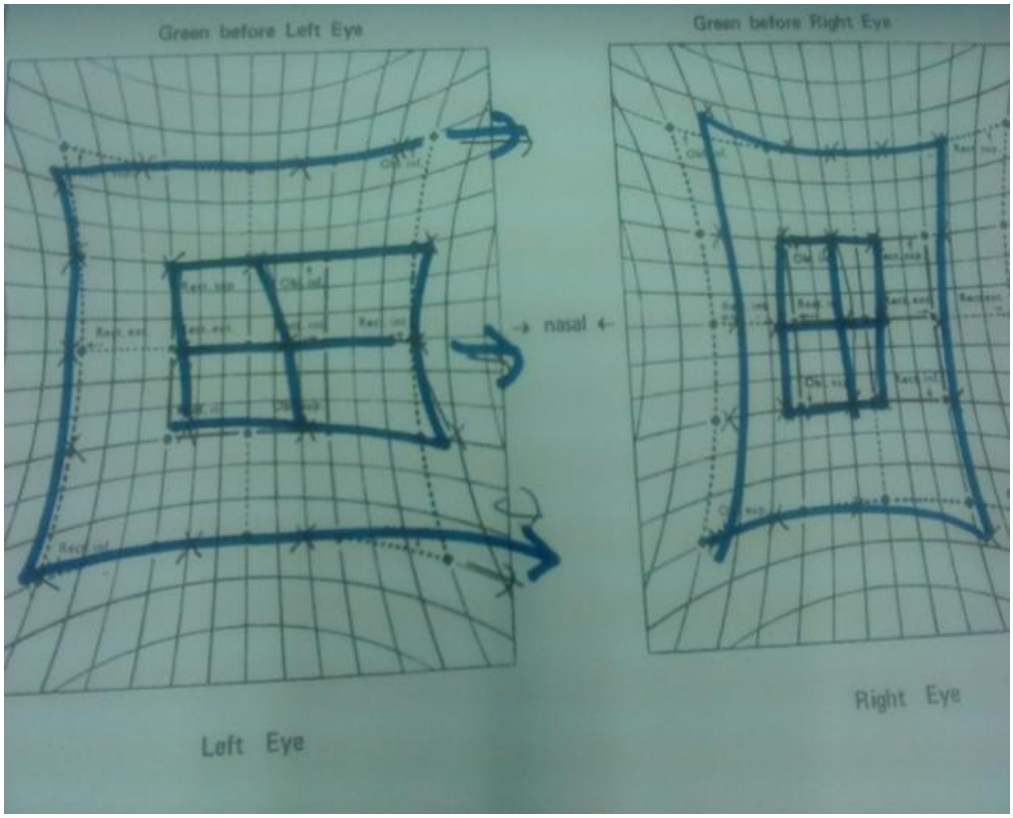

Figure 1. HESS chart at presentation.

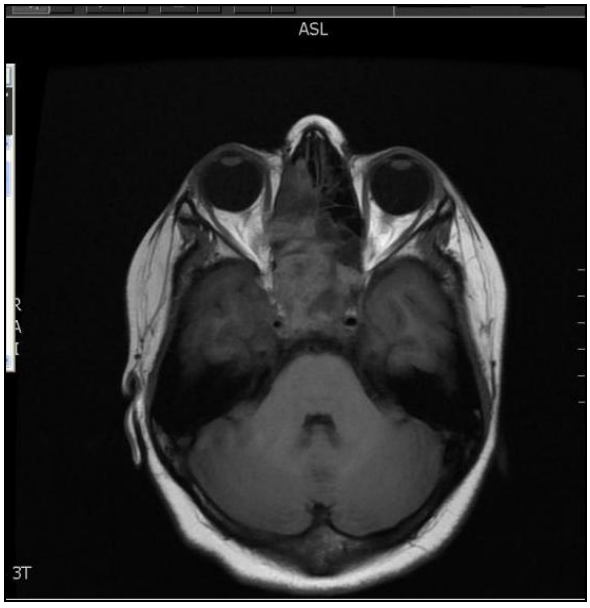

Figure 2. MRI showing diffuse mucosal thickening and enhancement with fluid noted within the sphenoid sinus extending anteriorly into the ethmoid and the right frontal sinus. The sphenoid sinus is expanded with erosion of the sphenoid bone. Possible erosions occur into the base of anterior cranial fossa with associated diffuse thickening of meninges in the right cerebral hemisphere suggestive of intracranial extension.

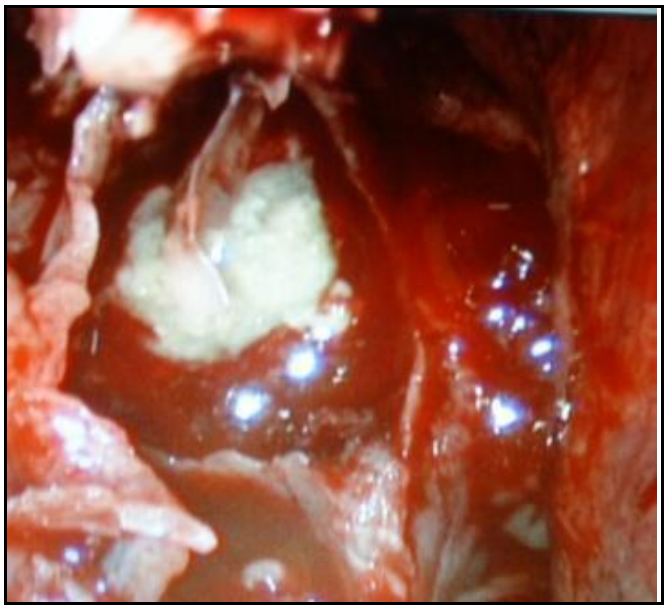

Figure 3. Yellowish, fluffy, fungal material seen intraoperatively.

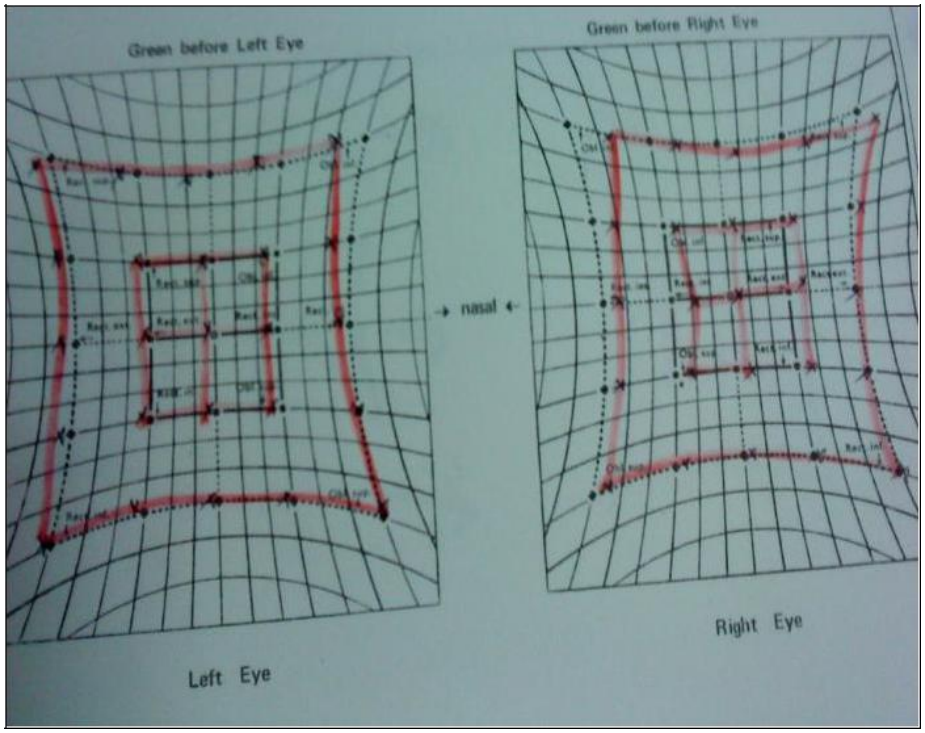

Figure 4. HESS chart post operatively showing recovery of the disease.

and sellar extension (Figure 3). Histopathological examination (HPE) and tissue culture however were negative for organisms or malignancy, due to known difficulty of the fungi to be cultured. Despite lack of conclusive evidence on HPE based on MRI and intraoperative findings, a presumptive diagnosis of mucormycosis infection was made. The patient was treated with intravenous amphotericin B for 30 days. She responded well to treatment and regained almost complete right extraocular motility, documented on Hess chart (Figure 4). The MRI which was performed at 1 month post operatively showed complete resolution of the

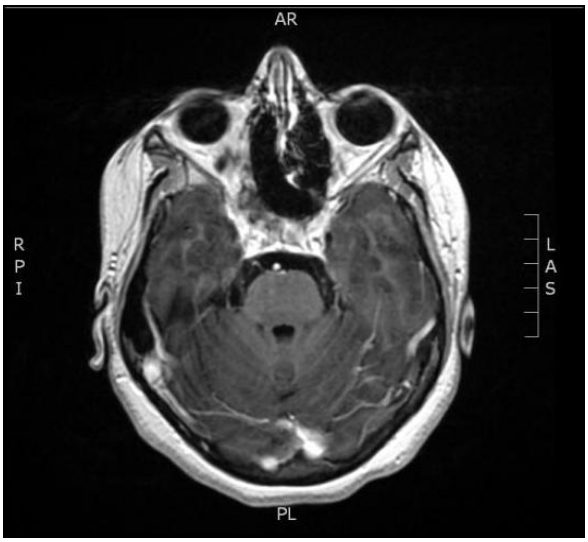

Figure 5. MRI 1 month post-operative showing resolution of the disease.

disease with no recurrence (Figure 5).

DISCUSSION

Mucormycosis infection of the sinuses (also called rhinocerebral mucormycosis) is a rare occurrence in healthy individuals. It occurs mostly in immunocompromised patients. Exposure occurs through inhalation of the spores from the environment. A spore which gets converted into hyphae becomes invasive and may track through the paranasal sinuses into the brain and/or orbits (Rumboldt and Castillo, 2002). Possible direct soft-tissue invasion leading to formation of cerebral abscesses are frequent. Any extension along the blood vessels can lead to thrombosis (Rumboldt and Castillo, 2002). In recent times, incidences of such cases of infection caused by fungal pathogens in healthy individuals are being noted (Sridhara et al., 2005; Prasad et al., 2008; Jain et al., 2011). A recent literature review revealed a worldwide distribution with India having the most reported cases (Mignogna et al., 2011). Increasing trend of reported cases are seen, with more than half reported in the last decade. Postulations are made on whether or not this points to a rise in global poverty with limited access to medical or surgical intervention. A high index of suspicion is warranted in cases of atypical headache, especially if associated with other neurological signs.

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