

Case Report

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Crossed Nerve Palsies in Sinonasal Mucormycosis: A Diabetic's Facial-Hypoglossal Puzzle

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ABSTRACT

Introduction: Sinonasal mucormycosis is an acute invasive fungal infection of the nose and paranasal sinuses, which is rare, opportunistic and potentially fatal, occurring in both normal and immunocompromised individuals. Uncontrolled diabetics and immunocompromised patients are more prone to this invasive fungal infection. Sinonasal mucormycosis is approximately 1.7 cases per 1,000,000 inhabitants per year. Incidence of VII Cranial nerve palsy is 11% in these cases, and coexistence of ipsilateral Facial nerve and contralateral hypoglossal nerve is not documented in existing literature.

Case Report: A 53-year-old woman was diagnosed with Sinonasal Mucormycosis with extension to the skull base and Infratemporal fossa with involvement of ipsilateral facial nerve palsy and contralateral hypoglossal nerve palsy, in the setting of newly diagnosed and uncontrolled diabetes mellitus. She was promptly started on Liposomal Amphotericin-B and underwent "Endoscopic trans-nasal trans-maxillary trans-pterygoid debridement".

Conclusion: Mucormycosis is a rare condition that is still fatal in the era of medical and surgical advancements. A combination of endoscopic surgical debridement followed by intravenous Amphotericin B therapy is quintessential in the management of such patients and has less morbidity compared with conventional treatment. Rare presentations like multiple cranial nerve palsy, which would otherwise raise the suspicion onwards granulomatous disease, need to be scrutinised and aspects ruled out in effective treatment of patients.

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Introduction

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Case Report

A 53-year-old woman was diagnosed with Sinonasal Mucormycosis with extension to the skull base and Infratemporal fossa with involvement of ipsilateral facial nerve palsy and contralateral hypoglossal nerve palsy, in the setting of newly diagnosed and uncontrolled diabetes mellitus. She was promptly started on Liposomal Amphotericin-B and underwent "Endoscopic trans-nasal trans-maxillary trans-pterygoid debridement".

Medical Intervention

The patient was treated with Liposomal Amphotericin (5 mg/kg intravenous for 1 week but later tapered to 3 mg/kg when she developed Takotsubo cardiomyopathy). Takotsubo cardiomyopathy reversed after reducing the dose of amphotericin. The total cumulative dose given was 4050mg [Figure 1 & 2].

Surgical Intervention

The patient also underwent Endoscopic trans-nasal trans-maxillary trans-pterygoid debridement under General Anesthesia (GA). All the necrotic tissues of the nasal cavity and bilateral maxilla, Pterygopalatine fossa, and Infratemporal fossa were debrided (Figure 3a&3b). Granulations were also seen along the edges of the septal perforation with bare cartilage at places [Figure 3c]. Histopathology reports were suggestive of "Fungal infection of the maxillary sinus – Mucormycosis" [Figure 4].

The patient was discharged on tab posaconazole 300mg OD for 2 months. At 4 weeks, facial nerve and hypoglossal nerve palsy improved, and the nasal cavity was well healed, with no crusts and post-op status present (Figure 5).



Figure 1: Pre-op Images of House Brackmann Grade IV left-sided LMN Type Facial palsy (1A) and Right Hypoglossal Nerve Palsy (1B)

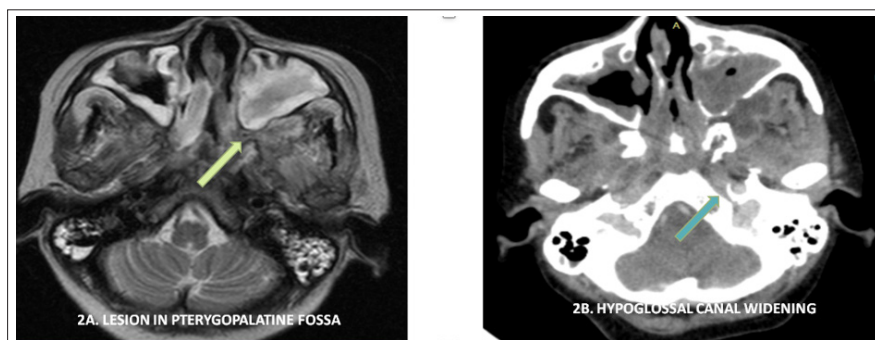


Figure 2: 2A: CEMRI Axial Section Showing Lesion in Pterygopalatine Fossa, 2B: CECT Axial Section Showing Widening of Hypoglossal Canal

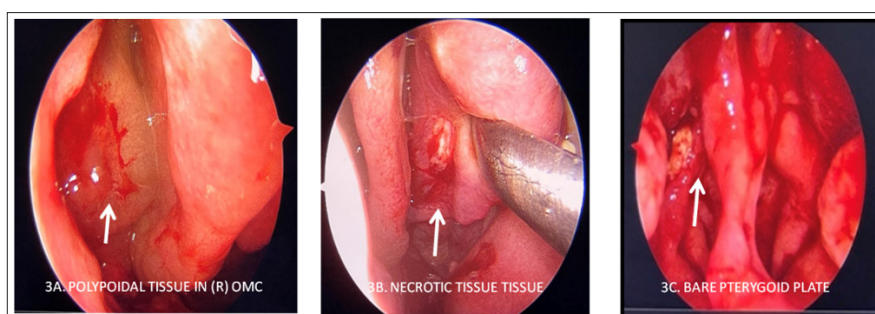


Figure 3: Intra-op Images

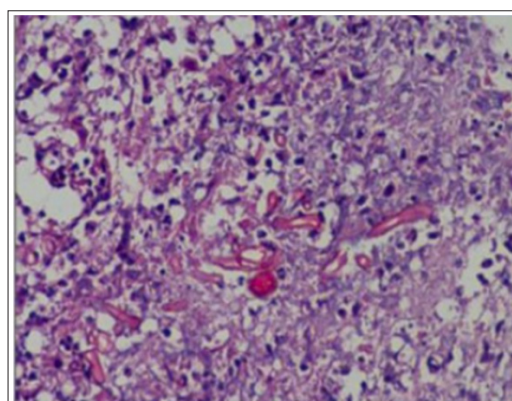


Figure 4: HPE Images

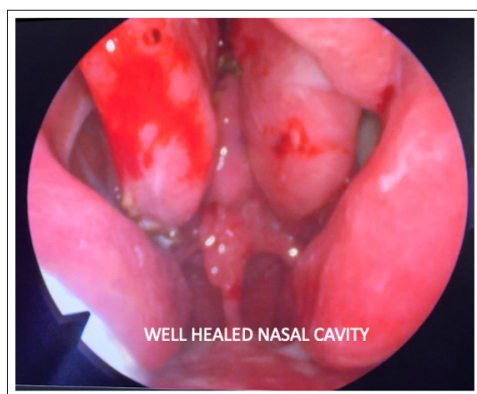


Figure 5: Post-op image

Discussion

Invasive mucormycosis is a serious infection and requires prompt surgical treatment to improve patient outcomes. Fungal invasion of paranasal sinuses of a susceptible host causes consistent symptoms of sinusitis or periorbital cellulitis and facial numbness, followed by the onset of conjunctival suffusion, blurry vision, and soft tissue swelling, followed by eschar formation and necrosis of the Naso facial region [1-3]. The fungi causing mucormycosis are opportunistic and can become fatal in debilitated patients if not treated in time. They tend to grow into blood vessels and lymphatics, causing the formation of mucor thrombi, resulting in ischemia and infarction of the affected organ. The infection may spread rapidly into the orbit from adjacent sinuses and may even extend intracranially or direct hematogenous route. The fungus may cause cavernous sinus thrombosis, leading to either unilateral or bilateral [4]. A clinical suspicion of mucormycosis requires confirmation by radiological examination, preferably a CT scan of the maxilla and orbit, which commonly shows a membrane and periosteal thickening with bony disruption [5]. Imaging findings may be contradictory statements and include unilateral or bilateral pansinus inflammatory changes such as polypoid mucosal thickening. Foci of hyperdensity in the affected sinus on CT scans are highly suggestive of a fungal disease [6].

The involvement of cranial nerve VII in mucormycosis is very rare, and its pathogenesis is not well understood. However, we propose that it may be caused by mycotic emboli or thromboembolic phenomenon to the vasa vasorum of the facial nerve [7].

In advanced disease, symptoms include chemosis, ptosis, proptosis, ophthalmoplegia, blindness, and multiple cranial nerve palsies (function of cranial nerves II, III, V, VI, and VI may be lost or impaired) [8, 9]. The classical clinical presentations are facial pain, an irregular black eschar on the palatal or nasal mucosa, and pus discharge from the eye and nose.

Despite recent tremendous advancements in sinus surgery, invasive fungal sinusitis still has a high mortality rate. Available research consensus recommends aggressive surgical debridement and long-term antifungal treatment [10]. However, more recently, it has become clear that aggressive surgery may also cause severe morbidity in the survivors. Therefore, many researchers have sought to improve treatment techniques to gain better clinical outcomes. Hence, our surgery, Endoscopic trans-nasal trans-maxillary trans-pterygoid debridement, plays an important role in improvement in neurological recovery as well as in removal of disease from the nasal cavity, sinuses and skull base.

Amphotericin B is the established pharmacotherapy in its liposomal form and has less toxicity than the conventional form. Hence, Liposomal Amphotericin B was the choice of drug in this case [11].

Conclusion

Mucormycosis is a rare condition that is still fatal in the era of medical advancements. A combination of endoscopic surgical debridement followed by intravenous Amphotericin B therapy is quintessential in the management of such patients and has less morbidity compared with conventional treatment.

A good clinical acumen is therefore essential to pick up this potentially fatal condition, especially in a diabetic patient. The treating team must have a high degree of suspicion, even if the diabetic status of the patient is of recent onset.

Rare presentations like Multiple cranial nerve palsy, which would otherwise raise the suspicion onwards granulomatous disease, need to be scrutinised and aspects ruled out in the effective treatment of patients.

Declarations

- The authors have no relevant financial or non-financial interests to disclose.
- The authors have no competing interests to declare that are relevant to the content of this article.
- All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest or non-financial interest in the subject matter or materials discussed in this manuscript.
- The authors have no financial or proprietary interests in any material discussed in this article.
- No identification of any patients done in the paper. Appropriate sections covered to hide identity.
- Consent from patient taken.
- Having done for an emergency situation prior sanction not taken and retrospective waiver taken in view of life saving procedure
- Data is available for the journal's perusal

Ethical Statement

- All the authors mentioned have contributed to the paper
- No funding has been received for the above study
- This paper is in accordance with 1964 Helsinki Declaration for compliance with ethical standards
- There is no conflict of interest for authors' contributing.
- No identity of the patients have been revealed while writing the paper.

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