

Mucormycosis presented with Painful Facial Nerve Palsy: a case report

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Abstract

Mucormycosis is a rare fungal infection with high morbidity and mortality which can present as rhino-cerebral, pulmonary, cutaneous, gastro-intestinal, orbital, and disseminated disease. We are presenting a case of maxillary mucormycosis with unilateral facial nerve palsy as a rare presentation. Clinico-anatomical basis of facial nerve involvement has not yet been completely discovered. We highlight the importance of understanding regional anatomical relationships along with the clinical suspicion which helps to uncover the unusual presentations of mucormycosis.

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Case report

A 45-year-old male was admitted with one week history of left-sided facial pain. He was previously diagnosed with type 2 diabetes, stage 3b chronic kidney disease, and chronic liver disease. Facial pain was associated with nasal blockade and headache. He denied a history of fever, trauma, rashes, or sore throat. Initially, he has been treated for possible maxillary sinusitis. The patient was re-admitted to Teaching Hospital Peradeniya due to worsening of the above symptoms.

On examination, the patient was in severe facial pain and headache. There was left-side maxillary tenderness with left-sided sub-centimetre size cervical lymphadenopathy suggestive of reactive lymph nodes. He had few carried teeth without significant surrounding inflammation. No palatal or nasal mucosal erosions were noted. His initial investigations showed mild neutrophil leukocytosis with $11.3 \times 10^9/L$ and marginally elevated CRP of 48 mg/L. X-ray sinus view showed features of maxillary sinusitis.

The next day patient developed lower motor type left side facial nerve palsy without other cranial nerve involvement. Infectious-related neuritis, demyelinating neuropathies, diabetes-associated mononeuritis multiplex, and mucormycosis causing erosive neuropathy were the initial differential diagnoses. Urgent non-contrast computer tomography of the brain with paranasal sinuses revealed paranasal sinus mucosal disease without bony erosions (Figure 1). The ENT surgical team performed a fiberoptic endoscopic sinus surgery. It showed unhealthy nasal mucosa

with blackish debris (Figure 2) and microscopic examination revealed fungal filaments, confirming the diagnosis of mucormycosis.

The patient was started on Liposomal Amphotericin B and interval sinus debridement was done. When pain relief was achieved, he was started on physiotherapy.

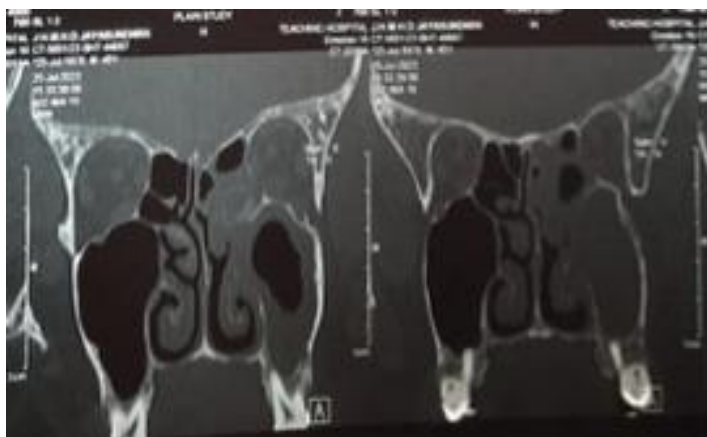


Figure 1: Severe left-sided maxillary sinusitis

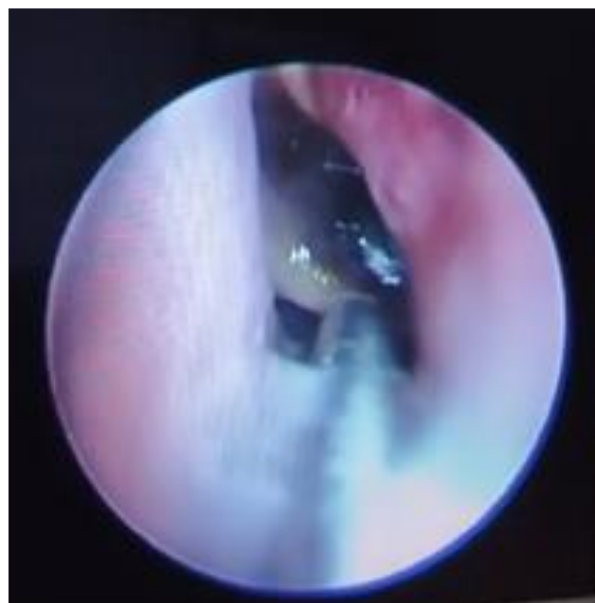


Figure 2: Fungal material visualized by Naso-endoscopy

Discussion

The incidence of mucormycosis reported in the literature varies among regions of the world, from 0.005 to 1.7 per 1,000,000 populations per year (1). Mucormycosis is an opportunistic, fulminant fungal infection caused mainly by *Rhizopus* species of the order Mucorales (2). It affects primarily the immunocompromised patients, specially those with uncontrolled diabetes mellitus. In addition, patients with organ transplant or hematopoietic stem cell transplant, corticosteroid therapy, neutropenia, or malignancy are at high risk (2,3,4).

Depending on the site affected, the disease may manifest in different forms, as rhino cerebral, pulmonary, cutaneous, gastro-intestinal, orbital, and disseminated disease. Rhino cerebral mucormycosis (RCM) is the commonest type accounting for more than 30% of cases (2, 4). This begins in the nasal cavity, spreads to paranasal sinuses, and then can invade into the retro-orbital region and cranium as well. The typical presentation of RCM includes nasal stuffiness, headache, retro-orbital pain, orbital swelling, ophthalmoplegia and visual loss in diabetic patients (2). Rarely can present with unilateral lower motor neuron facial palsy (2,5,6,7,8).

Since the maxillary sinus is closely related to important anatomical structures, this highly invasive fungus can spread fast and even can lead to diagnostic conundrums. The most common incident is, that this can present as a mimicker of cerebrovascular accident (CVA), which will ultimately delay the treatment leading to poor prognosis (5,6,7).

The exact mechanism or the anatomical basis of developing facial nerve palsy due to mucormycosis has not yet been described. Involvement of pterygopalatine fossa (9), perineural extension through the nerve of the pterygoid canal (Vidian nerve) (6), and direct spread through the Eustachian tube or the vascular channels into the middle ear (8) have been suggested as the anatomical basis for the isolated facial nerve palsy.

The pterygopalatine fossa is a small pyramid-shaped space lying between the posterior aspect of the maxilla (behind the maxillary sinus) anteriorly and the pterygoid process of the sphenoid bone, posteriorly (10, 11). Parts of the maxillary artery, vein, nerve, pterygopalatine ganglion, and fat are the main contents of this pterygopalatine fossa. The fungi in the maxillary sinus can easily reach the pterygopalatine fossa. Hosseini and Borghei have described pterygopalatine fossa as a reservoir of fungus and spread to retro global space of orbit and infratemporal space as well (9). The nerve of the pterygoid canal (Vidian nerve) is formed in the foramen lacerum by the union of the greater petrosal nerve which is a branch of the facial nerve and the deep petrosal nerve (a branch from the internal carotid sympathetic plexus) (11,12) (Figure 3). Perineural extension through the nerve of the pterygoid canal can cause isolated lower motor-type facial nerve involvement (6).

Mucormycosis is an angio-invasive organism that can invade arteries and veins leading to thrombosis (12). And perineural invasion of cranial nerves by this fungus also have been described (6). The pterygopalatine fossa contents including nervous and vascular

tissues, can act as a medium for direct angio-invasion and perineural invasion of the mucor up to the cranial tissue and other nearby structures. Mucormycosis, being a highly invasive organism in immunocompromised, the direct spread through the Eustachian canal is also a possibility (8).

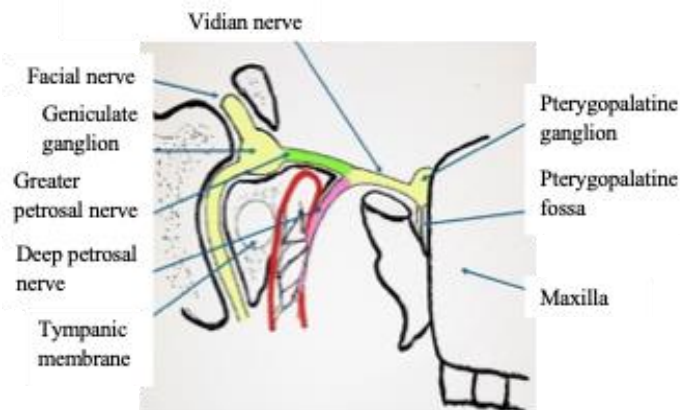


Figure 3: Connection between the facial nerve and the pterygopalatine fossa

In our case, there were no bony erosions evident in the bone window images of the CT scan even though the isolated facial nerve palsy and maxillary sinus mucormycosis were confirmed. Hence, direction erosive invasion through the maxillary sinus is a remote possibility. One postulated mechanism for our case would be, the spread from the maxillary sinus via the angio-invasive property of the fungus to the pterygopalatine fossa through the superior alveolar artery (a branch of the pterygopalatine part of the maxillary artery) supplying the maxillary sinus. And then from the pterygopalatine fossa via perineural spread to the facial nerve. Another possibility is, that since the pterygopalatine fossa can be a reservoir of the fungus, reactivation of the fungus with the background immunosuppression and spread via perineural

invasion without direct connection with the concurrent maxillary sinus infection.

Conclusion

Mucormycosis needs to be considered in patients with risk factors, presenting with lower motor type painful facial nerve palsy. Awareness on anatomical complexities of the region of interest will support early diagnosis of rare clinical presentations.

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