

Unilateral abducens nerve palsy secondary to isolated fungal sphenoidal sinusitis

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ABSTRACT

Background: Fungal sinusitis of isolated sphenoid sinus is a rare entity. Most of the sufferers referred with complications since the primary manifestations are non-characteristic.

Case presentation: We describe a 40-year old woman presenting with severe headache, diplopia, and limited right-eye movement. Further studies proposed isolated sphenoidal sinusitis, for which she underwent endoscopic sphenoidotomy. Microbiologic studies revealed extramucosal contamination with a saprophytic mucor. She enjoyed healthy life 5 weeks later.

Conclusion: Prompt diagnosis and treatment of isolated sphenoidal sinusitis is of utmost importance since it has non-characteristic manifestations. Noninvasive fungal sphenoidal sinusitis is best treated with sphenoidotomy. It seemed that abducens nerve palsy was associated with total sinus obstruction since patient condition improved promptly following the sphenoidotomy

Keywords: *Sphenoidal sinusitis, Fungal infection, Abducens nerve palsy.*

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INTRODUCTION

Fungal sinusitis of isolated sphenoid sinus is a rare entity with an incidence of less than 3% (1). Inflammatory pathogens are considered the most commonly reported etiologic factors, among which fungal agents have been detected in 31% of cases (2). *Aspergillus fumigatus* is the most frequent fungal pathogen isolated from paranasal sinus (3). Patients usually present with non-characteristic manifestations and refer to otolaryngologists during the end stage course of the disease when

complications occur. In the present report we describe a 40-year old woman presenting with isolated fungal sphenoidal sinusitis and unilateral abducens nerve palsy.

CASE PRESENTATION

A 40-year woman presented with nausea, fever, and severe headache primarily on right retro-orbital and temporoparietal regions. On admission laboratory indices were as follow: white blood cell count= 14000/mm³, polymorphonuclear (PMN) =70% and ESR=80. Lumbar puncture was performed; however, it was within normal range. One week following the admission, she complained of diplopia. Further ophthalmologic examination

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revealed abducens nerve palsy, but visual acuity was intact. Patients became afebrile and complementary neurologic examinations added nothing to the previous findings. Magnetic resonance imaging (MRI) revealed a thorough right sphenoidal sinus opacity which was isodense and hypodense in T1 and T2 section, respectively. Cavernous sinus was intact. During rhinoscopy sphenoidal recess was found to be erythematous, but other examinations were normal. Computed tomography (CT) showed right sphenoidal sinusitis with intact sinus wall. Other paranasal sinuses were normal. She received intravenous ceftriaxone, metronidazole, and vancomycin, nonetheless, the patient condition did not improve. Therefore, she was ordered functional sphenoidal sinus endoscopy through a transnasal approach. Sphenoidal recess was covered by polypoid tissue like superior turbinate. Anterior sphenoidal wall was dissected and a dense white mass was removed and sent for culture and smear. A saprophytic mucor was found with no bacterial growth. Pathological studies reported a chronic inflammation without evidences of granuloma, necrosis, mucosal or vascular invasion. Few hours following the procedure, improvement of eye motion started. She was discharged the day after the endoscopic procedure and one week later ESR was reported within the normal range.

During the next 5 weeks her condition improved and abducens nerve palsy alleviated thereafter. Follow up rhinoscopy was normal.

DISCUSSION

Sphenoidal sinusitis usually occurs in association with other paranasal sinusitis (4). Since it is adjacent to vital anatomic sites including dura, hypophysis gland, optic nerve, cavernous sinus, pterygoid nerve and canal, internal carotid artery, cranial nerves (III, IV, VI) and maxillary and ophthalmic branches of trigeminal nerve, its complications are of utmost importance.

Sphenoidal sinusitis could lead to superior orbital fissure syndrome, orbital apex syndrome, cavernous sinus thrombosis, and meningitis through direct invasion, hematologic dissemination or bone dehiscence (5). Prior investigators have proposed the following risk factors for isolated sphenoidal sinusitis: corticosteroid and/or immunosuppressive therapy, cocaine use, swimming in contaminated pools, craniofacial radiotherapy, obstruction of sinus ostium, and maxillofacial trauma (6); however, our patient had none the above mentioned risk factors. HIV test was negative. There was no history of Diabetes Mellitus, corticosteroid therapy or other immunocompromising factors in our patient.

The vast majority of sphenoidal disorders (i.e., 70-90%) are presented with severe headache which is usually localized at retroorbital region, however, it could be spread to vertex, frontal and temporal regions as well (9,8,7). Visual disturbance is the second most frequent complication of sphenoidal disorders with an incidence of 12% (9). Visual disturbance could be caused by cranial neuritis secondary to adjacent infections, ischemia due to an expansible lesion, or thrombophlebitis- or vasculitis- associated ischemic infarction (10). Optic nerve is appeared to be involved more often than any other cranial nerve. In the previously reported series the incidence was 12% for inflammatory lesions (9). Prognosis is poor in patients with optic neuritis; therefore, this should be considered a true emergency (10). Abducens nerve is the second most frequent involved cranial nerve with an incidence of 6%, partly due to its medial intracavernous position (9). Diplopia, caused by abducens nerve involvement, is among the earliest manifestations of sphenoidal sinusitis (9).

Involvement of other cranial nerves (III, IV, VI, VII) have also been described. Cranial nerves III and IV seem to be more fragile than optic nerve and their involvement occurs earlier. Lee et al demonstrated a satisfactory response rate (75%) among patients with abducens nerve palsy

secondary to isolated sphenoidal sinusitis when they referred during the first 96 hours. Furthermore, they stated that early improvement (during the first 3 months) in extraocular movement is associated with better outcome (10). Moreover, epistaxis and nasal obstruction may be the other presenting symptoms (11).

Disorders of sphenoidal sinus are usually diagnosed based on a thorough history, physical examination, rhinoscopy, CT scan, and MRI (10). Friedman et al reported their experience with 36 isolated sphenoidal sinusitis patients, among whom 10 presented with fungal ball, 2 with chronic invasive fungal sinusitis, and only one patient with fungal debris in sphenoidal recess (2). *Aspergillus* was the most common isolated fungus, a finding that is in accordance with other studies (12).

In our patient, mycologic studies revealed *mucor* while pathologic studies showed chronic inflammation without evidences of granuloma, necrosis and mucosal invasion, therefore, a noninvasive fungal infection could be proposed. To our knowledge, noninvasive fungal sinusitis of sphenoid sinus due to *mucor* has not been previously described. We have prescribed intravenous antibiotics for our patient, but her condition did not improve, so she underwent transnasal endoscopic sphenoidotomy in order to remove secretions, decrease mass-induced compression and infection eradication. Few hours following the endoscopic procedure, she felt better and her conditions alleviated thereafter.

In conclusion, prompt diagnosis and treatment of isolated sphenoidal sinusitis is of utmost importance since it has non-characteristic manifestations. Noninvasive fungal sphenoidal sinusitis is best treated with sphenoidotomy. It seemed that abducens nerve palsy was associated with total sinus obstruction since patient condition improved promptly following the sphenoidotomy.

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