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Case Report

Mucocele of the Sphenoid Sinus Caused by Aspergillus Fumigatus

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ABSTRACT

A 65 year old male presented with right sided frontal headache and pain in the right eye since 3 weeks. He also complained of diminution of vision in the right eye since 2 weeks. He was a known case of diabetes on treatment since last 12 years. Clinically this condition was diagnosed as ophthalmoplegia with benign Diabetic Retinopathy. On CT scan diagnosis was given as Sphenoid mucocele on right side secondary to fungal sinusitis. So based on clinical features & imaging study this condition was provisionally diagnosed as Sphenoid mucocele with optic neuritis. Functional endoscopic sinus surgery was done and pus was drained under general anaesthesia. Material was sent for histopathology and microbiology. Histopathological diagnosis was given as Aspergillosis. The appearance on wet mount was suggestive of Aspergillus .The fungal culture yielded Aspergillus fumigatus.

Keywords: Sphenoid mucocele, Aspergillus fumigatus, Aspergillosis

INTRODUCTION

Mucocele of the paranasal sinuses benign containing cystic are mucus formations covered respiratory by epithelium. They are caused by blockage of sinus ostia with subsequent the accumulation of mucus that eventually expands the sinus. The majority of paranasal sinus mucocoeles occur in the frontal and anterior ethmoidal sinuses. Mucocele of the sphenoidal sinus is uncommon and usually remains asymptomatic until it causes compression of the surrounding structures. The close proximity of optic nerve to sinus makes it vulnerable to get involved. Patients

with sphenoidal sinus mucocoele may present with intractable headache, decreased visual acuity, visual field defects exophthalmoses and endocrine disorder from seller extension.^[1] Urgent surgical management is mandatory if sphenoid sinus pathology is suspected to avoid untoward and irreversible damage like blindness. Histopathological examination and fungal culture play a crucial role in confirming the diagnosis and guiding the clinicians regarding management of these conditions.

CASE HISTORY

A 65 year old farmer presented with 3 weeks history of right sided frontal headache and pain in the right eye. He also gave a history of ptosis since 2 weeks. There was progressive loss of vision in the right eye since 2 weeks. He had a history of diabetes since last 12 years & was on treatment. His general examination was normal. All vital signs were normal. His fasting blood sugar was 62mg/dl and postprandial blood sugar was 379 mg/dl. His HbA1c was 10.6 % indicating uncontrolled diabetes for a prolonged period. There was nothing significant in other laboratory tests. Blindness was confirmed ophthalmologist. Their clinical impression was painful ophthalmoplegia with benign diabetic retinopathy. On CT scan of paranasal sinus the findings were suggestive of sphenoid sinus mucocele on right side secondary to fungal sinusitis causing destruction of its walls involving posterior ethmoidal air cells with likely focal optic neuritis & cavernous thrombosis on the right side. So based on the imaging & clinical picture provisional diagnosis of this condition was given as sphenoidal sinus mucocele with optic neuritis.

A week later patient was taken for right functional endoscopic sinus surgery. A bulge due to collection of pus at sphenoid ostium was noted. This pus along with fragmented tissue was sent microbiological histopathological & examination. KOH mount of the pus revealed hyaline septate hyphae with dichotomous branching. Histopathological examination revealed fragmented pieces of entangled fungal ball showing long fungal hyphae with acute angle branching and numerous round conidia [Figure 1]. It was diagnosed as aspergillosis in sphenoid sinus. The sample was cultured on Sabouraud dextrose agar at 37^oC.After 48 hours velvety colonies were grown which later turned smoky-green [Figure 3]. The reverse side was white to tan .The findings of lactophenol preparation cotton blue demonstrated dichotomously branched septate hyphae, smooth conidiophores, uniseriate vesicles and conidia covering upper half of the vesicle parallel to axis of stalk [Figure 2].

Post-surgery there was relief from the pain but vision was not improved.

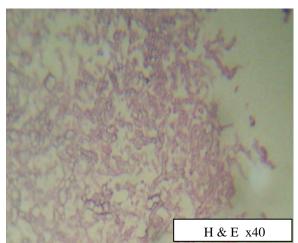


Figure 1:Section showing septate hyphae of *Aspergillus* showing acute angle branching.

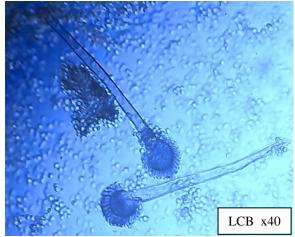


Figure 2: Conidiophores, Vesicles covered by single layer of phialides (uniseriate) on the upper half and Conidia of Aspergillus fumigatus.



Figure 3: Smoky green coloured colonies of Aspergillus fumigatus.

DISCUSSION

Berge in 1889 described the first case of sphenoidal sinus mucocele. Since then, around 150 cases have been reported so far in the literature. They constitute only 1-2 % of mucocele that occur in paranasal sinuses. Etiology of Sphenoid sinus mucocele is controversial. It occurs when a sinus ostium is obstructed, causing the sinus cavity to be filled and airless. The obstruction is often due to inflammation, prior trauma or surgical manipulation. In our case fungal sinusitis lead to the formation of mucocele.

The clinical manifestations of sphenoid sinus mucocele are determined by the direction of expansion and the involved surrounding structures. Headache is the most common symptom seen in 70 % to 90% cases^[3] followed by visual disturbance.^[4] It is the visual symptoms that alert the patient and the clinician to the seriousness of the problem.^[7] Our patient also presented first with headache and pain in the eye followed by progressive diminution of vision leading to blindness.

Investigations for diagnosing sphenoid mucocele include plain X ray of skull AP and lateral view and Computed tomography (CT) of the brain which in case

of sphenoid mucocele would show hypodense cystic lesions in sphenoid sinus. MRI scans of the brain can diagnose mucocele without doubt as a cystic homogenous lesion in relation to paranasal sinuses.^[8]

On reviewing the literature, it was seen that the visual loss occurring in cases of sphenoid mucoceles is usually irreversible hence early surgery is strongly recommended in cases where the vision is at stake. [8]

In this case by the time patient came for consultation there was total loss of vision. After surgery patient was symptomatically better, there was some relief from pain but there was no recovery in vision.

Mucocele of sphenoid sinus is extremely rare. Although imaging technique provides complimentary diagnostic evidence, demonstration of the fungus in tissue and isolation of the fungus confirms the diagnosis. This is a unique report of Sphenoid sinus mucocele caused by Aspergillus fumigates in which clinical, histopathological, imaging and microbiological findings correlated very well.

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