

FUNGAL MUCOCELE OF SPHENOID SINUS, AN UNUSUAL PRESENTATION IN AN IMMUNOCOMPETENT – A CASE REPORT

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Abstract

Background: Mucocele involving the sphenoid sinus is an uncommon entity and a diagnostic challenge. Rarely these lesions can be complicated by fungal infections, especially in immunocompromised individuals. In an immunocompetent individual this presentation is unusual. The symptoms are non-specific – in our case, the patient presented with the chief complaints of headache and dizziness, and the paranasal sinuses findings were identified incidentally on the Non Contrast Computed Tomography (NCCT) Head study. Radiological imaging, particularly multidetector computed tomography (MDCT), plays a crucial role in clinching the diagnosis. The aim of this case report is to add to the limited literature available and provide an account of the radiological features. Case presentation: A 20-year-old male presented with a 1.5-month history of headaches and giddiness. He did not have any co-morbidities. The paranasal sinus Computed Tomography scan revealed sphenoid sinus expansion with internal hyperdense (HU~ 130 to 150) areas, indicative of fungal sphenoidal mucocele. Additionally fungal sinusitis in the right sphenoid sinus and bilateral maxillary sinuses was also noted. Peripheral blood smear findings revealed eosinophilia, supporting the diagnosis of fungal etiology. A nasal swab confirmed *Curvularia* species through KOH mount analysis. Conclusions: Fungal mucocele of the sphenoid sinus is a rare condition, that too in an immunocompetent individual with only a handful of cases documented in literature. Understanding the distinctive radiological traits is crucial in notifying the clinician about the diagnosis. Timely identification and intervention are vital to prevent potential serious complications.

Keywords: Fungal, Mucocele, Sphenoid Sinus, Immunocompetent, Computed Tomography.

BACKGROUND

Though mucoceles were first described by Langenbeck in 1820(1), and the first case of sphenoid sinus mucocele was documented by Berg in 1889(2), very few cases have been documented till date in literature(2). Mucoceles are benign, expansile, cystic lesions found in the paranasal sinuses, that tend to silently invade and erode the adjacent bones and tissues(2). They are most often seen to involve the frontal sinuses, with the ethmoids being the next most common site. This can be attributed to the low incidence rate of sphenoid sinus mucocele, reported to be between 1-2%(3).

Although mucoceles usually secrete sterile mucoid fluids, pyocoeles can form as a result of secondary bacterial infections. However mucoceles with concurrent fungal infections are incredibly uncommon(4).

The aim of this case report is to add to the limited literature available and provide an account of the radiological features.

The clinical presentation is vague with symptoms ranging from headache, nasal obstruction, purulent nasal discharge, altered vision and neural deficits, depending on the structures involved by the fungal invasion(5).

A predilection for the development of fungal infection is seen in association with steroid abuse, chemotherapy and metabolic diseases like diabetes mellitus, as the

immunocompromised nature of the host facilitates the spread of fungal infections(9). However fungal ball is commonly found in immunocompetent patients as well, with the likely pathophysiology being impaired sinus ventilation(9).

Multidetector computed tomography is often the investigation of choice in suspected paranasal sinus pathologies with a high degree of agreement found between MDCT and histopathological diagnosis(10).

The computed tomography findings include complete opacification and expansion of the involved sinus, with thinning of the sinus walls. Bony erosion and extension into surrounding structures may also be seen(11,12).

Treatment options include endoscopic sphenoidotomy, which has low morbidity and recurrence rates(13,14).

Case Presentation

A 20-year-old male patient from a rural household presented with the complaints of headache and giddiness since one and half month and was referred to the department of Radiodiagnosis for brain CT scan study.

The general physical examination and the vitals were unremarkable. The patient was afebrile at the time of presentation. He was non diabetic and non hypertensive. Visual examination was unremarkable. No focal neurological deficit was present.

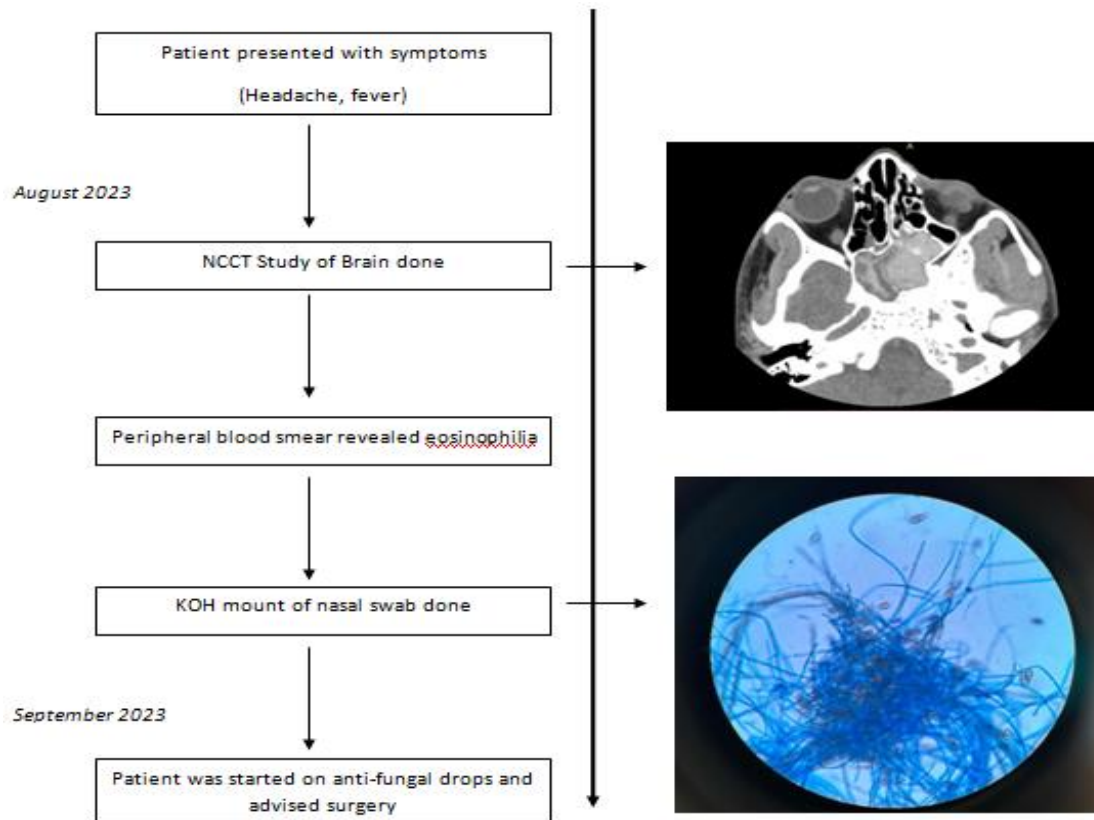
A Non-Contrast CT scan of the brain was performed on a 128-slice multi-detector CT scanner (OPTIMO WIPRO GE HEALTHCARE), which showed expansion of the left sphenoid sinus with hyperdense (HU~ 130 to 150) areas within. Bony erosion at places with thinning of walls is noted. Mucosal thickening with hyperdense areas are also noted in right sphenoid sinus and bilateral maxillary sinuses.

Mucosal thickening without hyperdense areas are noted in bilateral frontal sinuses and ethmoidal air cells. Left maxillary sinus also showed expansion with widening of osteomeatal complex. These features were indicative of left sphenoid sinus fungal mucocele with fungal sinusitis of right sphenoid sinus and bilateral maxillary sinus.

Further questioning revealed that the patient had the additional symptoms of intermittent fever since 2 months. The above CT findings were consistent with the clinical features of the patient.

Peripheral blood smear revealed eosinophilia, in keeping with the diagnosis of fungal etiology. A nasal swab was taken for KOH mount which revealed brown curved conidia with transverse septations suggestive of *Curvularia* species.

The patient was started on antifungal nasal drops for 15 days in view of the above mentioned findings, and was counselled regarding timely need for surgical exploration.



DISCUSSION

Mucoceles are expansile, benign, cystic lesions that rarely involve the paranasal sinuses, more commonly occurring in the frontal and ethmoidal sinuses. Sphenoid mucoceles are rare. Amongst the paranasal sinuses, the sphenoid sinus is the least frequently involved, comprising 1%–2% of all mucoceles (2,4). Less than 150 cases of sphenoid sinus mucocele have been described in the literature(5). In a study by Rombaux et al (6), sphenoid sinus mucocele was noticed in 1% to 8% of the paranasal mucoceles.

Early detection of any type of sphenoidal sinus pathology is challenging due to its anatomical placement (2).

Although the precise etiopathogenesis of the sphenoid sinus mucocele is yet unknown, obstruction of the sphenoid sinus ostium is typically assumed to be the etiology (3). The expansile mucocele's locally invasive nature can be accounted for by the high prostaglandin E2 levels leading to aggressive osteolytic processes. The production of proinflammatory cytokines (TNF- α and IL-1) exacerbates osteolytic bone resorption as well (3).

Despite being pathologically benign, sphenoid sinus mucocele can have an impact on important adjacent structures such as the internal carotid artery, cranial nerves (optic, oculomotor, abducent, and trigeminal nerves, dura, pituitary gland (3).

Sphenoid mucoceles associated with fungal ball are incredibly uncommon. Very few cases are reported till date in the literature (4,7,8). In these cases no mention of any associated co morbidities were made. Our case was of an immunocompetent individual.

The pathogenesis can be elucidated by the hypothesis that the fungus ball inside the sinus induces repeated infections, leads to obstruction of the sinus ostium and subsequently causes mucocele to develop (4).

Typically, sphenoid sinus mucoceles have vague symptoms on presentation. In addition to nonspecific headaches, other possible clinical manifestations depend on the adjacent structures involved and include purulent nasal discharge, retropharyngeal drip, nasal block, vision problems, and neurological deficits (5).

Although the diagnosis remains unusual, the incidence has been increasing, which can be attributed to the increased occurrence of metabolic disorders such as diabetes mellitus, utilization of broad-spectrum antibiotics, cancer patients receiving chemotherapies, and the rise in people with impaired immune systems brought on by steroid usage (9).

The deep seated nature of the sphenoid sinus makes physical examination a challenge, and pathologies are most often picked up by imaging (5).

Owing to its deep location, plain radiographs fail to provide clear visualization of the sphenoid sinus and imaging modalities such as (CT) computed tomography, (MRI) magnetic resonance imaging as well as endoscopic biopsy are crucial for comprehending the various diseases that impact this area (5).

Multidetector Computed Tomography provides a comprehensive picture of the paranasal sinus and demonstrates a high diagnostic precision in keeping with the histopathological diagnosis (10).

The CT scan findings in allergic fungal sinusitis include expansion and opacification of paranasal sinuses, with central hyperdense fungal component and peripheral hypodense mucosa. Bony involvement is also common in the form of thinning and remodelling of the bony walls of the paranasal sinuses (11).

On CT images a sphenoidal mucocele is seen as a hypodense, cystic lesion in the sphenoid sinus, which may cause sinus expansion and bony erosion of its walls, and extend to the neighboring sellar and parasellar regions. It must be distinguished from other cystic lesions observed at this site, including arachnoid cysts, cystic optic nerve gliomas, cystic pituitary adenomas, rhinopharyngiomas, and Rathke cleft cysts (12).

MRI findings in allergic fungal sinusitis most often include low signal intensity on both T1 and T2 weighted images, however high or mixed signal intensities are also sometimes seen on T1. (10) The T2-weighted images typically show peripheral hyperintensity indicative of inflamed mucosa, with central signal-void or hypointense areas due to the high concentration of fungal organisms and high concentration of protein with low concentration of free water in allergic mucin. Peripheral mucosal enhancement is also visible when gadolinium contrast is applied to T1-weighted images (11).

On MR imaging, mucocele is hypointense on T1-weighted images and hyperintense on T2-weighted images. The sinus mucosa enhances as a thin line surrounding the mucocele in contrast-enhanced MRI imaging. However their MRI appearance depends on the protein concentration of the mucoceles, which varies over time (11).

In our case, the patient was immunocompetent. NCCT showed expansion of the left sphenoid sinus, with a lobulated, heterogeneous, low-attenuation, cystic lesion of the left sphenoid sinus with central hyperdense areas of HU +190 to +200 indicative of

fungal content within the mucocele. Mucosal thickening with hyperdense areas were also noted in bilateral maxillary sinuses.

Inflammatory mucosal thickening is appreciated in bilateral frontal sinuses and ethmoidal air cells. Left maxillary sinus also showed expansion with widening of osteomeatal complex. These features were indicative of left sphenoid sinus fungal mucocele.



Figure 1: a,b,c: NCCT Axial, Coronal and Sagittal images showing expansion of the left sphenoid sinus with hyperdensities within indicative of fungal content within the mucocele (black arrows).

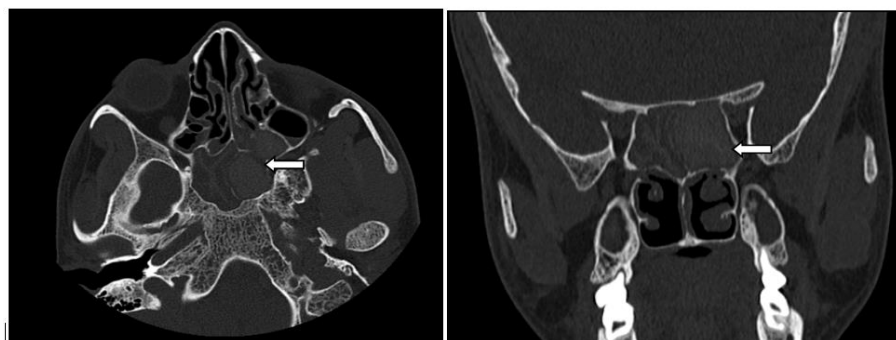


Figure 2: NCCT Axial and Coronal images showing expansion of the left sphenoid sinus (white arrows).

KOH mount of nasal swab revealed brown curved conidia with transverse septations suggestive of *Curvularia* species.

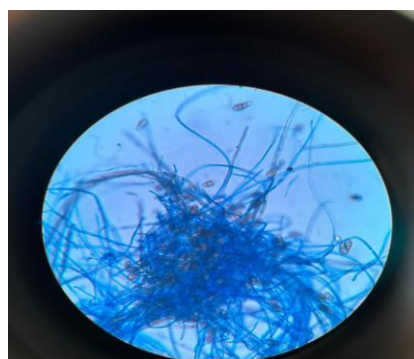


Figure 4: KOH mount showing brown curved conidia with transverse septations suggestive of *Curvularia* species.

Surgery is the definitive treatment for fungus ball within mucocele because it allows the afflicted sinus to be cleared of fungal debris and its proper drainage and ventilation to be restored (11, 14).

A safe and efficient therapeutic method is the endonasal endoscopic technique with drainage and marsupialization of the sphenoid sinus through a transnasal corridor (13,14).

CONCLUSIONS

The formation of a fungal ball within a sphenoidal mucocele is an extremely uncommon occurrence. Despite advancements in diagnostic techniques and imaging exams, diagnosis is still a difficult task. Knowledge of the characteristic radiological features is essential in alerting the clinician to the diagnosis. Early diagnosis and treatment are necessary to avoid potentially serious complications. This entity is very uncommon but should be kept in mind as a differential of expansile, cystic, sphenoid sinus.

List of Abbreviations

HU - Hounsfield Units

MDCT - Multi Detector Computed Tomography

NCCT - Non Contrast Computed Tomography

MRI - Magnetic Resonance Imaging

KOH - Potassium Hydroxide

TNF-a - Tumor Necrosis Factor-alpha

IL-1 - Interleukin-1

Declarations

Consent for publication:

Written informed consent was obtained from the patient.

Author contributions:

VRK reported the patient's CT scan, and directed and guided the manuscript preparation.

VD performed a review of the published literature, and prepared the manuscript of the case report.

The authors have read and approved the final manuscript.

Corresponding author – VRK

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