Isolated Sphenoid Fungus Ball Leads to Hemorrhagic Stroke

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Abstract

Objectives:
To describe a rare case involving fungal invasion of the cavernous carotid following endoscopic removal of an isolated sphenoid fungus ball.

Study Design:
Case report and literature review.

Methods:
Review of a case history including the patient's clinical course, surgery, radiographic images, histopathology and autopsy slides. A pertinent literature review is also presented.

Results:
The patient is a 68 year-old man with a history of inhaled and low dose oral steroid dependent asthma who presented with acute sinusitis. Following culture-directed antibiotic therapy, vertex headaches persisted, and a CT revealed an opacified sphenoid with a dehiscent right lateral wall, the latter of which occurs in up to 30% of sphenoids. He underwent uneventful awake endoscopic sphenoid sinus debridement with a perioperative increase in systemic steroids. Histopathology demonstrated a fungus ball, cultures were negative. 3 days postoperatively he developed a new onset pupil-sparing right third nerve palsy. An MRI showed minimal enhancement of posterior clinoids and CT showed an unremarkable sphenoid. 2 weeks postoperatively he suffered a hemorrhagic stroke and died. Postmortem evaluation revealed an edematous right third cranial nerve and invasion of the intima of the cavernous internal carotid artery with hyphae, consistent with *Aspergillus*, adjacent to bony dehiscence.

Conclusions:
Steroid use and a defect in the bony barrier of the sphenoid adjacent to the carotid artery may have predisposed this patient to carotid fungal invasion. Histopathology is critical in confirming diagnosis and antifungal therapy may inhibit fungal growth. Since this case, immunocompromised patients at our institution with a suspected fungus ball receive perioperative antifungals.

Introduction

Isolated sphenoid sinus fungal ball (FB) is the third most common pathology in isolated sphenoid sinus disease.[1] The most common presenting complaint is headache and the gold standard of management is surgical removal.[2] While removal in the awake patient is not uncommon, surgical manipulation of the sphenoid sinus is not without its risks as incidental bony dehiscence over critical structures such as the optic nerve and internal carotid artery may occur in upwards of 30% of patients.[3-5] Here we report a case of an isolated sphenoid FB that was complicated by an acute hemorrhagic stroke secondary to fungal invasion of the internal carotid artery.

Clinical Images

Figure 1. Non-contrasted, coronal section, CT scan demonstrating bony dehiscence of right lateral wall of sphenoid and sinus opacification.

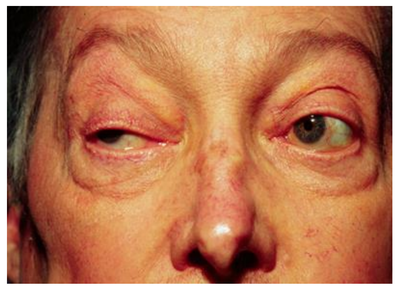

Figure 2. Patient with right eye ptosis, limitation of right lateral gaze and sparing of pupillary reactivity.

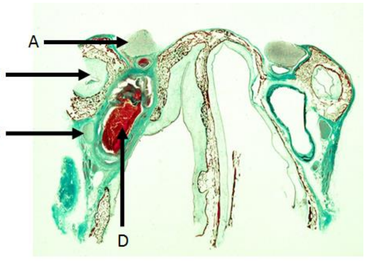

Figure 3. Optic nerve (A), anterior clinoid process (B), oculomotor nerve (C), and thrombosed internal carotid artery (D) in cross section from patient's autopsy slides depicting relationship of anatomical structures of the cavernous sinus.

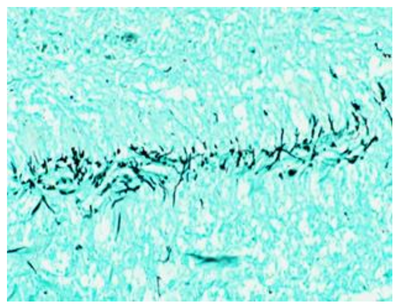

Figure 4. 100x magnification histopathological slide with silver stain that reveals invasion of fungal hyphae into carotid intima (Panel D).

Case Report

The patient is a 68 year-old on inhaled- and low dose oral steroids for asthma control who presented to the otolaryngology clinic with acute sinusitis, nasal discharge and worsening of asthma. Culture-directed antibiotics were administered and symptoms improved with the exception of vertex headache. A sinus CT revealed opacification of the right sphenoid sinus with a 1cm dehiscence in the right lateral sphenoid wall (Figure 1). The patient underwent awake, endoscopic removal of the sphenoid fungus ball in the operating room with a 45 degree elevation of his head. 3 days following this uneventful surgery, pathology revealed a fungus ball consistent with aspergillus. Fungal cultures did not grow. Concurrently, the patient presented with a right third nerve palsy. Neuroophthamologic exam confirmed a pupil sparing third nerve palsy (Figure 2); MRI showed enhancement of the posterior clinoids and sinus CT showed a clear sphenoid. 2 weeks post operatively he suffered a hemorrhage stroke and died. He was an organ donor and autopsy of the sphenoid showed edematous right third cranial nerve with associated cavernous sinus involvement (Figure 3) and invasion of the carotid with hyphae consistent with aspergillus (Figure 4).

Discussion

The convergence of several factors contributed to this catastrophic complication. Fungus balls are often asymptomatic, but may cause chronic drainage that worsens underlying asthma. Involvement of the sphenoid sinus classically causes vertex or temple pain.

Bony dehiscence of the sphenoid wall is a common finding that may occur is upwards of 30% of patients.[3-5] Many fungal species have a predisposition for vascular invasion. Aspergillus is associated with a specific stroke syndrome via hyphae invasion of small- and medium-size cerebral vessels, causing either a coagulative necrosis, which induces arterial thrombosis, or formation of mycotic aneurysms that may induce fatal hemorrhage.[6]

The autopsy histopathology from this immunocompromised patient demonstrates local invasion from what was initially just a fungus ball. To our knowledge, this is the first documentation of a pupil sparing 3rd nerve palsy caused by a fungus ball that became invasive. The combination of dehiscence of the bony wall and steroid use predisposed this patient to fungal invasion of the carotid artery. Perioperative use of antifungals may have prevented this postoperative complication.

Conclusions

Though typically a benign disease, fungus ball in an immunocompromised patient may represent a more aggressive pathology. Steroid use and a defect in the bony barrier of the sphenoid adjacent to the carotid artery may have predisposed this patient to carotid fungal invasion. Histopathology is critical in confirming diagnosis. Since this case, immunocompromised patients at our institution with imaging consistent with a fungus ball are given perioperative antifungals before undergoing ESS regardless of culture results. If pathology does not support presence of fungus, the antifungal is stopped.

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