CASE REPORTS



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Creative Commons (CC BY-NC-ND 4.0) Attribution - NonCommercial - NoDerivatives 4.0 International Blindness from Fungal Rhinosinusitis of the Paranasal Sinuses: A Case Report

ABSTRACT

Objective: To present a unique case of blindness resulting from fungal rhinosinusitis involving multiple sinuses mimicking a malignant process in a pregnant patient.

Methods:

Design: Case Report

Setting: Tertiary Government Training Hospital

Patient: One

Result: A 36-year-old pregnant woman developed unilateral blindness during her 20th week of gestation with a history of binocular diplopia, unilateral nasal obstruction and anosmia for 13 months during the pre-pregnancy period. Sphenoid sinus malignancy was suspected on imaging. The planned biopsy was intraoperatively shifted to endoscopic sinus surgery when clay-like materials were seen involving the left maxillary sinus and bilateral sphenoid and ethmoid sinuses. Histopathologic examination confirmed fungal growth. Postoperatively, nasal symptoms resolved but blindness of the left eye and blurring of vision of the right eye persisted.

Conclusion: Fungal rhinosinusitis rarely occurs in multiple sinuses and is commonly misdiagnosed. It can afflict pregnant patients and mimic a malignant process. A high index of suspicion early on, especially in the presence of nasal congestion and diplopia may prevent potentially irreversible complications.

Keywords: sinusitis; blindness; sphenoid sinus; maxillary sinus; pregnancy

Fungal rhinosinusitis is a rarely diagnosed disease affecting 12,000,000 globally¹ with an incidence of 5.4 and 8.2% in South Korea and Japan, respectively.² Sinus fungus ball is a non-invasive type that is predominantly unilateral in presentation and grows in wet, moist, cavities of the paranasal sinuses, mostly in females and elderly hosts with normal immunologic status.³ Since symptoms are non-specific, its presentation is indistinguishable from usual chronic bacterial rhinosinusitis and may only be discovered incidentally.⁴ In some cases, it may even mimic a malignant tumor especially when the orbit is involved.⁵ We present the insidious development of common non-specific nasal symptoms towards blindness in a pregnant patient with sinus fungus ball.

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CASE REPORT

A 36-year-old G4P2 (2-0-1-2) woman presented with a 13-month history of binocular diplopia and gradually progressing to blurring of vision. She also complained of occasional headache, left nasal obstruction and anosmia. No nasal discharge, epistaxis or difficulty of breathing were noted. She had an unremarkable past medical history and lived in a small apartment home with poor ventilation.

Her worsening blurring of vision prompted her to consult. A plain Cranial Computed Tomography (CT) scan revealed a lobulated soft tissue mass with coarse calcifications occupying the clivus, sphenoid, posterior ethmoid, sellar and suprasellar region (Figure 1A) and left maxillary sinus (Figure 1B) with primary consideration of a pituitary neoplasm. She was unable to obtain the further workups requested.

She was admitted under the Neurology service on her 20th week of gestation due to left eye blindness and severe headaches (VAS score 8/10). Ophthalmologic examination revealed absent light perception in the left eye and hand perception in the right eye. Both eyes were medially deviated with lateral rectus palsy more noticeable on the left. Fundoscopic examination revealed pale optic disc with no papilledema on the left eye. An MRI of the head with diffusion weighted imaging revealed bony expansion and thinning of bilateral sphenoid sinuses and the left maxillary sinus, heterogeneously-enhancing foci occupying the nasal area displacing the sella, pituitary gland, optic chiasm and nerves superiorly hence a suspicion of sphenoid sinus malignancy was entertained. (Figure 2A, B)

She was referred to ear, nose, throat - head and neck surgery (ENT-HNS) for evaluation of the suspected paranasal sinus mass. Physical examination revealed left nasal congestion with clear watery discharge on anterior rhinoscopy, and facial tenderness over the left maxillary area on palpation. Nasal videoendoscopy showed a suspicious nasopharyngeal bulge but punch biopsy revealed chronic inflammation only. Hence, maxillary antrostomy with exploration of the other sinuses via endoscopic sinus surgery was done. (Figure 3A) Intraoperative findings showed clay-like material filling the left maxillary sinus, bilateral ethmoid and sphenoid sinuses. (Figure 3B, C, D) Hematoxylin-Eosin and Gomori- Methenamine staining confirmed fungal growth of the acquired specimen. (Figures 4A and 4B, respectively). Serum galactomannan to determine the specific organism involved was not facilitated due to financial constraints. Both mother and fetus tolerated the procedure well. Antifungal medications were not given post-operatively due to pregnancy considerations.

Post-operatively, nasal congestion, facial tenderness, headache and anosmia resolved. Ophthalmologic examination revealed no

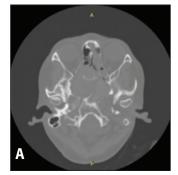
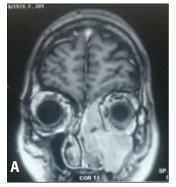




Figure 1. Plain cranial CT scan, Axial view: **A.** Lobulated soft tissue mass with coarse calcifications occupying the clivus, sphenoid, posterior ethmoid, sellar and suprasellar region; **B.** Lobulated soft tissue mass occupying the left maxillary sinus.



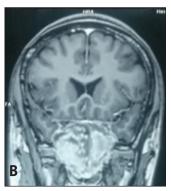
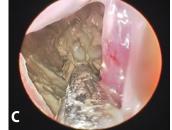


Figure 2. MRI of the head with DWI and contrast: **A.** Heterogeneously enhancing foci with bony expansion and thinning at the level of the left maxillary sinus; **B.** at the level of the sphenoid sinuses displacing the parasellar structures.







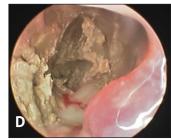
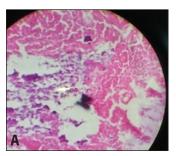


Figure 3. Intraoperative findings: **A.** Left maxillary sinus showing fungus ball upon opening of the anterior maxillary wall accessed thru gingivobuccal incision; and endoscopic views showing aggregates of fungus ball in the **B.** Left maxillary sinus, **C.** Sphenoid sinus and; **D.** Ethmoid sinus.

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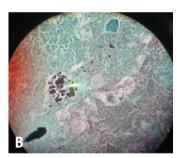


Figure 4. Histopathologic slides: **A.** Hematoxylin and Eosin stains showing fungal elements on high power view (white arrow) **B.** Histopathologic slides using Gomori methenamine staining showing collection of fungal elements in low power view (arrow).

improvement of her vision. She was discharged home with no other complications. On follow-up, she had delivered her baby at term via normal spontaneous delivery with no feto-maternal complications.

DISCUSSION

As shown in our case, blindness can be an unfortunate sequela of fungus ball. A MEDLINE (PubMed) search using the keywords "fungus ball" and "blindness" revealed a report by Kim *et al.* on a patient with blindness secondary to a sphenoid sinus fungus ball. Using the same keywords, a search of HERDIN Plus, the ASEAN Citation Index, Global Index Medicus (Western Pacific Region Index Medicus, WPRIM and Index Medicus of the South East Asia Region, IMSEAR), Directory of Open Access Journals (DOAJ) and Google Scholar yielded no other similar cases published locally.

The most common organisms causing fungus ball are *Aspergillus fumigatus, Aspergillus flavus, Alternaria*, and *Mucor* species.⁷ They are usually found in just one sinus; the maxillary sinus as the most frequently involved (94%) followed by the sphenoid sinus (4–8%).⁷ The ethmoid sinus (3%) is most often a continuous involvement from the maxillary sinus.⁷ Fungal rhinosinusitis rarely occurs in multiple sinuses and is commonly misdiagnosed.⁷

Mostafa *et al.* associated a small apartment floor, poor air conditioning, and exposure to dust and cockroaches with higher incidence of fungal rhinosinusitis due to humid environment and less exposure to sunlight.⁸ Our patient lived in similar circumstances with no other comorbidities that could lead to her condition.

Fungal rhinosinusitis has a non-specific presentation which may lead to confusion in diagnosis. In our case, an initial impression of a nasopharyngeal tumor was considered due to the nasal endoscopic findings of a nasopharyngeal bulge and the patient's diplopia. In about 57% of patients with nasopharyngeal carcinoma and diplopia, it was observed that the infiltrating nature of the mass to extend intracranially could have involved cranial nerve VI.9

In general, the visual disturbance in compressive lesions similar to our patient is attributed to the interruption of the blood supply to

the optic nerve with secondary ischemic optic atrophy, as confirmed by the fundoscopic findings of a pale optic disc.¹⁰ It has been posited that accumulation of viscous eosinophilic mucin by nasal congestion can cause obstruction of sinus outflow tracts, which then allows inflammatory mediators to cause gradual sinus expansion and bony erosion.¹¹ Hence, the bilateral bony expansion of the sphenoid sinus seen on MRI may have caused compression on the optic nerve, supported by the yellow-brownish friable fungus ball seen intraoperatively.

Definitive treatment for fungal rhinosinusitis involves restoration of normal sinus drainage by removal of the fungus ball, extirpation of allergic mucin, and long-term nasal steroids to prevent recurrence.¹¹ Antifungal medications were no longer initiated as they have not been proven to be beneficial in treatment¹¹ and could place the pregnancy and fetus at risk.

In conclusion, our case has shown that fungal rhinosinusitis can afflict a pregnant patient with non-specific nasal symptoms and ophthalmic complications mimicking a malignant process. Multiple sinus involvement of fungal rhinosinusitis, although rare, should also be considered as a differential diagnosis even in pregnant patients. A high index of suspicion early on, especially in the presence of nasal congestion and diplopia may prevent potentially irreversible complications.

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