Polish Annals of Medicine

Journal homepage: https://www.paom.pl

Case Report

Unveiling the mystery: Isolated sphenoid granulomatous fungal sinusitis causing nonspecific headache

Nur Izzaty Nadia Mohd Ariffin[1], Intan Kartika Kamarudin[2], Norasnieda Md Shukri[1]

[1] *Department of Otorhinolaryngology-Head and Neck Surgery, School of Medical Sciences, Universiti Sains Malaysia, Kelantan, Malaysia*
[2] *Universiti Teknologi MARA, Malaysia*

CORRECTED PROOF

Article info

Article history
Received: December 2, 2024-12-02
Accepted: February 8, 2025
Available online: November 18, 2025

Keywords
sphenoid sinus
nonspecific headache
sphenoid fungal sinusitis
myecetoma

Doi
https://doi.org/ 10.29089/paom/201137

User license
This work is licensed under a Creative Commons Attribution – NonCommercial – NoDerivatives 4.0 International License.

BY-NC-ND

Abstract

Introduction: Isolated sphenoid sinusitis is a rare paranasal sinusitis, accounting for less than 3% of sinus infections. Fungal infections cause 10% of these cases and are often missed due to nonspecific symptoms.

Aim: This case report highlights the importance of isolating sphenoid fungal sinusitis in patients with chronic, nonspecific headaches and emphasizes the need for early otorhinolaryngology referral.

Case study: A 67-year-old woman with well-controlled diabetes mellitus presented with a 10-year history of severe, nonspecific headaches. After multiple treatments and normal neurological examinations, a CT scan revealed isolated left sphenoid sinus soft tissue density with calcification and erosion. Endoscopic sphenoidotomy confirmed fungal balls, leading to a diagnosis of chronic granulomatous invasive fungal sinusitis.

Results and discussion: Isolated sphenoid fungal sinusitis predominantly affects immunocompetent individuals, particularly older women. Diagnosis relies heavily on imaging, as nasoendoscopy may not detect abnormalities. Endoscopic trans-nasal sphenoidotomy is the preferred surgical treatment for isolated sphenoid lesions.

Conclusions: (1) Chronic, nonspecific headaches unresponsive to conservative treatment should raise suspicion of isolated sphenoid fungal sinusitis. (2) Early otorhinolaryngology referral, proper endoscopic examination, and appropriate radiological imaging are crucial for timely diagnosis and treatment, preventing potentially severe complications.

Corresponding author: Norasnieda Md Shukri; Department of Otorhinolaryngology- Head and Neck Surgery, School of Medical Sciences, Universiti Sains Malaysia, 16150 Kubang Kerian, Kelantan, Malaysia.
E-mail address: asnieda@usm.my

1. INTRODUCTION

Paranasal sinusitis is common and affects 16% of the global population.[1] Isolated sphenoid sinusitis stands out due to its rarity and unique clinical challenges among the various forms of paranasal sinusitis. Since isolated sphenoid sinusitis only makes up 2.7% to 3.0% of all paranasal sinus infections, its infrequent occurrence distinguishes it and the array of nonspecific symptoms it can induce.[1] In 10% of cases, it is caused by fungal infection.[2] Fungal sinusitis is classified into invasive- defined by the infiltration of the mucosa and spreading to the surrounding structures, primarily affecting immunosuppressed patients; and non-invasive, typically affecting immunocompetent patients, without tissue invasion. In addition to causing a severe headache in the sphenoid region, it can produce occipital headache, retroorbital pain, otalgia, sleepiness, and symptoms resembling meningitis.[3]

2. AIM

We highlight the importance of considering isolated sphenoid fungal sinusitis in patients with chronic, nonspecific headaches. The enigmatic nature of these symptoms often leads to the misdiagnosis or delay in identifying sphenoid fungal sinusitis. The aim is to raise awareness of the need for early referral to an otorhinolaryngologist for a prompt diagnosis and proper management to avoid devastating complications.

3. CASE STUDY

A 67-year-old woman has been experiencing severe, nonspecific headaches for the past ten years. The pain is throbbing, mainly at the frontal region, progressively radiating to the occipital region. She has no nasal symptoms, other neurological deficits, or eye symptoms. The patient has underlying well-controlled diabetes mellitus. Despite seeking multiple treatments for her headache, her symptoms are still not relieved. Neurological examinations were normal. Nasoendoscopy shows bilateral inferior turbinate hypertrophy with no other pathological findings.

4. RESULTS

Serial computed tomography (CT) brain was performed to investigate her multiple attacks of severe headaches. After four years, it showed isolated left sphenoid sinus soft tissue density with calcification, which eroded the sinus's superior, lateral, and posterior walls (Figure 1). We performed endoscopic sphenoidotomy via trans-nasal approach. Intraoperatively, the sphenoid ostium was full of fungal balls and pus, which then was completely evacuated (Figure 2).

The histopathology examination was consistent with mycetoma. However, no fungal is isolated in the culture. The patient was diagnosed with chronic granulomatous invasive fungal sinusitis, given her immunocompetent status with fungal material in the sphenoid causing erosions of the surrounding area. Postoperatively, the intense headache is gradually relieved and remains well under follow-up.

5. DISCUSSION

Fungal sinusitis includes non-invasive and invasive forms. The non-invasive fungal ball (mycetoma) predominantly affects immunocompetent individuals, with a higher prevalence among older women, as documented by Leroux et al.[2] While the exact pathophysiology of the fungal ball is not entirely understood, it is widely accepted that inhaled spores

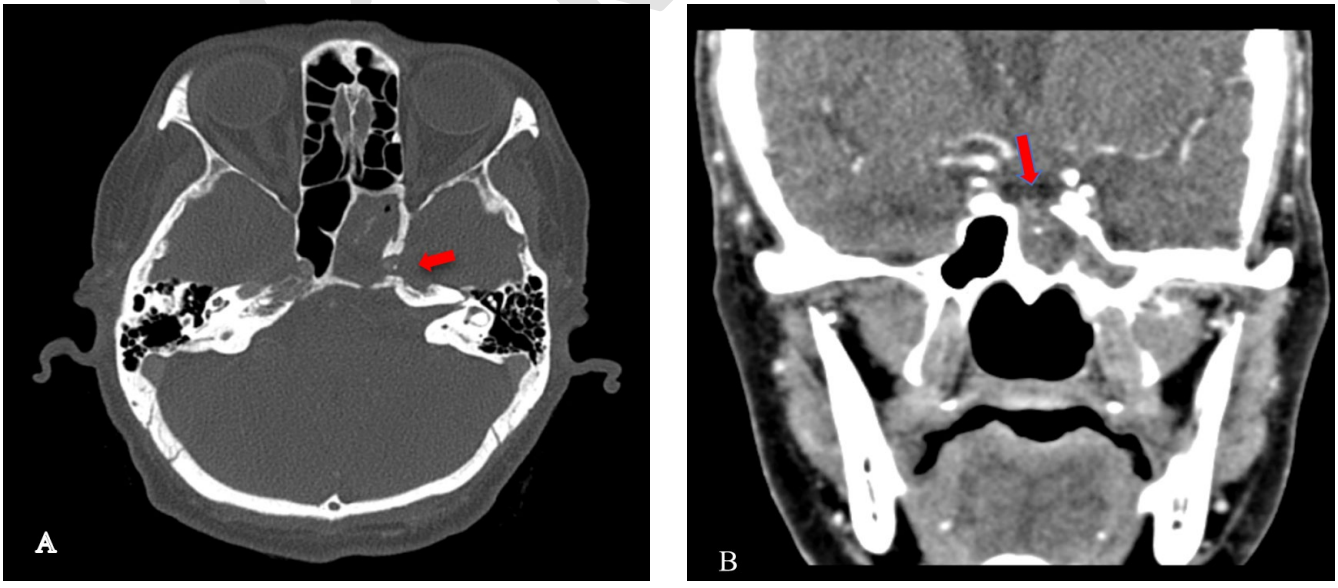

Figure 1. Axial CT image (A) shows isolated left sphenoid lesion with calcification within, and hyperostosis of left sphenoid sinus wall with erosion.

CORRECTED PROOF

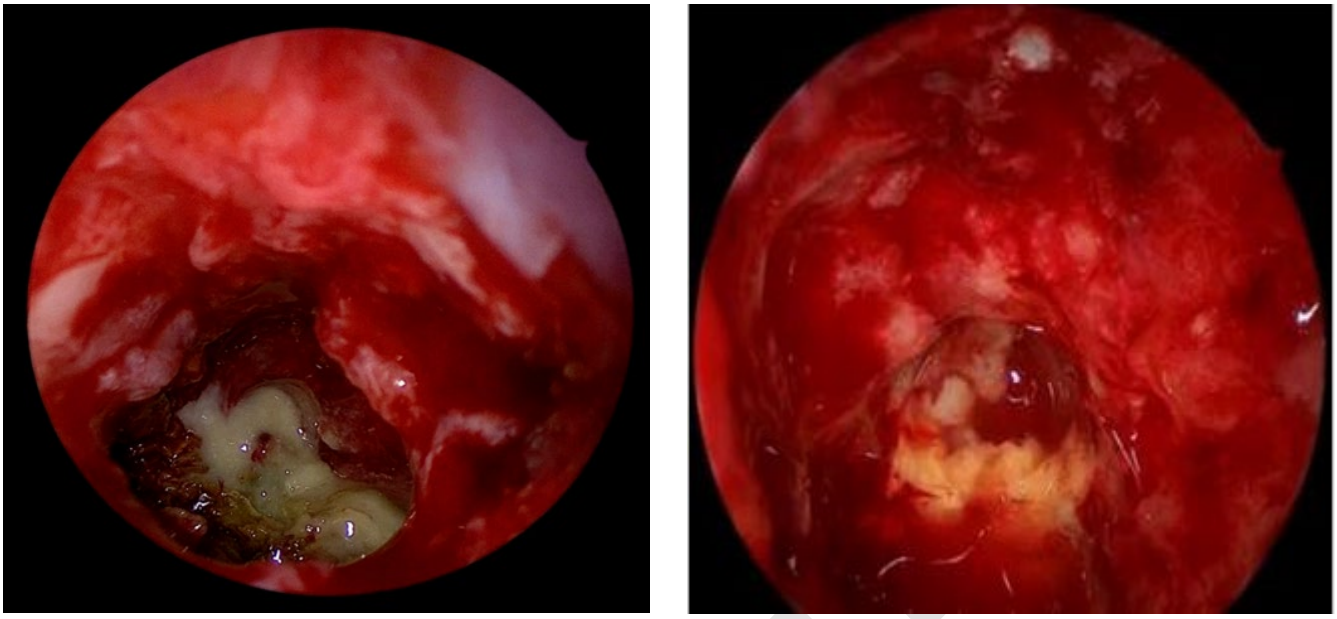

Figure 2. An intra-operative findings of fungal ball and pus inside the sphenoid sinus.

are the primary route by which fungi are introduced into the sinus. Previous mucosal injuries resulting from surgery may contribute to this process. Additionally, anatomical variations, such as ostial obstruction or chronic rhinosinusitis itself, may play a role in the pathophysiology of fungal balls.[4] In this particular case, the patient's well-controlled diabetes mellitus may increase her risk for fungal sinusitis, highlighting the importance of recognising systemic conditions as potential risk factors.

Isolated sphenoid fungal sinusitis, though rare, typically presents with atypical headache or rhinological symptoms. The headache usually has varying intensities and locations, which do not respond to analgesics and are worsened by head movement.[5] This symptom distribution correlates with the sinus's innervation via the trigeminal nerve and sphenopalatine ganglion afferent fibres.[3] In our case, headache is the only presenting symptom that makes the diagnosis difficult and causes delayed referral to us. The literature demonstrates that most patients will present with headaches, mainly described as being retro-orbital or frontal.[2,5,6] These cases, along with ours, highlight the rarity of the condition and the importance of including sphenoid sinus pathology in the differential diagnosis of chronic headaches.

Diagnosing isolated sphenoid fungal sinusitis is challenging due to its nonspecific manifesting symptoms. Nasoendoscopy is a crucial diagnostic tool for detecting pathologies in the paranasal sinuses. However, the findings in patients with isolated sphenoid sinus disease may not be prominent. Reports have shown that normal nasoendoscopy findings do not exclude any pathological abnormalities in 50% of the cases.[7,8] The diagnosis relies heavily on CT and magnetic resonance index (MRI). Imaging is crucial in pre-operative evaluation and post-operative surveillance. In our case, incidental findings from the CT brain due to the headache led to the diagnosis. A fungal ball usually manifests on CT as a hyperdense mass with central calcifications (punctuate or linear) confined to a single sinus due to matted fungal hyphae. Sclerotic, thickened, and bowed bony sinus walls without signs of invasiveness are commonly seen.[9] On T1 weighted MRI, fungal balls typically show intermediate/low signal intensity images and extremely low signal intensity on T2 weighted images.[9] Although CT is still the modality of choice for diagnosis, MRI may provide value to evaluate extra sphenoidal invasion. Early imaging studies focusing on the sphenoid sinus can facilitate timely diagnosis and management, preventing potential complications such as optic neuritis, cavernous sinus thrombosis, or intracranial spread.

Treatment options usually depend on the diagnosis. For isolated sphenoid lesions, the treatment of choice is surgery. The preferred surgical technique for inflammatory disease affecting only the sphenoid sinus is endoscopic trans-nasal sphenoidectomy,[10] as this technique has the advantage of sparing the ethmoid sinus mucosa.[4] At the same time, the trans-ethmoidal approach is commonly used for non-isolated sinusitis or sphenoid sinus tumours[10] when considered to combine with the adjacent extension to the posterior ethmoid sinus.[4] We chose the transnasal approach sphenoidotomy due to the isolated sphenoid lesion, which did not involve any nearby structures. Medical management, such as systemic antibiotics, can be initiated for infectious and inflammatory sphenoid sinusitis cases. Nonetheless, systemic antifungal medications can be started in invasive fungal sinusitis. Antifungal medication is not necessary in our case.

6. CONCLUSIONS

(1) Chronic, nonspecific headaches that do not respond to conservative treatment should raise a high suspicion of isolated sphenoid fungal sinusitis and necessitate referral to otorhinolaryngologist for further assessment.

CORRECT PROOF

(2) Early referral to the otorhinolaryngology team can prevent complications and prompt earlier curative surgery.

Conflict of interest
None declared.

Funding
None declared.

References
1 Rossi IR, Levinson J, Kuan EC, Tajudeen BA. Chronic Sphenoiditis With Deep Neck Space Extension: Case Report With Review of the Literature and Postulated Mechanisms for Extracranial Extension. *Ear Nose Throat J.* 2024;103(3):151–155. https://doi.org/10.1177/0145561321989453.

2 Leroux E, Valade D, Guichard JP, Herman P. Sphenoid fungus balls: Clinical presentation and long-term follow-up in 24 patients. *Cephalalgia*. 2009;29(11):1218–1123. https://doi.org/10.1111/j.1468-2982.2009.01850.x.

3 Ishak NL, Subha ST, Abu Bakar S. Isolated sphenoid sinusitis: A big headache. *Malays Fam Physician*. 2019;14(1):29–30.

4 Kim JK, Jung SJ. A Case of the Sphenoid Fungal Sinusitis Under Local Anesthesia. *World J Surg Surgical Res*. 2024;7:1548.

5 Ruoppi P, Seppä J, Pukkila M, Nuutinen J. Isolated sphenoid sinus diseases: report of 39 cases. *Arch Otolaryngol Head Neck Surg.* 2000;126(6):777–781. https://doi.org/10.1001/archotol.126.6.777.

6 Ravindra A, Shetty S, Devaraja K, Pujary K. Isolated sphenoid sinus disease; a retrospective review of underlying pathologies and their clinicopathological implications. *Int J Otorhinolaryngol Head Neck Surg.* 2022;8(3):217–222. https://doi.org/10.18203/issn.2454-5929.ijohns20220471.

7 Sethi DS. Isolated sphenoid lesions: diagnosis and management. *Otolaryngol Head Neck Surg.* 1999;120(5):730–736. https://doi.org/10.1053/hn.1999.v120.a89436.

8 Aydın E, Akıdil AÖ, Ateş Evren G, Sayın İ, Güneş S, Yazıcı ZM. Isolated Sphenoid Sinus Lesions. *Eur J Rhinol Allergy.* 2019;2(2):41–44. https://doi.org/10.5152/ejra.2019.203.

9 Popolizio T, Perri M, Balzano RF, et al. Isolated fungus ball in sphenoid sinus: tips and pitfalls of T1 hyperintense lesions. *BJR Case Rep.* 2018;4(2):20170081. https://doi.org/10.1259/bjrcr.20170081.

10 Villemure-Poliquin N, Nadeau S. Surgical treatment of isolated sphenoid sinusitis – A case series and review of literature. *Int J Surg Case Rep.* 2021;79:18–23. https://doi.org/10.1016/j.ijscr.2020.12.091.

CORRECTED PROOF