J M U H
Journal of Medicine and Urban Health

CASE REPORT

Fungal Ball of the Sphenoid Lateral Recess with Infraorbital Nerve Hypoesthesia: A Case Report and Literature Review

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Received 10 June 2025
Revised 11 September 2025
Accepted 11 September 2025
J Med Urban Health
2026;70(1):e7154
https://doi.org/10.62691/jmuh.2026.7154

ABSTRACT

A fungal ball in the lateral recess of the sphenoid sinus (LRSS) is a rare entity, particularly when it presents atypically as facial hypoesthesia. Due to its deep location and proximity to critical neurovascular structures, it can present with atypical symptoms and radiological features that can mimic skull base malignancy, complicating diagnosis. Only one similar case has been previously reported in the literature. We report a case of a 59-year-old male who presented with a one-month history of progressive headache and left facial numbness. Neurological examination revealed diminished pinprick sensation in the infraorbital nerve distribution. A computed tomography showed a hypodense mass with rim calcification and pterygoid bone erosion adjacent to the left sphenoid sinus, raising suspicion of an invasive skull base lesion. The patient underwent endoscopic transpterygoid sphenoidotomy, which revealed fungal concretions encased within a bony partition in the LRSS. Histopathological analysis confirmed *Aspergillus* spp. without mucosal invasion or malignancy. Postoperatively, the patient recovered from facial numbness and had an uneventful one-year follow-up.

Keywords: fungus, fungal ball, lateral recess, endoscopic sinus surgery, transpterygoid approach

INTRODUCTION

Fungal rhinosinusitis (FRS) is a spectrum of conditions classified into non-invasive and invasive forms, each with distinct clinical features, management strategies, and prognoses. Non-invasive FRS includes saprophytic fungal infestation, fungal ball, and allergic fungal rhinosinusitis (AFRS); these forms are typically confined to the sinus cavity without mucosal or bony invasion and often present with mild or nonspecific symptoms such as nasal congestion, purulent rhinorrhea, or facial pain.[1] AFRS frequently occurs in atopic individuals and is characterized by nasal polyposis and eosinophilic mucin containing fungal hyphae.[1,2]

In contrast, invasive fungal rhinosinusitis (IFRS) is categorized into acute and chronic forms, with the acute type rapidly affecting immunocompromised patients and often resulting in severe complications such as orbital or intracranial involvement, while the chronic form progresses more slowly and can affect immunocompetent hosts.[1,3] Invasive types are associated with a significantly worse prognosis and require immediate surgical intervention, antifungal therapy, and correction of immune status, unlike non-invasive forms, which rarely threaten life. This distinction is important to prevent potentially catastrophic outcomes.[1,3,4]

The fungal ball is a non-invasive form of FRS that causes mucosal inflammation and bone reaction without soft tissue or vessel invasion on histopathological examination.[1,4,5] It is commonly found in the maxillary sinus of immunocompetent, middle-aged women.[1,5] Meanwhile, sphenoid sinus fungal ball (SSFB) is the second most commonly involved sinus (approximately 10%) after the maxillary sinus, often occurring in elderly patients and in the smaller side of the sphenoid sinus.[1,5-9]

The sphenoid sinus is located deeply within the skull, adjacent to the middle cranial fossa and cavernous sinus. In a very well-pneumatized sphenoid, the lateral recess can be found in the adjacent pterygoid bone, lateral to the foramen rotundum and vidian canal. Although fungal ball in the sphenoid is not uncommon, a fungal ball confined exclusively to the lateral recess of the sphenoid sinus (LRSS), with almost complete ossified bone septation between the sinus and recess, has never been reported in the literature to our knowledge. The location of isolated LRSS is also challenging, as it cannot be readily accessed via conventional sphenoidotomy.

The fungal ball of the sphenoid sinus commonly presents with headache, retro-orbital pain, postnasal drip, or visual loss.[5,10] However, this report describes a fungal ball located in the LRSS, atypically presenting with numbness of cranial nerve V2 (CNV2), demonstrating calcified septation between the sphenoid sinus and the LRSS on computed tomography (CT), and requiring an endoscopic transpterygoid approach for removal of the fungus.

CASE REPORT

A 59-year-old Thai male presented with a progressive left-sided headache for one month. He also reported left-sided facial numbness but denied sinonasal symptoms, facial pain, facial palsy, ocular symptoms, or loss of smell. He had no significant past medical history, including absence of underlying disease, use of immunosuppressive therapy, recent antibiotics, or herbal medicines.

On physical examination, bilateral inferior turbinate hypertrophy was noted, with no evidence of discharge from the osteomeatal complex or sphenoethmoidal recess. No nasal polyps or mass were detected. Neurological examination revealed decreased pinprick sensation in the distribution of the CNV2. Ophthalmologic assessment showed bilateral visual acuity of 20/40 with full extraocular muscle movement, negative relative afferent pupillary defect, and normal light reflex.

A contrast-enhanced CT scan of the paranasal sinuses demonstrated almost complete opacification of the left sphenoid sinus with a hypodense mass surrounded by ring enhancement. Bony erosion was identified at the inferomedial plate of the pterygoid process. Based on these findings, the provisional diagnosis included a sphenoid sinus tumor with bony erosion and cranial nerve involvement **(Figure 1)**.

The operation started with left uncinectomy, maxillary antrostomy, complete anterior and posterior ethmoidectomy, and wide sphenoidotomy. After the sphenoid sinus was opened via the transethmoidal approach, the mucosa within the sinus was markedly swollen, making visualization and access to the LRSS difficult despite the application of topical decongestant. In routine sphenoidotomy, if the LRSS is not readily visible after lateral widening of the ostium, surgeons would avoid drilling or removing the lateral wall because of the risk of internal carotid artery (ICA) injury. Consequently, the left transpterygoid approach was performed by removing the pterygoid base that forms the anterior wall of the LRSS to directly access the pathology.

The lateral nasal wall mucosa over the palatine bone at the posterior wall of the maxillary sinus was elevated, and the sphenopalatine artery (SPA) was identified. The crista ethmoidalis of the palatine bone was removed using a Kerrison Rongeur, and SPA ligation was performed with bipolar cautery and subsequently cut. The periosteum and contents of medial pterygopalatine fossa were tracked laterally, until reaching the pterygoid base of the sphenoid bone, where the vidian nerve from the vidian canal and CNV2 from the foramen rotundum could be identified. However, the vidian nerve was inevitably sacrificed to achieve maximum exposure of the LRSS. Finally, the base of the pterygoid bone was drilled to provide access to the lateral recess and to fully delineate all walls of the LRSS.

Intraoperative findings revealed a fungal concretion encased within a bony partition separating the lateral recess from the sphenoid sinus cavity. Localized bony erosion was noted at the base of the pterygoid bone. The mucosa appeared mildly pale and polypoid without ischemic or necrotic changes. The fungal concretion and adjacent mucosa were sent for

 https://doi.org/10.62691/jmuh.2026.7154

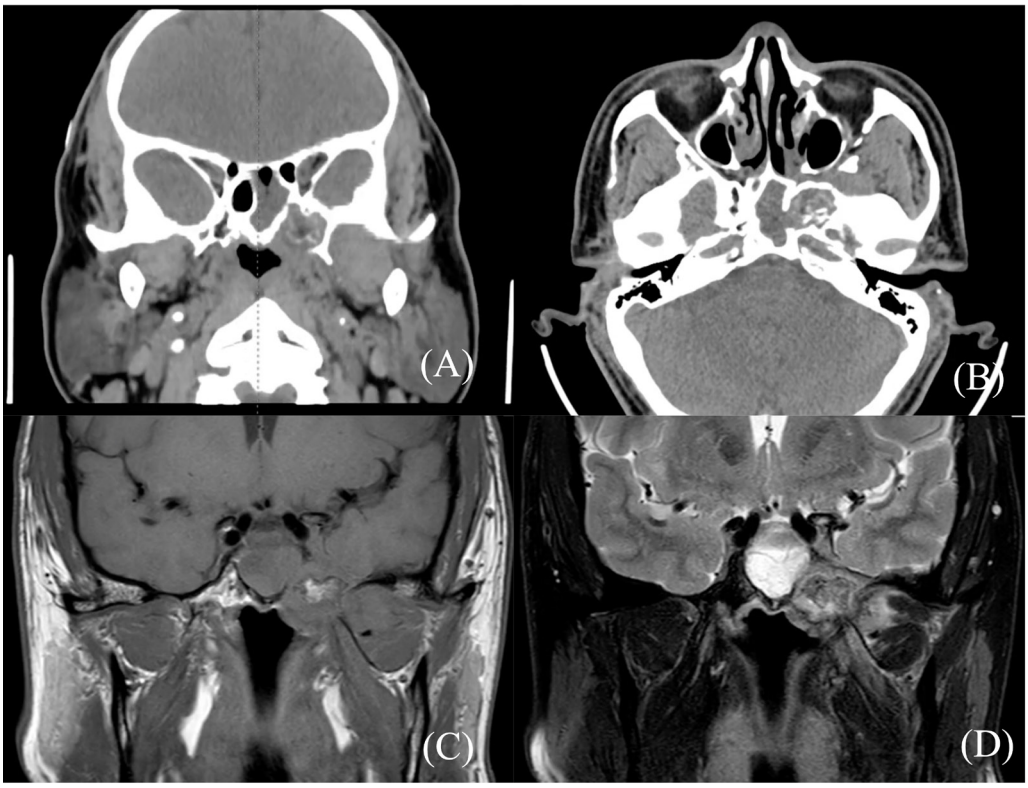

Figure 1 Computed Tomography scan showing opacification of the left sphenoid sinus with sclerotic bone change, a hypodense lesion with peripheral hyperdensity within the lateral recess of the sphenoid sinus (LRSS), and erosion of the greater wing of the sphenoid bone: coronal (A) and axial (B) views. Magnetic Resonance Imaging reveals a hypointense lesion within the left sphenoid sinus and lateral recess on a T1-weighted image (C), and a hyperintense lesion within the left sphenoid sinus together with a hypointense lesion within the left LRSS (D).

pathological examination, including Gomori Methenamine Silver (GMS) and Periodic Acid-Schiff (PAS) staining for fungal identification. The sclerotic septation bone between the lateral recess and the main sphenoid sinus was drilled to reconnect the sphenoid sinus cavity and the lateral recess **(Figure 2)**.

Postoperatively, the patient was advised to use budesonide 1 mg mixed with 250 mL normal saline for nasal irrigation once daily in the morning, and saline irrigation alone in the evening. Oral amoxicillin-clavulanic acid 1000 mg was also prescribed twice daily for 7 days.

The pathological report confirmed the presence of fungal hyphae consistent with *Aspergillus* spp. without evidence of mucosal invasion or malignancy. At the four-week follow-up, the patient developed dry eye, which was evaluated by an ophthalmologist and managed with artificial tear drops. At the three-month follow-up, the facial numbness had significantly improved, and the dry eye had completely resolved. There was no recurrence of the fungal ball, after one-year follow-up.

The study was approved by the Ethics Committee of Rajavithi Hospital (Trial No. 67015) on March 20th, 2024.

DISCUSSION

Fungal ball is classified as a secondary localized chronic rhinosinusitis according to the European Position Paper of Rhinosinusitis and Nasal Polyps 2020.[2] Fadda et al. outlined clinicopathological diagnostic criteria for fungal ball, which include sinus opacification with or without flocculent calcification, presence of cheesy or clay-like material within the sinus, matted and dense conglomeration of hyphae separate from sinus mucosa, chronic inflammatory response without eosinophil predominance, granulomatous response, or allergic mucin, and no histologic evidence of fungal invasion of mucosa, blood vessels, or underlying bone.[10] In clinical practice, the diagnosis of a fungal ball is primarily based on the presence of fungal concretions within the paranasal

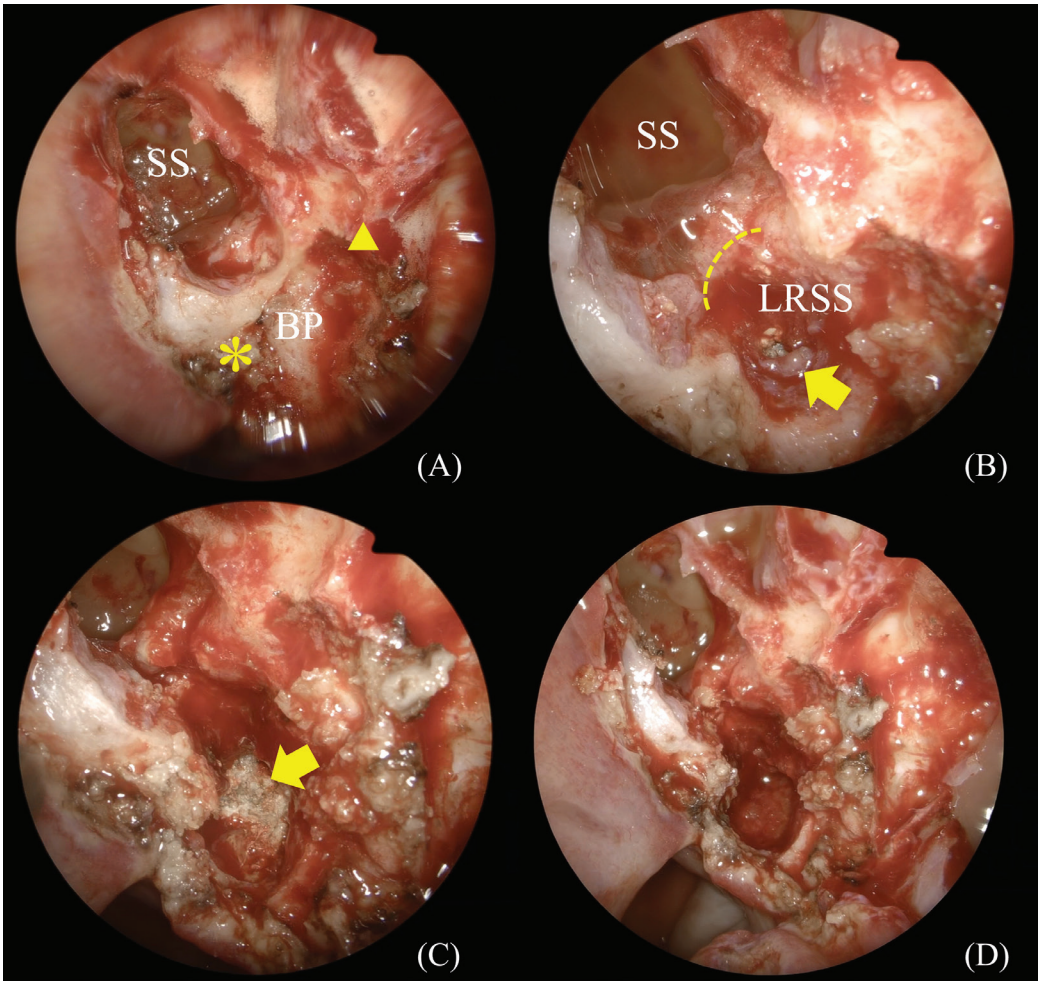

Figure 2 (A) Intraoperative findings of the transpterygoid approach showing the sphenoid sinus, base of the pterygoid process, vidian canal (asterisk), and foramen rotundum (triangle). (B) After drilling the base of the pterygoid process, the lateral recess of the sphenoid sinus (LRSS) was exposed, and the bony partition (dashed line) was visualized; the fungal ball was partially visible (arrow). (C) Surrounding soft tissue and bone were drilled, and the fungal ball was fully exposed (arrow). (D) The fungal ball was removed through irrigation and suction.

sinus, without the features of AFRS or IFRS. Despite our patient presenting with cranial nerve involvement and bony destructive lesion of the pterygoid bone, the diagnosis remains consistent with fungal ball based on the pathological criteria.

The sphenoid sinus represents the second most frequent site of fungal ball involvement following maxillary sinus, with reported prevalence rates ranging from 10% to 25% across studies.[5,7,8,11] Current demographic data indicate a predilection for immunocompetent elderly female patients, though the precise pathophysiological mechanisms underlying this epidemiological pattern remain under investigation.[7-9,12] While definitive risk factors remain unestablished, clinical observations suggest potential associations with prior sinonasal surgical interventions and diabetes mellitus in select case series.[5,6,12] Furthermore, Meerwein et al. found that the smaller sphenoid side may be more frequently affected (78%).[9]

Clinical presentations of SSFB include headache, retro-orbital pain, postnasal drip, or visual loss.[5,10] Furthermore, atypical presentations such as facial numbness, hyposmia, or hypogeusia have also been reported.[7] Some SSFB patients who visit the clinic without any symptoms may have incidental findings on CT or magnetic resonance imaging (MRI) performed

 https://doi.org/10.62691/jmuh.2026.7154

for other reasons. Facial numbness in the distribution of CNV2 is a distinctive clinical presentation in this case.

Jiang et al. reported the symptoms of 77 patients with SSFB including: headache (79.2%), rhinological symptoms (29.9%), eye symptoms (7.8%), and asymptomatic (9.1%).[6] Likewise, Leroux et al. observed in 24 patients that 62% had headache, 21% had rhinological symptoms, and 16% were asymptomatic. Notably, only one patient in their study presented with CNV2 hypoesthesia.[7] Hence, this rare presentation of CNV2 hypoesthesia in our patient with SSFB underscores the importance of considering a wide range of symptoms when evaluating patients with SSFB.

The bony walls of the paranasal sinuses can be affected by pathology within the sinus cavity. Chronic rhinosinusitis with or without nasal polyps produces persistent inflammation of the sinus wall, which consequently leads to proliferation of the periosteum, bone remodeling, and neo-osteogenesis, manifesting as osteitic or sclerotic bone changes on radiographic study.[13,14] Furthermore, benign sinonasal tumors such as inverted papilloma, mucocele, AFRS and fungal balls progressively increase intrasinus pressure against the sinus walls and similarly enhance neo-osteogenesis.[1,15,16] Conversely, in cases of sinonasal malignancy or IFRS, these diseases invade the sinus walls and compromise the vascular supply, resulting in bone resorption and erosion.[1,17] Considering this case, the pattern of bone erosion raises concern for aggressive disease, as it lacks the bony strut typically seen in inverted papilloma, the localized sclerotic bone found in a typical fungal ball, the diffuse osteoneogenesis characteristic of AFRS, or the sinus wall ballooning expansion observed in mucocele.

Specifically, osteolytic lesions in fungal ball cases are less common than sclerotic lesions. The prevalence of osteolysis compared to sclerosis in SSFB has been reported in several studies: Jiang et al. observed 41.6% vs. 93.5%, Leroux et al. found a prevalence of 25% vs. 44%, and Kim et al. reported 40% vs. 64%[6-8] The underlying pathophysiology of osteolytic bone lesions is not fully understood, but it is hypothesized to result from mechanical pressure, chronic inflammation, and/or fungal metabolites and enzymes.[11,15,18] In our case, the bone erosion may have resulted from a combination of the hypothesized mechanisms, whereby the fungal concretion was confined within a small partition of the sphenoid sinus characterized by marked mucosal inflammation and bony septation. This anatomical configuration limited effective drainage into the sphenoid antrum, thereby increasing pressure within the LRSS, while fungal enzymes further disrupted normal bone healing and resorption processes. Thus, fungal ball should remain a consideration in the differential diagnosis for sphenoid sinus wall erosion on CT or MRI, even though sinonasal malignancy and IFRS should be of greater concern.

Endoscopic sphenoidotomy is the preferred treatment for removing fungal balls in the sphenoid sinus.[7,8,12] Meier et al. reviewed forty-three patients with SSFB who underwent sphenoidotomy using different approaches, including transethmoidal sphenoidotomy (76.7%) and transnasal sphenoidotomy (23.3%), to provide sufficient access.[12] However, the transpterygoid approach may be necessary in cases where the tumors or tumor-like lesions are located in the LRSS.[19,20] This approach involves partial or total removal of the pterygoid bone, enabling access to deeper anatomical regions, including the pterygopalatine fossa, LRSS, petrous apex, Meckel's cave, infratemporal fossa, and the middle or posterior skull base.[21]

In this case, anterior removal of the pterygoid base was required to access the LRSS. This was achieved by first identifying the vidian nerve and CNV2, followed by drilling the anterior face of the LRSS between both important landmarks. Through this approach, the fungal ball was adequately removed without limitation from the bony septum between the sphenoid antrum and LRSS. This necessity justified selecting the transpterygoid approach over the transnasal or transethmoidal approaches, which are more commonly used for fungal balls of the sphenoid sinus but may not provide sufficient access in scenarios where the fungal concretion is not connected to the main part of the sphenoid sinus.

The major concern with the transpterygoid approach is the risk of intraoperative and postoperative complications. Injury to critical structures such as the ICA and orbit may occur, particularly in cases of limited surgical experience or poor visualization.[20,21] Good anatomical knowledge, surgical skills, and the use of navigation systems can help minimize these risks. Li et al. classified pterygoid process pneumatization into three types: 1.) no identifiable LRSS, 2.)

superolateral pneumatization toward the greater wing of the sphenoid without extension below the vidian canal, and 3.) pneumatization of both the greater wing and the caudal pterygoid process.[22] In some cases, the vidian nerve can be preserved; however, lesions involving the superior or lateral wall of the LRSS may necessitate vidian neurectomy to achieve full exposure and surgical freedom. In the present case, the vidian nerve was sacrificed to maximize exposure of the LRSS and to allow evaluation of its mucosa and bony walls. The study of Lyu et al. show 16.7% develop dry eye at 1-2 month after vidian neurectomy, but the symptoms spontaneously resolved by 3-4 months with only conservative management such as artificial tears.[23] Preoperative counseling and postoperative ophthalmologic monitoring for dry eye are therefore essential.

Histopathology is essential for diagnosing and differentiating FRS, particularly distinguishing non-invasive fungal balls from IFRS. Routine fungal staining, such as GMS and PAS, confirms fungal elements and identifies pathogens. Biopsy of surrounding mucosa and bone helps rule out malignancy or secondary infections, especially in cases with bony erosion. Early histopathological confirmation facilitates appropriate next surgical intervention, if needed, reducing the risk of disease progression and recurrence.[24] Fungal culture is less important in cases of fungal balls, as only 30% yield growth, and *Aspergillus fumigatus* is typically identified.[25] A fungal ball was finally confirmed in our case, even though the preoperative findings suggested IFRS or sinonasal malignancy.

The transpterygoid approach was successfully employed for the fungal ball in the LRSS in our case. At the three-month follow-up, the CNV2 hypesthesia had spontaneously resolved, and there was no disease progression or recurrence at one year. Consequently, the temporary CNV2 numbness was more likely attributable to nerve compression or fungal toxin effects than to direct invasion or permanent neural injury.

Our case presentation demonstrates a rare finding of unilateral SSFB confined to the LRSS. The lesion is localized with sclerotic septation between the lateral recess and the sinus, mimicking a tumor within the pterygoid bone. The case also involved facial numbness in the CNV2 area, which completely resolved after surgery.

CONCLUSION

SSFB can mimic invasive fungal sinusitis or malignancy due to bony erosion and cranial nerve involvement, but it remains a non-invasive entity. CNV2 hypesthesia can occur without foramen rotundum erosion, likely due to local inflammation, bone reaction, or toxin release. An endoscopic transpterygoid approach is recommended for direct access to the LRSS. The SSFB should be included in the differential diagnoses of skull base lesions with cranial nerve involvement and bony erosion.

Conflict of Interest

The authors declare that they have no conflict of interest.

Acknowledgments

We thank the patient and his family for their permission and consent, which made this case report possible. No funding was received for this study.

Author Contributions

Conceptualization: W.C.
Data curation: V.P.
Formal analysis: V.P., W.C.
Funding acquisition: W.C.
Investigation: V.P., W.C.
Methodology: P.S., Wo.C.
Project administration: P.T., Pa.T.
Resources: N.A., I.R.
Software: N.S., T.D.
Supervision: W.C.
Validation: V.P., W.C.
Visualization: V.P.
Writing – original draft preparation: V.P.
Writing – review & editing: W.C.

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