

Isolated sphenoid sinus lesions: A tertiary Hospital experience

Original Article

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Abstract

Objective: To review and report isolated sphenoid sinus diseases from a tertiary hospital, with special attention to the patients' presenting symptoms, diagnosis, and clinical outcomes.

Study Design, Material and Methods: Retrospective chart review performed on patients diagnosed with isolated sphenoid sinus disease, undergoing endoscopic sinus surgery between January 2017 and December 2021.

Results: 19 patients were included, with a mean age of 48.0 ± 18.7 years. The most common symptom was headache (38.9%) followed by nasal obstruction (33.3%). Cranial neuropathies occurred exclusively in the group of malignant neoplasms. Inflammatory etiology was the most frequent (57.9%) and included fungal sinusitis (27%), chronic rhinosinusitis (18%) and mucocele (5%).

Conclusions: The diagnosis of isolated sphenoid lesions requires a high degree of suspicion since they can present with nonspecific symptoms. Early surgical intervention, for diagnostic or therapeutic purposes, is generally indicated, with nasosinusal endoscopic surgery being the most appropriate modality in most cases.

Keywords: Sphenoid sinus; isolated lesion; sinusitis

Introduction

Isolated sphenoid sinus lesions account for 1% to 2.7% of all lesions of the paranasal sinuses.^{1,2} Most lesions are of an inflammatory nature, and neoplastic etiology accounts for only 15–16% of the lesions.^{3,4} The increasing use of imaging methods such as computed tomography (CT) and magnetic resonance imaging (MRI) has contributed to an increase in the frequency of diagnosis of isolated sphenoid sinus lesions. However, these lesions remain a diagnostic challenge due to the insidious and non-specific clinical presentation.^{1,5} The most common symptom is frontal or retro-orbital headache, which may be associated with sinonasal symptoms such as rhinorrhea, nasal obstruction, hyposmia, or epistaxis.^{6,7}

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Despite the close relationship of the sphenoid sinus with the dura mater, cranial nerve pairs (III, IV, V1, V2, and VI), optic chiasm, optic nerves, cavernous sinus, hypophysis, internal carotid artery, sphenopalatine ganglion, sphenopalatine artery, and pterygoid canal, the symptoms arising from the involvement of the adjacent structures are less frequent and appear at a late stage.⁷⁻⁹

In this retrospective study, the authors aimed to analyze the etiology, symptoms, diagnosis, and surgical outcomes of a sample of 19 patients with isolated lesions of the sphenoid sinus who underwent diagnostic or therapeutic sinonasal endoscopy.

Material and Methods

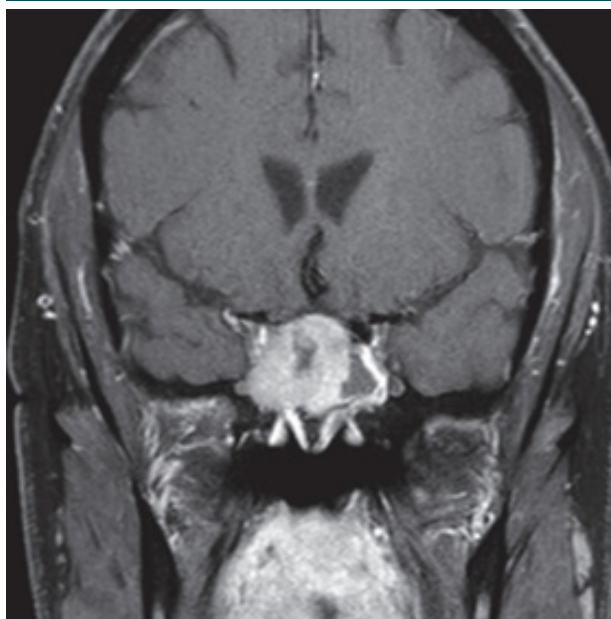
This was a retrospective and descriptive study of patients diagnosed with isolated lesions of the sphenoid sinus who underwent sinonasal endoscopy, with diagnostic or therapeutic intent, between January 2017 and December 2021. The following variables were analyzed: sex, age, presenting symptoms, means of diagnosis, etiology, microbiological and anatomic pathology study, surgical procedure, and surgical outcome of the macroscopic resection of the lesion, whenever applicable.

Results

The study included 19 patients, 73.7% (n=14) of which were men. Age varied between 15 and 77 years, with a mean age of 48.0 ± 18.7 years. The following lesions were identified (table 1): 11 lesions of inflammatory origin (57.9%), including six cases of fungal sinusitis (one invasive) and one case of mucocele, and seven neoplastic lesions (36.8%), of which three were malignant (metastatic secondary neoplasms). Additionally, one case of the spontaneous cerebrospinal fluid fistula was included without previous history of sinonasal surgery or brain injury.

The most frequent symptom was headache or retro-orbital pain (n=7, 38.9%), followed by nasal obstruction (n=6, 33.3%). Posterior rhinorrhea (n=1, 5.3%) and purulent anterior rhinorrhea (n=1, 5.3%) were reported only in

Figure 1
Coronal section magnetic resonance imaging (MRI) T1 weighted image with gadolinium of a patient with sphenoid myeloid sarcoma



cases of sphenoid mycetoma and fibrous dysplasia, respectively. Symptoms compatible with the involvement of the paired cranial nerve pairs occurred exclusively in the group of malignant neoplasms and included two patients with diplopia, one patient with eyelid ptosis, one patient with facial hypoesthesia, and one patient with diminished visual acuity. In five patients (27.8%), who were asymptomatic, the diagnosis was incidental following imaging exams. The distribution of symptoms according to the diagnosis group is listed in Table 2.

For the initial diagnosis, all patients underwent at least one imaging exam: 13 patients (68.4%) underwent CT complemented by MRI, while six patients (31.6%) only underwent CT. The group whose imaging evaluation was complemented by MRI includes all cases of malignant neoplasms, all cases of spontaneous cerebrospinal fluid fistula, and a single case of invasive fungal rhinosinusitis.

In patients with a diagnosis of fungal sinusitis and chronic rhinosinusitis without nasal polyps, the samples collected intraoperatively were submitted for bacteriological and mycological analysis, whose results are

Table 1
Distribution of the identified diseases, per category of diagnosis

Group of diagnosis		Number (%) of patients	Observations
Inflammatory disease (n=11, 57.9%)	CRSwNP	2 (9 %)	
	CRSwoNP	2 (9 %)	Iatrogenic (n=1), post-surgery for the excision of pituitary macroadenoma via transsphenoidal approach
	Fungal sinusitis	6 (27%)	Invasive (n=1), non-invasive (n=5, including four mycetomas and one AFRS)
	Mucocele	1 (5 %)	
Neoplastic disease (n=7, 36.8%)	Benign	4 (18 %)	Inverted papilloma (n=2), fibrous dysplasia (n=1), glomangiopericytoma (n=1)
	Malignant	3 (27 %)	Metastasis of urothelial carcinoma (n=1), paraclival metastasis of follicular thyroid carcinoma (n=1), myeloid sarcoma (n=1)
Others (n=1, 5.3 %)		1 (5.3 %)	Spontaneous cerebrospinal fluid fistula

CRSwNP – Chronic rhinosinusitis with nasal polyps; CRSwoNP - Chronic rhinosinusitis without nasal polyps; AFRS – Allergic fungal rhinosinusitis

shown in Table 3. *Aspergillus* sp. was the most frequently isolated strain in the mycological analysis. Additionally, cultures of the patient with allergic fungal rhinosinusitis showed *Schizophyllum* sp., *Klebsiella pneumoniae*, and *Streptococcus anginosus*. In four patients, including three with a diagnosis of sphenoid mycetoma, the cultures were negative. Except for one patient with invasive fungal rhinosinusitis and two patients with metastatic secondary neoplasms (endoscopy procedure with diagnostic intent), complete macroscopic removal of the lesions was

achieved via the endoscopic transnasal (n=12, 63.2%) or transethmoidal approach (n=9, 47.4%), without postoperative complications. Three deaths occurred during the follow-up period (15.8%): one patient with invasive fungal rhinosinusitis (10 days after surgery due to massive epistaxis); one patient with urothelial carcinoma metastasis (16 days after surgery due to progression of the underlying disease); one patient with thyroid follicular carcinoma metastasis (30 months after surgery due to progression of the underlying disease).

Table 2
Symptoms according to the diagnosis category

Diagnosis category (N, %)	Presentation symptoms (N, %)					
	Headache	Nasal obstruction	Posterior rhinorrhea	Anterior rhinorrhea	Cranial neuropathy	CSF
Inflammatory disease (n= 11, 57.9%)	rhinorrhea	3 (15.8%)	1 (5.3%)	0 (0)	0 (0)	0 (0)
Neoplastic disease (n= 7, 36.8%)	2 (10.5%)	3 (15.8%)	0 (0)	1 (5.3%)	2 (10.5%)	0 (0)
Other* (n=1, 5.3%)	0 (0)	0 (0)	0 (0)	0 (0)	0 (0)	1 (5.3%)
Total (N=19)	7 (36.8%)	6 (31.6%)	1 (5.3%)	1 (5.3%)	2 (10.5%)	1 (5.3%)

*Spontaneous cerebrospinal fluid fistula is included in this group. CSF, cerebrospinal fluid

Table 3
Strains isolated from cultures

Isolated strain	Number (%) of patients
<i>Staphylococcus aureus</i>	1 (12.5%)
<i>Pseudomonas aeruginosa</i>	1 (12.5%)
<i>Klebsiella pneumoniae</i>	1 (12,5%)
<i>Aspergillus sp.</i>	2 (25%)
<i>Mucor</i>	1 (12,5%)
<i>Schizophyllum sp.</i>	1 (12,5%)
Negative cultures	4 (50%)

Note: In some samples, more than one strain was isolated. The percentages were calculated based on the total number of patients who underwent cultures (n=8).

Discussion

Isolated sphenoid sinus lesions are rare, accounting for <3% of sinus lesions and <0.05% of malignant neoplasms of the paranasal sinuses.^{2,10} Historically, the sphenoid sinus was widely neglected because it is an anatomic area with difficult access and because its lesions have an insidious onset, with few signs or symptoms, which causes delayed diagnosis.¹¹ In 1973, Wyllie et al. published one of the first large case series of isolated sphenoid sinus lesions with only 63 cases retrieved over 37 years.⁸ A growing number of cases are being reported with the increasing availability of imaging exams such as CT and MRI. In 2000, Cakmak et al. completed the series previously published by Wyllie et al. to report the largest series available to date in the English literature, which included 182 patients.⁹

In this study, inflammatory disease was the most frequent cause of isolated sphenoid sinus lesions (57.9%), which is in line with the findings of previous studies that show a prevalence of 61–80%.^{1,2,4} In the most recently published case series, chronic rhinosinusitis without nasal polyps accounted for 28.3% of cases, followed by mucocele (20.3%), fungal sinusitis (12.5%), and chronic rhinosinusitis with nasal polyps (3.4%).⁷ In our sample, in the group of inflammatory diseases, fungal sinusitis was the most frequently diagnosed entity, with mucocele making up only 5.3%

of the diagnoses. This discrepancy may be a result of the small sample size.

In the group of neoplastic disease, the prevalence of malignant neoplasms was higher than that of benign neoplasms (27% and 18%, respectively), which is in line with the findings in the literature.^{4,7,8} Lawson et al., in 1987, and later Cakmak et al., in 2000, demonstrated a higher prevalence of primary malignant neoplasms than metastatic secondary disease.^{4,9} However, the analysis of this study sample showed that malignant sphenoid neoplasms occurred exclusively due to metastatic disease.

Cerebrospinal fluid fistulas represent 4.1%¹² to 8%³ of isolated sphenoid sinus lesions. This study only included one case of cerebrospinal fluid fistula in a patient with no previous history of sinonasal surgery or brain injury.

Headache was the most frequent symptom at clinical presentation (present in 38.9% of patients). In a study published by Moss et al., which is one of the most representative reviews in the literature to date with 1133 patients, the authors reported a headache prevalence of 71.4% and could not establish an association between the diagnosis and location of the headache.⁷

Most of the studies reported a higher prevalence of cranial neuropathy with neoplastic lesions. Friedman et al. demonstrated the involvement of cranial nerve pairs in 50% of patients with a diagnosis of sphenoid neoplasm, compared to 8% of patients with inflammatory disease.³ Wang et al. reported visual changes in 26% of patients with benign disease, compared to 50% of patients with a diagnosis of malignancy.² Diplopia and decreased visual acuity, due to the involvement of the oculomotor and optic nerves, respectively, are the most commonly reported impairments.⁷ The results of this study were similar, with cranial neuropathy occurring exclusively in the group of malignant neoplasms.

Brook showed a higher prevalence of *S. aureus* in acute sphenoid sinusitis, with anaerobes and gram-negative bacilli being the most common strains in the chronic forms

of the disease.¹³ Despite the variety of strains isolated in fungal sinusitis, *Aspergillus* sp. was the most frequently isolated¹⁴ strain in this study sample. However, the small number of cultures performed does not allow reliable extrapolation of the results. In four patients, including one with chronic rhinosinusitis without nasal polyps and three with sphenoid mycetoma, the cultures were negative. In the first case, the negative bacteriological exam can be explained by antibiotic therapy given prior to sample collection. In other patients, as the imaging and intraoperative findings were highly suggestive of sphenoid mycetoma, the results may be false negative due to inadequate sampling or slow-growing, fastidious agents.

CT is a key diagnostic tool, particularly in patients without visible alterations in nasal endoscopy. In the case of bone erosion or expansive lesion, especially if the intracranial or orbital extension is suspected, CT should be complemented by MRI.¹⁵

Conclusion

Isolated sphenoid sinus pathology is a rare entity accompanied by non-specific symptoms and has an insidious course. Early diagnosis relies on a high index of suspicion, complemented by the careful use of imaging studies. Early surgical intervention, for diagnostic or therapeutic purposes, is generally indicated. Endoscopic sinonasal surgery is safe and effective and is the most adequate surgical modality in most cases.

Conflicts of Interest

The authors declare that there is no conflict of interests regarding the publication of this paper.

Data Confidentiality

The authors declare having followed the protocols in use at their working center regarding patients' data publication.

Protection of humans and animals

The authors declare that the procedures

were followed according to the regulations established by the Clinical Research and Ethics Committee and to the 2013 Helsinki Declaration of the World Medical Association.

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Availability of scientific data

There are no datasets available, publicly related to this work.

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