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Solitary sphenoid sinus benign lesions: management and prognostic values as retrospective audit of seven case series

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Abstract

Background Solitary sphenoid sinus pathology is uncommon. The deep position of the sphenoid sinus makes symptoms related to solitary sphenoid sinus pathology non-specific and contributes to a significant diagnosis delay. Moreover, surrounding anatomical elements cause its pathology to be potentially serious and make surgical management challenging.

Methods This retrospective study includes 7 cases presented with primary benign sphenoid sinus pathology managed between January 2019 and January 2022. Epidemiological features, clinical aspects, and treatment modalities are presented.

Results Six female patients and one male patient were operated; their age ranges from 11 to 67 years old. Etiologies are divided into infection, tumors, and pseudo tumors. Sphenoid sinus approach was performed through the sphenoethmoid recess in 5 cases, through the septum in one patient and through the ethmoid in one patient.

Conclusion Endoscopic procedures have significantly improved the management of the sphenoid sinus pathology. They offer good access and control of the sphenoid pathology but require a perfect mastery of radiologic and endoscopic anatomy and its variations.

Keywords Sphenoid sinus, Endoscopic endonasal surgery, Sphenoid pathology

Background

Isolated sphenoid sinus pathology is uncommon [1]. Solitary sphenoid sinusitis is found in 2.7–3% of all patients presenting with rhinosinusitis [1, 2]. Also, tumors of this location count for 15–16% of all solitary sphenoid sinus pathology [2]. Symptoms tend to be non-specific especially in early stages. Thus, sphenoid sinus is called *dumb sinus* in literature. The main symptom is headache of various intensity and locations. Therefore, patients are prone to neglect the symptom or see another medical practitioner than an otorhinolaryngologist [3]. Hence, some affections remain completely asymptomatic until complications happened [1]. Sieskiewicz et al. [1] reported that the delay in diagnosis of isolated sphenoid pathology was 10.2 months; this delay was different for ORL and "non-ORL" practitioners. In fact, when the patient consulted an ORL, the diagnosis was made in 10.8 months, and that delay was 9.5 months for "non-ORL" practitioners [1]. These results might be related to the fact that ORL usually see patients referred from another medical practitioner but also because endoscopy failed to demonstrate any alteration of the sphenoethmoidal recess in 31.2 to 50% of cases in isolated benign sphenoid

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sinus pathology [1, 4, 5]. Therefore, diagnosis is mainly based on imaging techniques. CT scan and MRI benefited the diagnosis and the management strategy as they give a good mapping of the lesion and help considerably plan surgery. Endoscopic procedures completely transformed sphenoid sinus surgery. Its indications kept enlarging from simple sinus drainage to more complex approaches to skull base pathology.

Methods

We operated on seven patients with primary sphenoid sinus pathology through an endoscopic endonasal approach from January 2019 to January 2022. All patient underwent routine preoperative assessment. Pain level, if present, was scaled using a VAS. We used rigid 0° or 30°, 4 mm, endoscope for nasal cavity examination. We ordered a CT scan to confirm the diagnosis and evaluate the underlying pathology. Gadolinium MRI imaging was indicated in selected cases. Comprehensive ophthalmological and neurological examination and specific laboratory tests were ordered according to the patient's case.

Trans-nasal approach was the main procedure performed in our series followed by the trans-ethmoidal approach and transseptal approach. Postoperative care included nasal packing when bleeding control using endonasal bipolar was insufficient. Nasal packing is removed after 24 to 48 h. Nasal endoscopy and crust removal were scheduled 10 and 20 days after surgery.

Results

Case 1

An 11-year-old male teenager with no significant medical history presented to the pediatric emergency department for severe headache complicating a right intermittent facial pain evolving for the past 2 months. No other symptoms were reported. Physical examination disclosed trismus with no significant dental decay, no oculomotor palsy, or another cranial nerve paralysis. Endoscopic examination was uneventful. CT scan coronal view (Fig. 1A) exhibits a 6-cm hypodense lesion filling the right sphenoid sinus and expanding to the homolateral infratemporal fossa with skull base damage. Bone seems hypertrophic in many sites with frosted-glass aspect that is pathognomonic of fibrous dysplasia. MRI coronal view (Fig. 1B) confirmed the diagnosis of mucocele of the right sphenoid sinus expanded to the infratemporal fossa. The patient underwent endoscopic trans-nasal sphenoidotomy for the marsupialization of the mucocele. In the postoperative course, facial pain, headache, and trismus resolved, and the patient was referred to the pediatrics for further management of the fibrous dysplasia.

Case 2

A 17-year-old female teenager with chronic gastroesophageal reflux presented to the ORL outpatient clinic with a chronic mild headache with discreet posterior rhinorrhea and recent foul-smelling. Endoscopic examination disclosed minimal purulent secretions of the right spheno-ethmoidal recess. CT scan coronal view (Fig. 2A) revealed a thickening along the bony walls of the right sphenoid sinus with central calcification compatible with sinus fungal ball. The patient underwent a trans-ethmoidal approach to the sphenoid sinus because of a very narrow nasal corridor. The postoperative course was uneventful. The patient received antibiotics, pain

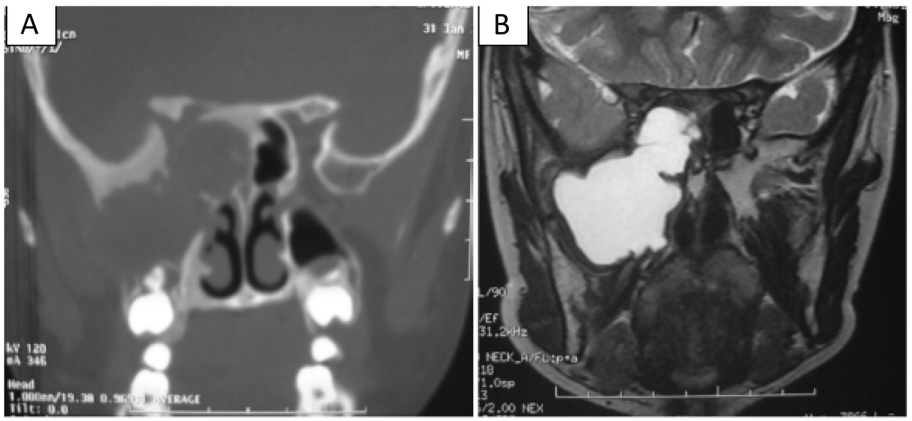

Fig. 1 Case 1. Right sphenoid sinus mucocele expanded to the infratemporal fossa. CT scan coronal view (**A** left) showing a 6-cm liquid area in the right sphenoid sinus and infratemporal fossa with skull base damage and several sites of hypertrophic bone with a frosted-glass aspect characteristic of fibrous dysplasia. MRI coronal view (**B** right) confirmed the inflammatory nature of the effusion (hypersignal on T2-weighted image), ruled out a diagnosis of meningocele, and confirmed the diagnosis of mucocele

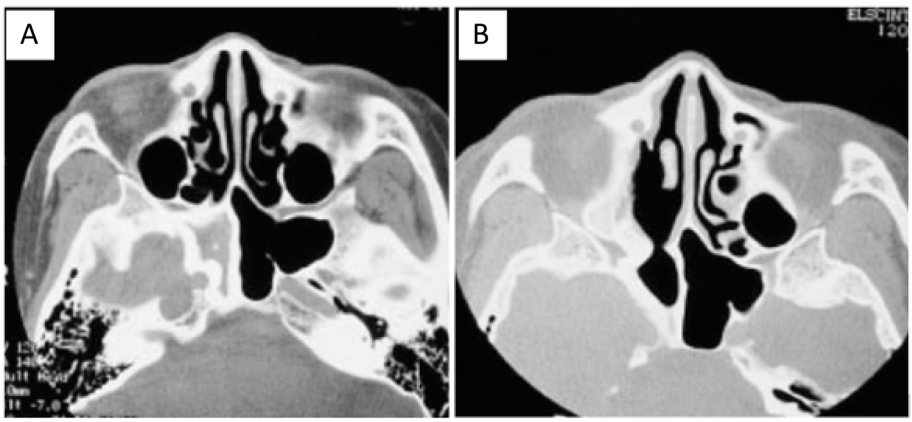

Fig. 2 Case 2. Right sphenoid sinus fungal ball. CT scan coronal view (**A** left) revealing a complete hypodense filling of the right sphenoid sinus associated with a regular thickening along the bony walls of the sinus with central hyperdensity. Postoperative CT scan (**B** right) of a transethmoidal approach to the right sphenoid sinus

medications, and saline nasal irrigation. Postoperative control CT scan was performed 2 months later (Fig. 2B).

Case 3

A 59-year-old woman with a history of high blood pressure and hyperuricemia was referred from her cardiologist to the ORL outpatient clinic for chronic headache rapidly worsening in the last 3 months and uncontrolled high blood pressure. Medical history revealed progressive nasal obstruction, sleep apnea, and exertional dyspnea. Endoscopic examination showed a submucosal tumor filling the nasopharynx. MRI sagittal view (Fig. 3A & B) showing a solid tumor of the sphenoid sinus expanded to the rhinopharynx with no intracranial involvement. The tumor was exclusively limited to the sphenoid sinus. The patient underwent endoscopic transseptal removal of the tumor. Pathology and immunochemistry examination revealed a chondroma. At 14-month follow-up, no evidence of recurrence was found on control endoscopic and radiologic examinations (Fig. 3C & D).

Case 4

A 19-year-old young women presented to the ORL outpatient clinic with a chronic unilateral permanent nasal obstruction associated with an intermittent unilateral rhinorrhea and mild headache. Endoscopic examination of the nasal cavity identified a pale translucid mass that seems to protrude from the right sphenoethmoid recess and nasopharynx. Left choanae and nasal fossa were normal. Sinuses computed tomography (Fig. 4A & B) revealed a soft tissue density mass of the right sphenoid sinus filling the choana and the posterior homolateral nasal cavity. We used an endoscopic trans-nasal approach for the removal of that polyp. Histopathology examination of the specimen is revealed as inflammatory polyp. There is no evidence of disease recurrence at 18-month follow-up.

Case 5

A 37-year-old female patient was admitted to the emergency department for an intense headache, diplopia, and vomiting. Investigation found a recent history of foul-smell and paroxysmal headache for the last 2 months. Physical examination disclosed a left internal rectus muscle palsy. Endoscopic examination revealed a bulging in the left sphenoethmoid recess. Neurological examination was unremarkable. Craniofacial CT scan (Fig. 5A) showed a total soft-tissue filling of the sphenoid sinus with clivus partial bone destruction. MRI T1-weighted sequence revealed an intermediate T1 signal total filling of the left sphenoid sinus (Fig. 5B). The lesion exhibited a hyposignal T2 and was surrounded by an inflammatory mucosa (Fig. 5C) with close contact to the surrounding vascular structures, all suggesting a pseudotumor invasive sinusitis.

Mass excision was performed through a trans-nasal sphenoidotomy. It exhibited a gray solid component with brown fluid surrounded by an inflammatory mucosa. Histopathologic examination of a biopsy specimen revealed mucosal infiltration with *Aspergillus* species without angioinvasion. The patient received systemic antifungal treatment based on voriconazole at 400 mg twice a day the first day and then 200 mg twice a day for 3 weeks with regular liver function tests. Postoperative CT scan (Fig. 5D) showed complete removal of the mass. However, the patient had persistent left internal rectus muscle paralysis.

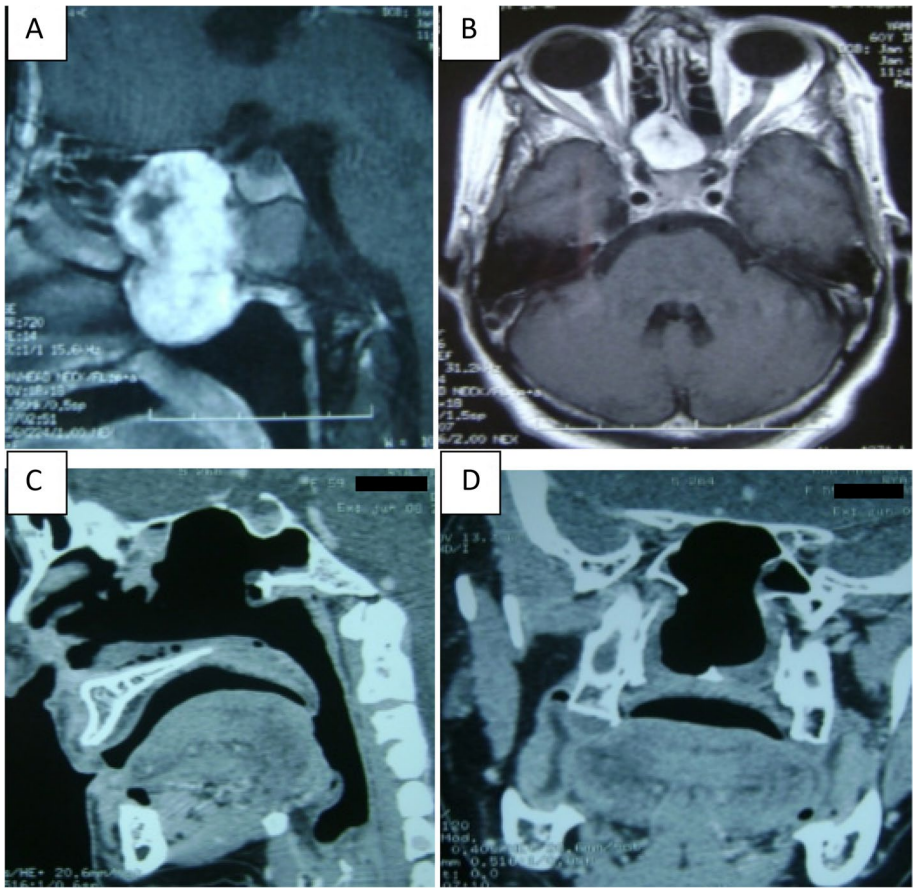

Fig. 3 Case 3. Sphenoid sinus chondroma. MRI sagittal view (**A** upper left) showing a tumor of the sphenoid sinus expanded to the rhinopharynx with no intracranial involvement. On MRI axial view (**B** upper right), the tumor does not involve surrounding structures. Postoperative CT scan sagittal view (**C** lower left) and coronal view (**D** lower right) showing a total removal of the sphenoid sinus tumor

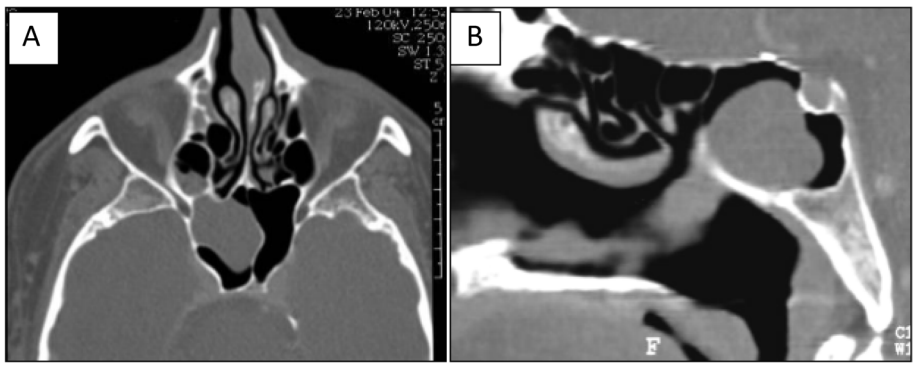

Fig. 4 Case 4. Spheno-choanal polyp. CT scan axial view (**A** left) showing a right sphenoid sinus mass with posterior ethmoidal cell's retention. Sagittal view (**B** right) showing a sphenoid sinus's well-defined mass expanded to the rhinopharynx suggesting a spheno-choanal polyp

Case 6

A 41-year-old female patient presented to the emergency department with severe headache, recent blurry vision, and vomiting, 5 days after the onset of a common cold episode. Symptoms did not resolve after antibiotics that the patient received for the couple prior days. Physical examination revealed a 38.5 °C fever; light perception and reflex were preserved with no stiff neck. Endoscopic examination showed an inflammatory sphenoethmoidal

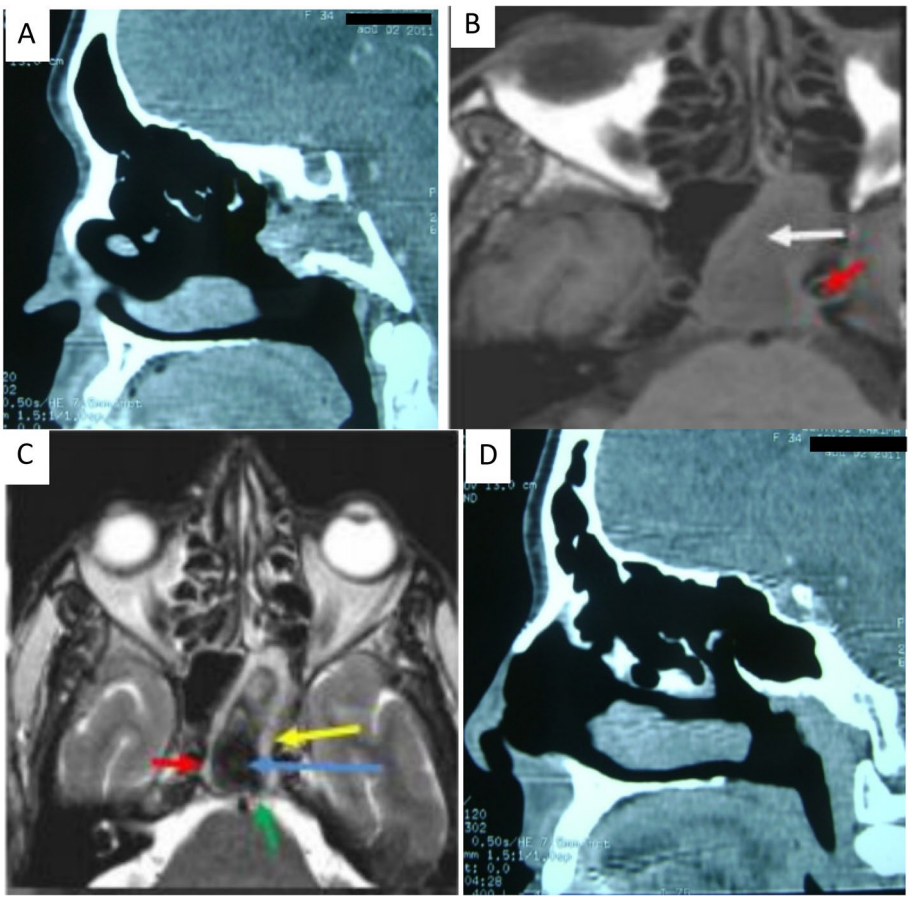

Fig. 5 Case 5. Pseudotumoral invasive sinusitis. CT scan sagittal view (**A** upper left) showing total tissue filling of the sphenoidal sinus with clivus partial destruction. MRI T1-weighted sequence axial view (**B** upper right) showing an intermediate T1 signal total filling of the left sphenoid sinus (white arrow). MRI T2-weighted sequence axial view (**C** lower left) showing a hyposignal total filling of the left sphenoid sinus (blue arrow) that encounters close contact with the internal carotid artery (red arrow), basilar trunk (green arrow), and an inflammatory mucosa (yellow arrow), all for a pseudotumoral invasive sinusitis. Postoperative CT scan sagittal view (**D** lower right) showing a complete removal of the sphenoid sinus pathology

recess with minimal purulent nasal discharge. Ophthalmology examination revealed 0.6 visual acuity in the left eye and 1 in the right eye with no visual field loss. Fundus examination disclosed a stage 1 papilledema according to Frisen scale. At laboratory tests, white cell count is 15,700/mm^3, neutrophils at 85%, normal kidney function, and CRP: 182 mg/l. Cerebral CT scan was normal apart from an uncomplete filling of the left sphenoid sinus (Fig. 6). Lumbar puncture results were normal. Surgical procedure was scheduled since the patient had ophthalmological complications. Saline irrigation, antibiotics based on levofloxacin, and oral corticosteroids were used as preoperative preparation. The patient underwent trans-nasal sphenoidotomy. Pus sample and mucosal biopsy were performed which results concluded an acute nonspecific infectious sinusitis with negative bacterial and fungal culture. The patient uses levofloxacin for 10

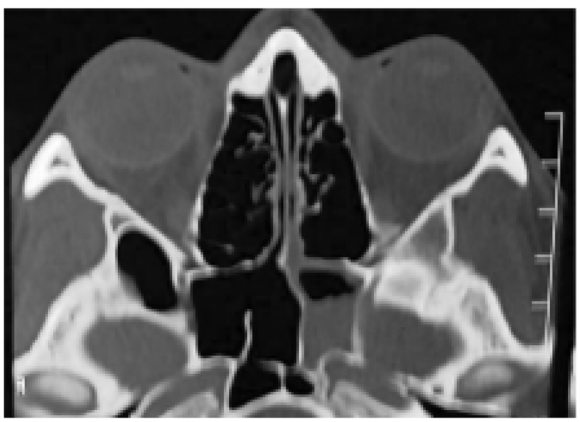

Fig. 6 Case 6. Left sphenoid sinusitis. CT scan axial view showing partial filling of the left sphenoid sinus without bone destruction or bone thickening

days postoperatively and achieved complete recovery with normal visual acuity and eye fundus examination.

Case 7

A 67-year-old female patient with a history of diabetes mellitus type 2 was admitted to the emergency department for severe headache, visual disturbance, and recent generalized weakness for the past 36 h. Physical examination found a confused patient, her breath had a strong odor of acetone, Glasgow Coma Scale at 12 points, fever at 38.6 °C, left eye blindness, and complete ophthalmoplegia. Endoscopic examination disclosed nasal congestion associated with thick purulent discharge in the superior left meatus and sphenoethmoidal recess with no evidence of mucosal necrosis. No neck stiffness or explicit motor impairment was found. Laboratory tests showed a white blood count of 25,000/mm^3, neutrophils of 88%, CRP: 354 mg/l, creatinine: 1.32 mg/dl, urea: 110 mg/dl, glucose 725 mg/dl, K: 2.5 mEq/l, Na: 135 mEq/l, and Cl: 85 mEq/l. The acetone of the urine was positive 4+. Brain CT scan without contrast revealed a heterogenous total filling of the left sphenoid sinus without cerebral oedema or tumors. Cerebrospinal fluid analysis ruled out the diagnosis of meningitis. The patient was admitted to the ICU to restore metabolic equilibrium. Empiric broad-spectrum antibiotics were started based on fluoroquinolones and carbapenems after appropriate microbiological sampling that revealed an *Enterobacter* septicemia. However, despite the recovery of metabolic and kidney impairment, the patient state remained altered. Cerebral MRI with gadolinium (Fig. 7) disclosed complete hyperintense T1 filling of the left sphenoid sinus with homolateral cavernous sinus thrombosis and skull base pachymeningitis. Infectiology team advised to start amphotericin B at 1.5 g/kg and schedule surgical treatment as soon as possible due to the high suspicion of mucormycosis. An endoscopic endonasal surgery was performed which found limited mucosa necrosis of the anterior wall of the sphenoid sinus and a congestive mucosa of the rest of the nasal cavity. We performed a trans-nasal approach for debridement of the necrotic tissue and sinus drainage. We harvested mucosa sample and sinus concretions for microbiological and histological study. Liposomal amphotericin B was continued postoperatively. Histopathology confirmed the diagnosis of invasive mycotic sinusitis with *Rhizopus* species' mucosae infiltration, angioinvasion, and mucosa necrosis. Bacteriological samples were sterile. Control laboratory tests revealed a noticeable improvement, white blood cells at 12,000/mm^3, CRP: 127 mg/l, glucose: 130–170 mg/dl. Kidney and liver functions were normal. However, neurological state of the patients remains unchanged. A week later, the patient developed an acute kidney injury with a creatinine of 3.2 mg/dl, amphotericin B was stopped, and oral posaconazole was administrated instead through feeding tube. However, the patient altered her kidney failure and developed a liver failure with PT at 20%. Subsequently, systemic antimycotic treatment was stopped. Unfortunately, the patient passed away of a refractory hemorrhagic syndrome.

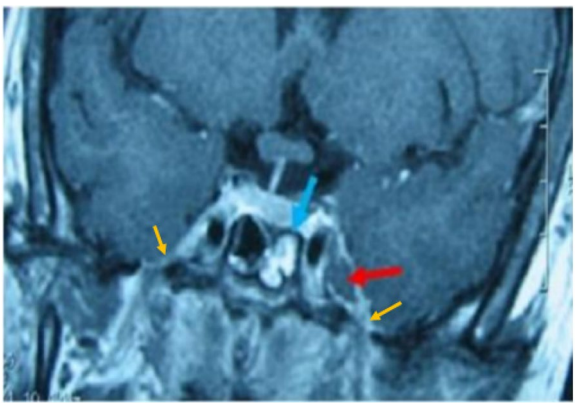

Fig. 7 Case 7. Left acute invasive fungal sinusitis with cavernous sinus thrombosis. MRI T1 gadolinium-weighted sequence coronal view showing a total filling of the left sphenoid sinus (blue arrow) with homolateral cavernous sinus thrombosis (red arrow) and skull base pachymeningitis (yellow arrow)

Table 1 summarizes our series demographics, clinic, and follow-up data.

Discussion

Among primary sphenoid sinus pathology, sinusitis whether bacterial or fungal is the most frequent pathology [6]. Solitary sphenoid sinusitis is uncommon but potentially induces serious complications such as cranial nerve palsy, encephalic abscess, empyema, or meningitis [7]. Headache is the most usual symptom, although no typical pattern is reported [7]. Some atypical presentations of isolated sphenoid sinusitis include unilateral VIth nerve paralysis in children, [8] IIIrd nerve paralysis, [9] bacteremia due to beta-hemolytic group C streptococcus [10], and isolated *Cryptococcus* sphenoiditis associated with skull base osteomyelitis, meningitis, and septicemia [11]. On the other hand, fungi are mainly responsible of chronic rhinosinusitis. Most fungal sinusitis is noninvasive, apart from when they manifest in immunocompromised person [12–14].

Noninvasive fungal sinusitis includes allergic fungal sinusitis and sinus mycetoma or fungal ball. Allergic fungal sinusitis is caused by *Aspergillus fumigatus*, *Bipolaris*, *Curvularia lunata*, or *Drechslera* species [15, 16]. It is suspected in patients with recalcitrant sinusitis and nasal polyposis. Other manifestations might include eye

Table 1 Our series demographic, clinic, and follow-up data recapitulation

Patient	Age	Gender	History	Symptoms	CT scan	MRI	Diagnosis	Procedure	Surgery complications	Follow-up
1	11	M	-	Headaches+++ Trismus+	Figure 1A	Figure 1B	Sphenoid sinus mucocele extended to intra-temporal fossa with fibrous dysplasia	Trans-nasal sphenoidotomy	None	-
2	17	F	GERD	Headaches++ Posterior rhinorrhea+ Cacosmia++	Figure 2A	-	Fungal ball	Trans-ethmoidal sphenoidotomy	None	Figure 2B
3	59	F	High blood pressure	Headaches+++ Nasal obstruction+++ Sleep apnea	Figure 3A	Figure 3B	Chondroma	Transseptal sphenoidotomy	None	Figure 3C & D
4	19	F	-	Headaches+ Nasal obstruction++ Rhinorrhea+	Figure 4A & B	-	Spheno-choanal polype	Trans-nasal sphenoidotomy	None	-
5	37	F	-	Headaches+++ Vomiting Cacosmia ++ Diplopia++	Figure 5A	Figures 5B & C	Pseudotumor fungal sinusitis	Trans-nasal sphenoidotomy	Persistent left medial rectus muscle paresis	Figure 5D
6	41	F	-	Headaches+++ Vomiting Decreased visual acuity++	Figure 6	-	Acute bacterial sinusitis	Trans-nasal sphenoidotomy	None	Complete recovery with normal visual acuity
7	67	F	Type 2 diabetes ketoacidosis	Headaches+++ Left eye blindness Ophthalmoplegia Fever confusion	Total filling of the left sphenoid sinus	Figure 7	Acute invasive fungal sinusitis	Trans-ostial sphenoidotomy	None	Deceased because of refractory hemorrhagic syndrome

GERD gastroesophageal reflux disease. *HP Helicobacter pylori*. +Minimal, ++mild, +++severe (according to a visual analogic scale)

muscle entrapment or proptosis. Sinus CT scan reveals opacification with concretions and/or calcifications [16, 17]. According to Lu-Myers et al. [18], patients with allergic fungal rhinosinusitis have higher quantitative serum immunoglobulin E levels and higher Lund-Mackay scale scores compared to patients with chronic rhinosinusitis [18]. *Aspergillus fumigatus* and *Dematiaceous* fungi most commonly cause fungal ball. Clinical manifestations of sinus mycetoma are similar to that of chronic sinusitis. Usually, sinus mycetoma is found accidentally on sinus CT scans [19].

Invasive fungal sinusitis includes the acute fulminant fungal sinusitis, the chronic fungal sinusitis, and granulomatous types. Fungi of Mucorales order, although saprophytic organisms, may cause acute invasive fungal sinusitis in immunocompromised patients [19]. Blitzer et al. [20] found the prevalence of diabetes to be 70%; only 4% had no identifiable risk factors. The other 26% (46/179) patients had immunocompromising conditions such as leukemia, kidney disease, infant diarrhea, immunosuppression medications after transplant, and pancreatitis [20]. Necrosis spots on the septum, turbinates, or palate are pathognomonic. Cavernous sinus thrombosis is typical of advanced disease stages.

Chronic invasive fungal sinusitis is caused only by *Aspergillus fumigatus*. This entity presents as chronic rhinosinusitis that might exhibit ophthalmological symptoms such as orbital apex syndrome [21].

Granulomatous invasive fungal sinusitis is described almost exclusively in immunocompetent African patients and caused by *Aspergillus flavus*. Commonly, patients present symptoms of chronic rhinosinusitis associated with proptosis. Nasal cavity examination can be inconclusive. Nevertheless, eye examination discloses remarkable findings. Surgery is the best management modality of fungal sinusitis. Nonetheless, systemic antifungal medications are essential in invasive forms [22].

Sphenoid sinus tumors, though rare, are very diverse. The most frequent benign tumor is osteoma which arises mostly from the sinus septa. They are commonly asymptomatic. On CT scan osteoma has a limited shape. Usually very dense, it seldom has a heterogeneous aspect [23].

The spheno-choanal polyp represents 3 to 6% of all nasal polyps [24, 25]. It occurs mainly in teenagers and young adults. The youngest case is a 4-year-old girl reported by Lim and Sdralis [26]. Most of these polyps are small, asymptomatic, and usually regress spontaneously. In some cases, the polyp can grow after a persistent infection and become clinically symptomatic [24].

Sphenoid chondroma is an exceptional benign cartilaginous tumor that develops from cartilaginous remnants of the skull base junctions, especially the ethmoido-sphenoidal junction [23]. Also, it can rise from the petrous apex [27]. Multiple locations occur in enchondromatosis [28]. Clinical features are delayed and non-specific, usually related to the involvement of adjacent structures. CT scan and MRI state the tumor extension but could not set up the diagnosis of chondroma. Chondroma and chondrosarcoma are quite difficult to distinguish in histopathological study. Complete surgical resection is the main treatment [23].

Seventeen percent of bony locations of fibrous dysplasia are craniofacial [29]. It affects the sphenoid in 19% of cases [23]. Pathology samples exhibit features of inflammation and hemorrhagic or cystic remodeling [23]. CT scan presentation is nonspecific. Fibrous dysplasia appears as low-to-intermediate signal lesion on MRI T1-weighted sequences and low signal intensity on T2-weighted images [30]. There are only two reported cases of sphenoid mucocele associated with fibrous dysplasia [31, 32]. Several pathophysiology hypotheses have been brought up such as mechanical obstruction of the sinus drainage pathways, inflammatory obstruction, or infectious triggers. The mechanical theory seems the most consistent in the case of sphenoid mucocele associated with fibrous dysplasia as the latter might induce sinus ostial obstruction [32].

Conclusion

Benign solitary sphenoid sinus pathology might not always be harmless. Some etiologies, especially infections, are very dangerous either because of the close contact with vital structures or because of their invasive character. Endoscopic sphenoid sinus surgery, in the hands of an experienced surgeon, offers a safe and efficient approach to sphenoid sinus benign pathology.

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Authors' contributions
NO was involved in diagnosis, literature review, and manuscript drafting; HL was involved in literature review and drafting of the manuscript; ZCH was involved in collecting data and reviewed the manuscript; HC was involved in literature review and study design and reviewed the manuscript; ZZ was involved in diagnosis, management of the cases, and study design; and MNA reviewed the manuscript for insightful remarks. The authors read and approved the final manuscript.

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Availability of data and materials
The datasets generated and/or analyzed during the current study are not publicly available due to patients' data confidentiality but are available from the corresponding author on reasonable request

Declarations

Ethics approval and consent to participate
The IRB approved our study, and informed written consent to participate to the study was signed by the alive participants (or their legal guardian for the

patient under 16) and by the deceased patient's relatives. Our IRB is CEHUF (comité d'ethique hospital-universitaire de Fès), and email is Comite.ethique.fes@usmba.ac.ma.

Consent for publication
An informed consent for publication purpose was obtain from the alive patients (or their legal guardian for the patient under 16) and from the deceased patient's relatives. Written consent is available.

Competing interests
The authors declare that they have no competing interests.

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