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An unusual case of complicated rhinosinusitis of the sphenoid sinus involving the cavernous sinus and skull base: Endoscopic sinus surgery and medical therapy

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Abstract

Isolated sphenoid sinus diseases are generally asymptomatic and relatively uncommon with the potential for serious complications. Patients with this condition should be monitored closely and treated aggressively and either diagnostic or therapeutic intent is often indicated. In the management of a complex, life-threatening condition that involves many different fields of expertise, the otolaryngologist plays a key role in orchestrating different specialists and gaining direct access to the affected area, thus taking the first and essential step towards diagnosis and therapy. Because of the superiority of computed tomography in defining the bony margins and the superior soft tissue resolution of magnetic resonance imaging, these two techniques should be used in a complementary manner in the evaluation of isolated sphenoid sinus disease in addition to mapping the lesion better and identifying intracranial and intraorbital extent. We report an unusual case of isolated rhinosinusitis of the sphenoid sinus involving the cavernous sinus, pterygoid fossae and masticatory space in an immunocompetent patient.

Keywords

Sphenoid sinusitis, isolated sphenoid sinus disease, fungal rhinosinusitis, cavernous sinus thrombosis

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Introduction

Sphenoid sinus disease usually results from a posterior ethmoidal spread.[1] Occasionally though, the sphenoid can be the only affected sinus (1%–2.7% of all diagnosed paranasal sinus disease).[2] Symptoms of isolated sphenoid sinus pathology are often subtle and non-specific: headache is the most common symptom (71.4%), followed by sinonasal disturbances (such as nasal congestion, rhinorrhea, hyposmia, and epistaxis). Cranial nerve neuropathy is also relatively common, occurring in 16.5% of cases and usually affecting the I and III cranial nerves.[3]

The most common cause of isolated sphenoid sinus opacification is infections: 28.3% of all cases are due to chronic rhinosinusitis without nasal polyps, 20.3% to mucoceles, 12.5% to fungal sinusitis, 7.7% to malignant neoplasms, 7% to intracranial lesions, 5.7% to benign neoplasms, 3.4% to chronic rhinosinusitis with nasal polyps and 4.7% to other lesions.[3]

Imaging plays a crucial role: the combined use of computed tomography (CT) and magnetic resonance imaging (MRI) scans is considered the gold standard to formulate a diagnostic hypothesis and to identify possible complications, such as intracranial and intraorbital extension,[4] and to exclude cavernous sinus extension.[5]

In fact, the sphenoid sinus occupies an anatomically critical position: its proximity to the cavernous sinus, the carotid artery and the III, IV, V1V2 and VI cranial nerves makes correctly diagnosing and treating sphenoidal pathology all

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the more crucial. At the same time, its limited accessibility makes obtaining a specimen challenging.

We present an unusual case of isolated rhinosinusitis of the sphenoid sinus, involving the cavernous sinus, pterygoid fossae and masticatory space in an immunocompetent patient.

Case report

Our patient was an 83-year-old man. In October 2016, he started to experience right mandibular pain radiating to the zygomatic and mastoid regions. The pain worsened progressively and required a combination of oxycodone, paracetamol and ketorolac to manage it. Since a trigeminal neuralgia was suspected, an empirical course of prednisone (50 mg per day) was started, but with no benefit.

Contrast-enhanced maxillofacial MRI and CT scans were performed (Figures 1 and (2). ENT evaluation of the patient by endoscopy of the upper airways showed a purulent discharge in the sphenoethmoidal recess. With the suspicion of sphenoid sinus disease with radiological complications and worsening right facial pain, the patient underwent right-sided endoscopic sinus surgery (ESS). After middle turbinectomy, a transnasal paraseptal sphenoidectomy was performed. The natural ostium of the sphenoid sinus was obstructed by a pyocele which was marsupialized and the content drained. This was sampled and, along with samples of the sphenoid mucosa, was sent for bacteriological and histopathological examination. The surgeon opened the natural ostium of the maxillary sinus and then the postero-anterior ethmoid sinuses was performed. The breach in the posterior-lateral sphenoid wall was repaired with medium turbinate mucoperiosteum. After careful haemostasis, a Merocel sponge was positioned in the right nasal fossa (Figure 3).

There were no postoperative complications and the patient was discharged after 2 days. Histopathological examination was negative for both hyphae and neoplastic cells. Cultures were also negative, both for bacteria and fungi. After an initial improvement, the right trigeminal neuralgia gradually returned. A follow-up MRI, performed 1 month after surgery, confirmed persisting inflammation of the sphenoid sinus, skull base and pterygoid fossa.

A consultation with an infectious diseases specialist was sought. He ordered a serological work-up, which was negative for galactomannan and positive for beta-D-glucan. Then empirical antifungal therapy with voriconazole was commenced. The initial dose was 800 mg on the first day, followed by a dosage of 200 mg twice a day. Voriconazole serum levels were then regularly monitored along with liver enzymes and kidney function. After approximately 1 month of antifungal therapy, a spike in liver cytolysis markers was detected. The possibility of voriconazole-induced acute hepatitis was investigated with a liver ultrasound and liver MRI, which were both negative. Hepatic enzymes also gradually

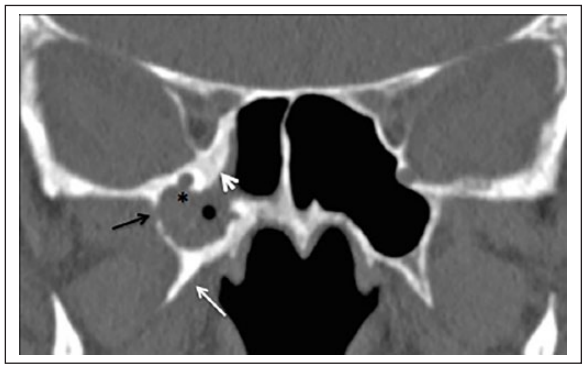

Figure 1. Preoperative coronal CT scan, showing an enlargement of the right sphenoid sinus with overdevelopment and opacity of the lateral recess. Thickening and sclerosis of the bone surrounding the right sphenoid sinus, the pterygoid process (white arrow) and the orbital apex (white arrowhead) can be observed. Focal interruption of the foramen rotundum (CNV2) (small star) and of the sphenoidal lateral recess wall is also present (black arrow).

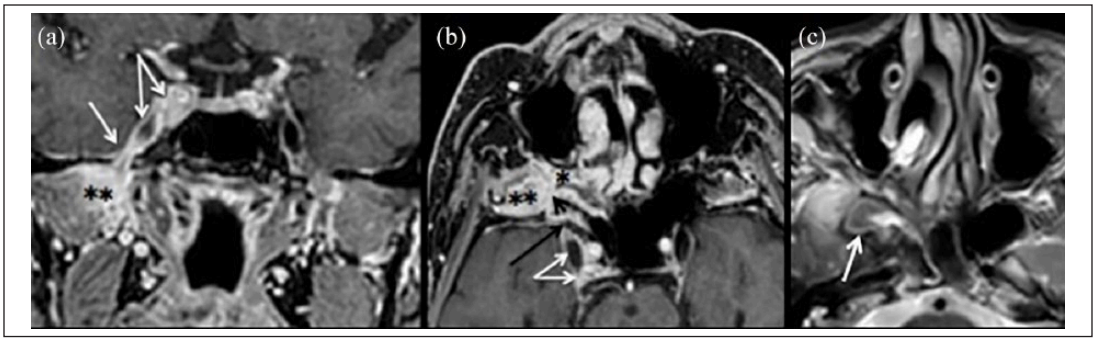

Figure 2. Preoperative (a) coronal and (b) axial gadolinium-enhanced T1-weighted MRI shows soft tissue protruding through the breach in the lateral recess wall of the sphenoid sinus (black arrowhead), invading the right internal pterygoid muscles, the masticatory fossa (double small star) and the pterygopalatine fossa (single small star) and infiltrating the II (CNV2) (black arrow) and III (CNV3) (white arrow) trigeminal branches. Endocrianially, the tissue extended into the cavernous sinus surrounding the Gasser ganglion (double angled arrow). The cavernous carotid artery appears unobstructed. The MRI show marked hypointense signal on (c) axial T2-weighted into the lateral recess of the sphenoid sinus with inflamed mucosa at periphery (white arrow).

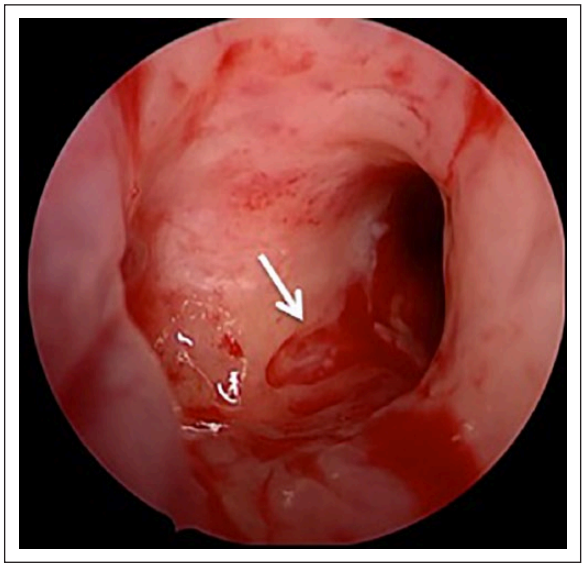

Figure 3. Intraoperative view during endoscopic sinus surgery shows an erosion of lateral recess wall of the right sphenoid sinus (white arrow).

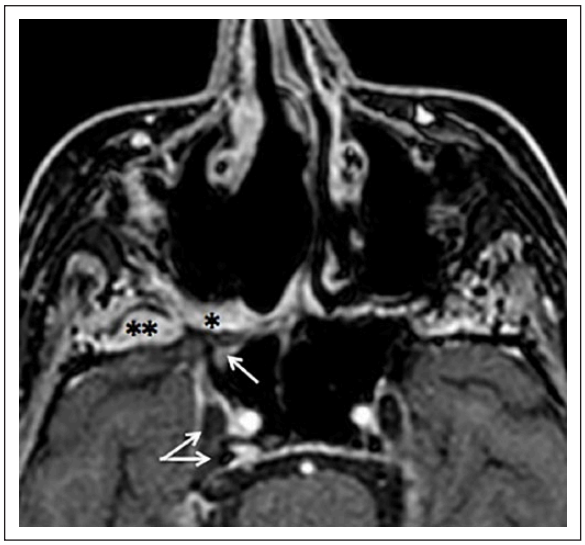

Figure 4. Axial gadolinium-enhanced T1-weighted MRI after 12 weeks of therapy shows good pneumatization of the right surgical cavity and a sharp decline in contrast enhancement of the lateral recess of right sphenoid sinus (white arrow), masticatory fossa (double small star) and pterygopalatine fossa (single small star). A similar decrease can be appreciated in phlogistic alterations of the Gasser ganglion and the cavernous sinus (double angled arrow).

decreased. Thus, a decision was made to continue the antifungal therapy.

Follow-up maxillofacial MRI, performed 1 month into the antimycotic chemotherapy, showed improvements, with shrinkage of both the sinus and the intracranial component of the phlogistic tissue. This was matched by an improvement in clinical condition: facial pains gradually decreased and finally disappeared.

A follow-up MRI performed after 12 weeks of therapy showed that the infection had almost completely resolved. Thus, antifungal therapy was discontinued and at the present time, 11 months after surgery, the patient is still symptom-free (Figure 4).

Discussion

Although isolated involvement of the sphenoid sinus is uncommon (1%–2.7% of all sinus infections), fungal infections play an important role: the incidence of sphenoidal fungal rhinosinusitis (FRS) ranges from 4.5% to 26.8%, according to different studies.[2]

The immunosuppressed population has a greater risk for FRS but it has to be noted how chronic FRS, as opposed to acute FRS, frequently occurs in immunocompetent or mildly immunosuppressed patients such as patients with poorly controlled diabetes mellitus, patients undergoing corticosteroid therapies and elderly patients in general.[6–9]

As in most cases with isolated sphenoid sinus involvement, presentation in our patient was subtle: he experienced no nasal obstruction or discharge. Worsening right facial pain was the foremost symptom, starting approximately 6 months before diagnosis. This is not uncommon in isolated sphenoidal FRS which may present with little or no sinonasal symptoms but with a variety of neurological or maxillary-facial signs: III, IV, V and/or VI cranial nerve impairment, focal neurological deficits, headache and facial swelling.[7,9,10]

In fungal rhinosinusitis, CT scans typically show a thickening of the affected sinus mucosa, obliteration of the affected sinus with metal-like signals within, and, especially in invasive forms, remodelling of sinus walls (sclerosis, thickening, expansion or irregular erosion). These features are not exclusive to invasive FRS (IFRS) and require a differential diagnosis with neoplasms. MRI shows iso- or hypodense signals in T1 weighted sequences, and markedly hypointense in T2w sequences up to the presence of a central 'signal void'.[4,7–11] Because our patient's imaging showed some of these typical features and his medical history was congruent, chronic fungal rhinosinusitis, possibly invasive, was our first diagnostic hypothesis.

After ophthalmologic and neurological consultation, the patient underwent ESS. Although the literature often recommends that surgical debridement in IFRS be as radical as possible, removing all apparently infected or necrotic areas until healthy tissue is reached and draining the thrombosed cavernous sinus,[7,9] the extent of debridement remains a debated topic with some authors advocating a patient-tailored approach.[12] We took into consideration the patient age and preferences, and decided to proceed with a debridement of the involved sinuses, without opening of the cavernous sinus. It has to be underscored, though, how in this field,

decisions are still made on a case-by-case basis: due to the relatively uncommon nature of the disease, there is a lack of large, standardized studies and most of the evidence is based on small series or case reports like this one.

Despite a good surgical outcome, our patient did not show a rapid radiological and clinical resolution of the disease. Also, histopathological examination found no hyphae within the sinus wall, but it was also negative for neoplastic cells. Unsurprisingly, due to the low sensitivity level, approximately 37.5% according to Pagella et al.[7] cultures were also negative.

Following advice from an infectious disease specialist, serologic testing was performed: galactomannan was negative while beta-D-glucan was positive. This suggested an active invasive mycotic infection and a course of empirical therapy was started, voriconazole being the medication of choice.

In the case of our patient, the decrease in beta-D-glucan levels and the clinical improvement suggested that a form of invasive fungal sinusitis had been responsible.

Conclusion

Isolated rhinosinusitis of the sphenoid sinus can be a subtle condition, presenting with little or no sinonasal symptoms. Differential diagnosis among different infectious forms and with neoplasms is crucial, and CT and MRI features can be of great help. Surgery has to be considered the option of choice: it has a curative, recurrence-preventive and diagnostic value, allowing the harvesting of specimens for both histopathological and microbiological examination. Management of invasive fungal forms must be started rapidly with a combination of long-term systemic antifungal therapy, endoscopic approaches with debridement of the affected sinus in order to block the progressive evolution and intracranial and intraorbital dissemination of the disease, and if possible, elimination of the predisposing conditions. The ENT specialist is right at the heart of this multidisciplinary approach, and should coordinate with other specialists (radiologist, neurologist/neurosurgeon, infectious disease specialist, microbiologist and histopathologist) in order to achieve diagnostic and therapeutic success.

Management of an invasive fungal form requires a combination of antifungal therapy, under the supervision of an infectious disease specialist, debridement of involved tissues and, if possible, elimination of predisposing conditions.

Declaration of conflicting interests

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Ethics approval

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Informed consent

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