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Clinical Record

Isolated inflammatory sphenoiditis with multiple unilateral cranial nerve palsies

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Abstract

Isolated sphenoidits is a rare entity that often presents with vague, non-specific symptoms. We present the case of a 36-year-old Middle Eastern man, who developed headache and a painful right eye. A diagnosis of acute sphenoiditis was made. Shortly afterwards, he developed diplopia due to isolated abducent nerve involvement. Within two months, the extent of cranial nerve involvement had increased to include cranial nerves II, III, and V. Subsequently, this was treated by functional endoscopic sinus surgical drainage and biopsy. Histology revealed inflammatory changes. The patient made a dramatic recovery post-operatively, with resolution in all symptoms.

Key words: Sphenoid Sinusitis; Cranial Nerve Diseases; Otorhinolaryngologic Surgical Procedures; Endoscopy

Introduction

Isolated sphenoiditis is a rare and potentially very serious condition that commonly presents with several non-specific symptoms. Pathology of the sphenoid sinus typically presents with headache and visual compromise, and these symptoms expand rather than focus the differential diagnosis, making early presentation unlikely.[1] The difficulty in visualising the sphenoid sinus directly requires that the clinician maintains a high index of suspicion when presented with the often vague features of sphenoiditis.

In this article, we present an interesting case of a man who developed sphenoid sinusitis, complicated by multiple cranial nerve palsies who underwent endoscopic computed tomography (CT) guided navigational surgery.

Case report

A 36-year-old Middle Eastern helicopter pilot developed an acute unilateral right sided retro-orbital pain with an associated pain in the vertex of the skull. A few hours after the onset of his pain, he noticed diplopia, for which prompt medical attention was sought in his home country. Examination revealed that he had VIth nerve palsy. He was admitted to a local hospital where ophthalmic tests and magnetic resonance imaging (MRI) of the head were conducted. A diagnosis of right sphenoid sinusitis was made, and he received intravenous antibiotic treatment for the next 23 days. Subsequently, his headache improved, but no change was noted in his diplopia. After the antibiotic treatment, he had surgery, the exact nature of which is unknown, but it sounded like endoscopic sinus surgery together with biopsy of the adenoid.

The patient had sought further treatment in the United Kingdom two months after the onset of his symptoms. Examination revealed that he now had a IIIrd nerve palsy and complete ptosis of the right eye in addition to the VIth nerve palsy. His acuity in the left eye was 6/4 and in the right eye was 6/6, indicating optic nerve involvement. He had decreased sensation to light touch and pin prick, and tingling was noted on the right side of his forehead and temple, indicating trigeminal (ophthalmic division) involvement.

A brain MRI and CT of the sinuses was carried out. The CT sinuses (Figure 1a and b and Figure 2) showed chronic mucosal thickening of the right sphenoid, loss of bone posterolaterally to the right within the sphenoid sinus, superiorly and posteriorly, resulting in there being no obvious bone between the right sphenoid sinus and the pre-pontine cistern posteriorly, and on the lateral wall of the right sphenoid sinus between the mucosa and the right internal carotid artery. The MRI scan (Figure 3a and b) revealed smooth abnormal enhancement of the right cavernous sinus with involvement down the superior wall of the clivus on the right.

He underwent endoscopic navigational surgery (Brainlab system) where a biopsy was taken from the sphenoid sinus, and the sinuses were drained. Intra-operatively, the surgical findings revealed a very thick right anterior sphenoid wall that proved to be impenetrable. Thus, the left sphenoid was opened followed by reduction of the intersinus wall, giving access to the right sphenoid. A biopsy was taken from the right sphenoid, which confirmed inflammation with no evidence of malignancy. Microbiology grew *Candida albicans*.

The patient made a dramatic recovery after the operation and with intravenous antifungal treatment. Two weeks after surgery, his eye symptoms had virtually disappeared, with normal eye movements, minimal diplopia, and normal pupils. All symptoms continued to improve.

Discussion

The sphenoid sinus is a development of out-growth of the sphenoid bone in the posteriosuperior segment of the

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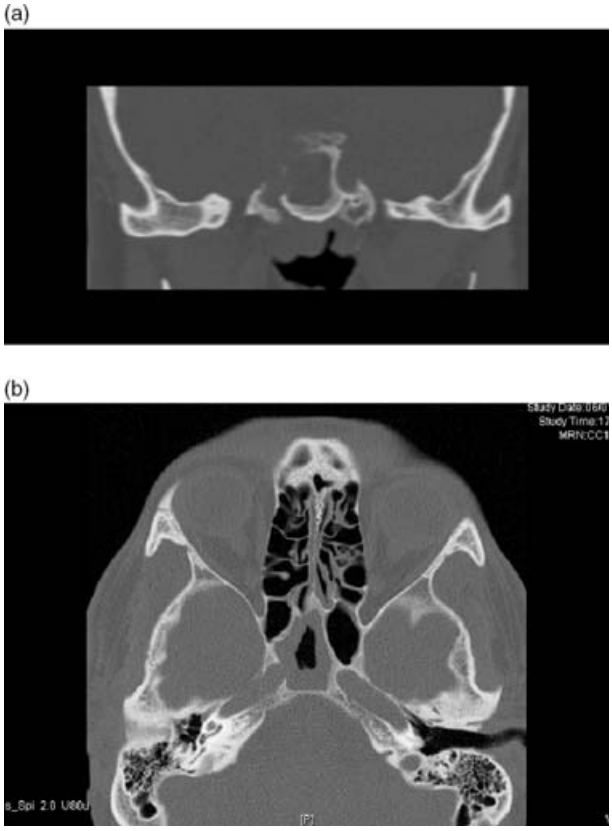

FIG. 1

Coronal (a) and axial (b) CT sinus image showing opacity in the right sphenoid sinus with loss of bone posterolaterally and no obvious bone between the right sphenoid sinus and the pre-pontine cistern posteriorly, and on the lateral wall of the right sphenoid sinus between the mucosa and the right internal carotid artery.

sphenoid recess. Pneumatisation of the sinus occurs in the sixth and seventh year of life and reaches its final size and shape between nine and 12 years of age.[1] Thirteen important structures are adjacent to the sphenoid sinus and include the dura, pituitary, cavernous sinus, pterygoid canal and nerve, internal carotid artery, and cranial nerves II, III, IV, VI, V_1 and V_2. [2–4] All of these structures are vulnerable to injury with sphenoid disease, and all the cranial nerves listed above, except for the cranial nerve V_2, were affected in our patient.

The signs and symptoms of sphenoid sinusitis are often vague and non-specific.[1–9] In our experience, headache is the most common presentation and therefore, such patients are often referred to the neurologist. Our patient presented with headache at the vertex of the skull and right retro-orbital pain, which is fairly typical of sphenoid disease. It is, however, the extent of his cranial nerve palsies that is of greatest interest. All the cranial nerves in the vicinity of the right sphenoid were affected.

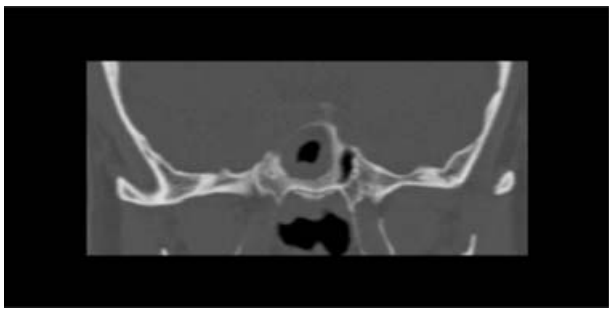

FIG. 2

Coronal CT showing circumferential mucosal thickening showing loss of bone laterally.

Visual disturbances ranging from, blindness and decrease of visual acuity to diplopia are the second most common cranial nerve complaint to be encountered. Lawson *et al.*, in their analysis of 132 cases, reported that the incidence of visual disturbance is 12 per cent for inflammatory lesions.[5] The optic nerve appears to be more often involved than any other cranial nerve.[1] The reason for the preponderance of optic nerve involvement is the local anatomy in the region.[3] Cadaver studies investigating the neurovascular relationship of the sphenoid sinus have shown that in 79 per cent of cadavers, the bony wall between the sphenoid sinus and the optic nerve is less than 5 mm thick, and is completely absent in 8 per cent.[7] It is also for this reason that the optic nerve is often the first nerve to be affected.[5] Visual loss in association with sphenoid sinusitis is a rhinology emergency. Our patient demonstrated a decrease of visual acuity in the right eye in keeping with right optic nerve involvement.

Diplopia was found to be the earliest signs of a diseased sphenoid, as was the case in our patient. However, he subsequently developed right optic neuritis secondary to osteitis. Lawson *et al.* found that in their series, the VIth nerve was involved in 6 per cent of the inflammatory cases. Oculomotor nerve palsy causing ptosis was seen in 7.5 per cent of all patients, but all were due to a neoplasm or in one case a mucocele, and not inflammation as in our patient. Trigeminal nerve deficit was seen in 4.5 per cent of all patients producing a retrobulbar and mid-facial pain and numbness.[5] It can be seen that cranial nerve lesions are uncommon in inflammatory sphenoid sinusitis, and in combination, to the extent seen in this case.

The treatment of isolated sphenoid sinusitis focuses mainly on appropriate antibiotic treatment, topical nasal decongestants and endoscopic sinus surgery. Various surgical approaches to the sphenoid sinus have been described in the past. The most popular approach includes the sublabial transseptal, transethmoidal and transpalatal. The disadvantages of all these are septal perforation, post-operative upper incisor numbness, scarring and prolonged hospital stay.[10] Today, the recommended treatment is via the endoscopic route: washout via the natural ostium, anterior sphenoid wall antrostomy and washout, intranasal, transseptal, transethmoidal and transantral sphenoidostomy.[3] Kieff *et al.* found that endoscopic sphenoidectomy without ethmoidectomy is sufficient in treating isolated sphenoiditis together with the removal of the inferior portion of the middle turbinate at the time of surgery and would enable direct access to the sphenoid sinus during surgery and post-operative inspection and care.[11] Brockbank *et al.* found that three out of seven of their ENT patients with sphenoiditis required repeated antral punctures before they were asymptomatic.[4]

Staphylococcus aureus is the common isolate in acute sphenoiditis and anaerobes are the most common isolates in chronic infection.[8] This is similar to the findings in maxillary sinusitis.[8] Partial blockage of the natural ostia results in poor drainage and increased intranasal pressure and the poor ciliary action observed in chronic disease reduces the oxygen content and pH of the sinus and supports the growth of anaerobic organisms.[8] Empiric broad-spectrum antimicrobial coverage is advised even in cases where negative cultures are obtained.

Our patient displayed a chronic picture of incomplete resolution of sphenoiditis due to incomplete opening of

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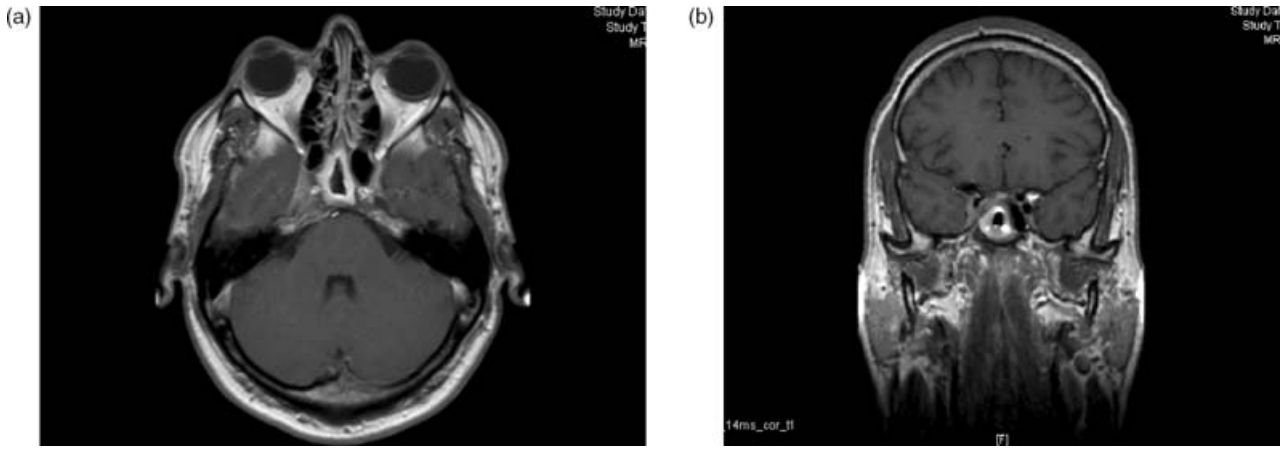

FIG. 3
MRI T1-weighted axial (a) and sagittal (b) images showing smooth circumferential enhancement of right cavernous sinus involving the superior wall of the clivus on the right.

the sphenoid sinus. He was treated initially with what appeared to be limited sinus drainage and antibiotics. On the second sinus surgery, no pus was found but *Candida albicans* was grown from the inflamed sinus mucosa.

In summary, we present the case of a 36-year-old man, who developed the symptoms of sphenoid sinus disease. He developed an isolated VIth nerve palsy, progressing to include II, III, IV, V, and VI as a result of cavernous sinus thrombosis. He underwent endoscopic sinus surgery, where the sphenoids were drained and found to be highly inflamed. Virtually all symptoms improved thereafter. Minimal diplopia was reported on upward and left sided gaze.

- **This paper describes the case of a 36-year-old man, who developed the symptoms of sphenoid sinus disease, complicated by multiple cranial nerve palsies (II, III, IV, V and VI) as a result of cavernous sinus thrombosis**
- **A CT scan of the sinuses is the gold standard investigation and MRI is an essential adjunct. Bone erosion requires MRI to determine the presence and extent of skull base and intracranial involvement**
- **The general consensus in management of isolated sphenoiditis is medical treatment in uncomplicated cases for 24–48 hours and surgical drainage for complicated cases or where medical treatment fails**

The sphenoid sinus is often regarded as the 'neglected sinus' due to the difficulty in visualising the ostium. Delay in making the diagnosis and initiating treatment of sphenoid sinusitis could result in a morbidity of 80–100 per cent with 23–29 per cent left with permanent disabilities.[4,12] ACT scan of the sinuses is the gold standard investigation and MRI is an essential adjunct. Bone erosion requires MRI to determine the presence and extent of skull base and intracranial involvement. In this particular case identification of the sphenoid sinus was aided using the navigational system. The general consensus in treatment of isolated sphenoiditis is medical treatment in uncomplicated cases for 24–48 hours and surgical drainage for complicated cases or where medical treatment fails.[1–12]

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Miss S M Keh takes responsibility for the integrity of the content of the paper.
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