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Case Report
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Isolated Sphenoid Sinus Infection with Polyneuritis Cranialis Masquerading as Migraine!

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Abstract

Background: Headache is one the commonest presentation in primary care settings especially preceding with any viral illness often poses a challenge to the treating physician perhaps a validated screening tool highlighting red flags increase the chance of predicting underlying etiology aids prompt diagnosis and management.

Case Report: A 40-year-old Emirati mother of four with no previous personal or family medical history of migraines, presented to local General Practitioner (GP) with a 4-month history of on and off headaches followed by mild flu like illness apart from mild runny nose no other obvious Sino nasal or cranial autonomic symptoms. A month after onset of the headaches, the patient began to notice a slight change in intensity with a worsening pattern, especially on exertion and bending. On examination, the patient was conscious and fully oriented. Cranial nerve examination showed normal visual acuities, right papilledema, reduced right afferent corneal reflex, with normal extraocular eye movements. no other signs of meningeal irritation were present, MRI of the brain and orbits showed mucosal thickening and enlargement of both right and left sphenoid sinus, right worse than the left with extension to the cavernous sinus. Trans nasal endoscopy showed a bilateral congested sphenoid sinus with purulent discharge, which was drained off and treated with IV Ceftriaxone and dexamethasone and the patient had dramatic clinical improvement.

Conclusion: Any headache associated with peri-orbital pain or retro-orbital pain associated with exertional element, requires thorough neurological examination, and warrants urgent appropriate investigations and specialist attention.

Keywords: Migraine with aura; Cavernous Sinus thrombosis; Sphenoid sinus; Retro-orbital pain; Afferent corneal reflex; Polyneuritis cranialis

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Background

Isolated sphenoid sinusitis is a rare condition associated with severe complications, including intracranial abscess, cavernous sinus thrombosis, and cranial neuropathies. Polyneuritis cranialis, or progressive inflammation of the cranial nerves, usually involves one or more cranial nerves IV, V, VI, and VII.

Case Presentation

This report describes a 40-year-old Emirati mother of four with no previous personal or family medical history of migraines, presented to local General Practitioner (GP) with a 4-month history of on and off headaches, according to the International Classification of Headaches Disorders (ICHD-3) migraine without aura. The headaches began to occur after she recovered from a mild flu like illness that subsided after a week, but the headaches remained at the same intensity with intermittent fluctuations apart from mild runny nose no other obvious Sino nasal or cranial autonomic symptoms and were not relieved by oral painkillers (Paracetamol 500 mg thrice a day × 7 days, antihistamine (Cetirizine 5 mg once at night) × 7 days, Domperidone 10 mg thrice a day × 7 days) prescribed by her General Practitioner (GP).

A month after onset of the headaches, the patient began to notice a slight change in intensity with a worsening pattern, especially on exertion and bending. During the second visit to the General Practitioner (GP) the patient was diagnosed to have relapse of migraine with aura and reassured

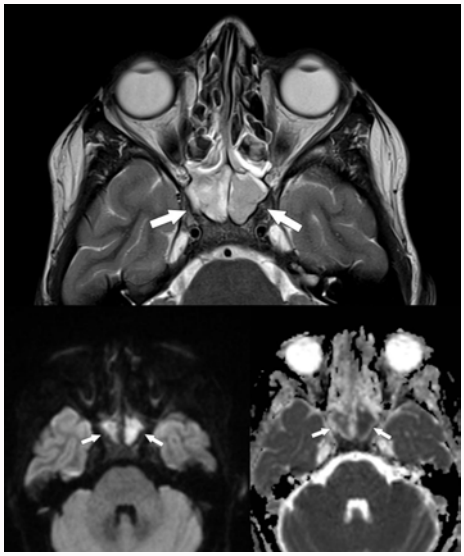

Figure 1a: Axial MRI T2-weighted image demonstrates complete occupation of the sphenoid sinuses bilaterally by material with intermediate T2 signal (arrows). DWI sequence at the same level shows significantly restricted diffusion (high signal in the b1000 images and low values in the ADC map). The findings are highly suggestive of purulent acute sinusitis.

Figure 1b: Contrast-enhanced MRI T1-weighted image demonstrates a filling defect in the right cavernous sinus (arrowheads), suggestive of the possible presence of thrombus.

Figure 1c: Posttreatment axial MRI T2-weighted image demonstrates normal pneumatization of the sphenoid sinuses.

Figure 1d: Posttreatment contrast-enhanced MRI T1-weighted images at the level of the right cavernous sinus do not demonstrate evidence of a filling defect (curved).

following preliminary blood tests (no inflammatory markers were performed) and a plain Computed Tomogram (CT) brain which was done at local Highstreet diagnostic center reported normal. As patient not keen to continue old pain medications or take any new pain medications was then referred for acupuncture.

The patient's symptoms continued to worsen in spite of acupuncture. The headaches became right hemi-cranialis nature with ipsilateral peri-orbital and retro-orbital pain and a worsening exertional pattern, then she was referred to King's College Hospital Dubai by a family friend.

On examination, the patient was conscious and fully oriented. Cranial nerve examination showed normal visual acuities, right papilledema (involving Optic nerve), reduced right afferent corneal reflex, (involving ophthalmic branch of Trigeminal nerve) with normal extraocular eye movements. Albeit patient complaining of pain with eye movements. Clinically no signs of meningeal irritation were present, rest of her neurological examination was normal. An infective cause was suspected cause of preceding flu like illness superior orbital fissure syndrome, orbital apex syndrome or cavernous sinus venous thrombosis were our top contenders for differential diagnosis. Urgent Magnetic Resonance Imaging (MRI) of brain and orbits with contrast and Computed Tomogram (CT) venogram, Computed Tomogram (CT) paranasal sinus views were organized.

Investigations

Apart from raise in inflammatory markers C-Reactive Protein (CRP 35) and Erythrocyte Sedimentation Rate (ESR 20), all other hematinics were normal, also her infective screen including Human

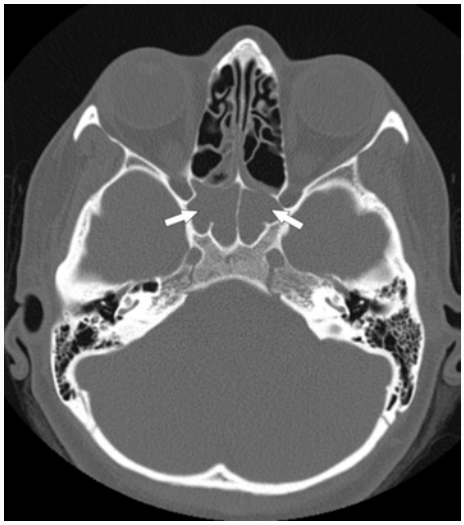

Figure 2a: Axial non-contrast CT performed at the level of the sphenoid sinuses demonstrates complete opacification of the sphenoid sinuses bilaterally.

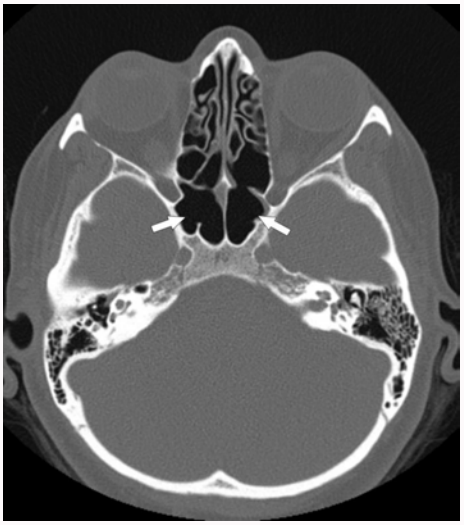

Figure 2b: Posttreatment non-contrast CT at the level of the sphenoid sinuses demonstrates normal pneumatization.

Immunodeficiency Virus (HIV) test was negative. Magnetic Resonance Imaging (MRI) of the brain and orbits showed mucosal thickening and enlargement of both right and left sphenoid sinus, right worse than the left (Figure 1a) with extension to the cavernous sinus (Figure 1b). Computed Tomogram (CT) showed complete opacification of the paranasal sinuses (Figure 2a).

Treatment

Computed Tomogram (CT) and trans nasal endoscopy showed a bilateral congested sphenoid sinus with purulent discharge, which was drained off and sent for culture. The final diagnosis of bacterial sphenoid sinusitis with orbital apex and cavernous sinus involvement was established and treated with IV Ceftriaxone and IV dexamethasone for 5 days with an oral weaning regimen. Surprisingly, no growth was cultured.

Outcome and Follow-Up

Five days after the intervention and completion of the initial course of antibiotics and dexamethasone, the patient had dramatic improvement in her headaches, including total disappearance of exertion, pressure symptoms and retro-orbital and peri-orbital pain, as well as normalization of the right optic disc, which was confirmed on 'Optical Coherence Tomography' (OCT). Follow-up scans (2 weeks) after intervention, Magnetic Resonance Imaging (MRI) brain and orbits demonstrates normal pneumatization of the sphenoid sinuses (Figure 1c) and no evidence of filling defect in cavernous sinus (Figure 1d) showed complete resolution of sphenoid infection on Computed Tomogram (CT) Paranasal sinuses (Figure 2b).

Discussion

The sphenoid sinus develops after age two and starts to pneumatize at age eight. The sphenoidal sinus drains into the sphenoethmoidal recess and is contained within the body of the sphenoid bone deep in the nasal cavity. The roof of the sphenoid sinus is related to the middle cranial fossa and the pituitary gland in the Sella turcica. Isolated sphenoid sinus involvement is relatively uncommon, isolated sphenoid sinus is a rare disease occurs at an incidence of 2.7% of all sinus infections [1,2] usually followed by nasal and ophthalmological nasal symptoms of which 70% are inflammatory, 15% are neoplastic, and around 10% are because of other causes such as cerebrospinal fluid leak and fibrous dysplasia [3,4]. If left untreated or delayed treatment may cause potentially devastating complications such as multiple cranial nerve involvement, brain abscess, and meningitis [1,2]. Any sphenoid sinus infection requires urgent attention, as there are many vital structures in close proximity, including the cavernous sinus, internal carotid artery, and adjacent dura mater posterior aspect of the orbital apex (1,11) which holds the oculomotor (III), trochlear (IV), abducens (VI) and ophthalmic branch of the trigeminal nerve (V1) palsies as well as the optic canal leading to optic nerve (II).

Although our patient's initial Presentation to her GP fulfills International Classification of Headache Disorders (ICHD-3) Migraine without aura [5] also absence of typical Sino nasal symptoms like nasal blockage, congestion, nasal discharge or anosmia, easy to override or misdiagnose sinusitis [6]. Presence of cranial nerve involvement on examination (ex. Right Papilledema, reduced ipsilateral afferent corneal reflex) argues against primary headache (migraine without aura). Among patients with orbital apex syndrome secondary to sinusitis, it has been found that those without radiographic findings of an orbital apex mass have a better prognosis and lower incidence of vision loss [7,8] therefore, when imaging reveals signs of mucosal invasion, early surgical debridement and directed accurate antibiotic therapy are crucial to prevent irreversible loss of vision. Other important and treatable conditions that present with painful recurrent ophthalmoplegia, such as Tolosa-Hunt syndrome, and immune-mediated systemic inflammatory disorders [9,10], such as Immunoglobulin G4 (IgG4)-related disease, new daily persistent headache were also in our differentials. However, MRI findings revealed features suggestive of an infection, rather than being inflammatory or immune mediated, as these cause more nonerosive lesions and negative histology (negative for plasma cells). Our patient was promptly diagnosed, investigated and treated with no delay, resulting in full recovery within 2 weeks.

Although most cranial nerve palsies associated with sphenoid sinus infection is 6th cranial (Abducens nerve) followed by 3rd (oculomotor nerve) cause of its direct extension of inflammatory process from sphenoid sinus expanding to cavernous sinus also can

cause compression of the optic nerve by sphenoid mass that expanding to the superior orbital fissure and cavernous sinus or orbital apex as was the cause of papilledema in our patient. Most of infective or inflammatory cranial neuropathies resolve with conservative treatment (intravenous antibiotics and steroids) sometimes also requires surgical drainage and debridement.

Conclusion

Any headache that too if associated with exertional element, aggravated by standing, walking, bending, coughing etc. or any unilateral peri-orbital pain or retro-orbital pain on exertion, requires thorough neurological examination, warrants appropriate investigations to rule out intracranial cause and urgent specialist attention. Any intractable or new-onset persistent headaches with intermittent severity on exertion or a patterned occurrence, and which interfere with sleep and are not relieved by simple analgesics, should alert one to the diagnosis of secondary or sinister headache and not necessarily migraine. Prompt diagnosis and appropriate treatment carry an excellent clinical outcome.

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