



Research Paper

Ocular cranial nerve palsies secondary to sphenoid sinusitis



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Abstract *Objective:* The clinical presentation of sphenoid sinusitis can be highly variable. Rarely, sphenoid sinusitis may present with cranial nerve complications due to the proximity of these structures to the sphenoid sinus.

Method: A case series from Rabin Medical Center and all cases of cranial nerves palsies secondary to sphenoid sinusitis that have been reported in the literature were reviewed.

Results: Seventeen patients were identified. The abducent nerve was the most common cranial nerve affected (76%), followed by the oculomotor nerve (18%). One patient had combined oculomotor, trochlear and abducent palsies. The most common pathology was isolated purulent sphenoid sinusitis in 64% followed by allergic fungal sinusitis (AFS) in 18%, and fungal infection in 18%. 94% had an acute presentation. The majority (85%) received a combined intravenous antibiotics and surgical treatment. The remainder received conservative treatment alone. Complete recovery of cranial nerve palsy was noted in 82% during follow up.

Conclusion: Sphenoid sinusitis presenting as diplopia and headaches is rare. A neoplastic process must be ruled out and early surgical intervention with intravenous antimicrobial therapy carry an excellent outcome with complete resolution of symptoms.

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Introduction

Isolated sphenoid sinusitis (ISS) is a rare disease which accounts for 1%–2% of all paranasal sinus infections.^{1–4} ISS can be difficult to diagnose because of its subtle onset and typical symptoms of headache and nonspecific facial pain.⁵

In severe cases, infection may spread to the nearby anatomical structures including cranial nerves (CN), cavernous sinus, internal carotid artery and pituitary gland, resulting in cranial neuropathy and death.^{6–8} Therefore, early antibiotic treatment or/and surgical intervention to the affected sinuses are warranted.^{2,9}

In rare cases, patients may present with diplopia and headache as the only symptoms. Ocular cranial nerve involvement is an uncommon complication of sphenoid sinusitis (SS) (6%–12%).¹⁰ The abducent nerve is the most common cranial nerve to be affected due to its medial anatomical location in the cavernous sinus and proximity to the sphenoid sinus, followed by the optic nerve and oculomotor nerve. Data regarding cranial neuropathy associated with sphenoid sinusitis, its clinical course and appropriate treatment are scarce and warrant further investigation.

In this study, we report a case series of ocular cranial nerve (III, IV and VI) palsies secondary to SS from our institution, and integrate additional cases published in the literature. We present data regarding the clinical presentation, imaging and outcomes in this patient group.

Materials

Following IRB approval, all cases of sphenoid sinusitis associated with ocular cranial nerve palsies (CN III, IV and VI) treated in our institution since 2000 were retrieved. In addition, a Pubmed search was conducted to identify all similar cases reported in the literature. Patients with diagnoses of benign or malignant tumors, acute invasive fungal infection and mucoceles were excluded. Patients with a diagnosis of cavernous sinus thrombosis on imaging were excluded as well. Thus, only patients with isolated ocular cranial nerve palsies and diplopia as their main symptom at presentation, associated with sphenoid sinusitis, were included.

Patient demographics, clinical data, imaging studies data, treatments modalities, follow up and outcomes were evaluated. Publications lacking patient-specific data were excluded.

Complete resolution was defined as a full recovery, subjective and objective, of cranial nerve deficits. Partial resolution was defined as subjective, patient-reported symptomatic improvement of diplopia and ocular motor restrictions. No resolution was defined as the absence of patients' subjective symptomatic improvement and the persistence of diplopia.

Results

A total number of 17 cases were identified, 4 cases from our institution and additional 13 reported in the literature (Table 1). The age range was 12–86 years. No gender

predilection was noted. The most common symptoms were diplopia in 100% of the patients followed by headaches and retro-orbital pain in 70% of the patients.

The most common sphenoid pathology associated with ocular CN palsies was purulent sphenoiditis (64%, $n = 11$). Seven of these 11 patients had isolated sphenoiditis. Three had a background of chronic rhinosinusitis, and one patient had contralateral sinusitis. In 18% ($n = 3$) of patients, allergic fungal sinusitis (AFS) of the sphenoid sinus was diagnosed, and in 18% ($n = 3$) a diagnosis of isolated chronic fungal infection was made. The majority (76%) of patients had isolated 6th CN palsy, 18% had isolated 3rd CN palsy and one patient (6%) had multiple CN palsies involving the 3rd, 4th and 6th nerves unilaterally. 12% ($n = 2$) had bilateral 6th CN palsy, one patient with AFS and the second with chronic invasive fungal infection. The right and left eyes were equally affected.

All patients underwent an imaging study. In none a neoplasm was suspected. Computed tomography scan was performed in 71% ($n = 12$) of patients and MRI was performed in remainder of patients. Complete opacification of the involved sphenoid sinus without bony erosion was noted in the majority of cases (88%). In the remaining patients, one had erosion of the sphenoid sinus roof and in the second, complete erosion of the lateral wall was noted. Interestingly, in this patient palsies of CN 3, 4 and 6 was noted. Abnormal signal intensities in both T1 and T2 within the sphenoid sinus, correlated to inflammatory process without signs of malignancy, were noted in those cases where MRI was performed.

Microbial culture was taken in 70% ($n = 12$) of patients. Positive culture was noted in 50% ($n = 6$). In two patients, *Staphylococcus aureus* and *Streptococcus pyogenes* were isolated concurrently. In two other patients, only a description of a gram(+) isolates was noted and in remaining two patients, *Aspergillus* was isolated.

In two patients the cranial neuropathy and symptoms resolved with conservative treatment alone (intravenous antibiotics and steroids) and complete recovery was noted after 3 weeks treatment. Fifteen patients underwent surgical treatment and concurrent intravenous antibiotics. 61% ($n = 8$) of the surgically treated patients underwent prompt surgical drainage and debridement of the affected sphenoid sinus within 1–5 days from onset of symptoms (mean = 2.5 days). All of these patients showed complete resolution of the affected cranial nerves following surgery. In 3 patients, delayed surgery (2–42 months following presentation of diplopia) was performed with two gaining full recovery and one patient with no recovery. In the remaining 4 patients, there was no data regarding the timing of surgical intervention from onset of symptoms but full recovery of CN palsy was noted in two patients and the remaining two patients had partial recovery during the follow up period. Overall, complete recovery was noted in 82% of patients while partial recovery was seen in 12%.

Discussion

Sphenoid sinusitis commonly presents as part of a multi-sinus infection with typical rhinosinusitis symptoms.

Table 1 Ocular cranial nerve palsies secondary to isolated sphenoid sinusitis.

Cases	Diagnosis	Ocular CN palsy	CT & MRI SCAN		Treatment	Time to resolution	Outcome	
			CT	MRI				
1	Isolated sphenoiditis	RT.III	Opacification of sphenoid w/o bone erosion		Surgical, Abx	1 week	Complete recovery	
2	CRSnNP	LT.VI	Pansinusitis w/o bone erosion		Surgical, Abx	2 weeks	Complete recovery	
3	Sphenoiditis	LT.III	Opacification of sphenoid w/o bone erosion	Abnormal enhancement of left.sphenoid	Surgical, Abx	2 weeks	Complete recovery	
4	Isolated sphenoiditis	RT.III,IV,VI	Opacification of sphenoid with bone erosion		Surgical, Abx, Steroids	1 month	Complete recovery	
5	Ada M et al ⁵ 2004	Isolated sphenoiditis	RT.VI	Opacification of sphenoid with petrous apicitis	Abx, Steroids	2 weeks	Complete recovery	
6	Siu J et al ¹¹ 2015	Isolated sphenoiditis	RT.VI	Opacification of sphenoid w/o bone erosion	Surgical, Abx	6 weeks	Complete recovery	
7	Safouris A et al ¹² 2013	Fungal sphenoiditis	LT.VI		Hyperintensity in left sphenoid sinus	Abx, Steroids	2 weeks	Complete recovery
8	Miller C et al ¹³ 2013	Isolated sphenoiditis	RT.VI	Opacification of sphenoid w/o bone erosion	Surgical, Abx	34 days	Partial recovery	
9	Dores A et al ¹⁴ 2014	Isolated sphenoiditis	LT.III	Opacification and bone erosion of left sphenoid	Surgical, Abx, Steroids	10 days	Complete recovery	
10	Fockaert N et al ¹⁵ 2014	AFS	LT.VI		Abnormal enhancement of sphenoid sinus	Surgical	1 month	Complete recovery
11	Miller C et al ¹³ 2013	AFS	BIL. VI	Opacification of sphenoid w/o bone erosion	Surgical	2 weeks	Complete recovery	
12	Miller C et al ¹³ 2013	AFS	RT.VI	Opacification of sphenoid w/o bone erosion	Surgical	2 weeks	Complete recovery	
13	Miller C et al ¹³ 2013	AFS	LT.VI	Opacification of sphenoid w/o bone erosion	Surgical, Abx, Steroids	1 month	Partial recovery	
14	Lee LA et al ¹⁶ 2002	Fungal ball	LT.VI		Abnormal signal intensity in the left sphenoid	Surgical, Anti-fungal	1 month	Complete recovery

(continued on next page)

Table 1 (continued)

Cases	Diagnosis	CT & MRI SCAN		Treatment	Time to resolution	Outcome
		Ocular CN palsy	MRI			
15 Lee LA et al ¹⁶ 2002	Chronic invasive fungal sinusitis	BIL. VI	Abnormal enhancement over bil.sphenoid	Surgical, Anti-fungal	17 months	Complete recovery
16 Lee LA et al ¹⁶ 2002	Sphenoiditis	LT.VI	Abnormal signal intensity in the left sphenoid	Surgical, Abx	No resolution	No resolution
17 Lee LA et al ¹⁶ 2002	Sphenoiditis	LT.VI	Moderate opacification of sphenoid w/o bone erosion	Surgical, Abx	4 months	Complete recovery

Isolated sphenoid sinusitis is an uncommon pathology that accounts for 1.0%–2.7% of all sinus diseases.^{1–4} Rarely, diplopia with ocular cranial nerve palsy may be the initial presenting symptom of sphenoid sinusitis. Diagnosis is typically achieved using imaging studies (CT and or MRI).

Of note, the majority of isolated sphenoid sinus lesions presenting with ocular cranial nerve palsy are associated with a neoplastic process in the sphenoid sinus,^{1,4} either arising in the sphenoid sinus or extending into it from an intracranial process. Thus, a neoplastic process should be ruled out in the event of such cases.

In this study, we reviewed 17 cases with ocular cranial nerve palsy secondary to sphenoid sinusitis. Four cases were from our institution and an additional 13 cases that were retrieved in our search of the literature. In the majority of patients, a diagnosis of bacterial sinusitis was made, and in the remainder AFS and fungal infection were diagnosed. Most cases had isolated disease in the sphenoid sinus. In approximately a third of bacterial sinusitis cases, infection occurred on a background of chronic rhinosinusitis without nasal polyposis.

The most common cranial nerve palsy associated with sphenoid infection in our series was the 6th nerve, followed by the 3rd nerve. Several possible mechanisms for development of cranial nerve palsy associated with sphenoid disease are suggested in the literature: 1. Direct extension of inflammatory process from sphenoid sinus to infect the nerve sheath and result in palsy. 2. Compression of the nerve by sphenoid mass that extending to the cavernous sinus or superior orbital fissure. 3. Vasculitis or cavernous sinus thrombosis causing an ischemic infarct of the cranial nerve.¹⁷ The 6th CN is suspected to be the most commonly involved due to its medial location in the cavernous sinus.

Interestingly, in one case a contralateral 6th CN palsy developed. A possible explanation for this was that the patient had a dominant contralateral sphenoid sinus crossing the midline and adjacent to the contralateral cavernous sinus. In two cases, a bilateral 6th nerve palsy was noted. In both a bilateral involvement of the sphenoid sinus was noted on imaging. Thus, clinical presentation and imaging findings may vary and physicians should be alerted to this during workup of such patients.

In rare cases, sphenoid sinus disease complicated by cranial neuropathy could progress to potentially life-threatening complications such as meningitis, cerebritis, cavernous sinus thrombosis or internal carotid artery involvement. Thus, prompt diagnosis and early surgical intervention is crucial to prevent these serious complications.

In this series, complete recovery was noted overall in 82% of the patients. Only one patient remained with ocular CN palsy following medical and surgical interventions. This was likely related to the delay in surgical treatment, since the patient underwent surgery 42 months following onset of diplopia and CN VI palsy, in contrast to the remainder of surgically treated patients who had surgical interventions with a mean of 2.5 days following onset of symptoms. Prompt surgical intervention was associated with complete recovery of the ocular cranial neuropathy.

Conclusion

Sphenoid sinusitis presenting as diplopia and headaches is rare. A neoplastic process must be ruled out and early surgical intervention with intravenous antimicrobial therapy carry an excellent outcome with complete resolution of symptoms.

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