

Endoscopic Sinus Surgery for Solitary Abducens Palsy in Patients with Isolated Sphenoid Sinus Disease: Report of Four Cases

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Solitary abducens palsy secondary to isolated sphenoid sinus disease (ISSD) is rare and early management is important. There is no report regarding the results of endoscopic sinus surgery (ESS) for prolonged abducens palsy due to ISSD. We present four cases of ISSD with solitary abducens palsy that received ESS from 1995 through 2000. The interval between onset of diplopia and ESS was longer than 96 hours (range, 4 days to 42 months). The sphenoid lesions were caused by Aspergillosis in two patients and inflammation in two. Three patients recovered completely from abducens palsy after 4-17 months and had no surgical complications. In conclusion, ESS is a safe and effective treatment for ISSD with abducens palsy. Recovery from abducens palsy is slow and progressive. Improvement of extraocular movement is an early sign of recovery. (*Chang Gung Med J* 2002;25:689-94)

Key words: isolated sphenoid sinus disease, abducens palsy, endoscopic sinus surgery.

Isolated sphenoid sinus disease (ISSD) accounts for 1% to 3% of all sinus lesions.⁽¹⁻³⁾ Isolated sphenoid lesion is easily misdiagnosed because of its subtle onset and vague symptoms.⁽¹⁻⁴⁾ Since the 1990's, this disease has been well documented and permanent cranial neuropathy and death have been reported.⁽¹⁻⁵⁾ Therefore, early antimicrobial therapy and/or surgical treatment are warranted.⁽²⁻⁴⁾ For cases in which the orbital complication resulted from sphenoidal sinus, surgical drainage or decompression of sphenoidal sinus and orbital suppuration are regarded as the quickest methods of treatment.⁽⁶⁾

Involvement of the sixth cranial nerve in ISSD is unusual.⁽¹⁾ Surgical intervention should be introduced as soon as possible after medical therapy failed.^(3,6) The role of endoscopic sinus surgery (ESS) for prolonged abducens palsy resulting from ISSD is still unclear. There have been no articles discussing this condition in English literature. Herein, we pre-

sent four rare cases of ISSD with solitary abducens palsy that lasted for more than 48 hours before ESS at Chang Gung Memorial Hospital from February 1995 through May 2000. We also briefly discuss a review of the literature about this disease.

CASE REPORT

Case 1

A 46-year-old healthy woman had suffered from intermittent headaches and bilateral retroorbital pain since May 1997. In June 1997, she presented with double vision for 2 days. After neurological and ophthalmologic examinations, bilateral abducens palsies were noted. Subsequently she was referred to our clinic for further examination. Physical examination revealed that a little mucopus had accumulated on the nasopharynx but no tumor. A magnetic resonance image (MRI) with gadolinium showed

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normal cerebrum and midbrain but abnormal enhancement over bilateral sphenoid sinuses. Endoscopic sinus surgery was performed under general anesthesia on the fourth day after presentation of diplopia. The bilateral sphenoid sinuses were filled with clay materials and a bony defect of the left sphenoid roof was also noted. Aqueous better-iodine and isotonic sodium chloride solutions were used for irrigation following aspiration of pus. The wound was filled with gentamicin ointment. The pathology showed *Aspergillus* infection and granulomatous inflammation of the mucosa. Therefore, invasive aspergillosis of bilateral sphenoid sinuses with bilateral abducens palsy was diagnosed. The antimicrobial drug was subsequently switched from penicillin G sodium to amphotericin B because the wound recovered slowly. She received regular debridement and sinusoscopic examination at our clinic and the sphenoidal mucosa healed within 6 months. Her extraocular movement (EOM) improved during the follow-up period. The diplopia had completely resolved within 17 months.

Case 2

A 45-year-old man received a Caldwell-Luc operation for chronic paranasal sinusitis in 1982. He had suffered from double vision since April 1995. He visited our neurology and ophthalmology outpatient clinic complaining of deteriorating diplopia in May 1998. A physical examination showed normal findings except for left sixth cranial nerve palsy. No preoperative medication was given. A MRI showed abnormal signal intensity in the left sphenoid sinus in both T1 and T2 weighted images as gray in comparison with the brain. He was referred to our clinic and neither nasopharyngeal tumors nor mucopus were noted. We performed an ESS to explore the sphenoid sinus in June 1998 (42 months after onset of diplopia). The redundant posterior end of the middle turbinate was partially resected for a better view of the sphenoidal recess. We found mild edema and diffuse fibrosis of sphenoidal mucosa after a left sphenoidotomy. The pathology revealed chronic inflammation but no malignant changes. Prophylactic antibiotic therapy (penicillin G sodium) was prescribed. He also received regular debridement at our clinic. Unfortunately, his diplopia did not resolve and an ophthalmologic surgery was undertaken in September 1998 to recess the left

medial rectus muscle and to resect the left lateral rectus muscle. His sphenoidal mucosa had healed within 6 months of the surgery.

Case 3

A healthy 67-year-old woman complained of intermittent headaches over the left temporal and frontal areas and progressive exacerbation of double vision since July 1998. She had taken acetaminophen for relief of her headache. One month after taking the medication, left abducens palsy and no other deficits were noted. She was referred to our clinic for further evaluation. An MRI showed an abnormal signal enhancement of the left sphenoid sinus in T1-weighted and T2-weighted images (Fig. 1). Endoscopic sinus surgery was performed under general anesthesia in October 1998 (3 months after the onset of diplopia). Fungal balls were found in the sphenoid sinus. After removal of the materials and diseased mucosa, the residual mucosa was clear and the bony wall was intact. Then the sphenoid sinus was irrigated with aqueous better-iodine and isotonic sodium chloride solutions, and then filled with gentamicin cream. The pathology was diagnosed as *Aspergillus* sphenoid sinusitis. Prophylactic antibiotics with penicillin G sodium were given for 7 days and intensive debridements



Fig. 1 Axial magnetic resonance scan with gadolinium showing abnormal enhancement of left sphenoid sinus (case 3).

were done. Her wound recovered well and no further complications developed. Her mucosa healed within 1 month and the left abducens palsy resolved completely within 6 months.

Case 4

A 69-year-old man received a Caldwell-Luc operation for chronic paranasal sinusitis in 1982. In April 2000, he presented with left intermittent retroorbital pain and progressive double vision for 1 month. Neither exophthalmos nor periorbital infection was found. The left sixth cranial nerve deficit was impressed. His pain was relieved by acetaminophen. A computed tomography (CT) scan showed moderate opacification of the left sphenoid sinus (Fig. 2). He was subsequently referred to our clinic. The nasopharyngeal mucosa was smooth. Endoscopic sinus surgery was performed in May 2000 (2 months after the onset of diplopia) and the left sphenoidotomy after ethmoidectomy was carried out. The thick jelly-like pus in the sphenoid sinus was aspirated. The inflamed mucosa was removed medially and inferiorly after irrigation with betteriodine and isotonic sodium chloride solutions. Pathology revealed chronic inflammation and no malignant cells. He received antibiotic therapy with penicillin G sodium for 3 days and regular debride-

ments. The mucosa healed within 3 months and the diplopia had disappeared within 4 months.

DISCUSSION

Sphenoid sinus disease is usually accompanied by an involvement of the other sinuses and ISSD is rare. Direct spreading from the ethmoid sinusitis, anatomic variation, or iatrogenic trauma may change the function of drainage and predispose the area to the disease.^(3,4) Several researchers have demonstrated that ISSD frequently occurs in middle aged (about 40 to 50 years old) and, as in our cases, elderly (mean age, 57 years old) patients.^(1,2,4) The etiologies of ISSD include inflammatory diseases, neoplasms, fibrous disease, encephalocele and internal carotid artery aneurysms.⁽¹⁾ In our report, inflammatory disease resulted in their disorders (fungal infection: 2; chronic inflammation: 2) and a previous sinus surgery seemed to be a predisposing factor for this disease because two cases had undergone Caldwell-Luc surgery (Table 1).

The presenting symptoms and signs in our cases were headache, retro-orbital pain, and visual disturbance. The onset of symptoms was subtle and the course progressed slowly. Therefore, patients usually had a prolonged course.⁽⁴⁾ In addition, indistinct symptoms and signs made it difficult to diagnose this disease earlier.⁽¹⁻⁴⁾ Lesions of the sphenoid sinus might involve the adjacent structures including the optic, abducens,⁽¹⁾ and oculomotor nerves.⁽²⁾ We should note that an involvement of the abducens nerve was more frequently associated with malignancies because half of the neoplastic cases presented with abducens palsy.⁽¹⁾ Endoscopic examination and MRI may provide a better view of the soft tissue.

The possible mechanisms for the development of abducens palsy from ISSD include 1) inflammation from sphenoidal sinusitis to infect abducens nerve sheath and to result in nerve palsy, 2) the sphenoid mass expanding to the cavernous sinus or superior orbital fissure and compressing the abducens nerve, and 3) vasculitis or cavernous sinus thrombosis causing ischemic infarction of the abducens nerve.⁽⁶⁾ We think the dorsal clival artery, which arises from the meningohypophyseal trunk and supplies the proximal portion of the intracavernous abducens nerve may play a significant role in the isolated abducens palsy.⁽⁷⁾

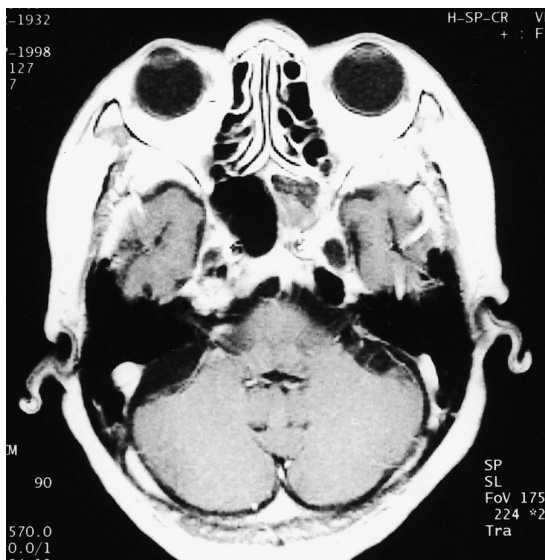


Fig. 2 Axial computed tomographic scan with contrast showing opacification of left sphenoid sinuses (case 4).

Table 1. Clinical Characteristics of 4 Patients with Solitary Abducens Palsy Due to Isolated Sphenoid Sinus Disease

Patient number	Age (Y)/ Sex	Predisposing factor	Symptoms	Interval**?	Diagnosis	Surgical techniques	Follow-up period (M)	Outcome
1	46/Female	-	Headache, retro-orbital pain, diplopia, nasal obstruction	4 D	Aspergillosis of sphenoid sinuses (Bil)	Sphenoidotomy	17	Complete recovery
2	45/Male	Operation*	Diplopia	42 M	Isolated sphenoid sinusitis (L)	Sphenoidotomy	6	Failure ; ophthalmic surgery after 3 months
3	66/Female	-	Headache, diplopia	3 M	Aspergillosis of sphenoid sinus (L)	Posterior ethmoidectomy and sphenoidotomy	6	Complete recovery
4	69/Male	Operation*	Diplopia, retro-orbital pain	2 M	Isolated sphenoid sinusitis (L)	Posterior ethmoidectomy and sphenoidotomy	4	Complete recovery

Abbreviations: Y: year; D: day; M: month; Bil: bilateral; L: left.

*: Caldwell-Luc operation 20 years ago;

** : interval between onset of symptoms and operation;

The common pathogens isolated from acute sphenoid sinusitis are *Staphylococcus aureus*, *Streptococcus* species, and *Aspergillus*. Gram-negative and anaerobic organisms are more associated with chronic sphenoid sinusitis.^(2,3) Broad-spectrum antimicrobial therapy is recommended as the first-line treatment of ISSD. However, surgical intervention should be performed when the symptoms become worse, they are sustained for 24 to 48 hours or complications appear.^(2,3,6) Various surgical techniques have been used to treat patients with ISSD. During the past 20 years, ESS is one choice of surgery for sphenoid sinus lesion.⁽¹⁻⁵⁾ Endoscopic sinus surgery is safe and feasible when performed by a well-trained otolaryngologist. As a rule of thumb, ESS for sphenoid sinus diseases should avoid damaging the nearby important structures, such as the optic nerve and internal carotid artery.⁽⁶⁾ Surgeons should clearly identify these vital organs and refrain from over-manipulation.

We tried to resolve the prolonged diplopia secondary to sphenoid lesions by ESS and antimicrobial therapy; the intervals between onset of diplopia and sphenoidotomy were from 4 days to 42 months. Fortunately, the outcomes of our patients were good. Three cases completely recovered within 4 to 17 months, however, one had no improvement. We believed that the prolonged interval between onset (42 months) in case 2 was probably the cause for his

irreversible neuropathy. As compared with the results reported by Muneer et al, the recovery time of our patients was longer (4 to 14 months versus 24 to 48 hours).⁽⁵⁾ It seems that the longer the diplopia persists, the longer the recovery time of is needed. Certainly, the degree of severity and duration of the cranial nerve deficit influenced the outcome. The course of recovery from the prolonged diplopia is slow and long. Therefore, this result accounts for the necessity of a long-term follow-up. We also suggest regular and continued debridements and sinusoscopic examinations to prevent further inflammation. In addition, an improvement of the EOM is a good sign for recovery. Finally, bleeding, cerebrospinal fluid rhinorrhea, and damage of nearby structures may also effect the prognosis.

In summary, although surgical interventions were delayed for more than 48 hours, the chance of complete recovery still existed after ESS. The course of recovery was slow and the improvement of EOM was an early sign of recovery.

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以內視鏡鼻竇手術治療源自孤立性蝶竇疾病之單獨的 外展神經麻痺：四例報告

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源自孤立性蝶竇疾病之單獨的外展神經麻痺是罕見的，並且及早處理是重要的。並沒有文獻報告以內視鏡鼻竇手術治療導因於孤立性蝶竇疾病的較長期之外展神經麻痺的結果。我們報告4例病患於1995年至2000年接受內視鏡鼻竇手術治療孤立性蝶竇疾病併單獨的外展神經麻痺。病人接受內視鏡鼻竇手術與複視出現的時間間隔大於96小時（範圍，4天到42個月）。其中，2個蝶竇病變為麴菌感染及2個為發炎所造成。有3位病人的外展神經麻痺經過內科及外科的治療4至17個月後完全恢復且沒有手術併發症產生。以內視鏡鼻竇手術來處理孤立性蝶竇疾病併外展神經麻痺是一個安全及有效的治療。外展神經麻痺的復原是緩慢且漸進的。而動眼機能的進步可視為復原的一個早期徵象。(長庚醫誌 2002;25:689-94)

關鍵字：孤立性蝶竇病變，外展神經麻痺，內視鏡鼻竇手術。