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Case Report

A rare case of septic cavernous sinus thrombosis as a complication of sphenoid sinusitis

*Ming-Chun Chen[a], Yu-Huai Ho[b], Pau-Nyen Chong[c], Jui-Hsia Chen[a]**

[a]Department of Pediatrics, Hualien Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation, Hualien, Taiwan, [b]Department of Infectious Disease, Hualien Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation, Hualien, Taiwan, [c]Department of Medical Imaging, Hualien Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation, Hualien, Taiwan

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Abstract

Isolated sphenoid sinusitis is a rare disorder and may present with severe complications due to its proximity to the orbital and intracranial areas. We report a 13-year-old boy hospitalized for septic shock with fever of unknown origin. Facial palsy was later noted. Brain magnetic resonance imaging showed a sphenoid mass and right cavernous sinus and internal jugular vein thrombosis. Biopsy revealed chronic rhinosinusitis. Complete recovery followed by an incision/drainage procedure and antibiotic treatment. Acute sphenoid sinusitis should be included in the differential diagnosis of septic manifestations mimicking central nervous system infection or cranial nerve palsy.

Keywords: *Acute sphenoid sinusitis, Cavernous sinus thrombosis, Children, Cranial nerve palsy*

Introduction

Acute paranasal sinusitis is a common disorder in the pediatric population, and orbital complications, mostly resulting from ethmoid sinusitis, account for 80% of cases [1]. Intracranial complications such as meningitis and cavernous sinus thrombosis accounted for only 13% of cases and were mainly secondary to frontal sinusitis and rarely induced by sphenoiditis [1]. We herein report a 13-year-old boy who presented with fever of unknown origin, septic shock accompanied by right neck pain, and facial palsy due to sphenoiditis-induced right cavernous sinus and internal jugular vein thrombosis. Complete recovery followed by an incision/drainage procedure and antibiotic treatment. Acute sphenoiditis should be included in the differential diagnosis of septic manifestations mimicking central nerve system (CNS) infection or cranial nerve palsy.

Case report

A 13-year-old boy with a pertinent medical history of only allergic rhinitis was hospitalized for septic shock with fever of unknown origin. He had gone diving 10 days prior to admission (PTA). Six-day PTA, he had a fever (38°C) followed by headache, nausea, and vomiting but no diarrhea. Pertinent serologic examination at another hospital revealed white blood cells (WBCs) 10,170/μL, segments 92.6%, and C-reactive protein (CRP) 28.67 mg/dL. The chest radiograph and urinalysis showed no abnormalities. The family refused a lumbar puncture. Intravenous (IV) penicillin was given and the fever gradually subsided. Four-day PTA, right neck pain occurred without a history of trauma or any particular findings in the aforementioned area. One-day PTA, the fever rose to 39°C; right neck pain progressed and he became drowsy. Upon admission, his body temperature was 36.9°C, pulse rate 96/min, respiratory rate 18/min, and blood pressure 70/30 mmHg. Physical examination showed no abnormalities except tenderness on the right side of the neck level II of the submandibular angle without nuchal rigidity, cervical lymphadenopathy, wound, or skin discoloration. Dopamine was given for his shock status on admission. Laboratory data were WBC – 7510/μL, bands – 49%, segments – 32%; platelets – 13,000/μL; CRP – 31.21 mg/dL; slightly elevated fibrinogen/D-dimer; and a normal prothrombin time/activated partial thromboplastin time (PT/aPTT). Fiberscopic examination revealed no evidence of deep neck infection. Under suspicion of atypical infection, IV penicillin and levofloxacin were started and his clinical condition improved. Dopamine was discontinued on the 2nd hospital day (HD), and his fever subsided on the 5th HD. However, right neck pain persisted accompanied by right peripheral facial palsy and ptosis but

**Address for correspondence:*
Dr. Jui-Hsia Chen,
Department of Pediatrics, Hualien Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation, 707, Section 3, Chung-Yang Road, Hualien, Taiwan.
E-mail: chenjh@tzuchi.com.tw

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no visual impairment. Tests for serum Lyme antibody IgG and IgM were normal. Brain magnetic resonance imaging (MRI) on the 6th HD showed a right sphenoid sinus mucocele with a right cavernous sinus and internal jugular vein thrombosis [Figure 1]. Endoscopic incision/drainage was performed. Pathology revealed chronic sphenoiditis. Blood cultures grew *Peptostreptococcus*; anaerobic pus cultures grew *Prevotella loescheii*, both sensitive to penicillin. Pus acid-fast stain and tuberculosis cultures were negative. After completing a 4-week course of penicillin and ceftriaxone (levofloxacin was shifted to ceftriaxone due to elevating liver enzymes), he was discharged with full recovery of cranial nerve palsy and no neck pain.

Discussion

A thromboembolic event is rare in the pediatric population. Central venous catheter-related thrombosis is the most common etiology of pediatric venous thrombosis followed by infection, inflammation, hypercoagulability, malignancy, and dehydration [2]. Our patient had no medical history of any inflammatory disease or thromboembolic events. Likewise, he had no central venous catheter insertion during hospitalization. Clinical and laboratory data showed no evidence of malignancy or dehydration. There was no thrombocytosis and PT/aPTT levels were normal, which were not consistent with hypercoagulation syndrome. His urinalysis was normal, excluding hypercoagulable state-induced thromboembolic complications of nephrotic syndrome. Thus, infection (sphenoiditis)-related right cavernous sinus and internal jugular vein thrombosis were most likely in our case.

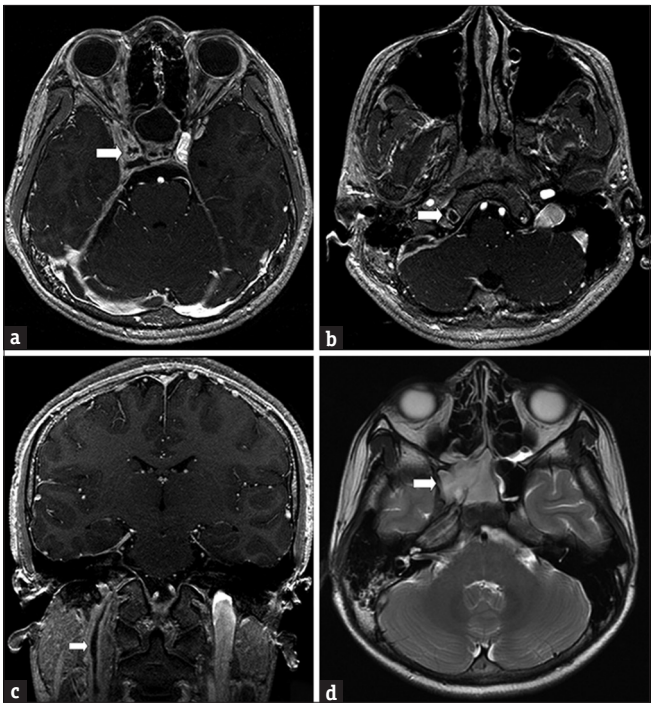

Figure 1: Axial slice on a T1-weighted image after gadolinium–diethylenetriaminepentacetate administration focusing on the cavernous sinus (a), jugular fossa (b), and coronal section of the jugular vein level (c). There is nonenhancement in the right cavernous and sigmoid venous sinus, contiguous with the jugular vein, compatible with sphenoiditis-resultant right cavernous sinus thrombosis and deep venous thrombosis of the right cerebrum and neck. T2-weighted image axial section at the mid sphenoid level (d). There is mucus in the right sphenoid sinus (white arrow), right mastoiditis, and a bright signal in the right sigmoid sinus and jugular fossa indicating nonflow void in the venous sinus and jugular vein

The sphenoid sinus has been referred as the "neglected sinus" and is absent until the age of 1 year. It becomes visible through imaging and is clinically relevant between the ages of 5 and 15 [3]. The low incidence of isolated sphenoiditis compared with infection in other paranasal sinuses may be due to fewer drainage problems secondary to few mucous secreting cells along the pseudostratified ciliated epithelium lining of the sphenoid sinus [4]. Thus, isolated sphenoid sinusitis (ISS) is uncommon, affecting only 1–2.7% of patients with paranasal sinusitis [5].

The diagnosis of ISS is usually missed or delayed until neurological symptoms become apparent [6,7]. In acute cases, 86% of patients have headaches, followed by nasal (35%) and ocular (30%) symptoms [8]. *Staphylococcus aureus* is responsible for 24% of acute sphenoiditis, followed by *Streptococcal* species. Anaerobes (*Peptostreptococcus, Prevotella*, *Fusobacterium,* and *Porphyromonas* species), Gram-negative bacilli (*Klebsiella pneumoniae*, *Escherichia coli, and Pseudomonas aurigunosa*) [3], and fungal pathogens (*Aspergillus xavus* and *Aspergillus fumigatus*) are mostly encountered in chronic sphenoiditis [9]. Diving/swimming is associated with acute severe sphenoid sinusitis, whereas allergic rhinitis is related to acute nonsevere forms [3]. Our patient had allergic rhinitis. This may have caused sinusitis leading to sphenoid mucocele formation. Minor trauma and forceful entry of water into the nose during diving may aggravate the progression of sphenoiditis, causing intracranial complications from right cavernous sinus and internal jugular vein thrombosis.

There are many recent reports of ISS presenting with headache and ipsilateral oculomotor nerve or abducens nerve palsy [6,7]. Because indistinct nasal symptoms are often encountered, ISS is difficult to diagnose in the acute stage, so a high index of suspicion is necessary for early and accurate diagnosis. Neurological symptoms including headache which does not respond to symptomatic treatment or septic manifestations mimicking CNS infections, tumors, or vascular diseases require further evaluation to exclude sphenoiditis. Another important disease in the differential diagnosis of child cranial nerve VII palsy is Lyme disease, and bilateral facial nerve palsy is virtually pathognomonic of this disease. However, the serum Lyme antibody test in our patient was negative. High-resolution computed tomography or MRI is the diagnostic study of choice for ISS [10]. Antibiotics for uncomplicated cases include the amoxicillin-clavulanate, cephalosporins, or fluoroquinolons, and treatment is given for about 4 weeks. Immediate surgical drainage is indicated for symptoms occurring >24 h or the presence of neurological/ocular complications, with transnasal endoscopic sphenoidotomy currently used most frequently [3].

In conclusion, acute sphenoiditis should be included in the differential diagnosis of headache not responding to symptomatic treatments or for septic signs and symptoms mimicking CNS infections or cranial nerve palsy.

Declaration of patient consent

The authors certify that the patient and his parents have obtained an appropriate patient consent form. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient and his parents understand that his name and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

References

1. Oxford LE, McClay J. Complications of acute sinusitis in children. Otolaryngol Head Neck Surg 2005;133:32-7.
2. Cox M, Epelman M, Chandra T, Meyers AB, Johnson CM, Podberesky DJ, et al. Non-catheter-related venous thromboembolism in children: Imaging review from head to toe. Radiographics 2017;37:1753-74.
3. Marseglia GL, Pagella F, Licari A, Scaramuzza C, Marseglia A, Leone M, et al. Acute isolated sphenoid sinusitis in children. Int J Pediatr Otorhinolaryngol 2006;70:2027-31.
4. Tan HK, Ong YK. Acute isolated sphenoid sinusitis. Ann Acad Med Singapore 2004;33:656-9.
5. Hnatuk LA, Macdonald RE, Papsin BC. Isolated sphenoid sinusitis: The Toronto hospital for sick children experience and review of the literature. J Otolaryngol 1994;23:36-41.
6. Giordano D, Perano D, Marchi S, Raso MG, Barbieri V. Unusual presentation of pediatric acute sphenoid sinusitis. Acta Biomed 2014;85:271-4.
7. Gupta R, Shukla R, Mishra A, Parihar A. Isolated acute sphenoid sinusitis presenting with hemicranial headache and ipsilateral abducens nerve palsy. BMJ Case Rep 2015;2015. pii: bcr2015209408.
8. Gilony D, Talmi YP, Bedrin L, Ben-Shosan Y, Kronenberg J. The clinical behavior of isolated sphenoid sinusitis. Otolaryngol Head Neck Surg 2007;136:610-5.
9. Friedman A, Batra PS, Fakhri S, Citardi MJ, Lanza DC. Isolated sphenoid sinus disease: Etiology and management. Otolaryngol Head Neck Surg 2005;133:544-50.
10. An YH, Venkatraman G, DelGaudio JM. Isolated inflammatory sphenoid sinus disease: A revisitation of computed tomography indications based on presenting findings. Am J Rhinol 2005;19:627-32.