

C A S E R E P O R T

Unusual presentation of pediatric acute sphenoid sinusitis

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Summary. *Background:* Sphenoid sinus lesions are rare entities, occurring in 2 - 3% out of all paranasal sinus lesions. Isolated oculomotor nerve palsy due to acute sphenoid sinusitis is very rare, with only few cases reported in literature. *Methods:* Retrospective report about a case of isolated acute sphenoid sinusitis in a child with a left-sided third cranial nerve paralysis as the only sign at presentation. *Results:* Isolated oculomotor nerve palsy can be the initial sign of an isolated acute sphenoid sinusitis in children and it requires a high index of suspicion in order to avoid a delay in diagnosis. *Conclusions:* Magnetic resonance imaging should be promptly performed. Functional endoscopic sinus surgery represents the treatment of choice in order to restore sinus drainage and avoid further intracranial or ocular complications. The need of serial postoperative debridement under general anesthesia should be adequately scheduled and previously discussed with parents.

Key words: sphenoid sinusitis; paranasal sinus; oculomotor nerve paresis; functional endoscopic sinus surgery; debridement

Introduction

Sphenoid sinus lesions are rare entities, occurring in 2 - 3% out of all paranasal sinus lesions [1,2]. Differential diagnosis includes a variety of conditions such as inflammatory diseases (acute or chronic bacterial and micotic infections), mucoceles, primary or secondary tumors (inverted papillomas, ossifying fibromas, pituitary adenomas) [3]. Headache is reported as the most common complaint, accounting for up to 80-100% of cases [4,5]. Ocular symptoms may be present in up to 22,5% of cases and they are represented by blurred vision, photophobia, progressive vision loss, and diplopia due to sixth nerve paralysis [6,7].

In the present paper we report about a case of isolated acute sphenoid sinusitis in a child with a left-sided third cranial nerve paralysis as the only sign at presentation. Considerations regarding this unusual presentation and concerns related to treatment and postoperative follow up in children have been discussed.

Case report

A 9-year-old boy presented with a 3-week history of diplopia. He had no history of previous diseases, fever, nasal congestion, or headache. Physical examination revealed an isolated superior and inferior rectus oculomotor muscles paresis in the absence of proptosis or orbital cellulitis. Cell blood count, C-reactive protein, and erythrocyte sedimentation rate were normal. The patient underwent neurological examination and electroencephalography that resulted to be otherwise normal. Three days after he developed a vertex headache and a left-sided ptosis. Ophthalmologic evaluation confirmed the presence of a left-sided ptosis and a 2/10 loss of visual acuity in his left eye. Pupil diameter was 5 mm bilaterally but left eye was non-reactive to light because of an afferent papillary defect (Marcus Gunn pupil). Flexible fiberoptic endoscopy of the nasal cavities showed a little inflammatory polyp in the sphenothmoidal recess of the left nasal cavity, masking the natural ostium of the sphenoid sinus. Then, magnetic resonance imaging (MRI) of the brain

and face was performed. The latter revealed an isolated complete opacification of the left sphenoid sinus. On coronal T2-weighted sequences sphenoid sinus appeared to be covered by a highly hyperintense edematous mucosa with central hypointense signal suggestive of empyema (Figure 1). The cavernous sinus could be seen in close proximity of the infectious focus, between the infected sphenoid sinus medially and the medial aspect of the left temporal lobe laterally (Figure 2). Ethmoid sinus was normal bilaterally. On axial T2-weighted sequences the optic nerve could be seen just superiorly and laterally to the sphenoid sinus (Figure 3). An inflammatory polyp was also visible in the left sphenothmoidal recess, just in anteriorly to the anterior wall of the sphenoid sinus (Figure 4). The patient underwent left endoscopic endonasal parasseptal sphenoidotomy that confirmed the presence of a purulent collection within the sphenoid sinus. Endoscopic inspection of the sinus showed a marked edematous inflammatory mucosa. Intravenous ceftriaxone 1 g once a day was then started. Bacteriology revealed a heavy growth of Gram positive cocci (*Streptococcus Milleri*). Patient's signs and symptoms resolved completely after surgery and he was discharged on 4th postoperative

day. After discharge he underwent endoscopic postoperative debridement under general anaesthesia on 8th, 16th, and 25th postoperative day in order to keep a

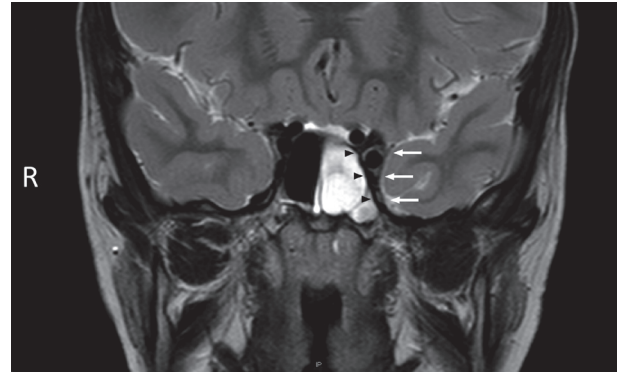


Figure 2. MRI coronal T2-weighted sequence showing the left cavernous sinus in close proximity of the infectious focus, between the infected sphenoid sinus medially (arrowheads) and the medial aspect of the left temporal lobe (arrows).

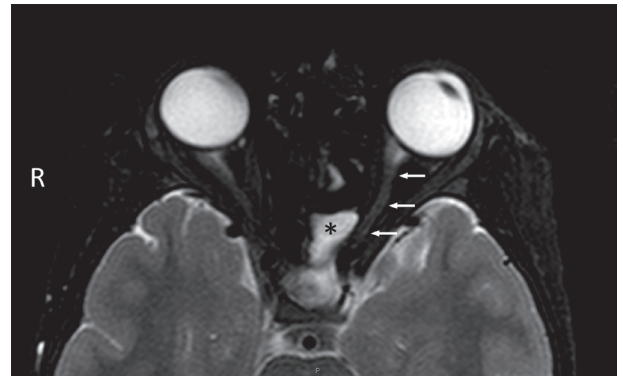


Figure 3. MRI axial T2-weighted sequence showing the optic nerve (arrows) running just superiorly and laterally to the sphenoid sinus, in close proximity to the infectious focus (asterisk).

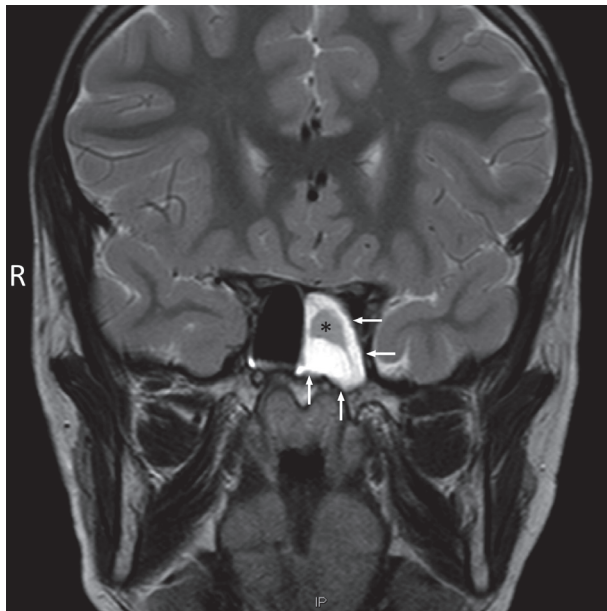


Figure 1. MRI coronal T2-weighted sequence showing the left sphenoid sinus covered by highly hyperintense edematous mucosa (arrows) with central hypointense signal suggestive of empyema (asterisk).

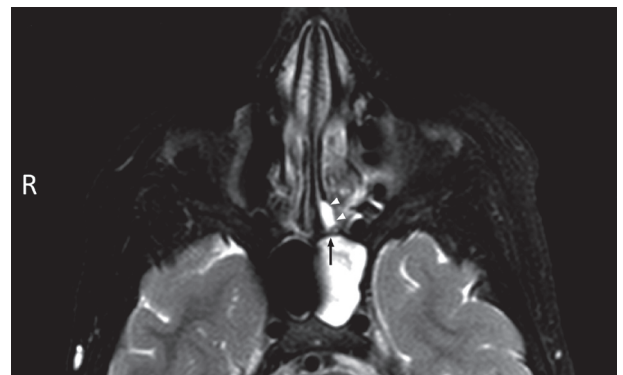


Figure 4. MRI coronal T2-weighted sequence showing the left cavernous sinus in close proximity of the infectious focus, between the infected sphenoid sinus medially (arrowheads) and the medial aspect of the left temporal lobe laterally (arrows).

proper healing and patency of his left sphenoid sinus ostium. Two months after surgery flexible fiberoptic nasal endoscopy showed a completely healed and patent left sphenoidotomy.

Discussion

Isolated oculomotor nerve palsy due to acute sphenoid sinusitis is very rare with only few cases reported in literature [8-11]. To the best of our knowledge, this is the first case reported in a child. Oculomotor nerve runs from the brainstem to the superior orbital fissure through the roof of the cavernous sinus, which is located just laterally to the sphenoid sinus, in contact with its lateral wall. Sinusitis can influence ocular motility by affecting single muscles or a combination of muscles and/or cranial nerves due to a local inflammatory tissue reaction. In the present case an inflammatory polyp in the left sphenoidal recess may have altered the sinus drainage through the natural ostium, thus triggering the infectious process. Based on a cadaveric study some authors have showed the presence of bony dehiscence in the lateral wall of the sphenoid sinus. Moreover, the wall of the optic canal may be thinner than 0.5 mm [12]. These findings may have represented "loci minoris resistentiae" that have favoured spread of infection to oculomotor and optic nerves. The possibility that infection can spread through diploic veins has been reported [13]. The latter may have represented an alternative mechanism that could explain the spread of infection from sphenoid sinus to the cavernous sinus.

History of rhinitis or nasal congestion, headache, fever, or visual disturbance are findings that can promptly suggest an inflammatory focus in the paranasal sinus. Nevertheless, unusual presentation is possible and it may cause a delayed diagnosis and requires a high index of suspicion in order to avoid further progression of ocular and intracranial complications [14]. In such cases imaging study of the brain and the face should not be delayed. MRI provides an excellent spatial resolution of this area and depicts with great accuracy the presence of ocular or intracranial involvement.

Endoscopic endonasal surgery represents the treatment of choice in order to restore the drainage

and release pressure within the sinus. Anterior sphenoidotomy can be performed through an endoscopic endonasal parasseptal approach, which minimizes iatrogenic damage to normal nasal mucosa and postoperative crusting. Nevertheless, it should be kept in mind that the narrow space of the nasal fossa in a pediatric patient, if compared to adults, may require excision of the middle turbinate or a transethmoidal approach in order to reach the sphenoidal recess. This situation should be adequately discussed with parents because it may increase the rate of complications and prolong the healing process.

Postoperative serial debridement is advocated by various authors as an essential part for long-term success of surgical treatment [15,16]. This allows to maintain sinus ostium patency during healing process, reduce pooling of mucus, prevent infection. Nevertheless, little evidence supports its need or defines the extent to which it is necessary [17]. Although postoperative debridement after endoscopic sinus surgery is generally well tolerated when performed in awake adult patients it represents a major concern in pediatric population. This procedure is generally poorly tolerated in the awake pediatric patient and it may require general anesthesia. The surgeon should keep in mind this concern in order to properly plan a serial postoperative debridement schedule, and appropriately inform and discuss its implications with parents.

In conclusion, isolated oculomotor nerve palsy can be the initial sign of an isolated acute sphenoid sinusitis in children and it requires a high index of suspicion in order to avoid a delay in diagnosis. In such cases MRI should be promptly performed. Functional endoscopic sinus surgery represents the treatment of choice in order to restore sinus drainage and avoid further intracranial or ocular complications. The need of serial postoperative debridement under general anesthesia should be adequately scheduled and previously discussed with parents.

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- Received: 20 February 2014
Accepted: 4 March 2014
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