



Intermittent esotropia and ataxia in a child with late diagnosis of empyema in the sphenoid sinus - a case report*

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Abstract

Background: Acute bacterial rhinosinusitis is a common condition often affecting children with inflammation of the mucous membrane in one or more nasal sinuses. Isolated sphenoid sinus infections can exhibit atypical or no symptoms before potentially severe suppurative complications develop. Awareness of early signs of complications is crucial to avoid delays in referral, diagnosis, and treatment. We present the first detailed report of a child with sphenoid sinus empyema with the symptoms intermittent unilateral esotropia and limb and gait ataxia. This case emphasizes the need to recognise atypical presentations, aiding early diagnosis and treatment to prevent severe, possibly life-threatening complications.

Case presentation: A 7-year-old girl, previously healthy, presented with a months-long decline in health marked by general malaise, significant weight loss, and recurrent upper respiratory infections. Despite multiple primary care visits no antibiotics were prescribed because infection parameters were found to be normal. Subsequently, the emergence of intermittent double vision prompted referral to the pediatric emergency department. Examination revealed a wide-based gait, ataxic finger-nose-finger and knee-heel tests, and intermittent esotropia of the left eye. Neurological examination was otherwise unremarkable. Imaging of the cerebrum was performed on the suspicion of intracranial pathology and found isolated empyema within the left sphenoid sinus. Endoscopic sinus surgery and antibiotic treatment led to a full recovery within a month. The accommodative esotropia, likely triggered by the infection, was identified, and successfully treated with spectacles.

Conclusion: This case recognises isolated empyema in the sphenoid sinus as a potential cause of intermittent esotropia and limb and gait ataxia. It serves as a reminder for clinicians to be vigilant for early signs of suppurative complications to commence prompt diagnosis and treatment to prevent delayed recognition and severe complications. It advocates the inclusion of images of the sphenoid sinus to accompany the conventional scans of the cerebrum, as patients risk referral on the suspicion of intracranial pathology on the basis of neurological symptoms.

Key words: acute bacterial rhinosinusitis complications, ataxia, case report, empyema, esotropia, *Haemophilus influenzae*, sphenoid sinusitis

Introduction

Acute bacterial rhinosinusitis (ABRS) is a condition characterized by bacterial inflammation of the mucous membrane in one or more nasal sinuses preceded by local viral or allergic inflammation ⁽¹⁾. Often children are affected with ~10% experiencing at least one episode of ABRS before the age of 3 years ⁽²⁾. The diagnosis of ABRS relies on clinical criteria that conform to one of three typical patterns; persistent rhinorrhoea and cough for >10 days, severe illness with purulent rhinorrhea and >38.5°C for >3 days, or double sickening with worsening of rhinorrhea, cough,

and fever after initial improvement of symptoms ⁽²⁾.

A considerable delay in the referral, diagnosis, and treatment of patients has been described for isolated bacterial infection of the sphenoid sinus ⁽³⁻⁴⁾. Awareness of early signs of suppurative complications and correct treatment is crucial to avoid detrimental complications ⁽⁴⁾. Patients often present with non-specific headache or even remain asymptomatic until complications occur ^(3, 5), as symptoms do not conform to the typical diagnostic ABRS criteria ⁽²⁾.

Complications affect approximately 0.7% of children with any ABRS seen in emergency departments but the exact incidence is unknown ⁽¹⁾. Complications develop when the infection spreads beyond the paranasal sinuses to neighbouring anatomical regions, orbital cavity, large blood vessels i.e. foremost the internal carotid arteries, and central nervous system. It leads to neuropathy, meningitis, impaired vision, thrombosis, sepsis, and in the worst cases, death (3% mortality from intracranial complications of ABRS) ^(2, 6). Treatment primarily involves aggressive management with antibiotics and, if necessary, surgery with endoscopic drainage ^(2, 6). Additional information on initial signs of complications is therefore encouraged to diagnose the disease as early as possible.

We describe a case in which intermittent esotropia, ataxia, and headache were symptoms of isolated empyema in the left sphenoid sinus. No study has described ataxia or intermittent esotropia for isolated empyema in the sphenoid sinus. This case report aims to improve the future diagnostic approach and increase the awareness of empyema of the sphenoid sinus in a young child with months of worsening general malaise.

Case presentation

A previously healthy 7-year-old girl had over the last few months experienced gradually worsening general malaise, including headaches, fatigue, dizziness, intermittent fever, 6 kg weight loss, and several upper respiratory infections. The headache described as located behind the left eye and radiating out into both temporal regions. In this period the girl had several visits to her own GP (general practitioner), but because infection parameters were found to be normal no antibiotics were prescribed. Instead, vitamin D supplementation was prescribed due to a slight deficiency. Intermittent double vision and headache then began to develop and two weeks later the girl was referred to the pediatric emergency department. Upon arrival, the girl had a wide-based gait, ataxic finger-nose-finger and knee-heel tests, as well as intermittent esotropia (inward deviation) of the left eye, which appeared episodically when gazing straight ahead. No nystagmus or loss of lateral gaze was found. The girl was otherwise neurologically intact without visual field deficits,

good contact, and normal vital parameters without fever.

The patient was investigated with a wide panel of blood tests including tests for Epstein-Bar virus (EBV) and Cytalomegavirus (CMV) with normal findings except for slightly elevated lymphocytes $(5.0 \times 10^9/l)$ and platelets $(713 \times 10^9/l)$. Due to suspicion of intracranial pathology, an acute computerized tomography (CT) of the cerebrum was performed which showed no intracranial changes. CT was selected due to unavailability of acute magnetic resonance imaging (MRI), and because it was deemed necessary to clarify intracerebral pathology without delay. A total opacification of the left sphenoid sinus was diagnosed (Figure 1). A supplementary MRI of the cerebrum, spinal cord, and paranasal sinuses confirmed an isolated empyema in the left sphenoid sinus without signs of meningeal enhancement (Figure 2). The girl was referred to our department for treatment of suspected ABRS. An ophthalmic consultation was also ordered.

Ophthalmologic examination found intermittent left esotropia with normal visual acuity in both eyes (3/3). Subsequent refractions with cycloplegia revealed mild hyperopia of +1/+1 dioptres and the esotropia could be corrected with spectacles. At the otorhinolaryngology department, the girl subsequently underwent computer-aided endoscopic sinus surgery using a paraseptal approach performing a wide opening to the left sphenoid sinus (Figure 3A). Thick malodorous pus under pressure was drained leaving an empty sphenoid sinus (Figure 3B). Postoperatively, the girl was treated with intravenous antibiotics (cefuroxime IV 750mg + metronidazole IV 250mg \times 3 daily for one day). The day after the surgery there was significant improvement in the girl's general condition as well as improvement in intermittent esotropia and ataxia. She was discharged with oral antibiotics (amoxicillin-clavulanic acid oral suspension 500mg × 3 daily for 7 days).

Microbiological culture and sensitivity analysis was positive for *Haemophilus influenzae* (untyped) and sensitive for the chosen oral antibiotics. The girl had followed the Danish Childhood Vaccination Program which protects against type b ⁽⁷⁾. Histological material from the mucosa showed oedema and inflammation without signs of malignancy. One-month follow-up showed the girl had recovered from the symptoms, had regained her appetite, and was functioning well both at home and in school.

Discussion

This case presents a rare and prolonged infection of acute bacterial rhinosinusitis (ABRS) with *Haemophilus influenzae* in a 7-yearold girl who developed isolated empyema in the left sphenoid sinus and intermittent esotropia and ataxia with inconspicuous biochemistry. This led to diagnostic difficulties with repeated physician visits over several months before CT and MRI of the ce-



Figure 1. Axial CT image with bone window setting demonstrates a total slightly heterogenous opacification throughout the left sphenoid sinus (*). Note the intact bony margins of the sphenoid sinus without signs of dehiscence.



MRI T2-weighted axial image with gadolinium contrast with isolated affection of the left sphenoid sinus (*) with hyperintense mucosal thickening (<-) suggestive of mucosal edema along with central hypointense material suggesting inspissated secretions.

rebrum on the suspicion of intracranial pathology coincidentally revealed isolated empyema in the left sphenoid sinus.

It is widely accepted that CT, possibly with a complimentary MRI, of the cranial sinuses, orbit, and cerebrum is indicated when there is suspicion of orbital or intracranial complications to ABRS, multiple cases of ABRS, or no improvement of ABRS after appropriate antibiotics ⁽²⁾. However, because isolated sphenoid ABRS often shows inconspicuous presentation, imaging is likely delayed until the onset of complications as adjacent anatomy becomes affected (e.g. the central nervous system). Imaging may therefore be conducted on the suspicion of primary intracranial pathology, which, as evidenced by this case, risks omittance of the nasal sinuses. Our case also underlines that despite an isolated opacification in the left sphenoid sinus is found by coincidence on a CT of the cerebrum, accurate diagnosis of ABRS can remain elusive. A retrospective study found 78 % of patients with opacification of the sphenoid sinus were not referred to an otorhinolaryngologist and suggested physicians underestimate the risks of isolated sphenoid disease and calling for increased awareness (3).

In this case, accommodative esotropia was determined as the likely underlying cause of the intermittent esotropia which

became symptomatic during ABRS. Accommodative esotropia is the most common form of esotropia affecting farsighted children, where coupling between accommodation and convergence drives the eye axes inward when objects are brought into focus – and is a relatively benign condition treated by spectacle correction in childhood (to which the child in this case was referred) ⁽⁸⁾.

It is unclear why intermittent limb and gait ataxia developed, and why it presented a synchronous relationship with the intermittent esotropia. General malaise, fever, dizziness, and reduced energy consumption may also have played a role. While the empyema was isolated to the sphenoid sinus, it is well-known isolated inflammation in the sphenoid sinus can cause abducens nerve dysfunction ^(9,10), as it generates surrounding inflammatory oedema due to limited venous and lymphatic drainage ⁽⁶⁾, and the nerve's long and free course in the cavernous sinus is sensitive to this adjacent inflammation which affects nerve signalling ⁽¹⁰⁾. The inflammatory oedema was also evident in the tissue biopsies from the girl in the case. As the lateral rectus muscle loses innervation from the nerve, ipsilateral esotropia develops given the unopposed antagonistic tone of the medial rectus muscle ⁽⁶⁾. Hence, affection of the abducens nerve is conceivable, but is less probable given the esotropia



Figure 3. Intraoperative endoscopic appearance during left-sided endoscopic transnasal sphenoidotomy during (A) and after (B) the drainage. The anterior wall of the sphenoid sinus is transected (SS), and thick malodorous pus under pressure is released (*) and subsequently removed through suction and irrigation (->). The septal wall is seen on the left side of the images (S), and the lateral wall is seen on the right with a resected superior turbinate (L).

persisted and the girl was hyperopic.

To the best of our knowledge, this is the first detailed report of an isolated sphenoid empyema causing ipsilateral intermittent esotropia and limb and gait ataxia. However, this study has limitations: this case was retrospective in nature and thus no clinical photography/video of the patient is obtainable, the perioperative endoscopic images are not optimal, and a more precise timeline of initial symptoms is unavailable.

Conclusions

Bacterial infection of the sphenoid sinus may be delayed or missed due to the non-specific clinical presentation, especially among children. The presented case report recognises the disease as a potential cause of intermittent esotropia and gait and limb ataxia. It emphasizes the importance of early diagnosis and treatment of isolated empyema in the sphenoid sinus. It advocates the inclusion of images of the sphenoid sinus to accompany the conventional scans of the cerebrum, as patients can be referred based on neurological symptoms i.e., intracranial pathology.

Abbreviations

ABRS: acute bacterial rhinosinusitis; GP: general practitioner); EBV: Epstein-Bar virus; CMV: Cytalomegavirus; CT: Computerized tomography; MRI Magnetic resonance imaging. Acknowledgments None.

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Authorship contribution

AO and CB conceived and designed the case report. All authors (AO, PT, and CB) were involved in manuscript drafting, critical revision for content, and approval of the final version for submission. All authors have actively participated in the research process and have provided valuable insights and expertise in their respective areas of expertise.

Ethics approval and consent to participate

Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient of the patient. A copy of the consent form is available for review by the Editor of this journal.

Availability of data and materials

Data sharing is not applicable to this article as no datasets were generated or analysed during the current study.

Conflict of interest

The authors declare that they have no competing interests.

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