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ABSTRACT

Background: Intracranial and orbital abscesses in combination together are rare complications of sinusitis. They can be life-threatening and can result in multiple sequelae.

Case presentation: A 9-year-old female presented with left periorbital swelling, gaze restriction and headache. Following scans, she underwent emergency endoscopic sinus surgery, evacuation of the intraorbital empyema and stereotactic minicraniectomy with the evacuation of the extradural empyema as a joint case. The patient recovered well and was discharged to complete intravenous antibiotics for 6 weeks.

Conclusion: In the pediatric population intracranial complications of acute sinusitis can have more devastating consequences. Therefore, prompt recognition and management are essential within a multidisciplinary team setting.

We also highlight the rarity of concomitant multi-site abscess formation and the need to be vigilant for same.

INTRODUCTION

Acute or chronic sinusitis complications can range from minor (headaches, flu symptoms) to loss of organ function (loss of vision) or can be life threatening (brain abscess). Early diagnosis and treatment is essential to prevent sequelae. A high index of suspicion is needed when assessing and treating the paediatric population in this setting.

In the paediatric setting paranasal sinuses have not completely developed until sometimes early in the teenage years. This is particularly the case particularly with frontal sinus development in the

Keywords

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paediatric population. Indeed, in this population group, frontal sinuses are the commonest to cause significant complications, especially after the age of 10 (10,16). This is mainly due to their vicinity with the orbit and the orbital content but also with the brain and meninges.

The lamina papyracea (a thin bony plate that separates the orbital content from the nasal cavity) is very thin and often dehiscent, allowing for direct infection spread between the two anatomical regions. Orbital complications alone are more common in patients younger than 5 years of age (16). So preseptal and orbital cellulitis, followed by subperiosteal and orbital abscess are the most common complications secondary to acute or chronic sinusitis. This can lead to compromised vision. Cavernous sinus thrombosis is the most serious of all, with intracranial spread of infection and possible contralateral orbital symptoms. In the paediatric population, periorbital swelling is the most common sign that brings the patient to hospital or their family doctor's attention. This is usually associated with upper respiratory upset, fever and headache as the most common but nonspecific symptoms (16). Typical nasal symptoms seen in the adult population (nasal blockage or discharge, postnasal drip and pain over the cheeks or frontal headache) are sometimes lacking. Also a history of previous nasal complaints is absent in most cases (14).

Extension of the abscess to the intracranial cavity is usually gained through the valveless venous system, direct extension through a previous dehiscent or eroded skull base, or due to septic emboli. Subdural empyema is the most common intracranial but extra axial complication, followed by subdural and brain abscess. All are severe complications and can be life threatening or have major sequelae (hemiparesis, aphasia or epilepsy). In the paediatric population the consequences of such complications can affect patients for the rest of their life and can be debilitating and life changing for their families too.

Simultaneous intraorbital and intracranial complications secondary to acute sinusitis is very rare. This was reported as 9.3% of their patients over the age of 7 with intraorbital complications by Hermann (12).

A high index of suspicion for intracranial complications needs to be maintained and regular

assessment of the child is mandatory to check for any deterioration in their symptoms and signs or failure to improve.

Hermann et al (12) found that children with intraorbital complications of sinusitis benefit from performing a MRI brain early due to a higher association with intracranial infection in:

- children older than age 7,
- males,
- those with neurological status changes,
- frontal sinus opacification on CT,
- superior or lateral position of orbital abscess,
- intraorbital abscess needing drainage.

Sinus infection may progress to intracranial infection even after appropriate empiric antibiotic therapy has been initiated (14,17,19). Lack of early recognition and urgent treatment can be fatal. Empirical antibiotic treatment is generally broad-spectrum and started under local microbiological guidelines and supervision.

Timing of surgical intervention is a controversial subject, with multiple studies being in favour of early rather than late surgical intervention (14,17).

Intraorbital abscess formation needs to be drained in conjunction with an otorhinolaryngologist (ORL) performing endoscopic sinus surgery and intracranial abscess formation needs prompt neurosurgical input and craniotomy with drainage of the infection.

CASE PRESENTATION

A 9-year-old female patient presented to the emergency department with a 2 week history of flu symptoms and headaches. She complained of right upper eyelid oedema prior to presentation that subsided followed by persistent left periorbital oedema and ophthalmoplegia. Prior to her hospital presentation, our patient has been already on 5 days of oral antibiotics started by her family.

Her medical history was non-contributory with no history of nasal symptoms or allergic rhinitis, nor any history of atopy. The child lived with her family in America and was on holiday visiting her relatives.

An initial assessment showed pain on eye movement with no focal neurological signs with a Glasgow coma scale (GCS) of 15 out of 15. The patient had neither neck stiffness nor photophobia. She was afebrile with stable vital signs. Intravenous access

was obtained and routine bloods were sent to the laboratory.

She was admitted with a diagnosis of preseptal cellulitis for intravenous antibiotics as per our local guidelines.

On admission her inflammatory markers were elevated. White cell count was 18.2 [normal 6.0-18.0($\times 10^9/l$)], with neutrophilia of 13.9 [normal 1.0-8.5($\times 10^9/l$)]. Her C-reactive protein (CRP) was 162 (normal 0-10mg/l).

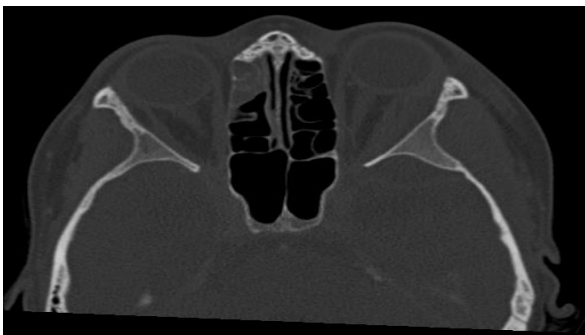
Despite 24 hours of intravenous Cefotaxime, she developed restriction of eye movement on elevation and downward gaze combined with drowsiness. Otorhinolaryngology was consulted and flexible nasal endoscopy did not reveal any intranasal pathology. An urgent ophthalmology consult was requested and vision was found normal in both eyes.

Our patient underwent urgent computer tomography (CT) of sinuses without contrast. The scan showed pansinusitis on the right side (maxillary, ethmoid and frontal sinuses) with left sided sinuses free of mucosal disease. Also she had a small left extraconal fluid collection (measuring 7x 15 x 25mm).

Pictures 1 and 2 shows the above findings on a coronal and axial CT cut:



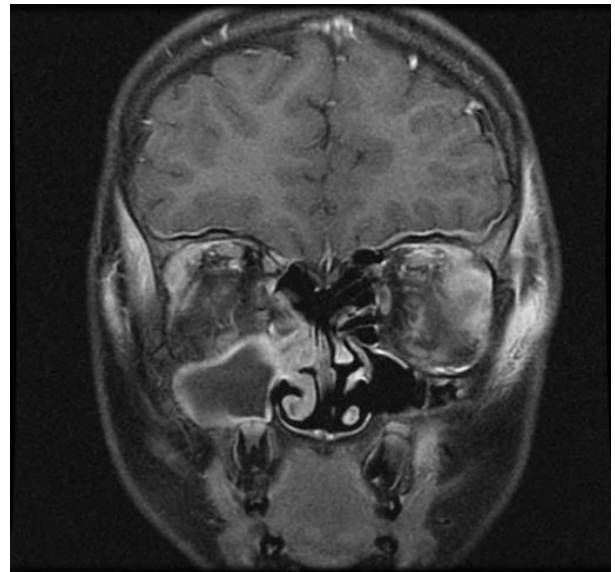
Picture 1. Coronal CT sinuses.



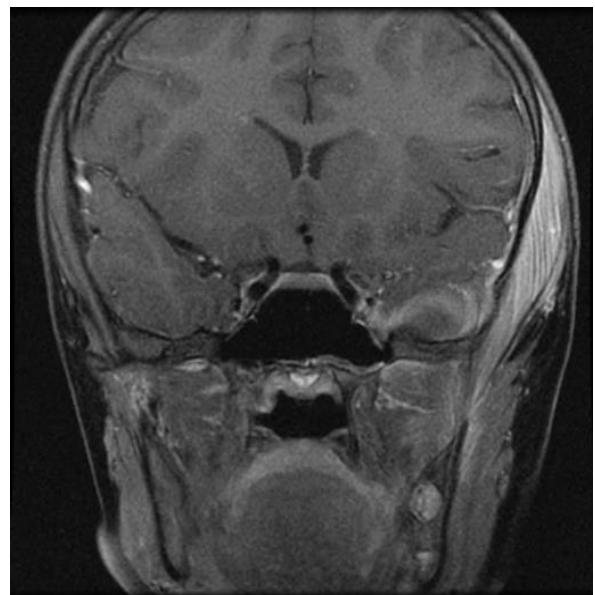
Picture 2. Axial CT sinuses.

Due to the high clinical suspicion of intracranial pathology and increased drowsiness an urgent magnetic resonance imaging (MRI) of brain with contrast was performed. This revealed the left orbital extraconal abscess with extension through the posterolateral orbital wall into the extradural space anterior to the left temporal lobe. The bone demonstrated osteomyelitis. There was also a small extracranial component adjacent to the pterion, with inflammatory changes in the left temporalis muscle.

All these findings are visible in the Pictures 3 and 4:



Picture 3. Coronal MRI showing left external orbital abscess.



Picture 4. Coronal MRI with contrast showing left extradural empyema.

After an urgent neurosurgical consult emergency surgery was planned as soon as possible. Endoscopic sinus surgery followed by left orbit abscess drainage and stereotactic mini-craniectomy with empyema evacuation was performed.

After the patient was prepped as per endoscopic sinus surgery protocol a left maxillary antrostomy was performed where frank pus was found. Similar findings were found in the left anterior ethmoidal sinus. On exploring the left posterior ethmoid only mucosa disease was found. Sinus surgery found normal sinuses on the right side despite the abnormal findings (pansinusitis) on the CT on this side.

The left external orbital abscess was drained via an external approach and a drain was left in situ.

A stereotactic mini-craniectomy was performed via a left curved pterional incision, followed by evacuation of the empyema and again a drain was placed. Pus for culture and sensitivity was sent from all 3 locations.

The patient tolerated the procedure well and no perioperative complications were encountered. No intensive care unit monitoring was needed postoperatively for our patient. She was commenced on intravenous Ceftriaxone and Metronidazole.

The approach to this case was a multidisciplinary one with Paediatricians, ORL, Neurosurgery, Ophthalmology, Infection Disease, microbiology and physiotherapy involved in this child's care.

Our patient recovered well from a neurosurgical and ORL point of view but was quite slow from an eye perspective. The intraorbital drain was removed after 12 hours and the intracranial drain after 24 hours. No temperature spikes were recorded postoperatively.

A repeated MRI brain was performed on day 2 postoperatively due to slow intraorbital progress and this revealed a small collection at the site of previous orbital abscess. Close ophthalmology review followed until resolution of the collection, 2 and half weeks after the surgery.

Culture from all sites grew *Streptococcus intermedius*. Inflammatory markers returned to normal values. A repeat MRI scan at 18 days postoperatively showed near complete resolution of the remaining collection.

The patient was hospitalized for 4 weeks duration and further intravenous antibiotics were arranged for 6 more weeks at home. During this period, our

patient was reviewed in multiple speciality clinics. She improved totally, with no neurological sequelae. A repeat MRI scan at 2 months postoperatively showed complete resolution of the collection and osteomyelitis.

Due to the fact that our patient lived in another country, further follow up was not possible but all images, documentation and advice on follow up were given to organise ORL follow up in her home country.

DISCUSSION

Simultaneous intraorbital and intracranial complications secondary to sinus disease are extremely rare. The incidence is unknown with only few cases in the literature (8,9,12,18,22) and a handful of case presentations from paediatric population (4,25).

Yeh presented the case of 17 years old male with longstanding orbital symptoms (1 month) treated with sinus surgery who developed 1 month after the treatment an inflammatory frontal mass needed excision with no neurological sequels (25).

Constantin et al (4) reported the case of a 12 years old male patient with sinusitis complicated later by orbital cellulitis ending developing frontal brain abscess. Sinus surgery and adenoidectomy was followed at 24 hours by neurosurgical intervention with collection excision.

Four retrospective studies presented the presented symptoms and management of their patients. Hermann et al (12) found a rate of 10% (4 patients) in his study of paediatric patients with simultaneous intraorbital and intracranial complications. All were males and the youngest was 14 years of age. They were treated surgically and all of the abscesses were found to be polymicrobial. He found that MRI helped identifying the intracranial complications after they were missed on CT. As a significant finding, in this study all patients had either a superior or lateral intraorbital located complication.

In his study Germiller et al (8) presented 7 cases of concomitant orbital and intracranial complications treated surgically with a focus on intracranial complications and outcome.

In her retrospective review of 10 patients younger than 18 years, Reynolds et al (18) found the youngest to be 7 years old and predominantly males. Also she

found that frontal lobe was involved in 8 out of 10 patients.

Goyita et al (9) found 14 paediatric patients to have intraorbital and intracranial complications, with a predominant male distribution in this group.

Our patient (age 9) was amongst the youngest ones from the previously reported cases.

An age greater than 7 was found to be associated with the development of sinogenic intracranial complication rather than intraorbital ones alone (5,14,17) but also with dual orbital and intracranial abscesses (11).

Our patient was a young female, although male adolescents were found to be more prone to intracranial complications alone and in association with intraorbital ones for reasons not yet elucidated (8,9,13,17,19,25). If there is a hormonal rather than an anatomical component it is to be discovered.

CRP, an inflammatory marker was found to be higher in children with brain empyema complicating rhinosinusitis than in uncomplicated rhinosinusitis, with a mean of 18.1 mg/dl (17).

While CT scan of the paranasal sinuses is the gold standard imaging modality in acute or chronic rhinosinusitis, used by surgeons worldwide to assess sinus disease, MRI brain with contrast remains the gold standard in identification of brain abscesses (9,12,21). This should be requested for patients with increased risk factors of intracranial disease or if any suspicion of intracranial complication exists. It should be performed early rather than later in the course of the disease.

Our patient's CT sinuses failed to show the true extent of the sinus disease. The scan revealed right sided pansinusitis and during endoscopic sinus surgery all these sinuses were free of disease. The left maxillary sinus seemed free of disease on the scan but frank pus was seen during surgery. Why there was such a non concordant finding it is not clear. The subsequent MRI brain showed clearly the presence and extent of the intracranial collection.

As demonstrated by multiple studies, CT sinuses findings are not absolutely accurate when compared with the endoscopic sinus surgery findings (2,20).

Another unique aspect of the case was the bacteria grown which was *Streptococcus intermedius*. The most common bacteria in uncomplicated sinusitis in children is *Haemophilus Influenzae* followed by *Streptococcus pneumoniae*, *Staphylococcus aureus*, *Staphylococcus epidermidis*,

and *Streptococcus pyogenes* (13,23). Mixed aerobic and anaerobic cultures are commonly seen also (12,23).

Streptococcus intermedius is a subspecies of *Streptococcus milleri*. It is highly pathogenic via multiple mechanisms. It is associated with deep, complicated abscesses including cerebral abscess as a complication of sinusitis (5,6,23). This could explain the aggressive course of this case.

Prompt recognition and aggressive medical and surgical treatment was the key to a complication free and smooth recovery. The role of early multidisciplinary surgical intervention was most useful in this case with all 3 specialities involved. Usually a third-generation cephalosporin plus metronidazole is most effective in fighting these types of complicated infections (7,24), but needs to be guided by local microbiology guidelines.

The role of surgery, although vital in some cases, still needs to be explored in finding the best technique and timing. The evidence in the literature is mainly based on retrospective reviews and case series. There is a lack of significant evidence in favour of one surgical treatment modality over the other (burr hole drainage, excision or stereotactic surgery) when analyzing their outcomes (1,15). The role and timing of endoscopic sinus surgery needs to be explored more, on larger samples to assess the full benefit these patients can gain. There seems to be some benefit in avoiding neurosurgical intervention if sinus surgery is performed first (3) but this was not reflected in all studies. We advocate that the focus of infection needs to be addressed first and at the same time as sinusitis being the „driver“ of the multicompartmental infection. While very small intracranial infections can be managed conservatively, any large intracranial collection needs to be evacuated or reduced due to the risk of spread, thrombosis and poor blood-brain barrier penetration of a lot of antibiotics.

Each case has to be individually evaluated and judged as no one procedure fits all.

Multidisciplinary input is common in paediatric practice but proved essential in this patient's care.

CONFLICTS OF INTEREST

Authors have no financial or other conflicts of interest to declare.

Authors have not received any financial sources of any kind and form.

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