Case Report

Parafalcine subdural empyema as an uncommon complication of acute odontogenic sinusitis: a case report and literature review

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Keywords

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Abstract

Parafalcine subdural empyema is a relatively uncommon but important suppurative infection of the central nervous system. We present a case of a 15-year old boy who developed headache with an altered level of consciousness, confusion, a right hemiparesis and fever three weeks after tooth extraction. Magnetic resonance imaging confirmed the diagnosis of a parafalcine subdural empyema on the left side. Intravenous ceftriaxone, metronidazole and acyclovir were administered, and emergency trepanation and functional endoscopic sinus surgery were performed. Dexamethasone sodium phosphate was given in the context of cerebral edema and an anticonvulsant was started preemptively. Culture from the empyema showed the presence of multisensitive Streptococcus anginosus. The hemiparesis improved gradually and 28 days after admission the boy was discharged. Neurological examination at discharge was normal. Parafalcine subdural empyema is an uncommon complication after tooth extraction and diagnosis can be challenging. An early multidisciplinary approach is important to prevent complications. Neurologic sequelae are common. Morbidity rate is 33% after six months and mortality rate is 5-10%.

Introduction

Intracranial subdural empyema (SE) is a suppurative infection localized between the dura and arachnoid matter (1). Most of the SE's are localized within the frontal lobe or convexity (2). They largely arise from direct extension of adjacent infection, hematogenous seeding, or trauma (3). Suppurative otitis media is regarded as the commonest cause of intracranial suppuration, with an incidence of approximately 39% (4). Bacterial paranasal sinusitis is the underlying cause in 3.7% to 11% (2, 5, 6). A parafalcine localization is only described in less than 20% of SE's and has a worse prognosis. Most patients present with non-specific symptoms such as (severe) headache, fever and vomiting (7). However, symptoms can be more subtle in case of a preexisting sinusitis, which makes the diagnosis of a SE challenging (8, 9). Neurological symptoms often consists of alteration of consciousness, focal deficit, hemiparesis, nuchal rigidity and seizures (1). It is important to diagnose and treat SE's promptly because life threatening complications, such as cerebral thrombophlebitis, cerebral edema and cerebral infarction, can cause increased intracranial pressure leading to coma and death within one to two days (8, 10). Parafalcine SE's, particularly, are associated with a worse prognosis (10). We present the case of a 15-year old boy with parafalcine empyema as result of an acute odontogenic sinusitis caused by tooth extraction.

Case presentation

A fifteen-year old Caucasian boy presented with recurrent episodes of fever in the last three weeks. The fever started five days after extraction of his dental elements 18, 28, 38 and 48. He started vomiting since a few days and had anorexia. One day before admission, he had headache, an altered level of consciousness with confusion and a right hemiparesis. Clinical examination revealed a pale apathic boy with a high fever and cognitive impairment. Cardiorespiratory parameters were normal. He had nuchal rigidity, photophobia and a right hemiparesis most pronounced at his right foot. Blood examination showed a leukocytosis of 24 800/µl (normal range 4000/µl-13000/µl) with neutrophilia and a C-reactive protein (CRP) of 86.5 mg/l (normal < 10 mg/l). A computed tomography (CT) of the cerebrum without contrast revealed a hypodense parafalcine structure, suspicious for empyema. Cerebrospinal fluid contained 49 leukocytes/field, mainly polymorphonuclear cells, an increased protein level of 73 mg/dl (normal < 45 mg/dl) and a normal glucose level of 78 mg/dl (with a serum glucose of 142 mg/dl). Intravenous ceftriaxone (100 mg/kg/day, max: 4 g/day), metronidazole (30 mg/kg/day) and acyclovir (1g/m2/day) were administered. A magnetic resonance imaging scan (MRI) confirmed the diagnosis of a parafalcine SE on the left side. (figure 1). There were signs of underlying cerebritis and cortical edema. The parafalcine SE was presumably caused by a breakthrough sinusitis of odontogenic nature.

Emergency trepanation and functional endoscopic sinus surgery (FESS) were performed to evacuate the SE. Post-operative neurologic controls were stable and anticonvulsive therapy with levetiracetam (20 mg/kg/day) was started preemptively. Dexamethasone sodium phosphate (0.25 mg/kg/day) was given in the context of the cerebral edema. Cultures from the empyema were positive for multisensitive *Streptococcus anginosus*. As blood cultures and culture from the cerebrospinal fluid remained sterile with negative PCR for the herpes simplex virus, acyclovir and metronidazole were ceased. Post-surgery MRI showed residual empyema, parafalcine, posterior, parietal to occipital with signs of frontal, parietal and occipital meningitis on the left side. The MRI also showed a small zone suspicious for hemorrhagic cerebritis and two small left frontal infarcts. Nevertheless, there was a favorable biochemical and clinical evolution with slow improvement of the right hemiparesis. On postoperative day eleven continuous headache reappeared. Blood examination showed a leukocytosis of 18 800/µl with neutrophilia and a CRP of 13,8 mg/L. MRI showed an overall decrease of the volume of the empyema but an increased lateral diameter of the dorsal component of the collection (Figure 2). The SE was rinsed through a left occipital trepanation on day fourteen. Sinus endoscopy showed clear sinuses. Peroperative cultures remained sterile. At day 21, the MRI showed no residual collection (figure 2).

Further immunological work-up was performed because of this impressive clinical picture. There were no severe infections in the boy's history. Familial history was negative. A previous immunological work-up showed a low T CD3+/HLA-DR+,

Figure 1 : *3T-MRI of the brain on admission. Axial T1-weighted images after Gadolinium administration show focal leptomeningeal contrast enhancement in the left frontal lobe (red circle in A), a finding compatible with meningitis. A left paramedian subdural fluid collection is seen along the falx cerebri (blue arrows in B) with increased signal on b1000-diffusion weighted imaging (C) and low signal on the ADC-map (images not shown), compatible with a subdural empyema.*

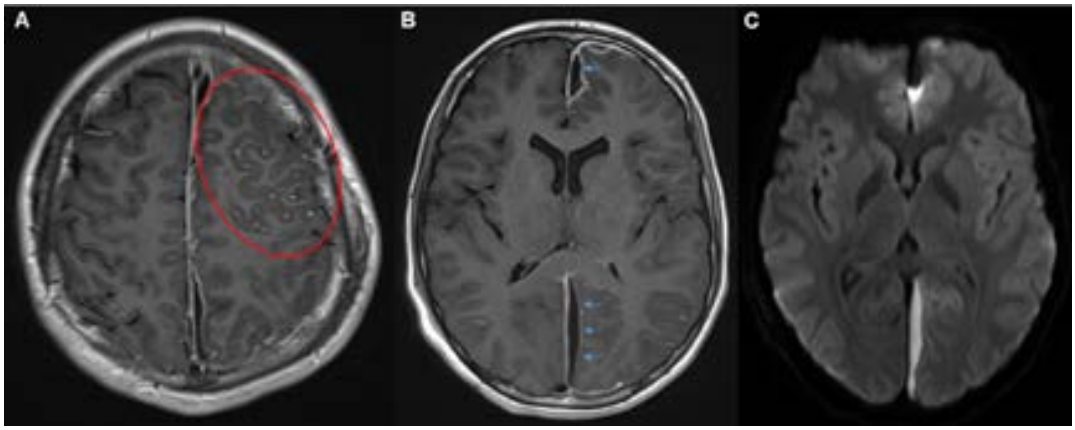
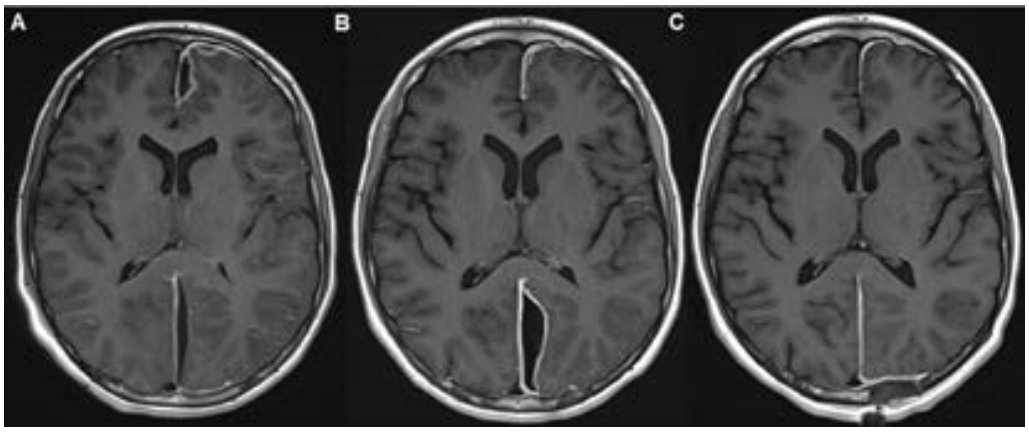


Figure 2 : *Evolution of the subdural empyema. Axial T1-weighted images after Gadolinium administration on admission (A), day 13 (B) and day 21 (C). Between the scan on admission (A) and day 13 (B) a clear volume-increase of the dorsal component of the subdural empyema can be seen. Control MRI on day 21 shows complete regression of the subdural empyema with some residual most likely reactive left parafalcine dural enhancement (C).*



T CD3+/CD8, B CD19 and lowered IgG and IgM at the age of two years. These values normalized by the age of three. Immunological testing after resolution of the disease showed a lower IgG of 5.4 g/l (normal > 7.12 g/l) with normal IgG2 and IgG3, normal IgA, IgM, total hemolytic complement (CH50) and complement fractions (C3 and C4). A mannose-binding lectin deficiency was detected. Pneumococcal antibody response after vaccination was normal.

The hemiparesis regressed progressively and 28 days after admission the boy was discharged. The neurological examination at discharge was normal. Levetiracetam and intravenous ceftriaxone therapy were continued during twelve weeks. MRI, six weeks after discharge, showed no residual collection and ceftriaxone was ceased. Electroencephalogram (EEG) was normal. Levetiracetam was continued for six months.

Discussion

We found only five articles concerning 82 children with parafalcine SE (10-14). In our case the parafalcine empyema was a complication of an acute odontogenic sinusitis caused by tooth extraction. In 3.7 % to 11% the cause of a SE is a sinusitis. Other known causes of SE include neurosurgical infection, head trauma, otitis media, osteomyelitis of the skull and meningitis (2). An important origin of sinusitis, especially unilateral maxillary sinus infection, is odontogenic. This is because the roots of the premolars and molars are situated just below the floor of the maxillary sinus (5). Evolution towards pansinusitis is possible through the osteomeatal complex. An oro-antral communication between the maxillary sinus and the oral cavity can be created during tooth extraction. This allows the passage of anaerobic bacteria into the sinus (15, 16). There are two major mechanism for the formation of SE from sinusitis. The most common mechanism is retrograde thrombophlebitis through the valveless diploe veins which causes translocation of bacterial seeds to the subdural space. Between the age of 7 and 20 years there is a peak in the vascularity of this diploe system and the development of the sinuses, especially the frontal sinus. This explains the peak prevalence of SE in the second decade of life (5, 17). The second mechanism is a direct breakthrough trough the facial bones with osteomyelitis and subsequent erosion into the epidural space (2, 8).

Patients with a SE mostly present with non-specific symptoms such as headache, recurrence of fever or vomiting (7). Neurological symptoms often consist of altered sensorium, focal deficit, hemiparesis, nuchal rigidity, photophobia and seizures (1). Seizures arise in 25-80% and are more common in SE than in any other intracranial complication of paranasal sinusitis (1, 2, 7, 8, 13, 17). However, when a sinusitis precedes the SE, it usually presents as an insidious process with purulent nasal discharge, headache and fever, as seen in our patient (8, 9). Once the infection reaches the intracranial space, neurologic deterioration follows rapidly because of the lack of anatomical constraints. Acute or progressive headache is the most important indicator of intracranial involvement (18). In patients with a parafalcine SE a typical presentation is the 'Falx syndrome' where the contralateral lower extremity is most affected.

The gold standard for the diagnosis of SE is MRI with gadolinium enhancement (9). However, CT scans are often the first choice of imaging modality because they are more available in the emergency setting (19). Additionally, CT is the best modality to visualize the paranasal sinuses and associated bony abnormalities. However, CT may fail to visualize intracranial complications (9). MRI with gadolinium and contrast enhanced CT are complementary examinations when evaluating complications of sinusitis (9, 19, 20).

Microbiological cultures of sinogenic empyema reveal in 67% bacteria belonging to the *Streptococcus milleri* group. *Streptococcus anginosus*, as in our patient, belongs to this group and accounts for 11% of intracranial abscesses (13, 17, 19). Anaerobic streptococci are known to cause invasive suppurative infections in different tissues. *Streptococcus anginosus* is the most frequently involved bacterium in intracranial complications of sinusitis and is related to a higher probability of neurosurgical intervention and long-term neurologic deficits (8, 19). Laboratory data are non-specific: white blood cell count, CRP and erythrocyte sedimentation rate may be elevated. Lumbar puncture is not indicated because of its poor diagnostic yield, technical difficulty and the risk of transtentorial herniation, neurologic deterioration and death. However, when a lumbar puncture is performed, the cerebrospinal fluid shows pleocytosis with polymorphonuclear predominance, elevated protein and normal glucose level. Cerebrospinal fluid cultures remain negative in more than 85% of the cases (2, 9, 17). Research indicates a possible association between mannose-binding lectin deficiency and increased susceptibility to recurrent and/or severe infections (21, 22).

The management and treatment of intracranial SE necessitates a multidisciplinary approach (23). Broad-spectrum intravenous antibiotic therapy consisting of a third-generation cephalosporine in combination with metronidazole should be initiated a soon as possible. The incidence of intracranial sinogenic complications has decreased dramatically since the advent of broad-spectrum antibiotics and improvement in diagnostic imaging. Antibiotic treatment however does not absolutely prevent intracranial suppurations and can sometimes delay diagnosis and treatment (2, 8, 17). Once the results of microbiological cultures are known, the antibiotic therapy can be adjusted. Intravenous antibiotic therapy should be continued for at least two weeks followed by up to six weeks of oral antibiotics. If osteomyelitis is present the therapy should be prolonged to eight weeks (8). However, the duration of the intravenous antibiotic therapy should be adjusted to the clinical circumstances and evaluated on a case-by-case basis. Adjuvant therapy should consist of preemptive anticonvulsants and corticosteroids to reduce the cerebral edema (2). Preemptive antiepileptic drugs are often administered due to the high incidence of seizures both before and after treatment. In Cowie et al, a case series of 89 patients, of those who had no early seizures, 42% had late seizures, the majority appearing within 16 months (24). The use and dosage of corticosteroids in brain abscess is still being debated. Dexamethasone is the corticosteroid of choice when treating brain abscess patients for associated vasogenic edema (25). Despite the anti- inflammatory and possible immunosuppressive properties of dexamethasone, the results of Simjian *et al.* suggested that there was no mortality benefit obtained from withholding dexamethasone (26). Because of the rapid progression, SE nearly always requires surgical drainage with a combined neurosurgical and rhinological approach (8, 9, 27). The most effective procedure is a direct and large drainage through craniotomy. An alternative approach through burr holes and saline irrigation is less effective and will often require additional surgery or conversion to craniotomy (19). Surgical intervention is the most important factor in the prognosis and subsequently a low threshold should be maintained (23). The goal is to evacuate the intracranial pus collection which improves the clinical condition, provides microbiological samples and allows managing of the source of infection. Drainage of the infected sinuses trough endoscopic sinus surgery should be done at the same time. Other approaches include maxillary irrigation, external fronto-ethmoidectomy, sphenoid sinusotomy, antral washout and frontal trephine (2). Because of the tricky localization of the pus in a narrow space, parafalcine empyema's represent a challenge to neurosurgeons. It is often necessary to redo the surgical procedure (8,15). Patel et al. reported that about 22% of children needed early revision surgery (17). Follow up MRI scans are therefore indicated four to seven days post-surgery (10, 13).

Neurologic sequelae after resolution are quite common, morbidity rate is estimated at 33% after six months and mortality rates are between 5-10% (28). Headache and limb weakness are frequently seen as short-term morbidities. On the long-term cognitive deficits, expressive aphasia and epilepsy have been reported (2, 8, 17, 23). Factors defining outcome include the level of consciousness and neurologic status at admission, time to diagnosis and treatment, a parafalcine localization, extent of the collection and underlying immunodeficiency (9).

Conclusion

Parafalcine SE secondary to tooth extraction is uncommon in children and diagnosis can be challenging as symptoms can be non-specific. MRI with gadolinium enhancement is the gold standard for the diagnosis. Early multidisciplinary approach is important to prevent complications. Management consists of neurosurgical interventions, intravenous broad-spectrum antibiotics and supportive care. Morbidity and mortality are high in SE with important neurodevelopmental consequences on the long-term. Early diagnosis and prompt treatment are key factors determining the outcome. Therefore, a good education of pediatricians and family physicians in combination with a high index of suspicion are of preeminent importance.

Declarations

Consent for publication: Informed consent was obtained from all individual participants included in the study.

Competing interests: The authors declare that they have no competing interests.

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