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Cavernous sinus thrombosis during pregnancy

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ABSTRACT

Cavernous sinus thrombosis (CST) represents a rare but devastating disease process that may be associated with significant long-term patient morbidity or mortality. Rapid diagnosis and aggressive medical and surgical management are imperative for patients with CST. We present the case of a 24-year-old pregnant woman with intraorbital abscess and CST secondary to *Streptococcus milleri*. Surgical intervention included orbital abscess drainage and dental extraction, medical therapy included intravenous antibiotic, heparin, and methylprednisolone and an elective cesarean section was performed. The latter was the key point to resolution the disease.

1. Introduction

Cavernous sinus thrombosis (CST) is a rare complication of orbital cellulitis and sinusitis that may be associated with significant long-term patient morbidity or mortality [1]. *Streptococcus milleri* is an anaerobic organism capable of causing significant morbidity. To our knowledge, this is the first case reported of a pregnant woman with orbital abscess and bilateral CST. This paper provides an insight into the current recommendations for the management of CST and the importance of early diagnosis and treatment of this condition.

2. Case report

A 24-year-old and 32 weeks pregnant woman presented to our institution in May 2012 with a history of severe right fronto-orbital headache, photophobia and vomiting. She reported no other neurological or otorhinolaryngologic symptoms and was otherwise healthy. The patient had no history of recent infections, new medications, or other systemic complaints. Physical examination was unremarkable. The blood tests showed no signs of infection. An emergency computed tomography (CT) scan was performed and showed no pathological findings (Fig. 1). The patient was discharged with analgesics.

Two days later she returned with similar complaints. The headache had worsened and associated neck pain and neck stiffness. A complete blood count revealed a white blood cell (WBC) count of 17,200/μL with 92% neutrophils, a erythrocyte sedimentation rate (ESR) of 90 mm/h, C-reactive protein (CRP) of 25 mg/dL and procalcitonin (PCT) of 8 ng/mL (Fig. 2). A lumbar puncture was performed because of suspected meningitis. Cytology, biochemical and microbiological analysis on cerebrospinal fluid were normal. The patient was admitted at the Neurology Department for monitoring, pain management and penicillin-based empirical antibiotic therapy.

During the admission, the patient worsened progressively; the headache got worse and severe right orbital pain, progressive right ophthalmoplegia and right mydriasis were objectified. Finally, the patient developed sign of sepsis, including tachycardia (pulse 95 beats per minute), tachypnea (respiratory rate 22 min^{-1}), hyperthermia 39 °C, hypotension

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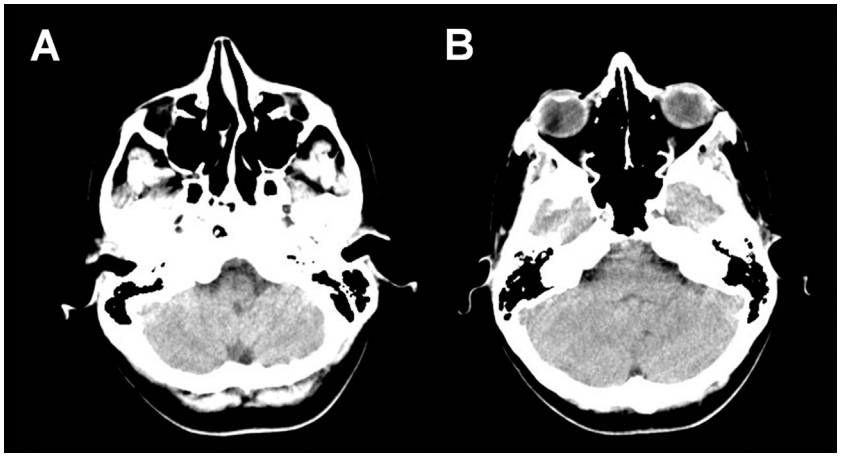

Fig. 1. Axial CT scan imaging shows absence of sinonasal and orbital pathology.

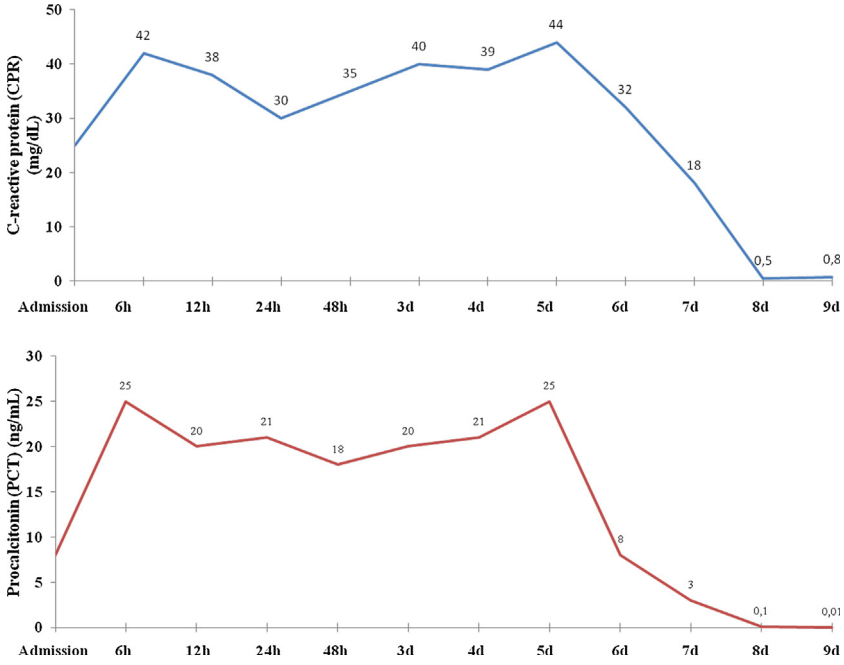

Fig. 2. Time course of serum levels of procalcitonin and C-reactive protein.

(80/62 mmHg) an impaired level of consciousness. A new complete blood count showed increased leukocytosis, CRP, PCT and fibrinogen (Fig. 2).

Because the gravity of the situation, 6 h after the admission, an emergency magnetic resonance imaging (MRI) was carried out and a diagnosis of bilateral CST and right orbital abscess was made (stage V, according to Chandler's Classification System) (Fig. 3) [1]. At no time during the course, signs of fetal distress were found.

Although the patient had reported having no history of infections, her family referred that she had noticed the appearance of the swelling, a few days before, on the right zigomatic region, probably caused by a dental infection, which was confirmed by the patient afterwards. Right maxillary molar extraction was done by the maxillofacial surgeon revealing a periapical abscess. However, there was no evidence suggestive of tooth abscess over right upper alveolar region in the MRI.

Given the findings of the MRI, we performed an emergent orbital decompression using a right lateral canthotomy and cantholysis approach (Fig. 4). Purulent material was obtained and sent for microbiological analysis. According to obstetricians, the patient was admitted to the intensive care unit for monitoring and she was treated by tracheal intubation, broad spectrum antibiotics (meropenem 2 g iv every 8 h), corticoids (dexametasone 8 mg iv every 8 h)) and low-molecular-weight heparin at therapeutic doses (enoxaparin, 100 IU/kg twice daily). It was isolated *S. millieri - group F* sensitive to beta-lactam antibiotics, in the purulent material.

Because the clinical situation (hypertermia, hypotension and hemodynamic instability), and laboratory parameters (WBC, CPR, PCT) did not improve and, due to high risk of severe fetal-maternal complications, 5 days after surgery, an elective cesarean section was performed at 34 weeks, uneventfully.

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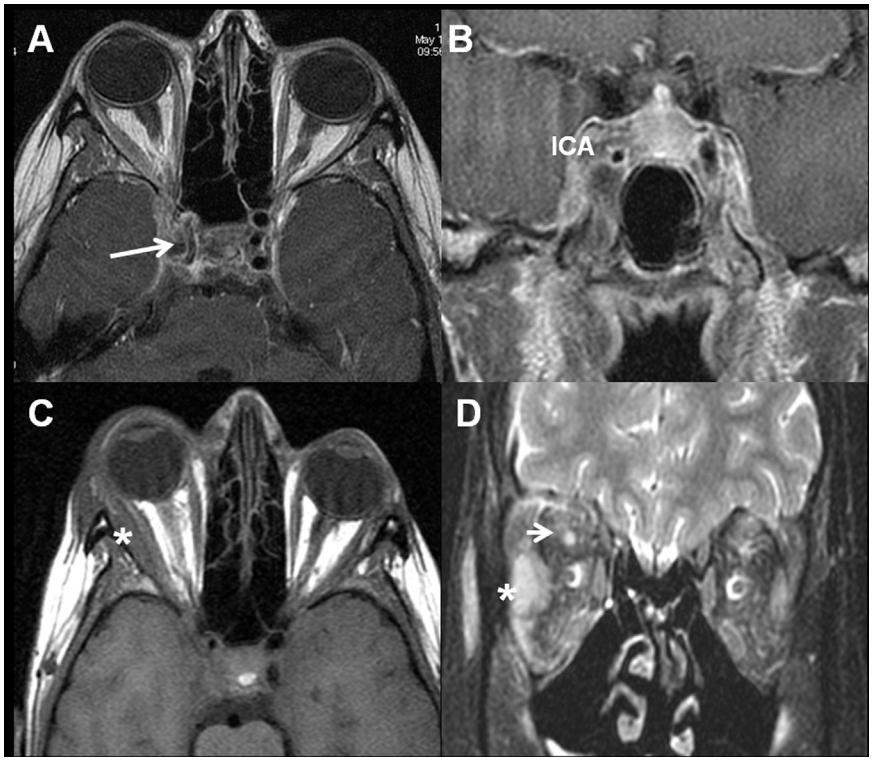

Fig. 3. (A) An axial 3-mm T1-weighted MRI scan reveals a bilateral cavernous sinus-filling defect, mainly in the right side *(arrow)*. There is thick dural enhancement bilaterally around the cavernous sinus regions. (B) A coronal high resolution T1-weighted magnetic MRI scan depicts cavernous sinus enlargement with small filing defects. An small abscess located in the right cavernous sinus is seen, around the internal carotid artery (ICA) which shows a decreased size. (C) An axial 3-mm T1-weighted enhanced MRI scan reveals the enlarging abscess in the right lateral canthal region (*). Right proptosis is seen. (D) A coronal high resolution T2-weighted MRI scan shows the abscess in the right lateral canthal region (*). Right superior ophthalmic vein is thrombosed *(arrow head)*.

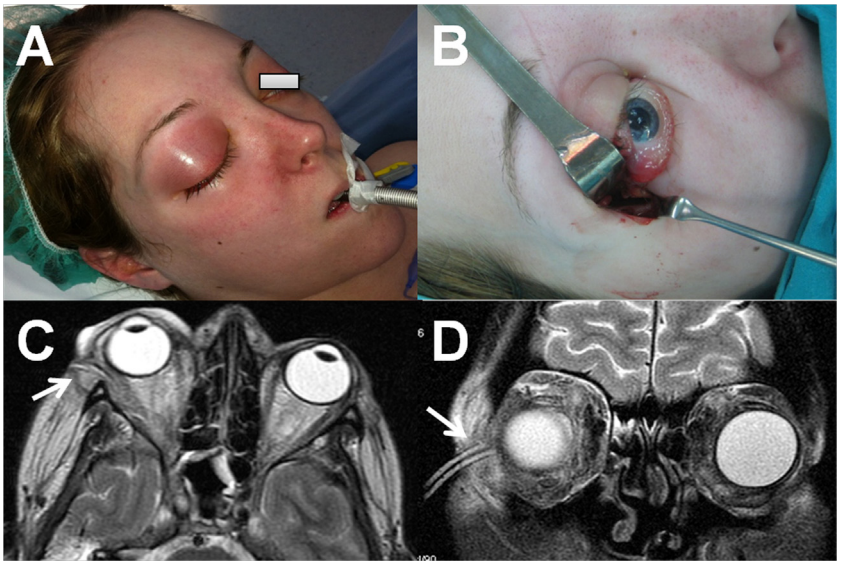

Fig. 4. (A) External view of patient. (B) Al right intraorbital abscess drained by canthotomy and cantholysis. An axial (C) and coronal (D) high resolution T2-weighted MRI scans show the drainage of the abscess *(arrows)*. There is no disease in the maxillary and ethmoid sinuses.

Since that time, she improved clinically and radiologically (Fig. 5), and she was admitted to the hospital ward to complete antibiotic treatment for 3 weeks. Tests for prothrombotic disorders did not revealed abnormalities. Anticoagulation with coumadin was continued on an outpatient basis. Currently, 3 years after the event, a right amaurosis is the only sequel, and the baby is healthy.

3. Discussion

CST is associated with significant morbidity, commonly reported as more than 50%, and has a mortality rate of up to 20–50% [2]. There are several situations that can favor the development of this condition, such as immunosuppression and hypercoagulability. Both conditions are present in our case

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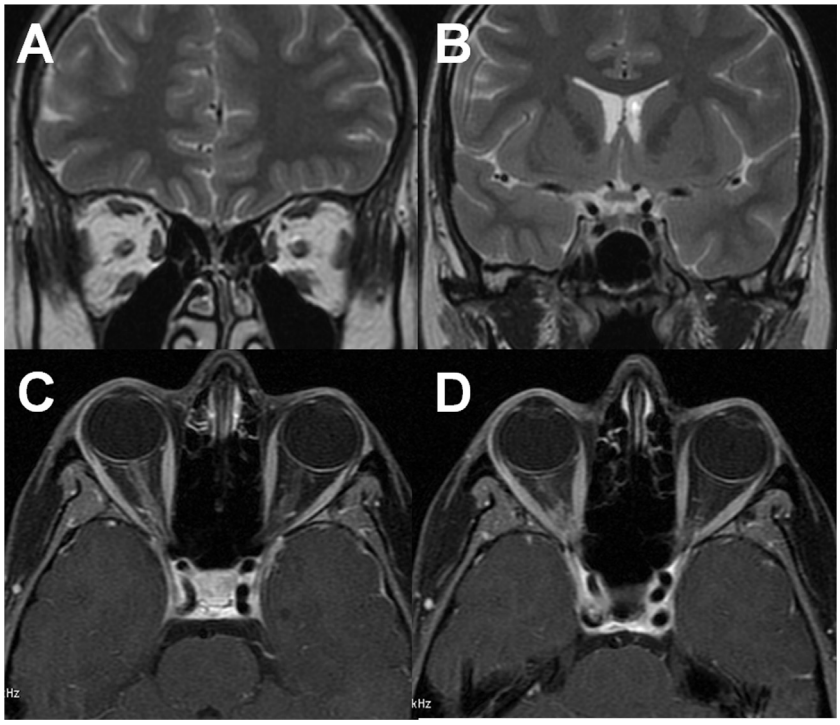

Fig. 5. Coronal high resolution T2-weighted MRI (A, B) and axial T1-weighted MRI (C, D) show the resolution of inflammation and the absence of abscess.

because pregnancy is characterized by a physiological immunosuppression and increases the risk of thrombosis 3-fold to 4-fold [3]. The causes of CST can be aseptic or infectious. Infections causes include upper respiratory tract infections, otitis and odontogenic and facial sources. As in our case, odontogenic origin have been reported to be responsible for up to 10% of cases [4]. CST may occurs secondary to the spread of infection by veins and by direct extension. Depending on the source of infection, causative organisms of CST may vary. In cases of sinusitis *Streptococcus* sp., *S. aureus*, gram-negative organism and anaerobes can be involved. Odontogenic infections are usually mixed flora, including *Streptococcus* sp. and anaerobes. *S. milleri*, included in the *Streptococcus viridans* group, is an infrequent pathogen in the head and neck. It can be highly virulent when it reaches pathogenic proportions, causing a rapid spread of the disease [5,6]. The dismal prognosis is linked to the severity of illness and the high level of virulence of this organism [5]. Outcome also depends on the time expended in making the correct diagnosis and beginning appropriate treatment [7]. The literature includes few references to this organism as a cause of orbital infection and only 3 cases of CST have been published so far [8–10]. The clinical course of CST include ''picket fence'' fever, headache, nuchal rigidity, signs of sepsis and, eventually coma, as was noted in our case. Eye signs secondary to orbital venous congestion and paralysis of the cranial nerves were also observed in our patient. CST diagnosis by MRI and CT is crucial in the evaluation of these patients. High-resolution contrast-enhanced CT is important in the diagnosis and classification of orbital infection. CT has been favored as the primary diagnostic test for CST, depicting filling defects and enlargement of CS. Direct signs of CST include expansion of cavernous sinus, convexity of the normally concave lateral wall, abnormal irregular filling defects and asymmetry. Indirect signs may be the dilatation of the superior ophtalmic vein, exophtalmos and thrombi in the veins and cavernous sinus tributaries [4,11]. MRI can detect all stages of thrombus formation. In the early stages, on T1-weighted images thrombus is isointense and on T2-weighted images is hypointense; later, thrombus becomes hyperintense on T1- and T2-weighted images. Most of the literature discussing CST management consists of case reports. This infection requires immediate treatment which usually includes antibiotics and incision and drainage of the involved sites, as soon as possible. Surgical drainage normalizes intraorbital pressure, removes purulent material and establishes aerobic conditions, which increase the potency of the antibiotics and host defenses. While awaiting culture results, intravenous antibiotic therapy should be initiated based on the common pathogens involved, depending on the source, as we stated above. Antibiotics should be used for an extended period beyond clinical resolution to treat the possibility of sequestration within the thrombus [12]. The use of steroids is controversial [13]. Steroids decrease orbital inflammation, cranial nerve and edema; however, they have potential immunosuppressive effects and prothrombotic properties. Further studies would be useful to review the use of steroids in this condition. Likewise, the role of anticoagulation in the setting of CST has been debated. Some authors argue that anticoagulation prevents the progression of the thrombus, promotes the penetration of antibiotics within the thrombus and decrease the morbidity [14]. Others assert that the clot limits and confines the spread of bacteria, which may result in dissemination of septic emboli, and anticoagulation therapy has the risk of intracranial hemorrhage [15]. In case the use of anticoagulation, it should be used until radiologic resolution of the thrombus [16].

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There are some interesting features of this case. This presentation is infrequent because the abscess presenting in this patient was intraorbital, as we can observe after draining the purulent material by lateral canthotomy and cantholysis. Currently, with the use of broad-spectrum antibiotic and the early diagnosis of orbital complications of sinusitis, intraorbital abscess are uncommon and they are usually seen in immunocompromised patients. The latter could be the predisposing cause of our case. Organism prone to form abscesses, such as *S. milleri*, also would be more likely to cause intraorbital abscess formation. Due to pregnancy, we attempt to keep a little aggressive treatment with antibiotic; however, due to worsening, we decided to perform a surgical drainage. Despite the surgical drainage and proper antimicrobial treatment, the situation did not improve, and an elective cesarean section was performed, after which, the patient improved significantly. Although the treatment was appropriate, the patient got worse and we think that the key factor for the improvement of the clinical situation was the effect of cessation of immunosuppression, after the completion of pregnancy. It has been suggested that pregnancy causes an immunosuppressive state that would facilitate fetal tolerance and result in an increased susceptibility to infection [17]. The immune system changes are not well understood but these changes may alter susceptibility to and severity of infectious diseases in pregnant women and they remains until delivery.

Despite the poor prognosis of this condition and the even grimmer outlook of the aggregate condition, the outcome of our patient was favorable and she only experienced complete loss of vision in her right eye. In the other 3 similar cases reported a complete loss of vision in one eye [12], a definitive left abducens nerve palsy [9] and a transient oculomotor and abducens palsies [2], were observed. In conclusion, this case presents the unusual finding of a CST due to *S. milleri*. The medical and surgical treatments were overwhelmed by the aggressive nature of the infection. The key factor for the improvement of the clinical situation was the cessation of immunosuppression, after the completion of pregnancy.

Conflict of interest

None.

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