CASE REPORT

Early cavernous sinus thrombosis following unilateral pansinusitis in a child

Ajay Mallick,¹ Som Dutt Pathak,¹ Sandeep Shankar,² Alok Sati²¹Department of ENT, Command Hospital, Kolkata, West Bengal, India²Department of Ophthalmology, Command Hospital, Kolkata, West Bengal, India

Correspondence to

Dr Alok Sati, aloksati_123@rediffmail.com

Accepted 31 March 2015

SUMMARY

Cavernous sinus thrombosis (CST) is a life-threatening entity with a high rate of mortality and lifelong morbidity. A strong clinical suspicion of the complication, early radiological detection and institution of timely, aggressive treatment are required to prevent permanent neurological disability. We present a 12-year-old girl with bilateral CST following unilateral pansinusitis. Clinical symptoms of headache and retro-orbital pain out of proportion to clinical signs on presentation prompted a suspicion of something beyond sinusitis and raised the clinical suspicion of cavernous sinus involvement. In spite of institution of an early medical treatment, she progressed to develop bilateral cranial nerve paresis. However, with the persistence of antibiotics and anticoagulation therapy, she was successfully managed with no residual neurological sequelae. The aim of this presentation is to highlight the grave consequences of cavernous sinus involvement following infections of paranasal sinus and the rare complete recovery from disability in this case.

BACKGROUND

CST is a rare neurological emergency. Incidence of infections of cavernous sinus has dropped dramatically in the recent years with the advent of potent antibiotics. The rarity of cases may lead to misdiagnosis and delayed treatment. In spite of aggressive medical management, the incidence of permanent disability is high. Visual deficit occurs in one sixth of cases and half of them may have residual cranial nerve deficits.¹ Valveless communications between facial and ophthalmic veins and the cavernous sinus help infections from the paranasal sinuses and orbit to spread to the cavernous sinus leading to the potential fatal cavernous sinus thrombosis (CST).¹ We report a rare case of CST following unilateral paranasal sinusitis in a child, managed successfully without any residual deficit.

CASE PRESENTATION

A previously healthy 12-year-old girl (figure 1A) presented with acute onset of unilateral left sided headache and facial pain of 2 days duration. The headache and pain were agonising in nature, and maximum in the frontal and left orbital regions. She also had fever for 2 days, which was of moderate intensity and accompanied with chills but no rigours. She developed swelling and puffiness of both upper eyelids on the day of presentation for which she reported to this centre. There was no history of redness of the eyes, difficulty in vision or problem in moving the eyeball. She had nausea and

vomited once on the day of presentation. The child gave a history of upper respiratory tract infection 10 days prior to onset of present symptoms.

Examination revealed a child crying due to severe headache and left orbital pain. She had diffuse swelling of periorbital regions, left more than right. She had tenderness on palpation of the left eyeball. Proptosis was present (left eye-23 mm, right eye-19 mm on Von Hertel's exophthalmometer). The IOP appeared to be normal on digital pressure. She had full range of eye movements, and pupils were equal and reactive to light. There was no corneal or conjunctival congestion. Corneal reflex was present and she had no loss of sensation on the face.

There was a grossly deviated nasal septum to the left with hyperaemic nasal mucosa. Nasal endoscopy did not reveal any mucopus in the nose or nasopharynx. The endoscope could not be passed through the left of the middle meatus due to the grossly deviated nasal septum. Endoscopic findings on the right nasal cavity were essentially normal. On external pressure, there was tenderness on the frontal and ethmoid regions. There was no involvement of the child's cranial nerves. She had an otherwise unremarkable systemic and neurological examination.

TREATMENT

She was clinically diagnosed as a case of left-sided pansinusitis with impending orbital cellulitis and was immediately started on intravenous antibiotics (injection of linezolid, amikacin and metronidazole) and supportive therapy in the form of parenteral analgesics and intravenous fluids, and cetirizine tablets and decongestant nasal drops.

A CT scan (figure 2A) was performed at the time of admission, which showed extensive mucosal thickening of the left paranasal sinuses. There was no expansion or erosion of bone. The right side sinuses were normal. There was mild proptosis of the left globe. However, the orbital contents were normal and there was no collection in the orbit. On day two of admission, external eye inspection revealed inability to abduct or move the left eyeball upwards. There was no other derangement in ocular movements. There was no conjunctival congestion. Pupils were normal in size and reaction. Funduscopy was normal. Visual acuity was 6/6 in both eyes. These features were consistent with left third and sixth cranial nerve paresis. The rest of the girl's cranial nerve examination was normal. She continued to be febrile. Her headache and

CrossMark

To cite: Mallick A, Pathak SD, Shankar S, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-208441

Figure 1 Digital photograph on the day of admission showing a toxic looking patient with bilateral ptosis (A) and restriction of ocular movements (B). Recovery of ocular movements after treatment (C and D).

vomiting (4–5 times a day) were persisting. She was clinically diagnosed to be having CST.

MR venogram scans performed the same day (figure 2B) revealed an enlarged and deformed left cavernous sinus with diffuse thickening of dural walls. The cranial nerves III, IV, V1, V2 and VI were pushed laterally. The internal carotid artery was normal. Postcontrast films showed filling defect in the left cavernous sinus. MRI of the orbit was essentially normal. The child was started on anticoagulation therapy with low-molecular-weight (LMW) heparin 40 mg subcutaneous twice a day. Diagnosis of CST was further established with development of contra lateral right eye III and VI sixth nerve paresis on day 4 (figure 1B). There was no loss of vision. Corneal reflexes were normal. Fundus examination showed mild papilloedema. There were no signs of meningeal irritation or other cranial nerve palsy, and no weakness of limbs or change in sensorium. Blood and nasal swab cultures were negative. Cerebrospinal fluid study was normal. Intravenous dexamethasone 4 mg 8 hourly was added to the treatment.

OUTCOME AND FOLLOW-UP

On day 6, the child was subjectively better with an increase in appetite, and reduction in intensity of headache and orbital pain. Her fever also settled. With improvement in her general condition, no further intervention by way of change of management was contemplated at this stage, and a wait and watch policy was adopted for progression of her clinical signs. On the

eighth day, she showed gradual lateral movement of her right eye. Left eye lateral movements slowly started recovering after the tenth day. Heparin injection was continued for a total of 12 days. Amikacin and metronidazole injections were stopped after 5 days. Linezolid injection was continued for 21 days. After 3 weeks of admission, the child's ocular movements gradually improved in totality. She had regained complete range of eye movements (figure 1C, D). She was discharged from the hospital the next day on 1 mg acitrom tablets once a day for 1 month. She has remained asymptomatic on 3-month follow-up. She is planned for corrective septosinonasal surgery at 6 months follow-up.

DISCUSSION

CST is a rare, potentially life-threatening complication of paranasal sinusitis. It was first reported by Duncan² in 1821. Mortality and morbidity rates associated with this disease were high in the preantibiotic era; they have improved markedly because of an early awareness of the condition and prompt initiation of parenteral antibiotics.³ Permanent morbidity, however, remains a concern, with ophthalmic or neurological deficits in over half the survivors.⁴

Infection is the common cause of this fatal disease. The valveless veins around the face and paranasal sinuses have connections with the cavernous sinus, and form a common source of infection.^{5–7} Infections of these areas induce local venous congestion and the creation of an inflammatory procoagulant

Figure 2 (A) CT scan on admission showing evidence of pansinusitis left and (B) MR venogram showing cavernous sinus involvement.

cascade with subsequent thrombophlebitis of the cavernous sinus.⁸ *Staphylococcus aureus* accounts for approximately 70% of all infections.^{9 10} *Streptococcus pneumoniae*, Gram-negative bacilli and anaerobes can also be seen. Fungi are a less common pathogen and may include *Aspergillus* and *Rhizopus* spp.

Our decision to start intravenous antibiotics empirically with linezolid, amikacin and metronidazole injections was based on the available clinical data on the aetiopathogenesis of the disease. We did not wait for the culture sensitivity reports, so as to start the treatment at the earliest. There was no clinical or radiological evidence of fungal infection and hence antifungals were not exhibited.

The most common signs of CST are related to anatomical structures affected within the cavernous sinus, notably cranial nerves III–VI, as well as symptoms resulting from impaired venous drainage from the orbit and eye. Classic presentations are abrupt onset of unilateral periorbital oedema, headache, photophobia and bulging of the eyes (proptosis). Compression of one or more cranial nerves III, IV, VI, V₁ and V₂ results in impingement of extraocular movement, as projections of these nerves lie within or against the cavernous sinus. Sixth nerve paresis is the most common. Sensory deficits of the ophthalmic and maxillary branch of the fifth nerve are common. Periorbital sensory loss and impaired corneal reflex may be noted. Papilloedema, retinal haemorrhages and decreased visual acuity and blindness, may occur from venous congestion within the retina secondary to impaired drainage to a thrombosed cavernous sinus.

The advancement from unilateral to bilateral involvement due to intercavernous communication is pathognomonic of CST. Further thrombophlebitic spread may give rise to meningitis or subdural empyema and hemiplegia. Transient pituitary enlargement with central hypogonadism secondary to bilateral CST and syndrome of inappropriate secretion of antidiuretic hormone associated with CST has been reported.^{11 12}

Differential diagnosis includes a variety of acute and indolent orbital and intracranial conditions. Superior orbital vein thrombosis is an acute painful orbital condition characterised by high intraocular pressure, retinal venous congestion and chemosis. It can be differentiated from CST by its dramatic onset.

Orbital and conjunctival swelling and ophthalmoplaegia of orbital cellulitis and orbital abscess make distinction from CST difficult. These conditions can also occur from spread of infection from the paranasal sinuses and may mimic or lead to CST. The pattern of ophthalmoparesis, orbital tenderness and degree of swelling, distinguishes these entities. Visual symptoms and unilateral involvement are more pronounced in orbital cellulites. There is less likelihood of profound sepsis, meningeal signs and early fixation of the globe compared with CST.

Sino-orbital fungal infections such as aspergillosis and mucormycosis, Tolosa Hunt syndrome, pseudotumours of the orbit and other conditions producing a superior orbital fissure syndrome or orbital apex syndrome may resemble chronic CST. Visual impairment occurs when the orbital apex is involved but not in lesions confined to the superior orbital fissure. Both conditions can be caused by extension of infections from the paranasal sinuses. Degree of swelling of the orbit and loss of vision is more marked in the orbital conditions than in CST. Bilateral orbital involvement and meningism favour cavernous thrombosis. CST needs to be differentiated from carotid cavernous sinus fistulas (CCF). CCF usually follows trauma, spontaneous rupture of an aneurysm or an atherosclerotic internal carotid artery. It manifests as a painful ophthalmoplaegia with pulsating exophthalmos and bruit over the eye. Neuroradiology helps in identifying and differentiating these conditions from CST.

In our case, certain features of CST such as absence of corneal reflex, weakness of limbs and signs of meningeal irritation were absent probably due to the early institution of treatment. Awareness and timely management are crucial to prevent morbidity and mortality in this condition.

Antibiotic and intensive supportive therapy is the mainstay treatment of this condition. At present, there is no scientific consensus regarding the issue of anticoagulant therapy in cavernous sinus thrombosis. Early anticoagulant therapy within 7 days of the clinical course was reported to reduce the morbidity in adults who had this condition twofold.¹³ Anticoagulation with LMW heparin should be considered since the goal is to prevent further thrombosis and to reduce the incidence of septic emboli. Heparin is, however, contraindicated in the presence of intracerebral haemorrhage or other bleeding diathesis.^{14 15} Corticosteroids are considered as an adjunctive therapy as they help to reduce inflammation and oedema.

Our case had a similar presentation, with excruciating retro orbital pain, headache and fever. The child had progression of unilateral to bilateral cranial nerve involvement following unilateral sinus involvement. A high index of suspicion of CST was thought of due to the severity of headache and agonising retro orbital pain, which were out of proportion to her clinical findings. With the early institution of treatment with intravenous antibiotics and adequate supportive therapy including steroids and LMW heparin, the disease was restricted to involvement of III and VI cranial nerves only. There was no further intracranial involvement of other structures. Subsequently, the child had a complete recovery from her cranial nerve palsy.

We have presented a case of CST following unilateral pansinusitis, who was successfully managed conservatively with no residual sequelae. Despite absence of orbital or neurological signs, the severity of symptoms aroused an index of suspicion of an intracranial complication. Subsequent neuroimaging confirmed the entity of CST. Awareness, early recognition and clinical diagnosis, administration of antibiotics without waiting for culture reports and proper supportive care are the key factors leading to a good outcome in CST.

Learning points

- ▶ Cavernous sinus thrombosis is a rare but potentially fatal complication of paranasal sinus infections requiring a high degree of clinical suspicion and a multidisciplinary, multimodality approach to identify the diagnosis.
- ▶ Overt ophthalmic and neurological signs may not be present in early cavernous sinus involvement from infections of the paranasal sinuses.
- ▶ Early institution of medical therapy and supportive measures without waiting for culture and imaging reports helps in limiting the condition and to avoid disabling morbidity or mortality.

Acknowledgements The authors acknowledge the contribution of Dr V Bhalla, Dr RK Thapar and Dr T Bose for their valuable contribution in managing the case.

Contributors AM, SDP were involved in the conception and design, acquisition of data or analysis and interpretation of data: AM, SDP, SS, AS were involved in the drafting the article or revising it critically for important intellectual content AM, SDP, SS, AS gave final approval of the version to be published.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- Mitchell R, Kelly J, Wagner J. Bilateral orbital complications of pediatric rhinosinusitis. *Arch Otolaryngol Head Neck Surg* 2002;128:971–4.
- Duncan A. Contributions to morbid anatomy. *Edinburg Med Soc J* 1821;17:319–25.
- Sofferman RA. Cavernous sinus thrombophlebitis secondary to sphenoid sinusitis. *Laryngoscope* 1983;93:797–800.
- DiNublie MJ. Septic thrombosis of the cavernous sinus. *Arch Neurol* 1968;45:567–72.
- Canon ML, Antonio BL, McCloskey JJ, et al. Cavernous sinus thrombosis complicating sinusitis. *Pediatr Crit Care Med* 2004;5:86–8.
- Baidya K, Bhaduri G, Mondal LK, et al. Atypical presentation of a unilateral septic thrombosis of cavernous sinus. *J Indian Med Assoc* 2005;103:437–8.
- Radulescu L, Poita I, Curca IA, et al. Thrombophlebitis of left cavernous sinus secondary to acute right sphenoid sinusitis. *Rev Med Chir Soc Med Nat Iasi* 2005;109:573–6.
- Esmon CT, Taylor FB Jr, Snow TR. Inflammation and coagulation: linked processes potentially regulated through a common pathway mediated by protein C. *Thromb Haemost* 1991;66:160–5.
- Price CD, Hameroff SB, Richards RD. Cavernous sinus thrombosis and orbital cellulitis. *South Med J* 1971;64:1243–7.
- Tveteras K, Kristensen S, Dommerby H. Septic cavernous and lateral sinus thrombosis: modern diagnostic and therapeutic principles. *J Laryngol Otol* 1988;102:877–82.
- Joubert M, Verdon R, Reznik Y. Transient pituitary enlargement with central hypogonadism secondary to bilateral cavernous sinus thrombosis: pituitary oedema? *Eur J Endocrinol* 2009;160:873–5.
- Feinfeld DA, Al-Achkar G, Lipner HI, et al. Syndrome of inappropriate secretion of antidiuretic hormone: association with cavernous sinus thrombosis. *JAMA* 1978;240:856–7.
- Levine SR, Twyman RE, Gilman S. The role of anticoagulation in cavernous sinus thrombosis. *Neurology* 1988;38:517–22.
- Coutinho J, de Bruijn SF, Deveber G, et al. Anticoagulation for cerebral venous sinus thrombosis. *Cochrane Database Syst Rev* 2011:CD002005.
- Visudtibhan A, Visudhiphan P, Chiemchanya S. Cavernous sinus thrombophlebitis in children. *Pediatric Neurology* 2001;24:123–7.

Copyright 2015 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- Submit as many cases as you like
- Enjoy fast sympathetic peer review and rapid publication of accepted articles
- Access all the published articles
- Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow