



Pediatric cavernous sinus and superior ophthalmic vein thrombophlebitis complicated by peri-venous orbital abscesses

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ABSTRACT

Purpose: This case report documents a 13-year-old male with bilateral cavernous sinus thrombosis and left superior ophthalmic vein thrombosis secondary to sphenoid sinusitis who subsequently developed peri-venous orbital abscesses.

Observations: Although the patient initially improved clinically with intravenous antibiotic therapy and therapeutic anticoagulation, his symptoms returned and repeat imaging demonstrated two well-circumscribed, rim-enhancing lesions in the left orbit concerning for abscesses. Surgical intervention revealed that these purulent collections were contiguous with the superior and inferior ophthalmic veins with the superior collection also containing hemorrhagic material. Despite aggressive management, the abscess in the inferolateral orbit recurred, requiring repeat surgical intervention. The patient was ultimately discharged on parenteral antibiotics and anticoagulation, and on outpatient follow-up, there was no evidence of recurrence.

Conclusions and Importance: This case report describes a pediatric patient with bilateral cavernous sinus thrombosis who developed the rare complication of multiple peri-venous orbital abscesses despite appropriate treatment with antibiotics. The patient's complex clinical course highlights the importance of prompt repeat imaging and possible surgical intervention upon clinical changes, especially in patients with infections from the *Streptococcus milleri* group.

1. Introduction

Septic cavernous sinus thrombosis (CST) is a rare but serious complication of sinusitis or orbital cellulitis. With modern treatment consisting of antibiotics and anticoagulation, mortality rates have fallen significantly in the last century while morbidity associated with CST has remained high.¹ Despite prompt treatment, there is still a significant risk for further complications. In this case, a patient with cavernous sinus and superior ophthalmic vein (SOV) thrombosis developed peri-venous orbital abscesses despite antibiotic treatment. The collection and evaluation of protected patient health information included in this case report were HIPAA compliant.

2. Case report

A 13-year-old male with a past medical history of septo-optic dysplasia and cerebral palsy presented to the emergency department with a one-day history of left eye redness and swelling in the setting of

recent fevers. The patient had been diagnosed with pneumonia the previous night and placed on oral antibiotics. The patient had also completed a course of oral antibiotics for sinusitis in the month preceding. At the time of presentation, the patient had signs concerning for sepsis, namely fever, tachycardia, and tachypnea. Significant laboratory findings included an elevated procalcitonin and C-reactive protein (CRP). Ophthalmic exam was significant for bilateral periorbital edema with conjunctival injection and chemosis of the left eye (Fig. 1A). Assessment of visual acuity was limited as the patient was non-verbal, but he was able to fixate and track. Pupils were reactive bilaterally without a relative afferent pupillary defect. Extraocular movements demonstrated a slight decrease in abduction of the left eye but were otherwise intact. Intraocular pressure was normal. No proptosis was noted at this time.

Initial imaging of the face (CT followed by MRI) showed bilateral sphenoid sinusitis, bilateral CST, and left SOV thrombosis (Fig. 1B). After imaging, the patient immediately underwent bilateral maxillary antrastomies, bilateral total ethmoidectomies, and bilateral

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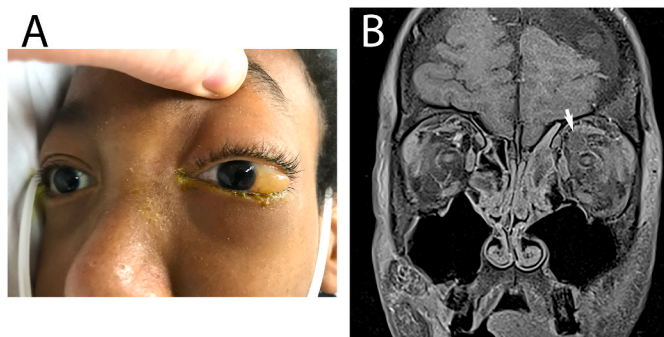


Fig. 1. 1A: External photograph demonstrating periorbital edema and chemosis of the left eye at presentation.
 1B: Coronal section of T1-weighted contrast-enhanced MR orbital study demonstrating left SOV thrombosis, evidenced by filling defects and lack of opacification, without abscess at presentation.

sphenoidotomies as well as collection of sinus cultures with the otolaryngology service. Antibiotic therapy was initiated with intravenous ceftazolin and metronidazole and later transitioned to intravenous ceftriaxone and oral metronidazole on day five per the recommendations of infectious disease. Blood cultures revealed *Streptococcus constellatus*, while cultures from the sinus contents revealed methicillin sensitive *Staphylococcus aureus*. Therapeutic anticoagulation with enoxaparin was begun after these initial studies.

The patient continued to improve until day eight when he was noted to have increased left eye proptosis and new onset pyrexia (Fig. 2A). Ophthalmic exam was otherwise unchanged. Nevertheless, these findings prompted repeat imaging, which demonstrated two rim-enhancing, well-circumscribed lesions in the left orbit concerning for abscesses. The larger, superior lesion was contiguous with the thrombosed SOV and

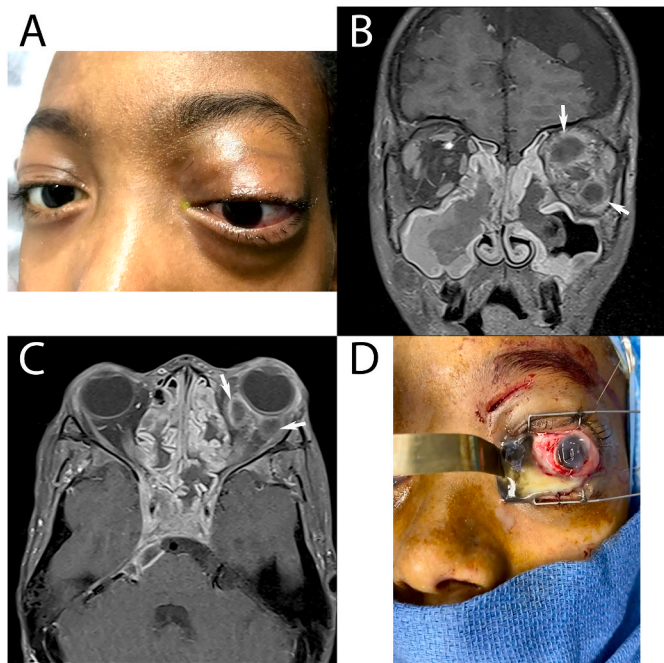


Fig. 2. 2A: External photograph demonstrating increased left eye proptosis on day 8.
 2B: Coronal section of T1-weighted contrast-enhanced MR orbital study demonstrating two orbital abscesses on day 8.
 2C: Axial section of T1-weighted contrast-enhanced MR orbital study demonstrating posterior extensions of both orbital abscesses on day 8.
 2D: Intraoperative external photograph demonstrating hemorrhagic and purulent material released during left medial frontal orbitotomy on day 8.

measured 2.3 cm × 1.8 cm, and the inferior lesion measured 1 cm × 1 cm (Fig. 2B and C). An additional lesion concerning for abscess was noted within the right preptentine cistern in addition to findings consistent with meningitis and skull base osteomyelitis. Finally, there was significant progression of maxillary sinus disease compared to initial imaging with the maxillary sinuses now completely or nearly completely opacified bilaterally.

The patient was subsequently taken to the operating room for abscess incision and drainage and additional sinus surgery including bilateral ethmoidectomy and sphenoidotomy and extensive removal of tissue from the bilateral sinuses. After the otolaryngology service completed their portion of the operation, an initial left medial sub-brow incision was made, but dissection down to the superomedial orbital rim revealed no infection or abscess. Next, after making a left perilimbal conjunctival incision and temporarily releasing the medial rectus from its insertion site, a left medial frontal orbitotomy was performed. A white walled off area contiguous with the left SOV was incised exuding purulent and hemorrhagic material, which was then cultured (Fig. 2D). A separate left inferolateral frontal orbitotomy was also performed with an initial incision at the left lateral canthal region extending horizontally across the left lower eyelid through the palpebral conjunctiva. A distinct white walled off area contiguous with the inferior ophthalmic vein was identified and incised. Purulent exudate from this collection was also cultured. Adequate irrigation and drainage were performed. At this time, infectious disease recommended continuing ceftriaxone and converting metronidazole from an oral to intravenous route of administration. They noted that if intraoperative cultures were to reveal an organism that the current regimen did not cover, they would broaden coverage. Cultures from both orbital peri-venous abscesses revealed mixed skin flora, and therefore, broadening antibiotic coverage was deemed unnecessary. There was also consideration of intrathecal antibiotics, which were ultimately decided against in light of the patient's unique anatomy and no definitive assurance of antibiotic penetration.

The patient initially recovered appropriately, but five days later was noted to have increased agitation and tachycardia. Although the ophthalmic exam was unchanged, repeat imaging showed recurrence of the inferior abscess in addition to resolution of the superior abscess (Fig. 3A and B). The patient underwent a repeat left frontal inferolateral orbitotomy via limbal peritomy with purulence noted intraoperatively. Cultures revealed *Streptococcus constellatus*. Post-operatively, the patient exhibited substantial clinical improvement and repeat imaging prior to discharge showed residual orbital fat stranding without any evident collection. The patient was discharged on parenteral antibiotic therapy and therapeutic anticoagulation. On outpatient follow-up one month later, the patient showed continued improvement as manifested by resolution of orbital signs without evidence of recurrence.

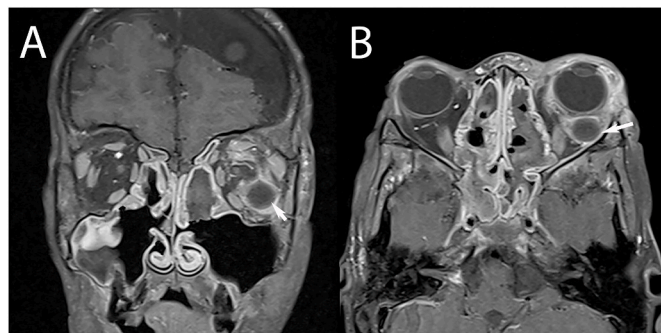


Fig. 3. 3A: Coronal section of T1-weighted contrast-enhanced MR orbital study demonstrating recurrence of inferior orbital abscess five days after initial surgical intervention (day 13).
 3B: Axial section of T1-weighted contrast-enhanced MR orbital study demonstrating posterior extension of recurrent abscess on day 13.

3. Discussion

Septic cavernous sinus thrombosis is a rare but serious complication of sinusitis, as in this case, or orbital cellulitis. Sphenoid sinusitis is often implicated in the development of CST as a result of the anatomical proximity of the sphenoid and cavernous sinuses along with the valveless venous system that drains the sinuses, face, and orbit.^{1,2} Ultimately, unfettered infection can then spread via thrombophlebitis or septic emboli from these regions to the cavernous sinus.¹ When the diagnosis of CST is suspected based on clinical and radiographic findings, rapid delivery of broad-spectrum intravenous antibiotics is the mainstay of treatment while the role of anticoagulation is disputable.^{3,4} Despite consensus that broad spectrum antimicrobial coverage is a cornerstone of the treatment regimen for CST, complications can still arise.

This report presents a case of multiple peri-venous orbital abscesses that developed in the setting of bilateral CST and SOV thrombosis despite appropriate antibiotic treatment and anticoagulation. This suggests secondary infectious phlebitis of the SOV, in addition to direct thrombotic extension. To our knowledge, one other case has reported a similar clinical situation in which a 17-year-old girl developed sphenoid sinusitis, CST, and SOV thrombosis.⁵ After an undefined period of antibiotic therapy, the patient was noted to have increased proptosis of the left eye. Repeat imaging demonstrated a new superomedial collection concerning for an abscess, which was drained operatively. As in our patient, microbial analysis revealed *Streptococcus milleri*.⁵ Although in the setting of orbital cellulitis, another case report describes a patient with septic SOV thrombosis that developed a peri-SOV orbital abscess despite receiving antibiotic treatment.⁶ A distinctive aspect of our case compared to these reports is the presence of hemorrhagic material in addition to purulent material in the abscess contiguous with the left SOV. This finding suggests hemorrhage of the thrombosed left SOV, leading to or complicating the peri-venous abscess. The hemorrhage may have been induced by the patient's therapeutic anticoagulation, which presents a serious complication risk associated with the common treatment adjunct. Importantly, both thrombosis and infection can spread in a retrograde manner in the setting of cavernous sinus thrombophlebitis.

Multiple cases in the literature describe patients with CST who developed orbital abscesses while on antibiotics, but without contiguity with venous vasculature.^{1,5,7} Interestingly, in a number of these cases, cultures identified a member of the *Streptococcus milleri* group (SMG), including *Streptococcus intermedius*, *constellatus*, and *anginosus*. These bacteria thrive in anaerobic conditions and are known to have greater sinus involvement compared to other bacteria often implicated in orbital cellulitis and to form multiple abscesses despite antibiotic treatment, emphasizing the challenge of eradicating these invasive infections.^{5,8–10} In a letter to the editor expanding upon the paper published by Watkins et al., the authors described six cases of orbital abscesses that grew bacteria from the SMG.⁸ They reported a high risk of persistent or recurrent disease as their patients required multiple operations to control the source of infection despite antibiotic coverage, thereby reiterating the danger of this bacteria.⁸ This underlines the importance of close monitoring and aggressive management to reduce morbidity and mortality, especially when bacteria from the SMG are implicated.

The blood cultures and final abscess culture of our 13-year-old patient revealed *S. constellatus*, a virulent member of the SMG. The patient required multiple operations despite antibiotic treatment to control the infection. In order to starve the bacteria of its anaerobic environment, source control and drainage of orbital collections were urgently

necessary. It is important to note that in cases with new onset worsening of ocular symptoms or visual acuity, repeat imaging to detect potential new orbital abscess formation and subsequent drainage has been shown to result in an acute improvement in symptoms and reduction in long-term sequelae.¹

4. Conclusions

This case report presents a rare complication of cavernous sinus thrombophlebitis in the antibiotic era and highlights the necessity of a multi-disciplinary team monitoring closely and making prompt adjustments as needed in the management of this disease. In certain infections, especially with bacteria from the SMG, antibiotics may not prevent the progression of disease, and providers must be prepared to quickly intervene to reduce morbidity in their patients. Additionally, it is important to remain vigilant despite appropriate initial management and investigate clinical changes prompting repeat imaging or surgical intervention.

Patient consent

The patient's legal guardian consented to publication of the case in writing/orally.

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