Case Report

CEREBRAL VENOUS SINUS THROMBOSIS SECONDARY TO ISOLATED MAXILLARY SINUSITIS AND MSSA BACTEREMIA: A CASE REPORT

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ARTICLE INFO

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Received:
July 2021
Accepted for publication:
July 2021

Keywords:

Cerebral venous sinus thrombosis, ptosis;
subdural effusion;
maxillary sinusitis;
life threatening;
early suspicion.

ABSTRACT

Cerebral venous sinus thrombosis (CVST) is a rare life-threatening condition related to high mortality and morbidity. The diagnostic challenge arises as patient may be asymptomatic or present with a variety of constitutional and neurological symptoms. We report a case of CVST in a young immunocompetent male. A 19 years old boy with no other comorbidity presented with sudden onset of fever, right jaw and temple pain, was referred to Ophthalmology team for right ptosis. Upon examination and prompt diagnosis, extensive dural venous sinus thrombosis with right cerebral convexity subdural effusion, right submandibular, parapharyngeal and retropharyngeal collections were detected through radiological imaging. Right maxillary sinusitis as the source of infection was also detected. The patient underwent maxillary sinus drainage and successfully treated with systemic antibiotics and anticoagulants. We highlight the importance of early suspicion of systemic involvement in patients presenting with eye symptoms. Prompt diagnosis, emergent radiographic evaluation, multidisciplinary approach and appropriate treatment with anticoagulants and antibiotics can improve survival and visual prognosis.

INTRODUCTION

Cerebral venous sinus thrombosis (CVST) is a rare life -threatening condition [1] related to high mortality and morbidity. It pose a diagnostic challenge as patient may be asymptomatic or may present with a variety of constitutional and neurological symptoms ranging from fever, headache, altered consciousness, cranial neuropathies or stroke, and often requiring radiological guidance [2,3]. Cerebral venous sinus thrombosis can be categorized etiologically into primary/idiopathic or secondary causes, further divided into infective or non-infective. Non-infective causes may be due to coagulation disorders, neoplasms, homocystinuria, vasculitis or head trauma [4]. Infective causes are more common, with the most common being acute sinusitis specifically the sphenoid and ethmoid sinusitis, commonly attributed to Staphylococcus Aureus infection [3,5]. Failure to diagnose and treat the primary infection may result in devastating outcome. Favourable outcome depends on recognition, prompt broad spectrum antibiotic treatment and surgical control of the primary focus [6].

We report a case of CVST in a young immunocompetent male with acute maxillary sinusitis, an uncommon site of spread of infection complicated with Methicillin sensitive *Staphylococcus aureus* (MSSA) bacteremia. Prompt investigations and diagnosis were made with multidisciplinary approach and with initiation of antibiotics and anticoagulants, severe life-threatening complications were prevented.

CASE REPORT

A 19-year-old young male with no past medical history was referred by Emergency Department to Ophthalmology team for mild right ptosis. He presented with high grade fever and excruciating right jaw pain extending to the right preauricular region and right temple. There were no other symptoms of increased intracranial pressure such as nausea or vomiting.

On examination, he was alert, conscious but febrile with a temperature of 38.5 degree Celsius. He had mild right conjunctival injection with mild ptosis and mild swelling over the right temple. All other vital parameters were unremarkable. Best corrected visual acuity (BCVA) was 6/12 and 6/9 in the right and left eye respectively. Neuro-ophthalmologic assessment showed no anisocoria, no relative afferent pupillary defect (RAPD), no visual field defect by confrontation test and colour vision was normal. The intraocular pressures were normal. There was no proptosis, or periorbital swelling. The anterior segments examination was unremarkable in both eyes. On fundoscopy, there was no evidence of optic disc swelling. He had limited

supraduction of the right eye with no limitation of other gazes and no diplopia were elicited (Figure 1).

He had right jaw tenderness extending to the right temple associated with inability to open his mouth. Dental and otorhinolaryngology examination was unremarkable.

Infective and thrombophilia screening was done. Total white cell count was 13.47x 10^9/L with neutrophil predominance, Erythrocyte Sedimentation Rate (ESR) of 71 mm/hour and C-Reactive Protein of 349 mg/dl. Viral and diabetic screening was negative. There was no hypercoagulable state, values of complement 3 and 4 were normal. International Normalized Ratio (INR) was 1.67, fibrinogen 445, and prothrombin time 19.4.

Contrast Enhanced Computed Tomography (CECT) of orbit and neck showed thrombosis in the superior sagittal sinus, cavernous sinus, right sigmoid sinus extending to the right proximal internal jugular vein (IJV) with no venous infarct or acute intracranial bleed. There was right cerebral convexity subdural effusion with mild cerebral oedema. There was also submandibular, parapharyngeal, retropharyngeal, carotid space and small temporalis muscle collections over the right side. Right masseter, temporalis and pterygoid muscles were bulky. Right Fossa of Rossenmuller (FOR) was obliterated with fullness of nasopharynx as well as oropharynx. Evidence of right maxillary sinusitis were also seen (Figure 2).

While awaiting for his blood culture and sensitivity (C&S), he was treated empirically with high dose of intravenous (IV) Ceftriaxone and anticoagulant Enoxaparin. He was also started on anti- epileptics IV phenytoin 100 mg thrice a day as prophylaxis against possible seizure.

On Day 3 of presentation, worsening of supraduction and abduction of right eye was seen associated with chemosis and injection in keeping with orbital cellulitis. The right intraocular pressure increased to 26 mmHg , with otherwise normal neuro-ophthalmologic assessment. There was no optic disc swelling. He was given additional topical moxifloxacin and timolol maleate.

Blood C&S grew Methicillin Sensitive *Staphylococcus aureus* (MSSA). Diagnosis of cerebral venous sinus thrombosis (CVST) secondary to maxillary sinusitis with MSSA bacteremia was made. Ophthalmologically patient had right orbital cellulitis with superior division third nerve palsy secondary to CVST.

He was then covered with IV Ceftriaxone and Cloxacillin for 6 weeks and was started on oral warfarin 3 mg 8 hourly, with dosage adjustments made based on daily INR. Otorhinolaryngology team performed the drainage of maxillary sinus.

After two weeks of medication, patient was afebrile and appeared more comfortable. Repeat Blood C&S showed no growth. Eye examination however showed persistence of mild right ptosis without any limitation in extraocular muscle movement. Chemosis and injection of right eye resolved with normal intraocular pressure. Fundoscopy however showed blurred disc margin of both optic disc. Enlarged blind spot on Bjerrum proves presence of papilloedema which was absent prior.

A repeat CECT brain and neck (Figure 3) showed worsening of right cerebral convexity subdural collections with suspicious intraparenchymal extension, dural and leptomeningeal involvement and cerebral oedema. However, there was resolving right submandibular, resolved superior sagittal sinus CVST with residual filling defects in right cavernous sinus, right sigmoid sinus extending to right IJV.

Due to worsening of cerebral collections and oedema, Infectious Diseases team started the

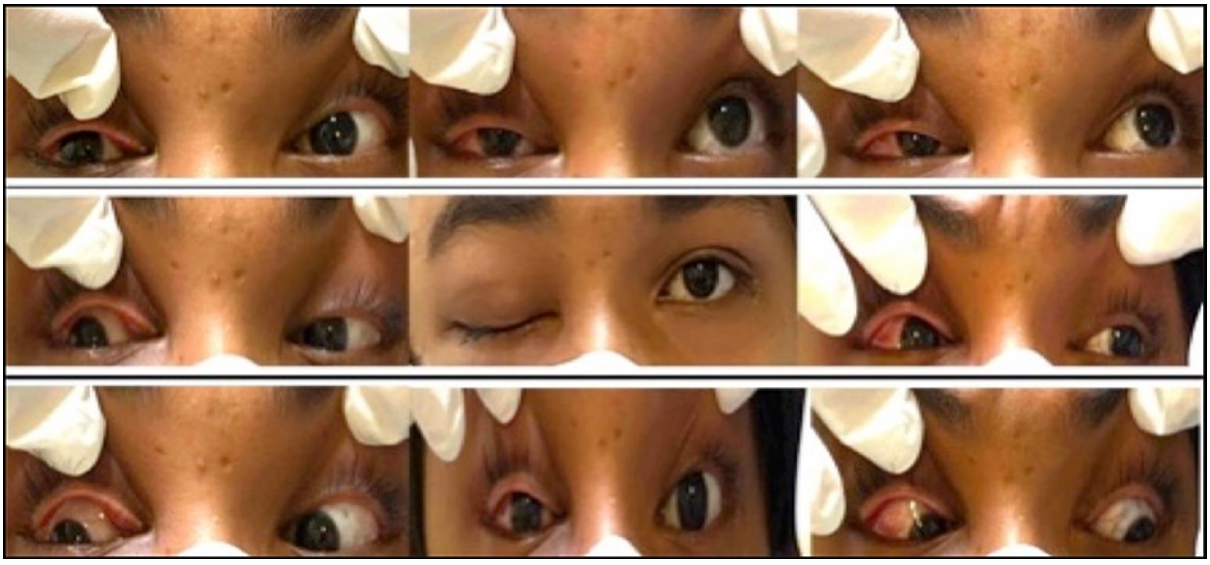

Figure 1: Shows 9 cardinal direction of gaze, right ptosis, mild injection of right conjunctiva and limitation of right supraduction.

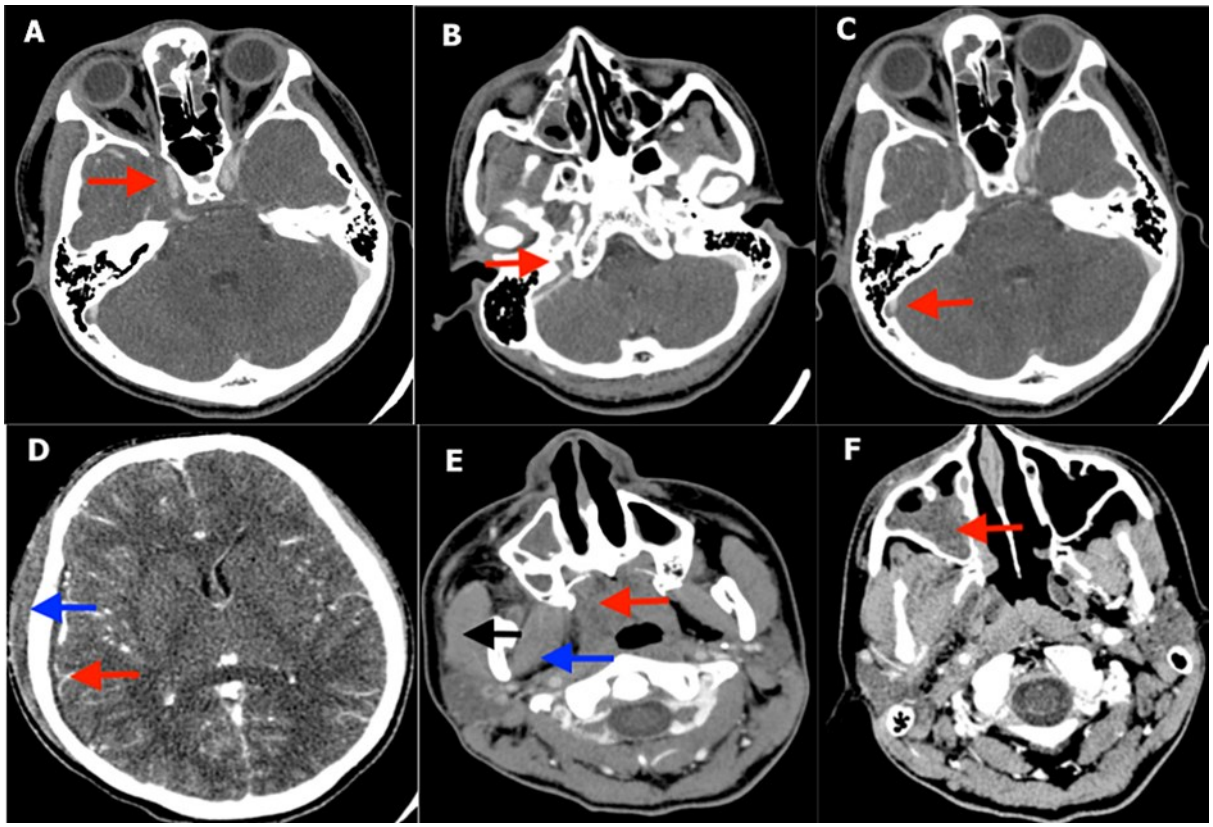

Figure 2: Axial brain Contrast Enhanced Computed Tomography:

A. Shows filling defect within right cavernous sinus
B. Filling defect within right jugular bulb
C. Filling defect within right sigmoid sinus
D. Right cerebral convexity with iso to hypodense subdural collection (red arrow) and swollen right temporalis muscle (blue arrow)
E. Right fullness of nasopharynx and oropharynx (red arrow), right bulky pterygoid muscle (blue arrow) and masseter muscle (black arrow)
F. Near total opacification of the right maxillary sinus, consistance with right maxillary sinusitis.

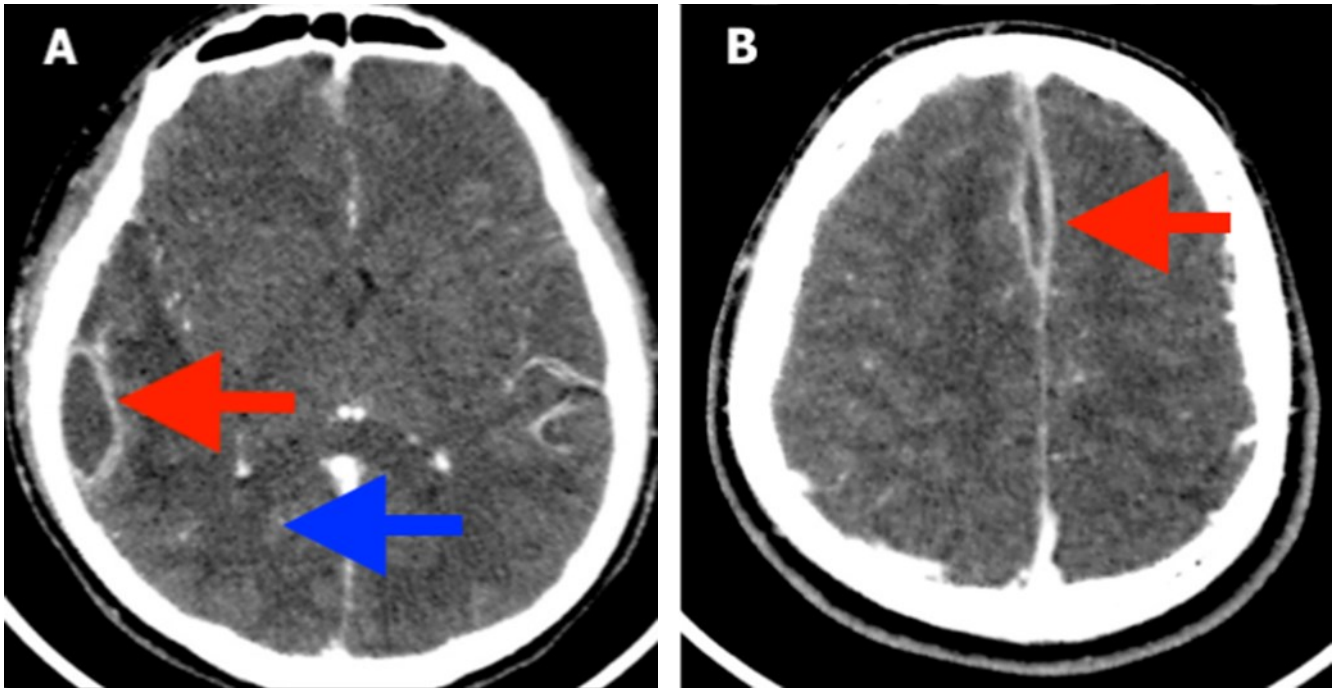

Figure 3: Axial brain CECT : **A**. Shows worsening right cerebral convexity hypodense subdural collection with suspicious intraparenchymal extension, dural and leptomeningeal involvement (red arrow) as well as cerebral oedema (blue arrow). **B.** Shows hypodense collection in the interhemispheric fissure.

patient on IV Meropenem which has better coverage in MSSA related complications and continued the IV Cloxacillin. Biopsy of right level 5 lymph done revealed chronic inflammation histologically. *Mycobacterium Tuberculosis* bacteria was not detected and neither was there evidence of tuberculosis infection radiologically or pathologically.

At completion of 6-weeks course of antibiotics, patient showed complete resolution of the ocular changes with no ptosis, no limitation of eye movements (Figure 4) and no papilloedema . He was afebrile and well.

DISCUSSION

In cerebral venous sinus thrombosis, prothrombotic condition is almost always present. In the absence of prothrombotic condition, as seen in our patient, it is nearly always related to infective causes [6]. The spread of infection is commonly from the paranasal sinuses, mid face and orbit [7]. The case we reported here had the spread of infection from the maxillary sinus which is rarely involved in CVST [4].

Our patient developed Methicillin sensitive *Staphylococcus aureus* septicaemia from maxillary sinusitis and progressed to extensive cerebral venous sinus thrombosis involving all major cerebral venous sinuses with collections extending into the pharyngeal spaces. One would expect a patient with an extensive involvement of CVST to present with increased intracranial pressure symptoms, pupillary dilatation and complete ophthalmoplegia with sinusitis symptoms like rhinorrhea, facial pain and tenderness of maxillary sinus. Our patient however did not present with such alarming signs.

All cerebral venous sinuses are valveless, thus they facilitate forward or backflow of blood allowing pathogens and neoplastic cells to travel to different parts of the brain. Infection coming from layers of the face can easily travel into the cavernous sinuses leading to the development of a thrombus resulting in cavernous sinus thrombosis. This condition may result in the swelling of the affected sinuses and damage to the surrounding cranial nerves [8]. Common symptoms include ptosis, proptosis, chemosis, ophthalmoplegia (cranial nerve [CN] III, IV, VI), and loss of sensation in the ophthalmic and maxillary divisions of the trigeminal nerve (V1 and V2) [7].

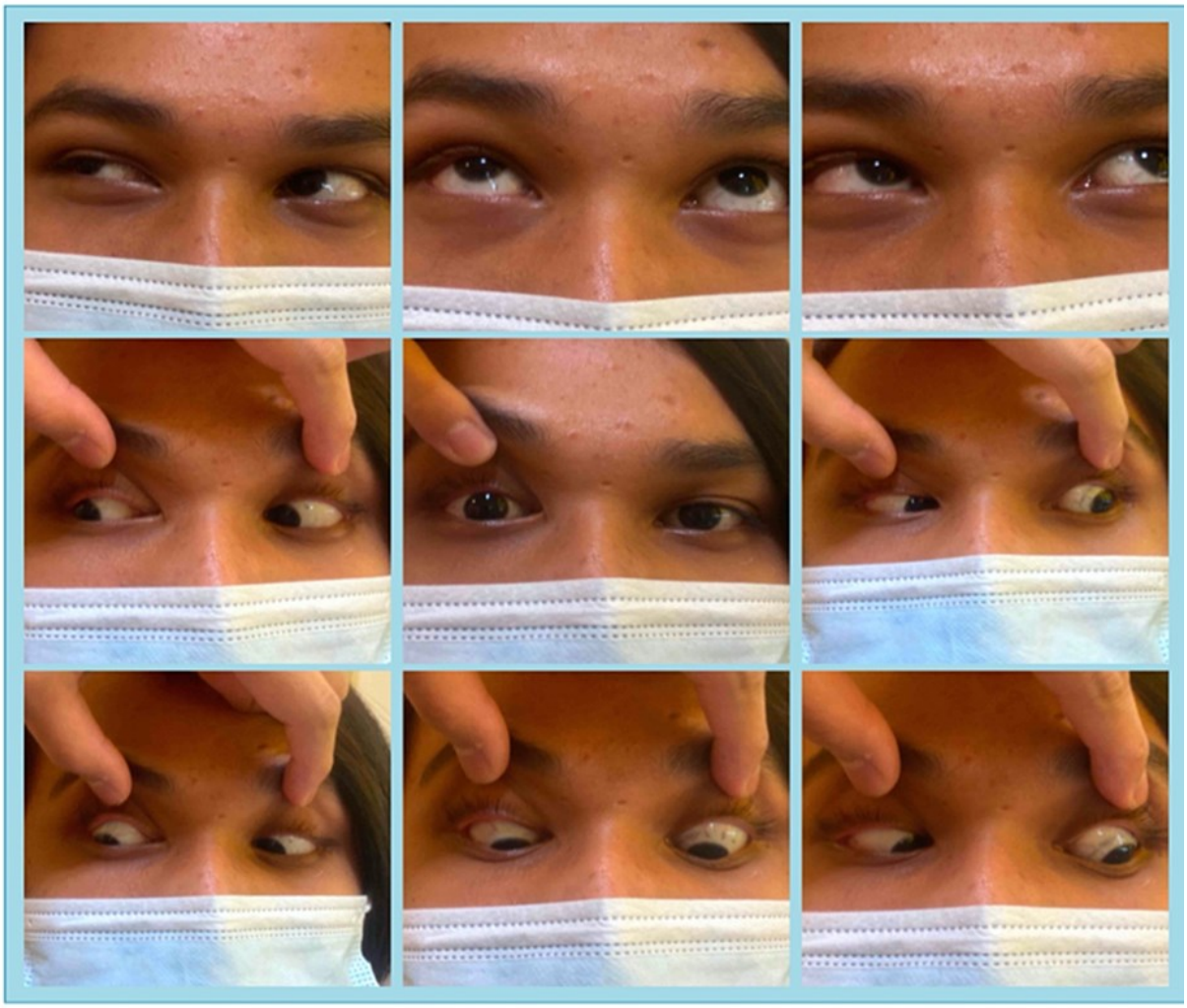

Figure 4: Shows resolution of ptosis and conjunctival injection as well as improvement of extraocular movements.
Repeated CECT brain and neck (Figure 5) showed resolved CVST. Patient was discharged with oral warfarin 3.5mg OD and INR of 2.20. He ultimately made a full recovery.

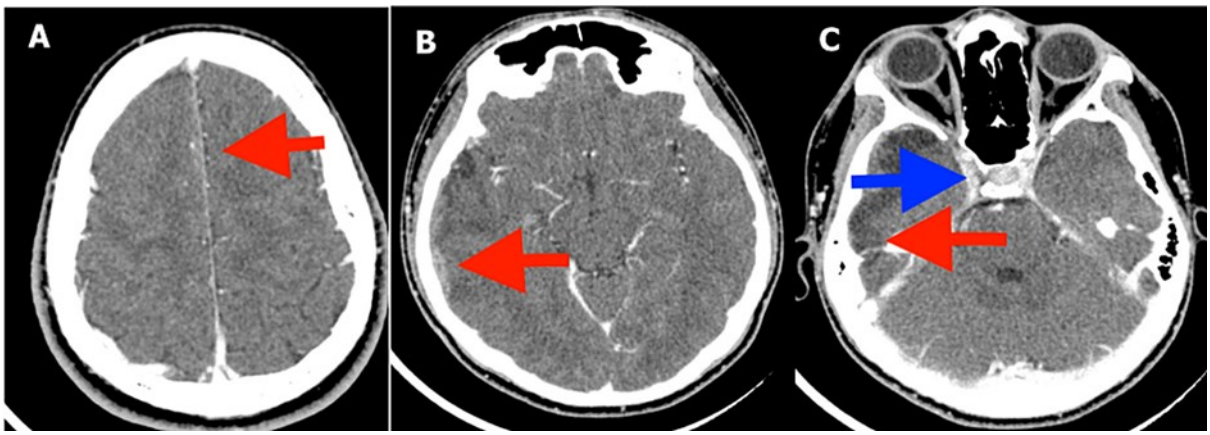

Figure 5: Axial brain CECT upon completion of treatment :**A.** Resolved interhemispheric collection **B.** Smaller residual of right cerebral convexity subdural collection

C. Resolving right temporal parenchymal collection with residual dural enhancement (red arrow) and minimal residual filling defect in cavernous sinus (blue arrow).

The outcome of CVST varies from a complete recovery to death. Although most patients with CVST are discharged with favourable outcome, 18.9% of patients succumb to death or dependency according to the International study on cerebral vein and dural sinus thrombosis (ISCVT) [9]. Most patients treated promptly with anticoagulants had good prognosis [10].

Our patient who presented with unilateral ptosis, fever and jaw pain, an immediate multidisciplinary team management was carried out and an immediate radio imaging aided us to the diagnosis and etiology (maxillary sinusitis). Prompt biologic investigations and initiation of antibiotics and anticoagulants had also helped us in treating the underlying MSSA and the CVST. The Otorhinolaryngology team had eliminated the source of infection by surgical drainage. Series of clinical and radiological examination had helped us in managing this patient with an excellent outcome for such an extensive CVST.

CONCLUSION

Early recognition and treatment of CVST has reduced mortality rate over time. Despite being a rare disease, CVST is frequently associated with catastrophic outcomes if it is not timely treated. Our case discussion highlights the importance of early suspicion of systemic involvement in patients presenting with ocular symptoms. Prompt diagnosis, emergent radiographic evaluation, multidisciplinary approach and appropriate treatment with anticoagulants and antibiotics can improve survival and visual prognosis.

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