

Case Report

Iatrogenic cerebral sinovenous thrombosis - well known but yet under reported

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ABSTRACT

Cerebral sinus venous thrombosis (CSVT) although rare, there is concern that cases of this potentially treatable condition are missed. The clinical manifestations can be life-threatening and cause long-term neurological deficits. As the symptoms and signs are non-specific, diagnosis is often delayed and may be missed, hence the onus lies on clinician to request for appropriate investigations. Oral contraceptives by inducing hypercoagulable state found to have strong causal association with CSVT.

Keywords: CSVT, Oral contraceptives

INTRODUCTION

Cerebral sinus venous thrombosis (CSVT) is a rare form of venous thromboembolism with an estimated incidence of 0.67 per 100,000 children per year.¹ CSVT is characterized by a highly variable clinical spectra, difficult diagnosis, variable etiologies and prognosis that require fine medical skills and high index of suspicion. CSVT affects the cerebral venous drainage and related anatomical structures. Prognosis depends on early detection. Proper enquiry of the generating cause and correcting the cause prevents the complications.

CASE REPORT

A 14 years old female presented with complaints of severe progressive headache since 2 months, fever and non-projectile vomiting for 3 days and two episodes of generalized tonic clonic seizures followed by altered sensorium. No history of (H/O) ear discharge or trauma. There was no significant past history. Child was developmentally normal, immunized appropriate for age. Vitals were stable. Neurological examination revealed right sided hemiparesis. Rest of neurological examination and that of other systems were unremarkable. Base line Investigations like blood sugar, complete hemogram,

CSF analysis, blood culture were normal. Coagulation profile (PT, aPTT, INR) within normal limits. Protein C and S was normal. MRI Brain findings concern for cerebral sinus venous thrombosis. MR Venogram (MRV) suggestive of filling defect in superior sagittal sinus and few cortical veins, left transverse sinus is hypoplastic/thrombosed. As we couldn't find definitive cause precipitating CSVT, we reassessed history and child's uncle has revealed about use of oral contraceptive pills (OCP) for a period of 8months, for complaints of menorrhagia without medical follow-up. Child was started on anticoagulation therapy to maintain INR (international normalized ratio) between 1.5-2.0. Neurological deficit recovered completely. She was discharged on oral warfarin for 3months, with regular follow-up. On follow-up child is symptom free and doing well. Follow-up MRV after 3 months has shown recanalization of superior sagittal sinus and cerebral veins and hypoplastic left transverse sinus

DISCUSSION

The incidence of CSVT is 0.67 per 100,000 children per year.¹ In children, CSVT most commonly affects a single dural sinus, the superficial sagittal and transverse sinuses represent 47.5% and 12% of cases, respectively; in the

same study, 30% of cases involved more than two sinuses.² There are numerous risk factors associated with paediatric CSVT, underlying infection is the most common risk factor, however, dehydration, coagulopathies, and malignancies are common predisposing conditions.¹⁻³

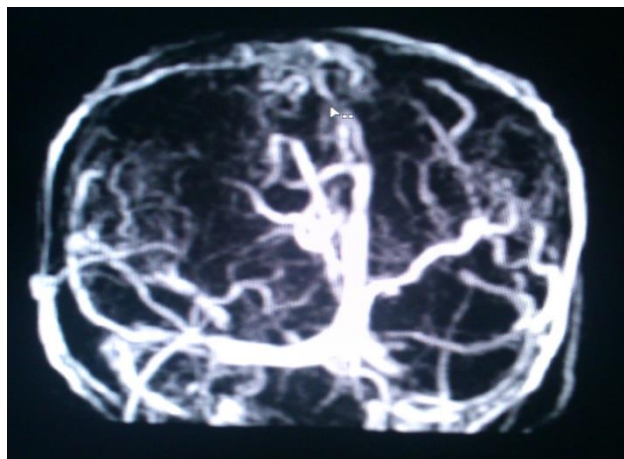


Figure 1: Non enhancement within the superior sagittal sinus and few cortical veins and Non visualization of left transverse sinus.

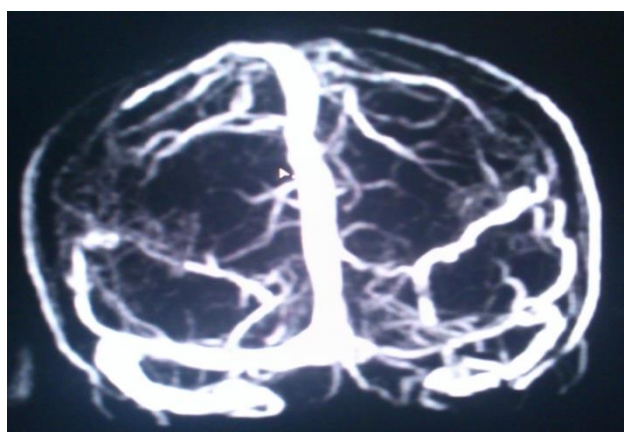


Figure 2: Follow-up imaging; complete recanalization of superior sagittal sinus with hypoplastic left transverse sinus.

Heller et al reported that CSVT in most cases were due to prothrombotic risk factors.⁴ There are case reports of CSVT due to usage oral contraceptives (OCP) in adults.⁵ This is the first case report of CSVT secondary to OCP use in children as far as our knowledge is concern. Few studies reported OCP's as predisposing factor in children in their study like Mallick et al (2 cases), Wasay et al (1 case), Kenet et al (1 case), Heller et al (4 cases).^{2,4,6,7}

The most common signs and symptoms of CSVT are severe headache, seizures (focal or generalized), altered sensorium or coma, papilledema, like in our case. CSVT in neonates present with seizures and nonspecific neurological signs.^{1,3} Although CT is typically the most

readily available neuroimaging technique, it is reportedly normal in 20-30% of CVST cases.^{8,9} The cord sign (hyper dense thrombosed veins on unenhanced CT scans) and the empty-delta sign (a filling defect in the superior sagittal sinus on enhanced CT scans) are considered pathognomonic for CVST.⁹ CT venography may improve the diagnostic yield when MRI is not available.

Although MRI more readily demonstrates associated parenchymal infarcts, care needs to be taking when interpreting MRI/MRV images, in particular where there is anatomical variation.⁹ In 75% of people the right transverse sinus is dominant over the left and 20-30% of transverse sinuses (usually the non-dominant side) have narrowed or atretic segments, as seen in our case.⁹

There is currently a consensus that in children beyond the neonatal period without hemorrhage, anticoagulation should be considered.^{10,11} Treatment regimens vary between centers, but many older infants and children receive anticoagulation in the acute setting with either parenteral unfractionated heparin, subcutaneous low molecular weight heparin (LMWH), or oral warfarin. Anticoagulation therapy improves outcome due to mortality and morbidities. CSVT mortality is less than 10%, but neurological impairments are present in 17% to 79% of survivors. Recurrence is seen in 20-40% of children with CSVT, requiring long term follow-up.^{6,12}

In our case after 3 months of follow-up MRI with MRV was repeated which has shown recanalization of superior sagittal sinus and cortical veins.

CONCLUSION

Cerebral sinus-venous thrombosis is a rare but life-threatening disease. Oral contraceptives are one of the leading iatrogenic causes. The true incidence of CSVT secondary to OCP's is not known. Growing knowledge about use of OCP's and its widespread availability has led to its usage without any medical supervision leading to complications. This case enlightens about importance of counselling and educating parents on early clinical signs and insisting on regular follow-up, could lead to early diagnosis and intervention.

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