Case Report

Cerebral Venous Sinus Thrombosis Associated Subdural Hematoma: A Case Series on Clinical Presentation and Management

Priyanka Chakkera, Manisha Gupta, Sandeep Marasakatla, Subasree Ramakrishnan, Girish Baburao Kulkarni

Department of Neurology, National Institute of Mental Health and Neurosciences (NIMHANS), Bengaluru, Karnataka, India

Abstract

Diagnosis and treatment of cerebral venous sinus thrombosis (CVT) associated with subdural hematoma (SDH) is challenging with an increased risk of rebleeding on using anticoagulation. There are no guidelines at present due to its rare presentation. In this report we describe three patients who presented with non-traumatic SDH and CVT over the last 3 years. Clinical assessment, investigations including neuroimaging, and management were reviewed both at time of admission and follow-up. These patients presented with varied CVT syndromes -isolated raised intracranial pressure (ICP), focal and diffuse encephalopathy. Neuroimaging helped in diagnosing CVT and SDH. Cases 1 and 3 had SDH alone, while case 2 had SDH along with intraparenchymal hemorrhage. Management of these patients was tailored individually as per mechanism of CVT. Case 1 was clinically stable, however, she had rebleeding after starting anticoagulation, requiring its discontinuation. Cases 2 and 3 underwent immediate neurosurgical intervention in view of deteriorated sensorium. Although CVT manifesting as SDH is rare, clinicians should have a high index of suspicion to accurately diagnose and manage these challenging cases. The decision regarding use of anticoagulation and apt time for neurosurgical intervention needs to be individualized depending on patients condition and response to treatment.

Keywords: Cerebral venous sinus thrombosis, management, neuroimaging, subdural hematoma

Introduction

Cerebral venous sinus thrombosis (CVT) usually presents as raised intracranial pressure (ICP) or hemorrhagic venous infarct.[1] CVT presenting as subdural hematoma (SDH) is very rare. Diagnosing such cases is challenging due to varied presentations. Also, their management is controversial and not well established due to rarity and risks associated with anticoagulation. Here, we present three cases of CVT with SDH and describe the management based on our experience it is not a review of literature article.

Case Description

Over the last 3 years, on reviewing medical case records three patients of CVT associated with SDH were found. A summary of all cases is provided in Table 1.

Case 1

A 25-year-old lady, presented with holocranial headache associated with vomiting for 6 days at first visit. There was no history of fever, seizures, and visual disturbances. Neurological examination was normal. Initial computed tomography (CT) scan of brain showed superior sagittal sinus (SSS), right transverse sinus (TS) and sigmoid sinus thrombosis with no intracranial bleeding. Patient was advised admission however she left against advice with treatment defaulted. Patient revisited 4 days later, with complaints of worsening headache, painless blurring of vision, double vision. Attenders noticed that the patient was unable to close eyes completely. There was no history of trauma. On examination, patient was conscious and oriented. Corrected visual acuity was 6/6 in both eyes. Fundus was showing bilateral grade III papilledema with bilateral lateral rectus palsy and lower motor neuron type of facial palsy. Rest of neurological examination was normal. Repeat CT brain showed CVT with SDH over the left frontoparietal (thickness ~ 9.3 mm) and bilateral tentorial leaflets [Figure 1a]. Magnetic resonance imaging (MRI) of brain showed CVT with features of raised intracranial pressure (ICP) [Figure 1b]. Risk factor identified was iron deficiency anemia (hemoglobin was 9.6 g/dL with microcytic hypochromic picture). Her coagulation profile, routine blood chemistry, Vitamin B12, homocysteine and folate were normal. Anti-nuclear antibody (ANA) profile, Anti-neutrophil cytoplasmic antibody (ANCA) and antiphospholipid antibodies (APL) were negative. Thrombophilia screening

Address for correspondence: Dr. Girish Baburao Kulkarni, Department of Neurology, National Institute of Mental Health and Neurosciences (NIMHANS), Bengaluru, Karnataka, India.
E-mail: gbk5000@rediffmail.com

Submitted: 20-Apr-2023 **Revised:** 30-May-2023 **Accepted:** 17-Jun-2023
Published: 25-Sep-2023

DOI: 10.4103/aian.aian_346_23

Table 1: Clinical Characteristics of 3 Patients with CVT associated with SDH

No.	Age/ sex	Presenting CVT Syndrome	Examination	Risk factor	Neuroimaging	Treatment	Outcome	Follow-up imaging
1.	25/F	Raised ICP syndrome	GCS: E4V5M6, bilateral grade III papilledema and lateral rectus palsy	Anaemia	CT and MRI Brain Plain and contrast: CVT of SSS, right TS and SS with SDH over left frontoparietal and bilateral tentorial leaflets and features of raised ICP Repeat CT brain after anticoagulation for 10 days: new SDH over left parietooccipital left cerebellar convexity	Conservative management with injectable followed by oral antiedema measures	Improved with complete resolution of headache and papilloedema	MRI at 6 months: markedly decreased SDH near total recanalization of sinuses
2.	65/F	Focal encephalopathy	GCS: E2V1M5, no focal deficits	Chronic alcoholic	CT Brain plain and contrast: Acute SDH along right fronto-parietal convexity and parieto-temporo-occipital intraparenchymal hemorrhage. CVT of SSS, right TS and SS	Surgical intervention followed by low dose injectable and oral anticoagulation	Sensorium improved with minimal persistent weakness of left upperlimb	CTV at 4 months: partial recanalization
3.	32/M	Encephalopathy	GCS E1M3VT, with asymmetrical pupil with paucity of movements on left side	Alcoholic, hyperhomocystienemia	CT Brain plain and contrast: thrombosis of SSS, bilateral TS and SS with bilateral symmetrical frontoparietal SDH Post Operative MRI: CVT and features suggesting intracranial hypotension with no spinal cord leak.	Surgical intervention followed by low dose injectable and oral anticoagulation	Sensorium improved and independent of daily activities	MRI at 6 months: complete recanalization with resolution of SDH

No.: Number, F: Female, ICP: Intracranial pressure, GCS: Glasgow Coma Scale, CT: computed tomography, MRI: magnetic resonance imaging, CVT: Cerebral venous thrombosis, SSS: Superior sagittal sinus, TS: Transverse sinus, SS: sigmoid sinus, SDH: subdural hemorrhage, CTV: Computed Tomography venography, M: Male

was not done in acute condition. Patient was managed conservatively with antiedema measures and started on a titrated dose of unfractionated heparin (UFH) (5000 IU twice a day subcutaneous (s.c.) up to qid for 10 days) gradually after 2 weeks. Patient complained of worsened headache after 10 days. Repeat CT brain showed new SDH over left parietooccipital (thickness ~ 6.8 mm) [Figure 1c] and left cerebellar convexity (thickness ~ 1.2cm). Anticoagulation was stopped and antiedema measures were continued. Neurosurgery team advised conservative management. At follow-up, she had complete resolution of headache and papilledema. She was continued on acetazolamide (1750 mg/day). Follow-up imaging at 6 months showed markedly decreased SDH [Figure 1d] with near total recanalization of involved sinuses but persisting features of raised ICP.

Case 2

A 65 year old diabetic and hypertensive lady with no prior history of use of antiplatelets, presented with a headache for 7 days and altered sensorium for 1 day. There was no history of seizures, focal deficit, or trauma. She was chronic alcoholic for 35 years. On examination, patient was drowsy, opening eyes to painful stimuli and localizing to painful stimulus with no focal deficits.

Her CT brain showed right parieto-temporo-occipital intraparenchymal hemorrhage with surrounding edema and mass effect with midline shift of 9.3 mm toward left side, uncal herniation with effacement of peri mesencephalic cisterns. Acute SDH noted along the right fronto-parietal convexity (thickness ~ 4.5 mm) [Figure 1e]. Post-contrast study showed filling defect in the SSS and right TS and sigmoid sinuses [Figure 1f]. Her hemogram and biochemistry parameters, Vitamin B12, folate, homocysteine were normal. She underwent right temporo-parieto-occipital decompressive craniectomy. Thrombophilia work-up was not done in acute phase. Cause of CVT was undetermined. Subsequently, the patient was treated with antiedema measures and anticoagulants (UFH 5000 IU qid a day s.c. started after 48 hours of surgery). During hospital stay, her sensorium improved. On follow-up, patient had minimal weakness of left upper limb(Modified Rankin scale (mRS - 2)) and was on oral anticoagulation for 4 months (T. Acenocoumarol 1 mg/day). Repeat CT venography at 4 months showed partial recanalization of venous sinuses.

Case-3

A 32-year-old male presented with history of headache, vomiting and altered sensorium for 3 days. There was history of binge alcohol intake one week prior. At admission, his GCS was E1M3VT, with asymmetrical pupil with paucity of movements on left side. His CT brain showed SSS, bilateral TS and Sigmoid sinus thrombosis with bilateral symmetrical frontoparietal SDH and diffuse cerebral edema with effacement of peri mesencephalic cistern [Figure 1g and h]. As his sensorium deteriorated, he underwent right frontoparietal burr hole with evacuation of SDH. Pre-operative MRI was

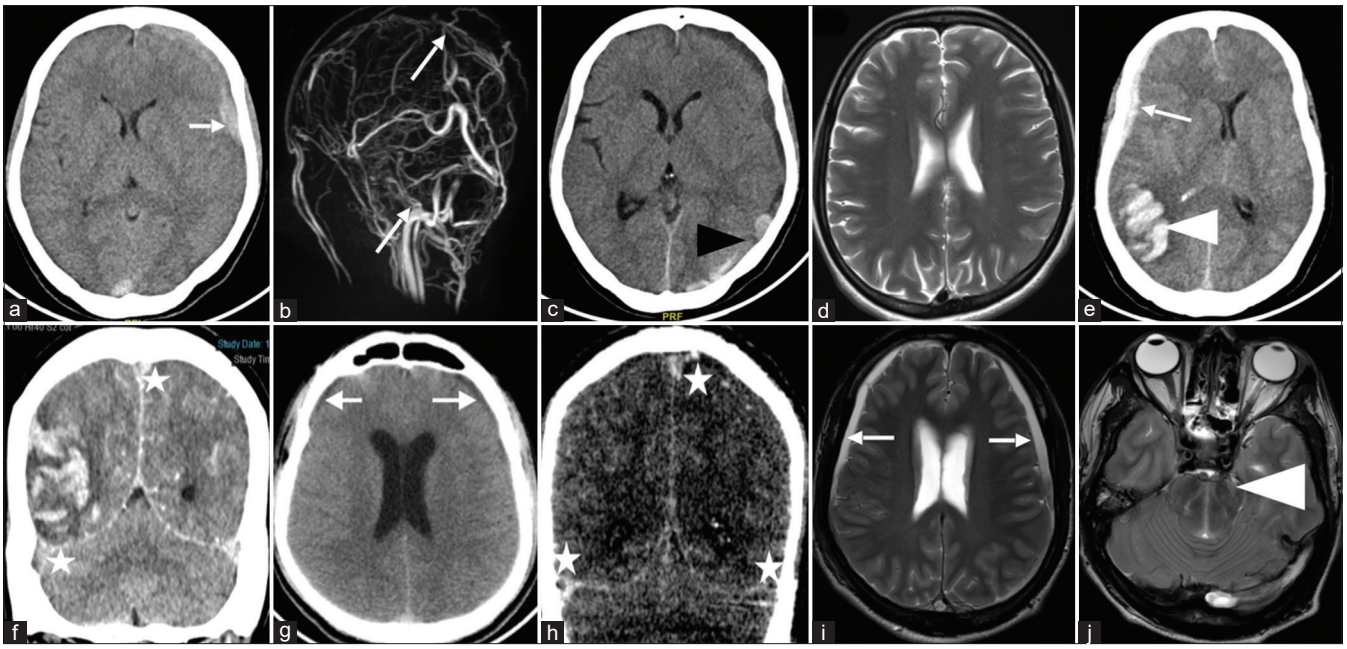

Figure 1: a-d: Neuroimages of Case 1: (a) CT brain showing left frontoparietal SDH (solid white arrow) and hyperdense confluence of sinus. (b) MRV showing superior sagittal sinus and bilateral transverse sinus thrombosis (solid white arrow). (c) Repeat CT brain on worsening of headache showed new SDH in left parietooccipital region (solid black arrowhead). (d) Follow-up MRI showing resolution of SDH; e, f: Neuroimages of Case 2: (e) CT brain showed right parietooccipital intracerebral hemorrhage (solid white arrowhead) with right frontoparietal SDH and midline shift (solid white arrow). (f) Post-contrast CT showing filling defects in SSS and Right transverse sinuse (solid white star); g-j: Neuroimages of Case 3: (g) Preoperative CT brain plain showing bilateral symmetrical subdural collections (solid white arrow) and (h) CT contrast showing thrombosis in superior sagittal sinus and bilateral transverse sinuses (solid white star). i and j) MRI Brain post-operatively showing bilateral symmetrical subdural collections (solid white arrow) with effacement of perimesencephalic cisterns and brainstem sagging (solid white arrowhead)

not done. Post Operative MRI was showing CVT and features suggesting intracranial hypotension with no spinal cord leak [Figure 1i and j]. Hence, it was concluded that CVT was secondary to intracranial hypotension. Thrombophilia profile was normal, ANA, ANCA, ACL antibodies were negative. He was managed with intravenous fluids for low ICP and low dose Inj. Unfractionated Heparin 5000 IU s.c. twice a day (after 48 hours of surgery followed by gradually increasing to qid). T. Acenocoumarol 1 mg/day was continued after discharge for 6 months. His sensorium improved and his follow-up mRS was 0 at 3 months. Repeat MRI at 6 months follow up showed recanalization of CVT with resolution of subdural collection.

Discussion

Very few cases of CVT presenting as SDH have been reported till now. It is surprising to find that none of the 624 patients were reported to have SDH in International Study on Cerebral Vein and Dural Sinus Thrombosis.[2] The incidence of SDH secondary to CVT was reported as 11% in a retrospective study of 260 patients with confirmed CVT over a period of 15 years done in Mexico and Europe.[3] Subdural hematoma can present in isolation or along with other intracranial hemorrhages. Isolated SDH was seen in Cases 1 and 3, while SDH associated with intraparenchymal hemorrhage was seen in Case 2. Subdural hematoma with CVT might be secondary to obstructed venous outflow, dural artery venous fistula, complication of the treatment with thrombolytics, or following intracranial hypotension.[4-7] The bleeding manifestations are due to the rupture of bridging veins secondary to obstructed venous outflow as was seen in Cases 1 and 2. However, in Case 3, CVT with SDH might be secondary complication of intracranial hypotension as suggested by bilateral symmetrical fronto parietal SDH on CT (but presurgical MRI would have confirmed it) and later detected in post-operative MRI. Rare manifestations of CVT including subdural hematoma are easily detected by increased availability of MRI, MR venography and CT venography.[8] All patients should be investigated for predisposing conditions to thrombosis, particularly screening for thrombophilia should be done as it is usually associated with SDH secondary to CVT.[9] Screening for thrombophilias including: protein C, protein S, antithrombin III deficiency; prothrombin and Factor V Leiden gene mutation; lupus anticoagulant; anticardiolipin antibodies; anti beta2glycoprotein antibodies; homocysteinemia related to mutations in the methyltetrahydrofolate reductase gene; and hemoglobinopathies should be done.[9]

The management of CVT with SDH is challenging, due to conflicting benefits of management with anticoagulation against the risk of SDH expansion or recollection.[1] Due to the rarity of its presentation, there are no fixed guidelines for management. Khatib and Baviskar[10] proposed an algorithm to treat CVT complicated as SDH/SAH by

starting immediate anticoagulation if the patient is stable and has no mass effect in neuroimaging. However, they recommended immediate neurosurgical intervention with supportive care in case of neurological deficit with mass effect. They recommended re-evaluation of the patient two weeks post-operatively for risk of rebleeding and consider oral anticoagulation. In Case 1, we applied this algorithm. However, after increase in SDH, we considered the risks of restarting the anticoagulation are higher than the benefits, so we elected to manage the patient conservatively with clinico-radiological follow-up and without anticoagulation. There are no guidelines on when to start anticoagulation in CVT with intracranial hemorrhage. As per our institute protocol, we started anticoagulation early in Cases 2 and 3 but in low doses followed by close observation. Endovascular thrombectomy of CVT for patient with recurrent SDH as treatment option was suggested by Akin *et al*.[9] This helps in circumventing the potential risk of bleeding from systemic anticoagulation. Clinicians must be attentive to intracranial hypotension complicating SDH and CVT, wherein treatment approach is different as in our case 3.

Based on recent literature review of such cases done by Lim *et al*.,[11] following details are known. There is no defined age group and it is variable from the second to eighth decade. Most common presentation is headache, similar to other CVT presentations and most common risk factor found was thrombophilia. Majority had unilateral SDH in neuroimaging except for two patients. However, the literature remains divided on the management, between the use of anticoagulation versus conservative treatment. We individualized management of such cases based on our experience, patient's response and available literature.

Conclusion

Though SDH secondary to CVT is rare, clinicians should consider it as differential in patients presenting with non-traumatic SDH. The role of anticoagulation in the management of CVT complicated by SDH remains controversial requiring development of guidelines. A high index of suspicion is required to diagnose timely and accurately and manage this challenging disease to prevent further complications.

Declaration of patients consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Alharshan R, Qureshi HU, AlHada A, Shaikh M, Khalil A. Cerebral venous sinus thrombosis manifesting as a recurrent spontaneous subdural hematoma: A case report. Int J Surg Case Rep 2020;67:223-6.
2. Ferro JM, Canhão P, Stam J, Bousser M-G, Barinagarrementeria F; ISCVT Investigators. Prognosis of cerebral vein and dural sinus thrombosis: Results of the International Study on Cerebral Vein and Dural Sinus Thrombosis (ISCVT). Stroke 2004;35:664-70.
3. Afifi K, Bellanger G, Zuurbier SM, Esperon CG, Barboza MA, Costa P, *et al*. Features of intracranial hemorrhage in cerebral venous thrombosis. J Neurol 2020;267:3292-8.
4. Bucy PC, Lesemann FJ. Idiopathic recurrent thrombophlebitis with cerebral venous thromboses and an acute subdural hematoma. JAMA 1942;119:402-5.
5. Takamura Y, Morimoto S, Uede T, Yamaki T, Minamida Y, Yamamura A, *et al*. Cerebral venous sinus thrombosis associated with systemic multiple hemangiomas manifesting as chronic subdural hematoma — case report—. Neurol Med Chir (Tokyo) 1996;36:650-3.
6. Dhillon AK, Rabinstein AA, Wijdicks EF. Coma from worsening spontaneous intracranial hypotension after subdural hematoma evacuation. Neurocrit Care 2010;12:390-4.
7. Singh S, Kumar S, Joseph M, Gnanamuthu C, Alexander M. Cerebral venous sinus thrombosis presenting as subdural haematoma. Australas Radiol 2005;49:101-3.
8. Saposnik G, Barinagarrementeria F, Brown RD, Bushnell CD, Cucchiara B, Cushman M, *et al*. Diagnosis and management of cerebral venous thrombosis. Stroke 2011;42:1158-92.
9. Akins PT, Axelrod YK, Ji C, Ciporen JN, Arshad ST, Hawk MW, *et al*. Cerebral venous sinus thrombosis complicated by subdural hematomas: Case series and literature review. Surg Neurol Int 2013;4:85.
10. Khatib KI, Baviskar AS. Treatment of cerebral venous sinus thrombosis with subdural hematoma and subarachnoid hemorrhage. J Emerg Trauma Shock 2016;9:155-6.
11. Lim EYT, Pai V, Sitoh YY, Purohit B. Acute subdural haemorrhage complicating cerebral venous thrombosis in a patient with protein C deficiency. BMJ Case Rep 2020;13:e236745.