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Case Report

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Cerebral Venous Thrombosis Caused by Spontaneous Intracranial Hypotension: A Case Report

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Conflict of Interest
The authors have no financial conflicts of interest.

ABSTRACT

This case report presents a rare case of cerebral venous thrombosis (CVT) caused by spontaneous intracranial hypotension (SIH). The cause and prognosis of CVT can vary; CVT caused by SIH is uncommon and difficult to diagnose and treat. In this case, magnetic resonance imaging myelography showed definite cerebrospinal fluid leakage, and the patient's symptoms did not improve after conventional treatment. Furthermore, subdural hematoma occurred, causing mental deterioration; however, it improved dramatically after the blood patch procedure and burr hole drainage, which was performed after early cessation of anticoagulant therapy.

Keywords: Intracranial hypotension; Sinus thrombosis, intracranial; Anticoagulants

INTRODUCTION

Cerebral venous thrombosis (CVT) is a rare disease with various etiologies. Symptoms of CVT vary widely from headaches to intracerebral hemorrhage. In partial cases CVT may be cured without any sequelae. However, severe neurologic defects, including death, may occur in many cases.[3] For good clinical outcomes, the treatment strategy should be adjusted while considering the etiology and the status of the patient. However, it is not easy to establish clear clinical guidelines owing to its low incidence, difficulty in diagnosis, and diverse etiology and clinical outcome. Here, we report the case of a patient with CVT caused by spontaneous intracranial hypotension (SIH) with recurrent subdural hematoma (SDH) during treatment.

CASE REPORT

A 34-year-old man was admitted to the hospital because of a gradually aggravating and uncontrolled headache that had begun 2 weeks prior. Magnetic resonance imaging (MRI) and computed tomography (CT) demonstrated CVT along the superior sagittal sinus and right transverse sinus. Diffuse pachymeningeal thickening along both cerebral convexities and engorged venous sinuses implied intracranial hypotension (**FIGURE 1**). Thorough clinical examination and history taking represented no other definite cause for CVT in this patient.

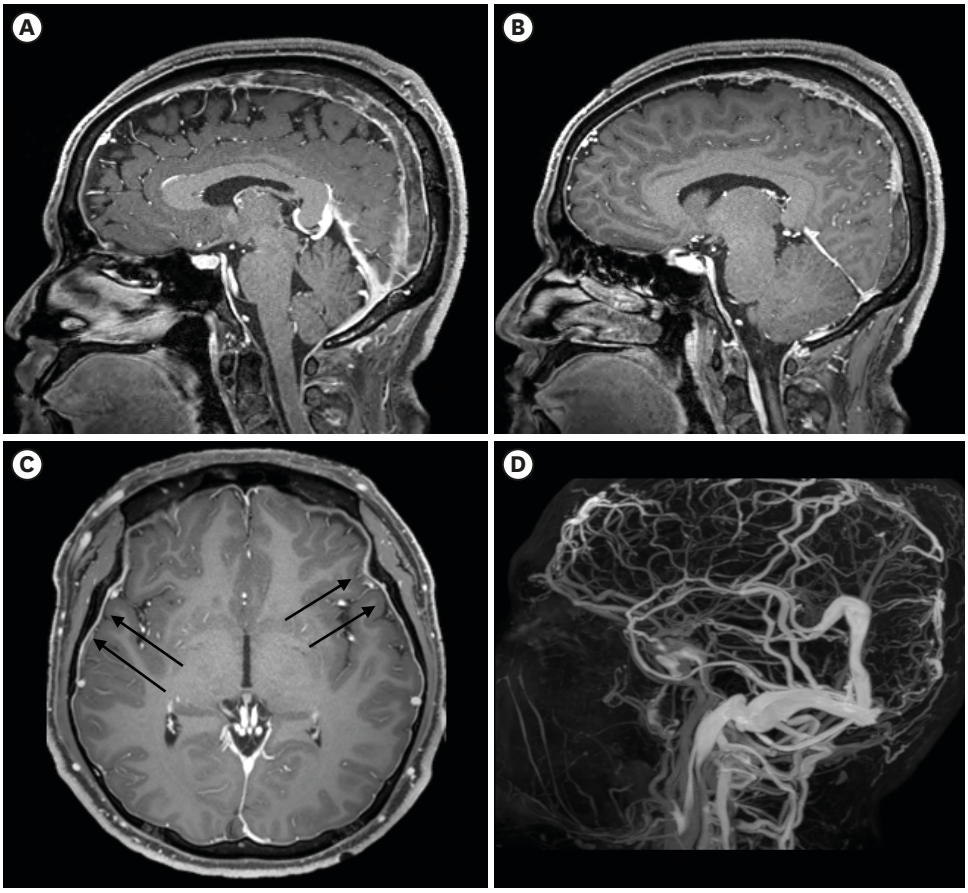

FIGURE 1. Brain magnetic resonance imaging. T1 gadolinium-enhanced imaging sagittal views (A & B), axial view (C) and MR venography (D). (A & B) Filling defects in sagittal and transverse sinuses are shown. (C) Black arrows indicate diffuse pachymeningeal thickening. (D) MR venography reveals non-visualization of the superior sagittal sinus.
MR: magnetic resonance.

The patient received immediate intravenous anticoagulation. Whole spine MRI myelography was performed under suspicion of SIH; however, there was no definite finding of cerebrospinal fluid (CSF) leakage. Transfemoral cerebral angiography was performed for detailed vessel evaluation (**FIGURE 2**). In a week, the patient's symptoms gradually improved, and intravenous anticoagulation was changed to novel oral anticoagulant (NOAC) medication (apixaban 5 mg bid). Although the patient was in a stable state, subdural effusion was detected on a follow-up CT scan (**FIGURE 3A**). The patient was in stable condition. However, orthostatic headache became clear with time. After 4 weeks, the patient experienced mental deterioration, and brain CT showed aggravated bilateral chronic SDH, compressing both lateral ventricles (**FIGURE 3B**). Anticoagulation medication was suspended, and emergency burr hole drainage was performed bilaterally. The patient recovered, and one week later, an epidural blood patch was performed bluntly on a suspicious CSF leakage site. Orthostatic headache considerably improved. The patient recovered after supportive care and was discharged without prescribing anticoagulants, although a mild headache persisted. After 2 months, follow-up MRI showed improvement in CVT; however, recurrent SDH occurred in the left hemisphere (**FIGURE 4**). At this point, the patient had no specific neurological condition, except for a mild orthostatic headache. The patient was readmitted, and a whole-spine MRI myelography was conducted under suspicion of persistent SIH, which revealed definite CSF leakage at the left T5 nerve root sleeve (**FIGURE 5**). Initially, a targeted autologous

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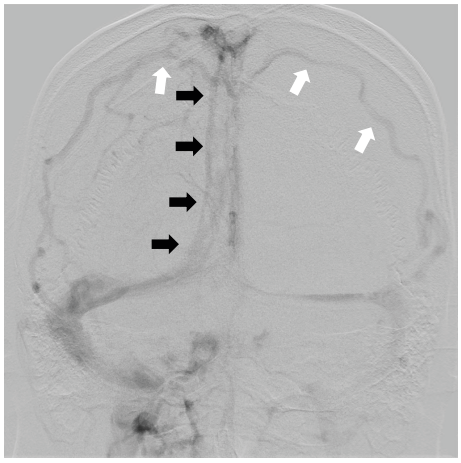

FIGURE 2. Venous phase of right internal carotid artery angiography finding. Black arrows indicate cerebral venous thrombosis in the superficial sagittal sinus. White arrows indicate some collateral venous flow from the right bridging vein to the left bridging vein.

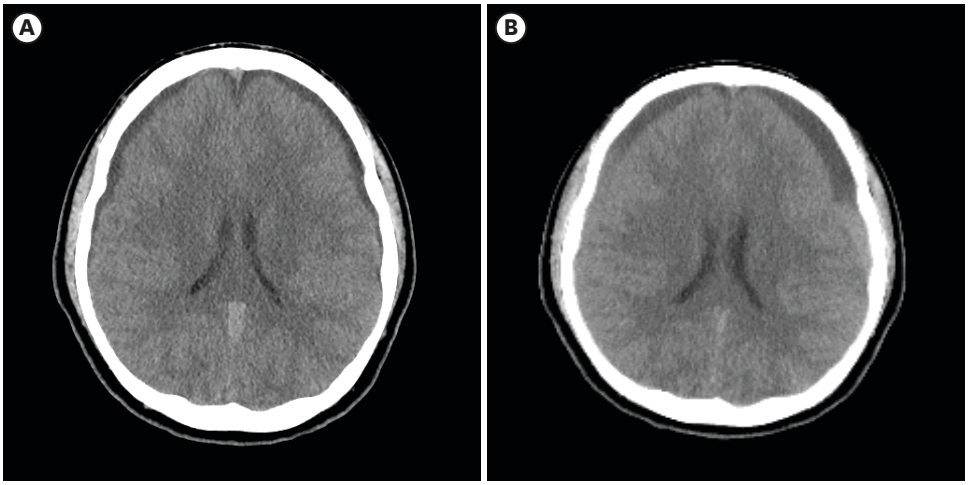

FIGURE 3. (A) On CT of week 1, bilateral subdural effusion was noted. (B) CT imaging after patient's mental deterioration 4 weeks after admission. Bilateral chronic subdural hematoma was compressing both lateral ventricles. CT: computed tomography.

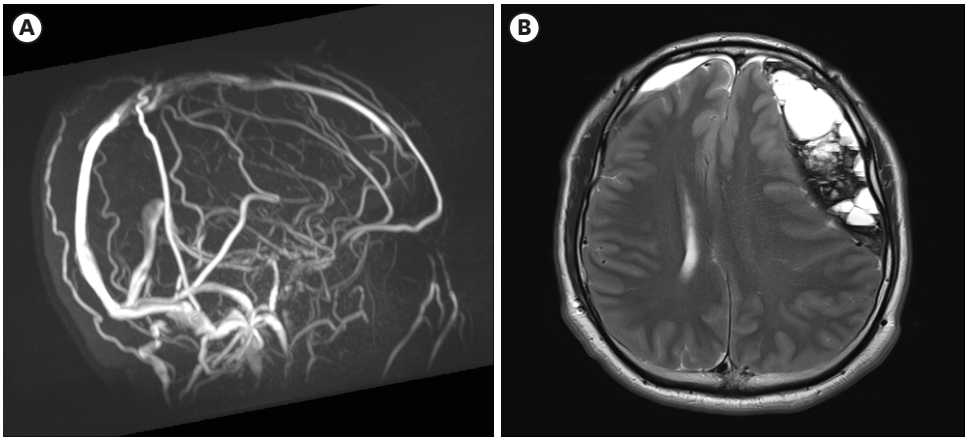

FIGURE 4. (A) Two months follow-up magnetic resonance imaging scan shows improvement in cerebral venous thrombosis, (B) newly occurred subdural hematoma in the left cerebral hemisphere.

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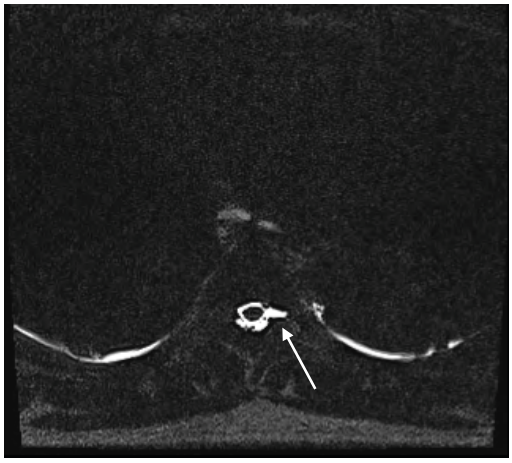

FIGURE 5. Whole spine magnetic resonance imaging myelography shows definite cerebrospinal fluid leakage in the left T5 nerve root sleeve (white arrow).

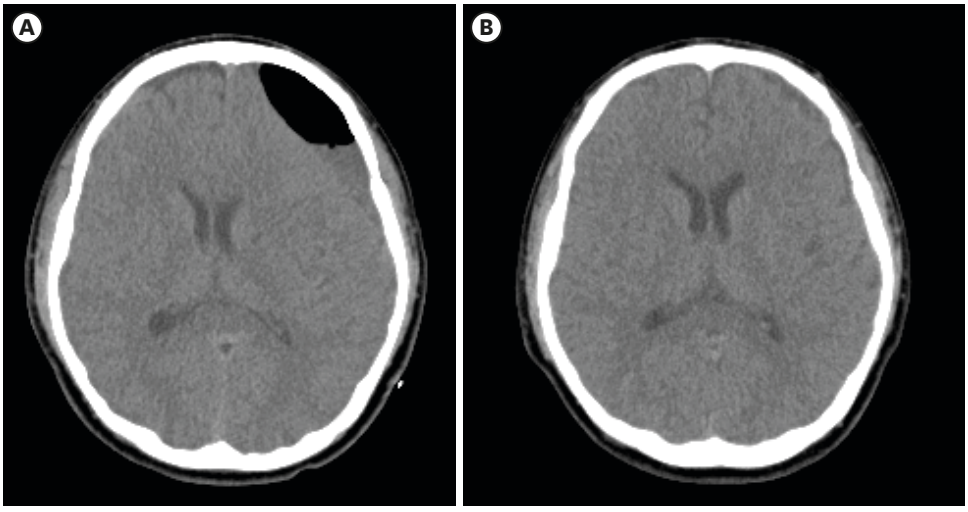

FIGURE 6. (A) Postoperative brain CT scan showing resolved chronic SDH in left cerebral hemisphere. (B) One-month brain CT scan showing near-completely improved state.
CT: computed tomography.

epidural blood patch was administered, after which, the patient's orthostatic headache improved significantly. Burr hole drainage of the left-sided chronic SDH was performed simultaneously. Serial follow-up brain CT showed no recurrence of SDH (**FIGURE 6**), and the patient was discharged. Currently, the patient is being followed up in the outpatient department for 3 months and is in a symptom-free state without any neurological deficiency.

DISCUSSION

CVT is a rare type of cerebrovascular disease, defined as thrombosis of the intracranial veins and sinuses, accounting for 0.5% of all strokes.[3] Common causes include deep vein thrombosis in the legs, genetic and acquired prothrombotic disorders, hematological diseases, infections, pregnancy and puerperium, certain drugs, in particular oral contraceptives, hormonal replacement therapy, and local causes such as brain tumors, arteriovenous malformations, and head trauma.[3] SIH is a condition that typically results

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from CSF leakage.[7] Intracranial hypotension can be caused by conditions such as a hypovolemic state, and traumatic and spontaneous CSF leaks.[7] Spontaneous CSF leak is the major cause of SIH and includes various etiologies such as meningeal diverticula, ectasia of the dural sac, joint hypermobility, and Marfan syndrome.[7] SIH is a risk factor for CVT, and approximately 2% of SIH patients develop CVT.[8]

CVT presents with various symptoms, including headaches, focal neurologic deficits, and/or partial seizures to altered consciousness.[1] Therefore, neuroimaging is essential for the diagnosis of CVT. MRI with magnetic resonance (MR) venography, CT venography, and catheter angiography are commonly used imaging methods. MRI is the most widely used technique and reveals direct visualization of the thrombus with absent flow on MR venography.[4,5]

Clinical complications of CVT vary from focal neurologic deficits, such as hemiparesis, dysphasia, and blindness, to severe and life-threatening complications such as hemorrhagic infarctions.[2] Cerebral hernia is the most common cause of death in patients with CVT. Therefore, its early detection and treatment is critical for patients with CVT. Anticoagulant therapy is the initial treatment for CVT. Both low-molecular-weight heparin and unfractionated heparin can be used to treat CVT. Oral anticoagulants can be administered thereafter.[6] Intravascular treatments such as chemical thrombolysis or mechanical thrombectomy may be required.[6] NOACs can also be used for the treatment of CVT. A recent multicenter study described that administering NOACs to CVT patients has fewer complications such as intracranial hemorrhage or gastrointestinal bleeding, compared to the use of warfarin, with similar therapeutic effects.[9]

Generally, the period for anticoagulation therapy for CVT is known to be: 3–6 months for CVT related to a transient risk factor, 6–12 months for idiopathic CVT, and indefinitely for recurrent or severe hereditary thrombophilia.[3] It is difficult to treat the combined condition, i.e., thrombosis and hematoma, as in this case. CVT requires immediate anticoagulation treatment, whereas anticoagulants must be suspended for blood patch procedures or burr hole operation. In our case, we initiated intravenous anticoagulation for one week, changed that to NOAC (apixaban), and stopped anticoagulation treatment just before the blood patch and burr hole procedures.

Early diagnosis is critical for the treatment of CVT and SIH; therefore, it is necessary to clinically suspect these conditions in such patients. Furthermore, personalized treatment strategies seem to be helpful in reducing complications. However, early detection and customization of treatment are difficult because of the low prevalence and varying symptoms of CVT and SIH. Therefore, attention should be paid to the concept of this disease and typical examples of treatment options. In this rare case, we achieved successful results after short-term anticoagulation treatment (4 weeks), followed by correction of the underlying cause of SIH. We believe that in the treatment of CVT with definite etiology, early suspension of anticoagulants for correcting the etiology of CVT should be considered. Furthermore, the use of NOACs in patients with CVT can be considered as an alternative. However, this single case report makes it insufficient to generalize our case, and further multicenter, prospective clinical research may be helpful.

CONCLUSION

We present a successfully treated case of CVT accompanying repeated SDH caused by SIH using short term anticoagulation and blood patch.

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