

Successful venous thrombectomy for extensive cerebral venous and sinus thrombosis after failed diagnostic lumbar puncture

Erfolgreiche venöse Thrombektomie nach ausgedehnter Sinus- und Hirnvenenthrombose

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ABSTRACT

Purpose With an incidence of 2–5 per million adults, cerebral venous and sinus thrombosis (CVST) is a rarity in the spectrum of cerebrovascular diseases. The etiology and symptomatic presentation are heterogeneous and diverse. CVST is, therefore, often underdiagnosed. In addition to therapeutic anticoagulation, venous thrombectomy is a last-resort therapy in individual cases.

Case Description A 42-year-old woman was admitted to the hospital with postural headache following an unsuccessful lumbar puncture. On suspicion of post-puncture syndrome, analgesic therapy with adjuvant caffeine tablets was initially chosen, leading to rapid improvement and short-term discharge. However, only one day later, the patient presented with a significant worsening of her symptoms and new onset

of paresthesia. Since distally accentuated paresis of the left arm also occurred within a very short period of time, a cerebral MRI examination was performed. This revealed an extensive CVST. Due to the extensive CVST and progressive worsening of symptoms, mechanical venous thrombectomy was discussed and performed on an interdisciplinary basis. Mechanical thrombectomy was successful and resulted in complete improvement of symptoms.

Discussion The pathophysiological diagnosis is cerebrospinal fluid leak syndrome after unsuccessful lumbar puncture. According to the Monroe-Kellie doctrine, the loss of CSF leads to compensatory dilatation and venous stasis, which in combination can lead to venous insufficiency and promote prothrombotic conditions. Although the available data on the performance of venous thrombectomy suggest caution, there should always be sufficient discretion for individual decision making. As our experience shows, there is a chance of successful venous thrombectomy in the early phase of CVST.

Key Points

- CVST is a rarity in the spectrum of cerebrovascular diseases.
- The etiology and symptomatic presentation are heterogeneous and diverse.
- A rare cause is the loss of CSF.
- Mechanical venous thrombectomy is a possible treatment option.

Citation Format

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ZUSAMMENFASSUNG

Ziel Mit einer Inzidenz von 2–5 pro Million Erwachsener ist die Hirnvenen- und Sinusthrombose (CVST) eher ein Kolibri im Spektrum der zerebrovaskulären Erkrankungen. Die Ätiologie und Symptomatik sind heterogen und vielfältig. Die CVST wird daher häufig unterdiagnostiziert. Neben der therapeutischen Antikoagulation ist in Einzelfällen auch die venöse Thrombektomie eine Ultima Ratio.

Fallbeschreibung Bei lageabhängigen Kopfschmerzen nach frustraner Lumbalpunktion erfolgte die stationäre Aufnahme

einer 42-jährigen. Bei Verdacht auf ein Postpunktionssyndrom entschied man sich initial für eine analgetische Therapie mit unterstützender Gabe von Koffeintabletten, was zu einer raschen Besserung und einer kurzfristigen Entlassung führte. Bereits einen Tag danach wurde die Patientin jedoch mit einer deutlichen Verschlechterung der Symptome neu aufgetretener Parästhesien erneut vorstellig. Da binnen kürzester Zeit zudem eine distal akzentuierte Parese des linken Arms auftrat, erfolgte eine zerebrale Bildgebung mittels MRT. Diese bot das Bild einer ausgedehnten CVST. Aufgrund der ausgedehnten CVST und der fortschreitenden Verschlechterung der Symptome wurde interdisziplinär eine mechanische Venenthrombektomie diskutiert und durchgeführt. Die mechanische Thrombektomie war erfolgreich und führte zu einer vollständigen Verbesserung der Symptome im Sinne einer restitutio ad integrum.

Diskussion Die pathophysiologische Diagnose lautet Liquor-Leck-Syndrom nach frustrierter Lumbalpunktion. Nach der

Monroe-Kellie-Doktrin führt der Liquorverlust zu einer kompensatorischen Dilatation und venösen Stase, die in Kombination zu einer venösen Insuffizienz führen kann und prothrombotische Zustände fördert. Obwohl die verfügbaren Daten zur Durchführung der venösen Thrombektomie zur Vorsicht raten, sollte immer ein ausreichender Ermessensspielraum für die individuelle Entscheidungsfindung bestehen. Wie unsere Erfahrung zeigt, besteht eine Chance auf eine erfolgreiche venöse Thrombektomie in der Frühphase der CVST.

Kernaussagen

- Die CVST ist eher ein Kolibri im Spektrum der zerebrovaskulären Erkrankungen.
- Die Ätiologie und Symptomatik sind heterogen und vielfältig.
- Eine seltene Ursache ist der Verlust von Liquor.
- Die mechanische venöse Thrombektomie ist eine mögliche Behandlungsoption.

Abbreviations

ADC	Apparent diffusion coefficient
CCT	Cerebral computed tomography
cMRI	Cranial magnetic resonance imaging
CSF	Cerebrospinal fluid
CVST	Cerebral venous and sinus thrombosis
DGN	German neurological society
DSA	Digital subtraction angiography
EEG	Electroencephalography
F	French
MRA	Magnetic resonance angiography
MIP	Maximum intensity projection
mRS	Modified Rankin Scale
ToF	Time-of-flight
SE	Status epilepticus
SRMA	Seizure-induced reversible MRI abnormalities

Background

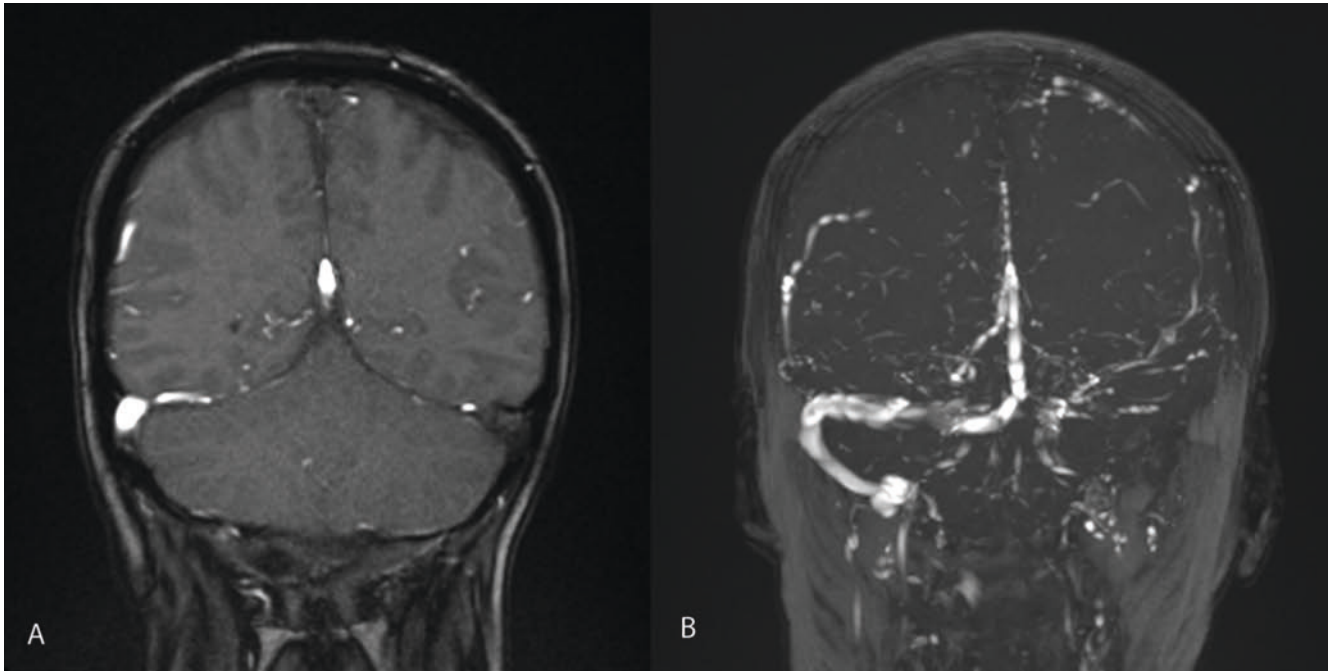
Cerebral venous and sinus thrombosis (CVST) is rare and, with an incidence of 3–5 per million, represents a small percentage of all cerebrovascular diseases worldwide [1, 2]. CVSTs are predominantly idiopathic, associated with sepsis or with coagulation disorders such as factor V Leiden mutation, prothrombin mutation, antithrombin deficiency, antiphospholipid antibody syndrome. There are also gender-specific predisposing factors, including use of contraceptives, pregnancy, and puerperium. CVST is rarely associated with intracranial hypotension or lumbar puncture [3, 4]. The clinical presentation of CVST is highly variable and depends on the extent and location of the venous occlusion and the availability of collateral venous drainage [5, 6]. The most common symptom is headache associated with hemiplegia, hemiparesis, or hemianopsia. Seizures are another rare clinical manifesta-

tion, especially in young patients. Imaging with cranial magnetic resonance imaging (cMRI) or cerebral computed tomography (CCT) often shows congestive hemorrhages and/or congestive infarcts [3, 4, 5, 6, 7].

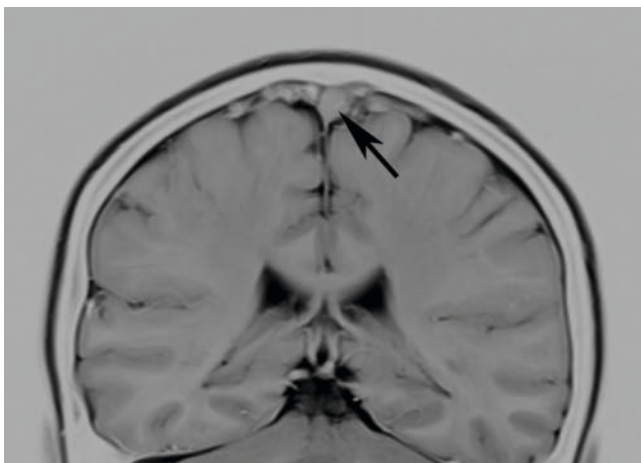
This is a rare case of CVST in an adult patient following a failed lumbar puncture. Mechanical venous thrombectomy was performed due to progressive worsening of symptoms, which completely resolved after mechanical thrombectomy.

Case Presentation

A 42-year-old woman presented to the emergency department with post-puncture syndrome after a failed diagnostic lumbar puncture. Clinically, she reported a positional headache that increased in quality and intensity in the upright position and decreased in the supine position. Initial cMRI revealed dural thickening and bilateral subdural hygroma. The patient was admitted to hospital and received analgesic therapy as well as supportive caffeine tablets. Following a rapid improvement in her symptoms, she was discharged after two days. However, just one day later, the patient presented again to the emergency department with worsening headache and new-onset paresthesia in both upper extremities and paralysis of the left arm. Cerebrospinal fluid hypotension syndrome was suspected, and the patient was admitted to the hospital and scheduled for blood patch therapy. However, the patient developed progressive arm paralysis and hyperreflexia of the lower extremities. Emergency cMRI was performed and showed extensive cerebral sinus venous thrombosis involving the entire superior sagittal sinus as well as left transverse and sigmoid sinus (▶ Fig. 1, ▶ Fig. 2). The cortical cerebral veins were also thrombosed (e.g., ▶ Fig. 3). In addition, diffusion-weighted imaging (DWI) showed an abnormally high signal in the right central region with a correlating decrease in the apparent diffusion coefficient (ADC) signal (▶ Fig. 4). The patient was admitted to the stroke unit and started intravenous therapy with weight-adjusted



► **Fig. 1** Venous ToF-MRA (A) with MIP reconstruction (B) in the coronary incision. Flow recesses are seen in the superior sagittal sinus and the transverse and sigmoid sinuses up to the left jugular vein bulb.



► **Fig. 2** Extensive venous thrombosis involving the cortical cerebral veins (black arrow).

heparin. The next morning, the patient developed left-sided hemiplegia and generalized seizures. A CCT scan showed supratentorial brain swelling. After a multidisciplinary team discussion, venous thrombectomy was carried out.

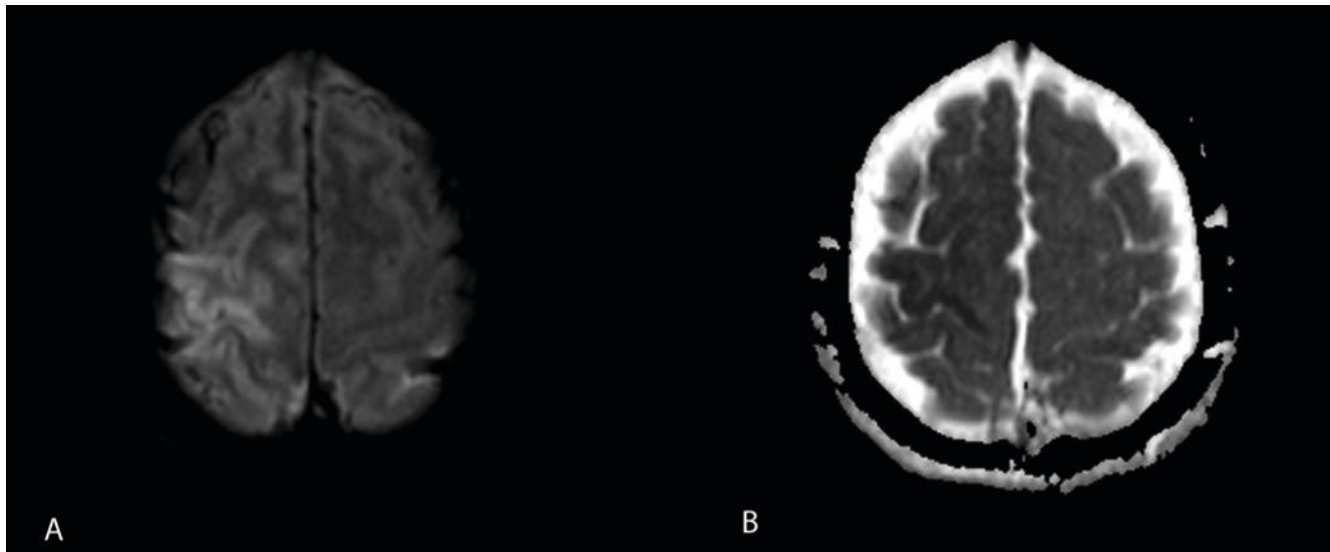
Mechanical Thrombectomy

The intervention was performed by the author. A 4French (F) sheath was advanced into the left femoral artery and a 4F catheter was placed in the left common carotid artery. On the right side, an 8F sheath was advanced into the femoral vein and an 8F guiding catheter was advanced into the left internal jugular vein. A coaxial

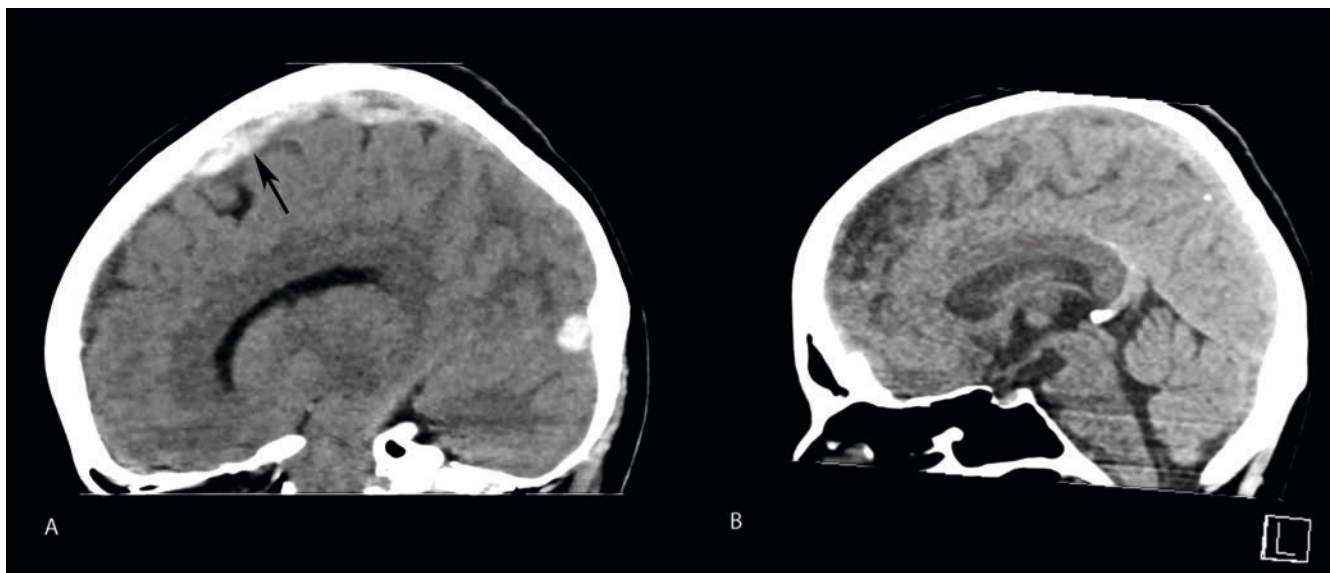
system including a 6F aspiration catheter with a standard 14 microwire as well as a 21 microcatheter was advanced into the left sigmoid sinus. The intracranial digital subtraction angiography (DSA) examination showed an extensive venous sinus thrombosis as described above. Venous thrombectomy was performed by advancing the microcatheter system and aspiration catheter into the sigmoid sinus, the left transverse sinus, as well as the superior sagittal sinus. After removal of the microcatheter and microwire, the aspiration catheter was withdrawn several times under continuous aspiration and a large amount of thrombus material was retrieved. The final DSA run showed complete recanalization of the entire previously occluded venous sinus. Some of the superficial cortical veins were not revascularized and remained partially occluded. No complications were observed on the post-interventional CCT scans (► **Fig. 5**, ► **Fig. 6**). The patient's clinical condition improved rapidly after the procedure and the subsequent hospital stay was uneventful. An electroencephalography (EEG) performed one day after the procedure showed no evidence of increased cerebral excitability, but intermittent right temporo-occipital brain dysfunction (delta slowing) was documented. After hematological workup and exclusion of hereditary or acquired thrombophilia, the patient was discharged without neurological deficit and oral anticoagulation was prescribed. At the 30-day follow-up after discharge, the patient had no neurological deficits.

Discussion

Our case demonstrates the safety and efficacy of venous mechanical thrombectomy in the treatment of heparin-refractory CSVT caused by failed lumbar puncture and CSF leak. As in this case, CSVT has a latency period [7]. Six days elapsed between the first



► **Fig. 3** A diffusion-weighted sequence was also performed as part of the rapid worsening of the findings and the first seizure event. This showed sub/cortical DWI signal increases (A) with correlating flat ADC signal decreases (B) in the right central region. These changes were no longer visible on follow-up imaging – i. e., completely reversible – and are therefore most likely to be interpreted as postictal changes during the seizure activity.

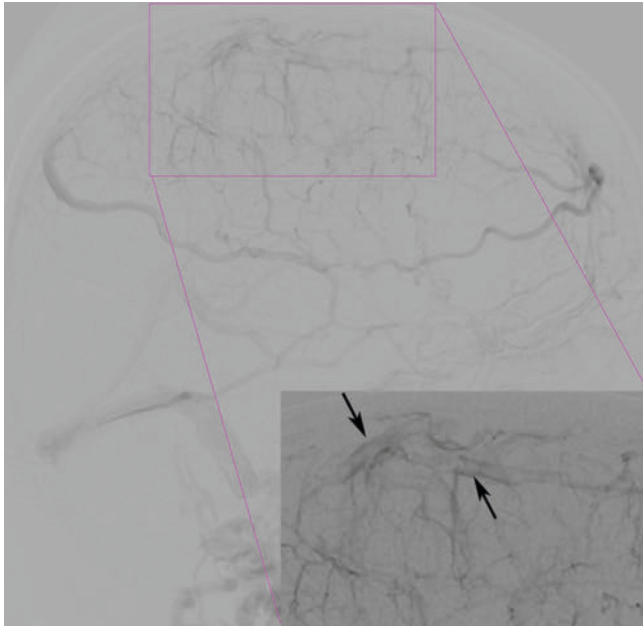


► **Fig. 4** Analogous to the MRI, this CCT also shows the cortical and cerebral venous thrombosis over long distances, which is caused by the dense vein sign in the area of the superior sagittal sinus and the left transverse sinus (A). On the postinterventional control CCT, the corresponding venous blood vessels are permeable again and the dense sign is therefore no longer visible (B).

clinical manifestation of symptoms and the onset of neurological deficits. This could be explained by the recruitment of collateral venous drainage before congestion of the venous system occurs. Due to the rapid clinical deterioration, extensive thrombosis of the venous system, and MRI findings including diffusion restriction in the right central region (► **Fig. 4**), endovascular thrombectomy was performed as a bail-out procedure. Mechanical thrombectomy and intravenous lysis for intracranial venous thrombosis are not currently standard of care according to the German Neurological Society (DGN) guidelines [4], which are based on the results of the TOACT trial [8]. This open-label, multicenter, blinded, randomized trial

compared patients receiving standard therapy for CVST with an intervention group. While the intervention group received either mechanical thrombectomy or local sinus application of alteplase or urokinase, or a combination of both procedures, the control group received only the standard treatment of therapeutic heparinization. When the cohorts were compared, both groups showed similarly good results in terms of mRS score at 12 months after CSVT [8]. There was also no statistically significant difference between the groups in terms of mortality and the incidence of symptomatic intracerebral hemorrhage [9]. A review by Ilyas et al., which included 17 studies with 235 patients, found that patients who underwent

mechanical thrombectomy tended to be more severely affected and had a higher intracranial thrombus burden, resulting in a less favorable outcome [8]. There is also evidence from an American retrospective study that a hormonal cause of CVST – such as in the postpartum state – is a predictor of a favorable outcome after mechanical thrombectomy [10].



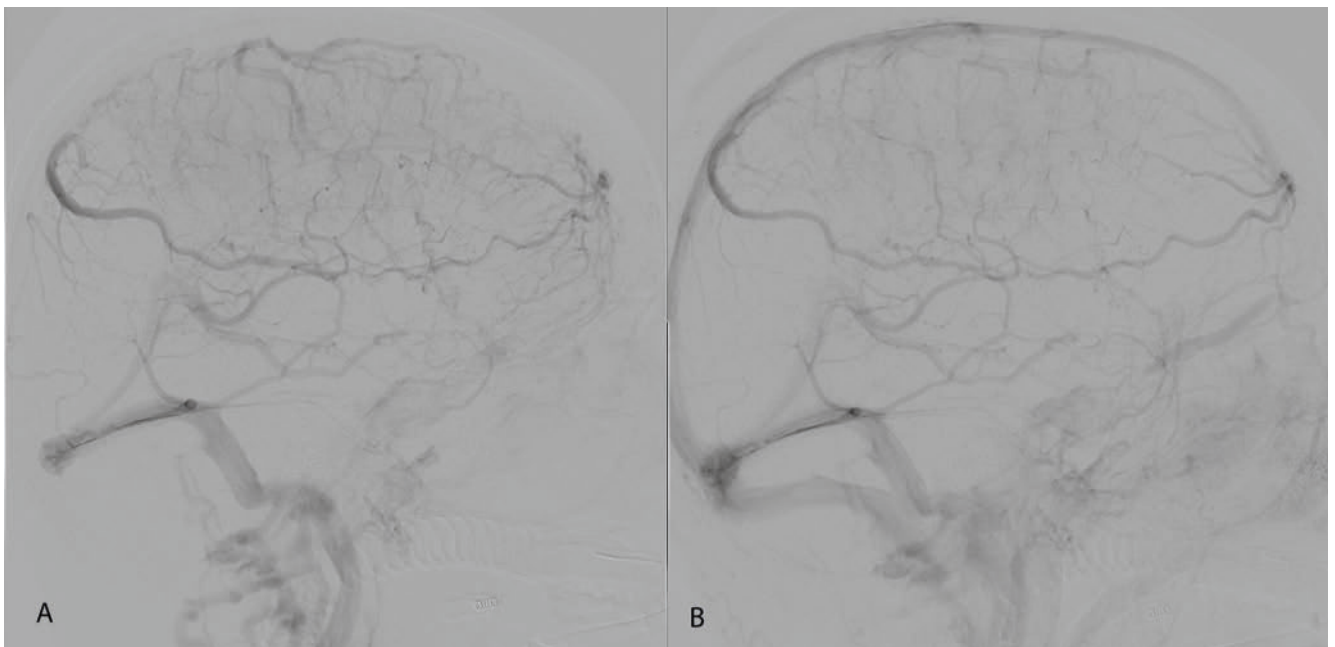
► **Fig. 5** Pre-interventional catheter angiography showing the extent of CVST. Enlargement of the encircling thrombi in the cortical veins (black arrows).

According to the Monro-Kellie theory, the loss of CSF causes a compensatory increase in venous blood volume, leading to dilatation of the cerebral venous system and slowing and stagnating venous blood flow, thus favoring thrombosis [10]. Kranz et al [11] found that patients with low intracranial pressure had dilated cerebral veins with a cross-sectional area 70% greater than the corresponding area in healthy subjects. The theory of abnormal CSF buoyancy states that the loss of CSF buoyancy leads to sagging of the intracranial tissue structure, resulting in a disturbance of venous hemodynamics, sometimes with stagnation of venous blood flow [12]. It has also been argued that CSF loss reduces CSF absorption by the venous system, which in turn leads to increased viscosity and hypercoagulability of the blood in the cerebral venous system, thus increasing the risk of thrombosis [12]. Canhao et al. found an average decrease in flow velocity of 47% in the superior sagittal sinus after puncture using Doppler sonography [3].

The MRI examination performed at the time of the first seizure and hemiparesis showed diffusion restriction in the right central region as mentioned above. These changes resolved completely on follow-up imaging, consistent with the patient's clinical improvement. Such changes were, therefore, transient and most likely seizure-related.

Conclusion

In our case, venous thrombectomy was performed as a salvage therapy for the treatment of heparin-refractory CVST caused by failed lumbar puncture and CSF leak. Complete, efficient, and safe recanalization of the sinus system was achieved, resulting in rapid relief of the congested venous system and complete resolution of the clinical symptoms. Although venous thrombectomy is



► **Fig. 6** Pre- (A) and post-interventional (B) sagittal plane DSA. Lack of contrast in the sagittal sinus, all cerebral veins, and the sinus confluence as well as the left transverse and sigmoid sinus up to the ipsilateral jugular vein bulb. After successful venous thrombectomy, contrast enhancement is again seen in these areas.

not yet standard of care, it could be considered as a treatment option in patients with deteriorating clinical symptoms and extensive venous thrombosis, especially in the early phase.

Conflict of Interest

The authors declare that they have no conflict of interest.

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