Journal of Taibah University Medical Sciences (2018) 13(1), 87–92

Taibah University

Journal of Taibah University Medical Sciences

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Case Report

Endovascular suction thrombectomy for severe cerebral venous sinus thrombosis: A report of two cases

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Received 13 January 2017; revised 16 June 2017; accepted 29 June 2017; Available online 21 July 2017

الملخص

تخثر الجيوب الوريدية الدماغية هي حالة سريرية خطيرة، تتظاهر بأعراض مختلفة، يمكن أن تكون خفيفة على شكل صداع، أو يمكن أن تكون مهددة للحياة مع ارتفاع الضغط والفتق داخل الجمجمة. وحتى مع العلاج الوريدي بمضادات تخثر الدم، لا يزال تخثر الجيوب الوريدية الدماغية يحمل خطر الوفاة عند حوالي ١٠٪ بسبب فشل إعادة استقناء الوريد بمضادات التخثر لوحدها. نعرض هنا حالتين لتخثر واسع للجيوب الوريدية الدماغية، وعلى الرغم من العلاج الوريدي بمضادات تخثر الدم تدهورت حالتهما العصبية تدريجيا. عولج المريضان بنجاح بإزالة الجلطة من داخل الأوعية الدموية باستخدام تقنيات استئصال الخثرة بالشفط مع إعادة فورية لتروية الجيوب الوريدية وسرعة التحسن الشعاعي. العلاج الموصوف يبدو فاعلا وآمنا، ولكن هناك حاجة لدراسات أخرى للتحقق من فاعلية هذا العلاج.

الكلمات المفتاحية: منع تخثر الدم؛ تخثر الجيوب الوريدية الدماغية؛ داخل الأوعية الدموية؛ استئصال الخثرة؛ تحلل الجلطة؛ النتيجة

Abstract

Cerebral venous sinus thrombosis (CVST) is a severe clinical condition that manifests with diverse symptoms that can be mild, taking the form of a headache, or can be life-threatening, with raised intracranial pressure and herniation. Despite systemic anticoagulation treatment, CVST still carries a mortality risk of approximately 10% because of the failure of venous recanalization by anticoagulants alone. This paper describes two cases with extensive CVST who presented with progressive neurological deterioration despite adequate systemic anticoagulation treatment. Both patients were successfully treated with endovascular clot removal using suction thrombectomy techniques with immediate recanalization of the sinuses and rapid radiographic improvement. The described therapy appears effective and safe; however, further studies are needed to validate the effectiveness of this treatment.

Keywords: Anticoagulation; Cerebral venous sinus thrombosis; Endovascular; Outcome; Thrombectomy; Thrombolysis

Introduction

Traditionally, cerebral venous sinus thrombosis (CVST), previously considered a rare diagnosis, is increasingly detected owing to the advent of modern imaging techniques.[1] The treatment for this potentially fatal condition has largely been anticoagulation. However, in few refractory cases or those with contraindication to anticoagulation, endovascular thrombolysis has been used in experienced centres.

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Peer review under responsibility of Taibah University.

Production and hosting by Elsevier

 http://dx.doi.org/10.1016/j.jtumed.2017.06.004

Approved by the Food and Drug Administration since 2006, the Penumbra Stroke system, a clot retrieval technology, has been used for the revascularization of patients with acute ischaemic stroke.[2] Its use in the management of cerebral sinus thrombosis is limited to tertiary care centres.

We report 2 cases with CVST successfully treated using manually aspiration thrombectomy using the Penumbra System and resulting in rapid radiographic improvement associated with a complete clinical recovery.

Case reports

Case 1

A 32-year-old female, not known to have a chronic medical illness, presented with a thunderclap headache and progressive deterioration of her level of consciousness. She was not pregnant or on birth control pills and was not known to have a hypercoagulable state. There was no history of trauma or family history of coagulation disorders. When she was first evaluated at another hospital, her Glasgow coma scale (GCS) was 12/15, and she was vitally stable. A computed tomography (CT) scan of the brain revealed diffuse oedema, and a magnetic resonance imaging (MRI) scan showed tiny multiple infarctions in the subcortical frontal lobes bilaterally with extensive thrombosis involving the superior sagittal, left sigmoid, and left transverse sinuses. She was started immediately on intravenous heparin therapy with adequate therapeutic anticoagulation monitoring for three days without improvement. The patient was then referred to our hospital for interventional therapy. Upon admission, she was unconscious with a GCS of 8, and her vital signs were stable. A general physical examination was unremarkable, and her cranial nerves examination was within normal, apart from sluggish pupillary reaction to light and bilateral papilloedema. She had mild withdrawal to painful stimuli on the right side more than the left, and her deep tendon reflexes were exaggerated. Her routine laboratory tests were within normal with the therapeutic heparin effect. Specific haematological and serological tests for an underlying thrombophilia were all within normal limits. The patient was intubated and stabilized, and a repeat brain MRI scan demonstrated diffuse persistent cerebral venous sinus thrombosis (Figure 1A). Magnetic resonance venogram (MRV) scans demonstrated the extensive thrombosis of the sagittal, left transverse and sigmoid sinuses (Figure 1B). The patient was taken to the angiography suite, and cerebral angiography showed an occlusion of the posterior part of the superior sagittal sinus (Figure 2A, B). A 6-French Neuron guide catheter (Penumbra, Alameda, California, USA) was carefully placed into the right transverse sinus, and a 0.41-inch Penumbra thrombectomy catheter was introduced into the occluded sinuses. Thrombolysis using a total of 10 mg of recombinant tissue plasminogen activator (r-TPA) in conjunction with the aspiration of the thrombus was successfully performed over a total of 23 min. The cerebral venogram showed complete reopening of the sinuses (Figure 2C). Within 24 h after endovascular therapy, her level of consciousness almost reached a GCS of 14, and she was extubated. She was discharged home after five days with mild left hemiparesis and continued oral warfarin 4 mg daily with adequate therapeutic anticoagulation monitoring. At the 3-month outpatient follow-up, the residual neurological deficit was completely recovered, and brain MRI and MRV scans showed a significant recanalization of the thrombosed sinuses (Figure 3).

Case 2

A 35-year old female patient, a known case of sickle cell anaemia, presented with a history of gradual onset headache for a duration of approximately 3 weeks that grew worse and reached maximal intensity on the day of her presentation to the emergency room. It was associated with photophobia, vomiting, dizziness and blurry vision. The patient complained of right-sided weakness and an inability to walk without assistance that started on the day of her presentation. There was no history of fever or any other signs or symptoms suggestive of infectious focus. She was non-smoking with no history of using oral contraceptives, and her last blood transfusion was 20 years ago.

In the emergency room, she was lethargic and tachycardic; she had a heart rate of 109 bpm, with blood pressure of 156/88 but afebrile. She was initially communicating and opening her eyes spontaneously with right gaze preference. She then deteriorated clinically, and her GCS dropped to 9/15, opening her eyes to painful stimuli only, with no vocal response and localize to pain as the best motor response,

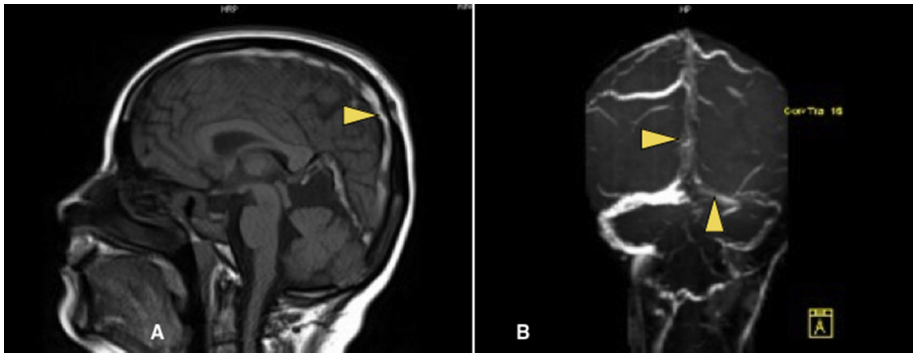

Figure 1: Sagittal T1-MRI (A) and MRV (B) scans demonstrating extensive filling defects in the superior sagittal and left transverse sinuses (arrowheads).

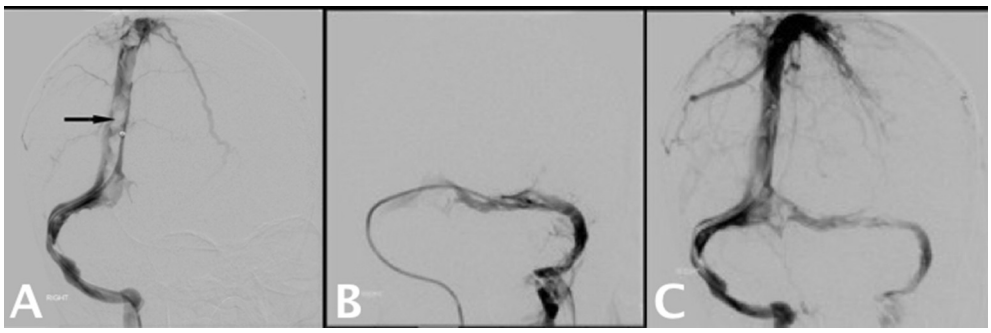

Figure 2: Digital subtraction venography (A, B, C) in the posterior aspect of the superior sagittal sinus showing extensive clots burden and an occluded left transverse sinus. Post-progressive manual suction and clot debulking, venography showing good opacification of the sagittal and transverse sinuses.

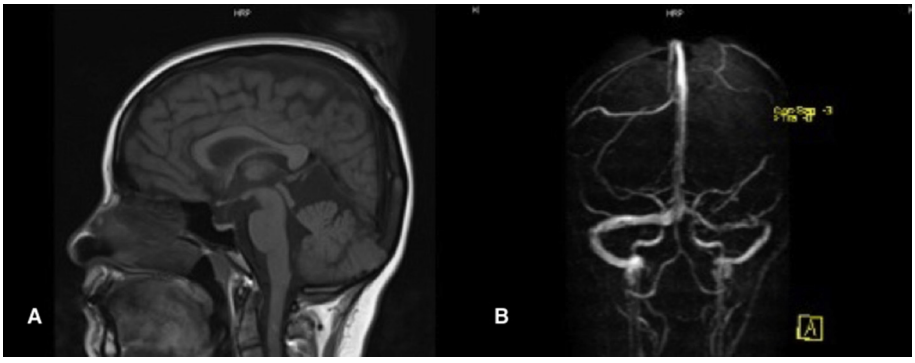

Figure 3: Follow-up sagittal T1-MRI (A) and MRV (B) scan showing complete recanalization of the venous sinuses.

moving the left limb more than the right. She had an intact cranial nerves exam; the fundi could not be visualized.

An initial head CT scan without contrast showed focal cortical haemorrhage at the right high frontal lobe, with a hyperdensity in the superior sagittal and bilateral transverse sinuses suggestive of a dural venous sinus thrombosis (Figure 4), in addition to acute infarction involving the right parietal lobe. Thus, venous infarct was suspected and later confirmed with a CT venogram, which showed extensive thrombosis of the straight sinus and both transverse sinuses. She was started on IV heparin. However, her clinical picture did not improve, and she began to become hypertensive and bradycardia, denoting progressively increasing intracranial pressure. She developed intractable seizures two days after the onset of anticoagulation that was managed medically; she was then intubated. The decision was made to proceed with mechanical thrombectomy. Arterial and venous accesses were obtained to the femoral artery and vein, respectively. Mechanical thrombectomy using manual suction and wire manipulation of the thrombus via a distal access catheter (5-French SOFIA Distal Access Catheter, Microvention, Tustin, California, USA) were used together with a small dose of local r-TPA pulsed at the thrombus interface (total of 12 mg). Several passes were made with the SOFIA catheter until the final venogram showed a good venous

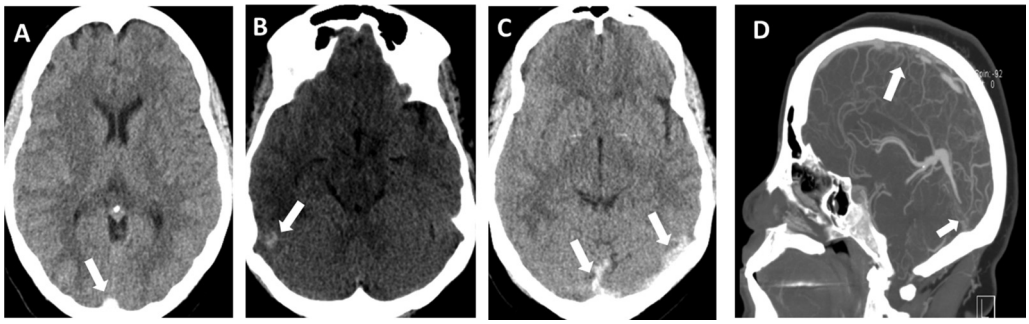

Figure 4: Unenhanced (A, B, C) axial cuts computed tomography (CT) scan images showing the hyperdense superior sagittal (A), right transverse (B) and the straight and left transverse sinuses (C) and CTV venography (D) showing a lack of opacification in the superior sagittal and straight sinuses.

outflow through the occluded sinuses despite some residual thrombus (Figure 5).

Immediately post procedure, the heart rate started to increase, and her blood pressure normalized without any pharmaceutical intervention. Over the next several hours after the procedure, the patient had a gradual clinical improvement in her level of consciousness, and she was successfully extubated the next procedural day. She became awake, oriented with intact speech; however, her headache was still persistent. She was discharged on warfarin for a target INR of 2.0–3.0. At discharge home, she was conscious, alert, oriented, with intact speech and improved power on the right side. During the next 3 months of anticoagulant therapy and follow-up, she returned to her baseline neurological state with no focal deficits and no new symptoms or recurrence.

Discussion

Intracranial venous sinus thrombosis is an important diagnosis to exclude in young to middle-aged patients with a sudden onset of neurological deficit, particularly women. Although it accounts for less than 1% of stroke cases,[2] it has a morbidity of 8.8–44.4% and a mortality of 6–10%.[1] The most common sinus affected is the superior sagittal and lateral sinuses (70%) individually involved in thrombosis, but in up to 30% of cases, both can be involved. The cortical and cerebellar veins can also be involved, although less commonly.[4] CVST can cause increased intracranial pressure, venous infarctions, haemorrhage or haemorrhagic infarction. More than 100 cases have been reported in the scientific literature.[4] However, no cause can be identified in approximately 20–25% of the cases.[4] CVST risk factors are common to other venous thrombotic phenomena including hormonal therapy such as birth-control pills, corticosteroids, and conditions promoting venous hypercoagulability such as pregnancy and cancers. Common presenting symptoms include a headache, seizures, paresis, an altered level of consciousness, visual disturbance, or papilloedema. Untreated cases will go into parenchymal oedema with venous infarction and haemorrhage in up to 50% of cases. CT or venography and digital subtraction angiography can be used individually or in various combinations to confirm the diagnosis. The outcome is highly variable, with death most often due to transtentorial herniation. Indicators of poor outcome include papilloedema, presentations with reduced GCS, coma, refractory seizures, age extremes, delayed presentation or diagnosis of more than ten days, the presence of haemorrhage, the involvement of the deep venous system or the straight sinus, increased intracranial pressure and the presence of an underlying cancer or sepsis.[4–6]

Treatment regimens include anticoagulation and, in rare circumstances, endovascular intervention or surgical decompression. Intravenous or subcutaneous anticoagulation using heparin and bridged by oral anticoagulants remains the first line of management even in the setting of intracerebral haemorrhage.[8] Anticoagulation is typically continued for three months to indefinitely, depending on the cause.[4] Fortunately, anticoagulants are all that is required in the majority of patients. Operative neurosurgical strategies are directed mainly to control increased intracranial pressure. These include external ventricular draining, ventriculoperitoneal shunting or decompressive craniectomy.[6] The novel use of the endovascular technique entails either performing direct mechanical thrombectomy using a rheolytic catheter or injecting a local thrombolytic agent.[7] The operative approach is attractive, particularly in patients who are rapidly deteriorating.[9]

The experience with venous thrombolysis dates back over 30 years using urokinase.[10] It has been advocated for use in children in some papers.[11] A particular difficulty in the evaluation of thrombolysis is that heparin is part of that treatment and is not readily separated. Thrombolysis can be systemic or local. Systemic intravenous thrombolytics have been tried but are seldom used.[6,12] Recently, pharmacological and pharmaco-mechanical local thrombolysis has been introduced. The use of thrombolysis is indicated in patients who deteriorate despite adequate heparinization and the exclusion of seizure activity and in those who do not improve despite a week of anticoagulation therapy.[2,4,6] However, evidence supporting its use for cerebral vein thrombosis is lacking.[3,8,6,12] However, its use along with endovascular thrombectomy can be considered in the presence of patients at high risk for malignant intracranial hypertension such as those with coma. For those with a mildly reduced level of consciousness, e.g., GCS $>$ 12, thrombolysis thrombectomy might be considered only after a failed attempt of anticoagulation. This scheme has been proposed in very few papers.[1] The recent advances in endovascular technology, local thrombolysis, and

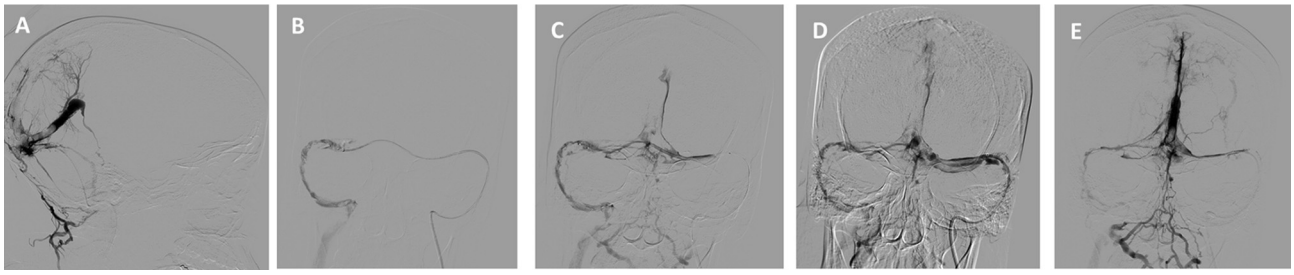

Figure 5: Digital subtraction venography lateral view (A) showing irregular partial filling of the straight and lack of opacification of the superior sagittal sinus. Frontal views (B, C, D) showing the distal access catheter crossing from the left to the right transverse sinus with continuous manual suction and clot debulking (E).

thrombectomy are increasingly used. This approach has the advantage of quick clot lysis and a rapid restoration of blood flow, with a subsequent reduction in ICP, and clinical improvement. On the other hand, it should be borne in mind the small but potentially fatal risks associated with these procedures such as haemorrhage, vascular injury or perforation, and pulmonary emboli. The current practice of direct thrombolysis is to gain access typically through the femoral vein[12] and to deliver the treatment at the thrombus interface using coaxially manipulated catheters. The objective is complete or near complete recanalization. Local r-TPA administration is preferred over other thrombolytic agents because it has a less systemic effect, is not antigenic, has a short half-life of 4–5 h, and produces fewer fibrinogen degradation products and a faster recanalization rate.[12] A recent paper has reported the safe and successful use of the Penumbra System in a case series of four patients with dural sinus thrombosis.[8] Another report has also described the use of the system in a case of extensive cerebral sinus thrombosis with good results.[12] Pharmacomechanical thrombolysis combines pharmacological thrombolysis with a mechanical fragmentation of the clot to increase the surface area exposed to the thrombolytic agent. The mechanical devices reported include balloon catheters, micro snare devices, a saline jet vacuum device (rheolytic catheter), balloon angioplasty, a Merci device, and a Solitaire stent.[13–18] Recently, a case report using a Pronto V3 catheter, a peripheral vascular catheter, for the management of CVST has been published.[19,20] These devices, specifically designed for intracranial circulation, have overcome many of the limitations and complications associated with the use of coronary angioplasty balloons. The Penumbra System (Figure 6) is a suction-based system that uses a catheter supplemented by a separator.[2] The combination macerates the thrombus and removes the pieces via an aspiration source. It can easily navigate the cerebral venous anatomy. The separator is an atraumatic, thin wire-like device that breaks the clot under the continuous suction of the clot debris via the reperfusion catheter. Its successful use in cerebral venous thrombectomy has been described in a handful of prior reports in adults[21–23] and children.[24] In one of our cases, the Penumbra System was used within ten days of presentation with rapid recanalization in 2 days. Manual suction has been previously reported with arterial thrombectomy.[25] However, the same principle would still apply in venous thrombi, as illustrated in our second case, as r-TPA was used with the suction device without balloon angioplasty. This promising approach for CVST treatment should be tempered by the careful consideration and understanding of its potential risks such as intracerebral haemorrhage and vessel injury.

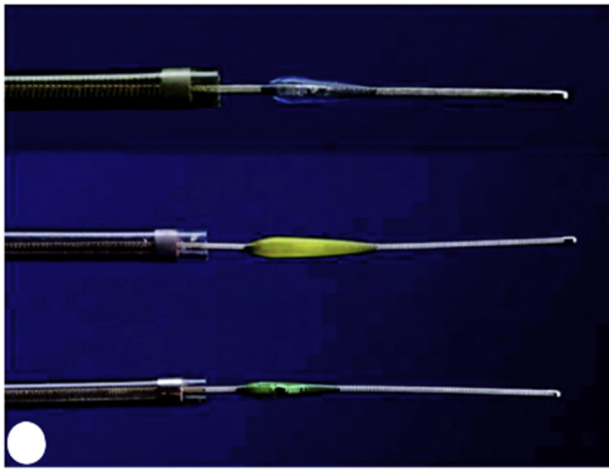

Figure 6: Penumbra reperfusion catheters with the separator wires.

Conclusion

Extensive CVST, particularly when associated with a decreased level of consciousness without improvement to aggressive systemic anticoagulation, carries a significant mortality. Successful immediate radiographic recanalization using the endovascular approach and aspiration thrombectomy, as shown in our two cases and others in the literature, has proven to be efficient and safe and associated with a good neurological recovery. Further studies in the form of large case series or clinical trials are encouraged to better define the role of endovascular interventions in such severe cases of CVST.

Authors' contribution

The manuscript has been read and approved by the authors. The requirements for authorship have been met by all authors, and each author believes that the manuscript represents honest work. SSB, AAH, AAN, and JKR conceived and designed the study, conducted the research, provided the research materials, and collected and organized the data. KIK and MAA analysed and interpreted the data. AAN and JKR wrote the initial and the final drafts of the article and provided logistic support. All authors have critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript.

Ethical approval

All procedures were in accordance with the ethical standards at our institution and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Conflict of interest statement

The authors have no conflict of interest to declare.

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How to cite this article: Najjar AA, Rasheedi JK, Kurdi KI, Hasan AA, Almekhlafi MA, Baeesa SS. Endovascular suction thrombectomy for severe cerebral venous sinus thrombosis: A report of two cases. J Taibah Univ Med Sc 2018;13(1):87–92.