

SUCCESSFUL TREATMENT OF CEREBRAL VENOUS THROMBOSIS ASSOCIATED WITH BILATERAL INTERNAL JUGULAR VEIN STENOSIS USING DIRECT THROMBOLYSIS AND STENTING: A CASE REPORT

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Cerebral venous thrombosis (CVT) is not uncommon, but CVT associated with bilateral internal jugular vein stenosis (BIJVS) is rare. Bilateral internal jugular vein stenting is also a rare procedure. We report on a patient with CVT associated with BIJVS, who was treated successfully using direct thrombolysis and bilateral internal jugular vein stenting.

Key Words: cerebral venous thrombosis, internal jugular vein stenosis, thrombolysis, stenting
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The outcome of cerebral venous thrombosis (CVT) varies widely, and a poor prognosis is usually associated with papilloedema, deteriorating consciousness, coma, cerebral hemorrhage, old age, and delayed diagnosis [1,2]. Internal jugular vein stenosis may further complicate CVT and result in the worst outcome. We report a case of CVT associated with bilateral internal jugular vein stenosis (BIJVS) that was successfully treated by using a combination of direct intravenous endovascular thrombolysis and bilateral internal jugular vein stenting.

CASE PRESENTATION

A 68-year-old male was sent to the emergency room due to sudden-onset right lower limb weakness associated with

aggravating non-throbbing headache. His medical history revealed hypertension, alcohol consumption and smoking. He was considered to have suffered a minor stroke and was hospitalized in the neurologic ward. Emergency brain computed tomography (CT) failed to detect any lesion.

On the second admission day, the right lower limb weakness rapidly progressed to total right paralysis, followed by right upper limb focal seizure and delirium with a Glasgow coma scale (GCS) score of 12 (E3V4M5). Repeated brain CT revealed a hemorrhagic lesion in the left parietal lobe (Figure 1A). The patient was immediately transferred to the neurologic intensive care unit (NICU), where there was rapid deterioration in consciousness to stupor (GCS score 7: E1V1M5) and the patient began to vomit. Contrast-enhanced brain CT revealed enhancement of the superior sagittal sinus and an empty delta sign (Figure 1B). Immediate magnetic resonance imaging and magnetic resonance venography (MRV) disclosed obstruction of the superior sagittal sinus and right transverse sinus. Cerebral angiography on the third admission day showed stenoses in bilateral distal internal jugular veins (Figure 2A and B) (Table).

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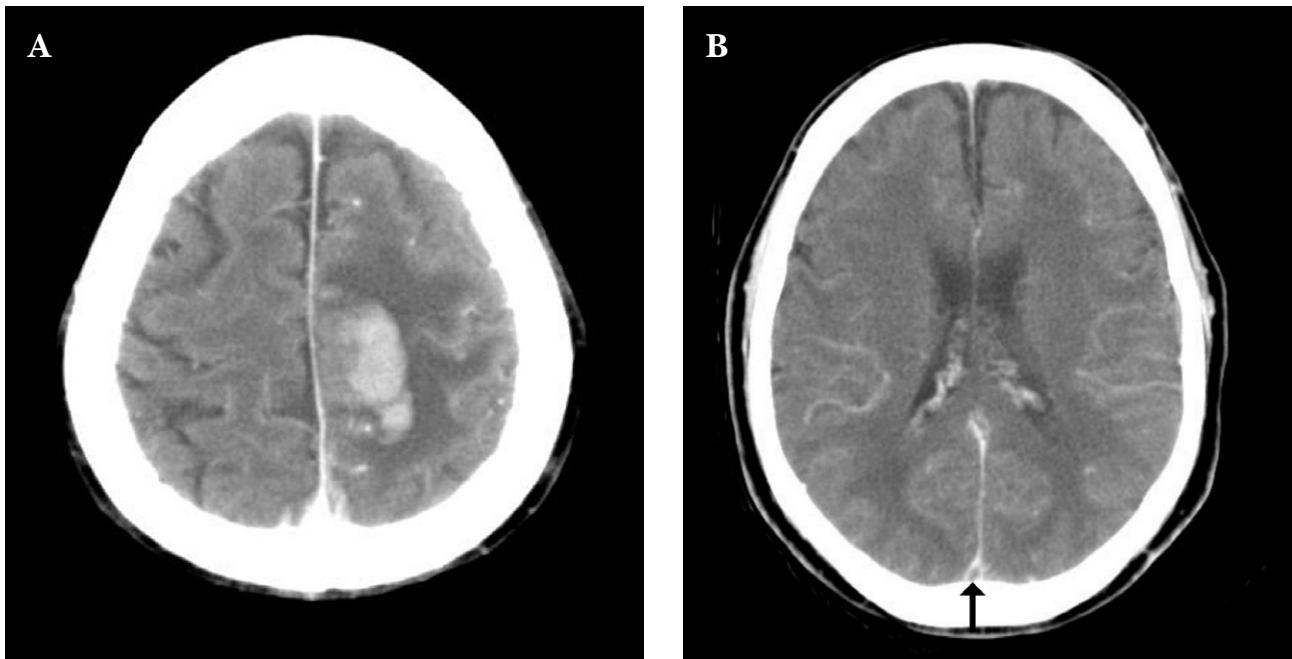


Figure 1. (A) Brain computed tomography (CT) shows hemorrhage in the left parietal lobe with perifocal edema. (B) Enhanced brain CT reveals an empty delta sign (arrow).

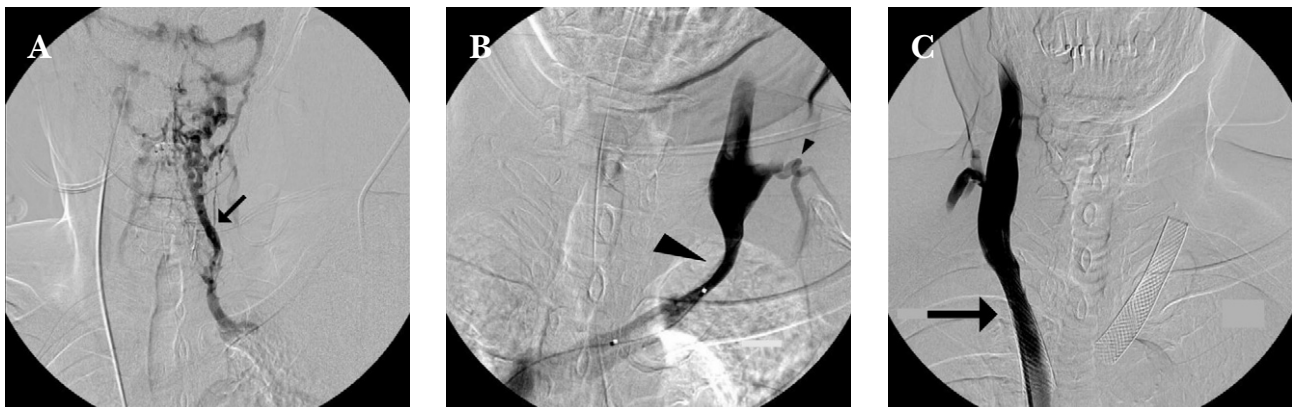


Figure 2. Stenosis in the internal jugular vein: (A) drainage from the external jugular vein (arrow) and (B) stricture in the left internal jugular vein (large arrowhead) and collateral circulation (small arrowhead). (C) Recovery of blood flow in the right internal jugular vein after stenting (arrow).

Table. Devices used in cerebral angiography

Left femoral sheath, 7F/9F (Terumo)		
Guiding catheter	Multipurpose, 7F (with Terumo regular and exchange guidewire)	
Microsystems	Renegade Hi-Flo + Transcend 0.010" guidewire Balt COPERNIC balloon catheter × 2	
	Stenting and percutaneous transarterial angioplasty (PTA)	Terumo Sailor dilatation balloon for PTA: 10 × 40 mm Superstif supporting guidewire: 180 cm Wallstent: 14 × 60 mm

Direct intravenous endovascular thrombolysis with infusion of urokinase 750,000 IU during cerebral angiography successfully facilitated partial recanalization of the sinus. On the fourth day of admission, the patient became more alert (E3V4M6) and headache and vomiting were greatly relieved. Because BIJVS might cause further CVT and counteract the effect of direct thrombolytic therapy, an endovascular procedure was used on the fifth day of admission for both CVT and BIJVS. Urokinase 250,000 IU was infused into the superior sagittal sinus, followed by balloon thrombectomy and deployment of a wallstent (14 × 60 mm) in the left distal internal jugular vein. The venous pressure dropped from 26 to 12 mmHg. Thus, stenting of the right internal jugular vein successfully restored blood flow (Figure 2C).

Heparin was administered initially to prevent further venous thrombosis. Treatment was changed to warfarin several days later. After treatment, the patient regained full consciousness (E4V5M6), partially recovered his right muscle power and was free of headache, vomiting and seizure. An extensive search for the cause of the CVT revealed nothing specific except the BIJVS, the etiology of which could not be identified.

DISCUSSION

Clinical symptoms and signs of CVT vary widely, as in our case who initially presented with only mild right lower limb weakness, making correct diagnosis difficult. The premise that brain CTs may be normal in CVT is emphasized by this case. Thus, MRV should be performed whenever CVT is suspected.

Treatment of CVT varies according to the clinical condition of the patient. Heparin was first encouraged over 50 years ago, and has since proved effective, even in cases associated with intracerebral hemorrhage [2–4]. Although the types of heparin and the need for and duration of oral anticoagulation is still controversial in some cases, heparin has been upheld as the first-line treatment for CVT. This use was due to heparin's safety, feasibility and efficacy [2,4,5], until the recent introduction of direct endovascular thrombolysis. Urokinase, streptokinase and recombinant tissue plasminogen activator (rtPA) are most frequently used in such thrombolytic therapy. Urokinase was first used to treat CVT in 1971, and the results were satisfactory in a few small studies [2,4–6]. More recently, rtPA has been used due to its multiple theoretical advantages, although there is no evidence that it results in a better clinical outcome than urokinase or heparin [4,6].

In our case, urokinase was used because it has a similar effect to rtPA but costs much less, and because it causes fewer anaphylactic reactions than streptokinase [6]. Experience of combining direct endovascular thrombolysis and intravenous heparin in the treatment of CVT is limited. Recanalization of occluded vessels might be faster and more effective, but there lacks strong evidence that the clinical outcome is better [4]. Thus, further randomized trials of heparin injection alone versus direct endovascular thrombolysis in combination with heparin injection may be needed. Overall, the role of endovascular thrombolytic therapy remains controversial and it is generally indicated for patients who respond poorly to heparin administration, suffer from a worsening clinical course, or are most severely affected, due to its potential for hemorrhage [2–5].

CVT associated with internal jugular vein stenosis is a rare occurrence that has been reported infrequently [7–11]. Internal jugular vein stenosis may be idiopathic or caused by surgery, central venous catheterization, or trauma, and may elevate intracranial pressure and cause CVT. Thus, treatment of such a condition by deployment of a stent in the internal jugular vein may be beneficial. There have been a few case reports of successful treatment of bilateral internal jugular vein occlusions by stent deployment in the internal jugular veins [9,12].

Due to deteriorating consciousness, old age, increased intracranial pressure, intracerebral hemorrhage, seizure, and focal deficits (all of which indicate a poor outcome), we treated this patient using urokinase instead of heparin in the initial endovascular thrombolysis. However, the associated BIJVS had the potential to cause further CVT and minimize the effect of direct endovascular thrombolysis. Thus, stents were deployed in bilateral distal jugular veins to provide better venous outflow, which is helpful in the recovery of CVT. After stenting, we administered heparin to prevent further thrombosis. Finally, after the condition of the patient stabilized, heparin was switched to oral anticoagulant for further prevention of CVT.

To the best of our knowledge, few reports have documented successful treatment of CVT associated with BIJVS. We adopted all current methods in the treatment of CVT and the result was satisfactory. We conclude that early stenting may result in better outcome, especially in patients with rapid or severe neurologic decline. However, venous stenting is not generally accepted and further randomized trials may be needed to support this experience. In addition, BIJVS was likely to have been the cause of CVT and this raises the possible need for more routine inspection for jugular stenosis.

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以血栓溶解術和支架置放術 成功治療大腦靜脈栓塞合併兩側 內頸靜脈狹窄：一案報告

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大腦靜脈栓塞在目前並不少見，然而，大腦靜脈栓塞合併兩側內頸靜脈狹窄卻是很罕見的。兩側內頸靜脈支架置放術也是少見的技術。我們報告一個罹患大腦靜脈栓塞合併兩側內頸靜脈狹窄的個案，並且成功的以血栓溶解術和兩側內頸靜脈支架置放術治療此病患。

關鍵詞：大腦靜脈栓塞，內頸靜脈狹窄，血栓溶解術，支架置放術
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