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Successful hybrid endovascular treatment for refractory cerebral venous sinus thrombosis in pregnancy: A case report

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ABSTRACT

Background: Cerebral venous sinus thrombosis (CVST) in pregnancy was common and endovascular treatment (EVT) could be an effective and safe treatment for patients with severe and refractory CVST. However, the efficacy and safety of hybrid EVT (craniotomy + endovascular treatment) for CVST were unknown. We represented a rare case of hybrid EVT through the incision of the superior sagittal sinus in a pregnant woman with CVST who failed to EVT through the femoral vein pathway.

Case presentation: A 26-year-old woman, in her second month of pregnancy, complained of a headache for 5 days and aggravation with coma combined with convulsions for 2 days. She was diagnosed with CVST in the local hospital by digital subtraction angiography (DSA) and treated with anticoagulation. She had no history of illness and the biochemical tests were normal. Hybrid EVT (craniotomy + EVT) was attempted after failing to conduct EVT through the femoral vein pathway due to difficulty to reach the target cerebral venous sinus. Briefly, a small hole was made in the frontotemporal head to expose the superior sagittal sinus and a 6F sheath was inserted into 2cm of superior sagittal sinus incision and fixed on the scalp, after repeated aspiration by 5F intermediate catheter and balloon dilatation of stenosis in the right transverse sinus and right sigmoid sinus, the cerebral venous system got successful recanalization. No obvious complications were found and the patient recovered very well after the surgery.

Conclusion: Anticoagulation was the standard treatment for CVST. EVT could rapidly restore venous flow and improve the prognosis for refractory and severe CVST. EVT by hybrid surgery through the superior sagittal sinus incision may be safe and effective for desperate patients with severe CVST.

1. Introduction

Cerebral venous sinus thrombosis (CVST) accounts for about 0.5 %–1.0 % of all strokes [1] and is more common in pregnant women, women taking oral contraceptives, and young people under 45 years of age [2]. CVST could obstruct venous vessels, and as the

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thrombus progressed and expanded, it could cause a severe block of venous drainage, leading to brain edema, intracerebral hemorrhage, and venous infarction. Generally, mild to severe headache is the most common symptom and more severe symptoms include disturbance of consciousness, epileptic seizure, and focal neurological deficits. The diagnosis of CVST is recognized from the clinical syndromes and confirmed by computed tomography venography (CTV), MRV, and digital subtraction angiography (DSA) that show the absence of flow in a venous sinus and the existence of intraluminal venous thrombus [3,4]. Systematic anticoagulation is deemed the standard treatment for acute CVST even for those with intracerebral hemorrhage (ICH) [5]. Most patients with CVST who are diagnosed and treated early have a generally long-term satisfied prognosis [6]. However, it may not be the only choice for refractory and complicated cases with severe neurological deficits, coma, or contraindications to anticoagulation. Severe and refractory CVST could rapidly progress to cause ischemic and hemorrhagic stroke, brain edema, mass effect to hernia, and eventually death [7]. These patients may benefit from endovascular treatment (EVT) such as intra-sinus thrombolysis (IST) [8], mechanical thrombectomy (MT) [5], and even craniotomy decompressive surgery [9].

Hybrid surgical treatment (craniotomy + EVT) is an innovative treatment for severe and refractory CVST with few studies in the literature. Here we reported a rare case of hybrid surgical treatment (craniotomy + EVT) for CVST in pregnancy when mechanical thrombectomy failed through the femoral vein pathway.

1.1. Case presentation

A 26-year-old woman, with a BMI of 22.5 (54kg in weight, 155cm in height), in her second month of pregnancy, complained of a headache for 5 days and aggravation with coma combined with convulsions for 2 days. She had no past medical history and she was diagnosed with cerebral venous and sinus thrombosis (CVST) in the local hospital by digital subtraction angiography (DSA) and treated with anticoagulation (Warfarin 3mg po qd + Low molecular weight heparin 5000iu ic bid). The patient was in a coma, intubated with a ventilator to assist respiration, with bilateral pupils of equal size and equal circle with 2.5mm in diameter, with light reflex and bilateral frontal lines of equal weight. Other cranial nerve examinations could not cooperate to complete. The examination of muscle strength of the extremities could not cooperate to complete and the muscle tension was low, the tendon reflex of the extremities was weighed, and the Babinski sign of the right lower limb was positive. A blood routine test revealed a normal white blood cell count of 8.91 × 109/L, red blood cell count of 3.9 × 109/L, hemoglobin level of 116.4 g/L, and platelet count of 165 × 109/L. The coagulation test revealed a normal partial thromboplastin time (APTT) (26.9s), elevated prothrombin time (PT) (31.5s), and elevated international normalized ratio (INR) (2.8) and D-dimer (2118 ng/mL). Blood chemistry and serology showed the following: normal alanine aminotransferase (ALT) level of 7 U/L, normal aspartate aminotransferase (AST) level of 15 U/L, normal total bilirubin level of 9.1 μmol/L, normal serum creatinine level of 63.5 μmol/L, normal blood urea nitrogen level of 5.4mmol/L, normal low-density lipoprotein cholesterol level of 1.42 mmol/L, normal total cholesterol level of 2.73mmol/L and a reduced serum albumin level of 26.4 g/L. Brain CT showed a high-density sign in the right transverse sinus, obvious edema in the right cerebral hemisphere, low-density lesions, and a

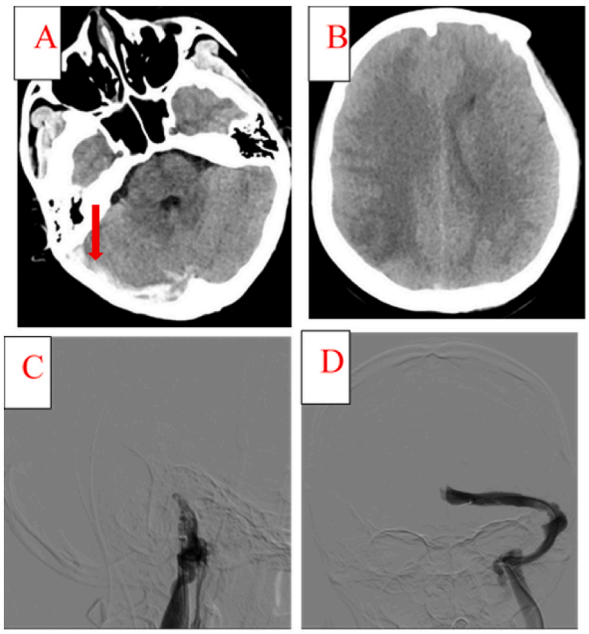

Fig. 1. Image characteristics of CVST.
A, Brain CT showed a high-density sign in the right transverse sinus (arrowhead) B, Brain CT showed tissue edema in the right cerebral hemisphere. C, DSA showed the occlusion of the intracranial segment of the right internal jugular vein. D, DSA showed no visualization of the superior sagittal sinus in the angiography of the left internal jugular vein.

little intracerebral hemorrhage (ICH) in the right frontal and parietal lobes (Fig. 1A and B).

The patient's symptoms progressed rapidly with anticoagulant therapy. We considered there was a high risk of brain edema, aggravated cerebral hernia, and death at any time for the young pregnant woman if EVT was not performed as soon as possible. Emergent EVT was performed, but it was difficult to reach the superior sagittal sinus after repeated attempts during the operation. We suspected the left transverse sinus did not connect to the superior sagittal sinus and the right transverse sinus, and the right jugular vein was blocked by the organization of the thrombosis or right jugular vein was stenosis (Fig. 1C and D). Hybrid EVT (craniotomy + EVT) was attempted and briefly, a small window in the superior sagittal sinus area was opened to expose the dura mater (Fig. 2A). The 6F artery sheath was inserted about 2cm into the superior sagittal sinus through the incision of the dura mater (Fig. 2A). After repeated aspiration by 5F intermediate catheter and balloon dilatation of stenosis in the right transverse sinus and right sigmoid sinus (Fig. 2B), the superior sagittal sinus, right transverse sinus, right sigmoid sinus, and right internal jugular vein were successful recanalization (Fig. 2C), and the amount of venous thrombus from the superior sagittal sinus was removed (Fig. 2D)After the surgery, the micro-catheter in the superior sagittal sinus from the femoral vein sheath was continuously pumped with 2u/h urokinase for three days. On the third day after the surgery, the disturbance of consciousness was well improved and changed from coma to lethargy. On the fourth day after the surgery, the disturbance of consciousness was further improved and changed to sleepiness. On the seventh day after the surgery, the patient became conscious and the endotracheal intubation was removed. At 17 days after the surgery, contrast-enhanced magnetic resonance venography (MRV) revealed a nearly normal appearance of cerebral venous and sinuses (Fig. 3A), while T2-flair showed hyper-intensity in the frontoparietal lobe (Fig. 3B) and magnetic susceptibility weighted imaging (SWI) showed some bleeding in the frontoparietal lobe (Fig. 3C). She was discharged 20 days after the surgery with a modified Rankin Scale (mRS) of 1. No adverse events such as skin bleeding, nasal bleeding, gingival bleeding, gastrointestinal bleeding, increase in cerebral bleeding, and significant thrombocytopenia were observed during the treatment. The timeline results of the blood routine test and coagulation test are shown in Table 1. After two weeks of follow-up, she recovered very well, with an mRS of 0. The patient expressed free of headache and seizure three months after discharge through telephone follow-up. She continued to take Warfarin (3.25mg qd) as we asked and monitored INR between 2 and 3 without any evidence of bleeding.

2. Discussion

CVST was a severe neurological disease with approximately 13 % poor functional outcomes, in terms of death or disability, even

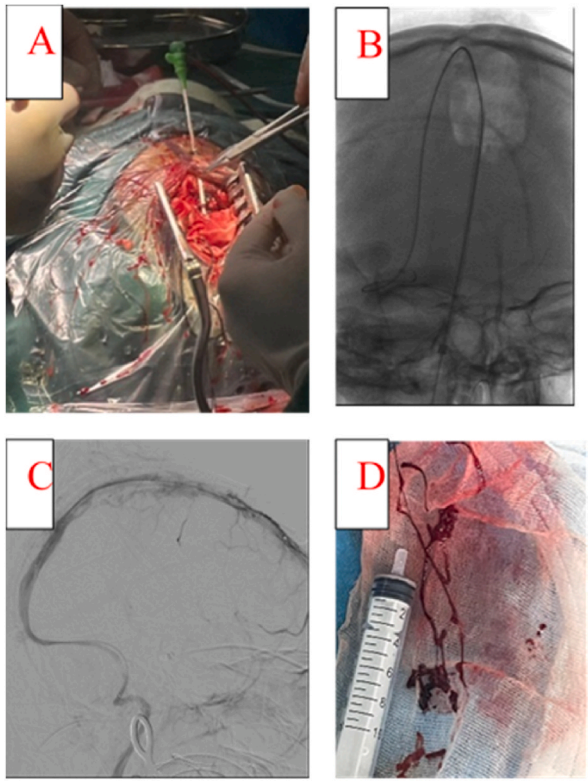

Fig. 2. Operation images.
A, the 6F artery sheath was inserted about 2cm into the superior sagittal sinus through the incision of the dura mater. B, 5F intermediate catheter got through the sheath into the superior sagittal sinus. C, DSA showed the recanalization of the superior sagittal sinus, right transverse sinus, right sigmoid sinus, and right internal jugular vein. D, Venous thrombus from the superior sagittal sinus aspirated by 5F intermediate catheter

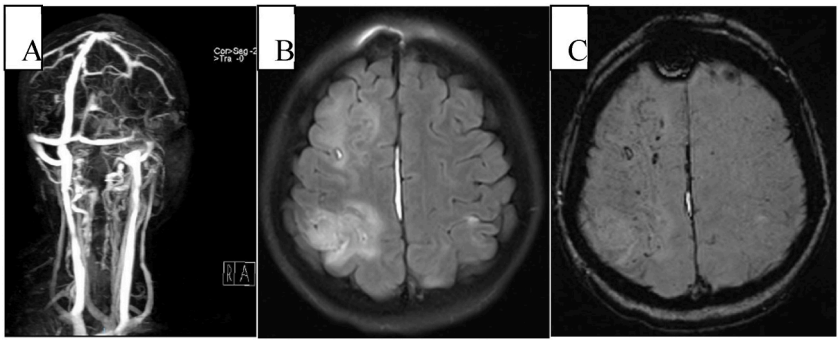

Fig. 3. MRV, MRI, and SWI findings at 17days after the surgery.
A, MRV shows significant improvement in the cerebral vein and sinus. B, Hyperintensity with cortical and subcortical white matter involvement is seen in the right parietotemporal lobe. C, Petechial bleedings are seen in the right frontoparietal lobe.

Table 1
Timeline results of laboratory test.

	PT(s)	APTT(s)	INR	WBC (10^9/L)	RBC (10^{12}/L)	PLT (10^9/L)	HGB (g/L)
Day1 (on admission)	31.5	26.9	2.8	8.91	3.9	165	116.4
Day2	29.6	39.4	2.64				
Day4	16.3	53.1	1.48	6.73	1.8	91	54
Day5	13	31.9	1.18	10.7	2.06	150	66
Day6	12.2	29.6	1.11	10.2	2.32	142	74
Day9	13.3	29.4	1.21	11.2	2.69	239	82
Day11	18.1	37.6	1.63				
Day12				11.7	2.69	252	81
Day14	16.1	36.1	1.45	9.61	2.73	271	82.6
Day16	19.7	41.6	1.77	8.88	2.71	265	82.8
Day18	27.5	40.5	2.43	6	2.64	267	83.1
Day20	42.4	42.6	3.68	5.7	2.54	268	80
Day21	41.5	41.6	3.61				
Day22 (discharge)							

PT, prothrombin time; APTT, activated partial thromboplastin time; INR, international normalized ratio; WBC, white blood cell; RBC, red blood cell; PLT, platelet; HGB, hemoglobin.

after treatment with anticoagulation [10].

Anticoagulation was deemed the standard treatment for acute CVST even for those with ICH. However, refractory and complicated cases of CVST with severe neurological deficits, coma, or contraindications to anticoagulation may bring neurologists into dilemma.

Endovascular treatment such as intra-sinus thrombolysis (IST) and/or mechanical thrombectomy (MT) might be a choice for these patients, and could rapidly restore the venous sinus blood flow [10,11]. However, the evidence supporting endovascular treatment for CVST was lacking. Some case reports and case series showed evidence of the efficacy and safety of MT and/or IST in patients suffering from CVST [5,8]. TO-ACT (Thrombolysis or Anticoagulation for Cerebral Venous Thrombosis) was a randomized clinical trial to evaluate the efficacy and safety of endovascular treatment in patients with a severe form of CVST, but the results showed that EVT with standard medical care did not appear to improve functional outcome of patients with CVST [12]. Given the small sample size of this study, although no conclusion was reached that EVT was superior to standard medical treatment, after careful screening and strategy improvement, it could be confirmed that some patients might still benefit from endovascular in future studies. A recent meta-analysis suggested endovascular treatment may be safe and efficacious in treating patients with severe CVST [13]. More studies with large sample sizes are needed to provide reliable data on the efficacy and safety of endovascular treatment for CVST.

Unsuccessful recanalization of CVST after the endovascular treatment was 4.7 %, a condition with a high mortality rate (83 %) [14]. Craniotomy decompressive surgery was a salvage therapy for these patients with a high risk of brain hernia secondary to brain edema, and intracerebral hemorrhage with CVST. Some case reports showed that craniotomy decompressive surgery could be life-saving in selected CVST patients with parenchymal lesions impending herniation [15–17]. However, it was a challenge for clinicians to treat patients who needed mechanical thrombectomy and/or endovascular thrombolysis but they failed to reach the target vessel through the femoral vein for various reasons. What could we do in this situation? In our case report, the young pregnant woman diagnosed with CVST was in a coma with an epileptic seizure, brain edema, and intracerebral hemorrhage, with no signs of hernia (her bilateral pupils were equally large and round and no midline shift was found in brain CT), so there was no indication for craniotomy decompressive surgery. Her symptoms gradually got worse with anticoagulation. Whether it was effective and safe for hybrid EVT through the superior sagittal sinus incision and direct thrombectomy? Previous case reports of similar hybrid surgery had been reported in the literature. Lechanoine et al. [18] described a case of a 45-year-old woman presenting with severe CVST which was

resistant to anticoagulation and endovascular therapies (thrombolysis). Bilateral hemicraniectomy combined with open surgical thrombectomy through the superior sagittal sinus was conducted when her clinical symptom aggravated to *trans*-tentorial herniation. She recovered very well after surgery with an mRS of 1 at discharge. Wang et al. [19] describe two cases of malignant CVST in pregnancy with clinical signs of impending herniation. One underwent bilateral coronal incision for craniotomy, the other one received frontotemporal bone flap craniotomy and they both underwent a superior sagittal sinusotomy and sagittal sinus thrombolysis during decompressive craniectomy through the incision of superior sagittal sinus and insertion of the suction tube. After the surgery, they both recovered very well with an mRS of 0 and 2 in three months after the operation. However, our case report showed there was no indication of craniotomy decompressive surgery. Instead, a small hole was made in the frontotemporal head to expose the superior sagittal sinus, and a 6F sheath was inserted into 2cm of superior sagittal sinus and fixed on the scalp, and then endovascular mechanical thrombectomy was successfully performed. After the hybrid EVT, no obvious complications were found and the patient recovered very well with an mRS of 0 in the follow. Our case report first showed EVT by hybrid surgery through the superior sagittal sinus incision was safe and effective in patients with CVST who failed to endovascular treatment through the femoral vein pathway.

3. Conclusion

Anticoagulation was deemed the standard treatment for acute CVST and EVT might be an alternative choice for these patients, and could rapidly restore the venous sinus blood flow and improve the prognosis. EVT by hybrid surgery through the superior sagittal sinus incision may be safe and effective for desperate patients with severe CVST who failed to EVT through the femoral vein pathway.

Ethics statement

Written informed consent was obtained from the individual for the publication. Any patients whose data or images are included in your publication have consented specifically for all images and clinical data and other data included in the manuscript to be published. The study was approved by the ethics committee of Zhongnan Hospital of Wuhan University (No.2023043K).

Data availability statement

Data will be made available on request.

CRediT authorship contribution statement

Renwei Zhang: Writing – review & editing, Writing – original draft, Visualization, Validation, Software, Resources, Project administration, Methodology, Investigation, Formal analysis, Data curation, Conceptualization. **Dong sun:** Resources, Investigation, Data curation. **Xinjun Chen:** Resources, Investigation, Data curation. **Yu Xie:** Resources, Investigation, Data curation. **Bitang Dan:** Resources, Investigation, Data curation. **Yumin Liu:** Writing – review & editing, Writing – original draft, Validation, Supervision, Investigation, Funding acquisition, Conceptualization. **Bin Mei:** Supervision, Methodology, Investigation, Funding acquisition. **Huagang Li:** Writing – review & editing, Validation, Resources, Methodology, Investigation, Formal analysis, Data curation.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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