Case Report

Mechanical Thrombectomy for Cerebral Venous Sinus Occlusion Due to Ewing Sarcoma: A Case Report

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Objective: To assess the efficacy of mechanical thrombectomy (MT) for cerebral venous sinus thrombosis associated with Ewing sarcoma invasion into the venous sinus.

Case Presentation: A 48-year-old woman presented to our hospital with left hemifacial paralysis. The patient had undergone surgical treatment for left subclavian Ewing sarcoma 24 years ago. Furthermore, the patient had undergone tumor resection for very late metastasis in the left occipital bone and mastoid air cells 3 years prior to her admission. As the Ewing sarcoma had extended to the left transverse sinus, the sinus was resected during craniotomy. The tumor remained in the mastoid air cells, and radiotherapy was administered. On admission to our hospital, FLAIR revealed hyper-intensities in the straight sinus and the superior sagittal sinus. Magnetic resonance venography revealed no signal in the posterior part of the superior sagittal sinus and the bilateral transverse sinuses. The patient was diagnosed with cerebral venous sinus thrombosis, and anticoagulation therapy was administered. However, the swelling and pain on the left side of the face worsened, and intraocular pressure (IOP) increased. Therefore, an MT was performed. After endovascular treatment, the patient's facial swelling and pain were reduced, and IOP returned to normal. Pathological examination of the captured thrombus revealed tumor cells, suggesting venous sinus invasion of Ewing sarcoma.

Conclusion: Although it was a palliative treatment, MT was effective in confirming the diagnosis of tumor invasion into the venous sinus and improving the patient's quality of life.

Keywords ▶ Ewing sarcoma, cerebral venous sinus thrombosis, mechanical thrombectomy

Introduction

Cerebral venous thrombosis (CVT) is a rare cause of stroke that occurs primarily in individuals younger than 55 years of age, with two-thirds of cases occurring in reproductive women.[1,2] The most common risk factors for CVT are oral contraceptives, pregnancy, and the postpartum period. Although mechanical factors such as compressive lesions of venous sinuses (primarily caused by meningiomas) have been reported,[2] there have been no previous reports of mechanical thrombectomy (MT) for CVT caused by Ewing sarcoma invasion into the venous sinuses. Herein, we report a case in which MT was effective in confirming the diagnosis and improving the quality of life.

Case Presentation

A 48-year-old Japanese woman presented to our hospital with left hemifacial paralysis. The patient had previously been diagnosed 24 years ago with left clavicle Ewing sarcoma and treated with surgical resection and preoperative and postoperative chemotherapy. However, metastasis to the left occipital bone was detected 3 years ago (very late metastasis, 21 years after the initial diagnosis). Ewing sarcoma had already extended to the left transverse sinus (TS), sigmoid sinus (SS), and mastoid air cells, and the left TS was occluded. A craniotomy and subtotal tumor

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excision were performed. Postoperatively, intensity-modulated radiation therapy (IMRT) at 61.2 Gy was administered in 34 fractions. MRI after IMRT showed a residual tumor that was smaller in size. The patient was discharged without a neurological deficit. CyberKnife radiosurgery was performed 20 and 27 months after IMRT, and local control of the residual tumor was achieved.

On admission to our hospital, MRI revealed edema of the left temporal lobe due to residual tumor, while FLAIR revealed hyper-intensities in the straight sinus (StS) and the posterior portion of the superior sagittal sinus (SSS) (**Fig. 1A** and **1B**). Magnetic resonance venography (MRV) showed no signal in the bilateral TS (**Fig. 1C**). DSA showed occlusion of the StS, the right TS, and the posterior part of the SSS in the venous phase (**Fig. 1D**–**1G**). Venous flow from the anterior portion of the SSS to the diploic vein and scalp vein was markedly prominent. In the deep venous system, venous flow from the internal cerebral vein was drained through the cavernous sinus. As these findings were only minimally related to the chief complaint upon admission, the patient was diagnosed with an incidentally discovered CVT, and no urgent emergency endovascular therapy (EVT) was performed. From a retrospective view, the facial nerve palsy on admission was presumed to be due to tumor extension in the mastoid air cells. Anticoagulation therapy with unfractionated heparin was continued for 1 week followed by transition to rivaroxaban 15 mg daily. On Day 8, T1-weighted imaging revealed hyper-intensities in the StS and SSS (**Fig. 2A**), suggesting the presence of a thrombus. Gadolinium-enhanced MRI and T2*-weighted imaging revealed a new metastasis on the left cerebellar tent. In addition, there were findings suggestive of tumor invasion from the right TS into the confluence of the sinus (**Fig. 2B** and **2C**). As there was no worsening of neurological symptoms, anticoagulation therapy was continued. However, left abducens nerve palsy and left facial swelling appeared on Day 18. Despite the addition of anti-edema therapy with glycerol, intraocular pressure (IOP) increased to 25 mmHg on the right and 31 mmHg on the left, and papilledema appeared. Owing to rapid worsening of pain and swelling on the left side of the face, DSA was performed on Day 21. The left inferior petrosal sinus (IPS), patent on admission, was no longer visible (**Fig. 3A** and **3B**), suggesting occlusion of the drainage route to the left internal jugular vein via the left IPS. This led to venous flow being redirected to the left ophthalmic vein, causing the sudden worsening of left facial pain and swelling. Therefore, we decided to perform emergency EVT.

EVT was performed as follows: under local anesthesia, a 4 Fr sheath was inserted into the left femoral artery. A 4 Fr JB-1 102 cm (Medikit, Tokyo, Japan) was placed in the right internal carotid artery for control angiography. An 8 Fr sheath was inserted into the right femoral vein, and an 8 Fr Optimo 90 cm (Tokai Medical Products, Aichi, Japan) was placed in the right jugular vein. RED72 (Penumbra, Alameda, CA, USA) was used as the aspiration catheter. First, RED72 was guided to the right TS near the confluence of the sinus, and MT was performed using a direct aspiration first-pass technique. However, recanalization was not obtained. Subsequently, Solitaire X 6 × 40 mm (Medtronic, Minneapolis, MN, USA) was deployed in the posterior half of the SSS and retrieved with aspiration from the RED72, which showed some thrombus in the stent but no recanalization. Finally, Trevo Trak 21 (Stryker, Kalamazoo, MI, USA) and Chikai black (Asahi Intecc, Aichi, Japan) were placed at the SSS, and EmboTrap III 6.5 × 45 mm (CERENOVUS, Johnson & Johnson Medical Devices, Irvine, CA, USA) was deployed (**Fig. 3C**). Using a combination technique with clot aspiration from the RED72 catheter and EmboTrap III stent retrieval methods, a significant clot was captured (**Fig. 3D**). Right internal carotid angiography in the venous phase revealed recanalization of the SSS and the right TS, and stenosis of the distal part of the right TS (**Fig. 3E** and **3F**). SHIDEN 6.0 × 20 mm (Kaneka Medix, Osaka, Japan) and Chikai black were placed at the site of stenosis in the right TS, and percutaneous transluminal angiography (PTA) was performed. Although the balloon dilated easily in the right TS (**Fig. 3G**), the TS recoiled to the state before PTA. The final right internal carotid angiography in the venous phase showed the antegrade venous flow from the SSS to the right TS (**Fig. 3H**). Although the StS remained occluded, this occlusion was present before the patient's symptoms worsened. The absence of impaired consciousness or edema in the deep brain suggested that it was unlikely to be related to the worsening of the patient's symptoms. Left internal carotid arteriography revealed occlusion of the left IPS (**Fig. 3I**). However, the procedure was terminated at this point because we assumed that recanalization of venous flow from the SSS to the right TS had reduced the burden on the left ophthalmic vein and facial vein (**Fig. 4**).

Although left facial pain and swelling improved immediately after EVT, dilation of the pupil and ptosis appeared on the left side. MRI showed no cerebral infarction, but tumor extension in the left cavernous sinus was detected (**Fig. 5A**–**5C**), which was considered the cause of the left

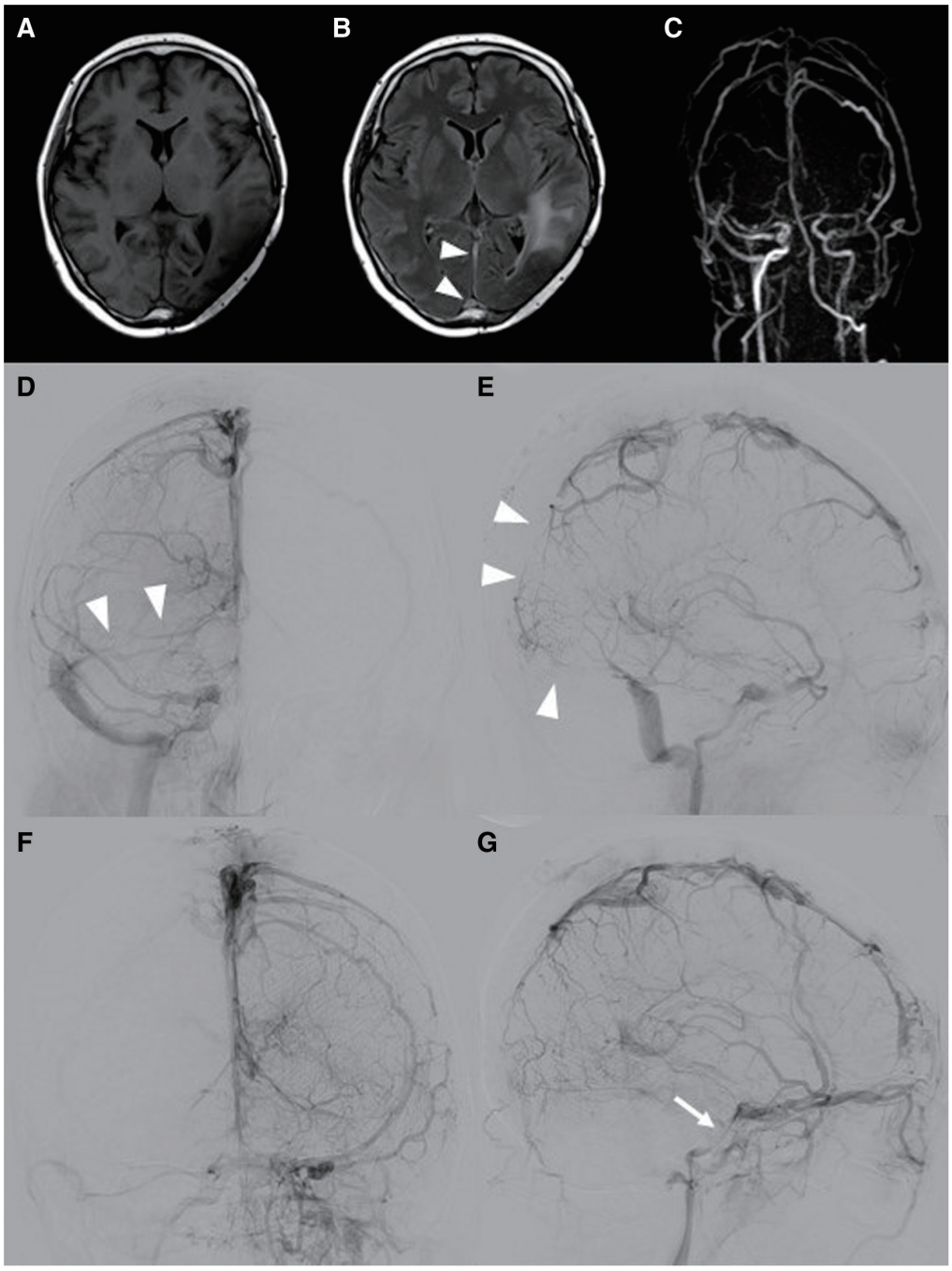

Fig. 1 T1-weighted imaging on admission showed no lesions in the dural sinuses (**A**). However, FLAIR revealed hyper-intensities (arrowheads) in the StS and SSS (B). MRV showed no signal in the SSS and bilateral TS (**C**). Right internal carotid angiography in the venous phase showed occlusion of the StS, the right TS, and the posterior part of the SSS (arrowheads) (**D, E**). Left internal carotid angiography showed the patency of left IPS (arrow) (**F, G**). IPS, inferior petrosal sinus; MRV, magnetic resonance venography; StS, straight sinus; SSS, superior sagittal sinus; TS, transverse sinus

oculomotor and abducens nerve palsy. IOP decreased to 17 mmHg on the right and 15 mmHg on the left. Pathological examination of the captured thrombus revealed small round cells with scant cytoplasm on hematoxylin–eosin staining (**Fig. 6A** and **6B**). CD99 immunohistochemistry showed strong membranous staining (**Fig. 6C**), and the cytoplasm was positive for periodic acid-Schiff (**Fig. 6D**). Therefore, we diagnosed cerebral venous sinus occlusion secondary to impaired venous flow caused by venous sinus invasion of Ewing sarcoma. Because the patient decided

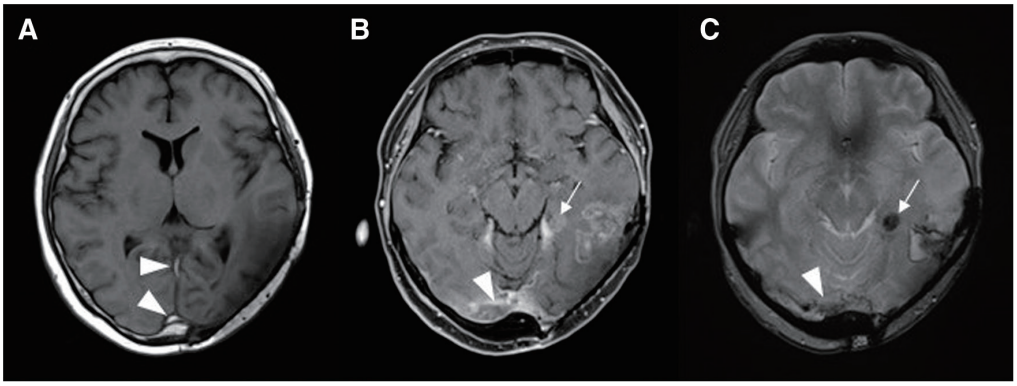

Fig. 2 On Day 8, T1-weighted imaging revealed hyper-intensities in the straight sinus and superior sagittal sinus (arrowheads), suggesting the presence of a thrombus (**A**). Gadolinium-enhanced MRI (**B**) and T2*-weighted imaging (**C**) revealed a neoplastic lesion on the left cerebellar tent, indicating a new metastasis (arrows), and findings suggestive of tumor invasion from the right transverse sinus into the confluence of the sinus (arrowheads).

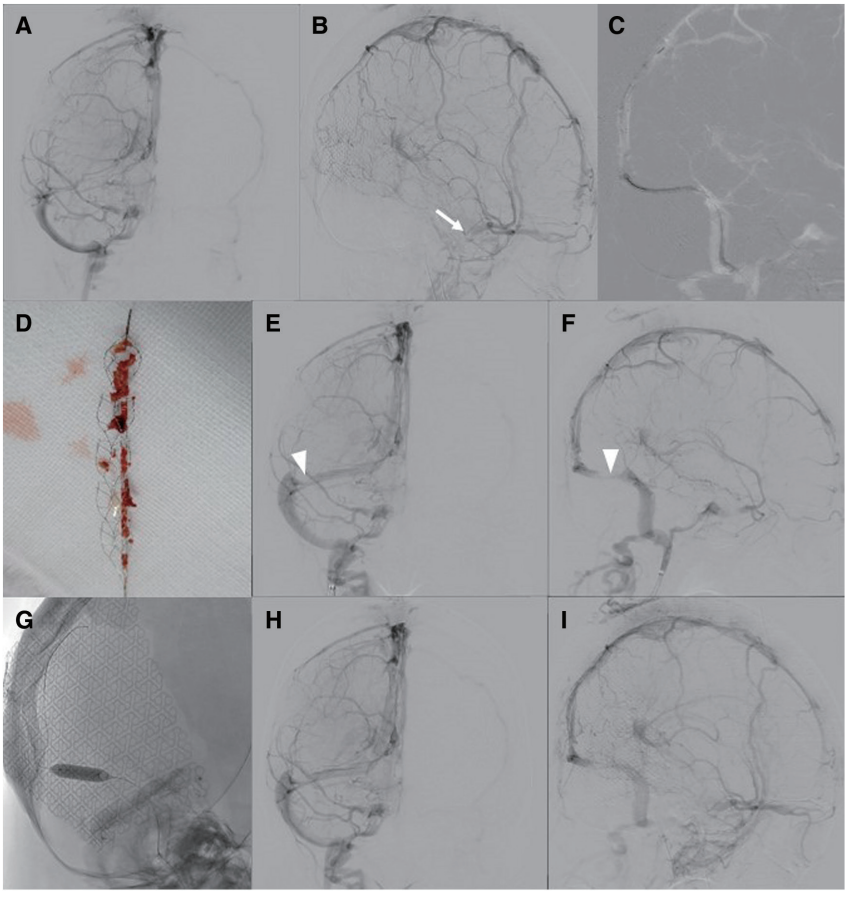

Fig. 3 Initial right internal carotid angiography in the venous phase depicting occlusion of the right TS and SSS (**A**). Left internal carotid angiography revealing occlusion of the left IPS (arrow) (**B**). A stent retriever was deployed at the SSS (**C**), and MT was performed. Using a combination technique with an aspiration catheter and stent retriever, a significant clot was captured (**D**). Although right internal carotid angiography in the venous phase revealed recanalization of the right TS from the SSS, there was a residual stenosis in the distal TS (arrowhead) (**E**, **F**). The balloon dilated easily in the right TS (**G**); however, the distal TS was recoiled (**H**). The final left internal carotid angiography showed the recanalization of the right TS from the SSS (**I**). IPS, inferior petrosal sinus; MT, mechanical thrombectomy; SSS, superior sagittal sinus; TS, transverse sinus

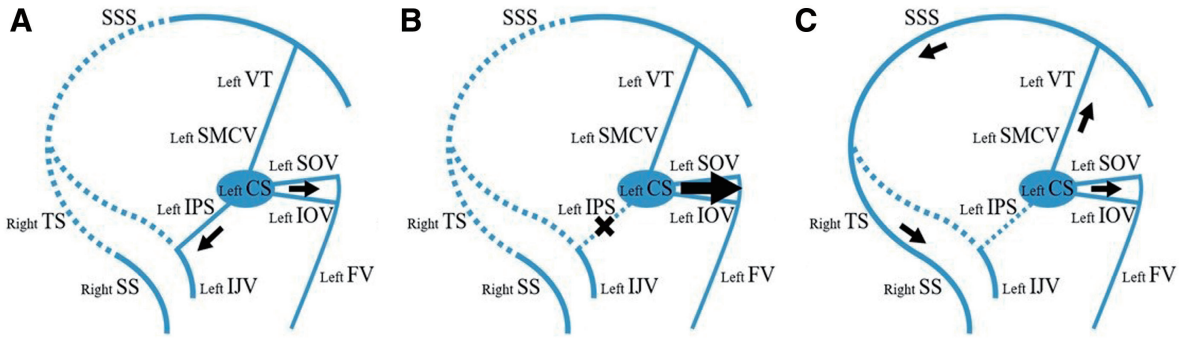

Fig. 4 On admission, the left IPS was patent and venous flow from the left CS drained through the left internal jugular vein via the left IPS (**A**). After the left IPS was occluded by the tumor extension, venous flow from the left CS converged to the left ophthalmic vein and then to the left facial vein. Edema and pain on the left side of the face were presumed to have manifested as a consequence (**B**). After MT, venous flow from the left CS was able to access the drainage route from the superior sagittal sinus to the right transverse sinus via the left superficial middle cerebral vein and vein of Trolard. Therefore, the venous flow to the left ophthalmic and facial veins was reduced, leading to an improvement in symptoms (**C**). CS, cavernous sinus; FV, facial vein; IJV, internal jugular vein; IOV, inferior ophthalmic vein; IPS, inferior petrosal sinus; MT, mechanical thrombectomy; SMCV, superficial middle cerebral vein; SOV, superior ophthalmic vein; SS, sigmoid sinus; SSS, superior sagittal sinus; TS, transverse sinus; VT, vein of Trolard

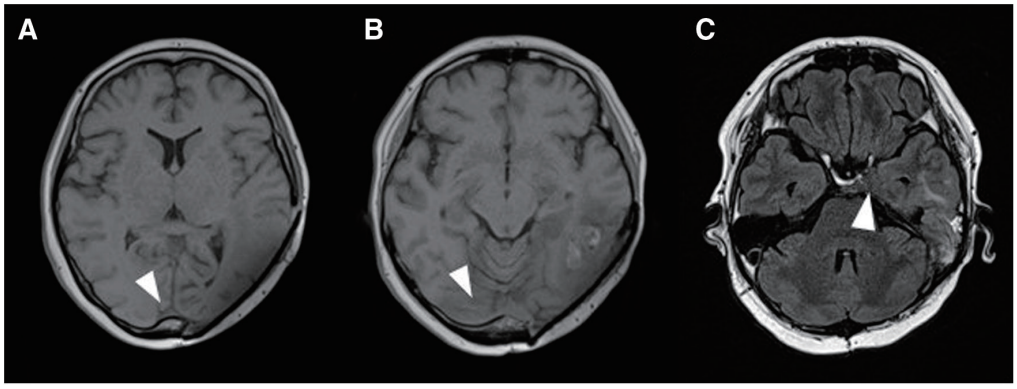

Fig. 5 MRI after MT revealed the disappearance of the hyper-intensities in the superior sagittal sinus and right transverse sinus on T1-weighted imaging (arrowhead) (**A**, **B**); however, FLAIR showed a new tumor in the left cavernous sinus (arrowhead) (**C**). MT, mechanical thrombectomy

not to undergo additional treatments, including radiotherapy, we opted for the best supportive care. The patient presented with impaired consciousness due to an enlarged tumor in the left cerebellar tent and had difficulty with oral intake after 6 weeks of treatment. Therefore, anticoagulation therapy was discontinued. MRV after 8 weeks of treatment showed no signal in the SSS to the right TS.

Discussion

Herein, we report a case of CVT caused by Ewing sarcoma invasion into the venous sinuses. CVT is typically treated with anticoagulation therapy, including with low-molecular-weight heparin.[2,3] EVT is indicated as salvage therapy in cases of clinical deterioration, or when standard therapy is ineffective or contraindicated.[2,4] In this case, anticoagulation therapy with heparin was initially administered. However, EVT was eventually performed because of clinical deterioration. There is currently no evidence to determine which EVT techniques (e.g., stent retrievers, microcatheters, aspiration catheters, aspiration pump systems) are superior to other therapeutic strategies.[2]

Although there has been a previous report of Ewing sarcoma of the occipital bone resulting in non-thrombotic partial venous sinus occlusion,[5] there have been no reports so far of MT for CVT associated with Ewing sarcoma invasion. Nevertheless, there are several reports of venous sinus stenting for meningioma invading the dural sinuses.[6–11] In all these cases, there was papilledema, suggesting increased intracranial pressure (ICP). The optimal indication for stenting has not been reported. Although stenting for venous sinus stenosis due to tumor compression has

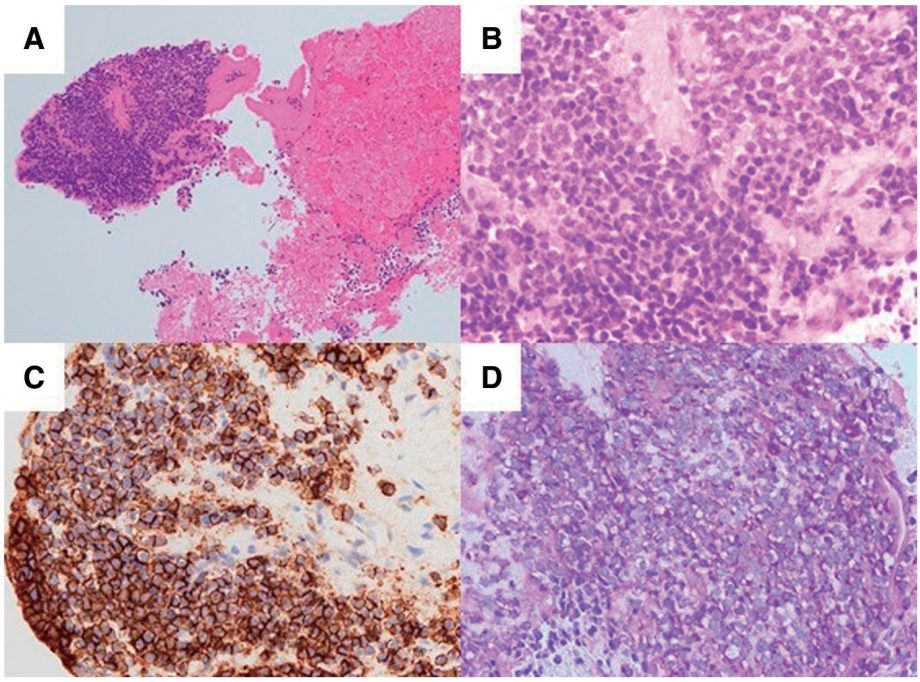

Fig. 6 Hematoxylin–eosin staining of the captured thrombus showed small round cells with little cytoplasm (**A** ×100, **B** ×200). Immunohistochemical staining revealed a membranous expression of CD99 (**C** ×200). The cytoplasm was typically periodic acid Schiff-positive, as it contained glycogen (**D** ×200).

been reported with favorable results, the long-term outcome remains unclear, and stenting requires the administration of antiplatelets or anticoagulants. Moreover, there are concerns about the influence of stenting on additional radiotherapy. By contrast, a case involving a meningioma with venous sinus stenosis reported a good outcome without complications after PTA, feeder occlusion of the tumor, and stereotactic radiosurgery.[12] However, in this case report, PTA was not successful in effectively dilating the dural sinus. In addition, the TS also recoiled to the state before PTA. The presence of a septation within the venous sinus may be one of the reasons why PTA did not provide sufficient dilation. The TS reportedly had 0–4 septations per side, with a mean of 1.75.[13] The septations were most commonly located where the sinus changed direction, with an average diameter of 6.21 mm on the right side and 6.30 mm on the left.[13] In the present case, contrast-enhanced MRI revealed at least one septation, with a diameter on the right TS of 6.9 mm. Stenosis of the right TS was not consistent with the tumor location as seen on MRI and was probably due to the septation. The presence of a septation and abundant anatomical variations may complicate EVT for CVT. Stenting or PTA has been considered in cases of obvious focal stenosis or occlusion such as meningiomas. However, in cases of diffuse stenosis/occlusion or long lesions due to tumor invasion or secondary thrombus occlusion, as in the present case, MT with a stent retriever may be an effective treatment. In addition, in this case, EmboTrap III (CERENOVUS) after retrieval showed that many thrombi and tumors were trapped in the inner channel as well as in the distal closed mesh (**Fig. 3D**). The venous sinus was thought to have narrowed considerably because of the tumor invasion. Therefore, Solitaire X (Medtronic) with its unique overlap structure may not have been able to provide sufficient radial force. However, EmboTrap III's segmental structure and inner channel with high radial force may have been effective.

One limitation of this treatment is the possibility of hematogenous metastasis to the lungs or other organs due to the intravenous spread of tumor cells. In this case, the risk of metastasis was explained to the patient, and EVT was preferred in view of the progressive worsening of symptoms. In addition, intraoperative monitoring of sinus pressure, as used for hemodynamic evaluation during balloon test occlusion,[14] may have been useful in evaluating treatment efficacy and determining whether to add PTA or stenting.

In the present case, after MT recanalized the venous sinus, IOP decreased steadily, and the left facial swelling and pain were reduced. Currently, the gold standard for ICP measurement is lumbar puncture or implantation of a pressure sensor

in the ventricle. However, these invasive measurements of ICP are accompanied by the potential risk of intracranial hemorrhage and infection.[15] Considering the risk associated with lumbar punctures, and the results of recent studies which suggest a strong correlation between ICP and IOP,[16–18] lumbar puncture was not performed in this case. We used IOP instead of ICP to estimate the efficacy of EVT.

Conclusion

We present a case of CVT due to Ewing sarcoma invasion into the dural sinuses, in which MT was performed, and recanalization was achieved. Although it was a palliative treatment, MT may help confirm the diagnosis, improve venous flow, and enhance the patient's quality of life in cases where CVT is suspected due to venous sinus invasion by the tumor.

Informed Consent

We obtained informed consent from the patient and her family to publish this clinical report.

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Disclosure Statement

The authors declare that they have no conflicts of interest.

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