CASE REPORT

Combined use of Solitaire FR and Penumbra devices for endovascular treatment of cerebral venous sinus thrombosis in a child

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ABSTRACT

A pre-teenager with newly diagnosed ulcerative colitis presented to an emergency department with acute headache, altered mental status and bilateral lower extremity weakness. Head CT demonstrated acute thrombus in the vein of Galen and straight sinus, and the patient was started on a heparin infusion. The patient clinically deteriorated and became unresponsive. In view of the rapid deterioration despite anticoagulation therapy, the patient was taken for endovascular treatment. A novel endovascular approach was performed with combined use of Solitaire FR and Penumbra devices to enhance access to the straight sinus and to limit intraprocedural blood loss. The post-treatment head CT demonstrated a decrease in hyperattenuation within the vein of Galen and straight sinus. The neurologic status improved within 24 h. The patient was discharged home with a normal neurologic examination.

BACKGROUND

Cerebral venous sinus thrombosis (CVST) in children is a rare disease with an incidence of only 0.67 per 100 000 children per year.[1] It may affect children of all ages and is often difficult to diagnose, but its potentially fatal outcome makes timely identification and effective treatment of the condition essential. Consequently, clinicians should be well aware of this entity and the potential usefulness of endovascular treatment in its management.

Multiple predisposing and comorbid conditions can lead to CVST, including infection, hypercoagulable states, trauma and chronic medical conditions including inflammatory bowel disease. The superficial venous system is most often involved by CVST, with the most common sites of involvement including the transverse sinus, superior sagittal sinus and sigmoid sinus.[2 3]

In contrast, deep venous system involvement, particularly involvement isolated to the deep system, is uncommon in both children and adults. Nevertheless, the outcome of untreated CVST of the deep system may be devastating due to the critical brain structures on which drainage through this system depends. In addition, endovascular access to thrombosed deep system vessels, particularly the proximal straight sinus, may be difficult. Techniques for endovascular treatment of deep system CVST in children should emphasize flexible devices to achieve maximal access and minimal blood loss. We report a patient with CVST isolated to the deep venous system whose rapidly deteriorating clinical presentation warranted endovascular treatment. A novel endovascular technique was used for access to and treatment of the deep venous sinus thrombosis using a combination of Solitaire FR and Penumbra devices.

CASE PRESENTATION

A pre-teenager presented with headache, altered mental status and bilateral lower extremity weakness. The patient had been diagnosed with ulcerative colitis 8 weeks prior to admission and 2 weeks prior to admission had experienced an ulcerative colitis flare treated with prednisone and mesalazine (Pentasa). Two days before admission the patient complained of frontal headache which improved with acetominophen. One day before admission the headache worsened and the patient experienced nausea and appeared confused. On the day of admission the patient was brought to an outside hospital emergency department. On arrival the patient was initially shaking their head to answer questions, and then developed weakness in the lower extremities bilaterally.

The hemoglobin value was 7.5 g/dL, but had been 10.1 g/dL on a test performed 2 months previously.

INVESTIGATIONS

Head CT demonstrated acute thrombosis of the vein of Galen and straight sinus. The patient was given a 75 unit/kg bolus of heparin, started on a heparin infusion and transferred to our institution. Upon arrival at our institution a repeat head CT again showed thrombus in the straight sinus and vein of Galen unchanged from the prior scan. Over the next hour the patient became unresponsive. MRI at that time showed thrombosis of the vein of Galen and straight sinus without intracranial hemorrhage. Edema within the basal ganglia was present bilaterally without diffusion restriction.

TREATMENT

A conventional angiogram to delineate the anatomy was not performed since the location and amount of clot was well delineated on MR venography (MRV). This mirrors practice by other investigators such as Stam *et al*[4] who prospectively studied 20 patients with severe cerebral sinus thrombosis treated with thrombectomy or thrombolysis. While

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some of their patients underwent conventional angiography, in others a diagnosis of CVST was confirmed by MRI and MRV or CT venography.[4] Furthermore, in pediatric patients we are very careful with the amount of contrast and radiation used. Therefore, when there is high-quality non-invasive imaging available to evaluate the venous anatomy adequately, an angiogram is not performed. In this case we also did not believe that an angiogram would fill the venous structures due to the large clot burden seen on non-invasive imaging. Therefore, the venous side was only accessed during this emergency procedure.

A 6 Fr 80 cm Shuttle sheath was positioned via the right common femoral vein in the distal right internal jugular vein. A 032 Penumbra reperfusion catheter coaxially placed within a 054 Penumbra reperfusion catheter was navigated into the proximal straight sinus using a Synchro-2 microwire. Contrast injection demonstrated straight sinus thrombosis with impairment of outflow. The 032 Penumbra catheter was removed and replaced by a Marksman catheter coaxially through the 054 Penumbra catheter. A 6×30 mm Solitaire FR device was deployed within the proximal straight sinus for 10 min without suction applied to minimize blood loss. The Solitaire FR device was then removed under continuous suction through the Penumbra catheter. Post-stent angiography demonstrated improvement in venous drainage into the transverse sinus from the straight sinus. The 6×30 mm Solitaire FR device was again deployed for 10 min without suction and again removed under continuous suction. Post-thrombectomy angiography demonstrated marked improvement in venous drainage without significant residual clot. Less than 20 mL of blood was collected in the Penumbra suction container during the procedure (figure 1).

OUTCOME AND FOLLOW-UP

Head CT after the procedure showed a decrease in the previously seen hyperattenuation in the straight sinus and vein of Galen. A post-procedure MRI was not performed by the primary team. The patient was continued on a heparin infusion over the first post-procedure day, after which the patient was transitioned to low molecular weight heparin therapy. The patient's neurologic status rapidly improved and the patient was discharged home after 6 days with a completely normal neurologic examination. Follow-up MRV/MRI was performed 2 months after discharge and demonstrated resolution of the thrombus.

DISCUSSION

This case illustrates the use of a novel endovascular technique for the treatment of deep system CVST and also highlights some specific aspects of pediatric CVST in which these techniques might warrant consideration.

In addition to treating any predisposing cause for CVST, systemic anticoagulation represents first-line therapy. Despite the widespread use of anticoagulation, it lacks support of large randomized trials and outcomes after treatment remain variable. CVST-specific mortality has been reported to be less than 10% in adults with long-term disability or neurologic deficits in 17–79% of survivors.[3] Although most patients have good clinical outcomes, mortality ranges of 5–30% are reported and 15–25% of patients survive with significant neurological deficits.[5] In addition, permanent neurocognitive disabilities are major sequelae in up to 44% of survivors.[6]

CVST involvement of the deep system is uncommon in both adult and pediatric populations but may carry a particularly poor prognosis. For example, Sebire *et al*[2] found only six cases of isolated deep system involvement in their review of 42 cases of pediatric dural sinus thrombosis.[2] Although uncommon, compromise of the deep venous system by thrombosis typically impairs thalamic and basal ganglionic function resulting in confusion and other alterations of consciousness including coma.

Coma has been noted to be a predictor of poor outcome in a large series of patients with CVST.[7] In addition, coma has recently been found by Sebire *et al*[2] to be a significant predictor of death in childhood CVST.[2] These authors also found that, although pediatric patients treated with systemic anticoagulation had a reduction in mortality, this reduction was not statistically significant.[2]

Importantly, differences exist between adults and children in the effects of CVST on brain parenchyma. This has been demonstrated by imaging of parenchymal abnormalities associated with CVST. Adults demonstrate a relatively poor correlation between the location and size of brain parenchymal abnormalities and the site and extent of CVST.[8] In contrast, Teksam *et al*[9] demonstrated good correlation between the site and extent of sinus thrombosis and the location of parenchymal lesions in children.

These findings suggest that adults might better tolerate CVST without parenchymal compromise than children. This may occur because pediatric patients lack protective mechanisms to prevent transmission of increases in venous pressure to the parenchyma or because the immature blood–brain barrier is more susceptible to disruption in a setting of sinus thrombosis.[9] The findings also suggest that parenchymal dysfunction, including that which may be irreversible, may develop earlier in children than in adults.

The combination of coma as a predictor of mortality, the uncertain prevention of mortality with conventional anticoagulant treatment and evidence of early onset parenchymal compromise in children suggest an aggressive approach to children with CVST who demonstrate decreases in level of consciousness. Because deep venous system involvement may underlie this presentation, particular attention should be given to endovascular treatment of patients with coma and deep venous system involvement since these patients may not do well with systemic anticoagulation treatment.

Endovascular treatment of CVST has been described in adults for a number of years in situations characterized by clinical deterioration in the face of standard anticoagulant treatment. Although no randomized trials exist, data from an adult series indicate that favorable outcomes can be achieved in over 90% of patients.[10] The role of endovascular techniques in pediatric patients has been less frequently reported. Recently, Mortimer *et al*[11] reported using endovascular techniques in nine pediatric patients with worsening clinical deterioration. They used an intrasinus infusion of recombinant tissue plasminogen activator (rt-PA) in eight patients, with the use of the Penumbra device in five combined with angioplasty in two. A favorable outcome was achieved in eight of the nine patients treated.[11] A review of the current literature shows only 15 additional pediatric cases of CVST treated with endovascular techniques.[12–19]

Most of the pediatric patients reported have been treated using the endovascular technique of intrasinus infusion of medication, either urokinase or rt-PA. In our case t-PA was not used because of the large clot burden and acuity of the patient's symptoms with a rapidly worsening clinical status. From our experience, large amounts of t-PA need to be infused in order to treat a large clot burden. In this pediatric case the burden was thought to be too large to be adequately treated using t-PA without increasing the risk of t-PA-related complications. The use of endovascular thrombolytic devices has been relatively

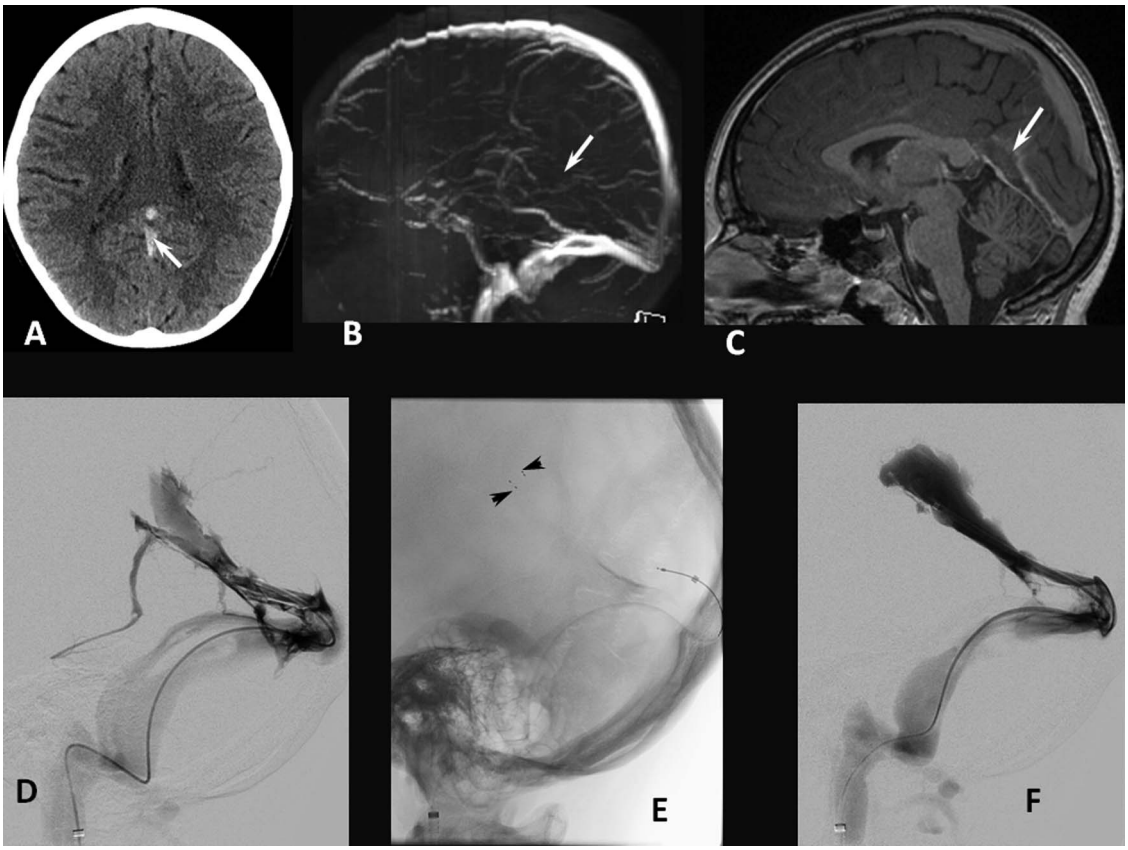

Figure 1 (A) Axial unenhanced CT showing hyperdense clot within the vein of Galen and straight sinus (arrow). (B) Sagittal reconstruction of two-dimensional time-of-flight MR venography with no visible flow-related enhancement within the expected location of the vein of Galen or straight sinus (arrow). (C) Sagittal T1 MPRAGE post-contrast imaging showing non-enhancing thrombus within the vein of Galen and straight sinus (arrow). (D) Straight sinus venography (lateral view) showing poor filling and intraluminal filling defects compatible with thrombosis. (E) Lateral view after deployment of the Solitaire FR device (arrowheads). (F) Straight sinus venography (lateral view) showing complete filling and rapid washout following thrombectomy.

limited, with five cases using the Penumbra device and one using the AngioJet device (Possis; now Medrad, Warrendale, Pennsylvania, USA).[11 19 20]

The continued reliance on intrasinus infusion with limited reports of the use of thrombectomy devices probably reflects the more recent development of these devices and also the difficulty of working within the often small dural sinuses of the pediatric population and the need to use the most flexible low profile devices. In addition, the dural sinus anatomy may be challenging, particularly when patients require treatment of the straight sinus, the major drainage pathway of the deep venous system. In the majority of reported cases, thrombolysis or thrombectomy has been limited to the sigmoid, transverse or superior sagittal sinuses.

The frequently acute angulation at the junction of the confluence of sinuses (CS) and straight sinus may make selective catheterization of the straight sinus particularly difficult. For example, in the series reported by Mortimer *et al*,[11] although straight sinus involvement was present in six cases, selective catheterization of the straight sinus was reported in only one with removal of clot confined to the distal straight sinus in one other case. The authors specifically noted that difficult anatomy precluded catheterization of the straight sinus in one case.

Park *et al*[21] evaluated the morphology of the CS in a cadaver study and concluded that the CS should not be regarded simply as the drainage site for the superior sagittal and straight sinus or the origin of the transverse sinuses, but rather as a complex system of anastomosing sinuses.[21] These investigators determined the direction, communication and type of inflow for the straight sinus at the CS, features which could impact on the feasibility of selective catheterization of the straight sinus from the transverse sinus. They found that, in 58.1%, the direction of straight sinus inflow was toward the center of the CS, in 35.5% it was toward the left transverse sinus, in 3.2% it was toward an occipital sinus and in only 3.2% was it toward the right straight sinus.[21] This anatomy suggests that in many cases, particularly from a right transverse sinus approach, a relatively acute reverse turn must be navigated in order to enter the straight sinus.

For this reason, the most maneuverable and flexible devices should be considered for straight sinus thrombolysis and thrombectomy. In addition, as with the procedural treatment of most pediatric disorders, differences in technique from that used in adults should be given significant emphasis. Consequently, some thrombolytic devices that have been found useful for adult CVST (such as Angiojet) may be less suitable in children because of size and stiffness.[15] In order to accomplish the often difficult catheterization of the straight sinus, we used a Penumbra thromboaspiration device. The relative flexibility of this device, particularly when used in a coaxial fashion, enabled placement into the proximal straight sinus at the junction with the vein of Galen.

Because of the small size of many patients undergoing pediatric neurointerventional procedures, an additional consideration is blood loss. The Penumbra device, particularly the larger bore catheters, if used with suction activated over the often long period of time required for clearance of a dural sinus, can result

in intraprocedural blood loss that may be significant in children, particularly patients with anemia associated with chronic disease. For this reason, we combined a Solitaire FR stent retriever device with the Penumbra.

The Solitaire FR device is a self-expanding retrievable nitinol stent approved by the FDA for flow restoration in patients with ischemic stroke due to large intracranial vessel occlusion. Its use has also been reported to be effective in the treatment of dural sinus thrombosis.[22] There is also one case in the literature of the combined use of the Penumbra and Solitaire devices for deep venous sinus thrombosis in an adult.[23] In our case a Solitaire FR was deployed through the Penumbra device and left expanded within the straight sinus thrombus for approximately 10 min without suction applied to the Penumbra device to minimize blood loss. After this period, suction was activated and the Solitaire FR was withdrawn into the Penumbra device. This permitted the relatively large amount of clot adherent to the Solitaire FR to be removed by a short period of suction through the Penumbra. This technique was repeated until the clot was removed.

Our preliminary experience with the combined use of the Penumbra and Solitaire FR device for the treatment of CVST was successful and without complication.

Although indications for endovascular therapy in the pediatric population remain unclear, poor prognostic features such as coma and clinical deterioration despite systemic anticoagulation should warrant early consideration of endovascular treatment. The use of current thrombectomy devices and novel endovascular techniques for limiting blood loss in children appear effective and should be of high importance when undertaking clot removal in the cerebral venous system. The combination of the Solitaire FR device with the Penumbra system proved to be a safe and effective way of clot removal from the deep venous system in this patient.

Key messages

- In patients with cerebral venous sinus thrombosis, poor prognostic features such as coma and clinical deterioration despite systemic anticoagulation should warrant early consideration of endovascular treatment.
- The combined use of a Solitaire FR device and Penumbra device is effective in treating cerebral venous sinus thrombosis in the pediatric population.
- Maneuverable and flexible devices should be used in the pediatric population due to their anatomic differences from adults.
- Limiting continuous suction through Penumbra devices to appropriate times during endovascular therapy may prevent excessive blood loss in pediatric patients.

Contributors HS and RWH were primarily involved with the endovascular approach of this case, gathering background data, performing extensive literature review, and writing this manuscript. DL was involved with clinical patient care, obtaining consents, and performing a literature review. BAP and AM were involved with the practical parts of this case and helped with deciding treatment during intervention and following patient in the hospital.

Competing interests None.

Patient consent Obtained.

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