Turk Neurosurg 34(2):362-366, 2024

Case Report

DOI: 10.5137/1019-5149.JTN.29642-20.2

Received: 26.02.2020
Accepted: 06.07.2020
Published Online: 13.02.2024

Embolization of a Non-Galenic Pial Arteriovenous Fistula with Assistance of a Solitaire™ Stent: Case Report

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ABSTRACT

This study aimed to describe a novel endovascular strategy to help control blood flow used successfully to treat an infant with high-flow pial arteriovenous fistula (AVF).

Here, a single-hole high-flow nongalenic pial AVF was diagnosed in a 2.5 year-old infant is presented. After coil packing failure despite temporal balloon occlusion, we deployed a Solitaire™ stent in the fistula. By twisting with detachable coils, we achieved satisfactory blood flow control, and obliteration of the fistula was achieved with Onyx injection.

In conclusion, Solitaire™ stent deployment in the high-flow pial AVF can help stabilize the coils in the fistula, leading to satisfactory blood control. This strategy may be a valuable addition to currently available options.

KEYWORDS: Pial arteriovenous fistula, Vascular disorder, Embolization

ABBREVIATIONS: NGPAVF: Nongalenic pial arteriovenous fistula, **AVF:** Arteriovenous fistula, **CT:** Computed tomography, **NBCA:** N-butyl cyanoacrylate

INTRODUCTION

Intracranial nongalenic pial arteriovenous fistulas (NGPAVF) are rare cerebrovascular lesions (prevalence ranging between 0.1 in 100,000 to 1 in 100,000) (2). These lesions are one or more arterial connections to a single venous channel without intervening nidus. Patients with NGPAVF can exhibit various manifestations originating from the steal phenomenon, rupture of a fistula orifice, venous hypertension, or mass effects of the expanded varix. If untreated, the prognosis of high blood flow of AVFs is poor, with mortality reported as high as 63% (6).

The treatment goal for an NGPAVF is disconnection of the arteriovenous shunt, but removal of the entire lesion is thought to be unnecessary. Consequently, with recent improvements in technique and materials, endovascular obliteration has become the preferred treatment for most pial AVFs due to its reduced level of invasiveness (4,7). However, treatment of high-flow AVF is always challenging when satisfactory blood control cannot be achieved.

In this case report, we described the novel application of a Solitaire™ stent to stabilize coils in the fistula and facilitate blood control in the embolization of an NGPAVF in an infant.

CASE REPORT

History

The patient was a 2.5-year-old boy with no significant birth history. He experienced sudden onset of unconsciousness

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and limb twitch starting 2 months prior, and was treated with anti-epileptic drugs at a local hospital. A head CT scan showed encephalatrophy and a large temporal mass (Figure 1). He was then transferred to our hospital for further treatment, and was found to have increased head circumference during neurological examination after admission.

Investigation

A CT scan revealed a high density mass in the left temporal lobe, about 3cm × 5cm in size in the coronary and sagittal section, with obvious encephalatrophy (Figure 1A, B). Also, a dilated transversal sinus was observed (Figure 1C). MRA indicated a pial AVF supplied by enlarged middle cerebral artery and left posterior cerebral artery (Figure 1D).

Angiograms and Endovascular Therapy

Embolization was performed under general anesthesia. Heparin (0.66 mg/kg) was administered for anticoagulation after femoral access was established. An angiogram revealed an NGAVF in the left temporal lobe. Feeding arteries from left middle cerebral artery (MCA) trunk, left MCA temporal branch, and left posterior cerebral artery (PCA) were visualized, and these arteries confluence into a single-hole fistula, and drained by a proximal vein followed by a giant varix. Downstream of the giant varix, venous drainage occurred primarily through a dilated transverse sinus (Figure 2A-F).

Our treatment strategy was to embolize the fistula and proximal draining vein without affecting the varix. Since high blood flow was observed, we attempted to embolize the fistula with coils and Onyx-18 (ethylene vinyl alcohol copolymer, ev3, Irvine, USA) under balloon-assisted flow arrest. Bi-femoral access was established. One Envoy DA guiding catheter (Codman, MA, USA) was placed in the cavernous segment of left internal carotid artery, and One Chaperon guiding catheter (Microvention, Terumo, USA) was put in the V3 segment of the left vertebral artery. Two Hyperform balloons (Hyperform 4/20,4/15, ev3, Irvine, USA) were delivered into left MCA trunk and left PCA, respectively. One Echelon-10 microcatheter (ev3, Irvine, USA) was advanced through the anterior circulation into the fistula, in preparation for introduction of coils and subsequent Onyx injection. After inflating the balloons, we introduced one Cosmos 14/51 (Microvention, Terumo, USA) in the proximal draining vein, in hopes that the coils can be kept in the fistula or proximal draining vein close to the fistula. However, the introduced coil constantly migrated into the varix, because of the flared morphology of the proximal draining vein and possible collateral blood flow into the fistula, despite balloon occlusion in both MCA and PCA trunks. Given that coil embolization in the varix will result in a much higher cost and might prevent potential shrinking of the thrombosed varix, we modified the treatment strategy. The balloon in the MCA was retracted and a Solitaire™ stent 4/20 (Medtronic, California, USA) was delivered across the fistula. The distal part of the stent was deployed in proximal draining vein, while the proximal marker was in the feeding artery of MCA trunk. Then, the Cosmos coil was stabilized precisely around the fistula after twisting with the stent. A total of six coils were introduced followed by an injection of about 2 ml Onyx-18. With blood control satisfactorily achieved, the Onyx displayed excellent dispersion. The stent was subsequently detached and the cast was stable (Figure 3A-F). In the final angiogram control, the lesion was totally occluded with obvious contrast stasis in the varix and transverse sinus. There was no migration of embolic agent into the distal draining vein or the varix. After embolization, one dural arteriovenous fistula was found in the sinus confluence and was scheduled for staged treatment (Figure 4 A–F)

Post-treatment outcome and follow-up

Low-molecular-weight heparin was administered subcutaneously after the procedure for 3 days. Blood pressure was strictly controlled after embolization. The mean arterial pressure was kept at approximately 75 mm Hg for 48 hours after embolization. Patient course was uneventful after operation, and was discharged 5 days later. Magnetic resonance venogram before discharge showed successful obliteration of the draining vein and left transverse sinus (Figure 4 G). During clinical follow-up two months later, the patient was stable with no neurological deficit.

DISCUSSION

Endovascular treatment of the pial AVF has evolved over the past decade. Embolic agents including N-butyl cyanoacrylate (NBCA) and Onyx have commonly been used to occlude

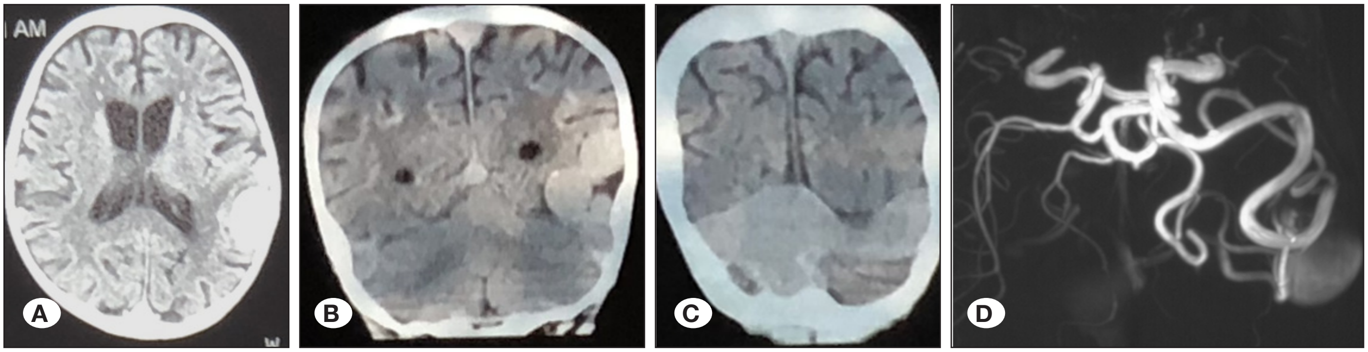

Figure 1: Images of the infant with NGAVF before admission. A CT scan revealed a high density mass in the left temporal lobe, about 3 cm × 5 cm in the coronary and sagittal section, and obvious encephalatrophy was observed **(A, B)**. A dilated transversal sinus was also observed **(C)**. MRA indicated a left temporal pial AVF supplied by enlarged middle cerebral artery and posterior cerebral artery **(D)**.

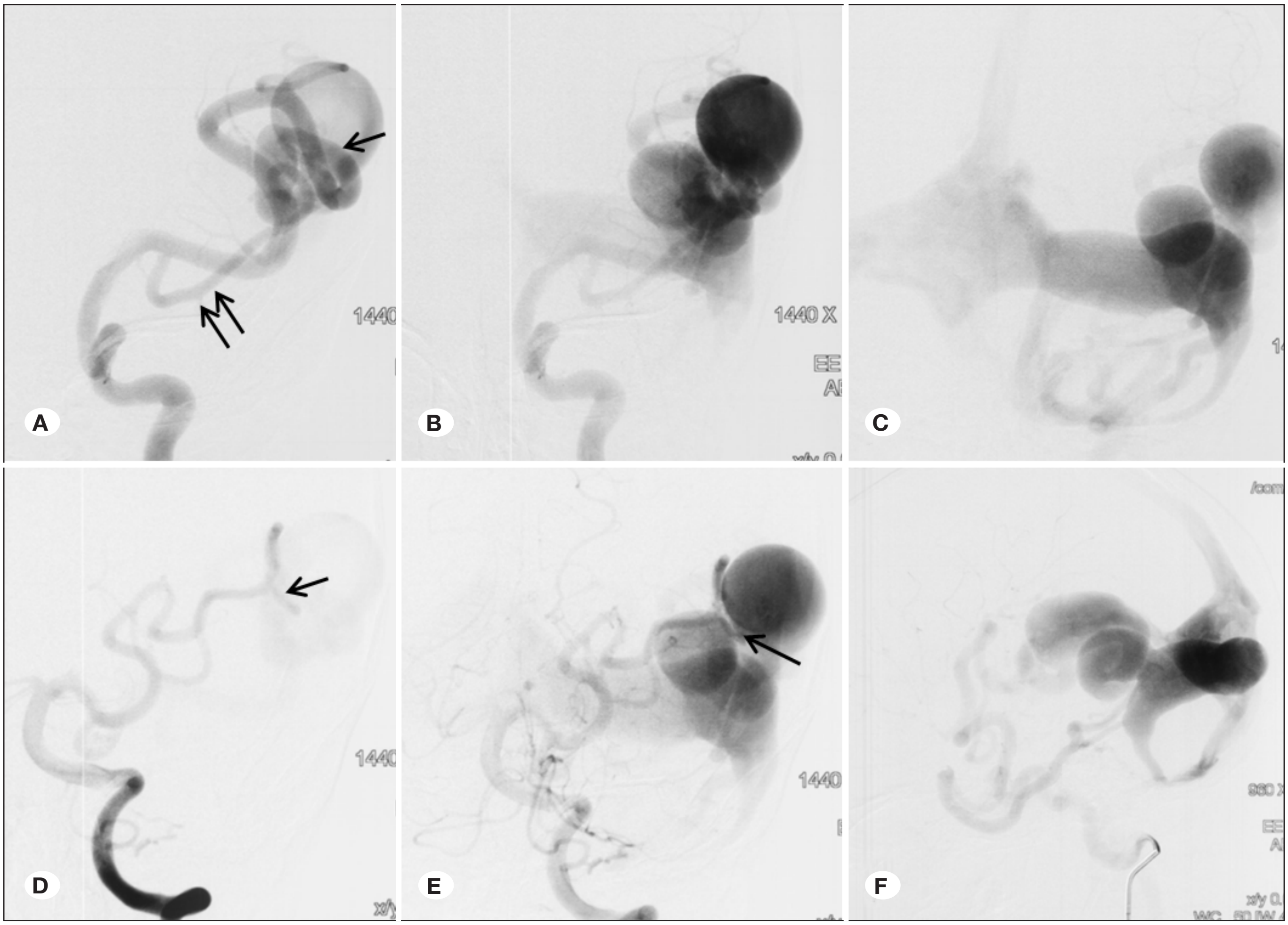

Figure 2: Angiogram performed prior to embolization. Angiograms in the left internal carotid artery **(A-C)**, and left vertebral artery **(E-F)** demonstrated a left temporal lobe NGAVF. Feeding arteries from left middle cerebral artery trunk (black arrow, A), left MCA temporal branch (double arrow, A) and left posterior cerebral artery (black arrow, E, F) were visualized. These arteries confluence into a single-hole fistula, and drained via a proximal vein, followed by several giant varices. Downstream of the giant varix, venous drainage occurred primarily through dilated transverse sinus **(B, C, E, F)**.

the fistula point. However, it was always technically difficult to deliver the embolic agent precisely to the shunt point due to high-flow dynamics when the blood flow was not well controlled. This may lead to undesirable migration of embolic material into the draining vein before complete obliteration of the fistula, and thus lead to disastrous consequences. Therefore, adequate control of blood flow is the key to successful NGPAVF treatment.

Several methods including coils and balloons have been proposed to better control blood flow. Balloon-assisted Onyx or NBCA embolization are often used, but sometimes cannot block flow from all collateral branches. Additionally, these procedures are complicated and rely heavily on the experience of the clinician, as repeated “deflation” and “inflation” are needed to ensure the fistula is completely obliterated (5). There are also worries about material migration after deflation of the balloon. Coils are another option as they can provide better positioning and are not easily carried away by high-velocity blood flow. However, in some lesions, stabilization of detachable coils may be challenging due to the special vascular morphology adjacent to the fistula site. In such cases, many coils have to be placed inside the giant venous varix to achieve flow control and stable placement of the coils.

In this case, we do not want to place the coils into the varix since it will cost a lot, and the coils may potentially prevent the varix from shrinking. So, we attempted balloon dilation to control the blood flow prior to placing the coils in the fistula or proximal draining vein. However, the coils cannot be stabilized in the proximal draining vein due to unfavorable morphology and the high-velocity of collateral blood flow. After failure of coil assisted embolization, we adopted a new method to help stabilize the coils and embolic agents. To our best knowledge,

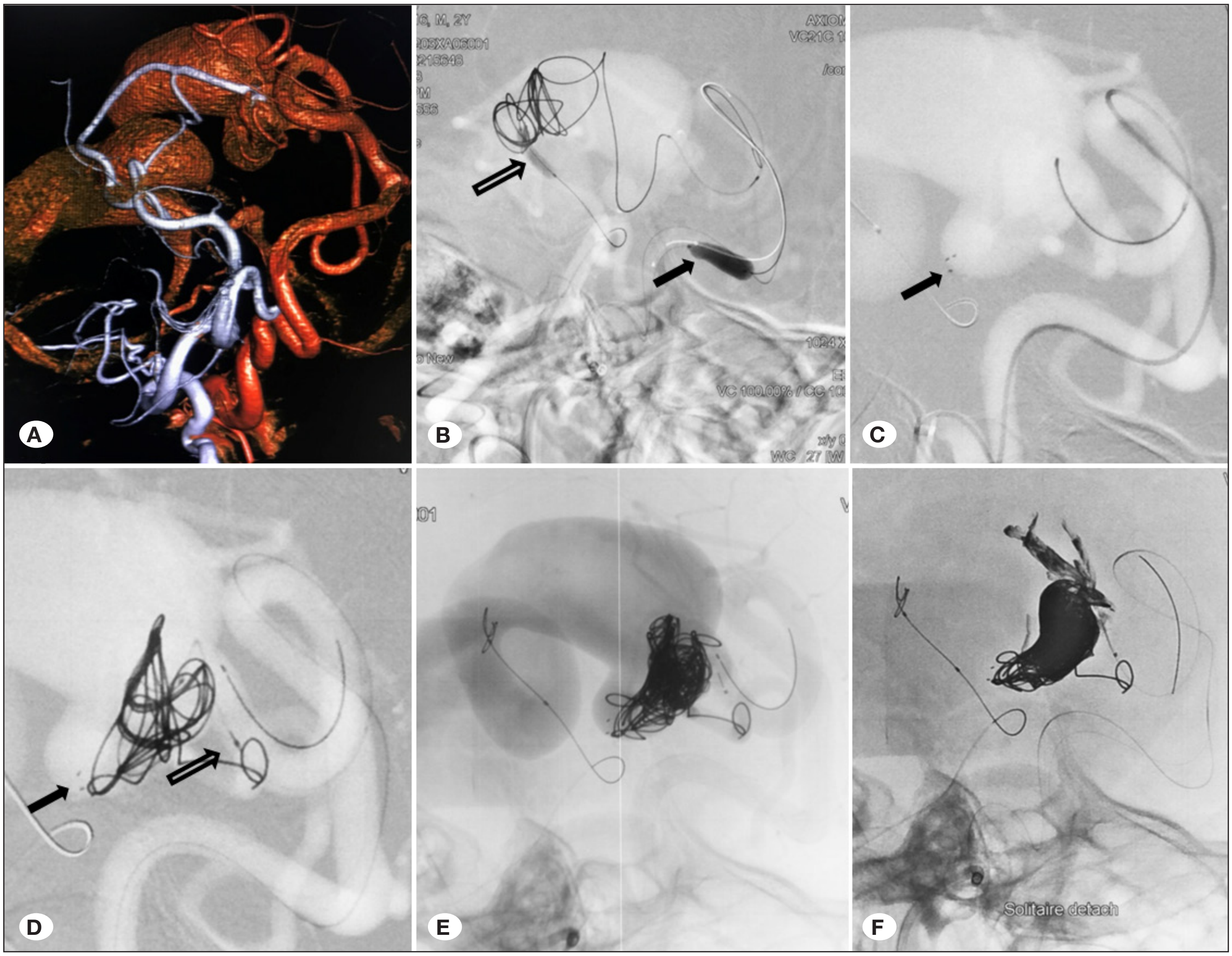

Figure 3: Treatment process. 3D reconstruction of the fistula clearly showed the structure of the fistula **(A)**. Three enlarged arteries confluence into a single-hole fistula followed by draining vein and varices. Two balloons were delivered into left MCA trunk (Hyperform 4/20) and left PCA (Hyperform 4/15), respectively, After inflating the balloons, we introduced one Cosmos 14/51 (Microvention, Terumo, USA) in the fistula, but the coil that we introduced constantly migrated into the varix **(B)**. Given that Coil migration into the varix will result in a much higher cost, and perhaps work against potential shrinking of the thrombosed varix, the treatment strategy was modified. a Solitaire™ stent 4/20 (Medtronic, California, USA) was delivered across the fistula. Distal part of the stent was deployed in proximal draining vein (arrow, C), while the proximal marker was in the feeding artery of MCA trunk (arrow, D). Then, the Cosmos coil was precisely stabilized around the fistula after twisting with the stent **(D)**. A total of six coils were introduced **(E)**. With satisfactory blood control achieved, the following Onyx injection displayed excellent dispersion **(F)**.

this is the first report of a successful embolization of a high-flow AVF with assistance of a stent.

The Solitaire™ stent was initially designed to treat wide-necked intracranial aneurysms, and later used widely in thrombectomy (1,3). It offered an unique overlapping and electrical detachment design. When we deployed the Solitaire™ stent in the fistula and proximal draining vein, twisted with the coils and helped reduce blood flow, which allowed us to inject Onyx-18 to occlude the fistula, now that we have deployed this “dam up” technique. We believe that injection of Onyx allows more precise targeting and delivery of embolic material to the fistulous point, consistent with previous work (5). The stent was successfully detached after the complete obliteration of the fistula. One potential limitation to the stent-assisted embolization is the worry of stent delivery. In many patients with high-flow AVFs, the supplying arteries are always tortuous, which adds difficulty in stent delivery to the target position. So, we used one Envoy DA guiding catheter to provide support for stent delivery.

CONCLUSION

We report the successful treatment of an infant non-Galenic pAVF with assistance of a Solitaire™ stent. To our knowledge, this is the first such case reported in the medical literature.

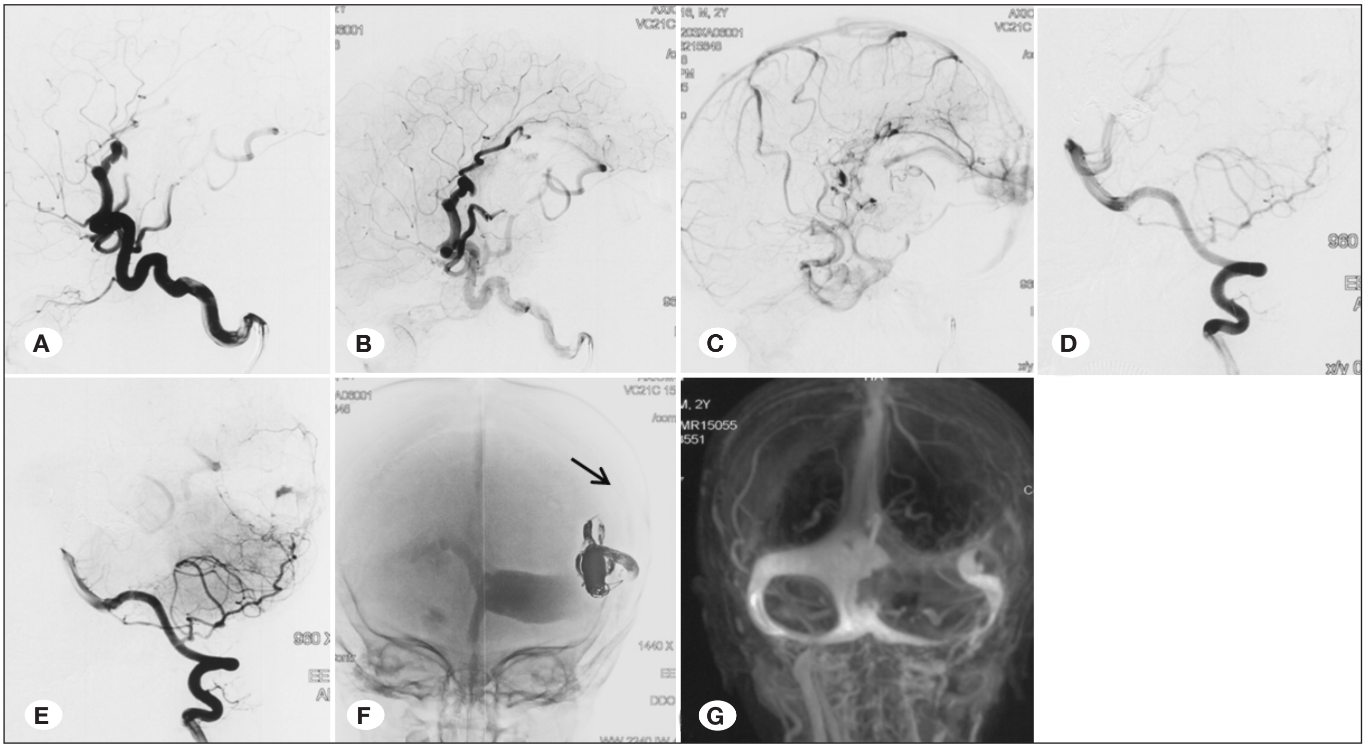

Figure 4: Angiogram after embolization. Angiograms in left internal carotid artery **(A-C)** and left vertebral artery **(D, E)** demonstrated that the AVF was completely occluded. An obvious contrast stasis was observed in the varix and transverse sinus **(F)**. However, one dural arteriovenous fistula was found in the sinus confluence, and was scheduled for staged treatment (arrow). Magnetic resonance venogram before discharging showed obliteration of the draining vein and left transverse sinus **(G)**.

Solitaire™ stent deployment in the high-flow pial AVF can help stabilize coils in the fistula and lead to satisfactory blood control. This treatment strategy adds a valuable addition to the currently available options. Future studies in more patients and longer-term follow-up are needed.

AUTHORSHIP CONTRIBUTION

Study conception and design: JL, YX
Data collection: YZ, QL, JL
Analysis and interpretation of results: YZ, QL, JML, YX
Draft manuscript preparation: YZ, QL
Critical revision of the article: JL, JML, YX
All authors (YZ, QL, JL, YX, JML) reviewed the results and approved the final version of the manuscript.

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