



Hybrid Operation of a Ruptured Aneurysm Associated with a Developmental Venous Anomaly

Jingwei Li, Shiwei Du, Liyong Sun, Feng Shang, Hongqi Zhang, Guilin Li

■ BACKGROUND: As a normal variation of the cerebral venous angioarchitecture, developmental venous anomalies (DVAs) represent a rare cause of intraparenchymal hemorrhage. Different from mixed vascular lesions, such as capillary malformations, arteriovenous malformations, arteriovenous fistulas, and thrombosis, here we describe the first case of a ruptured hemodynamic aneurysm associated with a DVA and introduce the first use of hybrid operation for the treatment of a hemorrhagic DVA.

■ METHODS: A 14-year-old girl suffered from sudden onset of headache, aphasia, and left hemiplegia. On the way of her transfer to our center, she suddenly fell into a coma. Computed tomography scan demonstrated an enlarged intraparenchymal hemorrhage in the left temporal lobe. Angiography revealed a large frontal DVA with an associated hemodynamic aneurysm. Superselective angiography of left middle cerebral artery confirmed that the aneurysm was located on the turning site of arteriovenous transition. **RESULTS:** Considering the large hematoma and the possible occlusion of surrounding collecting veins, we attempted transarterial embolization but were unsuccessful. Hematoma evacuation and aneurysm isolation were performed in a hybrid operation room. Intraoperative angiography was used to confirm the location of the aneurysm and to recheck the result. The patient woke up 1 day later and the symptoms were relieved entirely 1 year later.

■ CONCLUSIONS: Associated aneurysm may be a cause of intraparenchymal hemorrhage in DVAs and routine imaging monitoring is needed. Hybrid operation is a possible

treatment for such complicated mixed lesions in DVA, which proved to be safe and effective in this patient.

INTRODUCTION

Developmental venous anomalies (DVAs) are the most common form of vascular malformations, with an incidence of 2.6% in brain autopsies and 0.48%–1.7% via magnetic resonance imaging detection.^{1,2} They are considered as a normal variation of the venous angioarchitecture, consisting of a cluster of thickened and hyalinized veins with little smooth muscle and elastic tissue, rather than a pathologic entity.³ Considering the high incidence, DVA is rarely a cause of intraparenchymal hemorrhage, and thus, most reports suggest a conservative approach to treat DVA.^{4,5} However, it is still associated with potential hemorrhagic risk on the basis of the histologic and angioarchitectural characteristics. In addition, several studies indicate that DVAs mixed with other cerebrovascular abnormalities, such as arteriovenous malformations (AVMs), capillary malformation, arteriovenous fistulas (AVFs), and thrombosis, carry an increased risk of hemorrhage.^{3,6-10}

Here we describe the first case of a DVA with a ruptured hemodynamic aneurysm, which was located on a turning site of arteriovenous transition. The successful use of the hybrid operation for a hemorrhagic DVA is illustrated for the first time as well. We present the case along with a discussion of the natural history and management of DVAs.

CASE REPORT

Medical History and Examination

A 14-year-old girl suffered from sudden onset of headache, aphasia, and left hemiplegia. Computed tomography of the brain

Key words

- Aneurysm
- Arteriovenous fistula
- Developmental venous anomaly
- Hemodynamic
- Hybrid operation
- Intraparenchymal hemorrhage

Abbreviations and Acronyms

- AVF:** Arteriovenous fistula
- AVM:** Arteriovenous malformation
- DVA:** Developmental venous anomaly

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in local hospital showed a left temporal intraparenchymal hematoma (**Figure 1A**). On the way to our hospital, she suddenly fell into a coma, which turned out to be a rehemorrhage by the second computed tomography scan (**Figure 1B**). The Glasgow Coma Scale at admission was 6. After the informed consent form was signed by her parents, she was transferred to the hybrid operation room immediately.

Standard cerebral angiography demonstrated a left frontal DVA, which was fed by branches of left middle cerebral artery and anterior cerebral artery and predominantly drained to the left internal cerebral vein. On one perforating branch of MCA, a 3.5-mm aneurysm was found. Superselective angiography revealed that it was located on a proximal turning site of arteriovenous transition and there was no venous stenosis or thrombosis, indicating it was likely a hemodynamic aneurysm (**Figure 1C–G**). Taking the large hematoma and surrounding collecting veins into consideration, we attempted transarterial embolization but were unsuccessful. Then, hematoma evacuation and aneurysm isolation were performed successfully with the guidance of intraoperative angiography (**Figure 1H–J**). The patient woke up 1 day later and the symptoms were relieved entirely 1 year later.

DISCUSSION

According to the radiologic and autopsy series data, DVAs occur in 2.5%–3.0% of the population and are the most common vascular malformations in central nervous system (60% in autopsy series).^{1,11,12} As the meta-analysis shows, 61% of DVAs are discovered as incidental findings and they are usually treated as benign entities. However, they are indeed associated with potential angioarchitectural weakness, and about 6% of them

may present with symptomatic hemorrhages.¹³ Therefore, DVA, a risk factor of hemorrhage, should be carefully recognized by the caregivers.

Apart from the capillary malformations, other coexisted vascular abnormalities (such as AVMs, AVFs, stenosis, and thrombosis) also will influence the balance between the inflow and outflow of DVAs and thus may play a hemodynamic role in the mechanism of hemorrhage.⁷ The upstream AVM or AVF may increase the inflow of DVAs, whereas the downstream stenosis or thrombosis may decrease the outflow of DVAs. In those cases, the blood flow load as well as the venous pressure load will be increased obviously and thus may cause the rupture of the thin-walled vessels. AVF-associated aneurysms have been reported in very rare cases. However, those aneurysms were all located on the distal end of the transitional vessels. There seems to be a relatively narrow tract between the aneurysm and the collecting vein, indicating that the decrease of outflow may play more important role in the formation and rupture of aneurysm.^{7,14,15}

The aneurysm in our case was located on a proximal turning site of arteriovenous transition, and there was no stenosis or thrombosis on its outflow tract. Therefore, we suppose that the aneurysm was mainly caused by the blood flow load. The pathomechanism may be similar to the AVM-associated aneurysms on their feeding artery.^{16–19} It is possible that these weaker vessels with little smooth muscle and elastic tissue have a decreased responsiveness to flow changes in both short and long term. With the fast blood flow rushing, hemodynamic aneurysm may occur at the turning site or the bifurcation.

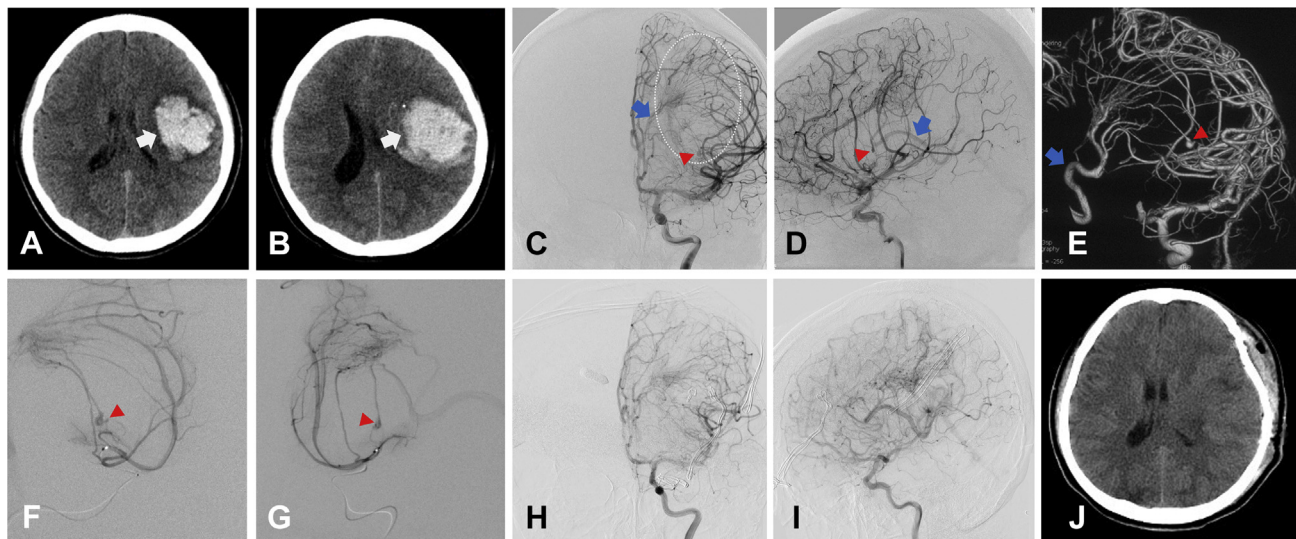


Figure 1. Axial computed tomography (CT) scan at onset (**A**) and 8 hours later after admission (**B**) showing an enlarged intraparenchymal hematoma (white arrow) in the left temporal lobe, which indicated that a rehemorrhage occurred. Anteroposterior view (**C**), lateral view (**D**), and 3-dimensional view (**E**) of left internal carotid artery (ICA) angiography (late arterial phase), revealing a developmental venous anomaly (typical caput medusae sign, white oval) fed by branches of left middle cerebral artery (MCA) and anterior cerebral artery and predominantly drained to the left internal cerebral vein (blue arrow). The aneurysm (red arrowhead) was located on one perforating branch of MCA (**F**, **G**). Anteroposterior view (**H**) and lateral view (**I**) of left ICA postoperative angiography showing the aneurysm was obliterated and surrounding transitional vessels were well preserved. At 3 days after the operation, axial CT scan of the brain (**J**) demonstrating the hematoma was completely removed and there was no infarction or developmental venous anomaly-related calcification.

For symptomatic DVAs, Pereira et al.¹⁵ classified them into 3 types: mechanical (20.3%), flow-related (71.0%), and idiopathic (8.7%). The microshunt presented in our case, increasing the inflow into DVA, belongs to the flow-related type and can be found in 15.9% of the symptomatic DVAs and 22.4% of the flow-related type. In addition, two thirds of the DVAs with increased inflow may present with symptomatic hemorrhage.¹⁵ Therefore, to identify the microshunt before hemorrhage and to detect the potential risk changes, routine imaging examination may be needed. The symmetrical dilated capillary veins in the medullary zone of DVAs, where the associated aneurysm may occur, should be detected with more attention.

Successful neurosurgical or neurointerventional management of these mixed DVAs would target the related components, thus reducing the hemorrhagic risk of the tissue-dependent DVAs.²⁰ The surrounding transitional vessels should be preserved to maintain the parenchymal perfusion; otherwise, the venous infarction or rehemorrhage may occur.^{5,21} Endovascular embolization for a mixed lesion in DVA may have a reliably positive effect on its angioarchitecture and hemodynamics. However, in our case, superselective angiography showed the microcatheter could hardly reach the aneurysm and the embolization of

surrounding transitional vessels seemed to be unavoidable. In addition, the large hematoma was also an important factor for the choice of hybrid operation. When accompanied with intraparenchymal hemorrhage, the search for a small aneurysm is difficult and might cause some unintentional injury to the surrounding tissue. Given the difficulty in locating the aneurysm, a marker (i.e., aneurysm clip) can be placed in the suspected site, and the angiography can show the relative location between the marker and the aneurysm. In this way, the least deviation can be introduced.

CONCLUSIONS

In summary, this case provides an extremely rare example of a ruptured hemodynamic aneurysm associated with a DVA and the first use of hybrid operation for the treatment of a hemorrhagic DVA. It reminds us of the possibility that the blood flow-related aneurysm may cause intraparenchymal hemorrhage in DVAs, and routine imaging monitoring may be needed for those symptomatic DVAs. In addition, for such complicated mixed lesions in DVA, the hybrid operation may be a possible treatment, which proved to be safe and effective in this patient.

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