

CASE REPORT

Stroke intervention for middle cerebral artery thrombus in a young patient with an ipsilateral Spetzler—Martin grade V arteriovenous malformation

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ABSTRACT

A patient (in their late 20s) was admitted with a right frontal stroke, left hemiparesis and hemianopsia, and a National Institutes of Health Stroke Scale (NIHSS) score of 11. CT perfusion imaging revealed an ischemic penumbra. A CT angiogram showed a Spetzler—Martin grade V arteriovenous malformation (AVM) in the right frontal lobe and a second, smaller AVM in the medial occipital region. Successful mechanical thrombectomy for middle cerebral artery thrombi improved flow from a Thrombolysis in Cerebral Infarction score of 0 to 2b. Following endovascular mechanical thrombectomy, the NIHSS dramatically improved from 11 to 2 and the patient was discharged home in 7 days. Evaluation 1 month after treatment disclosed a nearly complete recovery, with mild residual arm weakness (NIHSS 1). This case illustrates acute stroke intervention in the setting of an ipsilateral, large, high flow AVM in a young adult and is the first such reported case to our knowledge.

INTRODUCTION

Spontaneous acute ischemic stroke in the setting of arteriovenous malformation (AVM) is a rarely reported phenomenon; in infants and children, however, AVMs are the leading cause of hemorrhagic stroke, with an incidence of 1 per 100 000.¹ Potential causes of ischemic infarction associated with AVM in young patients are multifold; however, vascular steal has been implicated as a potential source.² Whether vascular steal can indeed cause stroke is unclear. A study of 152 patients with AVMs concluded that focal neurologic deficits, especially progressive ones, are exceedingly rare and could not be attributed to a steal mechanism.³

Here we describe the case of a young patient who presented with a right middle cerebral artery (MCA) stroke, with CT perfusion evidence of an ischemic penumbra who underwent endovascular intervention for recanalization. The report highlights the unique circumstance of navigating a microcatheter and microwire into an occluded vessel in the setting of an ipsilateral, large, high flow AVM.

CASE REPORT

Presentation

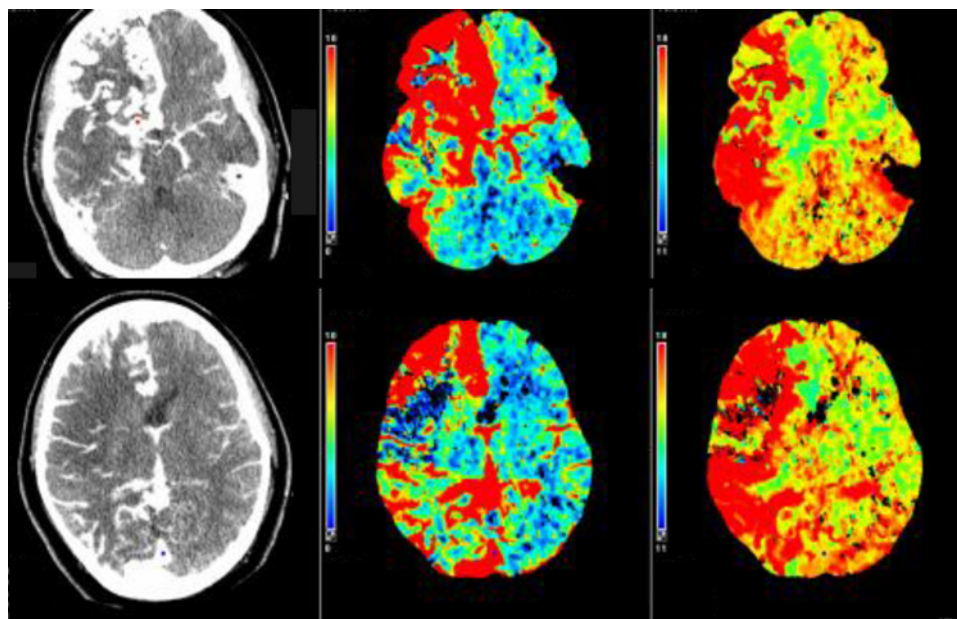
A patient in their late 20s presented with wake up stroke with left hemiparesis (more affected in the arm), facial droop, hemianopsia and hemisensory changes. National Institutes of Health Stroke Scale (NIHSS) score was 11. The patient was last seen normal in the late afternoon on the previous day. The patient was being treated medically for hypertension with metoprolol; systolic blood pressure was 130 mm Hg on admission, and the patient had adequate β blockade with a heart rate between 80 and 90 beats/min. Otherwise, the past history was medically unremarkable, including no previous history of smoking, alcohol or drug use. ECG was negative for atrial fibrillation or other abnormal rate or pathologic finding.

Because of the low clinical suspicion for the presence of a seizure disorder and the absence of abnormal laboratory findings, stroke evaluation proceeded. A cranial CT angiogram (CTA) with CT perfusion imaging showed a high time to peak value in the right frontoparietal cortex with a penumbra located anterior and posterior to an area of decreased cerebral blood volume, consistent with infarction (figure 1). CTA also demonstrated a diffuse right frontal AVM, Spetzler—Martin (SM) grade V, as well as a smaller (<2 cm), right, medial occipital periventricular AVM, SM grade II, situated posterior to the ischemic territory at risk. The high time to peak, representing perfusion deficit in the region of the large frontal AVM, was difficult to interpret, but a clear penumbra zone was present posterior to the area of decreased cerebral blood volume. MRI on admission showed a positive diffusion weighted imaging signal in the right frontal, temporal and insular lobes (figure 2).

Owing to the young age, symptoms and ischemic territory still at risk, the patient was taken emergently for endovascular intervention. The patient was outside the treatment window for intravenous tissue plasminogen activator (tPA) because of the time last seen normal. A current exclusion for tPA is symptom onset outside the 4.5 h window although some data show that such therapy may be safe for patients who wake up with stroke, with a hemorrhage rate of 4.3% and acceptable outcomes.⁴ The patient declared that

Ischemic stroke

Figure 1 Preoperative CT angiogram and CT perfusion images show high time to peak in the right middle cerebral artery distribution with a central area of completed stroke. Top panel, from left to right: non-enhanced sequence, cerebral blood flow sequence, time to peak sequence. Bottom panel: same sequences at a higher axial cut.



the AVM had been previously diagnosed but was being observed and was not causing symptoms.

Intervention

Given the young age, ongoing ischemia and territory or penumbra at risk, intervention was undertaken despite the lack of a clear time of onset and despite the presence of an AVM in the affected hemisphere. The patient consented to the procedure after associated risks and benefits were explained and alternative options of no treatment and/or heparinization were presented. The NIHSS score remained 11 at the time of treatment.

Using a standard endovascular technique, we obtained access to the right internal carotid artery (ICA) with a 9 French Concentric guide catheter (Concentric Medical-Stryker Neurovascular, Mountain View, California, USA) positioned at the C1 spinal level. On the initial right-sided ICA cerebral angiogram, a diffuse large frontal AVM and a smaller periventricular AVM were seen with a filling defect and stasis in the arterial phase of two separate MCA branches originating from a common M2 vessel, consistent with thrombus (figure 3A–E). After the

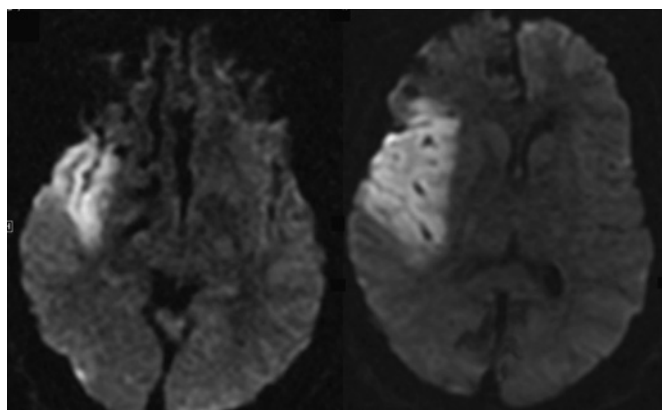


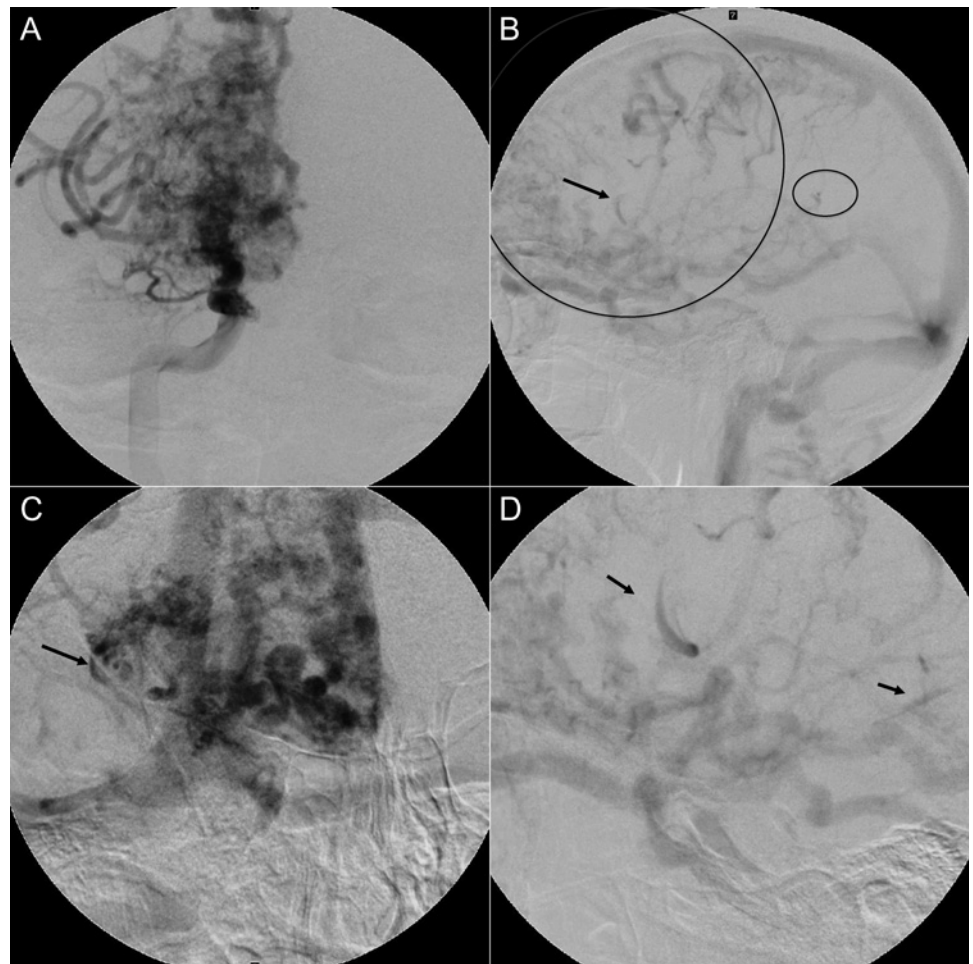
Figure 2 Preoperative MR images (higher axial cut on the right) show a diffusion weighted imaging positive stroke in the right middle cerebral artery distribution.

patient was appropriately heparinized with a weight based dose of 6000 units to an activated coagulation time of 299 s, a microcatheter (Excelsior SL-10; Boston Scientific, Natick, Massachusetts, USA) and microwire (Synchro 2 standard; Boston Scientific) were used to enter the intracranial circulation. The microwire was advanced under direct fluoroscopic guidance to the area of arterial stasis. The microcatheter was then advanced over the microwire and positioned at the M2–M3 junction, proximal to the occlusive sites. A Penumbra 0.040 microcatheter system (Penumbra Inc, Alameda, California, USA) was advanced over the microwire. A microcatheter angiogram showed slowed filling beyond the two areas of thrombus. The Synchro-2 microwire was then passed beyond the posterior occlusive lesion (figure 4). Given the smaller size of the M3 vessel, we replaced the microwire with a BMW exchange wire (Abbott Vascular, Santa Clara, California, USA) and then exchanged the entire Penumbra microcatheter system with a smaller Penumbra 0.032 microcatheter and brought the catheter closer to the lesion. The process of exchanging the larger Penumbra microcatheter with a smaller one and the associated microwire manipulation resulted in vessel revascularization (figure 5), without use of the aspirator. Further angiographic runs (figure 6) showed recanalization of both branches that previously were occluded with thrombus, albeit with slower flow (Thrombolysis in Cerebral Infarction 2b result).⁵ The entire procedure was performed under conscious sedation. The patient tolerated the procedure and remained hemodynamically and neurologically stable during the procedure.

Postprocedure

In this patient with a preprocedure NIHSS of 11 and right MCA acute ischemic stroke and AVM, CT perfusion imaging and MRI findings prompted the decision to perform endovascular intervention to recanalize the occluded arterial segments. Two days after the intervention, quantitative MR angiography (MRA), which was implemented with Non-invasive Optimal Vessel Analysis (NOVA) software (VasSol Inc., Chicago, Illinois, USA), was performed to evaluate the nature of the AVM, whether high or low flow and to document recanalization. MRA demonstrated high flow in both the right ICA territory and the

Figure 3 Digital subtraction angiogram. (A) Large right frontal arteriovenous malformation (AVM) (large encircled area in B) seen on anteroposterior (AP) angiogram. (B) Arrow indicating evidence of middle cerebral artery thrombus before treatment. Smaller circle indicates additional AVM. (C) Pretreatment thrombus seen with AVM in the AP angiogram (arrow). It is apparent how the AVM can mask the occlusive findings in a patient with stroke. (D) Pretreatment thrombus seen in two vessels that show evidence of stasis in lateral angiography (arrows show two different areas of stasis).



posterior circulation (figure 7). No preoperative MRA was performed for comparison. The postoperative evaluation of MCA recanalization was difficult to discern given the resolution of these vessels on both MRA and CTA, but flow to the right MCA measured 381 ml/min (normal range 86–230 ml/min for ages 18–40 years), which involves flow to the AVM. Notably, cerebral blood flow in the right ICA intracranially measured

958 ml/min (normal range 147–377 ml/min for ages 18–40 years). Basilar artery flow was 482 ml/min (normal range 101–231 ml/min for ages 18–40 years). The patient achieved an NIHSS score of 2 after the intervention and was started on aspirin 325 mg daily and rosuvastatin 20 mg daily. CT perfusion imaging showed improvement of the ischemic penumbra in the posterior temporo-occipital-parietal region (figure 8). The stroke did not enlarge on post-treatment MRI.

While in hospital, the patient underwent an evaluation to determine the stroke origin. An ECG with bubble study did not show right to left shunt. A hypercoagulability panel showed normal values for protein C and S and cardiolipin antibodies. Factors 8, 9, 11 and 12 activity levels were within normal limits. The antithrombin III level was low at 71% of normal (normal acceptable range is 80–120% of the normal value). However, antithrombin III levels are usually 50% of normal or less in type I or II antithrombin deficiency (which this patient did not demonstrate).⁶

The patient was discharged home 7 days after admission. Evaluation 1 month post-treatment disclosed a nearly complete recovery with an NIHSS of 1 due to very mild residual arm weakness.

DISCUSSION

Imaging from this case illustrates potential difficulties in localizing occluded vessels during endovascular acute stroke intervention in the setting of a complex intracranial lesion (see figures 3 and 4). An understanding of vascular anatomy is requisite for proper navigation to occluded vessels during acute stroke

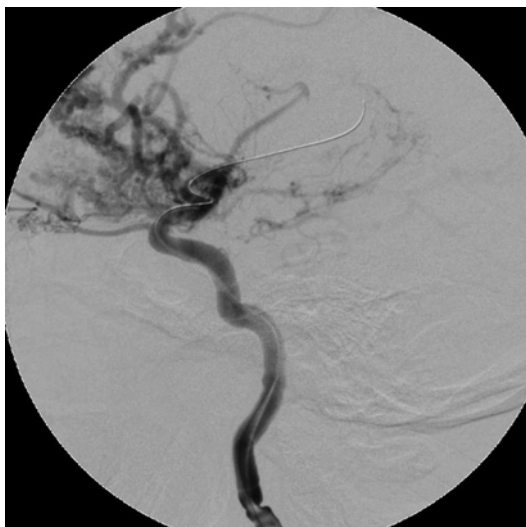
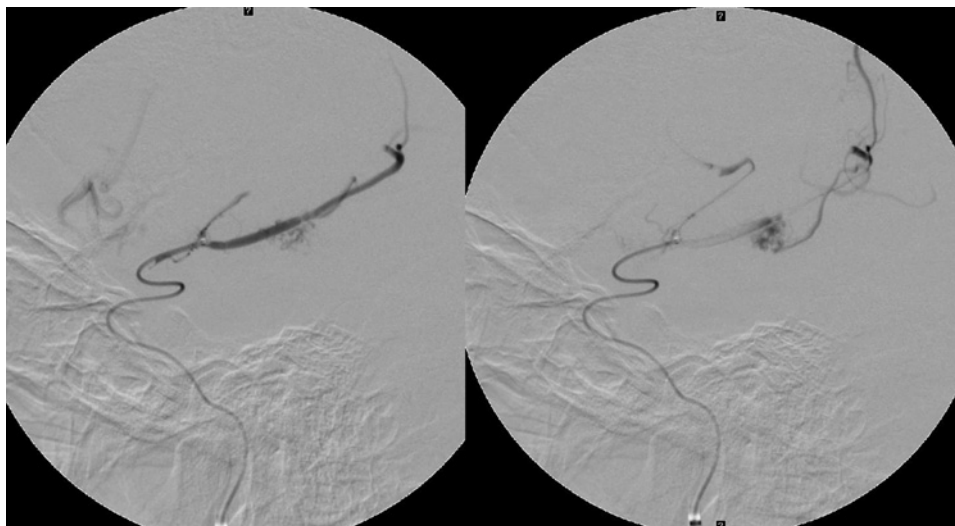


Figure 4 Angiogram showing the microwire further out in the occluded vessel.

Ischemic stroke

Figure 5 Microcatheter runs in the early (left) and later (right) arterial phases show flow distal to the occluded segments.



intervention, especially in the setting of nearby dilated and distracting AVM vessels.

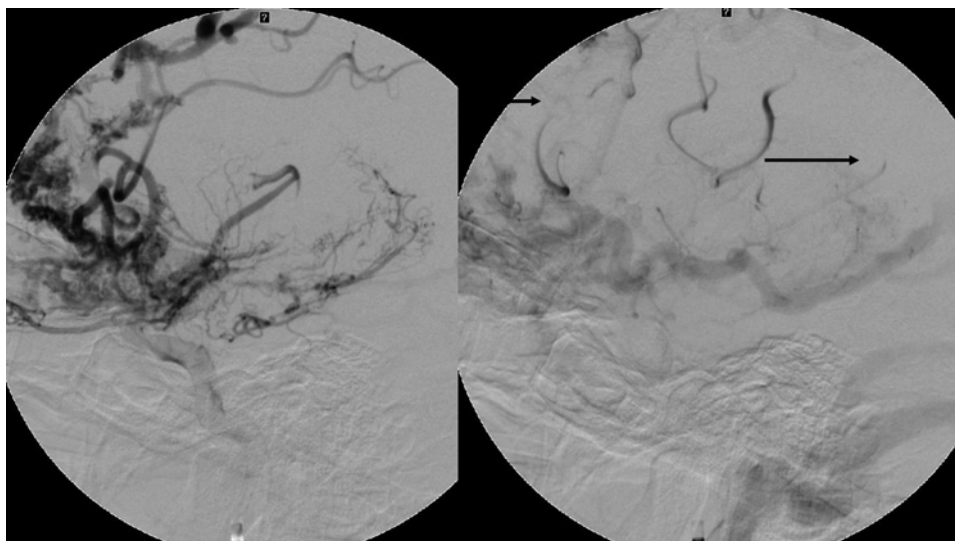
Scattered cases of stroke treatment in the setting of AVM have been described. Previously, iatrogenic MCA (M1) thrombus in the setting of an AVM embolization procedure was reported.⁷ In that case, intravenous tPA was administered to restore flow. Another report described moyamoya-like patterns of occlusion and infarction in the setting of AVM.⁸ Vascular occlusions could occur with acquired dysplastic vessel pathology that may be induced in the setting of high flow states, such as an AVM.⁸⁻¹⁰ According to several reports, these occlusions frequently occurred in the proximal artery that supplied the AVM, only on the ipsilateral side, and were possibly related to turbulent and/or flow induced intimal hyperplasia.⁸⁻¹⁰ In contrast with moyamoya-like patterns,⁸ the occluded MCA segments in the present case occurred distal to and between branches feeding the AVM. These branches supplied normal brain, as demonstrated by the perfusion deficit in the territory of the angular MCA branch.

The source of stroke in this patient remains cryptogenic. In young patients, ischemic strokes are often seen in our practice in those patients who have moyamoya disease or those who

experience arterial dissections of the brachiocephalic tree. One explanation for stroke in this patient is vascular steal, which results in shunting of blood away from normal vessels to the high flow vessels supplying the AVM. This shunting may create slower flow and the potential for stasis of blood within the vessels supplying normal brain. However, there is speculation as to whether this pathophysiology is truly responsible for neurologic findings in AVM patients.³ Another potential cause, hypercoagulability, is less likely given the near normal values of antithrombin levels. A cardioembolic source is suspected.

The MCA filling defect may have occurred due to the AVM's high flow, which increased the percentage of cardiac output seen in the ipsilateral ICA. A greater proportion of cardiac output equates to increased probability for thrombus of cardioembolic sources to end in this vascular territory. We would expect that when thrombus embolizes from a systemic source, it would go toward the higher flow vessels within the AVM, rather than to MCA vessels supplying normal brain tissue, as was seen here. In this case, the thrombus may have embolized to the AVM vessels but washed away due to the high flow and dilated nature of the vessels in the AVM. As an AVM does not supply normal brain tissue, potential

Figure 6 Post-treatment lateral angiograms (left, early; right, late arterial phase) showing Thrombolysis in Cerebral Infarction 2b perfusion. Flow distal to the previously occluded segments can be seen on the right (arrows).



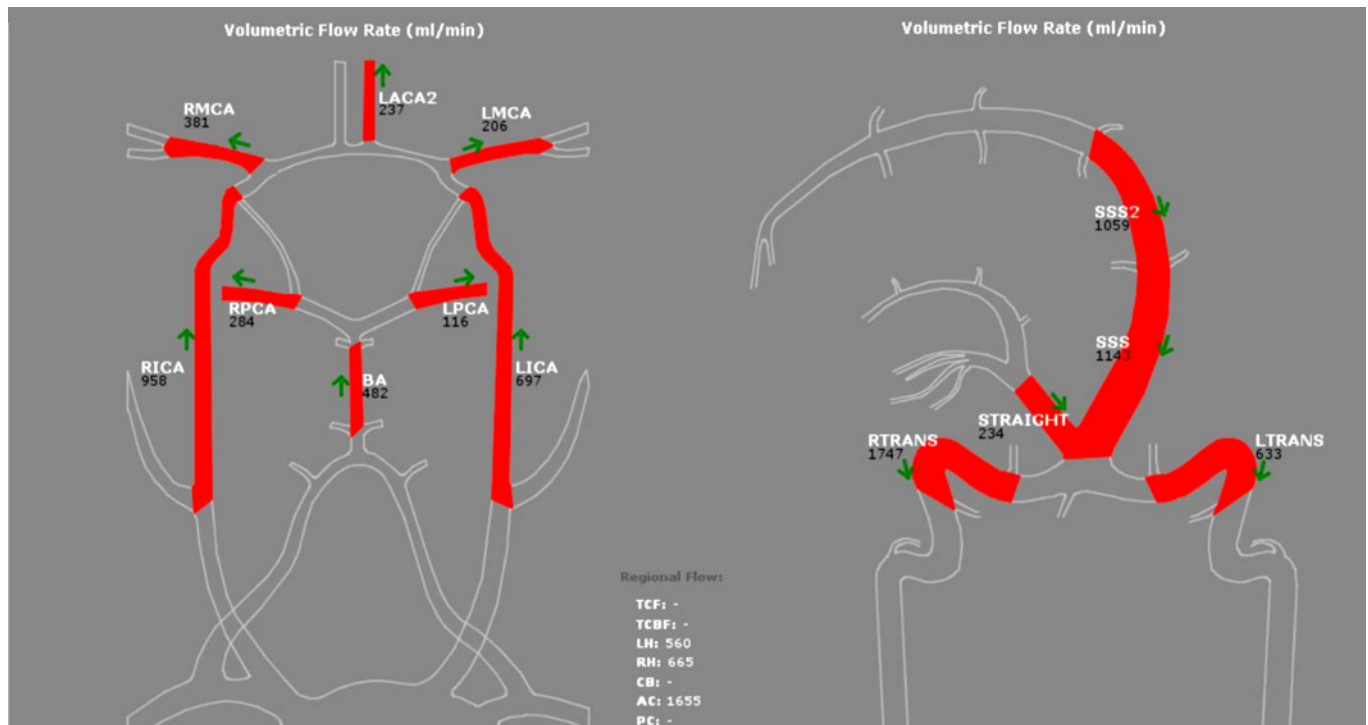


Figure 7 Quantitative MR angiography (Non-invasive Optimal Vessel Analysis (NOVA)), arterial (left) and venous (right), shows high cerebral blood flow in the right hemisphere compared with the left hemisphere and much elevated over the physiologic range ($\cong 150$ ml/min).

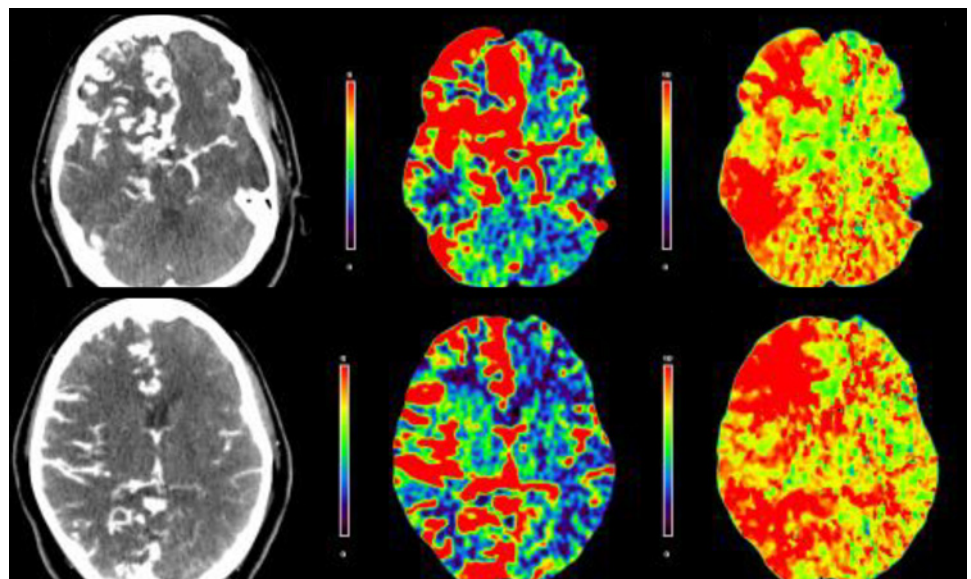
thromboemboli to AVM vessels should not infarct normal brain and should remain clinically silent.

CONCLUSION

We have presented a unique case illustrating acute MCA stroke intervention in a patient with an unruptured, untreated SM grade V AVM. This case of acute ischemic stroke in a young patient was confounded by an ipsilateral AVM with MCA occlusion and an unknown time of stroke symptom onset. The patient presented with a wake up stroke. Although these events are outside the traditional window for intravenous tPA therapy, attempts at intervention in the setting of wake up stroke are made at our

hospital,¹¹ and we are further investigating interventions in this setting by conducting a prospective randomized trial. The patient was felt to have salvageable brain tissue on the basis of CT perfusion imaging, and these findings extended the time window for treatment in this case. Successful MCA recanalization was accomplished via mechanical thrombectomy ipsilateral to a known but asymptomatic and untreated large AVM. In this case, the use of diagnostic angiography was essential in that it improved the ability to discern an occluded vessel in an ischemic stroke patient with an AVM ipsilateral to the occluded segment; MRA and CTA are largely unreliable in discerning thrombus and/or an occluded vessel in the setting of an ipsilateral AVM.

Figure 8 Postprocedure CT perfusion images during hospitalization show some resolution of the perfusion defect, mostly in the angular gyrus. Top panel, from left to right: non-enhanced sequence, cerebral blood flow sequence, time to peak sequence. Bottom panel: same sequences at a higher axial cut.



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Competing interests LNH receives grant/research support from Toshiba; serves as a consultant to Abbott, Boston Scientific,* Cordis, Micrus and WL Gore; holds a financial interest in Access Closure, Augmenix, Boston Scientific,* Claret Medical Inc, Micrus and Valor Medical; has a board/trustee/officer position with Access Closure, Claret Medical Inc and Micrus (until September 2010); belongs to the Abbott Vascular speakers bureau; and receives honoraria from Bard, Boston Scientific,* Cordis, Memorial Healthcare System, Complete Conference Management, SCAI and Cleveland Clinic. EIL receives research grant support (principal investigator: Stent-Assisted Recanalization in acute Ischemic Stroke, SARIS), other research support (devices) and honoraria from Boston Scientific* and research support from Codman and Shurtleff Inc and ev3/Covidien Vascular Therapies; has ownership interests in Intratech Medical Ltd and Mynx/Access Closure; serves as a consultant on the board of Scientific Advisors to Codman and Shurtleff Inc; serves as a consultant per project and/or per hour for Codman and Shurtleff Inc, ev3/Covidien Vascular Therapies and TheraSyn Sensors Inc; and receives fees for carotid stent training from Abbott Vascular and ev3/Covidien Vascular Therapies. He receives no consulting salary arrangements. All consulting is per project and/or per hour. AHS has received research grants from the National Institutes of Health (coinvestigator: NINDS 1R01NS064592-01A1, Hemodynamic induction of pathologic remodeling leading to intracranial aneurysms) and the University at Buffalo (Research Development Award); holds financial interests in Hotspur, Intratech Medical, StimSox and Valor Medical; serves as a consultant to Codman and Shurtleff Inc, Concentric Medical, ev3/Covidien Vascular Therapies, GuidePoint Global Consulting and Penumbra; belongs to the speakers bureaus of Codman and Shurtleff Inc and Genentech; serves on an advisory board for Codman and Shurtleff; and has received honoraria from American Association of Neurological Surgeons courses, an Emergency Medicine Conference, Genentech, Neocore Group LLC and from Abbott Vascular and Codman and Shurtleff

Inc for training other neurointerventionists in carotid stenting and for training physicians in endovascular stenting for aneurysms. He receives no consulting salary arrangements. All consulting is per project and/or per hour. (*Boston Scientifics neurovascular business has been acquired by Stryker.)

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