Rolandic arteriovenous malformations: improvement in limb function by IBC embolization.

A J Fox, J P Girvin, F Viñuela and C G Drake

AJNR Am J Neuroradiol 1985, 6 (4) 575-582
http://www.ajnr.org/content/6/4/575

Rolandic Arteriovenous Malformations: Improvement in Limb Function by IBC Embolization

Allan J. Fox[1, 2]
John P. Girvin[2]
Fernando Viñuela[1, 2]
Charles G. Drake[2]

Three patients with arteriovenous malformations in the rolandic region and significant limb deficit showed virtually complete functional recovery after awake operative embolization of most of the malformations using isobutyl-2 cyanoacrylate. Two of these patients, with functionally useless hands, had sustained the deficits months earlier as the result of a specific brain-damaging event: one as a result of surgery and the other as a result of a hemorrhage. Both of these showed significant return of function during the awake operative embolization procedure. The other patient had had progressive leg weakness over a 2 year period. The theory of steal phenomenon as an explanation for progressive neurologic deficits in association with large arteriovenous malformations must be extended to explain apparently stable deficits after some brain trauma (surgery or hemorrhage). These results suggest that some patients with arteriovenous malformations and without clinical deficits who are near a critical level of "near ischemia" may be thrown out of balance by an acute interceding event.

Progressive neurologic deficits in patients with large arteriovenous malformations (AVMs) have been explained as the phenomenon of ischemic steal [1–6]. This theory is that an AVM acts as a sump, successfully competing for regional blood flow by diverting flow from the surrounding tissue. The relative ischemia in the brain immediately surrounding an AVM may increase with time as flow to the AVM increases, thus explaining the progressive nature of the deficit. For AVMs of large size in important functional brain regions, progressive deficit due to steal has been considered to be an acceptable indication for partial treatment by sphere embolization [7–9], and stabilization or even reversal of deficits by embolization has been reported [7, 8].

Our series of patients with large, mainly rolandic AVMs treated by awake intraoperative embolization with isobutyl-2 cyanoacrylate (IBC) [10] includes three patients in whom severe limb impairment was reversed. Two of these had stable deficits; one had a progressive deficit. The differences in the etiology of the deficits, the course of improvement during and after treatment, and the angiographic correlations allowed us to extend what previous reports have referred to as the "steal phenomenon."

Materials and Methods

Over a 2½ year period, 15 patients with moderate or large rolandic and near-rolandic AVMs underwent 24 embolization procedures at University Hospital, London, Ontario. Eighteen embolization procedures were carried out by directly catheterizing vessels at craniotomy. In 17 of 18 embolizations local anesthesia was used in awake patients using a technique reported elsewhere [10]. The other six embolization procedures were carried out using the calibrated-leak balloon transfemoral approach [11, 12]. During the 24 embolization procedures in 15 patients, 62 IBC injections were done into the arterial feeders of the AVMs. Five patients had limb deficits; two had functionally useless hands, one had severe paresis of the leg with a foot drop, one had moderate sensory and motor deficit in the arm and leg, and one had

Received July 25, 1984; accepted after revision December 7, 1984.

Presented in part at the annual meeting of the American Society of Neuroradiology, San Francisco, June 1983.

[1] Department of Diagnostic Radiology, University Hospital and University of Western Ontario, Box 5339, Postal Stn. "A," London, Ontario N6A 5A5, Canada. Address reprint requests to A. J. Fox, Neuroradiology Section.

[2] Department of Clinical Neurological Sciences, University Hospital and University of Western Ontario, London, Ontario N6A 5A5, Canada.

AJNR 6:575–582, July/August 1985
0195–6108/85/0604–0575

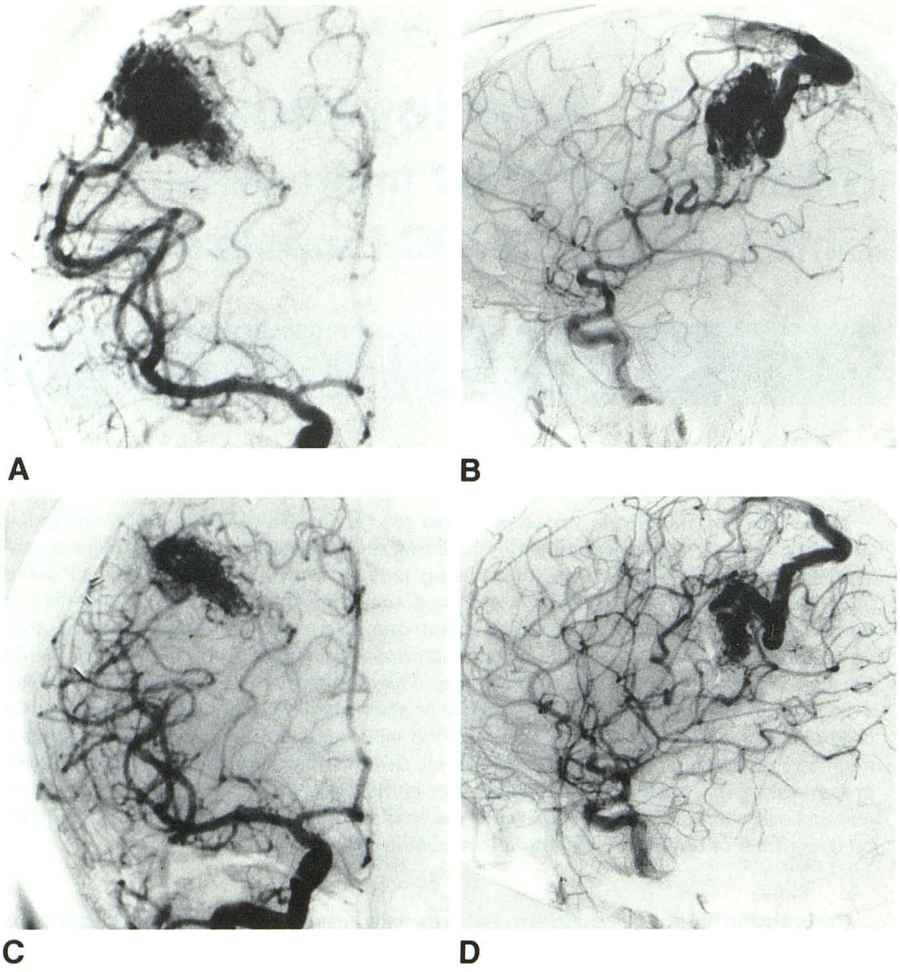


Fig. 1.—Case 1. Right rolandic AVM, residual from previous surgery. Anteroposterior (AP) (**A**) and lateral right (**B**) carotid angiograms before embolization (10 months after partial resection). AVM is fed by middle cerebral cortical branches, cortical branches from anterior cerebral via leptomeningeal collaterals, and lenticulostriate arteries. **C** and **D**, 1 week postembolization. Residual nidus of AVM is fed by small middle and anterior cerebral branches and lenticulostriate artery. Big draining vein is smaller than before embolization.

moderate left hemiparesis. All three patients with severely impaired limbs showed return to virtually normal function either during the embolization procedure or subsequent to it. In the other two patients, one had a mild residual arm deficit in late follow-up, while the other had a permanent foot drop and decreased finger dexterity after intraoperative embolization and AVM resection. The three patients who virtually improved completely from their severe deficits are the subject of this report.

Case Reports

Case 1

A 28-year-old man (Case 6 in Girvin et al. [10]) was seen elsewhere with focal sensory motor seizures of 1 year's duration due to a right rolandic AVM. Two months later he had an incomplete surgical approach (details uncertain) to the posterior aspects of the AVM at another institution. Postoperatively he was left with a functionally useless left hand that was thought to be from loss of both cortical sensation modalities and motor ability. For the next 9 months the patient noticed no continuing recovery from surgery, and it was believed he had a stable post-operative deficit likely caused by surgical trauma or local ischemia.

Angiography on admission to our institution 10 months after surgery disclosed a residual, moderate-sized AVM in the right rolandic region (figs. 1A and 1B) fed by both middle and anterior cerebral branches.

The patient underwent awake embolization at craniotomy with appropriate prior stimulation and clinical monitoring. Two middle cerebral feeders and one anterior cerebral feeder coming over the top were successively catheterized, monitored angiographically and functionally, and embolized with IBC. A total of 1.2 ml of IBC was injected. There was functional improvement of the patient's left hand during the procedure, and he was able to move his hand and fingers successively better after each of the three IBC injections. Postoper-

atively he exhibited very transient impairment of hand function, although at a level still much better than before surgery. The next morning the improvement noted during surgery was again seen, leaving him, on discharge 1 week later, with nearly normal function in his hand.

Postoperative angiography (figs. 1C and 1D) 1 week after embolization showed that at least half of the nidus of the AVM was obliterated by the IBC. A small remaining part of AVM was filled mostly by a lenticulostriate artery feeder. Draining veins of similar configuration but smaller size than before embolization remained.

Case 2

A 28-year-old woman (case 9 in Girvin et al. [10]) had focal seizures involving the left leg beginning 7 years before admission. These became intractable over time. For 1 year before admission she developed a progressively increasing motor deficit, and on admission to our institution she had a left hemiparesis with severe impairment of function in the left leg including a foot drop. Of interest, she reported occasional 30-sec periods of normal function, the last one being 1 month before admission. Angiography (figs. 2A–2D) showed a large right parasagittal rolandic AVM fed mainly by anterior cerebral branches, although leptomeningeal collateral feeders from middle cerebral and posterior cerebral branches to the anterior cerebral circulation were seen also.

The patient underwent awake operative embolization through a craniotomy. Three anterior cerebral branches were catheterized and embolized with a total of about 1.5 ml of IBC mixture. A remaining anterior cerebral branch was not embolized because increasing weakness was noted intraoperatively, although it was realized that this was likely caused by retraction of the medial aspect of the hemisphere during the embolization. Postoperatively she developed a hemiplegia within 24 hr, with rapid improvement 1 week later. Angiography (figs. 2E–2H) 1 week before surgery showed a reduction in the major part of the AVM nidus. Over the next few weeks her hemiparesis virtually cleared. She regained the ability to hop on her foot and was able to jog some miles a day. A 6 month follow-up examination showed essentially normal function on the left side, especially her foot. Seizures were markedly reduced in frequency. Angiography at this stage (figs. 3A–3D) showed a change in the filling pattern of the residual AVM compared with 1 week after the operative embolization. Transdural (meningeal) feeders to the AVM residua were also present.

The patient returned 1 year after treatment. For about the previous 3 months her leg function had gradually worsened with reappearance of the foot drop. Her motor power was still better than before the original operative embolization but clearly had worsened since the last follow-up. Angiography disclosed no significant change in comparison with the previous angiography. A decision was made to reoperate to embolize the anterior cerebral branch not embolized previously. Surgery was performed under general anesthesia since cortical mapping had been done previously, and the intention was to embolize just one branch. The procedure was carried out, and the patient had some improvement in her left lower limb function by the next day. Over the next week there was further rapid improvement in the motor function of the foot. Angiography 1 week after the second embolization (figs. 3E–3H). showed a further decrease in the residual AVM nidus. Improvement of the foot continued, though 6 months later she noticed weakness beginning again.

Case 3

A 17-year-old girl (case 10 in Girvin et al. [10]) had headaches of many years' duration, occasionally accompanied by nausea and vomiting, as well as presumed right focal sensory seizures. Three months before admission to our institution she suffered a coma-producing intracerebral hemorrhage associated with right hemiplegia and dysphasia. This led to the diagnosis of a large left rolandic AVM fed by middle cerebral arteries, including some leptomeningeal collaterals from the anterior cerebral artery (figs. 4A–4C). On examination she demonstrated residual sensorimotor hemiparesis, especially impairment of fine finger movements. Her hand did not improve during the previous month, and she was considered to have a stable posthemorrhagic deficit.

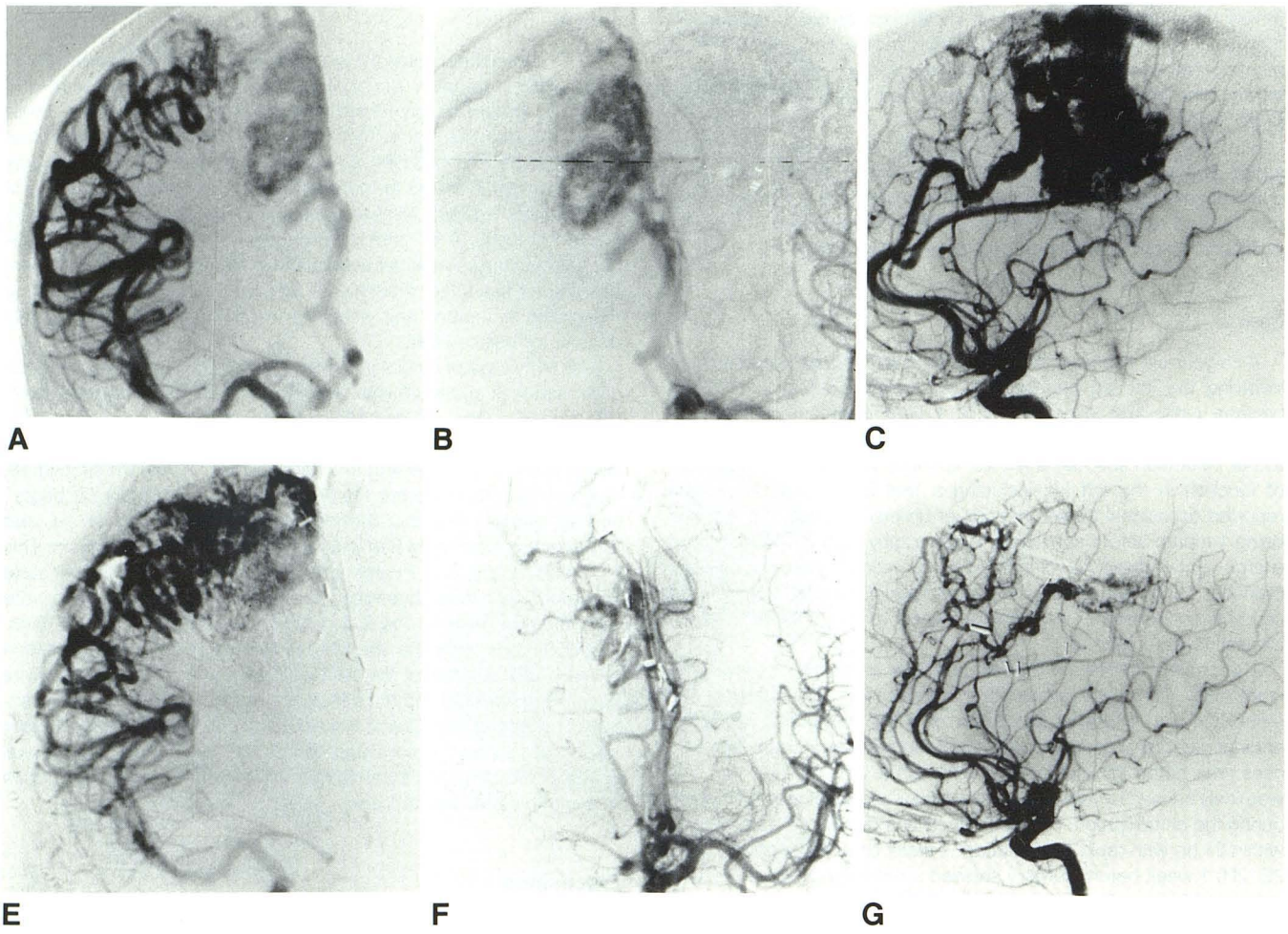
The first stage of embolization was performed via the transfemoral calibrated-leak approach with two injections of IBC into middle cerebral branches (figs. 4D and 4E). The patient remained neurologically unchanged, and 1 week later underwent awake operative embolization of five branches feeding the malformation. During the procedure, she was able to move her hand much better than before it began, and her grip at the end of the procedure was strong. Over the next day she developed right hemiplegia and asphasia due to edema. This improved rapidly, and 1 week after embolization her functional state was better than it had been preoperatively, and she was able to write with her right hand for the first time in 3 months. Angiography (figs. 4F–4H) 1 week after the operative embolization showed obliteration of most of the nidus of the AVM. On 6 month follow-up, she was functioning normally without deficit, and the angiograms were essentially unchanged from those 1 week after surgery. She was jogging some miles a day; she continued to function normally with occasional epileptic seizures. Ten months after embolization she had a sudden fatal intracerebral hemorrhage.

Discussion

The progressive deficit that may arise in some AVM patients, as particularly epitomized by our case 2, is considered to result from increasing relative *ischemia* [1–6]. If this phenomenon was seen only in older patients then it might be proposed to be caused by the "aging" process, in which neurons are progressively lost with advancing age, thus making what was at one time subclinical ischemia important clinically. However, the fact that it occurs in young patients would seem to rule out this explanation.

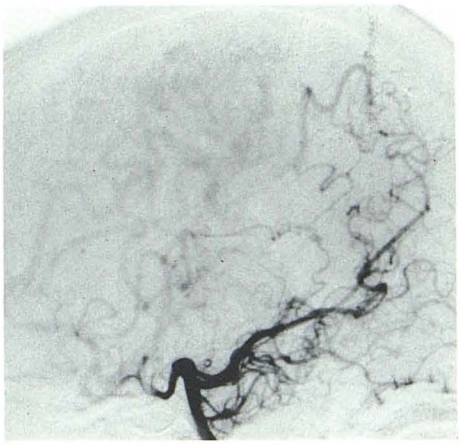
Given the fact that relative ischemia is at the root of the deficit, the deficit occurs progessively rather than in a stuttering or strokelike fashion. Thus, the intuitive assumption is that at any given time a part of the deficit is reversible and not dependent on a combination of neuronal dropout and/or gliosis. Rather, the deficit is due to a reversibly altered metabolic state, thus bringing about dysfunction of the population of neurons.

The angiographic picture of a prominent "sump effect" of large arterial feeders going only to the AVM with poor filling of normal brain vessels [13, 14] has been suggested as a correlate of the "steal phenomenon" [15, 16]. It is interesting,

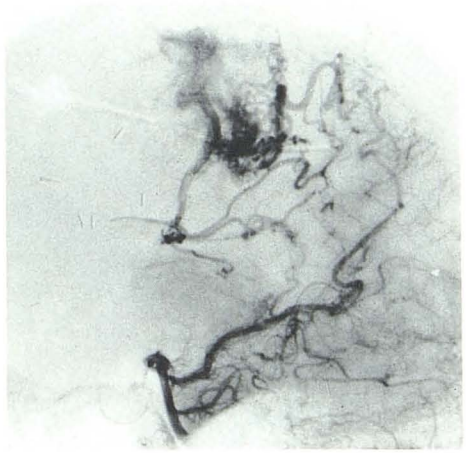


however, that while case 2 initially had a prominent angiographic sump effect (figs. 2A–2D), the residual AVM on the 6-month follow-up angiograms (figs. 3A–3D) did not show significant sump effect when the patient was clinically well, and the improved pattern did not change when she again began to worsen. The lower blood pressure in large feeding arteries to AVMs is assumed to induce maximal active dilatation of arteriolar capacity with a constant effort to maintain adequate flow in the surrounding regions [5].

The idea that patients without clinical symptoms of ischemia may actually be quite near a critical-level pressure flow [5] needs further discussion in light of cases 1 and 3. The mode of onset of the deficits in these cases is not easily explained by steal without invoking the existence of an asymptomatic critical level that may be thrown out of balance by an acute interceding event. In case 1, the neurologic deficit was a complication of surgery 10 months earlier, there was no continuing recovery for nine months, and stable permanent brain damage was presumed to be from either surgical trauma or ischemia associated with clipping of some of the branches to the AVM. Case 3 had no deficit until the sudden onset of coma-producing hemorrhage. The residual deficit after initial improvement was stable for some weeks before the embolization and was presumed to represent permanent brain damage due to hemorrhage. The remarkable improvement in limb function in these two cases necessitates rethinking the etiology of deficits of sudden onset in patients with AVMs. Some deficits considered to be irreversible, stable, and permanent, as was presumed in these two cases, may be reversible to some degree. The steal reversal can be instantaneous, as was seen intraoperatively in cases 1 and 3. Presumably these two patients had been near a critical level of pressure and flow in the brain near the AVMs and moved beyond that level due to surgical trauma (case 1) on hemorrhage (case 3). The degree of improvement in these cases was a pleasant surprise in the outcome of treatment.



D



H

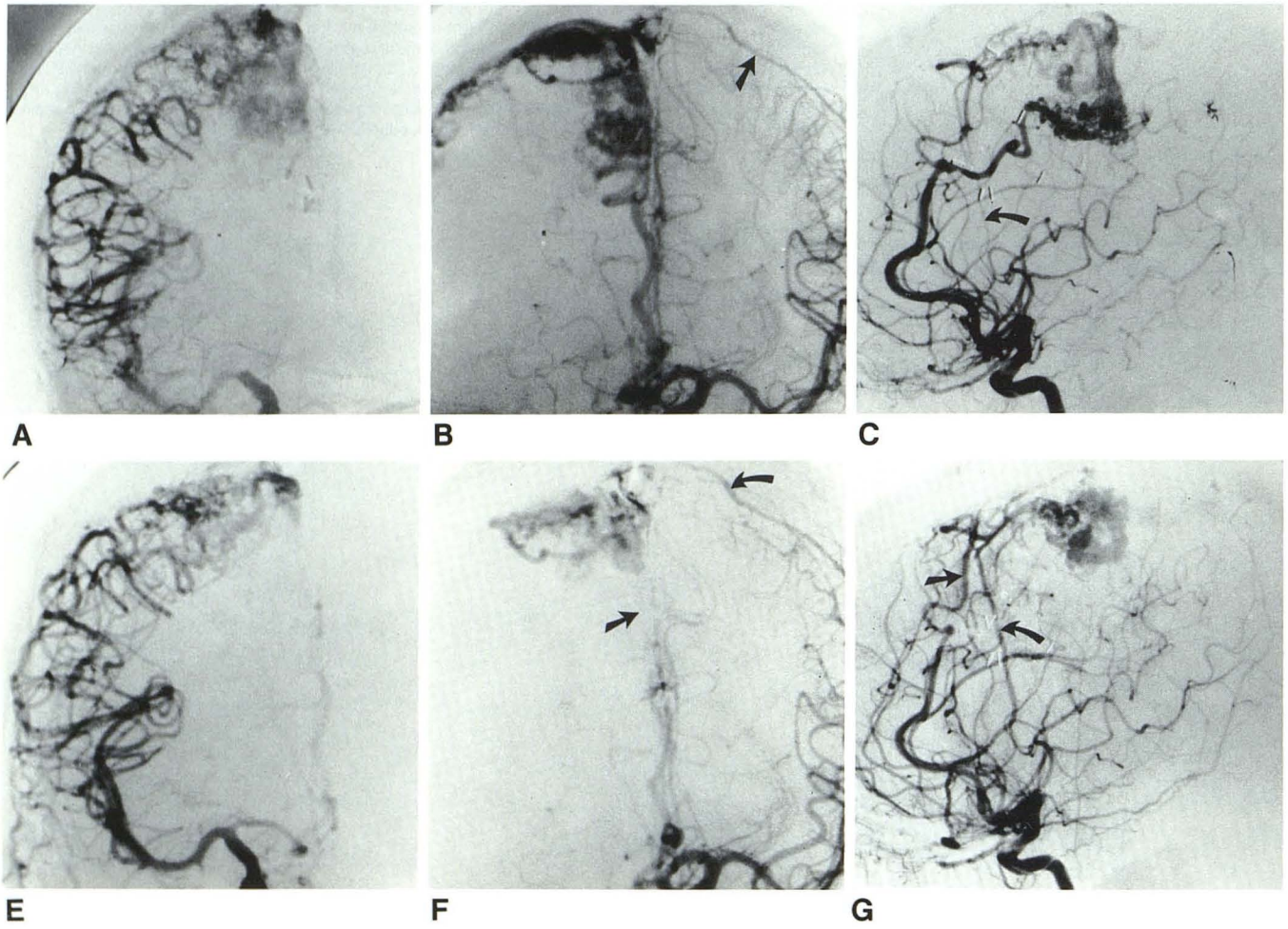
Fig. 2.—Case 2. Large right rolandic parasagittal AVM. AP right carotid (**A**), AP left carotid (**B**), lateral left carotid (**C**), and lateral vertebral (**D**) angiograms before treatment. Malformation is fed essentially by multiple anterior cerebral branches, though some leptomeningeal collateral from middle and posterior cerebral is also seen feeding lesion. AP right carotid (**E**), AP left carotid (**F**), lateral left carotid (**G**), and left vertebral (**H**) angiograms 1 week postembolization. Marked reduction of filling of AVM nidus after embolization of three anterior cerebral feeders. Middle cerebral leptomeningeal collateral pattern remains prominent, as does posterior cerebral pattern, which supplies more of AVM than before.

Obliteration of much of the AVM nidus with IBC embolization in these three cases must have changed significantly the abnormal perfusion physiology in the region of the brain surrounding the AVM. The improvement in the perfusion of brain is likely similar to that recorded with changes measured by fluorescein angiography, microregional blood flow measurements [2], and pressure-flow studies related to AVM resection [5].

For case 2, in fact, significant reversibility of the deficit was predictable on the basis of the fact that as recently as 1 month before initial treatment the patient had at least a 30 sec period during which she was able to use her leg quite normally, only to lose it completely at the termination of this very brief period. Such instances had also occurred over the previous 6 months. The incidence of these episodes of "normality" is uncertain from other reported series.

Another possible explanation for the progressive deficit is some type of subclinical, focal status epilepticus that renders the involved area functionless, or partly functionless, on the basis of long-lasting postictal depression. Electroencephalograms are notoriously abnormal in cases of large AVMs, but in our experience they have usually failed to show clear-cut evidence of epileptogenic abnormalities. On the other hand, scalp electroencephalograms may very well fail to show a very focal epileptogenic abnormality, particularly when it exists deep in a sulcus or on the medial aspect of a hemisphere, based on simple volume-conduction theory. Although subclinical status epilepticus with a prolonged interictal postictal state would provide an alternate explanation for the deficit, we believe on the basis of the available evidence that this is unlikely.

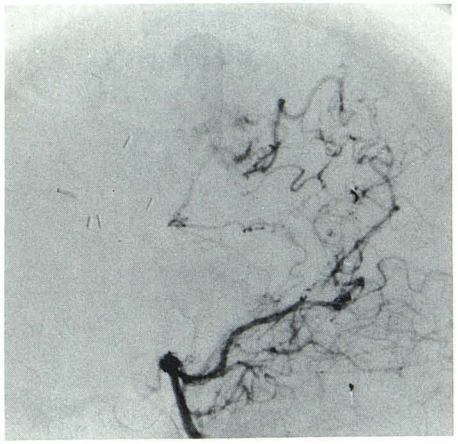
Neurologic dysfunction in cases of cerebral AVMs may, alternatively, not result from ischemic arterial steal but rather from venous hypertension (or a combination of the two). In fact, case 1 showed that the venous drainage had diminished after embolization. This was presumably a direct correlate of



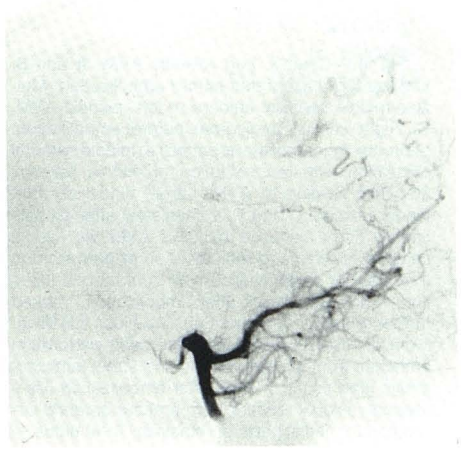
decreased venous outflow and pressure due to the embolization, which may in turn have correlated well with the neurologic improvement.

REFERENCES

1. Amacher AL, Allcock JM, Drake CG. Cerebral angiomas: sequelae of surgical treatment. *J Neurosurg* **1972**;37:571–575
2. Feindel W, Yamamoto YL, Hodge CP. Red cerebral veins and the cerebral steal syndrome: evidence from fluorescein angiography and microregional blood flow by radioisotopes during excision of an angioma. *J Neurosurg* **1971**;35:167–178
3. Michelsen WJ. Natural history and pathophysiology of arteriovenous malformations. *Clin Neurosurg* **1979**;261:307–313
4. Norlen G. Arteriovenous aneurysms of the brain: a report of ten cases with total removal of the lesion. *J Neurosurg* **1949**;6:475–494
5. Nornes H, Grip A. Hemodynamic aspects of cerebral arteriovenous malformations. *J Neurosurg* **1980**;53:456–464
6. Pool JL. Treatment of arteriovenous malformations of the cerebral hemispheres. *J Neurosurg* **1962**;19:136–141
7. Kusske JA, Kelly WA. Embolization and reduction of the "steal" syndrome in cerebral arteriovenous malformations. *J Neurosurg* **1974**;40:313–321
8. Luessenhop AJ, Presper JH. Surgical embolization of cerebral



D



H

Fig. 3.—Case 2. AP right carotid (**A**), AP left carotid (**B**), lateral left carotid (**C**), and left vertebral (**D**) angiograms 6 months postembolization. Size of middle cerebral leptomeningeal collateral feeders is markedly reduced (**A**) in comparison with fig. 2E, and contribution from posterior cerebral (**D**) is also smaller than before (fig. 2H). Feeders at this time include transdural meningeal vessel (*straight arrow*) that had enlarged since previous angiography and that on lateral view (*curved arrow*) is identified as meningeal branch of ophthalmic artery. AP right carotid (**E**), AP left carotid (**F**), lateral left carotid (**G**), and lateral left vertebral (**H**) angiograms 1 week after second embolization, with one anterior cerebral branch injected with IBC. Marked further decrease in residual AVM filling. Apart from one anterior cerebral feeder (*straight arrows*), anterior cerebral feeding to malformation is virtually gone, as is posterior cerebral part (*H*). Ophthalmomeningeal supply (*curved arrows*) and middle cerebral feeding (**E**) are still present.

arteriovenous malformations through internal carotid and vertebral arteries. Long-term results. *J Neurosurg* **1975**;42:443–451

9. Stein BM, Wolpert SM. Arteriovenous malformations of the brain. II: Current concepts and treatment. *Arch Neurol* **1980**;37:69–75
10. Girvin JP, Fox AJ, Viñuela F, Drake CG. Intraoperative embolization (IBC) of cerebral arteriovenous malformations in the awake patient. *Clin Neurosurg* **1984**;31:188–247
11. Debrun G, Viñeula F, Fox A, Drake C. Embolization of cerebral arteriovenous malformations with bucrylate: experience in 46 cases. *J Neurosurg* **1982**;56:615–627
12. Viñuela F, Fox AJ. Interventional neuroradiology and the management of arteriovenous malformations and fistulas. *Neurol Clin* **1983**;1:131–154
13. Boulos R, Kricheff II, Chase NE. Value of cerebral angiography in the embolization treatment of cerebral arteriovenous malformations. *Radiology* **1970**;97:65–70
14. Wolpert SM, Stein BM. Factors governing the course of emboli in the therapeutic embolization of cerebral arteriovenous malformations. *Radiology* **1979**;131:125–131
15. Malik GM. Surgical treatment of large cerebral arteriovenous malformations. In: Smith RR, Haerer AF, Russell WF, eds. *Vascular malformations and fistulas of the brain.* New York: Raven, **1982**:77–99
16. Spetzler RF, Wilson CB, Weinstein P, Mehdorn M, Townsend J, Telles D. Normal perfusion pressure breakthrough theory. *Clin Neurosurg* **1978**;25:651–672

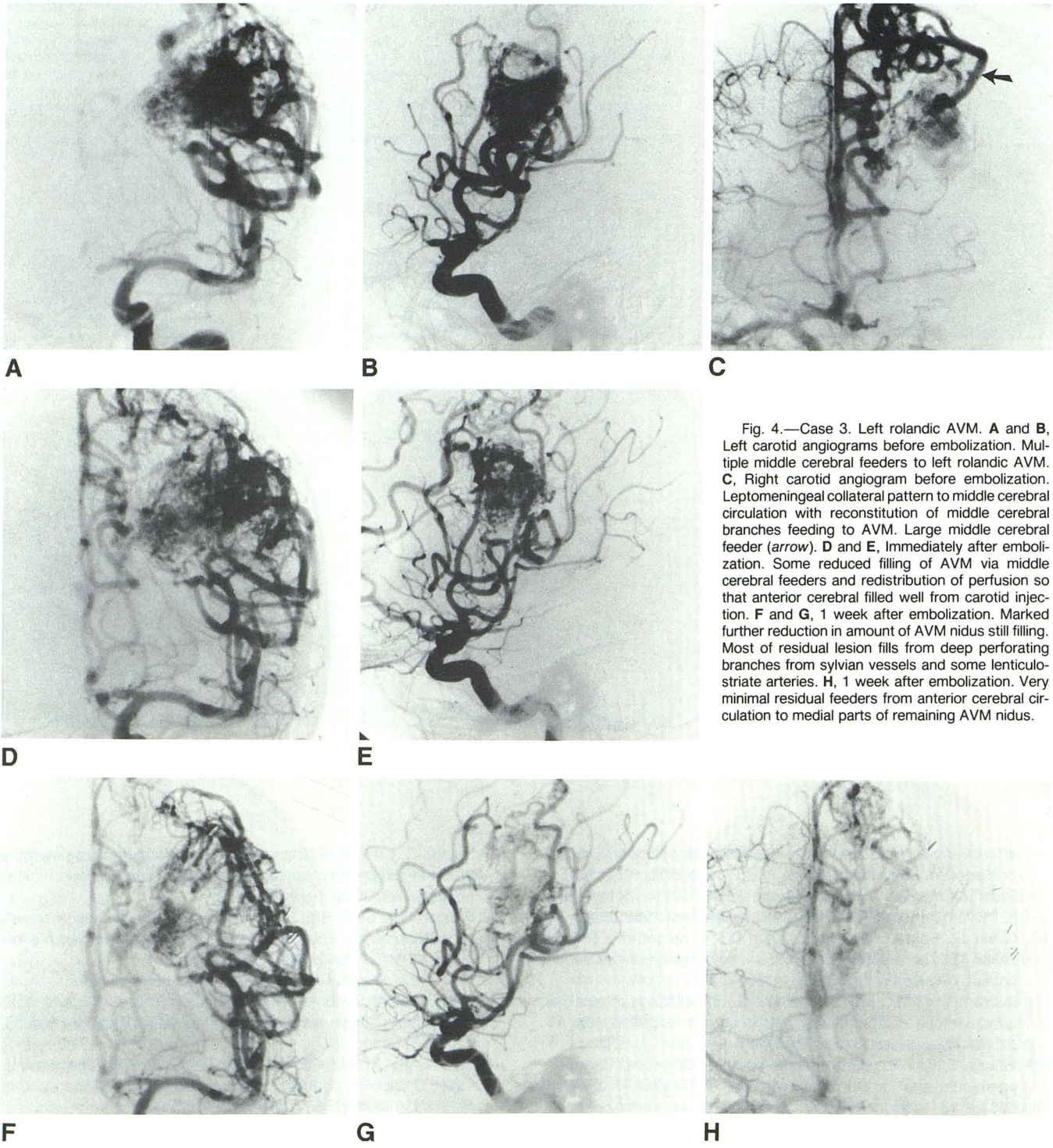


Fig. 4.—Case 3. Left rolandic AVM. **A** and **B**, Left carotid angiograms before embolization. Multiple middle cerebral feeders to left rolandic AVM. **C**, Right carotid angiogram before embolization. Leptomeningeal collateral pattern to middle cerebral circulation with reconstitution of middle cerebral branches feeding to AVM. Large middle cerebral feeder (*arrow*). **D** and **E**, Immediately after embolization. Some reduced filling of AVM via middle cerebral feeders and redistribution of perfusion so that anterior cerebral filled well from carotid injection. **F** and **G**, 1 week after embolization. Marked further reduction in amount of AVM nidus still filling. Most of residual lesion fills from deep perforating branches from sylvian vessels and some lenticulostriate arteries. **H**, 1 week after embolization. Very minimal residual feeders from anterior cerebral circulation to medial parts of remaining AVM nidus.