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Acquired Pial Arteriovenous Fistula Following Cerebral Vein Thrombosis

Constantine C. Phatouros, MBBS, FRACR; Van V. Halbach, MD; Christopher F. Dowd, MD; Todd E. Lempert, MD; Adel M. Malek, MD, PhD; Philip M. Meyers, MD; Randall T. Higashida, MD

Background—We report a unique case of an acquired pial arteriovenous fistula occurring after an asymptomatic thrombosis of a superficial cerebral vein.

Case Description—A cerebral angiogram performed in a 51-year-old man with subarachnoid hemorrhage revealed a 10-mm ruptured anterior communicating artery aneurysm and a thrombosed left superficial middle cerebral vein. Coil embolization of the anterior communicating aneurysm was performed. Follow-up angiography 18 months later revealed a new, asymptomatic, pial arteriovenous fistula between the previously thrombosed left superficial middle cerebral vein and a small sylvian branch of the left middle cerebral artery.

Conclusions—This case provides evidence that pial arteriovenous fistulas may develop as acquired lesions and furthermore may rarely follow cerebral vein thrombosis. Several cases of dural arteriovenous fistulas, as well as a single case of a mixed pial-dural arteriovenous fistula, occurring after dural sinus thrombosis have been reported previously. However, to our knowledge, this is the first report of an acquired pial arteriovenous fistula following a cerebral vein thrombosis. (*Stroke*. 1999;30:2487-2490.)

Key Words: cerebral arteriovenous malformations ■ etiology ■ sinus thrombosis ■ subarachnoid hemorrhage

Fistulas occurring between pial arteries and cortical veins are almost always congenital in etiology. The vast majority occur within the niduses of pial arteriovenous malformations. Isolated pial arteriovenous fistulas are rare congenital lesions, usually presenting during infancy or early childhood, that may occur sporadically or may be associated with hereditary vasculopathies such as Rendu-Osler-Weber disease.¹ Acquired fistulas between pial arteries and cortical veins are very rare. We report for the first time an acquired pial arteriovenous fistula occurring after a cortical vein thrombosis.

Case Report

A 51-year-old man with hypertension and chronic obstructive pulmonary disease presented with a 3-day history of sudden onset of severe bitemporal headache and nausea. Physical examination revealed mild meningismus without any neurological deficit (Hunt and Hess grade 1). A CT scan demonstrated subarachnoid hemorrhage with blood in the anterior interhemispheric fissure and both lateral and sylvian fissures (CT Fisher grade 1). Ventricular size was normal. Diagnostic cerebral angiography revealed a 10-mm-diameter anterior communicating artery aneurysm arising at the left A1/2 junction and a 2-mm-diameter right middle cerebral artery bifurcation aneurysm. Endovascular occlusion of the anterior communicating artery aneurysm was performed with the use

of 6 electrolytically detachable coils. After coil embolization, a small, nonocclusive embolus straddling the left middle cerebral artery bifurcation was noted. Full systemic heparinization was continued until the following day, when a diagnostic cerebral angiogram demonstrated resolution of this embolus. The patient remained neurologically intact throughout and was discharged home on postoperative day 7. The patient did not undergo a craniotomy or insertion of an intracranial pressure monitor or ventricular drain.

Follow-up angiography performed 18 months later (Figures 1 through 3) revealed mild coil compaction within the anterior communicating aneurysm and unchanged appearances of the unruptured, small, right middle cerebral artery aneurysm. There was a new left sylvian fissure arteriovenous pial fistula. Arterial supply was from a small sylvian branch of the left middle cerebral artery with venous drainage into the left superficial middle cerebral vein. Selective injection of the left external carotid artery demonstrated a coexistent low-flow dural arteriovenous fistula at the junction of the left vein of Labbé with the left transverse sinus. Arterial supply to the dural arteriovenous fistula was via a small posterior branch of the anterior division of the left middle meningeal artery. Retrospective analysis of the previous arteriogram showed nonopacification of the left superficial middle cerebral vein and left vein of Labbé during the late venous phase compatible with thrombosis. No dural arteriovenous fistula

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From the Division of Interventional Neurovascular Radiology, University of California at San Francisco Medical Center.

Correspondence to Constantine C. Phatouros, MBBS, FRACR, Division of Interventional Neurovascular Radiology, UCSF Medical Center, 505 Parnassus Ave, Room L-352, San Francisco, CA 94143-0628. E-mail con.phatouros@radiology.ucsf.edu

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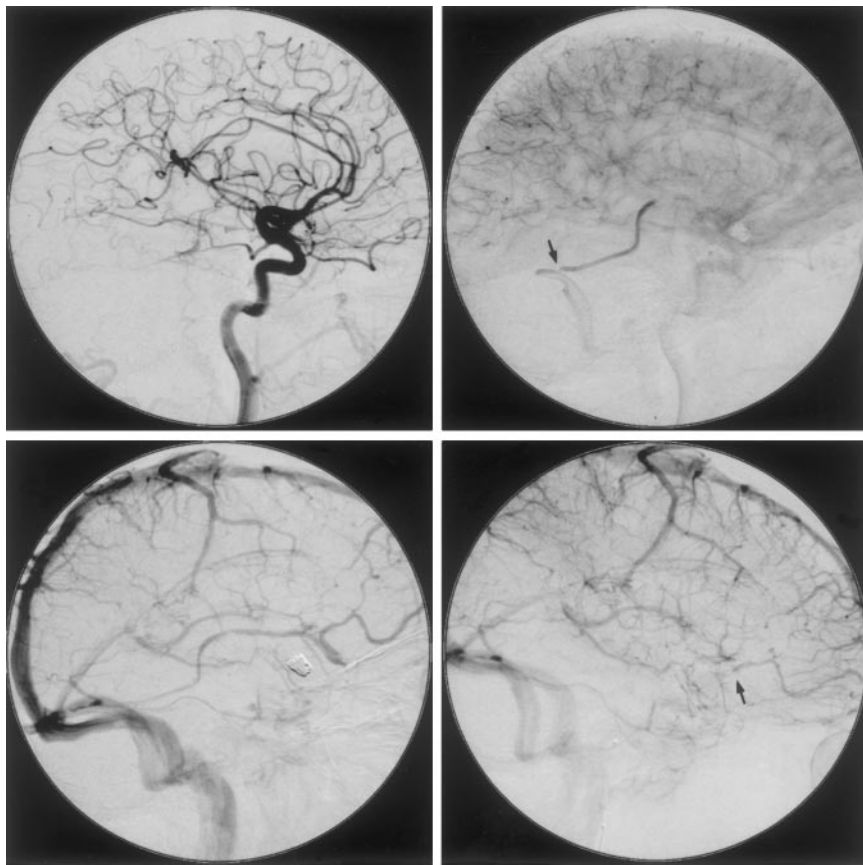


Figure 1. Top left and right and bottom left, Lateral views of 18-month follow-up left internal carotid angiogram showing early opacification of the left superficial middle cerebral vein compatible with a new pial arteriovenous fistula. Venous drainage is into the left vein of Labbé. A focal stenosis is present between the left vein of Labbé and the transverse sinus (arrow). Bottom right, Lateral view of the original left internal carotid angiogram during the late venous phase showing nonopacification of the left superficial middle cerebral vein and vein of Labbé compatible with thrombosis. Note the presence of a left frontal vein that ends abruptly at its junction with the thrombosed left superficial middle cerebral vein at the left sylvian fissure (arrow).

was identified on the previous arteriogram; however, a selective left external carotid artery injection was not performed. Therefore, the presence of a small, unrecognized, preexistent left dural arteriovenous fistula cannot be entirely excluded.

In view of the low-risk nature of both the pial and dural arteriovenous fistulas, no endovascular or surgical treatment

was performed. Attempted coil embolization of the small anterior communicating artery remnant was not technically possible.

Discussion

The unusual observation reported in this case is the development of a fistula between a previously thrombosed cerebral vein and a small cerebral artery. Newton and Cronqvist² classified intracranial arteriovenous malformations into 3 categories on the basis of their arterial supply: pial, dural, and mixed pial-dural. Pial arteriovenous malformations receive arterial supply from the cerebral and cerebellar arteries and usually drain into cerebral or cerebellar veins. Since a discrete nidus was not present in this lesion, the most accurate angiographic description is a pial arteriovenous fistula. Acquired pial arteriovenous fistulas are rare lesions in any event. However, we are not aware of a previous clinical report documenting an etiological association between cerebral vein thrombosis and pial arteriovenous fistula. Numerous clinical reports have documented the development of dural arteriovenous fistulas occurring in association with or after dural sinus thrombosis.³⁻¹⁹ Venous hypertension occurring secondary to sinus thrombosis is believed to represent the primary responsible mechanism. Experimentally induced venous hypertension without thrombosis has been shown to produce dural arteriovenous fistulas in rats.^{20,21} Venous hypertension may foster the growth of microscopic arteriovenous shunts found within the vasa vasorum of the normal pachymeninges and/or may stimulate the release of angiogenic factors.²²⁻²⁵

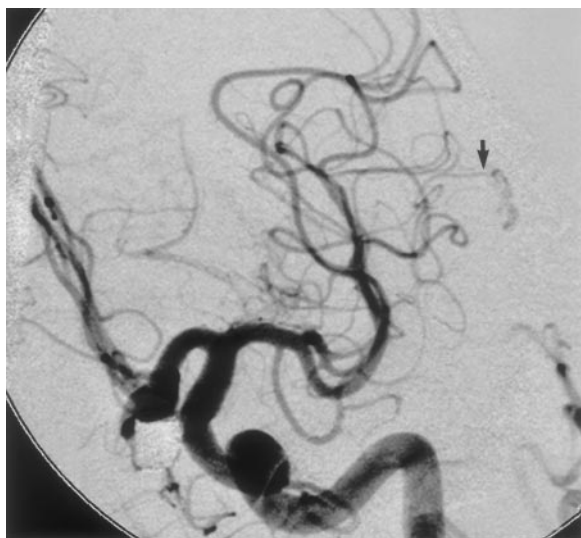


Figure 2. Anteroposterior view of 18-month follow-up left internal carotid angiogram showing a small sylvian branch of the left middle cerebral artery supplying the pial arteriovenous fistula (arrow).

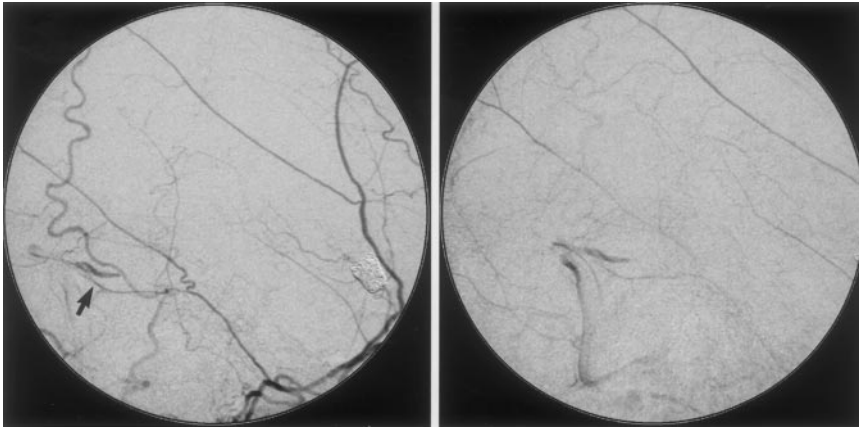


Figure 3. Lateral views of 18-month follow-up selective left external carotid angiogram showing early opacification of the left transverse sinus via a small posterior branch of the left middle meningeal artery (arrow) compatible with a dural arteriovenous fistula.

Ozawa et al²⁶ recently reported a case of a pial arteriovenous malformation with dural supply occurring after an episode of dural sinus thrombosis. In their case, dural sinus thrombosis resulted in a remote, pial arteriovenous malformation with meningeal arterial supply. The authors postulated that obliteration of the connections between the cortical veins and dural sinuses occurred as a result of retrograde thrombus propagation and that consequent elevated cortical venous pressures from impaired venous drainage subsequently led to the development of a pial arteriovenous malformation. As noted by the authors, very few reports have definitively documented the de novo development of a pial arteriovenous malformation on the basis of a previously negative cerebral angiogram—only 4 in our review.^{27–30} However, in 2 of these reports the pial arteriovenous malformation may have been preexistent but unrecognized on initial angiography because of a synchronous dural arteriovenous malformation²⁷ or an acute intraparenchymal hematoma.²⁸

Our patient suffered a small subarachnoid hemorrhage (CT Fisher grade 1), the CT blood pattern of which was compatible with a ruptured anterior communicating artery aneurysm. The initial cerebral angiogram demonstrating occlusion of the left superficial middle cerebral vein was performed 3 days after ictus. Therefore, the subarachnoid hemorrhage likely played an inconsequential role in the pathogenesis of both the cerebral vein thrombosis and the subsequent pial arteriovenous fistula. Importantly, no surgical craniotomy or insertion of a ventricular drain was performed.

A coexistent remote dural arteriovenous fistula at the junction of the left vein of Labbé and the transverse sinus was also present on the follow-up cerebral angiogram. Indeed, the original cerebral angiogram had demonstrated thrombosis of the left superficial middle cerebral vein and its draining tributary, the left vein of Labbé. Therefore, it is possible that this dural arteriovenous fistula also developed as a consequence of the cerebral vein thrombosis. A left common carotid injection performed on the original angiogram, which satisfactorily opacified the left middle meningeal artery, did not demonstrate evidence of a dural arteriovenous fistula. However, since a selective left external carotid injection was not performed at that time, the possibility that this dural arteriovenous fistula was preexistent, although unlikely, cannot be completely excluded.

References

- Garcia-Monaco R, Taylor W, Rodesch G, Alvarez H, Burrows P, Coubes P, Lasjaunias P. Pial arteriovenous fistula in children as presenting manifestation of Rendu-Osler-Weber disease. *Neuroradiology*. 1995;37:60–64.
- Newton TH, Cronqvist S. Involvement of dural arteries in intracranial arteriovenous malformations. *Radiology*. 1969;93:1071–1078.
- Witt O, Pereira PL, Tillmann W. Severe cerebral venous sinus thrombosis and dural arteriovenous fistula in an infant with protein S deficiency. *Childs Nerv Syst*. 1999;15:128–130.
- Chaudhary MY, Sachdev VP, Cho SH, Weitzner I, Jr, Puljic S, Huang YP. Dural arteriovenous malformation of the major venous sinuses: an acquired lesion. *AJNR Am J Neuroradiol*. 1982;3:13–19.
- Cataltepe O, Berker M, Gurcay O, Erbeni A. An unusual dural arteriovenous fistula in an infant. *Neuroradiology*. 1993;35:394–397.
- Iwakawa J, Umehara F, Nishizawa T, Eiraku N, Osame M. Dural arteriovenous fistulas of the posterior fossa associated with lateral sinus thrombosis presenting as progressive visual impairment [in Japanese]. *Rinsho Shinkeigaku*. 1996;36:1095–1099.
- Iwata A, Nakamura K, Nukina N, Kanazawa I, Iwata M. A case of dural arteriovenous fistula accompanied by sinus occlusion: a serial study with CT scan [in Japanese]. *Rinsho Shinkeigaku*. 1998;38:133–137.
- Koshimae N, Iwanaga H, Imanishi M, Okuchi K, Tokunaga H, Aoki H, Boku E, Tsujimoto M. Dural AVF of the posterior fossa associated with sinus occlusion presenting as intracerebral hemorrhage: case report [in Japanese]. *No Shinkei Geka*. 1995;23:163–167.
- Kutluk K, Schumacher M, Mironov A. The role of sinus thrombosis in occipital dural arteriovenous malformations: development and spontaneous closure. *Neurochirurgia (Stuttgart)*. 1991;34:144–147.
- Mayberg MR, Zimmerman C. Vein of Galen aneurysm associated with dural AVM and straight sinus thrombosis: case report. *J Neurosurg*. 1988;68:288–291.
- Mironov A. Classification of spontaneous dural arteriovenous fistulas with regard to their pathogenesis. *Acta Radiol*. 1995;36:582–592.
- Mironov A. Pathogenetical consideration of spontaneous dural arteriovenous fistulas (DAVFs). *Acta Neurochir (Wien)*. 1994;131:45–58.
- Pierot L, Chiras J, Duyckaerts C, Jason M, Martin N. Intracranial dural arteriovenous fistulas and sinus thrombosis: report of five cases. *J Neuroradiol*. 1993;20:9–18.
- Preter M, Tzourio C, Ameri A, Boussier MG. Long-term prognosis in cerebral venous thrombosis: follow-up of 77 patients. *Stroke*. 1996;27:243–246.
- Smith TP, Higashida RT, Barnwell SL, Halbach VV, Dowd CF, Fraser KW, Teitelbaum GP, Hieshima GB. Treatment of dural sinus thrombosis by urokinase infusion. *AJNR Am J Neuroradiol*. 1994;15:801–807.
- Sugiura Y, Miyamoto T, Takehara S, Sumiya K, Nozaki T. Multiple dural arteriovenous fistulas following extensive sinus thrombosis: a case report [in Japanese]. *No Shinkei Geka*. 1996;24:379–383.
- Malek AM, Higashida RT, Balousek PA, Phatouros CC, Smith WS, Dowd CF, Halbach VV. Endovascular recanalization with balloon angioplasty and stenting of an occluded occipital sinus for treatment of intracranial venous hypertension: technical case report. *Neurosurgery*. 1999;44:896–901.
- Tajima Y, Minami N, Okumura H, Miyasaka K, Moriwaka F, Tashiro K. An unusual case of superior sagittal sinus thrombosis accompanied with dural AV fistula [in Japanese]. *No To Shinkei*. 1991;43:981–985.

19. Touho H, Ohnishi H, Komatsu T, Furuoka N, Karasawa J. Dural arteriovenous fistula caused by sinus thrombosis: case report. *Neurol Med Chir (Tokyo)*. 1994;34:543–546.
20. Herman JM, Spetzler RF, Bederson JB, Kurbat JM, Zabramski JM. Genesis of a dural arteriovenous malformation in a rat model. *J Neurosurg*. 1995;83:539–545.
21. Terada T, Higashida RT, Halbach VV, Dowd CF, Tsuura M, Komai N, Wilson CB, Hieshima GB. Development of acquired arteriovenous fistulas in rats due to venous hypertension. *J Neurosurg*. 1994;80:884–889.
22. Kerber CW, Newton TH. The macro and microvasculature of the dura mater. *Neuroradiology*. 1973;6:175–179.
23. Terada T, Tsuura M, Komai N, Higashida RT, Halbach VV, Dowd CF, Wilson CB, Hieshima GB. The role of angiogenic factor bFGF in the development of dural AVFs. *Acta Neurochir (Wien)*. 1996;138:877–883.
24. Lawton MT, Jacobowitz R, Spetzler RF. Redefined role of angiogenesis in the pathogenesis of dural arteriovenous malformations. *J Neurosurg*. 1997;87:267–274.
25. Hamada Y, Goto K, Inoue T, Iwaki T, Matsuno H, Suzuki S, Matsushima T, Fukui M, Miyake E. Histopathological aspects of dural arteriovenous fistulas in the transverse-sigmoid sinus region in nine patients. *Neurosurgery*. 1997;40:452–456; comment 456–458.
26. Ozawa T, Miyasaka Y, Tanaka R, Kurata A, Fujii K. Dural-pial arteriovenous malformation after sinus thrombosis. *Stroke*. 1998;29:1721–1724.
27. Albright AL, Latchaw RE, Price RA. Posterior dural arteriovenous malformations in infancy. *Neurosurgery*. 1983;13:129–135.
28. Miyasaka Y, Kurata A, Saegusa H, Yuzawa I, Utsuki S, Ohwada T. Dural-pial arteriovenous malformation with unusual venous drainage. *Neurol Med Chir (Tokyo)*. 1996;36:91–95.
29. Sasaki T, Hoya K, Kinone K, Kirino T. Postsurgical development of dural arteriovenous malformations after transpetrosal and transtentorial operations: case report. *Neurosurgery*. 1995;37:820–824; comment 824–825.
30. Schmit BP, Burrows PE, Kuban K, Goumnerova L, Scott RM. Acquired cerebral arteriovenous malformation in a child with moyamoya disease: case report. *J Neurosurg*. 1996;84:677–680.