Resection of a Giant Intracranial Dural Arteriovenous Fistula with the Use of Low-flow Deep Hypothermic Cardiopulmonary Bypass after Partial Embolization: Technical Case Report

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- OBJECTIVE AND IMPORTANCE: To describe the surgical resection of a giant intracerebral arteriovenous fistula with involvement of dura mater and surrounding bone. Intraoperative bleeding was controlled by hypothermic circulatory arrest.
- CLINICAL PRESENTATION: This 46-year-old woman complained of persistent headache for 1 year; her diagnostic workup revealed the presence of an arteriovenous fistula in the dura mater of the left temporal region fed by the meningeal artery of the external and internal carotid arteries, with normal run-off into Labbé's and Trolard's veins. Magnetic resonance imaging depicted a Chiari I malformation that was most likely a result of insufficient cerebral venous drainage.
- INTERVENTION: In preparation for surgery, staged embolization of feeders from the left meningeal artery and the left occipital artery was performed under controlled hypotension. This procedure failed to achieve a significant reduction in flow because of the immediate recruitment of internal branches of the internal carotid artery and dural branches of the right external carotid artery. Surgical treatment was undertaken without further embolization. Because of involvement of surrounding bone and the high risk of uncontrollable bleeding, the procedure was carried out with the patient under deep hypothermic cardiopulmonary bypass. Forty-five minutes of low flow (1.5 L/min) at 18°C allowed total resection of the involved dura mater and surrounding bone. Postoperative recovery was marked by left brain edema that disappeared within 10 days. Findings on follow-up angiography were normal, and the patient was discharged with no neurological deficit.

CONCLUSION: Low-flow deep hypothermic cardiopulmonary bypass can be used to control intraoperative bleeding for surgical excision of a giant intracerebral dural arteriovenous fistula.

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Key words: Cardiopulmonary bypass, Chiari malformation, Circulatory arrest, Dural arteriovenous fistula, Giant aneurysm, Hypothermia

A variety of modalities have been pro-posed for the management of a dural arteriovenous fistula (DAVF). In a recent review of the English literature, Lucas et al. (6) described indications and outcomes with the use of different modalities regarding function of localization. For DAVFs with extensive feeders, combined embolization and surgery is often necessary to avoid a recurrence because of further recruitment. Surgical coagulation (or excision) of the shunt and venous drainage achieve a permanent cure. This report describes a patient in whom a giant DAVF involving the cranial base in the left temporal fossa was treated by embolization and surgical removal. Because of the unusual location and vascularization of the lesion, cardiopulmonary bypass was used to help with surgical removal.

CASE REPORT

A 46-year-old right-handed woman was referred to our institution for treatment of an osteodural vascular malformation located in the left temporal fossa. The patient had complained of left temporal headaches for 1 year. Physical examination revealed moderate left proptosis. Visual field and acuity were normal. An ophthalmoscopic examination disclosed no papilledema. The patient's cardiac examination was normal. Cardiac output was not measured before endovascular treatment. Magnetic resonance imaging (MRI) disclosed tortuous dilation of the superficial veins, a highly enhanced area in the dura of the left temporal fossa, and leftto-right parenchymal displacement (Fig. 1A). A Chiari I malformation was also visualized (Fig. 1B). Cerebral angiography depicted a left temporobasal arteriovenous fistula ascending the vault and corresponding to the enhanced area disclosed by MRI. The fistula was fed mainly by the left middle meningeal artery and was also supplied by meningeal branches of the right occipital artery, the right and left anterior and posterior meningeal arteries, and the left internal carotid artery. No feeding from the parenchymal branches of the

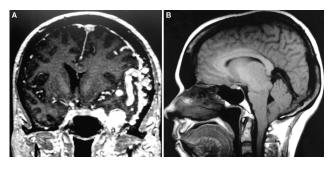
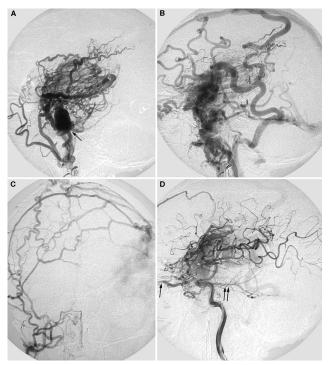


FIGURE 1. Preoperative T1-weighted MRI scans. *A*, coronal scan with diethylenetriamine penta-acetic acid gadolinium showing tortuous dilation of the superficial veins, a highly enhanced dural area of the left temporal

fossa, left-to-right parenchymal displacement, and the presence of an aneurysm close to the cavernous sinus at the base of the temporal fossa. *B*, sagittal scan showing a Chiari I malformation most likely acquired because of poor venous drainage.

internal carotid artery or the vertebral artery was observed. Angiography also visualized a large aneurysm measuring 1 cm in diameter and 2 cm in length at the level of the foramen spinosum. Venous drainage was via meningeal veins and a huge reflux into cortical veins draining first into Labbé's and Trolard's veins and then into the dural sinuses in an antegrade direction (*Fig. 2*). Contrast injection into the internal carotid arteries revealed delayed and poor venous drainage of the brain. Computed tomographic scanning disclosed bone erosion of the inner table of the left calvaria close to the malformation. The outer table was intact. A bone defect in relation to the aneurysm of the foramen spinosum was visualized (*Fig. 3*). In view of the cortical drainage of this fistula and the high-grade cortical venous steal resulting in poor venous drainage of the brain, staged embolization in preparation for surgical resection was planned.



lard's veins and then into the dural sinuses in an antegrade direction. *C*, frontal view of the right external carotid artery showing a feeder crossing the midline from the right meningeal artery. *D*, lateral view of the left internal carotid artery showing feeders coming from meningeal branches of the internal carotid artery. The *single arrow* indicates the ethmoidal artery, and the *double arrow* indicates Bernarsconi's artery.

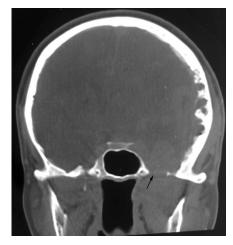


FIGURE 3. Computed tomographic scan in bone window showing bone erosion of the inner table of the left calvaria close to the malformation. The "defect" corresponds to enlargement of the foramen spinosum at the level of aneurysmal dilation of the left middle meningeal artery (*arrow*).

In the first procedure, distal embolization of the middle meningeal artery was performed with cyanoacrylate glue (Histoacryl; B. Braun Melsungen AB, Melsungen, Germany) with the patient under general anesthesia and controlled hypotension. The left superficial temporal artery and left occipital artery dural feeders were also occluded with glue. In the same procedure, the large aneurysm of the middle meningeal artery at the level of the foramen spinosum was occluded with the use of detachable mechanical platinum coils. Postembolization angiograms showed very little reduction in flow through the fistula, which continued to be fed by the anterior and posterior meningeal artery, the meningeal branches of the internal carotid artery, and branches of the contralateral middle meningeal artery. The patient's headaches persisted, and no change was observed in parenchymal displacement or venous drainage. Because of persistent shunting, prompt regrowth of small meningeal shunt feeders seemed likely. In addition, increased flow in the internal carotid artery after the initial procedure was predictive that further embolization of the external carotid artery would fail (Fig. 4).

We decided to undertake surgical treatment without further embolization.

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FIGURE 2. Preoperative angiograms.

A, lateral view in the

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left external carotid

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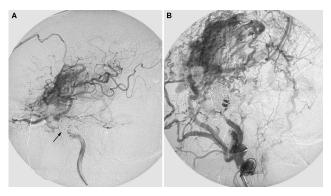


FIGURE 4. Postembolization angiograms with lateral views of the left external carotid artery (*A*) and the left internal carotid artery (*B*). High-flow shunting persisted despite coil embolization of the aneurysmal dilation of the middle meningeal artery (*arrow* in *A*)

and cyanoacrylate glue embolization of other meningeal branches of the left middle meningeal artery. Increased flow in the dural branches of the left internal carotid artery was also noted.

Because of the high risk of uncontrolled intraoperative bleeding, operative planning included low-flow deep hypothermic cardiopulmonary bypass in collaboration with the cardiothoracic surgery team. Preoperative pulmonary artery catheterization revealed mildly elevated cardiac output (6 L/min). No aortic insufficiency, atrial or ventricular hypertrophy, or other valvulopathy was observed. Based on these findings, a femorofemoral bypass was performed. A detailed description of the technique and team coordination is available in an article describing a series of eight patients operated on for complex aneurysm in our center (7). Flow rate was maintained at 1.5 L/min, and body temperature was kept at 16.5°C. Blood loss was high but controlled. The use of a blood salvage system compatible with neurosurgery (Cell Saver 5 Autologous Blood Recovery System; Haemonetics Corp., Braintree, MA) permitted recovery and transfusion of 3.6 L of autologous blood (2). The 45-minute duration of bypass was sufficient to perform craniotomy and excision of the dura around the malformation. There was no adherence between the brain, the fistula, and venous drainage. The major draining vein was coagulated and divided proximal to its junction with Trolard's and Labbé's veins. Forty-five seconds of circulatory arrest was needed to allow inspection at the base of the aneurysm and additional coagulation of arterial feeders at this level. The large dural defect was replaced with Durapath (Ethicon, Issy-les-Moulineaux, France). Bone plasty was not performed during the same procedure.

Immediate postoperative computed tomographic scanning disclosed left temporal edema. A postoperative neurological examination revealed mild aphasia that resolved within 10 days. The patient was released from the intensive care unit 9 days after the operation. Angiography performed 16 days after the procedure disclosed few tortuous meningeal arteries without shunting. Venous drainage of the brain was satisfactory (*Fig. 5*). Postoperative MRI on Day 20 revealed a decrease in parenchymal shifting and improvement in the Chiari I malformation (Fig. 6). Cranioplasty with copolymer of methylmethacrylate was performed on postoperative Day 35. The patient was discharged from the hospital on postoperative Day 45 with no neurological deficit and with normal visual field. Cardiac output measured immediately before discharge was 5 L/min. A histological examination of the surgical specimen disclosed large vascular channels consistent with a DAVF. An arteriovenous malformation with a nidus was not observed, although it should be noted that the specimen was extensively coagulated during the surgical procedure.

DISCUSSION

The current classification of DAVF groups lesions according to type of drainage (3). In Type I, the DAVF drains directly into a sinus or a meningeal vein in a normal flow direction. In Type II, the DAVF presents sinus drainage accompanied by reflux into cortical veins or into another sinus. In Type III, the DAVF is characterized by direct drainage into a cortical vein with retrograde

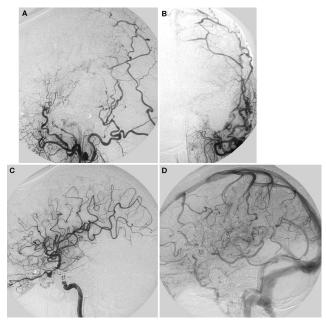


FIGURE 5. Postoperative angiograms. A, lateral view of the left external carotid artery. B, frontal view of left external carotid artery. These views confirm disappearance of the shunt. C, view of the left internal carotid artery after contrast injection in the arterial phase. D, view of the left internal carotid artery after contrast injection in the venous phase. The disappearance of the shunt is confirmed, and normal venous drainage not visible on preop-

erative cortical and venous sinus views is shown. Labbé's and Trolard's veins are not visible on these views, most likely because of postoperative thrombosis resulting from a dramatic decrease in flow.

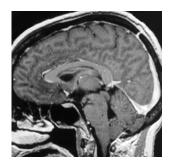


FIGURE 6. Postoperative sagittal T1-weighted MRI scan showing improvement of the Chiari I malformation. The tonsils are less tapered and higher than on preoperative MRI scans.

flow. In Type IV, the DAVF involves drainage into a dural venous lake that may be expansive.

Because all arterial feeders were issued from meningeal vessels, the lesion described in this article can be assumed to be a dural-seated shunt. However, it presented several features that prevented standard classification. In this patient, drainage into cortical veins was direct, but the flow was antegrade. Another unusual aspect of the case was the association of an arterial feeder with aneurysmal dilation. Two explanations can be proposed for these unusual features. The first is that the fistula was at an early stage of development in a location close to the dural penetration of a sylvian vein in the sphenoparietal sinus. These veins are frequently identified close to the tip of the lesser sphenoid wing during dissection of the sylvian fissure in a standard pterional approach. The absence of encasement of the cavernous sinus in the venous drainage could be explained by thrombosis of the sphenoparietal sinus. Sinus thrombosis is a possible explanation of the formation of a DAVF (4). According to this hypothesis, our case could be associated with a Type III DAVF. The second possible explanation for the unusual features observed in our case could be that the lesion described here was not a DAVF but rather a dural arteriovenous malformation; however, this hypothesis is not supported by the histological findings in which no malformed nidus was seen. The second hypothesis is consistent with displacement of the dura mater and extensive involvement of the cranium.

Therapeutic management of dural arteriovenous malformations is challenging. A range of modalities can be proposed, depending on the location and pattern of vascularization (6). Successful surgical treatment of two large parenchymal arteriovenous fistulae was recently described with low-flow cardiopulmonary bypass (1, 8). Similar success has been reported with the use of cardiopulmonary bypass with deep hypothermia for complex or giant aneurysms (especially of the posterior circulation), hemangioblastomas, glomus jugular tumors, and ischemic disease (5, 7, 9, 11).

In our patient, staged transarterial embolization of the DAVF was attempted as a means of reducing blood flow. This approach was quickly abandoned because the endovascular procedure failed to achieve a significant reduction in flow and was followed by an immediate increase of flow in the meningeal branches of the internal carotid artery. This suggested that further embolization not only would fail to control surgical bleeding but also would increase the complexity of the fistula by fostering recruitment of new feeding arteries. Another reason for abandoning staged embolization was the ischemic risk associated with occlusion of meningeal feeders from the internal carotid artery. In any case, embolization allowed occlusion of the large aneurysm in the middle meningeal artery. Because this lesion presented in an irregular dome shape and was not directly accessible, clipping would have prolonged surgical time and increased blood loss dramatically.

Cardiopulmonary bypass with deep hypothermic protection seemed to be the best solution for controlling operative blood loss. In a large review including 62 circulatory arrest procedures, Lawton et al. (5) underlined the importance of the duration of circulatory arrest but found no statistical correlation between this parameter and outcome. In many situations, complete circulatory arrest is unnecessary. In this regard, a recent study by van der Linden (10) suggested that cerebral metabolism and energy state were better after low-flow than after no-flow bypass. Moreover, reduced but persistent flow allows better identification of small vessels and more thorough coagulation. This reduces intraoperative bleeding and facilitates hemostasis at the end of the procedure.

Transient temporal lobe edema and aphasia in the present case were related more to venous drainage disorders than to surgical injury. Prompt resolution of these complications can be attributed to elimination of venous steal and the reestablishment of proper venous drainage of the left temporal lobe. The dramatic decrease in arterial flow observed in cortical veins (Trolard's and Labbé's veins) after surgery can explain the delayed thrombosis of these veins. Normal drainage via these thrombosed veins was not significant.

CONCLUSION

This case is the first one to describe the surgical management of a giant osteodural arteriovenous fistula. Preoperative embolization did not achieve sufficient occlusion of some arterial feeders to ensure control of intraoperative bleeding. Low-flow cardiopulmonary bypass was necessary to reduce flow in the fistula and to allow surgical resection. This experience emphasizes the need for a multidisciplinary approach for these complex fistulae that includes cooperation with cardiothoracic surgeons.

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REFERENCES

- Bendok BR, Getch CC, Frederiksen J, Batjer HH: Resection of a large arteriovenous fistula of the brain using low-flow deep hypothermic cardiopulmonary bypass: Technical case report. Neurosurgery 44:888–891, 1999.
- Cataldi S, Bruder N, Dufour H, Lefevre P, Grisoli F, François G: Intraoperative autologous blood transfusion in intracranial surgery. Neurosurgery 40:765–772, 1997.
- Djindjian R, Merland JJ: Super-selective Arteriography of the External Carotid Artery. New York, Springer-Verlag, 1978.
- Houser OW, Campbell JK, Campbell RJ, Sundt TM Jr: Arteriovenous malformation affecting the transverse dural venous sinus: An acquired lesion. Mayo Clin Proc 54:651–661, 1979.

- Lawton MT, Raudzens PA, Zabramski JM, Spetzler RF: Hypothermic circulatory arrest in neurovascular surgery: Evolving indications and predictors of patients outcome. Neurosurgery 43:10–21, 1998.
- Lucas CP, Zabramski JM, Spetzler RF, Jacobowitz R: Treatment for intracranial dural arteriovenous malformations: A meta-analysis from the English language literature. Neurosurgery 40:1119–1132, 1997.
- Mesana T, Collart F, Caus T, Pomane C, Graziani N, Bruder N, Dufour H, Grisoli F, Monties JR: Centrifugal pumps and heparincoated circuits in surgical treatment of giant cerebral aneurysms. Artif Organs 24:431–436, 2000.
- Meyer FB, Grady RE, Abel MD, Nichols DA, Caminha SS, Robb RA, Bates LM: Resection of a large temporooccipital parenchymal arteriovenous fistula by using deep hypothermic circulatory bypass: Case report. J Neurosurg 87: 934–939, 1997.
- Solomon RA, Smith CR, Raps EC, Young WL, Stone JG, Fink ME: Deep hypothermic circulatory arrest for the management of complex anteriorand posterior circulation aneurysm. Neurosurgery 29:732–738, 1991.
- van der Linden J: Cerebral hemodynamics after low-flow versus no-flow procedures. Ann Thorac Surg 59:1321–1325, 1995.
- Williams M, Rainer G, Fieger H, Murray I, Sanchez M: Cardiopulmonary bypass, profound hypothermia and circulatory arrest for neurosurgery. Ann Thorac Surg 52:1069–1075, 1991.

COMMENTS

In this report of a successful treatment of a massive dural arteriovenous fistula (DAVF), the authors wisely predicted that, even after embolization, bleeding at the time of craniotomy could have been catastrophic had they not used cardiopulmonary bypass. It is well known that several deaths occurred in the early days of surgical resection of these massive fistulae.

Apparently, this fistula drained mostly into a single venous connection with the leptomeningeal venous system. Knowing this, I am sure that the authors entertained the notion of treating the fistula with progressive embolization and, when the flow was low enough, doing nothing more than occlude the vein connected to the leptomeningeal venous system. We know that this is curative in patients in whom the only venous drainage is in fact a single venous connection to the leptomeningeal system. This situation is particularly common with fistulae in the area of the superior petrosal sinus and the tentorial edge, and I have treated close to a dozen of these fistulae by simple interruption of the single venous drainage. It is clear that the case presented here had multiple other venous channels into the dura and also into scalp veins; it is therefore not likely that this fistula would have been cured by interruption of the leptomeningeal venous connection. However, it is quite likely that the neurological symptoms, including brain swelling, would have resolved with this simple maneuver. The problem, of course, would be how to get to the vein without performing a craniotomy and incurring the danger of massive bleeding. Could this venous connection have been occluded endovascularly by retrograde venous catheterization? The authors decided on a more radical course and attempted a cure, which they achieved with only temporary morbidity. I believe that this was a good decision, and I probably would have done the same in this particular case.

I agree with the authors that the most likely explanation for the temporary neurological deterioration was retrograde venous thrombosis. We all have found this occasionally in cases of both dural fistula and brain arteriovenous malformations when there is an abrupt interruption by surgery or sometimes even by embolization of the arterial inflow into the veins. This patient was fortunate to have recovered promptly from this problem which, at times, can lead to major permanent deficits. The authors used a thoughtful and successful approach in treating this patient.

> **Roberto C. Heros** *Miami, Florida*

This case report describes a patient in whom the authors used low-flow hypothermic circulatory bypass to successfully treat a giant intracranial DAVF. We too have found circulatory bypass to be beneficial in treating complex dural and parenchymal vascular malformations (1). This case report emphasizes that complex cranial base DAVFs typically require multimodal approaches for successful treatment, including detailed preoperative angiography, preoperative embolization, complex intraoperative surgical techniques such as low-flow hypothermia, and quite possibly postoperative stereotactic radiation for residual unobliterated fistulae or malformations. Accordingly, these complex vascular lesions need to be approached from a multispecialty perspective commencing at the initial evaluation of the patient.

> Fredric B. Meyer Rochester, Minnesota

The authors describe the use of lowflow deep hypothermic cardiopulmonary bypass as an adjunct to reduce blood loss in the resection of a complex DAVF. Preoperative embolization is usually sufficient for the management of these lesions. However, the authors have brought to our attention a case in which cardiopulmonary bypass was of benefit. They appropriately highlight that blood loss can be considerable during resection of these lesions, even with the use of these adjunct techniques. The authors also emphasize the potential for neurological deficits (transient in this case) as a result of venous thrombosis. It is interesting that the Chiari malformation improved in this patient, which suggests that venous outflow obstruction may be a common etiology for arteriovenous fistulae and some Chiari malformations.

> Sam Javedan Michael Horgan Robert F. Spetzler Phoenix, Arizona

Meyer FB, Grady RE, Abel MD, Nichols DA, Caminha SS, Robb RA, Bates LM: Resection of a large temporooccipital parenchymal arteriovenous fistula by using deep hypothermic circulatory bypass: Case report. J Neurosurg 87:934– 939, 1997.