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Normal Perfusion Pressure Breakthrough Occurring During Treatment of Carotid and Vertebral Fistulas

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Of the 185 carotid and vertebral fistulas treated by the authors over the past 10 years, five developed neurologic deficits after abrupt closure of their fistulas. The earliest case, treated initially by proximal surgical carotid occlusion, presented 32 years later with cerebral steal symptoms from the large, long-standing carotid cavernous fistula. Upon completion of a surgical trapping procedure, there was immediate massive cerebral edema, brain herniation, and death. In the remaining four patients (three vertebral fistulas and one with carotid cavernous fistula), all treated by transvascular embolization techniques, neurologic deficits occurred coincidentally with the abrupt closure of the fistula and resolved with reestablishment of fistula flow. This indicated that the cerebral vasculature is unable to tolerate the reestablishment of normal cerebral perfusion after abrupt closure of the fistula. All four patients were treated with staged or slow occlusion of their fistulas, which resulted in complete fistula closure without permanent neurologic sequelae. All five patients who developed symptoms consistent with normal perfusion pressure breakthrough had large, long-standing fistulas, ranging in duration from 9 to 32 years. Two of the five patients developed slowly progressive neurologic deficits consistent with cerebral steal prior to treatment. This sign was not observed in the 180 patients who did not develop symptoms during treatment.

We conclude that patients with carotid or vertebral fistulas of long duration, particularly those with cerebral steal symptoms, are at risk to develop neurologic deficits related to perfusion breakthrough if their fistulas are abruptly closed. Staged or gradual closure may prevent this potentially devastating complication.

Normal perfusion pressure breakthrough theory was described in an attempt to explain the hemorrhages that occurred in normal surrounding brain after the resection of large, high-flow arteriovenous malformations (AVMs) [1, 2]. Rapid alterations of blood flow may result in malignant cerebral edema or hemorrhage in adjacent brain that has lost normal autoregulation because of the chronic steal into the adjacent AVM. This sequence of events was documented experimentally in cats with surgically created carotid-jugular fistulas [1]. Subsequently, ischemic and hemorrhagic complications secondary to embolization and surgical resection of AVMs and after carotid endarterectomy have been explained by this theory [3-9]. There have been no reports of this syndrome occurring after the closure of carotid or vertebral fistulas even though the experimental support for this theory was a carotid-jugular fistula created in cats [1]. Our early experience with a surgically treated case of chronic, long-standing carotid cavernous fistula complicated by massive cerebral edema and death made us acutely aware of this potentially lethal complication. Since that early experience, we have identified four more patients who developed symptoms of normal perfusion pressure breakthrough during transvascular occlusion of their fistulas. We describe the findings in these patients and the alterations in their treatments required to prevent permanent neurologic sequelae.

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Materials and Methods

The clinical and radiologic records of 165 carotid and 20 vertebral fistulas treated by the authors were reviewed to determine the number of normal perfusion pressure breakthrough syndromes occurring during embolization.

We defined this syndrome as the development of potentially reversible neurologic deficits that occurred coincidentally with or immediately after closure of the fistula. Only reversible neurologic deficits directly related to occlusion of the fistula were included to exclude other possible sources.

The age range among the 20 patients treated with vertebral fistulas was 20 to 61 years, with a mean of 33 years. The duration from onset of the fistula to treatment ranged from 6 hr to 28 years. The fistulas were closed via transvascular embolization with detachable silicone or latex balloons or with silicone spheres with preservation of the parent artery if possible. The procedures were performed under local anesthesia with IV sedation to allow continuous neurologic monitoring. Fifty-five percent of the fistulas were caused by penetrating wounds, 10% followed blunt trauma, and the rest were spontaneous in origin. Three of the 20 patients developed neurologic symptoms during embolization consistent with normal perfusion pressure breakthrough. In all three patients, alterations in treatment were required to achieve complete closure of the fistula and to preserve neurologic function.

The age range of the 165 patients treated for carotid cavernous fistulas was 6 to 76 years, with a mean of 44 years. One hundred and seven patients (65%) were treated by transarterial balloon embolization, 21 patients (13%) were treated by a transvenous route, 31 patients (19%) were treated by compression therapy, and six patients (3%) were treated by direct surgical exposure of the cavernous sinus and embolization. Only one patient had surgical clipping of the supraclinoid carotid artery 32 years after proximal carotid ligation to attempt cure. This patient and one other were the only patients who developed symptoms consistent with normal perfusion pressure breakthrough during treatment of their fistulas.

A comparison of the mean age and sex of the patients, and the size, location, and duration of their fistulas was made between the five patients who developed normal perfusion pressure breakthrough during treatment and the 180 patients who did not.

Results

Frequency

Normal perfusion pressure breakthrough occurred in two patients with carotid cavernous fistulas out of 165 treated (1.2%). This syndrome occurred in three of 20 patients with vertebral fistulas (15%). The overall occurrence of both vertebral and carotid fistulas was five of 185 (2.7%). Since our awareness of this syndrome, we have treated several large fistulas with staged occlusion to prevent normal perfusion pressure breakthrough from occurring. Because none of these patients developed symptoms, their risk of developing this syndrome is purely speculative.

Age

There was no significant difference between the ages of patients who developed normal perfusion pressure breakthrough and those who did not.

Location

There was no difference between the location of the fistulas in patients who developed normal perfusion pressure breakthrough and those who did not.

Duration and Size of Fistula

There was a strong association between the duration of the fistula and the chance of developing normal perfusion pressure breakthrough during treatment. Two of the three patients with vertebral fistulas had symptoms since birth. The third patient had symptoms for 9 years. The patient with the carotid cavernous fistula treated by balloon occlusion had symptoms for 15 years before definitive treatment. The patient with a carotid cavernous fistula treated by surgery had the fistula for 32 years. In both the vertebral and carotid fistula cases, the five patients who developed symptoms of normal perfusion pressure breakthrough had fistulas of the longest duration.

Exact quantification of fistula size was angiographically difficult; however, all five patients were judged to have the largest fistulas. In all five patients there was marked enlargement of the caliber of the feeding arteries.

Symptoms

Prior to treatment two patients developed slowly progressive neurologic deficits attributed to steal by the large fistula. The first patient, with a large, long-standing carotid cavernous fistula had internal carotid ligation 32 years earlier. There was poor filling of the normal intracranial vessels. She had ischemic symptoms in the ipsilateral hemisphere with the development of progressive contralateral body weakness over 4 months. The second patient had a bruit and neck pain since birth. At age 23 she developed slowly progressive weakness and clumsiness in her left leg and arm.

None of the remaining 183 patients had symptoms related to cerebral steal.

Representative Case Reports

Case 1

A 55-year-old woman developed a traumatic carotid cavernous fistula at age 23 and was treated shortly thereafter with proximal internal carotid artery ligation with transient improvement in symptoms. She continued to have signs and symptoms of a persistent fistula for the next 32 years. Four months before final treatment she presented with slowly progressive contralateral body weakness secondary to cerebral steal from the fistula. An angiogram (Fig. 1) revealed a large carotid cavernous fistula supplied by retrograde flow in a massively dilated ophthalmic artery. There was substantial supply to the fistula through enlarged anterior and posterior communicating arteries. Because of tortuosity of these arteries a transvascular treatment was not attempted. In an attempt to complete a trapping procedure of the carotid artery, surgical exposure was performed and a surgical clip was placed on the supraclinoid carotid artery. There was immediate massive brain swelling out of the operative site

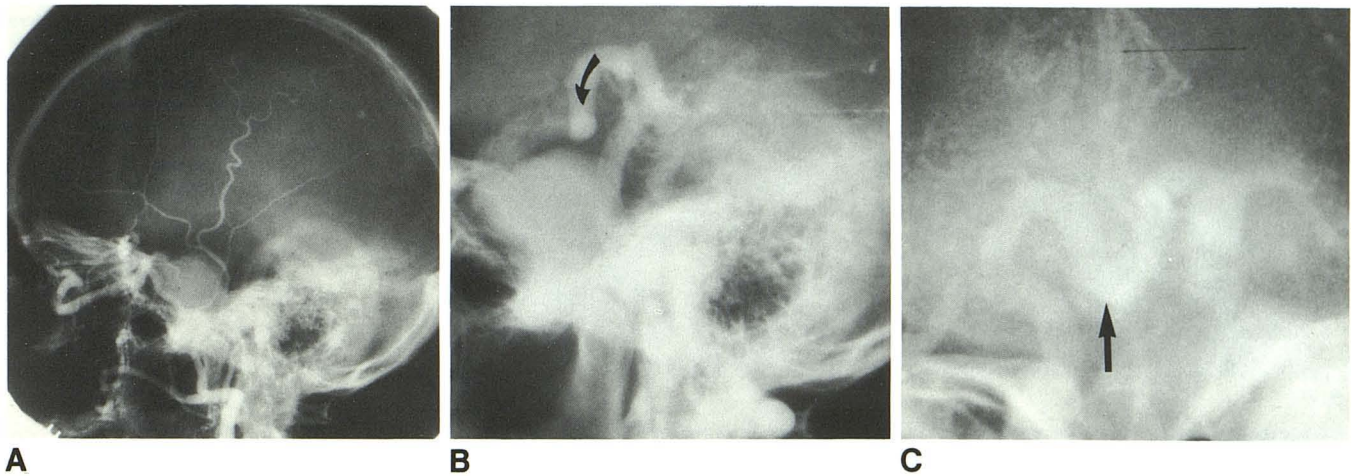


Fig. 1.—A, Left common carotid artery injection, lateral view, showing filling of a carotid-cavernous fistula via the ophthalmic artery supplied by external carotid collaterals.

B, Vertebral artery injection, lateral view. There is filling of the fistula through an enlarged posterior communicating artery (arrow).

C, Right internal carotid artery injection, anteroposterior view. There is filling of the fistula from an enlarged anterior communicating artery (arrow).

that prevented the clip's removal. Brain herniation ensued and the patient died. Autopsy revealed diffuse cerebral edema of the entire hemisphere. This was attributed to normal perfusion breakthrough in the chronically ischemic hemisphere.

Case 2

A 23-year-old woman presented with a history of lifelong bruit centered in the back of her head and intermittent severe headaches diagnosed as migraines. The bruit and headaches were exacerbated by stress and exercise, and the headache was often preceded by a prodrome of vertical lines in both visual fields described as a "moving picket fence." Treatment with ergot alkaloids worsened the headaches. Two years before treatment she began developing weakness and clumsiness in the left arm and leg. Several months before referral for treatment, an angiogram demonstrated a left vertebral fistula located at C2 (Fig. 2). Injection of both the ipsilateral carotid artery and the contralateral vertebral artery demonstrated steal down the basilar artery and the involved vertebral artery, respectively. A large, tortuous, left deep cervical artery arising from the costocervical trunk was the major supply to the fistula. A detachable silicone balloon was flow-directed through this vessel into the fistula and inflated, resulting in complete closure of the arteriovenous connection. Twenty seconds after closure the patient developed severe headache, vertigo, and disorientation. One minute after closure she developed a dense left homonymous hemianopsia. With deflation of the balloon and reestablishment of the fistula flow, her symptoms resolved completely within 5 min. These symptoms were reproduced whenever the fistula was abruptly closed. It was therefore decided to slowly occlude the fistula to allow gradual reestablishment of cerebral autoregulation.

The balloon was positioned through the deep cervical artery, across the fistula to the venous side and slowly inflated until the patient redeveloped the symptoms described above. The balloon was then deflated slightly until the symptoms abated, and it was maintained at that inflation volume for 20 min. Further inflation resulted in the reestablishment of symptoms, and again the balloon was slightly deflated. This gradual incremental inflation was repeated for 2 hr until

the fistula was completely closed without neurologic deficits. Another balloon was positioned on the venous side and detached, and a third balloon was positioned at the site of the fistula. A control angiogram revealed complete closure of the fistula and reestablishment of antegrade flow in the distal left vertebral and basilar artery. The patient's weakness and clumsiness gradually improved over the next few weeks.

Case 3

A 30-year-old man presented with a long history of neck pain and bruit. The year prior to treatment he developed exercise intolerance. An angiogram documented a large left vertebral fistula located at C1 (Fig. 3). Transarterial balloon occlusion was performed via the left vertebral artery. When the balloon was inflated, producing abrupt occlusion of the fistula, the patient developed severe headaches and vertigo. To prevent the development of normal perfusion pressure breakthrough, a staged closure was performed. Seven balloons were positioned in the venous side of the fistula and detached, producing subtotal occlusion of the fistula. A follow-up angiogram 7 weeks later revealed an interval decrease in caliber of the ipsilateral vertebral artery and diminished flow in the fistula. Three more balloons were positioned in the fistula, producing complete closure of the fistula without symptoms.

Case 4

A 36-year-old man developed a traumatic carotid cavernous fistula at age 21. At age 29 he was treated with surgical exposure of the fistula and placement of copper wires within the cavernous sinus in an attempt to thrombose the fistula and improve his declining vision. The fistula remained patent and his condition slowly deteriorated to total blindness. At age 36, 15 years after the onset of his fistula, he presented for treatment of severe headaches related to the fistula. An angiogram (Fig. 4) showed a large direct fistula with enlargement of the caliber of the involved carotid artery. During the initial transvascular balloon embolization procedure, several balloons ruptured

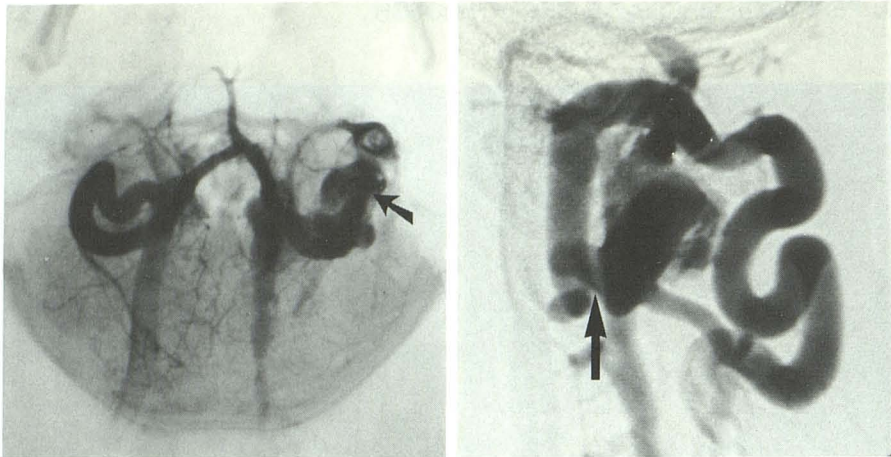


Fig. 2.—A, Right vertebral artery injection, submental vertex projection. There is retrograde flow down the distal left vertebral artery into a fistula (arrow) located at C2.

B, Left deep cervical artery injection, lateral view. This tortuous vessel supplies the same fistula (arrow).

C, Same injection. A balloon (arrows) has been positioned in proximal draining vein and inflated to cause partial occlusion.

D, Same vessel. There is complete occlusion of the fistula by three detached balloons, and antegrade flow up the basilar artery.

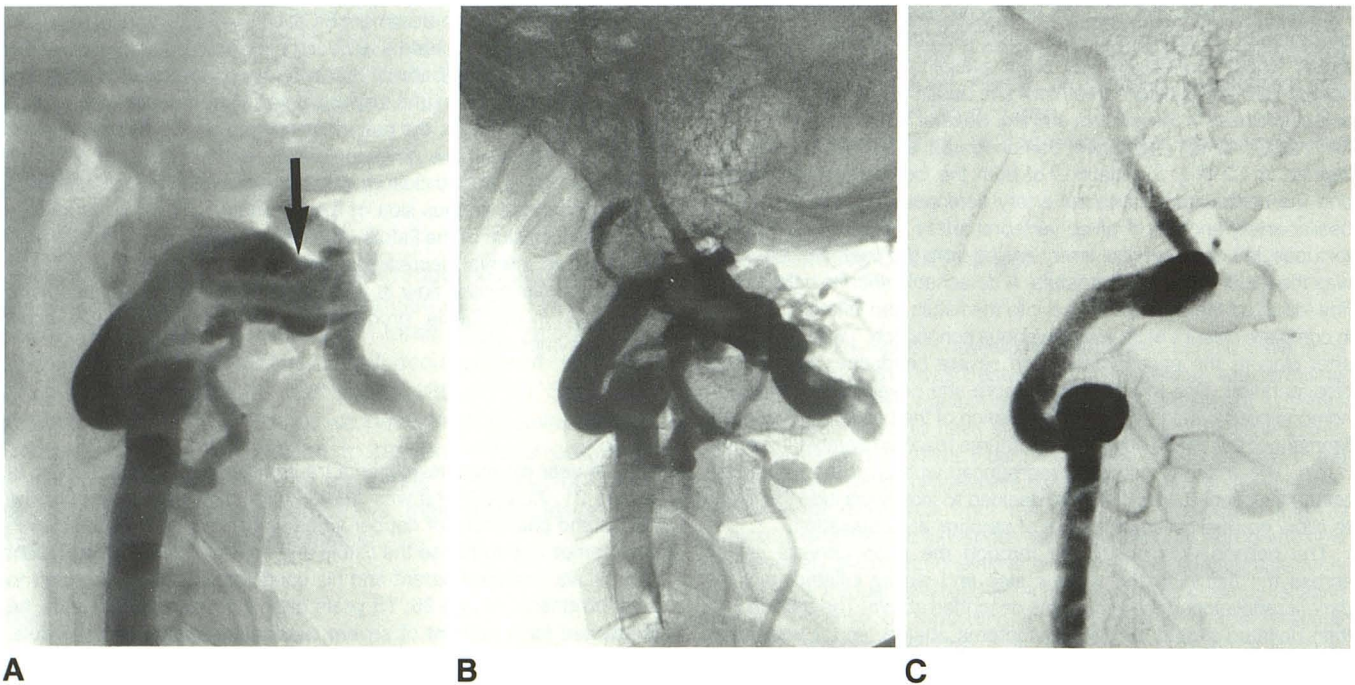
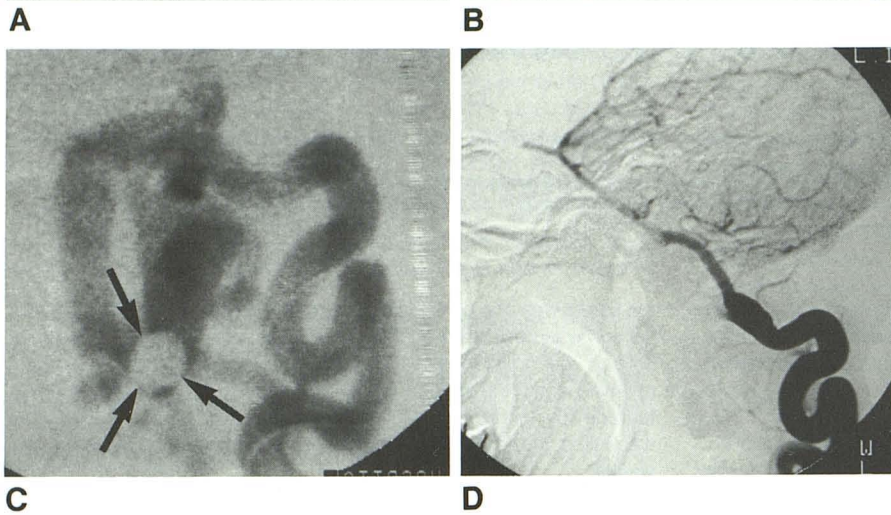


Fig. 3.—A, Left vertebral artery injection, lateral view. A large vertebral fistula (arrow) is located at C1.

B, Same vessel. Seven detachable balloons have been positioned on venous side of fistula and detached, causing partial closure.

C, Left vertebral injection, lateral view, 7 weeks later. Several balloons have deflated and three more balloons have been positioned to completely occlude the fistula without symptoms. Notice the interval decrease in size in caliber of the vertebral artery.

Fig. 4.—A, Selective right internal carotid artery injection, lateral projection showing large carotid cavernous fistula with dilated cavernous sinus. Note large caliber of internal carotid artery. Short pieces of copper wire project within and around the cavernous sinus.

B, Same vessel and projection after a staged closure procedure with reestablishment of intracranial flow and complete closure of fistula.

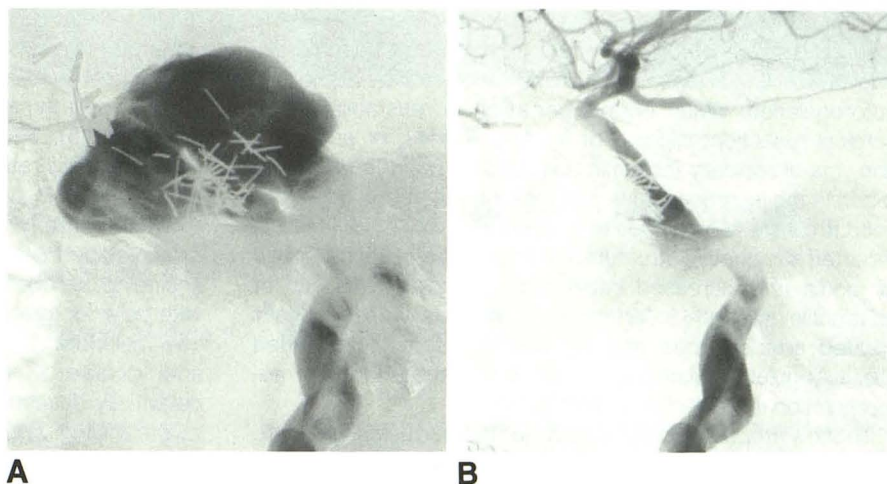
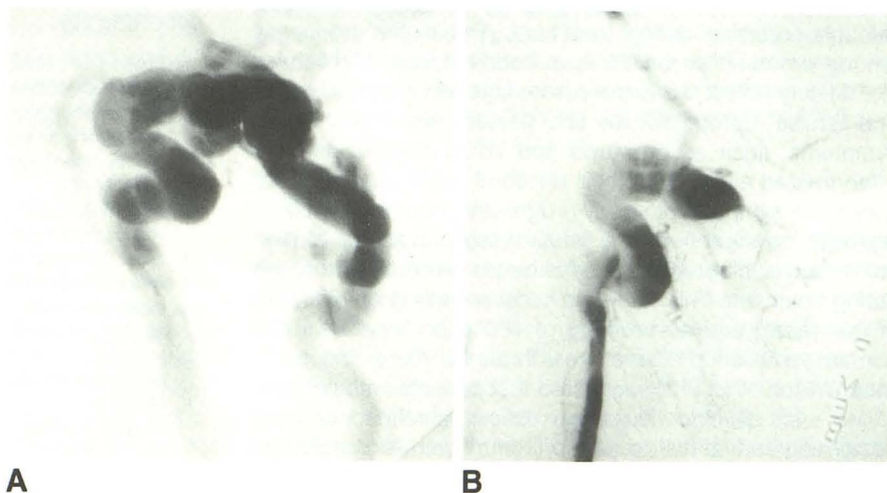


Fig. 5.—A, Right vertebral artery injection, lateral view, shows a large fistula located at C1. Note enlargement of the feeding artery.

B, Same vessel, same projection, postembolization. Gradual occlusion of fistula was performed, yielding complete fistula closure without permanent neurologic deficits.



because of the copper wires projecting into the cavernous sinus. Therefore, the subsequent balloons were inflated slowly and observed for deformation of their shells. The final inflation volume was kept to a minimum to reduce stress on the balloon shell. Transvascular balloon occlusion was performed, and with complete closure of the fistula, the patient developed a severe headache, nausea, vomiting, and altered mental status. A subtotal closure of the fistula was performed to allow for the development of autoregulation in the affected hemisphere. Four days later an additional balloon was placed and detached with complete closure of the fistula. No neurologic symptoms developed.

Case 5

A 61-year-old woman had a history of neck pain and bruit for approximately 20 years. She had no history of trauma or collagen-deficiency diseases. An angiogram demonstrated a large arteriovenous fistula located at C1 on the right (Fig. 5A). The caliber of the feeding vertebral artery was enlarged. A detachable silicone balloon was inflated in the fistula, producing abrupt occlusion. Several minutes later the patient developed severe quadraparesis. The balloon was deflated and flow was reestablished in the fistula; the quadra-

paresis completely resolved over the next 10 min. This symptom complex was reproduced whenever the fistula was completely occluded, and it resolved whenever the fistula was reopened. Therefore, to allow the reestablishment of cerebral autoregulation, a balloon was positioned in the venous side and inflated to produce subtotal occlusion. No neurologic deficits developed. Three weeks later a repeat embolization procedure was performed. With abrupt occlusion of the fistula the patient again developed quadraparesis. Therefore, a gradual occlusion was performed. The balloon was slowly inflated until the patient developed neurologic deficits, then it was deflated slightly until the deficits resolved and maintained at that volume for 20 min. It was then inflated slightly until the symptoms recurred, then deflated slightly and held at this new inflation for another 20 min. Using this slow, progressive step-wise inflation technique, the fistula was occluded over a 2-hr period without permanent neurologic sequela (Fig. 5B).

Discussion

Normal perfusion pressure breakthrough theory was described by Spetzler and Wilson [1] in 1978 to explain hemorrhage from adjacent normal brain after successful removal

of large AVMs. They postulated that chronic high-flow AVMs induced reactive hypotension in adjacent normal brain parenchyma resulting in chronic arteriolar dilation and loss of normal autoregulation. When normal perfusion is reestablished by surgical resection, clipping of feeding arteries, or embolization, the chronically ischemic surrounding brain is unable to regulate the increased flow, and malignant cerebral edema or uncontrollable hemorrhage may occur. Folkow et al. [10, 11] reported an adaptive structural change consisting of reduction of media and increased intraluminal diameter with loss of contractile strength in arterioles in animals with surgically created arteriovenous fistulas. Spetzler et al. [1] created carotid-jugular fistulas in cats and demonstrated loss of autoregulation in the ischemic hemisphere.

The low frequency with which normal perfusion pressure breakthrough occurred during the treatment of carotid and vertebral fistulas in our series may correspond to the length of time between the development of the fistula and treatment. There was a strong correlation between the duration of the fistula and the chance of normal perfusion pressure breakthrough occurring during treatment. The higher frequency among vertebral fistulas (15%) as opposed to carotid fistulas (1.2%) may reflect the larger percentage of congenital vertebral fistulas. Carotid fistulas also present with more severe symptoms, such as proptosis and visual decline, and are often treated earlier than their vertebral counterparts. In our series, the symptom of slowly progressive neurologic deficits representing steal from the fistula was an indicator of high risk in developing normal perfusion pressure breakthrough during treatment. This symptom had a sensitivity of 40% (two of five cases) and a specificity of 100% (both patients had normal perfusion pressure breakthrough). Along the same lines, Wilson et al. [12] suggested that patients with cerebral AVMs who develop neurologic deficits unrelated to prior hemorrhage are at risk to develop normal perfusion pressure breakthrough after surgical resection.

Neurologic deficits occurring during transvascular or surgical closure of carotid or vertebral fistulas may result from a number of causes. Ischemia may develop secondary to spasm, narrowing, or occlusion of surrounding vessels supplying or draining normal parenchyma. Thrombus formation on catheters or propagation of clot may cause distal embolization, which can result in neurologic deficits. Toxicity from repeated injections of contrast material can also cause neurologic sequelae. The development of neurologic deficits immediately after the abrupt closure of a fistula, and prompt resolution of those deficits after reestablishment of fistula flow, indicate that the ischemic vascular territory is unable to accommodate the restoration of perfusion. Gradual and staged occlusion were used in our series to allow restoration of normal autoregulation. A similar approach is recommended in the surgical management of large cerebral AVMs to prevent the development of normal perfusion pressure breakthrough. Many authors have suggested staged surgical clipping of

feeding vessels or preoperative embolization [1, 2, 6, 13], while others have advocated preventive measures, including lowering of systemic pressure by drug-induced hypotension or proximal internal carotid stenosis with a Selverstone clamp [1, 2, 13-15]. Mullan et al. [6] recommended the use of reversible clips rather than permanent ligatures during resection of AVMs in patients at risk to develop perfusion breakthrough. Similarly, in case 2, to allow percutaneous puncture if delayed perfusion breakthrough occurred, we left the critical occluding balloon filled with contrast rather than solidify it with polymer (our standard practice).

In conclusion, normal perfusion pressure breakthrough after closure of carotid and vertebral fistulas is a rare but potentially devastating occurrence. The presence of a large, long-standing fistula with associated cerebral ischemia indicates a high probability that this phenomenon will develop. Gradual or staged closure may prevent this complication by permitting the restoration of normal cerebral autoregulation.

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