Transarterial Occlusion of Solitary Intracerebral Arteriovenous Fistulas

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Congenital solitary arteriovenous fistulas unassociated with arteriovenous malformations are rare. Five patients with this condition were treated by endovascular procedures: three by transarterial balloon occlusion and two by coil and silk suture embolization. Of the three patients treated by balloon occlusion, complete angiographic obliteration was achieved in two, and one patient had subtotal occlusion resulting from proximal balloon placement. Two of these patients subsequently underwent surgical excision without incident. There were no complications. Two patients were treated with a combination of platinum coils and silk suture embolization, which resulted in complete angiographic obliteration in both. There was one asymptomatic complication consisting of a coil migrating through the fistula and lodging in the lung.

While detachable balloons have advantages in allowing test occlusion prior to detachment, the traction required for detachment can limit accurate deposition and place the feeding arteries under undue stretch. Coil and silk suture embolization enable more precise deposition and are probably the agent of choice for closure of solitary arteriovenous connections within the central nervous system.

Congenital arteriovenous connections within the CNS can be classified microscopically into capillary telangiectasias, cavernous angiomas, and venous and arteriovenous malformations (AVMs) [1]. The latter category accounts for the majority of clinically apparent intracerebral connections and most are composed of numerous small channels between arteries and veins without intervening capillaries. The size of the arteriovenous connections that occur in AVMs can vary widely and reach considerable size in some instances. While solitary congenital arteriovenous fistulas are common in the scalp, there are relatively few reports of this condition within the CNS.

Of the 320 AVMs treated by the authors over the past 10 years, five (1.6%) had a single arteriovenous connection. All five were treated by transvascular embolization techniques, three with detachable balloons and two with a combination of platinum coils and silk sutures.

Materials and Methods

Five patients (two males, three females) ages 4 to 25 years old with solitary intracerebral arteriovenous connections were treated by transarterial embolization techniques. The presenting symptoms were hemorrhage (cases 2 and 3), headaches (cases 1, 4, and 5), and neurologic deficit (cases 1 and 4). One patient (case 2) had two unsuccessful attempts at neurosurgical excision that were complicated by hemorrhage from arterialized draining veins. All five patients were treated from a transfemoral arterial access. Three patients (cases 1–3) were treated with detachable silicone balloons.* The balloons are manufactured in three detachment strengths: low, medium, and high. Low-detachment balloons require only 20–25 g of force for detachment from the 2-French catheter and were used in all three cases. This allowed for traction detachment at the desired site with relatively little force transmitted to the malformation or feeding arteries. Conversely, this small amount of force required to

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detach makes navigation and delivery more hazardous because of the risk of premature detachment. Considerable care and experience are needed to safely deliver this device. Following systemic heparin anticoagulation, a 7.3-French polyethylene catheter was placed in the distal carotid or vertebral artery. A low-detachment (20 g of force) silicone balloon attached to a 2/4-French coaxial polyethylene catheter system was placed through the 7.3-French catheter. The balloon was navigated to the fistula site and inflated with slightly hypertonic metrizamide (200 mg%) to produce complete fistula occlusion. Careful neurologic evaluation was performed and, if tolerated, the balloon was detached by gentle traction. Additional balloons were detached (in case 3) to ensure fistula closure should the first balloon deflate. Systemic anticoagulation was reversed with protamine sulfate (1 mg reverses 100 units of circulating heparin) given slowly over 15 min. Postembolization angiograms were obtained from all potential collateral arterial sources.

Two patients (cases 4 and 5) underwent transarterial embolization with platinum coils and pieces of silk sutures. A 5.5-French polyethylene catheter was positioned in the distal internal carotid artery and systemic anticoagulation achieved with IV heparin. A 3.2-French Tracker catheter with a 0.014-in. platinum guidewire was navigated to the fistula site. Superselective angiograms were obtained to ensure proper placement. Platinum coils measuring 4 mm in diameter were delivered to the fistula site with the use of a Teflon coil pusher. After the coils were placed, short pieces of 4-0 silk suture (range, 2–4 cm in length) were injected through the tracker catheter proximal to the fistula to entangle with the platinum coil and produce thrombosis. When fistula obliteration was observed, the catheters were removed, systemic anticoagulation reversed with IV protamine sulfate, and postembolization angiograms obtained from all potential arterial collaterals.

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Fig. 1.—Case 1. 4-year-old boy with headaches and left arm weakness.

A, Contrast-enhanced CT shows large enhancing structure above right tentorium.

B, Left vertebral angiogram, Towne's projection, shows a fistulous connection supplied from the right posterior temporal artery draining into a large varix.

C, Same injection and projection, after balloon embolization, shows complete obliteration of fistula.

D, Follow-up noncontrast CT scan shows high-density balloon (curved arrow) and high-density acute thrombus within varix (straight arrow).

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Representative Case Reports

Case 1

A 4-year-old boy presented with left arm weakness and headaches. A contrast-enhanced CT scan (Fig. 1A) demonstrated a dilated vascular structure medial to the posterior temporal lobe. An angiogram (Fig. 1B) showed a fistula between the right posterior temporal artery and a tortuous varix. A single detachable balloon was placed in the connection and detached. A follow-up angiogram (Fig. 1C) and a noncontrast CT scan (Fig. 1D) demonstrated complete closure of the fistula and thrombosis within the varix. The patient subsequently recovered complete strength, his headaches resolved, and is doing well 5 years later.

Case 2

A 25-year-old woman had a subarachnoid hemorrhage associated with an arteriovenous connection supplied by the right parietooccipital artery (Fig. 2A). Two surgical attempts at resection were unsuccessful because of excessive hemorrhage from arterialized draining veins. A single detachable balloon was navigated to the fistula site, but migrated proximally during detachment. The immediate follow-up angiogram demonstrated near complete obliteration of the fistula (Fig. 2B). A 6-week follow-up angiogram (Fig. 2C) demonstrated increased fistula flow supplied by transcortical collaterals distal to the balloon. Surgical excision was performed without incident.

Case 4

A 14-year-old girl developed severe unilateral headaches and diplopia. An angiogram (Fig. 3A) demonstrated a solitary arteriovenous connection supplied by two branches of the right posterior temporal artery. A 3.2-French Tracker catheter was navigated to the fistula site. A platinum coil was deposited at this site; however, because of the rapid flow, it passed through the fistula site and lodged in a small left upper-lobe pulmonary artery without symptoms. Additional platinum coils were then deposited just proximal to the fistula site and short pieces of silk sutures were entangled with the coils to promote thrombosis. A postembolization angiogram (Fig. 3B) demonstrated complete fistula obliteration, and the patient has had complete resolution of symptoms. A follow-up angiogram at 3 months (Figs. 3C and 3D) confirmed complete cure.

Case 5

A 10-year-old girl presented with unilateral headaches. An angiogram revealed a solitary connection supplied by a parietal branch of the left middle cerebral artery (Fig. 4A). Several platinum coils were deposited at the fistula site and silk suture pieces were placed proximally, resulting in complete fistula obliteration (Fig. 4B). A 2-month follow-up MR image disclosed complete fistula thrombosis, and the patient has had complete resolution of her symptoms.

Results

Three patients were treated with transarterial detachable balloons, and complete angiographic obliteration was documented in two (cases 1 and 3). Case 3 had subsequent surgical excision without incident. The remaining patient (case 2) had subtotal fistula obliteration resulting from the occlusion balloon being proximal to the fistula site. This patient was treated with surgical excision without incident. In the two patients treated by surgical excision after balloon embolization, postsurgical angiograms documented fistula closure. The third patient (case 1) had a follow-up noncontrast CT scan (Fig. 1D) that demonstrated thrombosis of a varix. This patient has had complete resolution of his presenting symp-

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Fig. 2.—Case 2. 25-year-old woman with subarachnoid hemorrhage in whom surgical ligation was attempted without success. A. Right vertebral angiogram, Towne’s projection, shows fistula (curved arrow) supplied by parietooccipital artery. B. Left vertebral angiogram, after embolization, shows a balloon (straight arrow) proximal to fistula site. The distal parietooccipital artery fills slowly from cortical collaterals (open arrow). C. Follow-up angiogram, 6 weeks later, shows increased filling of fistula (open arrow). The balloon (straight arrow) is unchanged.

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Tombs of arm weakness and headaches, and remains asymptomatic. The clinical follow-up period ranged from 30 to 66 months (mean, 52 months).

Two patients (cases 4 and 5) were treated by transarterial deposition of platinum coils and silk suture emboli. Both patients had complete obliteration of fistula flow on their postembolization angiograms. One patient (case 5) had a follow-up MR scan at 2 months that demonstrated complete

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Fig. 3.—Case 4. 14-year-old girl with cranial nerve palsies and headaches.
A. Right internal carotid angiogram, Towne's projection, shows solitary fistula supplied by two posterotemporal branches of right posterior cerebral artery.
B. Same injection and projection, after embolization of two distal posterotemporal arterial feeders with platinum coils (arrowheads) and silk sutures, show complete fistula obliteration.
C. Same injection and projection, at 3-month follow-up, show persistent thrombosis of fistula by coils (arrows).
D. Vertebral injection, Towne's projection, also shows occlusion of fistula.

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Fig. 4.—Case 5. 10-year-old girl with headaches.
A. Left internal carotid angiogram, left anterior oblique projection, shows fistula supplied by enlarged parietal branch of middle cerebral artery.
B. Same injection and projection, after platinum coil (arrow) and silk suture embolization, show complete fistula obliteration.
varix thrombosis. The second patient (case 5, Fig. 4) had a follow-up angiogram at 3 months that showed complete occlusion of the fistula. Both patients have had resolution of their presenting symptoms. The clinical follow-up period was short, only 7 months in each patient.

There were no symptomatic complications resulting from either detachable balloon or coil embolization. Case 5 had a single 4-mm platinum coil pass through the fistula site and lodge in the lung without symptoms.

Discussion

Direct arteriovenous connections between both the vertebral and carotid arteries and surrounding veins have been effectively treated with detachable balloons [2–4]. Intracerebral arteriovenous connections without an associated nidus are rare. The relatively young age of onset of symptoms in our series of five patients may reflect the enormous flow that occurs with a large solitay connection as opposed to typical AVMs, which are reported to present at a later age. Conversely, it may reflect the earlier diagnosis afforded by newer imaging technologies. Vinuela et al. [5] reported a congenital intracranial arteriovenous fistula supplied by the posterior inferior cerebellar artery treated successfully by detachable balloon therapy.

The choice of treatment for solitary arteriovenous fistulas is limited. While surgical ligation of the fistula is sometimes possible, the large draining veins can obscure and interfere with the exposure of the fistula. Because of the long-standing shunt, arterIALIZATION and thickening of the draining veins can occur, making identification of the exact fistula site difficult. In case 2, two surgical attempts at fistula ligation were complicated by excessive bleeding. After subtotal occlusion by a detachable balloon, surgical excision was performed without difficulty.

Balloon embolization of cerebral AVMs was first described by Serbinenko [3]. Others [5–7] have noted that collaterals develop within the malformation, and proximal occlusion by balloons or ligatures is largely ineffective in the long-term treatment of AVMs. Detachable balloons have been used effectively preoperatively to reduce the pressure within the nidus, to occlude deep or inaccessible feeding arteries, and to prevent normal perfusion pressure breakthrough [5, 8, 9]. Balloons deposited by transarterial methods must be detached by traction. If too much traction is needed to detach the balloon, then stretching or tearing of the surrounding vasculature can occur. If too little traction is needed to detach the balloon, then undesirable, premature detachment during placement is possible. The balloon may migrate proximally during detachment if insufficiently inflated. Conversely, if the balloon is inflated too much, pressure necrosis of the wall may occur. Platinum coils delivered through small catheters can be more accurately deposited than balloons. However, the catheters have no flow advantage in their delivery as do their balloon counterparts, which may make their placement difficult. In addition, test occlusion of the pedicle cannot be performed with platinum coils. The earliest platinum embolic agents were the ends of used platinum guidewires [10]. Recently, more sophisticated occlusion devices have been made available with complex coils** and coils with thrombogenic synthetic fibers.††

When a rare solitary arteriovenous connection exists, transarterial occlusion with balloons or platinum coils can be curative. None of the five patients in this series developed neurologic deficits resulting from the treatment. Care must be taken to position the occlusive device as close as possible to the arteriovenous connection. In case 2, the balloon was detached proximal to the fistula, resulting in recanalization through transcortical collaterals. In case 3, subsequent surgical excision of the fistula site was performed despite complete angiographic obliteration after balloon embolization. This was done, in one of our earliest cases, because of concerns about delayed recanalization, which can occur in true AVMs after balloon embolization [5–7]. We are now confident that if the fistula site is completely occluded with a balloon or platinum coil, then subsequent surgical excision is not necessary. If the occlusion device were to be detached distal to the fistula, increased pressure could be transmitted to draining veins, resulting in cortical venous hypertension or hemorrhage. The occlusion device can possibly dislodge and pass into larger draining veins or to the lungs, as occurred in case 4. In all five cases, there was a narrowing at the junction point between the artery and vein. There was also a secondary site of narrowing in the draining vein in case 3, where the vein entered the transverse sinus. None of the patients developed neurologic deficits resulting from the occlusion. In cases 1 and 4, the presenting neurologic deficits resolved after closure of the fistula. The mechanism of the deficits may be venous hypertension associated with the large shunt or arterial ischemia.

Several potential hazards may occur with the abrupt closure of a large intracerebral connection. Alterations in flow may overperfuse adjacent normal parenchyma that has lost the ability to autoregulate (normal perfusion pressure breakthrough), which could result in hemorrhage [11, 12]. Venous thrombosis can occur with closure of an arteriovenous fistula draining into a single vein or varix [13]. Although none of these complications occurred in this small series, it is important to be aware of them as potential hazards. Careful, repeated neurologic examinations after occlusion of the fistula are important to detect symptoms that could herald these complications. In our series, test occlusion of the fistulas was performed in cases 2 and 3 for 30 min without change in neurologic function.

In conclusion, direct solitary intracerebral arteriovenous connections are rare; can present with hemorrhage, headaches, or neurologic deficits at a young age; and can be treated by either detachable balloons or platinum coil and silk suture embolization techniques.

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