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Treatment of Vertebral Arteriovenous Fistulas

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Twenty patients with vertebral arteriovenous fistulas (eight spontaneous, six traumatic without vertebral artery transection, and six traumatic with vertebral artery transection) were treated by transvascular embolization techniques, resulting in complete fistula closure in all patients. The fistulas were located at C1–C2 in 45%, C2–C3 in 25%, C4–C5 in 15%, C5–C6 in 10%, and C6–C7 in 5%. Trauma was the most common cause: 30% followed knife wounds, 20% followed gunshot injuries, and 10% followed blunt trauma. Eight patients had spontaneous fistulas, two associated with fibromuscular dysplasia. Three patients—all with large, long-standing fistulas—developed neurologic deficits coincident with the abrupt closure of the fistula, which resolved with reestablishment of fistula flow. Two of these patients were treated by staged closure; the other one by gradual closure. In all three cases the result was complete fistula closure without neurologic sequelae. The remaining spontaneous fistulas were all closed by balloon embolization with preservation of the vertebral artery and without deficits. The six patients with traumatic fistulas without transection were cured by balloon embolization, without deficits; in four there was also preservation of vertebral flow. The other six patients had traumatic fistulas with transection and were all cured by balloon embolization with preservation of flow in two. Four patients required bilateral approaches to the fistula to achieve complete fistula closure. The only complication was a mild residual Wallenberg syndrome after occlusion of the posterior inferior cerebellar artery in the treatment of a transection located at C1.

In our opinion, transvascular techniques are the treatment of choice for vertebral arteriovenous fistulas.

Vertebral arteriovenous fistulas are abnormal connections between the extracranial vertebral artery or its branches and neighboring veins. They are uncommon lesions, with only 81 cases reported prior to 1977 [1]. The most common cause of these fistulas is penetrating neck injuries, usually knife wounds or gunshot injuries. Iatrogenic causes include the sequelae of direct puncture of the carotid or vertebral artery for diagnostic angiography [2, 3], inadvertent vertebral damage during the insertion of venous catheters [4, 5], or complications of anterior interbody fusion [6]. Diseases associated with vertebral arteriovenous fistulas include fibromuscular dysplasia [7–9] and neurofibromatosis [10–12]. Blunt trauma in association with cervical spine fracture has been reported as a cause [13, 14]. Many cases are spontaneous [15] and often thought to be congenital. Symptoms often related to the fistula are bruit and neck pain, but brain and spinal cord dysfunction related to steal, venous hypertension, or mechanical compression have also been reported [1, 16]. Traumatic fistulas can present with massive hemorrhage, neurologic deficits, expanding hematomas, pseudoaneurysms, and airway obstruction. Treatments have included proximal ligation, trapping procedures, direct surgical exposure and closure, combined balloon embolization and surgical ligation, and, more recently, embolization with balloons or steel coils [16–20]. Over the past 7 years we have treated 20 vertebral arteriovenous fistulas by transvascular embolization techniques, and we report the results of those treatments.

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Materials and Methods

Twenty patients with vertebral fistulas were treated by using transarterial embolization. All procedures were performed with local anesthesia and IV sedation to allow continuous neurologic monitoring. The femoral arterial access was used for all diagnostic and therapeutic procedures. A diagnostic angiogram with catheter tip located near the fistula was obtained via the involved vertebral artery, and rapid filming was used to delineate the site of the fistula, presence of arterial narrowing or damage, and draining venous structures. If antegrade flow up the involved vertebral artery above the fistula was not observed, then gentle advancement of a soft-tipped guidewire was attempted to place the diagnostic catheter above the level of the fistula. If successful, an angiogram was obtained with the catheter tip located just above the fistula site. Angiograms of the contralateral vertebral, ipsilateral carotid, thyrocervical, and costocervical arteries were obtained as indicated. If continuity of the artery was maintained (no transection) then a 7.3/5-French coaxial catheter system was placed in the artery with the safest access to the fistula, usually the involved vertebral artery. Systemic anticoagulation with heparin, usually 5000 units bolus and 2500 units/hr in a 70-kg patient, was given unless contraindicated. If the patient had a pseudoaneurysm or expanding hematoma, then anticoagulation was not used. The inner 5-French catheter was removed and a 4/2-French coaxial polyethylene catheter system with a silicone detachable balloon attached to the 2-French catheter was placed through the 7.3-French catheter. The space between the 4- and 7.3-French catheters was perfused with heparinized saline through a sidearm attachment. Injection of contrast material can also be made through this space during balloon placement. The balloon was advanced under fluoroscopic guidance into the fistula and inflated. If in satisfactory position, the balloon was detached by traction or coaxial technique. Attempts were made to preserve vertebral arterial flow if possible.

If vertebral transection was present, an attempt was made to place the catheter system into the distal transected segment via the ipsilateral vertebral artery.

If a vertebral transection was present and the diagnostic catheter could not be advanced into the distal vertebral artery, then a contralateral approach was initially used. The balloon was flow-directed up the contralateral vertebral artery across the junction of the vertebral arteries, down the distal ipsilateral vertebral artery, and to a point above the level of the fistula. If in satisfactory position, the balloon was then detached by gentle traction. The 7.3-French catheter was then placed in the ipsilateral vertebral artery and balloons were positioned in the fistula site and ipsilateral vertebral artery near the site of the fistula. After final detachment, a control angiogram was obtained from all potential collateral sources that could supply the fistula. If the fistula was occluded, the heparin anticoagulation was reversed with IV protamine sulfate (1 mg of protamine sulfate counteracts 100 units of circulating heparin) given slowly over 5 min. Follow-up angiograms were obtained if the patient had symptoms related to the fistula or if the control angiogram demonstrated subtotal occlusion. The patients were followed by the authors at 1-month, 6-month, and yearly intervals after treatment. The follow-up period was 5–72 months (average, 35 months).

The age and gender of each patient, location and cause of the fistula, symptoms, presence of vertebral transection, treatment, and complications are summarized in Table 1.

Results

Nineteen of 20 patients were treated by transarterial balloon embolization. In the other patient (case 12) the orifice of the fistula was too small to permit the balloon to enter the fistula; therefore, a balloon was transiently positioned in the vertebral artery above the fistula and silicone spheres were embolized behind (proximal to) the balloon into the fistula. All 20 patients had complete closure of their fistulas by transvascular treatments.

TABLE 1: Summary of Vertebral Fistulas in Patients Treated with Transvascular Embolization

Case No.	Age	Gender	Site	Cause	Symptoms/Findings	Vertebral Artery Transection/Occlusion	Treatment
1	61	F	R C1	Spontaneous	Bruit	Occlusion	Three balloons
2	23	F	L C1	Spontaneous	Weakness	No	Three balloons
3	29	F	R C4–C5	Spontaneous	Neck pain, radiculopathy	No	One balloon
4	27	M	L C2–C3	Gunshot wound	Hemorrhage	Occlusion	Three balloons
5	24	M	R C1	Gunshot wound	External hemorrhage	Transection/occlusion	Six balloons
6	20	M	L C1	Blunt trauma	Bruit, pain	Transection	Seven balloons
7	20	M	R C5–C6	Spontaneous	Subarachnoid hemorrhage	No	One balloon
8	64	F	L C2–C3	Fibromuscular dysplasia	Bruit, pain	No	One balloon
9	35	M	L C1	Spontaneous	Bruit, pain	No	Two balloons
10	20	M	R C1	Blunt trauma	Bruit	Transection/occlusion	Ten balloons
11	30	M	L C1	Spontaneous	Exercise intolerance	No	Four balloons
12	26	F	L C2–C3	Fibromuscular dysplasia	Pain, bruit, quadriplegia	No	Silastic spheres
13	60	M	R C6–C7	Gunshot wound	Hematoma	No	One balloon
14	61	M	R C4–C5	Stab wound	Bruit	No	One balloon
15	25	M	R C1	Gunshot wound	Bruit	Transection	Six balloons
16	28	M	L C2	Stab wound	External hemorrhage	Transection/occlusion	Five balloons
17	21	M	L C1	Stab wound	External hemorrhage	Transection/occlusion	Three balloons
18	28	M	L C2–C3	Stab wound	External hemorrhage	No	Three balloons
19	26	M	L C4–C5	Stab wound	Pain, hematoma	Occlusion	Two balloons
20	29	M	L C5–C6	Stab wound	Radiculopathy, pain	No	Four balloons

Note.—Treatment resulted in fistula closure in all cases. There was only one complication, occlusion of the posterior inferior cerebellar artery in case 6. R = right; L = left.

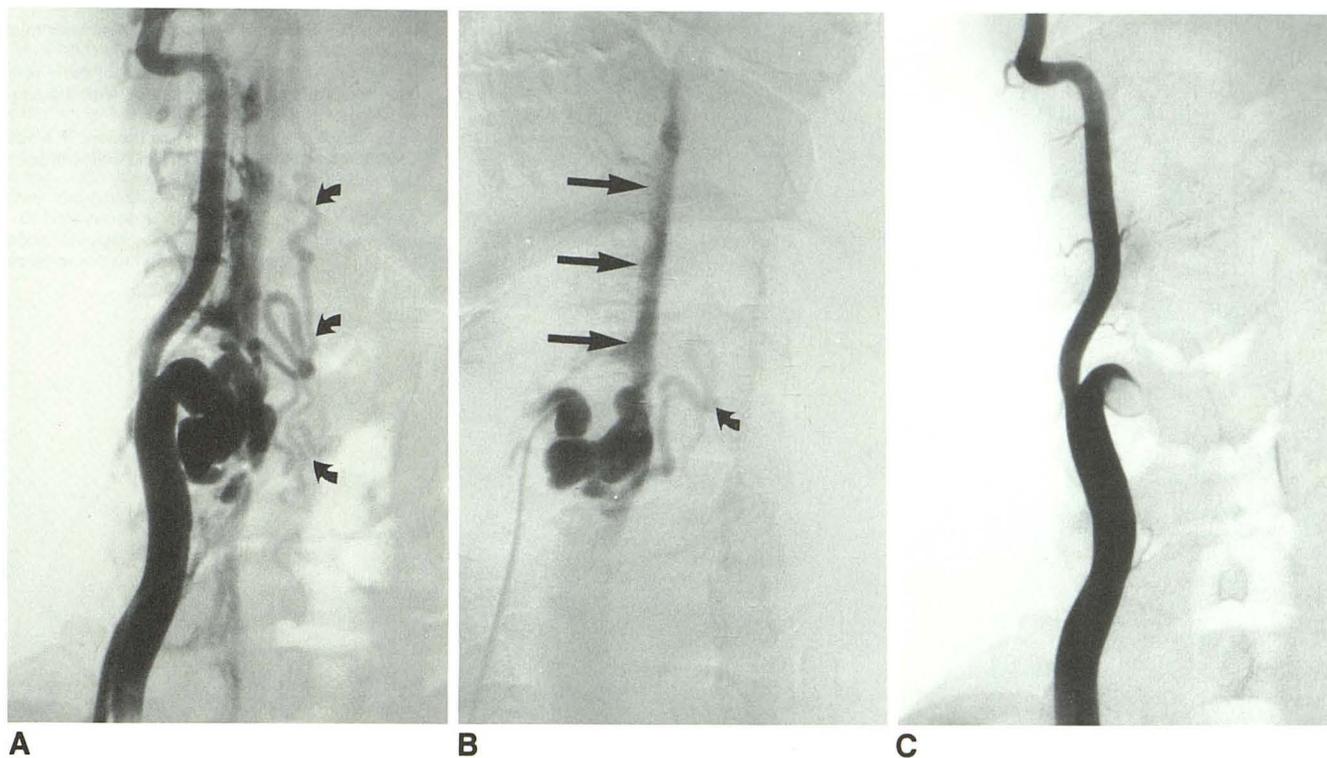


Fig. 1.—Case 7: 20-year-old man with spontaneous vertebral fistula at C5–C6 who presented with massive subarachnoid hemorrhage. A, Anteroposterior view of right vertebral artery injection shows vertebral fistula with venous drainage to epidural venous plexus and medullary veins (arrows). B, Selective radicular artery injection, oblique view, shows site of fistula with venous drainage to epidural venous plexus (straight arrows) and medullary veins (curved arrow). C, Selective right vertebral artery injection, anteroposterior view after embolization, shows balloon in fistula with preservation of vertebral flow.

Spontaneous Fistulas

In spontaneous fistulas the diameter of the involved feeding arteries is often enlarged, presumably secondary to long-standing fistula flow. Of the eight patients with spontaneous fistulas, three developed reversible neurologic deficits coincident with the abrupt closure of the fistula. With reestablishment of fistula flow the neurologic deficits promptly resolved. Fistula closure was staged in two patients, and in the third the occlusion was gradual over a 2-hr period. None sustained permanent neurologic sequelae.

In all eight patients, transarterial occlusive procedures resulted in complete closure of the fistula and preservation of flow in the involved vertebral artery. Case 7 (Fig. 1) was a 20-year-old man who had subarachnoid hemorrhage secondary to a fistula supplied by a radicular branch of the right vertebral artery with venous drainage to the epidural plexus and medullary veins. A single detachable balloon was positioned at the fistula with complete closure and no further episodes of hemorrhage. Case 1, with a large, high-flow fistula, developed delayed thrombosis in the large vertebral artery weeks after balloon embolization. This event was associated with mild pain, but without neurologic deficit, and was presumed to be secondary to the slow flow in the dilated vessel after closure.

In the two patients with fibromuscular dysplasia, complete closure was achieved without deficits.

Traumatic Fistulas Without Transection

In the six patients with traumatic fistulas without transection, the involved vertebral artery was preserved in four. In the other two patients narrowing and intimal damage in the involved artery prevented preservation of flow. All procedures were performed from the involved vertebral artery and none resulted in neurologic deficits.

Case 14 (Fig. 2) was a 61-year-old man who developed a right vertebral fistula after a penetrating trauma. A single balloon was positioned within the fistula and detached, resulting in complete closure of the fistula.

Traumatic Vertebral Fistulas with Transection

Six patients had complete transection of the vertebral artery. In four patients penetrating injuries were responsible: two knife wounds and two gunshot injuries. In the other two patients, blunt trauma was responsible with severe whiplash in one and fracture of C1 in the other. With complete transection there is retraction of proximal and distal ends of the severed artery away from the site of the injury. There is usually a pseudoaneurysm associated with the fistula. The walls of the pseudoaneurysm are made of clot and offer little structural support unless surrounded by firm structures such as bone or ligaments. Our earliest experience with balloon

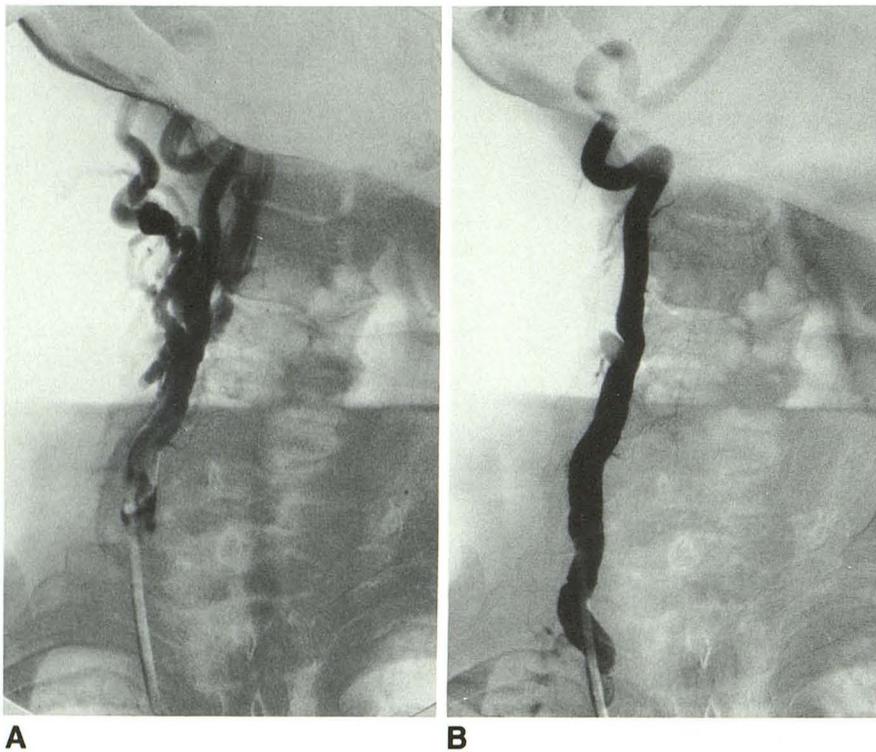


Fig. 2.—Case 14: 61-year-old man with neck injury.

A, Right vertebral artery injection, early arterial phase, shows fistula at C4-C5 with drainage to paravertebral veins.

B, Same vessel after embolization. A single balloon has been placed at fistula site with preservation of vertebral artery flow.

placement within the pseudoaneurysms was that the balloons dissected into the walls of the pseudoaneurysm causing expansion and sometimes mass effect. In case 5 (Fig. 3) there was a vertebral transection with pseudoaneurysm at the level of C1 secondary to a gunshot injury. Several balloons were positioned within the pseudoaneurysm with enlargement of this structure and persistent fistula flow. To achieve closure of the fistula, balloons were positioned in both the proximal and distal severed vertebral artery segments and detached. Because the flow in the distal vertebral is retrograde into the fistula, vertebral artery and fistula occlusion were tolerated in all six patients who required vertebral artery occlusion.

Case 17 (Fig. 4) was a 21-year-old man who developed a left vertebral transection after a knife wound. Injection of the involved vertebral artery showed filling of a pseudoaneurysm at the level of C2. Injection of the contralateral vertebral artery showed retrograde flow down the involved vertebral artery and entry into the same pseudoaneurysm at a higher level. Because of the discontinuity of the severed ends and retrograde flow down the distal left vertebral artery, treatment could not be rendered entirely from the involved side. Therefore, a balloon was flow-directed up the right vertebral artery and then down the distal left vertebral artery. It was detached above the fistula with preservation of flow in the posterior inferior cerebellar artery (PICA). Several more balloons were then detached in the distal left vertebral artery and in the site of the fistula, with complete closure of the fistula without deficits.

Case 10 (Fig. 5) was a 20-year-old man with transection at C1 with pseudoaneurysm formation. The distal vertebral artery could not be reached from an ipsilateral approach. There-

fore, a trapping procedure consisting of contralateral followed by ipsilateral occlusion resulted in a complete cure. Note the acute angle between the two vertebral arteries, which required a sharp curve in the 2-French catheter to negotiate this bend. Despite the proximity of the PICA to the fistula site, flow in this vessel was preserved.

The only complication in our series occurred during treatment in case 6, when the PICA origin was occluded. In one of our first cases, previously reported [21], the balloon could not be positioned below the PICA origin because of the proximity to the fistula. Although the patient tolerated test occlusion of the PICA for 20 min, he developed a delayed mild Wallenberg syndrome.

Discussion

The long intraforaminal course and posterior location of the vertebral artery accounts for its rare involvement in penetrating injuries of the neck. Injuries to the short first segment, from its origin to its entry into the foramen transversarium (usually at C6) usually involve other vital structures and have a high morbidity. Most traumatic vertebral fistulas involve the long second portion (intraforaminal), whereas spontaneous fistulas usually involve the third portion (where the artery leaves the foramen of the atlas to where it enters the foramen magnum) [15]. Surgical approaches to these regions have been described but are not without morbidity [22-25]. The earliest attempts at surgical control have involved ligation of the involved vertebral artery [17]. This often has resulted in decreased flow to the fistula but it also could potentiate steal down to the distal vertebral artery and result in cerebral steal

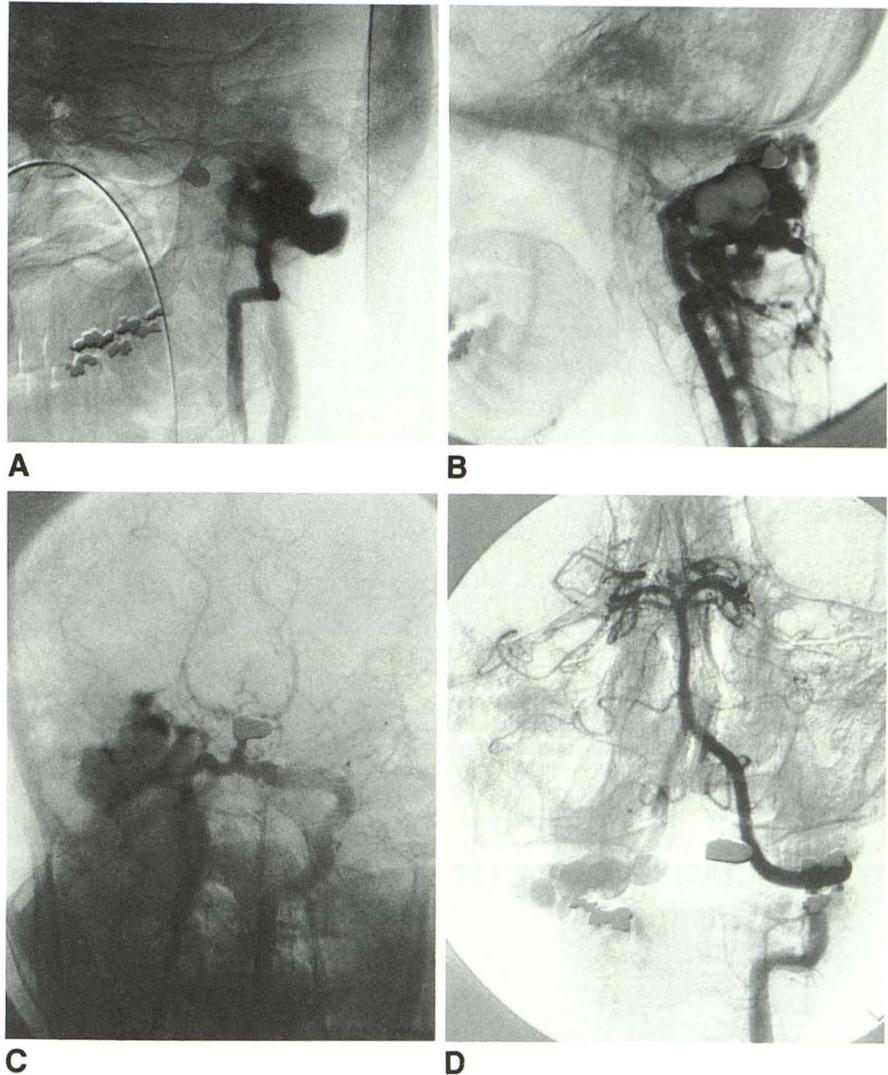
Fig. 3.—Case 5: 24-year-old man.

A, Right vertebral artery angiogram, oblique view, shows fistula at C1 with pseudoaneurysm.

B, Same vessel, lateral view, after partial embolization with persistent fistula filling and diversion of venous flow to paravertebral veins.

C, Left vertebral artery injection, Towne projection, with steal down distal right vertebral artery into fistula.

D, Left vertebral artery injection after embolization. Occlusion of fistula with preservation of posterior inferior cerebellar artery.



symptoms [26, 27]. Trapping procedures with ligation of the vertebral artery on either side of the fistula may be effective; however, collaterals may maintain fistula patency [28]. Surgical exposure of the fistula involving the second portion of the vertebral artery (intraforaminal) can be difficult. The surrounding arterialized Batson plexus can cause extensive hemorrhage during surgical exposure, and damage can occur to surrounding structures such as the phrenic nerve, brachial plexus, and cervical spinal cord [29, 30]. In an attempt to minimize the hemorrhage during surgical exposure, several techniques have been employed, including temporary occlusion of the fistula site with angioplasty catheters [31], intraoperative placement of balloon catheters at the fistula site, and hypotensive anesthesia [30]. Combined surgical and balloon embolization techniques have been used to complete trapping procedures [32–34]. More recently, trapping procedures with detachable balloons and balloon embolization with preservation of vertebral flow have been described [35–37]. Occlusion by spring coils has also been reported [38, 39].

Of the eight spontaneous fistulas in our series, three de-

veloped reversible neurologic deficits coincident with the abrupt closure of the fistula. We postulated that the chronically ischemic cerebral vasculature was unable to regulate cerebral blood flow once normal perfusion was reestablished. This has been documented experimentally in animals as well as after surgical resection of large cerebral arteriovenous malformations and is described as normal perfusion-pressure breakthrough [20]. One of our three patients presented with progressive cerebral steal symptoms before treatment, which can signify the cerebral vasculature's loss of normal autoregulation. All three patients had large, long-standing fistulas. In these three patients slow, gradual occlusion in one and staged occlusion in the other two allowed complete fistula closure without the development of permanent neurologic deficits. In addition to the three patients with vertebral fistulas we have also reported the development of normal perfusion-pressure breakthrough during the closure of carotid cavernous fistulas [40]. In all eight patients with spontaneous fistulas in our series, vertebral flow was preserved and complete fistula closure was achieved with transvascular embolization.

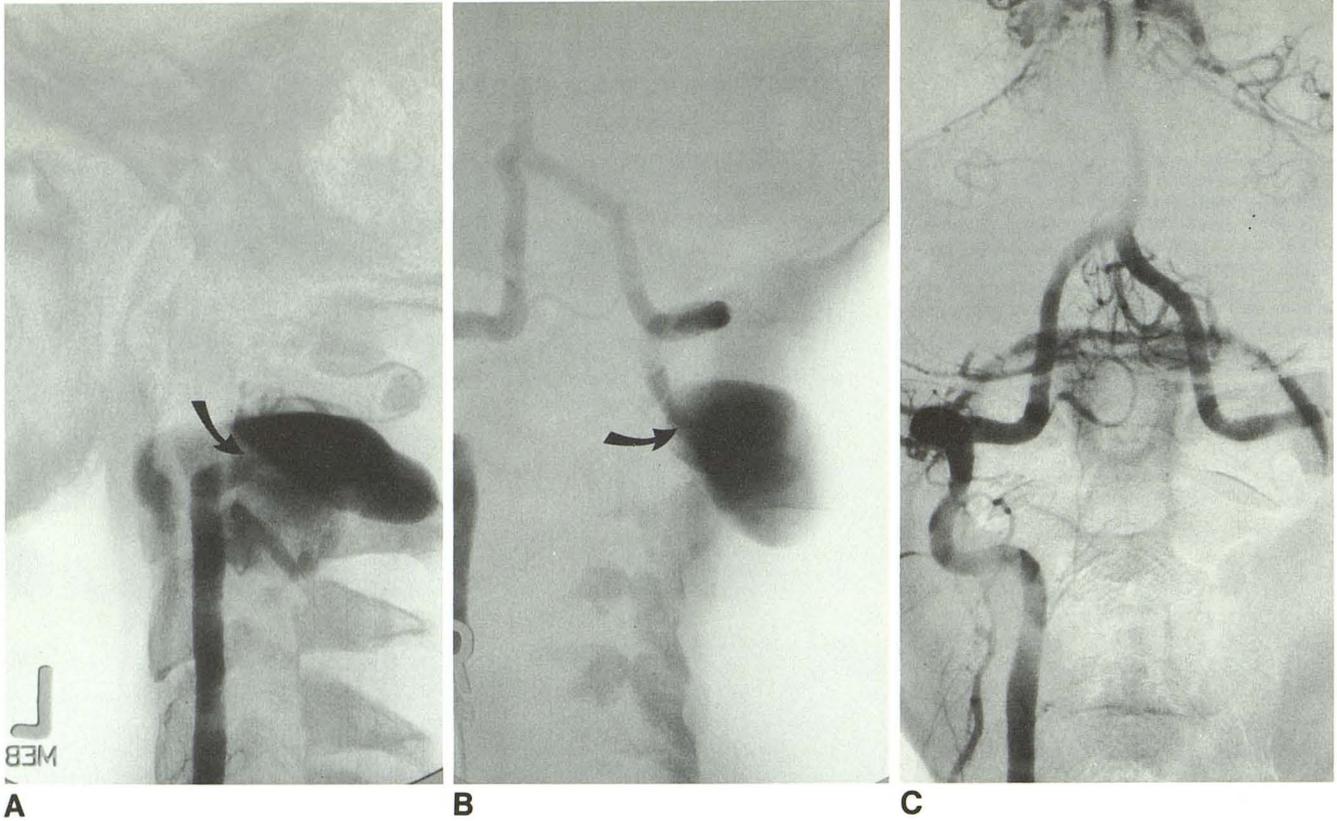


Fig. 4.—Case 17: 21-year-old man with penetrating neck injury.
 A, Left vertebral artery injection, lateral view, shows vertebral artery fistula (arrow) draining into pseudoaneurysm.
 B, Right vertebral artery injection, anteroposterior view, shows retrograde steal down distal left vertebral artery into same pseudoaneurysm (arrow). Higher level of entry into pseudoaneurysm is consistent with vertebral transection.
 C, Right vertebral artery injection, anteroposterior view, after balloon embolization. Both contralateral and ipsilateral embolization approaches have been used to effect complete fistula closure.

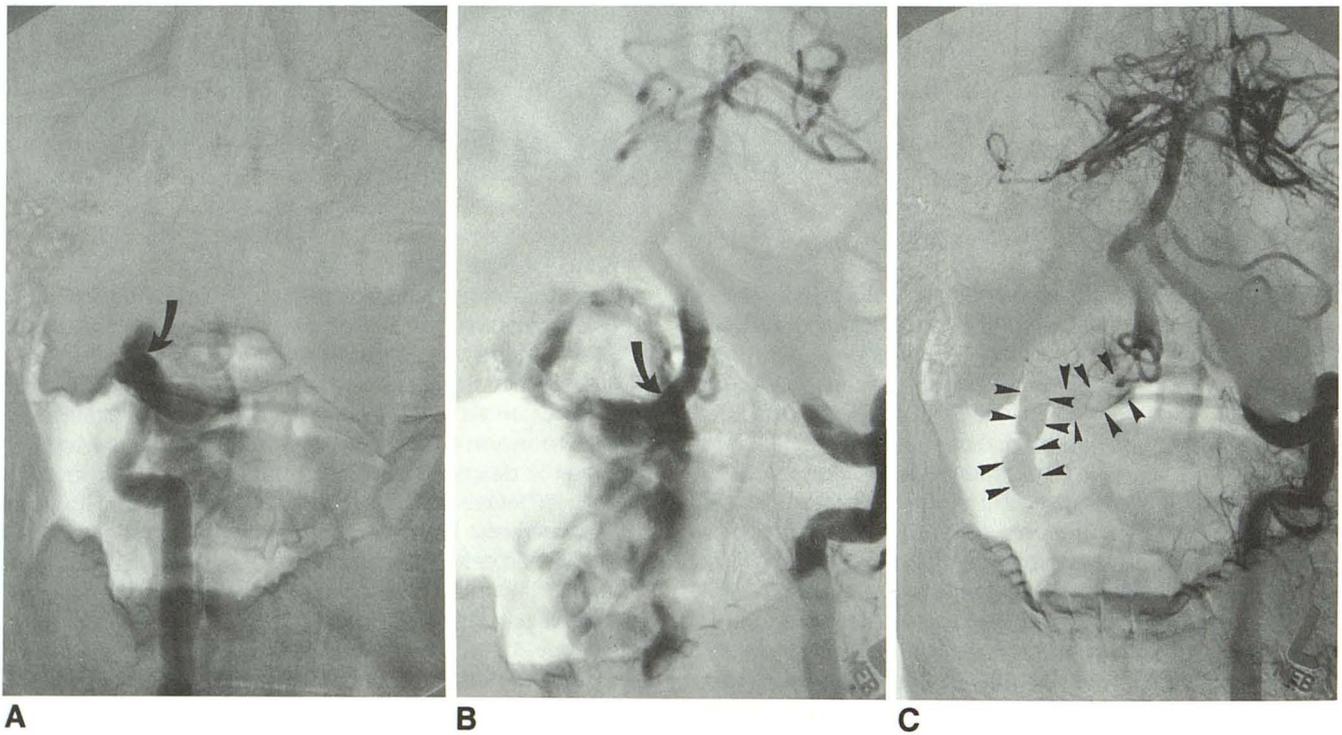


Fig. 5.—Case 10: 20-year-old man with blunt trauma.
 A, Right vertebral artery injection, anteroposterior view, shows site of fistula (arrow) at C1.
 B, Left vertebral artery injection, anteroposterior projection, shows transection (arrow) and drainage into pseudoaneurysm.
 C, Same vessel, after embolization. By using both contralateral and ipsilateral approaches, balloons (arrowheads) have been positioned in both proximal and distal vertebral segments as well as within fistula, resulting in complete cure and preservation of posterior inferior cerebellar artery.

Patients in our series with traumatic fistulas without transection were all treated with transvascular embolization with preservation of flow in four of six involved vertebral arteries.

Vertebral artery transection often presents a diagnostic and therapeutic challenge. Because the cause is often trauma (100% in our series), presentation is generally acute, with external hemorrhage or expanding pseudoaneurysms. The retracted severed ends of the vertebral artery make preservation of flow difficult or impossible. In the six patients in our series with transection, vertebral patency could be preserved only in two. The retracted ends of the vertebral artery and associated pseudoaneurysm often require trapping procedures to achieve fistula closure; these were performed in four patients. Great care must be taken to ensure that the fistula is closed before or coincident with the closure of the vertebral artery. If the proximal vertebral artery is closed before the fistula is obliterated, then increased steal from the distal vertebral artery into the fistula could compromise cerebral circulation. Therefore, in complete vertebral transection where preservation of the artery is not technically possible, the distal vertebral artery should be occluded first. Because of the retraction of the severed ends of the vertebral artery, retrograde flow in the distal vertebral segment, and turbulent flow within the fistula, it is often technically difficult to reach the distal vertebral artery across the fistula site from the ipsilateral approach. It is sometimes necessary to approach the distal vertebral segment from the contralateral vertebral artery to perform a trapping procedure involving the fistula. This maneuver must be undertaken with extreme caution and carries several potential hazards. Occlusion of the contralateral vertebral artery increases the steal from the posterior fossa circulation and may cause neurologic deficits if prolonged. The angle between the two distal vertebral arteries is often acute and negotiation may be difficult. Often, a sharp curve in the distal 2-French catheter is necessary to execute this maneuver. Because of the tortuosity of the distal vertebral arteries, coaxial detachment is impossible. Traction detachment is necessary when the balloon is positioned in the distal vertebral segment above the fistula, and it is important that the final balloon position be below the origin of the PICA. The only complication in our series (in one of our earliest cases) followed occlusion of the PICA, which resulted in the development of a delayed mild Wallenberg syndrome with left face and body hypalgesia and residual clumsiness in the ipsilateral upper extremity.

In addition to the trapping of both the proximal and distal ends of the involved vertebral artery, balloons are placed within the fistula itself to prevent fistula patency from potential collaterals. Both silicone and latex balloons are adequate for occlusion; however, only silicone balloons were used in our series.

In conclusion, transvascular balloon embolization is currently the method of choice in the treatment of vertebral fistulas. In patients with spontaneous or traumatic fistulas without transections, it is often possible to preserve vertebral arterial flow. Patients with long-standing fistulas may be at risk to develop neurologic deficits if abruptly occluded; these can be averted with gradual or staged fistula closure. In patients with transection, a trapping procedure is often necessary to achieve complete fistula closure and can be performed from a transvascular route.

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