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Percutaneous Obliteration of a Postoperatively Persistent Vertebral Arteriovenous Fistula

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Traumatic vertebral arteriovenous fistulae (AVFs) are relatively uncommon lesions that often follow penetrating wounds of the neck or iatrogenic arterial trauma [1–7]. Such posttraumatic single-hole fistulae differ from congenital arteriovenous malformations, which are composed of multiple communications between the cervical arteries and an abnormal vascular nidus [8]. True single-hole vertebral fistulae may be treated surgically by ligation of the abnormal communication [9, 10] or by transcatheter embolization (arterial or venous approach), often with preservation of flow in the vertebral artery [11–15]. Surgical ligation or trapping of the vertebral artery segment supplying the fistula without obliteration of the fistulous communication is ineffective, since collateral flow from other cervical arteries will continue to supply the fistula, and the vertebral artery is unnecessarily sacrificed [2, 6, 8, 16–19]. Also, once direct intravascular access to the fistula site has been obliterated by unsuccessful ligation, there may be no remaining arterial route available to attempt cure by embolization.

We present here a case of a posttraumatic vertebral AVF previously treated (unsuccessfully) by ligation of the vertebral artery proximal and distal to the site of the fistula. Unusual transspinal interarterial collateral blood flow from the nonaffected left vertebral artery toward the affected right vertebral artery resulted in a neurologic disturbance related to a vertebralbasilar “steal syndrome” [20, 21]. In addition to the absence of an adequate intraarterial route for embolization, no venous route was available, since the right vertebral vein was thrombosed at the time of the initial injury. Total occlusion of the fistula hole was, therefore, accomplished by direct puncture of the blind segment of the affected vertebral artery (between the previously placed surgical ligatures) with introduction of Gianturco steel coils at the fistula site [22–24]. All neurologic symptoms and signs disappeared after the fistula was obliterated. We believe this case to be unique, although

our technique should have broader applicability in other cases of postoperative persistence of single-hole AVFs.

Case Report

A 52-year-old woman presented with neurologic symptoms referable to the posterior circulation, and right-sided tinnitus. Twelve years prior to admission she was hospitalized for 14 weeks with septic complications and right arm paresis following a penetrating neck wound. A right cervical radiculopathy was also present for several months and then subsided along with the paresis. Four years prior to admission a right neck bruit was discovered, but no further work-up was done. Two years prior to admission she developed dizziness and unsteady gait with a burning sensation in the tongue and lower face bilaterally. Angiography at this time revealed an AVF between the right vertebral artery (RVA) and the vertebral venous plexus. The proximal and distal (C1 level) RVA was surgically ligated in two separate operations within 10 months. Each time, her symptoms abated temporarily for 3–4 months, although the bruit never was eliminated. Because of recurrent neurologic symptoms, the patient was admitted to our hospital for evaluation.

Neurologic examination revealed prominent right lateral gaze nystagmus. Gait performance was normal, except for tandem walk. Reflexes were 2+/4 and symmetrical. The Nylan-Barany maneuver elicited immediate persistent nystagmus with complaint of increased dizziness when the head was in the down position. A prominent bruit was heard throughout the right side of the neck.

Superselective angiography revealed complete occlusion of the RVA at its origin (the point of prior surgical ligation). Collateral branches of the thyrocervical and costocervical arterial trunks, and the occipital branch of the right common carotid artery reconstituted a blind segment of the RVA within the foramina transversaria extending from C3 to C6. Subselective thyrocervical trunk injections (Fig. 1) revealed a single-hole AVF between the C5 level of the blind RVA segment and an intervertebral foraminal vein, which then filled the intraspinal longitudinal epidural vertebral venous plexus [25]. Owing to prior traumatic thrombosis of the right vertebral vein, venous drainage was cephalad through the jugular bulb and then down the jugular vein (Fig. 1). Selective injection of the left vertebral artery

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(LVA) (Fig. 2) demonstrated filling of the blind RVA sac (and the fistula) by collateral flow through transspinal interarterial anastomotic branches. A vertebral to vertebral steal was thus documented by a route bypassing the prior ligation of the distal RVA at the C1 level.

In view of clinical evidence that the persistent fistula was related to the patient's neurologic complaints and signs, particulate embolization or transarterial fluid embolization was considered but rejected as untenable, since many relatively small collateral arteries communicated with the larger blind RVA sac. Such materials would lodge within the collateral circulation and not thrombose the sac or the fistula. An alternative approach was chosen. The patient was brought to the angiographic suite and neuroleptic analgesia was employed for sedation. The right carotid artery was catheterized and its relative position noted fluoroscopically. The patient was then rotated so that the carotid lay toward the left of the proposed direct percutaneous approach to the blind RVA segment. The angiographic catheter was then placed within the ascending cervical branch of the right thyrocervical trunk, which collateralized to the fistula (Fig. 3A). Check injections within this artery served to fluoroscopically monitor the exact location of the fistula hole and allowed triangulation of a direct percutaneous approach to the fistula. After local anesthesia, a 19-gauge Seldinger needle was employed for direct puncture of the blind RVA segment proximal to the fistula; however, the initial puncture and check contrast injection revealed filling of the carotid artery. The needle was withdrawn and the neck compressed for 10 min. A more lateral repuncture and a check injection revealed the blind segment

of the vertebral artery and the fistula, with subsequent filling of its venous drainage. At this point, two 3×4 -mm Gianturco steel coils were introduced through the needle and were lodged within the inferolateral portion of the blind RVA sac, near but not at the fistula hole (Fig. 3B, compare with Fig. 3A). The needle was withdrawn and the neck compressed. Angiography via the right thyrocervical trunk revealed the laterally placed coils obliterating the inferolateral portion of the blind sac but allowing continued filling of the more medially located fistula (Fig. 3B). The blind segment was again repunctured, this time more medially under direct fluoroscopic control; the needle was guided by the initial deposition of coils. The sac was punctured directly at the site of the fistula, and two additional 3×4 -mm Gianturco coils were introduced, resulting in obliteration of the fistula hole. Check angiography, performed by way of the right thyrocervical trunk collateral route, revealed slow filling of the blind RVA sac with late stasis and no filling of the fistula or the venous drainage (Fig. 4). A muscular collateral vessel from this trunk was now noted to fill the distal RVA. This anastomotic route was visible in retrospect on preocclusion films but was clearly more dilated after occlusion. The LVA was again injected after occlusion, and there was no supply to the fistula. Immediately after the procedure, the patient noted absence of tinnitus. Follow-up neurologic examination at 24 hr revealed disappearance of nystagmus and the positive Nylan-Barany maneuver; 5 months later there were no neurologic symptoms and no audible bruit. Right lower cervical pain was present for 2 weeks after embolization, but resolved completely thereafter.

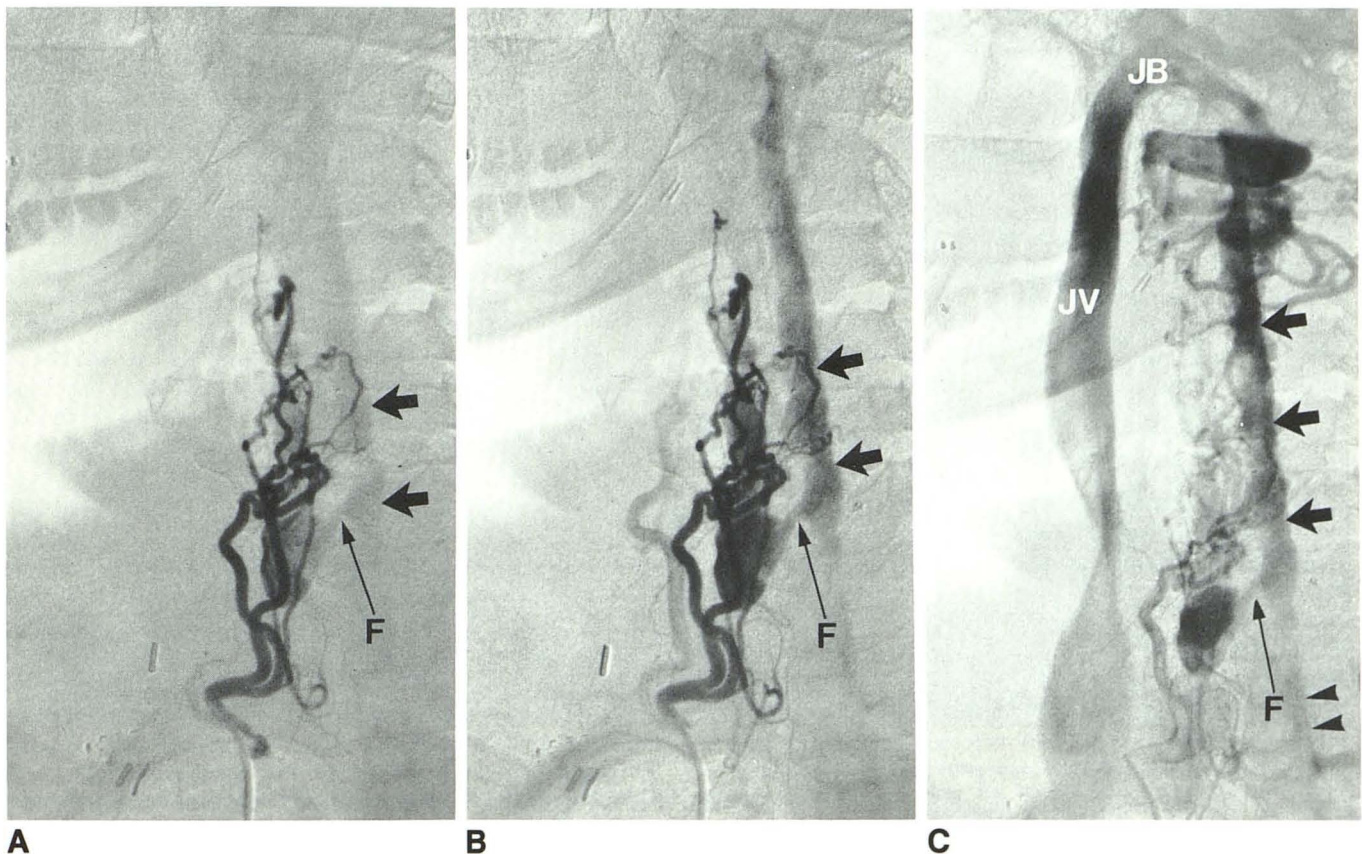


Fig. 1.—A–C, Oblique projection of right thyrocervical artery branch injection; early arterial phase (A) and later phases (B and C). Branches of trunk collateralize to a dilated, blind, mid-cervical vertebral artery segment that fills fistulous communication with foraminal vein (F) and intraspinal longitudinal venous sinus (arrows). Prior postthrombotic obliteration of lower right vertebral vein (arrowheads, C) resulted in preferential cephalad venous flow toward mastoid emissary system, jugular bulb (JB), and jugular vein (JV).

Discussion

Operative ligation of vessels leading to single-hole AVFs and malformations may result in transient or persistent decrease in direct arterial flow to the abnormal communication, but rarely results in cure. The persistently open fistulous hole will attract collateral flow from any regional collateral route [2, 5, 6, 16–18]. The arterial ligatures prevent access to the fistula for interventional techniques, such as detachable balloon occlusion therapy; techniques that have been shown to be effective in curing these lesions while preserving normal arterial flow. Collateral vessels that develop after failure of operative intervention are small vessels draining toward large arteries and/or AV communications. Detachable balloons cannot be navigated through such communications and, therefore, other embolic materials have been employed with limited success. In our case, the absence of a suitable arterial or venous route for transcatheter embolization prompted us to attempt direct introduction of materials for the obliteration of the fistula hole and the portion of the blind vertebral arterial segment near the hole. We believed this to be a safe and

effective approach, particularly in view of difficulties in surgical reexploration of a previously traumatized and operated site, and that preservation of flow within the RVA could not be accomplished because it had been occluded by prior surgical ligations. Neurologic risks were minimal, since the RVA segment feeding the fistula did not communicate with the distal vertebral artery, which had been previously ligated at the C1 level. Success in this case depended on the introduction of a permanent embolic material exactly at the fistula site, since permanent closure of the hole would preclude the possibility that collateral vessels coursing toward the blind arterial sac would fill the fistula. The fistula hole lay between the vertebral artery and the perivertebral venous plexus, which in turn drained through an intervertebral foraminal vein to the longitudinally communicating intraspinal epidural venous sinus [25].

Gianturco coils were chosen as the embolic agent because they could be easily introduced through a Seldinger needle and would not pass through to the venous side of the fistula. Detachable balloons would have been equally effective in obstructing the fistula. We felt that these would be difficult to

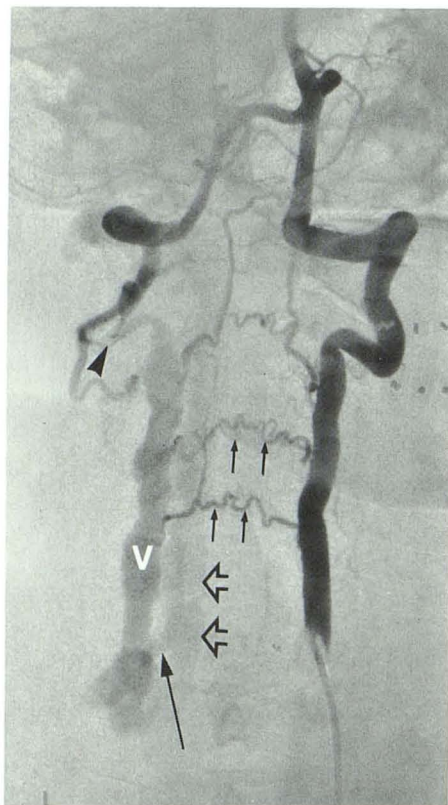


Fig. 2.—Left vertebral arteriogram, anteroposterior view with head turned toward left. Note transspinal segmental vertebral anastomoses (double arrows) at four cervical levels, providing flow to blind right vertebral arterial sac (V). Intraspinal anterior longitudinal venous sinus then fills (open arrows) from fistula hole via intervertebral foraminal (uncinate) vein at C6 level (long arrow). Note, also, collateral filling of distal right vertebral artery down to level of surgical clip (arrowhead) by flow across vertebrobasilar junction.

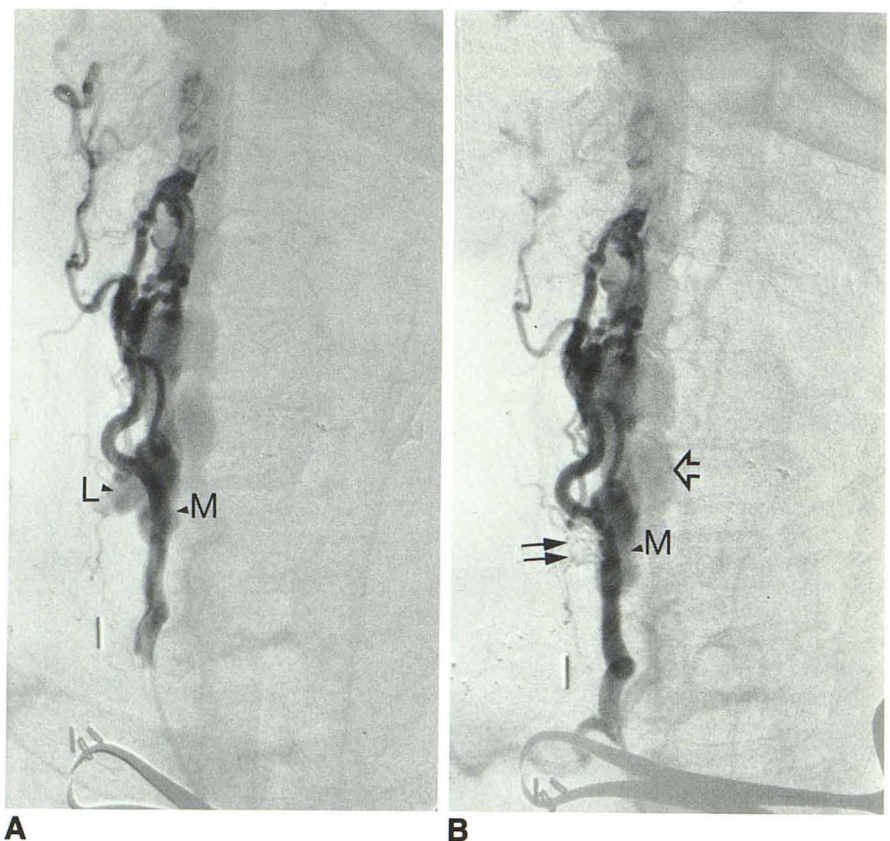
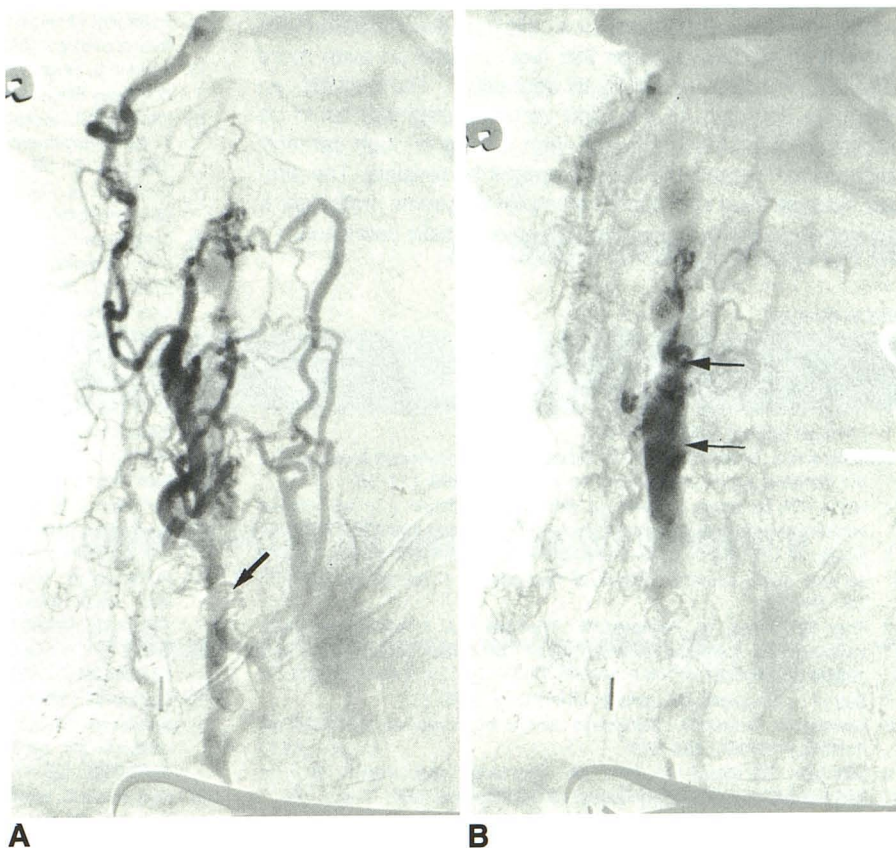


Fig. 3.—A, Selective injection of ascending cervical branch of right thyrocervical trunk performed at time of fistula occlusion (compare with B). Note segmented caudal portion of blind vertebral arterial sac; a broad medial portion (M) and a lateral locule (L).

B, Same injection as in (A), after direct percutaneous deposition of two 3 × 4-mm Gianturco coils. Lateral locule of blind vertebral segment is obliterated by coils (arrows); however, medial segment (M) still fills fistula and venous sinus (open arrow).

Fig. 4.—A and B, Right thyrocervical trunk angiogram performed after deposition of four Gianturco coils; intermediate (A) and delayed phases (B). Two additional coils are noted (arrow, A) inferomedial to previously placed coils (compare with Fig. 3). Note slowed filling of blind right vertebral arterial sac with flow stasis in the late phase (arrows, B). Fistula no longer fills and longitudinal venous sinus no longer opacifies (compare with Fig. 1).



introduce through a nonvascular tract, although balloons have been introduced within feeding arteries through a 16-gauge needle [26]. While liquid embolic agents have been employed percutaneously for treatment of AVFs [17], these agents are difficult to deposit accurately and may pass through a large fistula hole.

The coils were deposited in the arterial sac, not within the foraminal vein (the venous side of the fistula), since we felt that distension of the vein in the foramen might cause radiculopathy, and the artery had already been surgically sacrificed. A short-lived radicular pain did develop anyway, perhaps because of root irritation by the intraarterial coils. Cervical radiculopathy has been reported to occur in association with vertebral AVFs, presumably because of root compression by enlarged foraminal veins [4]. It was particularly gratifying that the patient's vertebrobasilar symptoms subsided after closure of the fistula. Neurologic manifestations of vertebral AVFs are infrequent but well reported [3, 26–29]. As summarized by Reizine et al. [26], nonradicular symptoms may be categorized as focal hemispheric signs due to steal from adjacent circulations, brainstem and cerebellar signs due either to vertebrobasilar steal or increased venous pressure within posterior fossa veins, or focal spinal signs. The latter may be due to medullary compression by dilated veins [27], or medullary or cord ischemia related to either increased venous pressure or steal from the anterior spinal arterial system [29].

When direct vascular access for detachable balloon therapy is available it should be the procedure of choice, since selec-

tive elimination of the AVF with *preservation of normal arterial flow* is the preferable goal. However, we have shown that a direct approach is potentially safe and effective in cases in which no intraarterial route remains after surgical ligation. The technique may also be appropriate if prior embolization has obstructed feeding arteries but has failed to result in cure of a fistula. Our case also illustrates the well-known observation that proximal and distal ligation of the vertebral artery without fistula closure is ineffective because of the extensive anastomotic network between the external carotid, subclavian, and contralateral vertebral arteries and the affected vertebral circulation. Such collaterals most frequently arise from the ipsilateral ascending cervical artery of the thyrocervical trunk, since embryologically this vessel and the vertebral artery both originate as longitudinal anastomoses between the first six pairs of embryonic dorsal intersegmental arteries, which are paired branches of the primitive aortic arches [8, 30].

We believe that our case is the first one reported of a direct percutaneous puncture approach for cure of a vertebral AVF. This percutaneous approach may be less effective in cases of true AV malformations, since a large number of feeding arteries and a large vascular nidus exist, and a cure is less readily accomplished by the placement or local deposition of embolic materials. Although we employed steel occlusion coils alone, percutaneous approaches might incorporate other materials such as isobutyl 2-cyanoacrylate (bucrylate); although as noted, fluid embolic agents generally entail a higher degree of risk of embolization of normal tissue territories and the

venous drainage. Steel occlusion coils may be pretreated with thrombin [31], and perhaps this would make an even more effective embolus for localized deposition. Alternatively, an operative approach to the blind vertebral segment could be employed, and direct puncture with a large enough catheter to introduce detachable balloons might be feasible. The simpler nonoperative approach we employed seems preferable if the vertebral artery has already been surgically sacrificed.

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