Case Reports

Arteriovenous Fistula of Vertebral Vessels in the Neck*

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The occurrence of a fistula between the vertebral artery and vein in the neck is extremely rare. Only 20 cases have been found in a review of the literature. All of these fistulae occurred following some type of trauma; the majority were the result of war injuries. Two developed subsequent to blunt injury of the neck; 1, 2 resulted from a percutaneous arterial puncture. 3 Most of the cases were reported either prior to or immediately following World War II, hence there are only 2 cases verified by angiography reported in the literature. 4 It is conceivable that some of the earlier cases may not have been true arteriovenous fistulae but traumatic, false arterial aneurysms. It is the purpose of this report to present 2 cases of arteriovenous fistula of the vertebral artery. Both were verified angiographically and successfully obliterated surgically.

Case Reports

Case 1. U.H. #962663, L.D., a 35-year-old man, had 2 episodes of hemiparesis, each affecting a different side. Both attacks lasted about 3 hours and subsided spontaneously. The 2nd episode occurred on July 27, 1960. He had a left hemiparesis and hyperreflexia and a mild left central facial palsy. His blood pressure was 154/104 mm. of Hg. Bilateral carotid angiography revealed no abnormality. A right subclavian angiogram failed to demonstrate the vertebral artery. A left percutaneous vertebral angiogram revealed excellent filling of the left as well as the right vertebral artery. The vertebral artery was punctured at the C5–C6 level. It was felt that the patient had cerebrovascular disease and that his two episodes of alternate hemiparesis were the result of transient cerebral ischemia. He was given a vasodilator and a tranquilizer and sent home.

The patient was first seen at the University Hospitals in January 1961. At this time he complained of a noise in his neck. He claimed that this noise had been present since the angiographic examination referred to previously. This was described as a "whistling noise" occurring at the same rhythm as his heart beat. It would disappear if he turned his head to the right or if he hyperextended his neck.

Examination. There was a loud bruit with systolic and diastolic components heard in his neck bilaterally, the left being more prominent. There was no mass or thrill. Turning of the head to the right, hyperextension,

digital compression of the carotid artery and almost any pressure applied to the left side of his neck could obliterate the bruit. His neurological findings were normal.

Bilateral carotid angiography was repeated without demonstration of any vascular abnormality. Subclavian angiography on the right revealed filling of the vertebral artery but there appeared a partial obstruction at the take-off of the right vertebral from the subclavian artery. This was thought to be the result of an atheromatous plaque. A left subclavian angiogram demonstrated an arteriovenous fistulous lesion of the vertebral artery at the C5–C6 level (Fig. 1).

Operation. A procedure planned to obliterate the fistula was done in March 1961 under general anesthesia and hypothermia of 86°F. Hypothermia was employed because of 1) history of cerebrovascular ischemia; 2) question of angiographic evidence of a plaque at the take-off of the right vertebral artery, and 3) anticipation that temporary occlusion of the left vertebral artery might be necessary.

An incision in the skin was made parallel to the
Fig. 2. Case 1. Postoperative vertebral angiogram, showing complete obliteration of the fistula.

The anterior border of the left sternocleidomastoid muscle. The first portion of the vertebral artery was dissected proximal to the subclavian artery. The intraforaminal portion of the vertebral artery was exposed by an anterior approach. The digitations of the deep cervical muscles were separated from the 6th, 5th and 4th transverse processes and retracted laterally, and the anterior walls of the transverse foramina were removed. This brought into view the large and pulsating venous channels enveloping the vertebral artery in the canal. In fact, these venous channels seemed to swell up with arterial blood as the foramina were being opened. It was impossible to locate the fistulous opening. Gradually, the numerous venous channels running anterior, medial and lateral to the vertebral artery from C6 up to the C4 level were dissected off the artery and coagulated. Eventually the vertebral artery was isolated and strips of Gelfoam were placed between the artery and the venous channels running posteriorly. Temporary occlusion of the vertebral artery proximal and distal to the fistula was not necessary because there was no serious bleeding. The fistulous opening or openings were never identified. Since the vertebral artery was isolated from the venous channels it was assumed that the fistulous communications were obliterated.

Postoperative course was uneventful. The noise in the neck disappeared. In January 1963, about 22 months after operation, a left vertebral angiogram was repeated using retrograde brachial-artery technique. There was no evidence of the arteriovenous fistula (Fig. 2). The patient has been followed in the clinic. He has not had any further neurological problem.

Case 2. U.H. #978810. C.L., a 14-year-old boy, was brought into the Emergency Room, University Hos-
pitals in October 1961, following a hunting accident in which he was struck in the back of the head and neck by a blast from a 12-gauge shotgun at a distance of about 20 yards.

Examination. He was awake and oriented. Blood pressure was 120/80 mm. of Hg. In the occipital and sub-
occipital region there were numerous perforations of shot. Pertinent neurological findings included paresis of the left upper extremity and impaired sense of position and tactile discrimination in the left arm. In the right upper extremity and over the upper thoracic segments there was hypalgiesia and hypesthesia.

Roentgenograms of the skull and cervical spine showed no fracture. There were multiple pellets in the head. By stereoscopic visualization 2 pellets were seen in the cerebral hemispheres, each just lateral to the midline about 2 in. anterior to the inion. In addition, 1 shot was believed to be inside the spinal canal against the left side of the 1st cervical vertebra. Another shot appeared to be located just left of the odontoid process. 

Course. Following the patient’s admission, his scalp was debrided. He was given tetanus toxoid and anti-
biotics. His neurological deficit improved gradually. Just prior to his discharge in November 1961, he noticed a “soft noise” in his head. The possibility of a vascular complication was entertained but because of some in-
duration and edema which were still present in the sub-
occipital region, it was felt that further investigation for this “soft noise” should be postponed. He was re-
ferred to the Department of Physical Medicine and Rehabilitation for therapy.

On follow-up examinations in February and March 1961, the “soft noise” was louder. There was a distinct audible bruit heard best over the right posterior auricular and cervical region. There was also a small suboccipital subcutaneous mass. A right subclavian angiogram showed an arteriovenous malformation ex-
tending from the occiput down to the level of the 3rd cervical vertebra (Fig. 3). The ascending branches of the right thyrocervical trunk fed into the malformation and were unusually large. The right vertebral artery was well demonstrated and it, too, was involved in the lesion. A right carotid angiogram showed no abnormality. A 2-stage surgical treatment of this lesion was planned.

Operations. In the 1st operation the external carotid artery, the superior thyroid artery and all the branches of the thyrocervical trunk on the right side were ligated and sectioned. This procedure reduced the bruit greatly but did not eliminate it completely.

Two days later, therefore, a 2nd operation was car-
ried out under general anesthesia. The incision was made in the suboccipital region behind the right ear. There was a mass, about 3 cm. in diameter, consisting of large arterial as well as venous channels, embedded in the suboccipital muscles. This was partially removed to gain access to the 3rd portion of the vertebral artery. Extending posteriorly from this portion of the artery was a glomery of vessels about 1 cm. in diameter. These vessels seemed to enter the circular sinuses at the foramen magnum. With further dissection it was possible to ligate these vessels which served as the fistulous open-
ing. Following this, the large angiomatous mass col-
lapsed and it was removed totally. At this time, a par-
tial laminectomy was done at C1, C2 and C3 levels. Intradural inspection did not reveal any further abnor-
mality.
Arteriovenous Fistula of Vertebral Vessels in Neck

Postoperatively the patient did well except for delayed healing of the wound probably caused by the extensive resection of the subcutaneous tissue. The bruit was eliminated entirely.

Follow-up examination in the out-patient clinic showed satisfactory neurological recovery. He has been back to school and is very active. Postoperative angiography was proposed but declined by the patient's parents.

Discussion

Several remarks can be made concerning Case 1. First, it is most probable that the fistula was iatrogenic resulting from the percutaneous puncture of the vertebral artery between the transverse processes of C5 and C6. As far as we can ascertain, this is the second report in the literature of an arteriovenous fistula involving the vertebral artery as a complication of angiography; the first was reported by Olson and his associates. We were as surprised as they in not being able to find in the literature additional reports of such complications. Secondly, the bruit in this case could be obliterated by pressure applied to the anterior aspect of the neck, by turning the head to the right and by hyperextension. In the literature it has been emphasized that the bruit of a fistula involving the vertebral vessels in the neck generally could not be obliterated by digital compression in the neck. It can only be speculated regarding the mechanism whereby the bruit disappeared in our case with these maneuvers as mentioned. It is possible that digital pressure in the neck could have occluded the venous return from the fistulous region to the subclavian, thereby decreasing the arteriovenous shunt. Turning of the head and hyperextension may have produced the same effect. Regardless of the mechanism involved in this instance, it is clear that the disappearance of the bruit with digital pressure in the neck cannot be taken as an indication that the fistula involves the carotid and not the vertebral vessels. Needless to say, angiographic examination is indispensable in the diagnosis and management of such cases.

An anterior approach to the infaeroraminal portion of the vertebral artery was used in Case 1. It is felt that this is a satisfactory approach which requires the least dissection across the transverse process of the cervical spine. The bellies and digitations of the prevertebral muscles are retracted laterally, thus avoiding possible trauma to the brachial plexus. In this operation a self-retaining retractor designed by Cloward for anterior fusion of cervical interbodies is invaluable. The prevertebral area is approached exactly as in an anterior fusion, staying medial to the carotid sheath. Since the serrated edges of the retractor are positioned beneath and limited by the bellies of the longus colli and the longus capitis, excessive traction of the carotid artery would not occur.

The rationale for the use of hypothermia in

Case 1 has already been stated. It is felt, however, hypothermic or hypotensive methods of anesthesia are generally not necessary during surgery. For extracranial arteriovenous fistulae wherein a gradually developing collateral circulation of the brain has already taken place, ligation of the artery proximal and distal to the fistula would not compromise and may, indeed, improve the cerebral circulation by elimination of the “steal.” In support of this is the fact that in the literature no report of neurological deficit following such a surgical procedure could be found. One of the patients reported by Elkin and Harris, with an arteriovenous fistula of the 3rd portion of the vertebral artery on the left, was treated successfully by ligation, in separate stages, of both vertebral arteries followed by total excision of the lesion. It should be remembered, however, that previous experiences with these lesions were limited almost exclusively to young soldiers whose vascular system otherwise presumably was in excellent condition. It should be cautioned further than an intracranial arteriovenous lesion is a completely different situation. Such a lesion has a propensity to demand the blood which otherwise
would go to the brain so that a proximal ligation of the feeding artery might permit the fistula further to rob the brain of part of its blood supply.

Case 2 presented a somewhat different problem in that we did not know the exact site of the fistula nor were we sure about the vessels involved. This was the reason multiple operations were used to obliterate the lesion. Fortunately, it was possible to ligate the fistulous communication between the vertebral artery and the circular sinus, thereby preserving the vertebral circulation. This, of course, is the ideal situation. If this were not possible, ligation of the vertebral artery, both proximal and distal to the fistula, and excision of the lesion should be done. It is important to excise the entire lesion because the vertebral artery has along its course in the neck, especially the intraradicular portion, numerous small radicular and spinal branches which, if left alone, would continue to feed the fistula. If the site of the fistula is known, as in Case 1, our procedure of choice is isolation of the lesion and then packing of this segment of the artery away from the venous channels with some type of hemostatic material. This would avoid ligation of the vertebral artery. If recurrence of the fistula should take place, ligation and excision of the lesion can then be done. It is apparent that each fistulous lesion involving the vertebral artery is different from the next and consequently the treatment instituted must be individualized.

Summary

Two cases of arteriovenous fistula involving the vertebral vessels in the neck have been presented. The rationale and technique of surgical treatment have been discussed together with a brief review of the literature.

References