Neurol Med Chir (Tokyo) 40, 211~215, 2000

Spontaneous Vertebral Arteriovenous Fistula

—Case Report—

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Abstract

A 57-year-old male presented with a rare case of spontaneous vertebral arteriovenous fistula manifesting as radiculopathy of the right arm, subsequently associated with pulsating tinnitus and vascular bruit in the nape. He had a past history of chiropractic-induced vertebrobasilar infarction. Angiography showed a simple and direct fistula between the third segment of the right vertebral artery and the epidural veins at the C-1 level, where the artery runs backward above the arch of the C-1 just proximal to the penetration of the dura. The fistula was successfully obliterated by coil embolization, resulting in rapid improvement of the signs and symptoms. Mechanical compression to the nerve roots by the engorged epidural veins with arterial pressure was considered to be the major cause of radiculopathy. Vertebral artery dissection induced by chiropractic manipulation is most likely responsible for the development of the fistula.

Key words: vertebral arteriovenous fistula, radiculopathy, coil embolization, vertebral artery dissection

Introduction

Vertebral arteriovenous fistula (AVF) is a rare vascular disease characterized by abnormal connections between the extracranial vertebral artery (VA) or its branches and the neighboring veins. Bruit and neck pain are common symptoms related to the fistula. Brain and spinal cord dysfunction may also occur, attributed to blood flow steal, venous hypertension, or mechanical compression of nerve roots and spinal cord.[7,9] Many vertebral AVFs are of traumatic origin, following incidents like penetrating neck injuries or medical interventions such as carotid artery or VA puncture.[7,10] Non-traumatic vertebral AVFs are usually divided into congenital and spontaneous types. The pathogenetic mechanism of spontaneous fistulas is usually unknown, but there may be a relationship with neurofibromatosis (NF).[1,4,6]

We report a case of spontaneous vertebral AVF probably originating from prior dissection of the VA caused by chiropractic neck manipulation.

Received September 13, 1999; Accepted December 1, 1999

Case Report

A 57-year-old male had suffered from diabetes mellitus for 5 years. He had no history of serious head or neck injuries nor familial history of any hereditary diseases including NF. In 1972, he had an attack of vertigo during chiropractic treatment of the neck, followed by muscle weakness in the right extremities. He soon lost consciousness and was taken to a nearby hospital by ambulance. Cerebral infarction was suspected but neither computed tomography (CT) nor magnetic resonance (MR) imaging were available at that time. Angiography was not performed. He regained consciousness, then continued to improve neurologically and returned to his previous job a couple of months later, with slight hemiparesis persisting on the right. He was doing well until 1997, when he started to suffer from gradually progressive muscle weakness in the right arm. He became aware of pulsating tinnitus on the right in February 1998. He went to a physician at a local hospital, where MR imaging showed an abnormality in the upper cervical spinal area. He was referred to our hospital.

On admission, loud vascular bruit was heard over the nape of the neck on the right. No subcutaneous tumor or blemishes were found. Neurological ex-

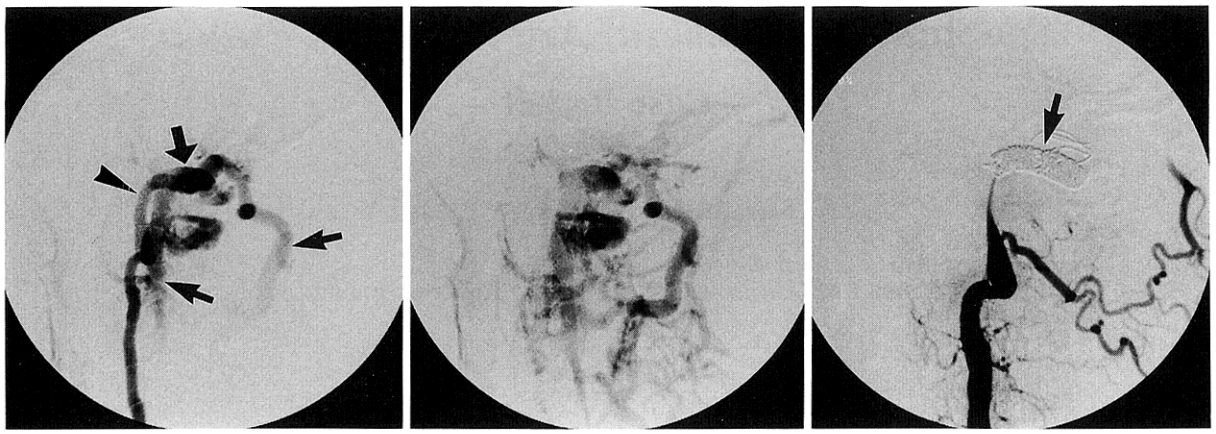

Fig. 1 **Right vertebral angiograms, lateral view, *left* and *center*: showing the fistula (*thick arrow*) located where the vertebral artery (*arrowhead*) runs above the arch of C-1 and connected with epidural venous plexus and suboccipital veins (*arrows*), and *right*: showing occlusion of the vertebral artery and fistula with coils (*arrow*).**

Fig. 2 ***left*: Right internal carotid angiogram, lateral view, showing a retrograde basilar flow via the posterior communicating artery feeding the fistula (*arrow*). *right*: Left vertebral angiogram, anteroposterior view, showing opacification of the fistula (*arrow*) through the retrograde vertebral flow.**

amination revealed 2/5 power in the deltoid and biceps muscles, 3/5 power in the triceps and wrist extension, and 4/5 power in the other muscles in the right arm. Sensory disturbance was also noticed in the right C-5 and C-6 dermatomes. The deep tendon reflexes of biceps, triceps, and brachioradial muscles were diminished on the right, whereas those of the bilateral lower extremities were exaggerated without Babinski's sign.

Angiography revealed an AVF where the right VA runs backward above the arch of C-1, just proximal to the penetration of the dura. The fistula was single and directly connected with the epidural veins which showed remarkable expansion in and around the spinal canal (Fig. 1 *left*, *center*). The left VA and bilateral internal carotid arteries also contributed to the shunt, showing the steal phenomenon (Fig. 2). CT (Fig. 3 *left*, *center*) and CT myelography (Fig. 3 *right*) clearly demonstrated the expanded draining veins, which had compressed the spinal cord and subarachnoid space, especially on the right. Brain MR imaging showed old multiple infarcts in the cerebellum and pons (Fig. 4).

The expanded epidural veins were considered to be responsible for the symptoms of the right arm through compression of the nerve roots and/or hindering the venous return. Thus, intravascular occlusion of the fistula was attempted. A microcatheter was advanced distal to the fistula coaxially through a 6F guiding catheter placed in the right VA. The distal segment of VA was occluded first with detachable and fibered platinum coils to stop the retrograde steal flow from the bilateral carotid arteries and left VA. Then, more coils were positioned in the VA distal to proximal to the fistula, including the orifice and the proximal VA. Angiography confirmed complete obliteration of the fistula (Fig. 1 *right*). The vascular bruit and tinnitus stopped immediately. The patient showed remarkable improvement both in muscle power and sensory disturbance on the day after treatment. He left hospital 2 weeks later. Postembolization MR imaging and CT showed shrinkage of the epidural veins and restoration of normal configuration of the spinal cord and cerebrospinal fluid space (Fig. 5).

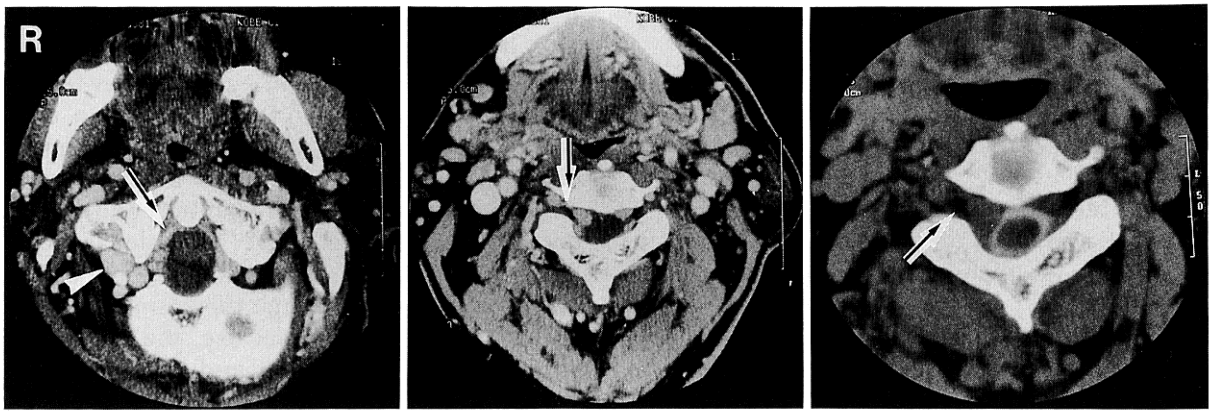

Fig. 3 *left* and *center*: Computed tomography (CT) scans with contrast media showing the fistula (*arrowhead*) above the arch of C-1 with the greatly enlarged epidural venous plexus (*arrow*). *right*: CT myelogram showing compression of the spinal cord and subarachnoid space by an epidural mass (*arrow*).

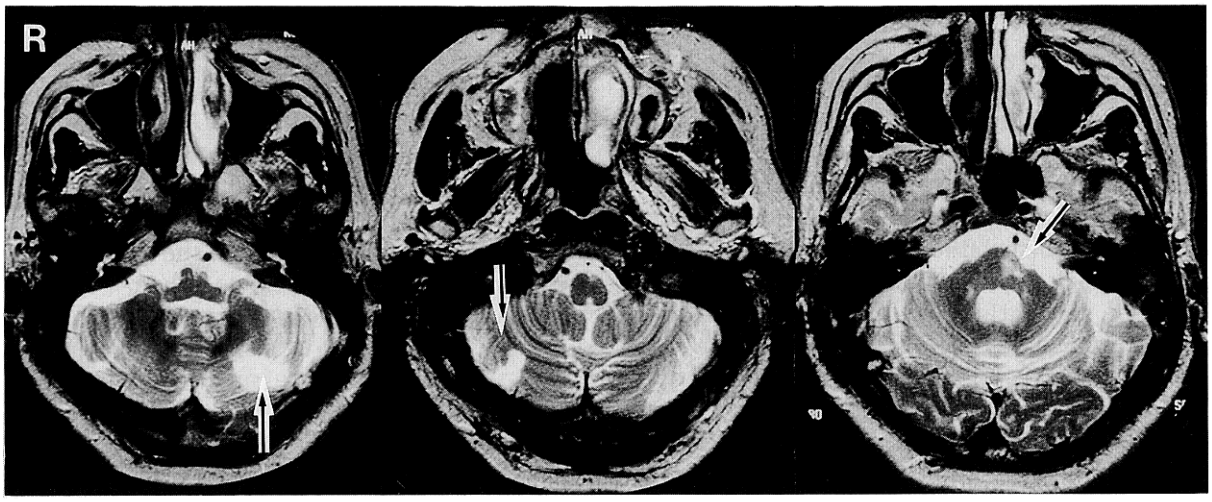

Fig. 4 T_2-weighted magnetic resonance images showing multiple infarcts (*arrow*) in the cerebellum and pons.

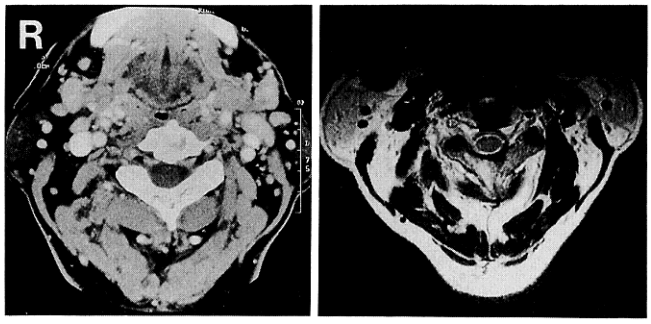

Fig. 5 Computed tomography scan with contrast medium (*left*) and T_2-weighted magnetic resonance image (*right*) after treatment showing shrinkage of the epidural veins and restoration of the normal configuration of the spinal cord and cerebrospinal fluid space.

Discussion

Our patient had radiculopathic symptoms in the right arm. Vertebral AVFs can cause radiculopathy, mostly due to mechanical compression by engorged epidural veins.[15] Nerve root compression by enlarged epidural veins has been confirmed during operation.[8] Impairment of venous drainage from nerve roots is another possible mechanism for radiculopathy.[3]

The pathogenetic mechanism of the fistula in our patient almost certainly was related to the stroke-like incident 25 years before. First of all, this event was very probably an ischemic attack in the vertebrobasilar territory. The symptoms and MR imaging evidence of old multiple infarcts in the brain stem and cerebellum support this assumption. The stroke occurred during chiropractic neck manipulation. At that time, he was as young as 30 years old with no risk factors for stroke. Even 25 years later, angiography showed very few atherosclerotic findings in his cerebral vessels. Considering these facts and the later development of the fistula, the incident 25 years ago was probably dissection of the VA caused by the chiropractic maneuver.

Chiropractic neck manipulation has been established as a cause for vertebrobasilar strokes. Study of 36 patients who sustained strokes during or after chiropractic neck manipulation showed a very high incidence of angiographical or autopsy findings, suggesting arterial dissection such as string sign, pseudoaneurysm, vessel occlusion, stenosis, and intramural hemorrhage.[6] Most of these findings were recognized at the third portion of the VA, where mechanical stretching and compression caused by neck rotation are maximum.[2,6]

Many non-traumatic, spontaneous vertebral AVFs are of unknown pathogenesis, but NF is involved in some cases.[1,4,5,12] Vascular changes such as aneurysmal dilatations, occlusions, and stenosis are well known to occur in patients with NF. Review of 16 cases of AVF associated with NF showed most were vertebral AVFs at the C1-2 levels.[1] Vertebral AVF associated with NF is caused by rupture of aneurysms or aneurysmal dilatations, which developed partly due to the pathological processes involved in NF, and partly due to the mechanical stress accompanying neck rotation. Angiography has provided evidence of a VA aneurysm at the third portion in a patient with NF, which later ruptured and an AVF developed at the same location.[14] These reports and the past history of our patient suggest that pseudoaneurysm or aneurysmal dilatation developed at the chiropractic-induced dissection at the third segment of the right VA, enlarged progressively, and finally ruptured into the surrounding epidural veins, resulting in an AVF.

Prior to development of endovascular techniques, various surgical methods were used to obliterate vertebral AVFs. However, surgical treatment was often unsatisfactory and difficult because of the many arterialized venous channels and direct inaccessibility of the fistulas.[9,11,13] Endovascular treatment is now the simplest, most reliable method for occluding AVFs. Although there are number of embolic materials in clinical use, the detachable balloon may be the simplest and quickest method to treat vertebral AVF, since the fistula is usually simple and direct.[3,7] However, obtaining reliable detachable balloon systems is not necessarily easy in Japan. This is the major reason for the use of coils in this case. Obliteration of just the fistula with preservation of the VA could be best, but we intentionally occluded the VA along with the fistula because of the rapid, drastic increase of the cerebral blood flow expected after obliteration of the high flow fistula. The "break-through phenomenon" occurring after abrupt closure of fistulas may cause new neurological symptoms.[7] Another concern was persistence of the retrograde steal, if the proximal side of the VA was accidentally occluded first. The additional embolization needed in such a case would require access through the contralateral VA, which is not always possible. Thus, we decided to occlude the VA from distal to proximal to the fistula.

References

1) Anegawa S, Hayashi T, Torigoe R, Iwaisako K, Sakae N, Ogasawara T, Utsunomiya H: [Symptomatic arteriovenous fistula in a patient with neurofibromatosis type I]. *No Shinkei Geka* 25: 373-378, 1997 (Jpn, with Eng abstract)
2) Barton JW, Margolis MR: Rotational obstruction of the vertebral artery at the atlantoaxial joint. *Neuroradiology* 9: 117-120, 1975
3) Beaujeux RL, Reizine DC, Casasco A, Aymard A, Rufenacht D, Khayata MH, Riche MC, Merland JJ: Endovascular treatment of vertebral arteriovenous fistula. *Radiology* 183: 361-367, 1992
4) Cluzel P, Pierot L, Leung A, Gaston A, Kieffer E, Chiras J: Vertebral arteriovenous fistulae in neurofibromatosis: report of two cases and review of literature. *Neuroradiology* 36: 321-325, 1994
5) Deans WR, Bloch S, Leibrock L, Berman BM, Skultety FM: Arteriovenous fistula in patients with neurofibromatosis. *Radiology* 144: 103-107, 1982
6) Frisoni GB, Anzola GP: Vertebrobasilar ischemia after neck motion. *Stroke* 22: 1452-1460, 1991
7) Halbach VV, Higashida RT, Hieshima GB: Treatment of vertebral arteriovenous fistulas. *AJR Am J Roentgenol* 150: 405-412, 1988

8) Kohno M, Takahashi H, Ide K, Ishijima B, Yamada K, Nemoto S: A cervical dural arteriovenous fistula in a patient presenting with radiculopathy. Case report. *J Neurosurg* 84: 119-123, 1996
9) Nagashima C, Iwasaki T, Kawanuma S, Sakaguchi A, Kamisasa A, Suzuki K: Traumatic arteriovenous fistula of the vertebral artery with spinal cord symptoms: case report. *J Neurosurg* 46: 681-687, 1977
10) Olson RW, Hillier LB Jr, Svien HJ: Arteriovenous fistula: a complication of vertebral angiography; report of a case. *J Neurosurg* 20: 73, 1963
11) Reizine D, Laouiti M, Guimaraens L, Riche MC, Merland JJ: Vertebral arteriovenous fistulas: clinical presentation, angiographical appearance and endovascular treatment—a review of 25 cases. *Ann Radiol (Paris)* 28: 425-438, 1985
12) Schievink WI, Piepgras DG: Cervical vertebral artery aneurysms and arteriovenous fistulae in neurofibromatosis type 1: case reports. *Neurosurgery* 29: 760-765, 1991
13) Schumacker HB, Campbell RL, Heimburger RF: Operative treatment of vertebral arteriovenous fistulas. *J Trauma* 6: 3-19, 1966
14) Ushikoshi S, Goto K, Uda K, Ogata N, Takeno Y: Vertebral arteriovenous fistula that developed in the same place as a previous ruptured aneurysm: a case report. *Surg Neurol* 51: 168-173, 1999
15) Willinsky R, ter Brugge K, Montanera W, Wallace MC, Gentili F: Spinal epidural arteriovenous fistulas: arterial and venous approaches to embolization. *AJNR Am J Neuroradiol* 14: 812-817, 1993

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