SNI
SURGICAL NEUROLOGY INTERNATIONAL
SNI: Neurovascular
OPEN ACCESS
For entire Editorial Board visit : http://www.surgicalneurologyint.com
Editor: James I. Ausman, MD, PhD University of California, Los Angeles, CA, USA

Case Report

Concurrent cervical dural and multiple perimedullary arteriovenous fistulas presenting with subarachnoid hemorrhage: The source of bleeding was invisible at initial angiography

Iku Nambu, Naoyuki Uchiyama, Kouichi Misaki, Masanao Mohri, Mitsutoshi Nakada

Department of Neurosurgery, Graduate School of Medical Science, Kanazawa University, Kanazawa, Ishikawa, Japan

E-mail: *Iku Nambu - namnamnambuiku@med.kanazawa-u.ac.jp; Naoyuki Uchiyama - naoyuki.uchiyama@med.kanazawa-u.ac.jp; Kouichi Misaki - misaki@med.kanazawa-u.ac.jp; Masanao Mohri - mmohri@med.kanazawa-u.ac.jp; Mitsutoshi Nakada - mnakada@med.kanazawa-u.ac.jp
*Corresponding author

Received: 16 June 16 Accepted: 30 November 16 Published: 19 January 17

Abstract

Background: We report the concurrence of a spinal dural arteriovenous fistula (DAVF) and multiple perimedullary arteriovenous fistulas (PAVFs) presenting with subarachnoid hemorrhage (SAH). Moreover, the bleeding site was detected 1 month after onset.

Case Description: A 56-year-old man was admitted to our hospital with an SAH. A DAVF and two PAVFs were detected at the C2 level by two rounds of digital subtraction angiography. The source of bleeding, an aneurysm on the feeding artery of PAVF, was detected at the second angiogram, which was performed 1 month after the onset of SAH. The aneurysm was not demonstrated at initial angiogram because of thrombosis in the aneurysm. The DAVF was interrupted by transarterial embolization, and the two PAVFs were subsequently treated with surgery.

Conclusion: A part of the whole AVFs or the source of bleeding may be invisible in the acute stage just after hemorrhage. Repeated angiography is necessary to diagnose such complex AVFs especially in case of an SAH and treatment should be performed during the subacute stage.

Key Words: Digital subtraction angiography, spinal dural arteriovenous fistula, spinal perimedullary arteriovenous fistula, subarachnoid hemorrhage

Access this article online
Website: www.surgicalneurologyint.com
DOI: 10.4103/2152-7806.198729
Quick Response Code:

INTRODUCTION

Spinal arteriovenous malformations are classified into four types, namely, dural arteriovenous fistulas (DAVF), epidural arteriovenous fistulas, perimedullary arteriovenous fistulas (PAVF), and intramedullary arteriovenous malformations. Here, we report a case of concurrent cervical DAVF and multiple PAVFs, presenting with subarachnoid hemorrhage (SAH), whose source of bleeding was detected 1 month after onset of the SAH. This case provides important observations regarding the diagnosis of such complex AVFs, as well as the appropriate timing of treatment for such cases with SAH.

How to cite this article: Nambu I, Uchiyama N, Misaki K, Mohri M, Nakada M. Concurrent cervical dural and multiple perimedullary arteriovenous fistulas presenting with subarachnoid hemorrhage: The source of bleeding was invisible at initial angiography. Surg Neurol Int 2017;8:2.
http://surgicalneurologyint.com/Concurrent-cervical-dural-and-multiple-perimedullary-arteriovenous-fistulas-presenting-with-subarachnoid-hemorrhage:-The-source-of-bleeding-was-invisible-at-initial-angiography/

CASE REPORT

A 56-year-old man experienced sudden onset of a severe headache, and was brought to our hospital. His Glasgow Coma Scale score was 15, and he had a stiff neck and abducens nerve paralysis.

Computed tomography of the brain revealed an SAH localized mainly at the anterior surface of the brain stem. Left vertebral artery (VA) angiography demonstrated a DAVF and two PAVFs at the C2 level [Figure 1a]. One PAVF was fed by a branch of the anterior spinal artery [Figure 1b]. An aneurysm on the distal side of the feeding artery was observed in the second angiogram that was performed 28 days after the onset of the SAH [Figure 1c]. This aneurysm appeared to be the source of bleeding. Another PAVF was fed by the descending artery from the left VA [Figure 1d], and the DAVF was fed by the C2 radicular artery [Figure 1e]. The PAVFs and DAVF shared the same drainage route into the anterior spinal vein. The patient became drowsy due to vasospasm and hyponatremia a few days after admission. We, therefore, made it a priority to treat the vasospasm and hyponatremia, and then performed the second angiography and surgery at the subacute stage.

Surgery was performed after transarterial embolization (TAE) of the DAVF with a coil [Figure 1f]. A C1 and C2 laminectomy with a suboccipital craniotomy revealed that the aneurysm was located under the C2 posterior nerve root, which was then cut. After gentle rotation of the spinal cord with the dentate ligament, the draining vein and an aneurysm on the distal side of the feeding artery could be observed [Figure 2a and b]. Two fistulous points of the PAVFs were detected with indocyanine green (ICG) videoangiography [Figure 2c]. We then coagulated and dissected the feeding artery of the PAVFs and draining vein. The final ICG videoangiography showed complete occlusion of the DAVF and PAVFs.

The histopathological features of the aneurysm demonstrated a thickened wall that was composed of fibrous tissue [Figure 2d and e]. These findings indicated that this was a true aneurysm.

Postoperative angiography showed complete obliteration of the DAVF and PAVFs. The patient was discharged without any neurological deficits after placement of a ventriculoperitoneal shunt for hydrocephalus.

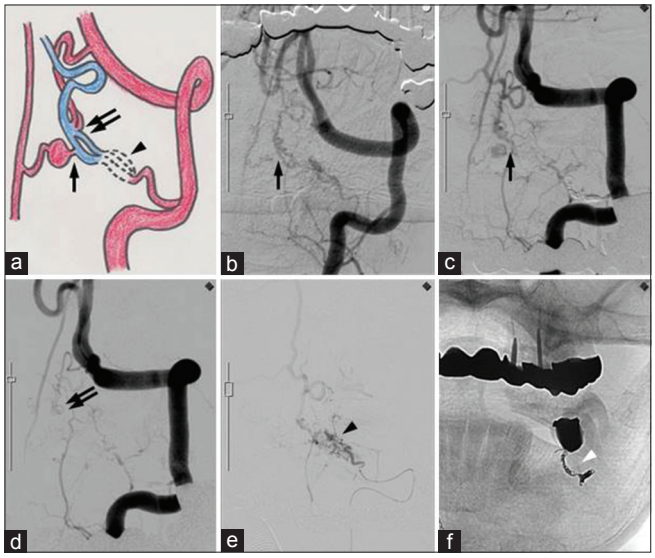

Figure 1: (a) Schematic representing a DAVF (arrowhead) and 2 PAVFs (arrow and double arrow). (b) Left vertebral angiogram showing one PAVF (arrow) that was fed by the branch of the anterior spinal artery. (c) A second angiogram that was performed 28 days after the onset of an SAH revealed an aneurysm on the distal side of the artery feeding the PAVF. (d) Another PAVF (double arrow) that was fed by the descending artery from the left vertebral artery. (e and f) Selective angiogram of the left C2 radicular artery showing a DAVF (arrowhead) and transarterial coil embolization (white arrowhead)

DISCUSSION

Patients with DAVFs or PAVFs that are located in the thoracolumbar region or conus medullaris usually present with gradual worsening of symptoms, such as pain and weakness of the legs.[2] However, DAVFs and PAVFs located at the craniocervical junction or cervical regions are associated with an increased incidence of SAH and cranial nerve dysfunction.[1,5] The concurrence of DAVFs and PAVFs is exceedingly rare, and there are only 11 reports of such cases occurring at the craniocervical junction or cervical region[9,11] and 4 such cases located at the thoracolumbar region.[3,6,8,10] Sato *et al.*[11] described 9 cases of concurrent DAVFs and PAVFs at the craniocervical junction. All of the concurrent DAVFs and PAVFs shared their main drainage route, and all of the cases presented with an SAH. Ruptured arterial aneurysms were detected in 8 of the 9 cases, and ruptured venous ectasia was detected in the remaining cases. In the case

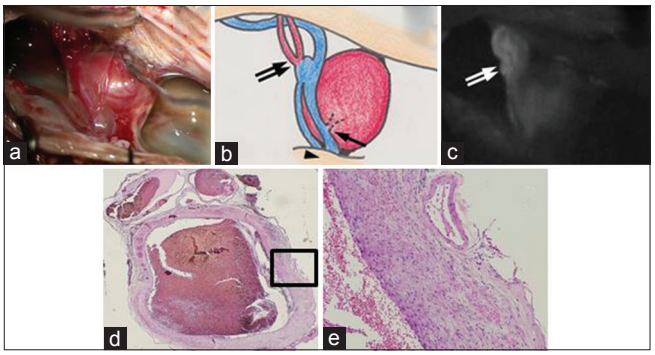

Figure 2: Intraoperative photograph (a), schematic (b), and indocyanine green videoangiography (c) demonstrating the location of 2 PAVFs (arrow and double arrow), a DAVF (arrowhead), an aneurysm on the distal side of the feeding artery, and draining veins of the DAVF and PAVFs. The histopathological features of the aneurysm (d) hematoxylin and eosin, ×20; (e) hematoxylin and eosin, ×100 demonstrating a thickened wall composed of fibrous tissue. The aneurysm retained a luminal structure

described in the present report, the DAVF and PAVFs were located at the C2 level, and bleeding occurred via an arterial aneurysm on the distal side of the artery feeding the PAVF.

The gold standard for diagnosis is DSA, which is useful for detecting feeding arteries, fistulous points, draining veins, arterial aneurysms, and varices. Moreover, important information on normal vessels that should be preserved can be obtained. Some cases of AVFs with a SAH are not identified on the first angiogram. Hayashi *et al*.[4] suggested a number of explanations for this, such as thrombosis of the AVF, a change in circulatory dynamics, compression by a hematoma, or vasospasm of the feeding artery. Maslehaty *et al*.[7] also revealed that approximately 10% of the patients with SAH of unknown etiology at first angiogram had a detectable source of bleeding on a second angiogram. In our case, one possible reason why the aneurysm on the distal side of the feeding artery was not detected in the first DSA was thrombosis of the aneurysm after rupture. Morgalla *et al*.[8] reported a case in which a second spinal fistula, which was not initially apparent, was revealed after closure of the first fistula. This may explain why the shunt volume of the second fistula could opens up after venous drainage was improved. Therefore, preoperative or postoperative repeated angiography is necessary for detecting the structure of the AVFs or the source of bleeding, especially in cases with SAH.

Spinal AVFs are treated with surgery, endovascular embolization, or both. Surgery has low recurrence rates and endovascular embolization is the less invasive approach. In our case, it was possible to occlude the entire shunt system with endovascular embolization using N-butyl cyanoacrylate (NBCA) or Onyx from the feeder of DAVF. However, we did not choose to use these for fear of several complications. If NBCA penetrates to the distal part of the drainer and the fistulous points of PAVFs are not occluded, there is a risk that the feeder aneurysm of PAVF may rupture at high pressure. On the other hand, Onyx can occlude the proximal part of the drainer followed by two fistulas of PAVFs. However, we were concerned that Onyx with the high penetrating ability may penetrate to brainstem supplying perforators, which are invisible on angiogram. We, therefore, decided to perform a surgery in which we could detect whole vascular structures by direct viewing. In the presurgical TAE, to decrease operation blood loss, we used coils instead of glue agents to avoid occluding the intra-dural portion of vessels.

When making a diagnosis, clinicians should be aware of the possibility of the coexistence of two or more spinal cord AVFs that have similar or different characteristics in a single patient, and fistulous points should not be overlooked. Moreover, in cases with SAH, some ruptured points may not be detected at the initial angiogram for several reasons. Therefore, repeated angiography is necessary to diagnose such complex AVFs, and treatment should be performed at the subacute stage, especially in case of SAH.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

REFERENCES

1. Aviv RI, Shad A, Tomlinson G, Niemann D, Teddy PJ, Moluneux AJ, *et al*. Cervical dural arteriovenous fistulae manifesting as subarachnoid hemorrhage: Report of two cases and literature review. AJNR Am J Neuroradiol 2004;25:854-8.
2. Cho WS, Kim KJ, Kwon OK, Kim CH, Kim J, Han MH, *et al*. Clinical features and treatment outcomes of the spinal arteriovenous fistulas and malformation: Clinical article. J Neurosurg Spine 2013;19:207-16.
3. Dam-Hieu P, Mineo JF, Bostan A, Nonent M, Besson G. Concurrent spinal dural and intradural arteriovenous fistulas. Case report. J Neurosurg 2001;95:96-9.
4. Hayashi K, Takahata H, Nakamura M. Two cases of spinal arteriovenous malformation presenting with subarachnoid hemorrhage [in Japanese]. No Shinkei Geka 2004;32:605-11.
5. Kai Y, Hamada J, Morioka M, Yano S, Mizuno T, Kuratsu J, *et al*. Arteriovenous fistulas at the cervicomedullary junction presenting with subarachnoid hemorrhage: Six case reports with special reference to the angiographic pattern of venous drainage. AJNR Am J Neuroradiol 2005;26:1949-54.
6. Krings T, Coenen VA, Weinzierl M, Reinges MH, Mull M, Thron A, *et al*. Spinal dural arteriovenous fistula associated with a spinal perimedullary fistula: Case report. J Neurosurg Spine 2006;4:241-5.
7. Maslehaty H, Petridis AK, Barth H, Mehdorn HM. Diagnostic value of magnetic resonance imaging in perimesencephalin and nonperimesencephalic subarachnoid hemorrhage of unknown origin. J Neurosurg 2011;114:1003-7.
8. Morgalla MH, Ernemann U, Gawlowski J, Deininger M, Bitzer M, Grote EH. Recurrent spinal fistula as result of a rare combination of a perimedullary and peridural spinal fistula: Case report. Surg Neurol 2004;61:347-52.
9. Onda K, Yoshida Y, Watanabe K, Arai H, Okada H, Terada T. High cervical arteriovenous fistulas fed by dural and spinal arteries and draining into a single medullary vein: Report of 3 cases. J Neurosurg Spine 2014;20:256-64.
10. Sasaki O, Yajima N, Ichikawa A, Yamashita S, Nakamura K. Deterioration after surgical treatment of spinal dural arteriovenous fistula associated with spinal perimedullary fistula. Neurol Med Chir 2012;52:516-20.
11. Sato K, Endo T, Niizuma K, Fujimura M, Inoue T, Shimizu H, *et al*. Concurrent dural and perimedullary arteriovenous fistulas at the craniocervical junction: Case series with special reference to angioarchitecture. J Neurosurg 2013;118:451-9.