

Acute Compressive Myelopathy Caused by Spinal Subarachnoid Hemorrhage: A Combined Effect of Asymptomatic Cervical Spondylosis

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Key words

- Arteriovenous fistula
- Cervical spondylosis
- Myelopathy
- Perimedullary
- Spinal subarachnoid hemorrhage

Abbreviations and Acronyms

- AVF:** Arteriovenous fistula
MRI: Magnetic resonance imaging
SAH: Subarachnoid hemorrhage

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Citation: *World Neurosurg.* (2016).

<http://dx.doi.org/10.1016/j.wneu.2016.08.031>

Journal homepage: www.WORLDNEUROSURGERY.org

Available online: www.sciencedirect.com

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INTRODUCTION

Spinal arteriovenous fistulas (AVFs) are rare vascular abnormalities. The prevalence of spinal AVFs is reported as 0.17 in 100,000 persons per year.¹ Among them, AVFs at the craniocervical junction are fed by an anterior spinal artery, radiculomeningeal artery, and radiculopial artery and are drained into a radiculopial vein. AVFs at the craniocervical junction present with subarachnoid hemorrhage (SAH), rather than venous congestive myelopathy caused by those at the thoracolumbar region.²⁻⁶ Patients with SAH by hemorrhagic AVFs usually present with meningeal signs, including headache and nausea, and focal neurologic deficit is found in rare cases. In this article, we report a case of acute compressive cervical myelopathy caused by hemorrhagic AVF at the craniocervical junction. The role of preexisting cervical spondylosis to facilitate symptom onset is discussed.

CLINICAL DESCRIPTION

A 73-year-old woman presented with sudden headache and subsequent left

BACKGROUND: Patients with subarachnoid hemorrhage (SAH) by hemorrhagic arteriovenous fistulas (AVFs) usually presents with meningeal signs, including headache and nausea, and focal neurologic deficit is found in rare cases. In this article, we report a case of acute compressive cervical myelopathy caused by hemorrhagic AVF at the craniocervical junction.

CASE DESCRIPTION: A 73-year-old woman was transferred to our hospital for sudden headache and subsequent left hemiparesis. Head computed tomography scan showed SAH exclusively in the posterior fossa, and catheter angiography revealed a perimedullary arteriovenous fistula at the craniocervical junction as a source of the SAH. Detailed neurologic examination showed the sensory disturbance of bilateral upper extremities and bladder and rectal disturbance, suggesting concurrent cervical myelopathy. Magnetic resonance imaging of the cervical spine showed disk herniation at the C4-5 level, spinal SAH deposition above the C4-5 level, and accompanying myelomalacia. No intramedullary hemorrhage was found.

CONCLUSIONS: Spinal SAH alone rarely causes focal neurologic deficit. However, this case suggests spinal SAH can cause acute compressive myelopathy when complicated with preexisting spinal canal stenosis.

hemiparesis and was transferred to our stroke center. Because she lived by herself, she had remained immobile at home for 2 days until she was found by a visitor. As a result, she had multiple decubiti, and her laboratory data revealed undernutrition. Her medical history included 2 SAHs of unknown origin in her fifties and a recent asymptomatic cervical disk herniation with left-sided predominance accidentally discovered during the examination of an unidentified complaint. She denied any trauma prior to the onset, and there was no sign of head or neck injury. Her Glasgow Coma Scale score was 15, and initial neurologic examination revealed left hemiplegia. Head computed tomography scan showed SAH exclusively in the posterior fossa, including the perimesencephalic, prepontine, and cerebellomedullary cisterns (Figure 1). Catheter angiography was performed to investigate the bleeding source. Right vertebral angiogram demonstrated perimedullary AVF fed by the anterior spinal artery and right radicular artery of

C1 and refluxed into the left petrosal vein through the spinal vein (Figure 2).

Head magnetic resonance imaging (MRI) was performed to investigate the cause of her left hemiplegia, which detected no ischemic stroke. Neurologic examination was repeated in detail, revealing acute cervical myelopathy, including bilateral upper sensory disturbance and bladder and rectal disturbance, in addition to her left hemiplegia. Cervical MRI was obtained and compared with her previous MRI when her cervical disk had been asymptomatic (Figure 3). Her previous MRI showed disk herniation at C4-5, with no evidence of spinal cord hyperintensity on T2-weighted sagittal imaging (Figures 3A and B). Her cervical MRI at symptom onset revealed a deposit of subarachnoid hematoma above the disk herniation and myelomalacia of the corresponding level of the left-sided predominance (Figures 3C and D). Although involvement of nontraumatic hyperextension spinal cord injury was not necessarily ruled out, we ultimately

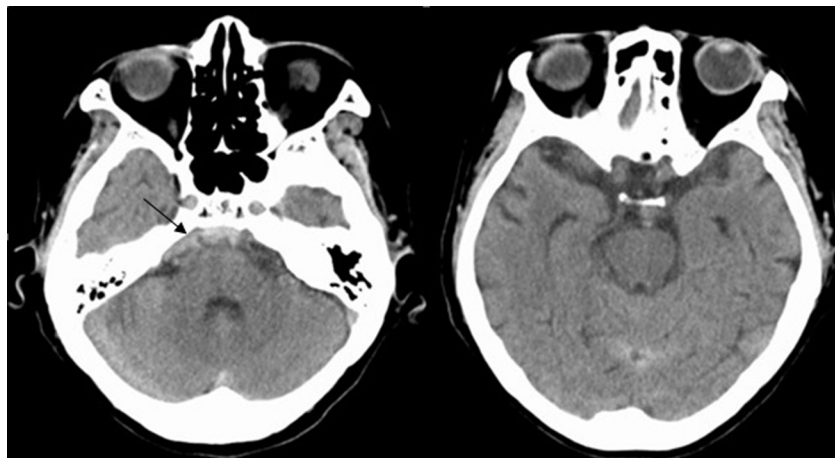


Figure 1. Head computed tomography scan showing subarachnoid hemorrhage predominantly in the posterior cranial fossa.

concluded that subarachnoid hematoma associated with AVF was blocked by the left paramedian disk herniation, raising the focal pressure in the spinal canal and adding acute mass effect on the chronic compression of the spinal cord caused by preexisting asymptomatic disk herniation.

Spinal surgeons in our institution were reluctant to undertake emergent decompressive laminectomy because decompression after 48 hours of onset was considered less effective and because her suboptimal general condition, including

decubiti and undernutrition, may not have tolerated surgical intervention. She also declined our proposal of surgical treatment for her AVF in the chronic stage and chose conservative therapy. Her symptoms partially recovered, and she was discharged to a rehabilitation center with modified Rankin scale score of 3.

DISCUSSION

Perimedullary AVFs are distributed along the entire axis of the spinal cord. Among

them, perimedullary AVFs at the cranio-cervical junction predominantly represent hemorrhagic presentation.²⁻⁶ On the other hand, AVFs at the thoracolumbar region present with progressive myelopathy by spinal venous congestion, which is traditionally known as Foix-Alajouanine syndrome.^{7,8} Previous articles demonstrated that ascending venous drainage from the cranio-cervical junction AVFs was more likely associated with hemorrhagic presentation, which is also true in our case, with venous drainage into the petrosal vein.³

Hemorrhagic perimedullary AVFs present with SAH in most cases, and they less frequently present with intramedullary hemorrhage.^{3,5,6} Intramedullary hemorrhage causes corresponding acute focal neurologic deficits, including motor weakness, sensory disturbance, and bladder and rectal disturbance. On the other hand, Endo et al.⁹ reported that all SAHs caused by cranio-cervical perimedullary AVFs revealed meningeal signs alone, including headache and nausea, and resulted in favorable outcomes. These meningeal signs caused by ruptured spinal vascular malformations have been traditionally known as Coup de poignard of Michon syndrome.¹⁰ The smaller clot volume associated with venous bleeding of AVFs and the diluting effect of longitudinal large subarachnoid spaces of the spine may contribute to these symptoms of lesser severity.¹¹ However, when cerebrospinal fluid obstruction is present at the upper cervical spine (e.g., because of cervical spondylosis), the downward distribution of the SAH is blocked and local spinal subarachnoid pressure increases, resulting in acute compressive myelopathy.¹¹

To our knowledge, this is the first report describing acute myelopathy by combination of spontaneous spinal SAH and spinal spondylosis. The rarity of this phenomenon should be caused by rarity of hemorrhagic spinal vascular malformation.¹ Although incidence of lumbar spondylosis is comparable with that of cervical spondylosis, the combination of spontaneous spinal SAH and lumbar spondylosis has never been reported, presumably because thoracolumbar spinal AVFs seldom bleed.^{3,5,6} Hanakita et al.¹¹ reported a case of acute



Figure 2. Anteroposterior (A) and lateral (B) view of digital subtraction angiography (DSA) reveals perimedullary arteriovenous fistula (PMAVF) with arterial feeders from the anterior spinal artery (ASA) and C1 radicular artery (arrow). The draining vein refluxes into the left petrosal vein (arrowheads).

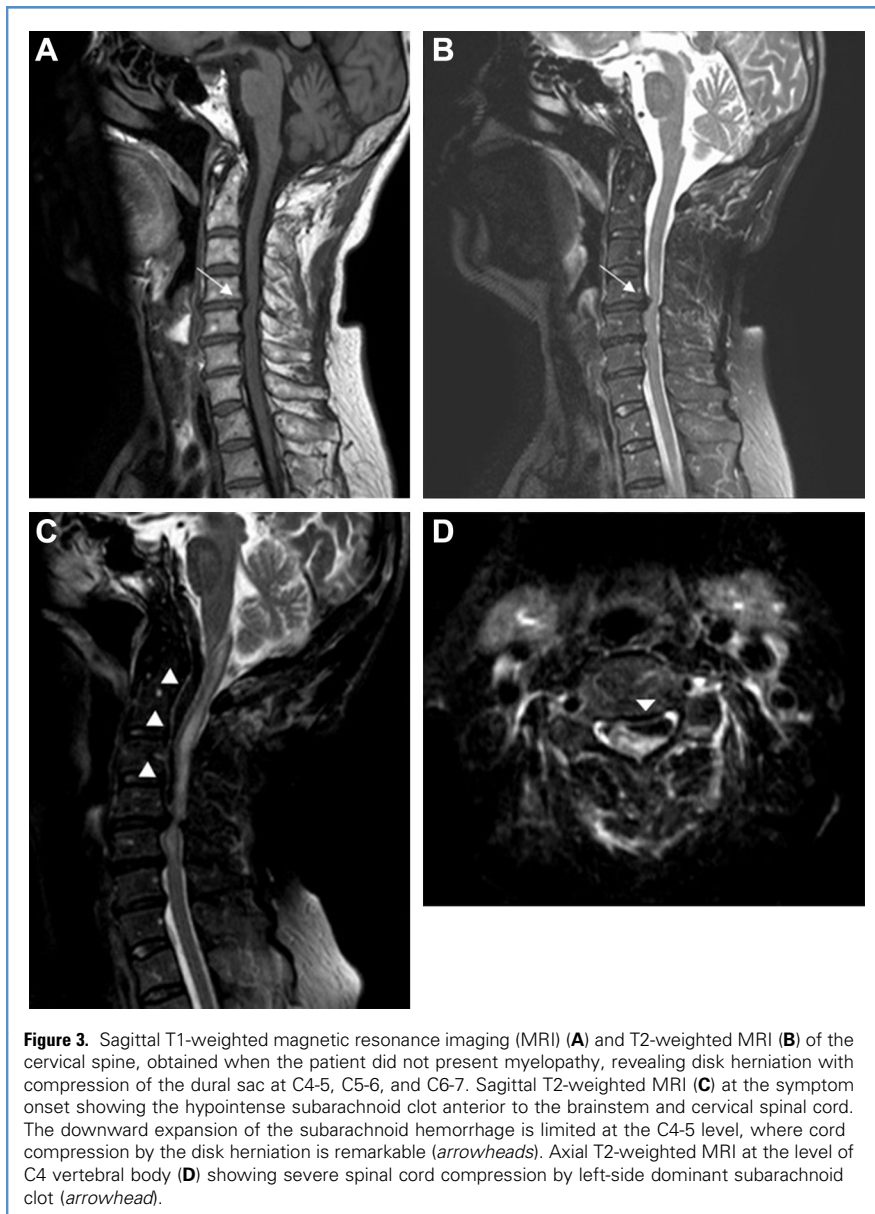


Figure 3. Sagittal T1-weighted magnetic resonance imaging (MRI) (A) and T2-weighted MRI (B) of the cervical spine, obtained when the patient did not present myelopathy, revealing disk herniation with compression of the dural sac at C4-5, C5-6, and C6-7. Sagittal T2-weighted MRI (C) at the symptom onset showing the hypointense subarachnoid clot anterior to the brainstem and cervical spinal cord. The downward expansion of the subarachnoid hemorrhage is limited at the C4-5 level, where cord compression by the disk herniation is remarkable (arrowheads). Axial T2-weighted MRI at the level of C4 vertebral body (D) showing severe spinal cord compression by left-side dominant subarachnoid clot (arrowhead).

compressive myelopathy caused by the combined effect of lumbar spondylosis and iatrogenic spinal SAH, in which the mechanism of symptom onset was analogous to that of our case. In the Hanakita case, failed lumbar epidural catheter insertion caused acute cauda equina syndrome, and the combined effect of iatrogenic spinal SAH deposition and lumbar disk herniation was revealed both by myelography and during decompressive laminectomy. This report suggested that lumbar region

spinal SAH could also cause acute myelopathy when combined with spondylosis.

In terms of rarity of this phenomenon, the reason why other hemorrhagic pathologies are less likely to cause acute myelopathy when combined with spondylosis must also be discussed. Spinal perimedullary AVF is associated with stagnant hypoxia of the spinal cord caused by venous hypertension.^{1,7} Therefore, it is possible that the spinal cord of perimedullary AVF was vulnerable to external

compression by disk herniation and SAH deposition. In addition, anatomic relationships between perimedullary AVF and disk herniation may also play a pivotal role for symptom onset. Facing antero-inferiorly at the upper cervical spinal cord, bleeding from craniocervical AVFs is more likely blocked by the disk herniation before diluted in a large amount of cerebrospinal fluid in the cisterna magna. Ruptured aneurysms in the posterior fossa (e.g., posterior inferior cerebellar artery aneurysms) could cause similar clinical manifestation to craniocervical junction AVFs caused by anatomic proximity to the cervical spinal cord. Although arterial bleeding from ruptured aneurysms is more severe than venous bleeding from AVFs, acute compressive cervical myelopathy associated with ruptured posterior inferior cerebellar artery aneurysm has never been reported. Because posterior inferior cerebellar aneurysms protrude posterosuperiorly into the cisterna magna, aneurysmal bleeding is immediately diluted in this large cerebrospinal fluid cistern and is less likely to cause intraspinal canal pressure elevation.

In the present case, a history of unknown SAH of approximately 20 years was noted. It is likely that her perimedullary AVF also bled at that time, but it was not detected as a bleeding source. The quality of angiography decades ago may not have been good enough to reveal the small arteriovenous shunt. Otherwise, located at the craniocervical junction, her AVF may have been out of the routine cranial angiographic range. Compressive myelopathy by SAH, which did not occur in her sixth decade, but rather in her eighth, may support the hypothesis that age-related degenerative change of her cervical spinal canal was involved in the distinct symptom onset.

In the present case, the patient presented with sudden headache and subsequent left hemiparesis. She was transferred to our stroke center, and meticulous head imaging was performed to detect the intracranial lesions. It was not until detailed neurologic examination was performed that cervical myelopathy was suspected. Even though the initial clinical manifestation and head imaging implied an intracranial lesion, a subarachnoid clot emptying into the upper

cervical spine could cause acute compressive myelopathy, therefore emphasizing the validity of detailed neurologic examination.

CONCLUSIONS

This case suggests that SAH by cranio-cervical junction AVF can cause acute compressive myelopathy when complicated with preexisting cervical spinal canal stenosis.

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Received 12 May 2016; accepted 8 August 2016

Citation: *World Neurosurg.* (2016).
<http://dx.doi.org/10.1016/j.wneu.2016.08.031>

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