

Cerebral Vasospasms After Intraventricular Hemorrhage From an Arteriovenous Malformation

—Case Report—

Shoji YOKOBORI, Akihiro WATANABE, Ryuta NAKAE, Hidetaka ONDA,
Akira FUSE, Shigeki KUSHIMOTO, and Hiroyuki YOKOTA

Department of Emergency and Critical Care Medicine, Nippon Medical School, Tokyo

Abstract

A 33-year-old female presented with a rare case of severe vasospasm following the rupture of an arteriovenous malformation (AVM) without subarachnoid hemorrhage. Initial computed tomography (CT) revealed a subcutaneous hematoma and cast formation of intraventricular clots without the deposition of subarachnoid blood in any basal cistern. Cerebral angiography revealed a small AVM located in the right parietal lobe without aneurysmal formations. Repeat CT demonstrated no evidence of subarachnoid clots expected with the presence of intraventricular clots and she was transferred to a general ward. She suffered sudden onset of motor aphasia and disturbance of consciousness on Day 17 after the hemorrhage. Magnetic resonance imaging indicated diffuse cortical infarction and subsequent magnetic resonance angiography revealed severe narrowing of the bilateral internal carotid arteries. Three-dimensional CT angiography on the same day indicated similar findings. She was transferred back to the intensive care unit for critical treatment. However, she suffered persistent mild right hemiparesis and motor aphasia. The characteristic features of vasospasm after intraventricular hemorrhage from AVMs are delayed onset, acute deterioration of consciousness, female predominance, and localization to the bilateral internal carotid arteries. Treatment of patients with AVM rupture should consider the risk of severe vasospasm, even if there is no subarachnoid clot.

Key words: vasospasm, arteriovenous malformation, intraventricular hemorrhage, cerebral infarction

Introduction

Cerebral arterial vasospasm often occurs in patients suffering from aneurysmal subarachnoid hemorrhage (SAH). Cerebral vasospasm is an uncommon but critical complication in patients with intraventricular hemorrhage (IVH) caused by arteriovenous malformation (AVM) rupture. Ruptured AVM can induce vasospasm, but rarely without SAH.^{14,15,17} We report a case of symptomatic vasospasm following IVH related to AVM.

Case Report

A 33-year-old female suddenly suffered headache, nausea, and loss of consciousness, and was transferred to our hospital. On admission, she was comatose with a Glasgow Coma Scale score of 13 and moderate left hemiparesis. Her blood pressure was 178/84 mmHg. Initial blood examinations, including clotting factors, found no abnormalities. She had no history of illnesses such as hypertension, and was not taking any medications on a regular basis.

Initial computed tomography (CT) indicated a right occipital subcutaneous hematoma and cast formation of intraventricular clots without blood accumulation in the basal cisterns (Fig. 1A). Her blood pressure was normalized with calcium antagonists to prevent deterioration of the hemorrhage, and she was admitted to the intensive care unit and observed closely.

The next day, her level of consciousness had slightly improved. Cerebral angiography performed on the following day revealed a small AVM located in the right parietal lobe, fed by the left anterior cerebral artery and drained by the right parietal cerebral vein, but no aneurysmal formations (Fig. 1B). Repeat CT at intervals of several days demonstrated no evidence of subarachnoid clots except for the presence of intraventricular clots. She was transferred to a general ward without severe neurological deterioration on Day 13 after admission.

Her verbal response suddenly decreased, and she appeared to have motor aphasia on Day 17 after the hemorrhage. Emergent magnetic resonance (MR) imaging indicated diffuse cortical infarction (Fig. 2A) and subsequent MR angiography revealed severe narrowing in the supraclinoid portions of the bilateral internal carotid arteries (Fig. 2B). Three-dimensional CT angiography per-

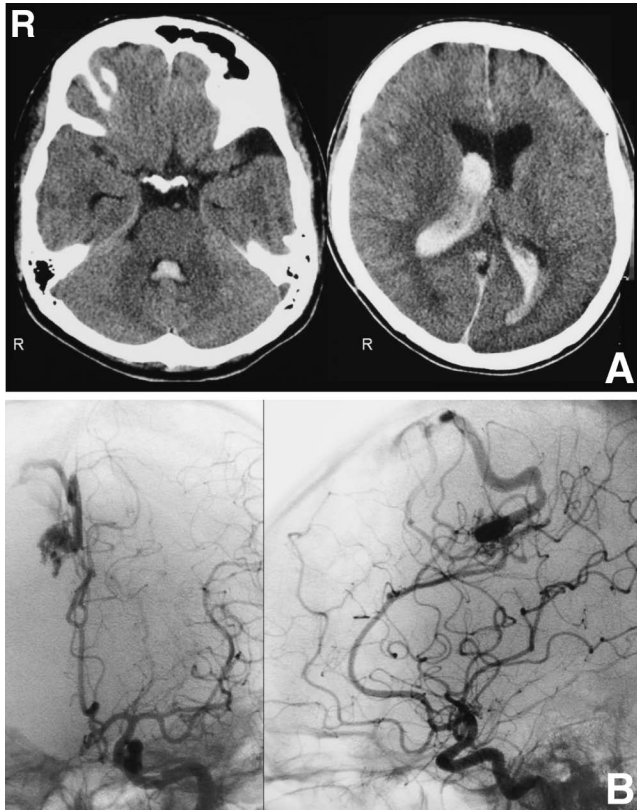


Fig. 1 A: Head computed tomography scans on admission showing right occipital subcutaneous hematoma and intraventricular clots in the lateral and fourth ventricles without accumulation of blood in the basal cisterns as subarachnoid hemorrhage. B: Left internal carotid angiograms performed on the day after admission revealing a small arteriovenous malformation in the right parietal lobe, fed by the left anterior cerebral artery and drained by the right parietal cerebral vein. No aneurysmal formation was seen. This series of bilateral carotid angiograms detected no spasm or occlusion of the cerebral vessels.

formed on the same day indicated similar findings. Her level of consciousness gradually deteriorated, and motor and sensory aphasia developed. She was treated with intravenous calcium antagonists, radical scavengers, and volume expanders, and was transferred back to the intensive care unit.

Despite intensive therapies for vasospasm, CT performed on Day 20 demonstrated low density areas indicating cerebral infarction in the left frontal lobe, left insular region, and right occipital lobe, according with the “watershed” area of the cerebrum. Her level of consciousness and aphasia improved during the 3rd week after the neurological deterioration. MR angiography on Day 24 demonstrated improvement of the severe spasms of the cerebral arteries (Fig. 2C).

Superselective embolization to the feeding artery and nidus was performed on Day 33. The size of the nidus was decreased and the flow of venous drainage reduced after embolization. However, she had persistent mild right

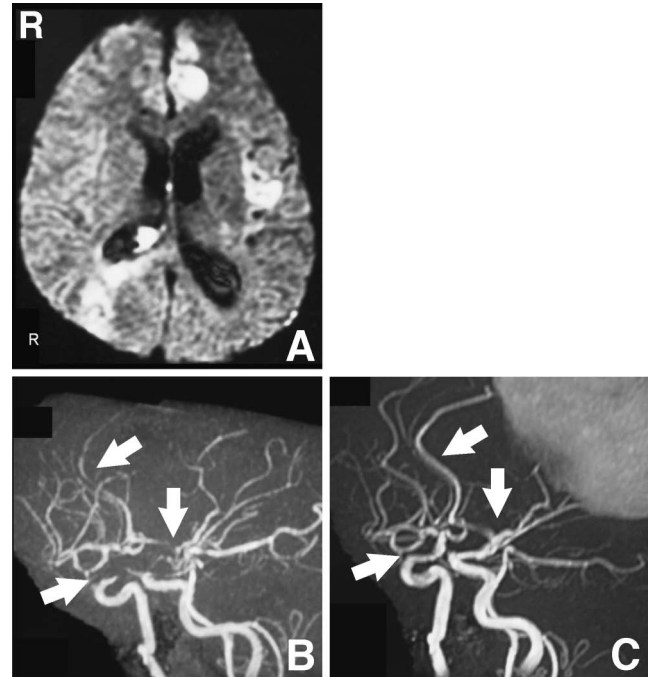


Fig. 2 A: Axial diffusion-weighted magnetic resonance (MR) image (repetition time/echo time 10,000/80 msec) on the 17th day of hospitalization demonstrating high intensity areas which indicated cerebral infarction in the bilateral hemisphere. B: MR angiogram performed on the same day revealing severe narrowing in the bilateral cerebral arteries, especially in the supraclinoid, A₁, and distal portions of the anterior cerebral arteries (white arrows). C: MR angiogram on the 24th day of admission revealing dramatic improvement of the vasospasms (white arrows).

hemiparesis and was transferred to another institute for rehabilitation on Day 40. She was discharged from the rehabilitation hospital and is planning to undergo gamma knife radiosurgical treatment of the residual nidus.

Discussion

Six cases of symptomatic vasospasm due to IVH following AVM rupture have been reported (Table 1).^{8,11,12,15,17} Initial CT demonstrated obvious IVH in all cases. Most cases had intraparenchymal hemorrhage as well but no cases demonstrated subarachnoid blood in the basal cisterns.

The size and thickness of subarachnoid blood in the basal cisterns are major predictors of vasospasm after aneurysmal bleeding,⁶ but the predictive value of IVH is not well known in patients with SAH. Several studies have found no significant association between IVH and the risk of vasospasm in the setting of aneurysm rupture,^{1,7} but others have demonstrated that IVH is an independent predictor of delayed vasospasm. Blood in the lateral ventricle might be a risk factor for delayed cerebral ischemia due to vasospasm in patients with SAH.³ The IVH was mainly distributed in the lateral ventricle in all previous cases (Table 1). Intraventricular clot was present for almost 2 weeks in our patient, so we suppose that such

Table 1 Reported cases of symptomatic vasospasm due to intraventricular hemorrhage (IVH) following arteriovenous malformation (AVM) rupture

Author (Year)	Country	Age (yrs)	Sex	IVH	ICH	SAH	Onset of vasospasm (days)	Location of intraparenchymal hemorrhage	Location of initial IVH	Location of AVM	Location of vasospasm	Infarct
Yanaka et al. (1992) ¹⁷⁾	Japan	11	F	+	-	-	3	-	lt lateral, third, fourth	lt MCA, lt ACA	bil A ₁ s, bil M ₁ s	lt parieto-occipital
Kothbauer et al. (1995) ¹²⁾	Switzerland	31	F	+	+	-	16	rt caudate nucleus	bil lateral, third, fourth	rt anterior callosal artery	bil ICAs, rt MCA	bil frontal
Maeda et al. (1997) ¹⁵⁾	Japan	40	F	+	+	-	6	rt thalamus	bil lateral, third	rt AchA	bil ICAs	lt parietal
Japan	Japan	14	F	+	+	-	17	rt putaminal	bil lateral, third	rt MCA	bil ICAs	unknown
Kobayashi et al. (2002) ¹¹⁾	Japan	26	M	+	-	-	17	-	rt lateral	rt AchA	bil MCAs, bil ACAs	rt temporo-occipital
Gerard et al. (2007) ⁸⁾	United States	41	F	+	-	-	10	-	lateral, third	rt PCA	bil MCAs, lt ICA, bil PCAs	lt parieto-occipital
Present case	Japan	33	F	+	+	-	17	rt occipital	lateral, third, fourth	lt ACA	bil ICAs, lt ACA	rt occipital, lt frontal, lt temporal

A₁: A₁ segment of anterior cerebral artery, ACA: anterior cerebral artery, AchA: anterior choroidal artery, PCA: posterior cerebral artery, SAH: subarachnoidal hemorrhage.

persistent clots in the lateral ventricles will stimulate the cerebral vessels continuously due to the circulation of bloody cerebrospinal fluid (CSF). Therefore, we also suggest that long-lasting clots are the main cause of vasospasm.

Vasospasm is also uncommon in patients with primary IVH. The first case of cerebral vasospasm associated with primary IVH suggested several mechanisms.⁴⁾ The main cause of vasospasms in patients with primary IVH could not be established, but subsequent hydrocephalus may have been involved. Hypothalamic dysfunction with extension of intracranial hemorrhage was also proposed as the cause of development of intracranial vasospasm.¹⁶⁾ Neither hypothalamic extension of intracranial bleeding nor remarkable severe hydrocephalus was present in our patient, so these mechanisms seemed unlikely causes of the vasospasm in our patient.

Bilateral carotid involvement also appears to be a characteristic feature of IVH-induced vasospasm after AVM rupture. The mechanism of vasospasm following IVH is unknown, but hypothetical mechanisms include recirculation of heme products into the subarachnoid CSF space with subsequent impairment of endothelium-dependent relaxation, decreased response to nitric oxide, increased sensitivity to the vasoconstrictor endothelin, and inactivity of potassium channels.⁵⁾

Remarkably, in 3 of the previous 6 cases, the onset of vasospasms occurred over 2 weeks after the hemorrhage (16-17 days after onset of hemorrhage). Our patient demonstrated late-phase vasospasm with onset on the 17th day after the hemorrhage. Such delayed onset may be a characteristic of vasospasm after only IVH is caused by bleeding from AVMs.¹⁵⁾ The cause of delayed vasospasm following IVH is not known. However, a large amount of IVH may delay CSF circulation and inhibit the washout of bloody CSF, thus provoking continuous cerebrovascular stimulation, resulting in delayed vasospasm.

The previous patients were almost all young females (Table 1). The incidence of AVM rupture in females is higher than in males.⁹⁾ No female predisposition for IVH has been described in patients with AVMs. However, female dominancy is known for the occurrence of vasospasm in patients with SAH,^{2,10)} but the mechanism is not well known. The effect of estrogen on changing vascular tone in coronary artery diseases may be important.¹³⁾ In our case, hormonal examination was not performed, so we cannot confirm any relationship between the hormonal balance and vasospasm. Analysis of the female hormones or sexual cycles is needed in patients with IVH from AVMs.

We conclude that the characteristic features of vasospasm after IVH from AVMs are delayed onset, acute deterioration of consciousness, female predominance, and localization to the bilateral internal carotid arteries. Treatment of patients with AVM rupture should consider the risk of severe vasospasm, even if there is no subarachnoid clot.

References

- 1) Brouwers PJ, Dippel DW, Vermeulen M, Lindsay KW, Hasan D, van Gijn J: Amount of blood on computed tomography as an independent predictor after aneurysm rupture. *Stroke* 24: 809-814, 1993
- 2) Carrera E, Schmidt JM, Oddo M, Fernandez L, Claassen J, Seder D, Lee K, Badjatia N, Connolly ES, Mayer SA: Transcranial Doppler for predicting delayed cerebral ischemia after subarachnoid hemorrhage. *Neurosurgery* 65: 316-324, 2009
- 3) Claassen J, Bernardini GL, Kreiter K, Bates J, Du YE, Copeland D, Connolly ES, Mayer SA: Effect of cisternal and ventricular blood on risk of delayed cerebral ischemia after subarachnoid hemorrhage: the Fisher scale revisited. *Stroke* 32: 2012-2020, 2001
- 4) Dull C, Torbey MT: Cerebral vasospasm associated with intraventricular hemorrhage. *Neurocrit Care* 3: 150-152, 2005
- 5) Faraci FM, Heistad DD: Regulation of the cerebral circulation: role of endothelium and potassium channels. *Physiol Rev* 78: 53-97, 1998
- 6) Fisher CM, Kistler JP, Davis JM: Relation of cerebral vasospasm to subarachnoid hemorrhage visualized by computed tomographic scanning. *Neurosurgery* 6: 1-9, 1980
- 7) Friedman JA, Goerss SJ, Meyer FB, Piepgras DG, Pichelmann MA, McIver JI, Toussaint LG 3rd, McClelland RL, Nichols DA, Atkinson JL, Wijdicks EF: Volumetric quantification of Fischer Grade 3 aneurysmal subarachnoid hemorrhage: a novel method to predict symptomatic vasospasm on admission computerized tomography scans. *J Neurosurg* 97: 401-407, 2002
- 8) Gerard E, Frontera JA, Wright CB: Vasospasm and cerebral infarction following isolated intraventricular hemorrhage. *Neurocrit Care* 7: 257-259, 2007
- 9) Graf CJ, Perret GE, Torner JC: Bleeding from cerebral arteriovenous malformations as part of their natural history. *J Neurosurg* 58: 331-337, 1983
- 10) Kaul S, Wozniak M, Rigamonti D, Sloan MA: Influence of race and gender on TCD diagnosis of vasospasm following subarachnoid hemorrhage. *Stroke* 27: 792, 1996
- 11) Kobayashi M, Takayama H, Mihara B, Kawase T: Severe vasospasm caused by repeated intravascular haemorrhage from small arteriovenous malformation. *Acta Neurochir (Wien)* 144: 405-406, 2002
- 12) Kothbauer K, Schroth G, Seiler RW, Do DD: Severe sympathetic vasospasm after rupture of an arteriovenous malformation. *AJNR Am J Neuroradiol* 16: 1073-1075, 1995
- 13) Kublickiene K, Luksha L: Gender and the endothelium. *Pharmacol Rep* 60: 49-60, 2008
- 14) Lobato RD, Gomez PA, Rivas JJ: Arteriovenous malformations and vasospasm. *J Neurosurg* 88: 934-935, 1998
- 15) Maeda K, Kurita H, Nakamura T, Usui M, Tsutsumi K, Morimoto T, Kirino T: Occurrence of severe vasospasm following intraventricular hemorrhage from an arteriovenous malformation. *J Neurosurg* 87: 436-439, 1997
- 16) Wilkins RH: Hypothalamic dysfunction and intracranial arterial spasms. *Surg Neurol* 4: 472-480, 1975
- 17) Yanaka K, Hyodo A, Tsuchida Y, Yoshii Y, Nose T: Symptomatic cerebral vasospasm after intraventricular hemorrhage from ruptured arteriovenous malformation. *Surg Neurol* 38: 63-67, 1992

Address reprint requests to: Shoji Yokobori, M.D., Department of Emergency and Critical Care Medicine, Nippon Medical School, 1-1-5 Sendagi, Bunkyo-ku, Tokyo 113-8603, Japan. e-mail: shoji@nms.ac.jp