Case Report

Arteriovenous Malformation Presenting with Acute Subdural Hemorrhage: Case Report and Literature Review

Keita Ueda* Jun Nomoto Masaaki Nemoto
Takaaki Nagao Daisuke Fukushima Kosuke Kondo
Naoyuki Harada and Nobuo Sugo

Department of Neurosurgery (Omori), School of Medicine, Faculty of Medicine, Toho University

ABSTRACT: We treated a patient with arteriovenous malformation (AVM) complicated by acute subdural hematoma (ASDH) resulting in cardiopulmonary arrest. A 12-year-old boy suddenly developed severe headache and disturbance of consciousness (3 on the Glasgow Coma Scale). He was in cardiopulmonary arrest, but heartbeat was restored during cardiopulmonary resuscitation in the ambulance. Computed tomography images of the head showed an AVM-induced intracerebral hematoma in the left occipital lobe and left ASDH, which was removed in an emergency procedure. Consciousness improved with normalization of intracranial pressure. The main feeder of the AVM was embolized on the 71st hospital day, and the AVM was resected on the following day. Postoperative course was satisfactory, and the patient was discharged with no neurologic abnormalities. Emergent treatment and a stepwise surgical strategy resulted in a satisfactory therapeutic outcome for AVM complicated by ASDH.

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KEYWORDS: arteriovenous malformation (AVM), acute subdural hematoma (ASDH), cardiopulmonary arrest (CPA)

The annual incidence of hemorrhage due to arteriovenous malformation (AVM) is 3.0%, and the incidences of initial and recurrent hemorrhage are 2.2% and 4.5%, respectively.[1] Most cases are complicated by intracerebral hematoma; acute subdural hematoma (ASDH) is rare.[2] For treatment of ASDH, early surgical intervention is essential, regardless of whether the condition is the result of trauma. Prognosis is poor.[3] We report a case of AVM resulting in ASDH and cardiopulmonary arrest (CPA) for which the outcome was favorable, and review the relevant literature.

Case Presentation

Patient: A 12-year-old boy

Chief complaint: Disturbance of consciousness

Past medical history: Unremarkable

Familial medical history: Unremarkable

History of present illness: None in particular

The patient developed acute headache during a swimming class. An ambulance was requested because his consciousness level rapidly deteriorated and he developed generalized tonic-clonic seizures. When the ambulance arrived, consciousness was III-300 on the Japan Coma Scale (JCS) and 3 (E1V1M1) on the Glasgow Coma Scale (GCS). Both pupils were dilated (6/6 mm), the light reflex and spontaneous respiration were absent, pulse was nonpalpable, and blood pressure was unmeasurable, indicating CPA. The ambulance paramedics began cardiopulmonary

6–11–1 Omorinishi, Ota, Tokyo 143–8541
*Corresponding Author: tel: 3(3762)4151
e-mail: ke-ueda@yj8.so-net.ne.jp

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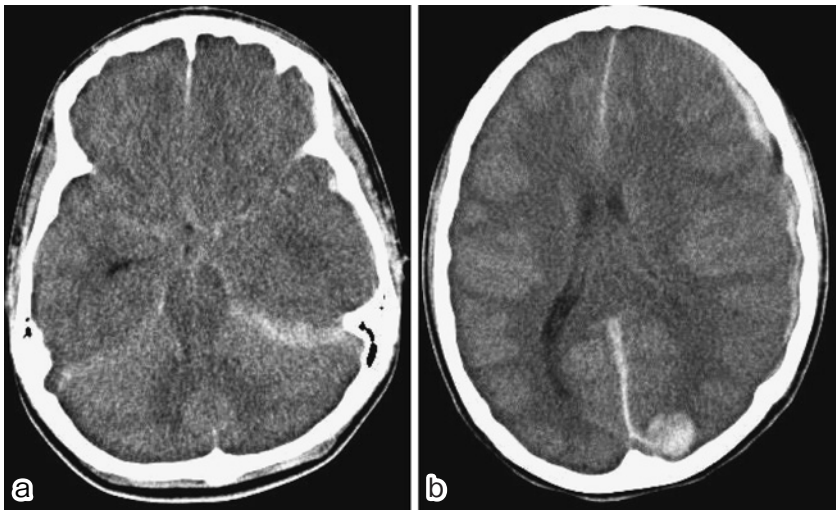

Fig. 1 Initial computed tomography (CT)
a: Subarachnoid hemorrhage in the basal cistern.
b: Shift of midline structures due to acute subdural hematoma (ASDH) over the left cerebral convexity, with ASDH.

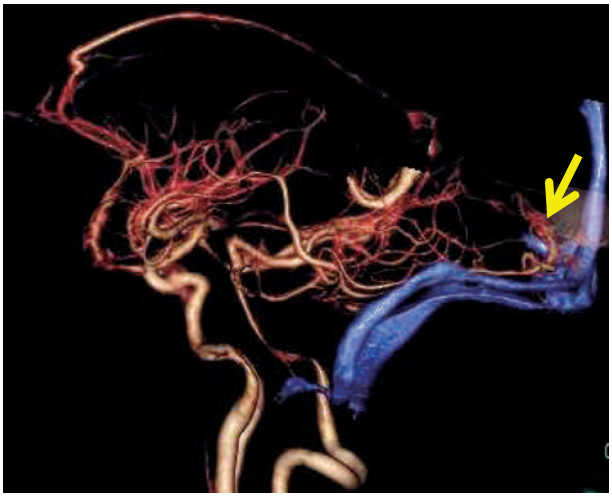

Fig. 2 Three-dimensional computed tomography angiography (3DCTA)
The nidus of the arteriovenous malformation (AVM) is in the left occipital lobe (arrow).

resuscitation. Because ventricular fibrillation was detected on electrocardiographic monitoring, an automated external defibrillator was used, and heartbeat resumed. The duration of cardiopulmonary arrest was estimated at 7 minutes.

Status on arrival

On arrival at our hospital, the JCS was III-300, GCS was E1V1M1, and pupil diameters were 6/6 mm. The light reflex was absent on both sides. No head trauma was noted. Systolic blood pressure was 108 mmHg, pulse was 115/min and regular, and respiratory rate was 40/min.

Examination findings on arrival

Results of arterial blood analysis (pH, 7.088; PCO2, 67.1 Torr; PO2, 121 Torr; HCO3, 19.4 mEq/l; BE, 11.9 mEq/l; and Lac, 7.5) showed marked respiratory acidosis. Computed tomography (CT) images of the head showed ASDH expansion in the left convexity and marked midline shift. An intracerebral hematoma was noted in the left occipital lobe, and the ASDH was in contact with the hematoma, along the falx and tentorium. Subarachnoid hemorrhage was observed in the basal cistern (Fig. 1).

Course after admission

Emergent 3-dimensional CT angiography (3DCTA) showed the nidus of the AVM at the site corresponding to the intracerebral hematoma in the left occipital lobe (Fig. 2). One-step craniotomy was not suitable for simultaneous treatment of the AVM in the left occipital lobe and ASDH quickly expanding in the left convexity. Because the patient's general condition was poor, due to his recovery from CPA, and the need for AVM treatment was not urgent, we selected stepwise treatment, *i.e.*, emergent removal of ASDH and external decompression to save his life, followed by AVM resection in the second step. Frontotemporal craniotomy through a skin incision was used for ASDH removal. Intracranial pressure (ICP) was high, and the dura mater was taut. A hematoma (thickness, 1 cm) was present directly below the dura mater. After ASDH removal, no brain contusion or hemorrhage from the su-

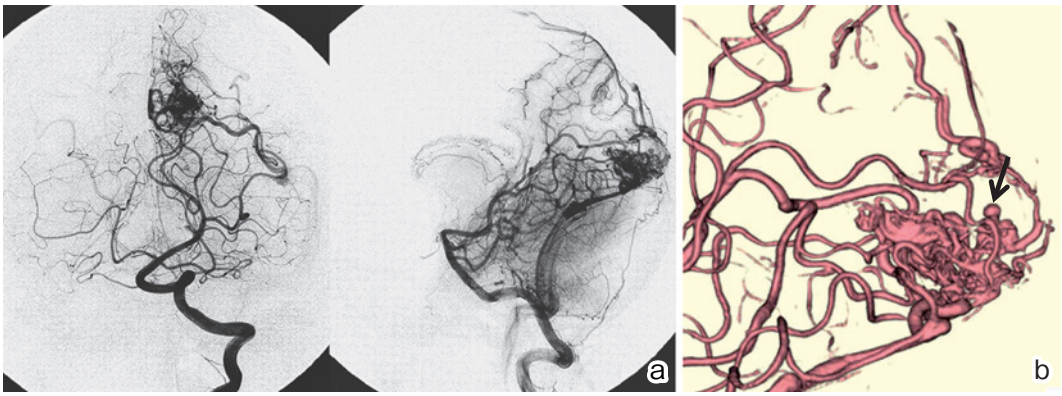

Fig. 3 Cerebral angiography
a: Angiogram of the left vertebral artery shows the nidus of the arteriovenous malformation (AVM). The main feeders are the calcarine artery and parieto-occipital artery. The drainers flow out into the superior sagittal and left sigmoid sinuses.
b: A 3-dimensional image showing an aneurysm on a feeder of the nidus (arrow).

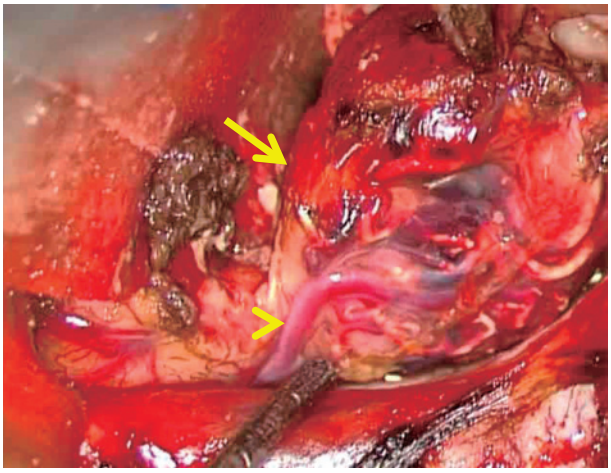

Fig. 4 Operative photograph of arteriovenous malformation (AVM) resection
Arrow: nidus of AVM, Arrowhead: drainer of AVM.

perficial blood vessels was noted on the brain surface, and there was no arterial or venous dilation associated with AVM. External decompression was applied, and surgery was completed. After confirming normalization of ICP on time-course CT after surgery, sedation was withdrawn on the ninth hospital day.

Arousal was rapid and his consciousness level recovered. No paralysis developed in any limb, and no neurologic deficit other than mild right facial palsy was noted. On the 20th hospital day, a flow void sign was noted in the left occipital lobe on T2-weighted magnetic resonance imaging (MRI) of the head, and the nidus was partially enhanced with gadolinium. On cerebral angiography performed on the 22nd hospital day, the AVM nidus was confirmed, as were 6 feeders. The main feeders were the calcarine artery and the parieto-occipital artery. Moreover, an aneurysm was present on a feeder of the nidus, the probable cause of hemorrhage. The drainers flowed out into the superior sagittal and left sigmoid sinuses (Fig. 3). Spetzler-Martin grade II AVM was diagnosed, and the main feeders (calcarine and parieto-occipital arteries) were embolized with Eudragit® (Evonik Industries AG, Essen, Germany) on the 71st hospital day. The nidus mostly disappeared, leaving only a small arteriovenous shunt on imaging. AVM resection was performed on the next (72nd) hospital day. Occipital craniotomy was performed with the patient in prone position. After incision of the dura mater, the nidus was partially exposed on the brain surface. The vessels of the nidus were obstructed, became white and hard, and were easily detached from adjacent brain tissue. The nidus was circumferentially dissected, and several deep, thin feeders were cut. Finally, the main drainers were processed, the nidus was excised en bloc, and surgery was completed (Fig. 4). Postoperative course was favorable: the right facial palsy noted immediately after the first surgery resolved, and the patient was discharged on the 90th hospital day with no neurologic sequelae.

Discussion

Most cases of ASDH are traumatic; nontraumatic ASDH accounts for only about 5% of cases.[4] Ruptured cerebral aneurysm, AVM, arteriovenous fistula, brain tumor, dural metastases of malignant tumors, abnormal coagulation, immunodeficiency, drug poisoning, and Moyamoya disease are reported causes of nontraumatic ASDH.[5] Because the

Table 1 Summary of cases of ASDH due to ruptured AVM[2, 11–14]

Author	Age	Sex	Consciousness	Grade	ASDH	AVM	Outcome
Oikawa (1993) [2]	13	M	ND	ND	removal	removal	ND
Datta (2000) [11]	48	M	GCS 3	Ⅱ	removal	removal	GR
Handa (2002) [12]	31	F	JCS 200	ND	removal	removal	GR
Inoue (2004) [13]	17	M	JCS 300	ND	-	-	death
Kominato (2004) [14]	42	M	ND	ND	-	-	death
This case (2014)	12	M	GCS 3	Ⅱ	removal	removal	GR

GCS: Glasgow Coma Scale, JCS: Japan Coma Scale, Grade: Spetzler-Martin grade, ASDH: acute subdural hematoma, AVM: arteriovenous malformation, M: male, F: female, ND: not determined, GR: good recovery

present patient had no past medical history of trauma and no evidence of trauma, such as brain contusion or vascular injury, on the brain surface during surgery, ASDH was likely due to hemorrhage from the AVM. 3DCTA and cerebral angiography showed an aneurysm in the AVM nidus, which suggests that it was the cause of the AVM rupture-induced hemorrhage. In about half of cases with concomitant aneurysm and AVM, the aneurysm is formed in the feeding artery, and this is an important risk factor for hemorrhage in AVM.[1] Turjman et al reported that a concomitant aneurysm was present in AVM in 5 of 58 patients and that the AVM manifested with hemorrhage in these 5 patients.[6]

Hemorrhage caused by AVM rupture perforates the pia mater and arachnoid, resulting in ASDH. A number of hypotheses have been postulated for this pathologic condition.[7–10] Specifically, idiopathic or traumatic adhesion between the nidus and arachnoid membrane causes hemorrhage in the subdural space. Because the present case was complicated by cerebral hemorrhage, hematomas formed bidirectionally in the brain and subdural space by means of the mechanism described above. In addition, CT showed that the ASDH was continuous with the intracerebral hematoma accompanying the nidus along the falx and tentorium. This suggests that the hematoma had expanded in the subdural space of the convexity through this pathway.

AVM rupture-induced ASDH is very rare: only 6 cases, including our patient, have been reported (Table 1).[2, 11–14] Consciousness level on arrival was poor (JCS200-300) in 3 of the 4 cases in which consciousness level was described, but the outcome was not necessarily correlated with state of consciousness on arrival or Spetzler-Martin grade of AVM. The factor believed to be most strongly associated with outcome is performance of emergent surgery for ASDH. ASDH was urgently removed and external decompression was applied in 4 of the 6 reported cases, and recovery was favorable in 3, excluding 1 case with missing information.[2, 11, 12] In addition, treatment was completed with AVM resection as the second step in all 4 of these cases.[2, 11, 12] As compared with the 2 patients with clearly reported treatments and outcomes, our patient had a good recovery, despite having the most severe complication (*i.e.*, CPA) (Table 1), perhaps because of rapid cardiopulmonary resuscitation by the ambulance paramedics.[11, 12]

Surgery for ASDH should be done immediately for decompression when there are signs of impending brain herniation caused by intracranial hemorrhage due to a ruptured aneurysm or cerebral arteriovenous malformation.[3] Ruptured AVM complicated by hematoma requires ICP control, and the anatomic characteristics and hemodynamics of AVM should be clarified for curative treatment of AVM, in view of the low re-rupture and fatality rates as compared with ruptured aneurysms, although rebleeding is a risk. Thus, only the hematoma should be removed, and subsequent two-stage multidisciplinary treatment should be implemented.[15, 16]

Our results suggest that it is important to remove the ASDH and apply external decompression in patients with enhanced ICP due to AVM-induced ASDH.[12] Rapid control of ICP may increase the possibility of a full recovery.

Conclusion

We reported a case of AVM complicated by ASDH resulting in cardiopulmonary arrest that culminated in a favorable outcome. The results suggest that emergent surgical intervention for ASDH is important in controlling ICP. Outcomes might be improved by a stepwise surgical strategy in which AVM is safely resected in combination with embolization after the general condition of the patient stabilizes.

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