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Case Report

A Pure Acute Subdural Hematoma Presenting with a Diploic Arteriovenous Fistula: Case Report and Literature Review

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Objective: We report a case of pure acute subdural hematoma (SDH) caused by a diploic arteriovenous fistula (AVF) and it is a first case report as far as we researched it.

Case Presentation: A 19-year-old man was admitted as an emergency to our hospital with headache and nausea. CT scan on hospital admission showed a right acute SDH. Because there was no history of head trauma, MRI, MRA, and DSA were performed to identify a source of bleeding. DSA disclosed an AVF. The shunt was located between a frontotemporal branch of the middle meningeal artery (MMA) and a diploic vein, and its shunting point formed an aneurysmal sac, which was considered to have ruptured. Endovascular treatment was administered rather than surgical treatment to prevent re-bleeding because the patient was conscious and alert, CT showed a small SDH, and the left MMA near the shunting point was accessible for catheterization. A diluted mixture of 25% n-butyl-2-cyanoacrilate was injected into a left frontoparietal branch just before the shunting point and the shunt, including the aneurysmal sac, was obliterated. The patient's postoperative course was uneventful and he was discharged without neurological deficits.

Conclusion: We experienced a patient with a pure acute SDH caused by diploic AVF. In patients with non-traumatic acute SDH, DSA is recommended to determine its underlying cause. Our review of published reports yielded few instances of non-traumatic pure acute SDH in young people. Possible causative factors should be investigated promptly and appropriate treatment provided immediately.

Keywords ▶ diploic arteriovenous fistula, non-traumatic, acute subdural hematoma, rupture point, endovascular surgery

Introduction

Non-traumatic acute subdural hematoma (SDH) is very rare in young people. It can be caused by hypertension, vascular malformations, coagulopathy, or anticoagulation and antiplatelet medication taken via the oral route.[1,2)]

Non-traumatic acute SDH often occurs in combination with intracerebral hemorrhage and/or subarachnoid hemorrhage,[3)] and a pure SDH is extremely rare.[3–5)] We treated a patient with a diploic arteriovenous fistula (AVF) presenting with a pure acute SDH. In our case, an aneurysmal sac, located at the shunting point, might have been the rupture source. We review the literature on AVFs associated with a pure acute SDH and discuss such a rare pathology.

Case Presentation

Present illness: A 19-year-old man was admitted to the emergency department of our hospital because of sudden onset of right temporal headache and nausea. He denied having suffered head trauma or taking anticoagulant and antiplatelet medication.

Neurological examination revealed a Glasgow Coma Scale (GCS) score of 15 but, otherwise, no remarkable

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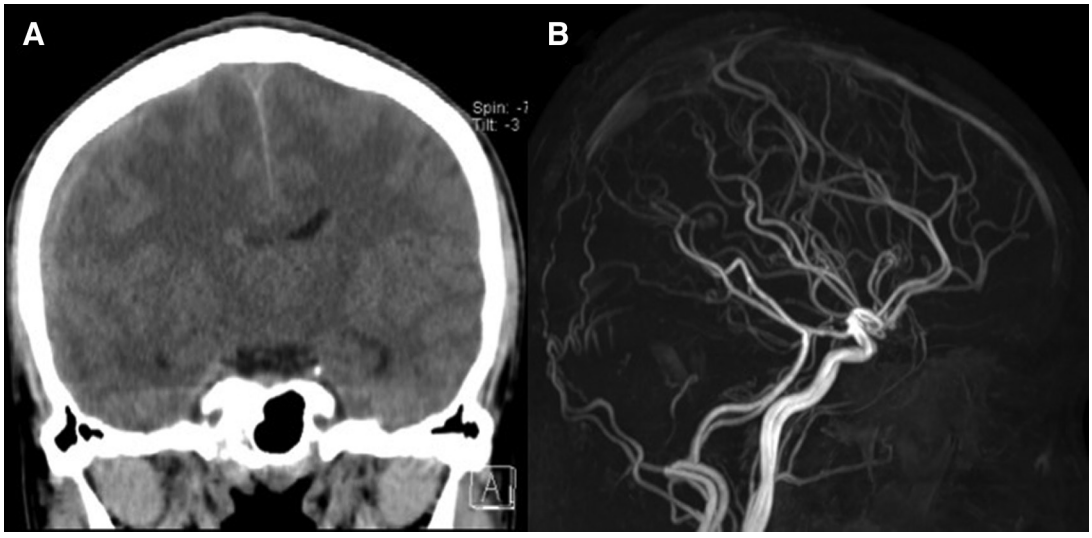

Fig. 1 (**A**) Coronal CT shows a thin acute SDH on the right cerebral hemisphere with a mild midline shift. (**B**) MRA shows the SSS and cortical vein. SDH: subdural hematoma; SSS: superior sagittal sinus

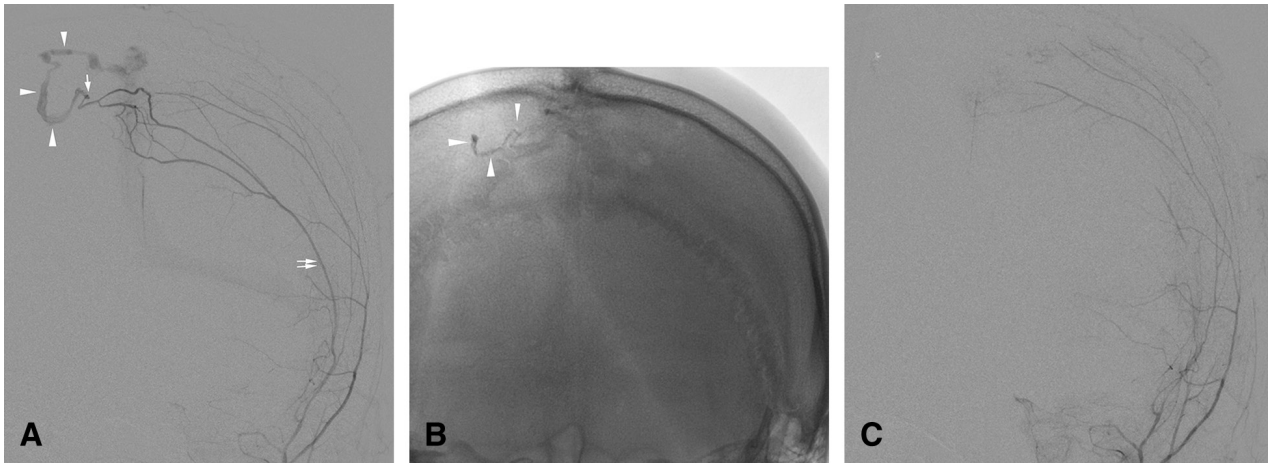

Fig. 2 Super-selective angiography of the left MMA in the arterial phase (left anterior oblique position) (**A** and **C**) shows an abnormal artery originating from a frontoparietal branch of the left MMA (double arrow) connected to a diploic vein (arrowheads) and its shunting point formed an aneurysmal sac (arrow). (**A**) Before embolization and (**B**) NBCA cast (arrowheads) after embolization, and (**C**) complete obliteration of the AVF after embolization. AVF: arteriovenous fistula; MMA: middle meningeal artery; NBCA: n-butyl-2-cyanoacrilate

features were noted upon hospital admission. CT upon hospital admission showed an acute SDH with a mild midline shift to the left (**Fig. 1A**). MRI and MRA of the brain were performed to exclude intracranial vascular malformations. The superior sagittal sinus (SSS) and cortical vein were revealed on MRA (**Fig. 1B**). DSA was planned to clarify the bleeding source for a non-traumatic acute SDH. Under local anesthesia, cerebral angiography was performed while confirming consciousness level. Angiography of the left and right external carotid arteries disclosed an AVF fed by a frontoparietal branch of the left middle meningeal artery (MMA) and right MMA, and drained into a diploic vein at the right convexity and then drained into the SSS via the venous lacuna (**Figs. 2A**, **3A**, and **3B**). Its shunting point had an aneurysmal sac directing to a subdural space (**Fig. 3A**). We suspected that this aneurysmal sac was the rupture source. Because the patient was conscious and alert (GCS score of 15), a small SDH was observed on CT, and the left MMA near the shunting point was accessible for catheterization, endovascular treatment was planned to obliterate the AVF and aneurysmal sac. The patient's consciousness level was carefully observed and he remained alert without deterioration until the following day, when endovascular treatment was performed under general anesthesia. A 7 Fr guiding catheter (Roadmaster; Goodman, Aichi, Japan) was placed in the left external

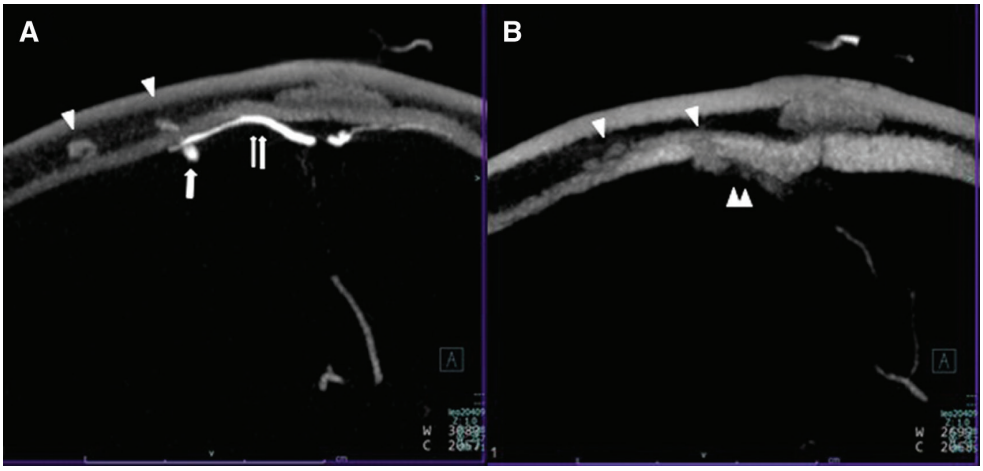

Fig. 3 Cone-beam CT shows (**A**) an AVF fed by a frontoparietal branch of the left MMA (double arrow). Its shunting point had an aneurysmal sac (arrow) directing to a subdural space, and drained into a diploic vein (arrowheads). (**B**) An AVFs drained into a diploic vein (arrowheads) and then drained into the SSS via the venous lacuna (double arrowhead). AVF: arteriovenous fistula; MMA: middle meningeal artery; SSS: superior sagittal sinus

carotid artery. A 4.2 Fr catheter (Fubuki; Asahi Intecc, Aichi, Japan) was navigated through this artery to just before the foramen spinosum level and a micro-catheter (Marathon; Medtronic, Minneapolis, MN, USA) was navigated to near the shunting point of a frontoparietal branch of the MMA using a microguidewire (Chikai 10, Asahi Intecc, Aichi, Japan). Then, 0.17 mL of a dilute mixture of 25% n-butyl-2-cyanoacrylate was injected into the left MMA. A small amount of n-butyl-2-cyanoacrylate glue reached into the aneurysmal sac and diploic vein penetrating the shunt (**Fig. 2B**). After embolization, angiography of the left external carotid artery showed complete obliteration of the AVF including the aneurysmal sac (**Fig. 2C**). Postoperative CT, performed just after the embolization, showed that the SDH had slightly diminished. His postoperative course was uneventful and the patient was discharged without neurological deficits. Twenty days after embolization, the SDH had disappeared on CT.

Discussion

We reported a pure acute SDH in a young man caused by a diploic AVF, the shunting point of which formed an aneurysmal sac that seemed to have ruptured.

Diploic AVFs are rare entities. The symptoms of diploic AVFs include intermittent tinnitus, head pain, head swelling (scalp hematoma), and headache.[6,7] Interestingly, it has been proposed that an aneurysmal bone cyst can be induced by diploic AVF.[8,9] The causative factors in nine previously reported diploic AVF cases were trauma in three cases,[7,10,11] spontaneous occurrence in five cases,[6,7,9,12,13] and occurrence during the postpartum period in one case.[8] The latter might be caused by dural sinus thrombosis, but the etiology of most diploic AVFs remains unclear.[7] The vascular structure of diploic AVF consists of abnormal intraosseous channels between meningeal arteries and diploic venous channels. They can be classified into three types according to patterns of venous outflow: (i) dural sinus drainage only; (ii) extracranial drainage only; and (iii) dural sinus and extracranial drainage.[7]

Our patient had a diploic AVF, which was supplied by a frontoparietal branch of the right and left MMAs, and drained into the diploic vein at the parietal convexity and then the SSS via the venous lacuna and is considered to be type I according to the classification created by Rivera-Lara.[7]

Of all reported acute SDHs, 2%–6.7% are non-traumatic.[1,2] Coombs et al. reviewed 193 cases of non-traumatic acute SDH reported in the literature[1] and found that the causes included intracranial vascular malformation, coagulation disorder, tumors, spontaneous intracranial hypertension, arachnoid cysts, and cocaine use.

Non-traumatic acute SDHs caused by intracranial AVFs are usually accompanied by intracerebral hemorrhage and/or subarachnoid hemorrhage, and pure SDHs are extremely rare.[3,14] We found nine cases of intracranial AVFs causing a pure SDH in the literature (including our patient).[3–5,14–17]

Cerebral angiography undertaken in seven cases demonstrated venous drainage to the SSS in four cases, to the diploic vein in one case (the present one), to the diploic and dural veins in one case, and to the frontal vein in one case.

Hemorrhage can result from rupture of a shunting point of these AVFs, a cortical artery at the site of adhesion with the dura mater,[18] a subpial vein, a fragile wall of a venous ectasias in the subdural space, a vein that is draining because of an increase in arterial or venous pressure,[3,5] or an arterialized dilated draining vein.[15] In our patient, a shunting point between a frontoparietal branch of the MMA and a diploic vein consisted of an aneurysmal sac, which was considered the source of bleeding. The presence of diploic AVF is apparent in angiography; however, dural AVF might not be depicted in acute phase.

The treatment can be determined by the state of patient and hematoma volume. In a conscious and alert patient with a small SDH on CT and accessibility of intracranial AVFs for catheterization, endovascular treatment can be initially undertaken to obliterate the bleeding point. The option of open surgery for removal of the hematoma can be considered under close observation of the patient.

Conclusion

We report an extremely rare case of a pure acute SDH caused by a diploic AVF involving the MMA and diploic veins draining into the SSS via the venous lacuna. Its shunting point had an aneurysmal sac, which was considered to have ruptured. The shunt (including the aneurysmal sac) was obliterated by endovascular treatment. In cases of non-traumatic pure acute SDH in young people, DSA is recommended to determine the underlying cause and appropriate treatment should be provided promptly. This case report may contribute to understanding the diversity of diploic AVFs.

Disclosure Statement

The authors declare no conflict of interest.

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