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Convexity Dural Arteriovenous Fistula without Cortical Venous Reflux Presenting with Pure Acute Subdural Hematoma

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Abstract

Hemorrhagic changes in a dural arteriovenous fistula are typically associated with cortical venous reflux and occur as intracerebral or subarachnoid hemorrhages. A convexity dural arteriovenous fistula (DAVF) usually flows directly into the cortical veins and exhibits cortical venous reflux. Herein, we report a rare case of a convexity DAVF without cortical venous reflux presenting with a pure acute subdural hematoma. A 19-year-old man complaining of headache without any history of head injury was diagnosed with a left acute subdural hematoma on magnetic resonance imaging (MRI) and referred to our hospital. The patient was conscious and exhibited no neurological signs. The MRI did not reveal any possible abnormalities leading to hemorrhage. Cerebral angiography revealed a dural arteriovenous fistula in the left parietal cranium with a feeder from the middle meningeal artery and a drainer into the main transverse sinus via a diploic vein. Part of the shunt blood flowed into the superior sagittal sinus from the meningeal vein; however, there was no reflux into the cortical vein or stasis of the cerebral vein, suggesting venous hypertension. A convexity DAVF was diagnosed as the source of bleeding, and transarterial embolization was performed. The patient recovered without any neurological deficits. In the absence of trauma, an acute subdural hematoma requires an appropriate evaluation of the vascular lesions and a treatment plan.

Keywords: dural arteriovenous fistula, convexity, acute subdural hematoma, transarterial embolization

Introduction

A convexity dural arteriovenous fistula (DAVF) often flows directly into the cortical veins, resulting in malignant symptoms, including intracranial hemorrhage, venous infarction, intracranial hypertension, and seizures.[1)] Cortical venous reflux (CVR) is a known risk factor for intracranial hemorrhage in DAVF.[2,3)] Intracerebral and subarachnoid hemorrhages are frequent hemorrhagic manifestations of DAVF. In contrast, a pure subdural hematoma is rare.[4)] Herein, we describe an unusual convexity DAVF without CVR presenting with acute subdural hematoma (ASDH).

Case Report

The patient was a healthy 19-year-old man without any history of head injury. He developed a headache without any apparent trigger and visited a local neurosurgical clinic 5 days later. Magnetic resonance imaging (MRI) of the head revealed a left subdural hematoma (Fig. 1A), and he was referred to our hospital. The patient had a headache but was oriented and alert. He had no nausea or vomiting, and abnormal neurological findings were absent. Blood tests showed no bleeding diathesis. The MRI revealed a subdural hematoma extending from the left parieto-occipital region to the interhemispheric fissure. Non-contrast computed tomography (CT) revealed an acute high-density hematoma. Contrast-enhanced CT revealed a prominent diploic vein on CT venography but no obvious vascular abnormalities (Fig. 1B, C). Cerebral angiography revealed a dural arteriovenous fistula on the left parietal cranium with a feeder from the left middle meningeal artery (Fig. 2A). Most of the shunt blood flowed anterogradely through the transverse sinus via the diploic

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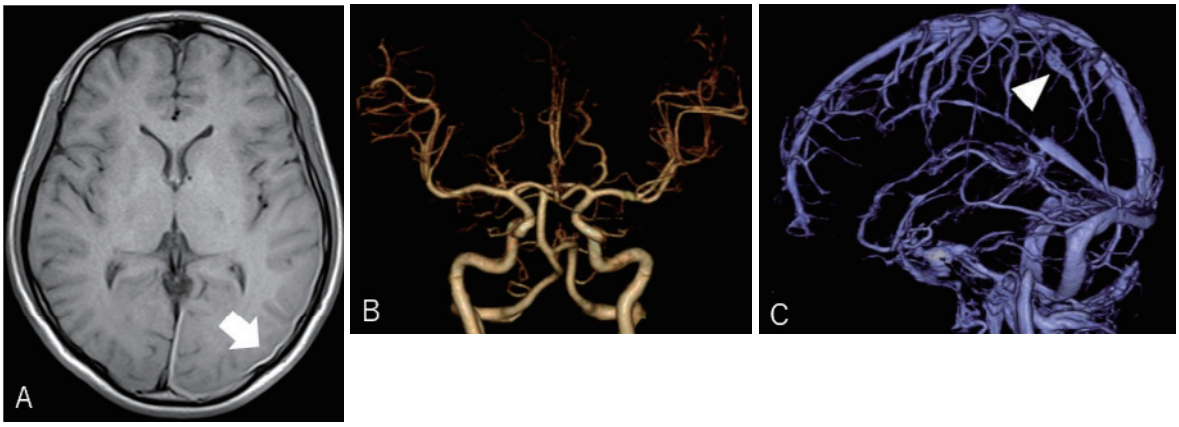

Fig. 1 Subdural hematoma (arrow) extending from the parieto-occipital region to the interhemispheric fissure detected on T1-weighted magnetic resonance imaging (A). Contrast-enhanced computed tomography (CT) reveals a prominent diploic vein (arrowhead) on CT venography without obvious vascular abnormalities (B, C).

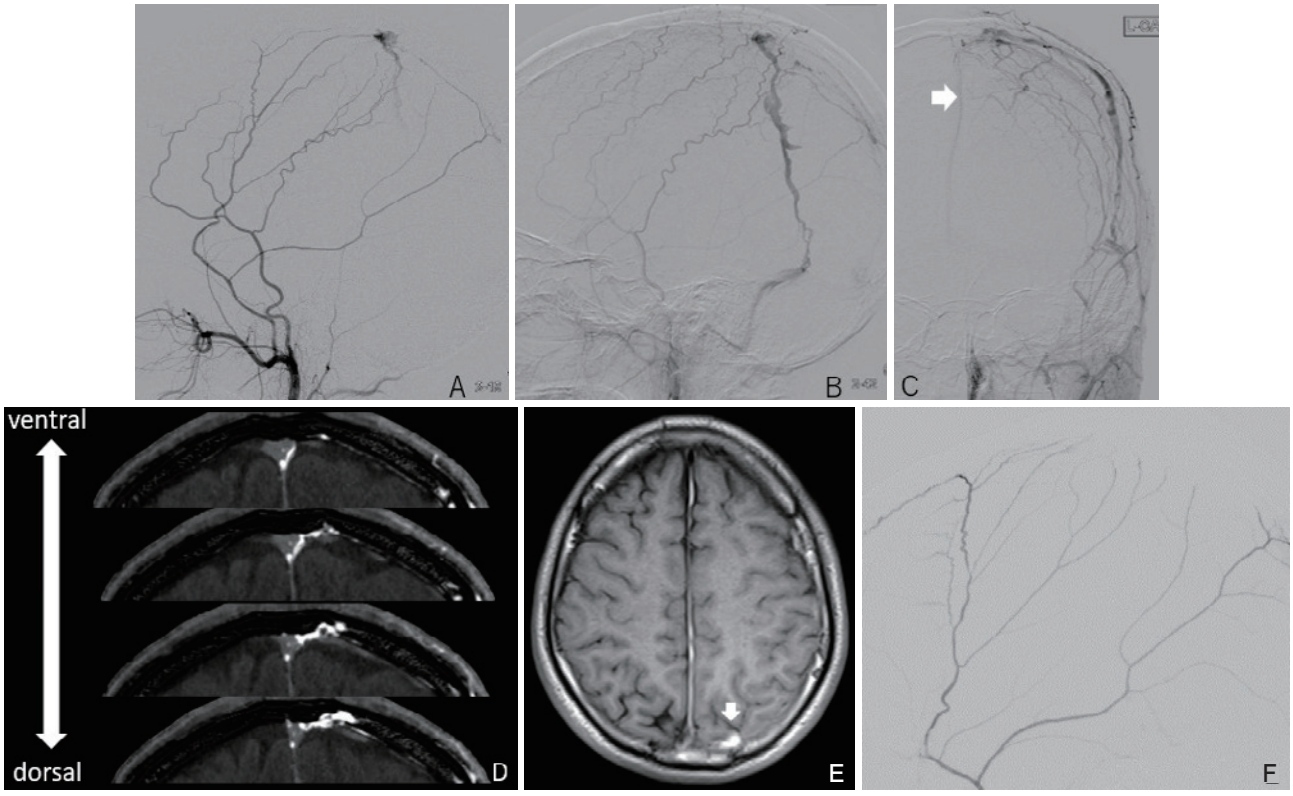

Fig. 2 The left external carotid angiogram shows the convexity dural arteriovenous fistula fed by the middle meningeal artery and draining into the transverse sinus via a diploic vein (A, B; lateral view). Some shunt blood flows into the superior sagittal sinus (SSS) (arrow) (C; anteroposterior view). Angiographic multi-planar reconstruction coronal images show that the shunt blood flows into the SSS via a dilated meningeal vein without involving a cortical vein (D). The thickest part of the hematoma coincided with the dilated meningeal vein on T1-weighted magnetic resonance imaging (arrow) (E). The arteriovenous fistula was occluded by two coils and 20% n-butyl-2-cyanoacrylate (F).

vein (Fig. 2B). Part of the shunt blood flowed into the superior sagittal sinus (SSS) and returned anterogradely (Fig. 2C). There was no CVR or cerebral venous stasis suggesting cerebral venous hypertension. There was also no stenosis or thrombosis observed in the dural sinuses. Angiographic multi-planar reconstruction (MPR) images showed that the shunt blood flowed into the SSS via the meningeal vein without involving the cortical vein (Fig. 2 D). The location of the dilated meningeal vein coincided with the thickest part of the hematoma, which was consid-

Table 1 Non-traumatic dural arteriovenous fistula without cortical venous reflux presenting with pure acute subdural hematoma

Author (years)	Age	Sex	Symptom	Side	Hemorrhagic phase	Findings on angiography			Cognard classification	Venous hypertension	DAVF Treatment	Outcome	
						Location	Feeding arteries	Draining veins				Radiographical	Clinical
Halbach (1988)	48	F	Weakness	R	N/A	SSS	bilateral MMA	SSS	N/A	N/A	Direct puncture of MMA, and TAE with IBCA	CO	good
Kohyama (2009)	60	M	Headache	L	Acute to chronic	L parietal convexity	bilateral MMA	SSS, PP	I	none	TAE with NBCA	CO	good
Ogawa (2010)	27	M	Headache	L	Acute	L parietal convexity	L OA	DV, MV	I	N/A	Resection	N/A	good
Kim (2016)	67	M	Headache	L	Chronic	L TS sinus	L MMA	TS sinus	I	N/A	TAE with PVA	CO	good
Yamauchi (2019)	29	M	Headache, nausea	L	Acute	L parietal convexity	L MMA	DV to SSS, sphenoparietal sinus	I	N/A	TAE with NBCA	CO	good
Tabibian (2021)	66	M	Headache, gait instability	R	Chronic	R parietal convexity	R MMA	MV to SSS	I	N/A	TAE with NBCA	CO	good
Present case	19	M	Headache	L	Acute	L parietal convexity	L MMA	DV to SS, MV to SSS	I	none	TAE with coil & NBCA	CO	good

CO, complete obliteration; DAVF, dural arteriovenous fistula; DV, diploic vein; F, female; M, male; IBCA, isobutyl-2-cyanoacrylate; L, left; MMA, middle meningeal artery; MV; meningeal vein; N/A, not available; NBCA, n-butyl-2-cyanoacrylate; PP, pterygoid venous plexus; PVA, polyvinyl alcohol; R, right; SS, sigmoid sinus; SSS, superior sagittal sinus; TAE, transarterial embolization; TS, transverse sigmoid

ered to be the source of bleeding (Fig. 2E). We diagnosed the patient with left convexity DAVF, classified as Borden type I and Cognard type I. The DAVF was considered the bleeding source of the ASDH. Therefore, transarterial embolization (TAE) was performed.

Under general anesthesia, a 6 Fr long sheath was inserted into the right femoral artery, and a 6 Fr Fubuki guiding catheter (Asahi Intecc, Aichi, Japan) was navigated into the left external carotid artery. A 3.2 Fr GuidePost (Tokai Medical Products Inc., Aichi, Japan) was guided to the left middle meningeal artery and placed at the foramen spinosum, and a Marathon (Medtronic, Minneapolis, MN, USA) was guided across the shunt point to the pouch of the diploic vein. Two coils were placed in the pouch, and 20% n-butyl-2-cyanoacrylate was injected to occlude the shunt (Fig. 2F). No SSS or cerebral vein stasis was observed.

The patient recovered well from general anesthesia and showed no new neurological symptoms. Three days after the intervention, the patient was doing well and was discharged. Cerebral angiography 1 year later showed no recurrence.

Discussion

This case led to two important conclusions. First, a DAVF without CVR can cause hemorrhage. Aggressive DAVFs are characterized by neurological symptoms and hemorrhagic changes, and CVR has been reported as a risk factor for the aggressive type, especially concerning hemorrhagic complications.[2,3] Another aggressive manifestation occurring without CVR is venous hypertension caused by a thrombus or stenosis of the dural sinus, resulting in venous infarction and hemorrhagic changes.[5] The convexity DAVF is usually classified as a nonsinus type, and the arterial blood usually flows directly into the cortical veins, resulting in CVR. The frequency of the aggressive type of convexity DAVF is high (52%).[1] In this case, the cortical vein or findings suggesting venous hypertension was not involved, and the patient developed a pure ASDH.

Second, a pure subdural hematoma may occur as a hemorrhagic complication of the DAVF. Regarding the hemorrhagic changes in the DAVF, a subdural hematoma is often associated with intracerebral and subarachnoid hemorrhages and rarely occurs alone.[4] Seven cases of DAVF complicated by a pure subdural hematoma have been reported, four of which were classified as convexity DAVF (Table 1).[6-11] The hemodynamic description of the present case was Cognard type 1, without CVR or venous hypertension. The hemorrhagic change was observed in the acute and chronic phases. Yamauchi *et al.* reported a case of an ASDH with a bleeding point on the inner surface of the dura detected after removing the hematoma after TAE. They considered that bleeding was caused by

meningeal vein dilatation, possibly obstructing another drainage route, as indicated by elevated d-dimer and fibrin degradation product (FDP) levels, suggesting thrombus formation.[10] Cortical veins generally drain into the SSS via bridging veins. Moreover, the veins of the falx cerebri and the meningeal veins, with traffic with diploic and emissary veins, also flow into the SSS. Other cortical veins join the meningeal veins in the dura matter 0.5-3.0 cm lateral to the SSS. The meningeal veins typically run on the inner surface of bone in an intradiploic or an intradural course.[12] In the present case, the possibility of obstruction in other drainage routes due to the hemorrhage cannot be ruled out due to the lack of measurement of d-dimer or FDP, and pathological examination was not performed. Angiographic MPR revealed that the meningeal vein draining into the SSS ran along the inner surface of the dura mater, suggesting meningeal vein dilatation, and the thickest part of the hematoma coincided with the dilated meningeal vein. Therefore, we speculated that the dilated meningeal vein was the source of bleeding.

The DAVF treatment can be categorized into direct surgery, endovascular therapy, and radiotherapy. Radiotherapy was inappropriate in this case because the patient had an intracranial hemorrhage requiring immediate shunt occlusion. Suppose intracranial hypertension occurred due to ASDH, resecting the dura mater may be considered along with hematoma removal; however, since headache was the only symptom, we chose endovascular treatment over direct surgery. When considering transvenous embolization, we anticipated difficulty advancing the microcatheter through the diploic vein. As the middle meningeal artery was not tortuous, the microcatheter could be guided to the shunt point, and the liquid embolization material could be used safely; therefore, TAE was selected. Coils were placed in the venous pouch to prevent the liquid embolization material from refluxing into the SSS via the meningeal vein. This coil-assisted TAE technique may be useful when a microcatheter can cross the shunt point and there is a migration risk of liquid embolization material.

Conclusion

A convexity DAVF without CVR or venous hypertension may cause a pure ASDH. The bleeding mechanism may differ from that of the well-known type resulting in hemorrhagic manifestations. In the absence of trauma, an ASDH requires an appropriate evaluation of the vascular lesions and a treatment plan.

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Abbreviations

DAVF: dural arteriovenous fistula
CVR: cortical venous reflux
ASDH: acute subdural hematoma
MRI: magnetic resonance imaging
CT: computed tomography
SSS: superior sagittal sinus
MPR: multi-planar reconstruction
TAE: transarterial embolization
FDP: fibrin degradation product

Author Contributions

JT conceived the manuscript idea and drafted the manuscript. SP and FO contributed to the manuscript idea and revised the manuscript. YT and YM contributed to the acquisition and interpretation of clinical information. All authors have read and approved the final version of the manuscript.

Informed Consent

Informed consent was obtained from the patient and his guardian to publish this case report and any accompanying images.

Conflicts of Interest Disclosure

The authors declare no conflicts of interest.

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