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CASE STUDY

Hemorrhagic Dural Arteriovenous Fistula with A Single Shunt Point on the Lateral Wall of the Cavernous Sinus: A Case Report and Literature Review

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Abstract We present a challenging case of a 52-year-old male with a hemorrhagic cavernous sinus lateral wall dural arteriovenous fistula (DAVF). The patient suffered a sudden consciousness disturbance, and imaging revealed a left temporal subcortical hematoma. Cerebral angiography unveiled a simple cavernous sinus DAVF with a single shunting point on the lateral wall. Initial attempts at trans-arterial embolization failed due to the inaccessibility of the shunt point. Subsequent rebleeding necessitated emergency direct surgery, involving disconnection of the venous drainage and hematoma removal. The draining vein was disconnected proximal to the varix due to the challenging accessibility of the origin of the draining vein. Post-surgery, angiography on day 17 revealed new shunt flow into the superior sagittal sinus, leading to a secondary direct occlusion of the draining vein just distal to the shunt point. Fortunately, the patient recovered without recurrence. Cavernous sinus DAVF with a single shunt and feeding artery is exceedingly rare. Our case underscores the complexity of treatment, requiring meticulous follow-up to prevent shunt flow recurrence, even in seemingly uncomplicated cases. Careful management post-treatment is essential to ensure a successful outcome.

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Key words: dural arteriovenous fistula; inferolateral trunk of the internal carotid artery; varix; lateral wall of the cavernous sinus.

Introduction

Cavernous sinus dural arteriovenous fistula (DAVF) is an abnormal connection between the internal carotid artery (ICA) and/or external carotid artery (ECA), and venous systems within the dura mater of the cavernous sinus. In Japan, cavernous sinus DAVF is the most common type of DAVF (25.8%)[1)]. Many cases of cavernous sinus DAVF have single or multiple shunt points at the intra-cavernous sinus, which are fed by multiple internal or external carotid arteries of the dural branches and drain into the superior ophthalmic vein, inferior petrosal sinus, superficial middle cerebral vein (SMCV), and intra-cavernous sinus connecting to the contralateral side of the cavernous sinus.

Cavernous sinus DAVF with a shunt point located at the lateral wall only is rare, with four cases being reported to date to the best of our knowledge[2-5)]. Three out of the four patients developed internal cerebral hemorrhage and deteriorated. We herein report a case of hemorrhagic cavernous sinus DAVF. An initial examination showed that it had a simple shunt point on the lateral wall of the cavernous sinus that was only fed by the inferolateral trunk (ILT) of the left ICA and refluxed into the SMCV with a ruptured varix. Trans-venous embolization was difficult and trans-arterial

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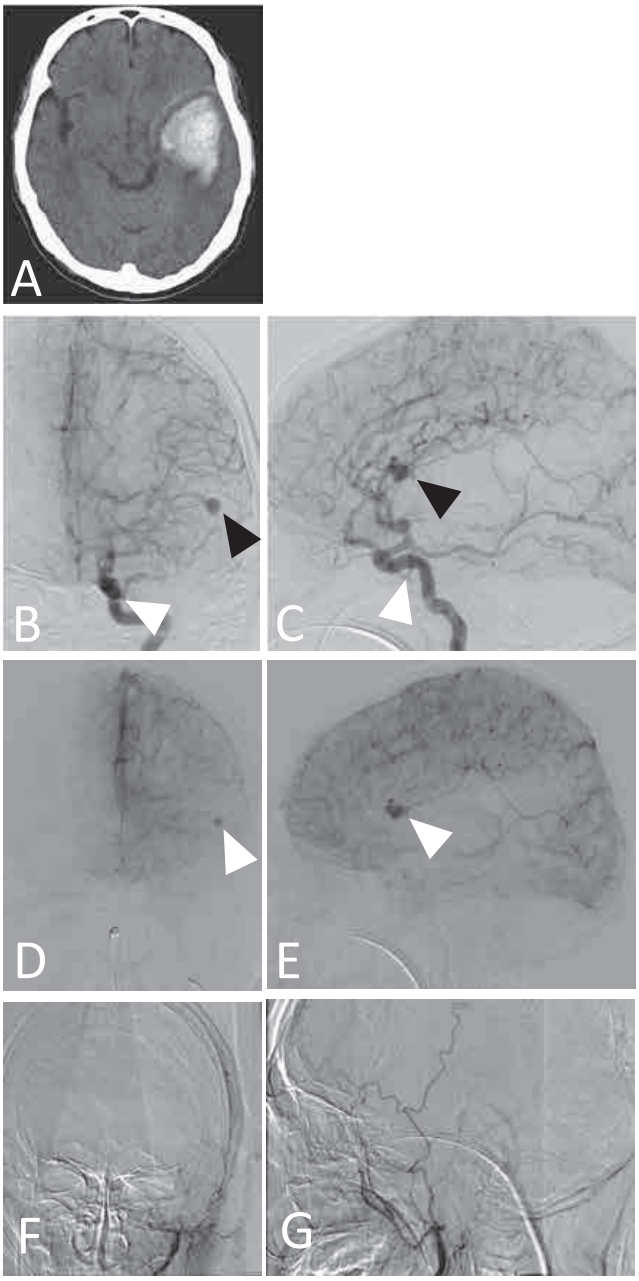

Figure 1 (A) Computed tomography (CT) image before attempts intravascular surgery showing a left temporal-parietal subcortical hematoma. (B~E) Cerebral angiography reveals that a dural arteriovenous fistula (DAVF) is fed by the left inferolateral trunk (ILT) of the left internal carotid artery (white arrows) and is draining into the left sphenoparietal sinus with reflux into the left superficial middle cerebral vein and a varix (black arrows), suggesting Borden type III (B, C: frontal views, D, E: lateral views). (F, G) Cerebral angiography reveals that DAVF is not fed by external carotid artery (F: frontal view, G: lateral view).

embolization was not accomplished. We emergently treated the patient by directly disconnecting the draining veins twice. We present the clinical course of the present case and difficulties associated with curative treatment as well as a literature review.

Case Report

A 52-year-old male with no history of head

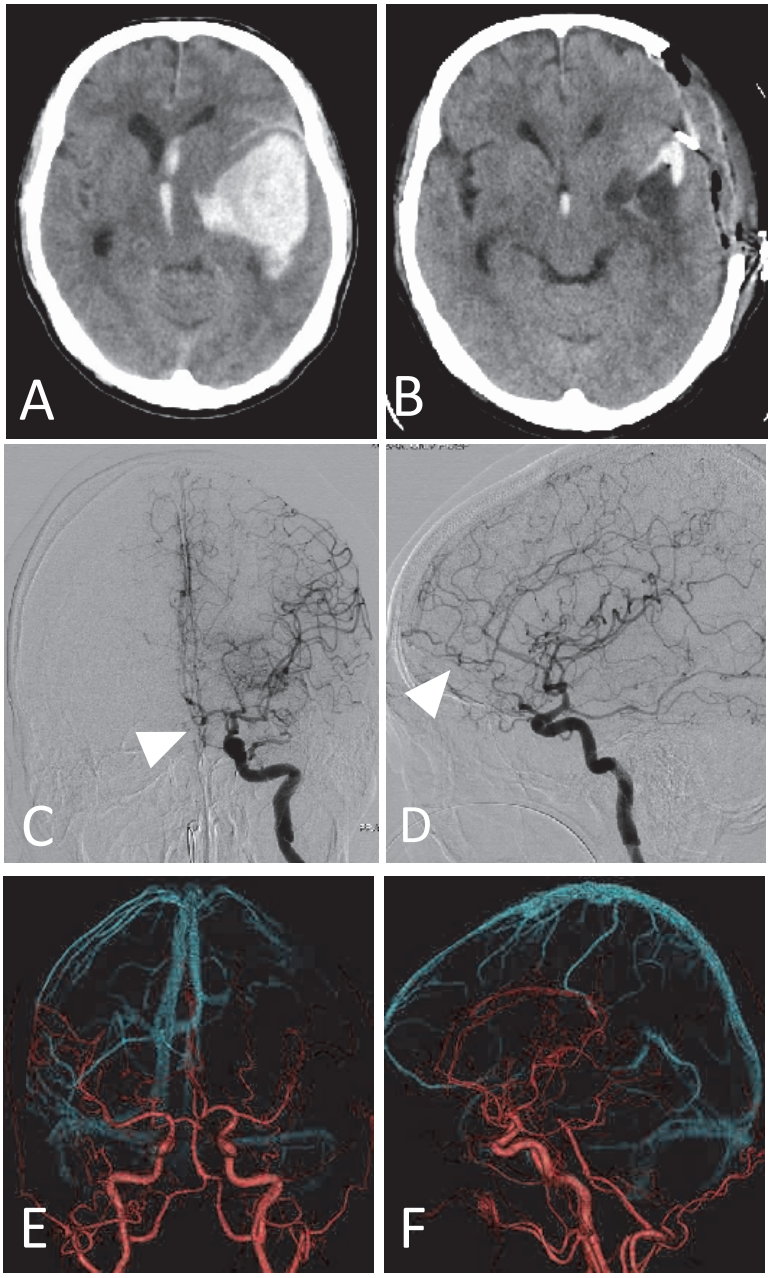

Figure 2 (A) Computed tomography image after attempts intravascular surgery showing the enlargement of the hematoma. (B) Computed tomography image after microsurgery showing that the temporal hematoma is evacuated and clipped. (C, D) Cerebral angiography 17 days after the first microsurgery shows a venous reflux to SMCV is remaining and a new drainage root to the superior sagittal sinus (white arrow). (E, F) Computed tomography image two years after the second microsurgery showing that the DAVF disappeared and there is no recurrence.

trauma was transferred to the emergency room with right hemiplegia, sensory aphasia and consciousness disturbance (Glasgow Coma Scale (GCS) 9). He had no ocular symptoms (diplopia, proptosis, and exophthalmos) before onset. Computed tomography (CT) showed a left temporal-parietal subcortical hematoma (Fig.1A). Cerebral angiography revealed that DAVF was fed by the left ILT of the left ICA, none ECA and drained into the left sphenoparietal sinus with

Table .

	Sex	Age	Feeder	Symptom	Shunt point	Drainer	Therapy
Inui et al	F	44	C4 portion of ICA	None	CS	Sylvian vein	Microsurgery
Kim et al	M	58	AMA ILT of ICA MMA Recurrent artery of the ophthalmic artery	ICH	CS	Sylvian vein BVR	TAE + microsurgery
Ushikoshi et al	M	76	ILT of ICA Recurrent artery of the ophthalmic artery	Subarachnoid hemorrhage	CS	Sylvian vein	Microsurgery
Uchiyama et al	M	40	ILT of ICA	ICH	CS	Sylvian vein	Microsurgery
Present case	M	52	ILT of ICA	ICH	CS	Sylvian vein	Microsurgery

BVR: basal vein of Rosenthal, CS: cavernous sinus, ICH: internal cerebral hematoma, ILT: inferolateral trunk, TAE: trans arterial embolization

reflux to the left SMCV and a varix, suggesting Borden type III (Fig.1B-G). An endovascular approach via the inferior petrosal sinus was difficult for trans-venous embolization and the ILT of the left ICA was the only feeding artery that was sufficiently wide for embolization. We selected a trans-arterial approach via the left ILT of the left ICA. However, the catheter was unable to reach the shunt point on the left wall of the cavernous sinus because the ILT of the left ICA was winding and narrow. We changed our strategy and discontinued the procedure. His consciousness deteriorated to GCS 7 just after endovascular therapy. CT images showed the enlargement of the hematoma (Fig.2A). We emergently planned venous disconnection of the draining vein of the left SMCV and removal of the hematoma. We performed front-temporal craniotomy and observed the varix of the SMCV. We exposed the SMCV from the varix and clipped the ruptured point of the varix. Difficulties were associated with exposing the exit of the draining vein from the shunting point at the lateral wall of the cavernous sinus and thrombosis of the single draining vein was expected after distal occlusion. The post-operative course was uneventful and CT one day after surgery showed the evacuation of the hematoma (Fig.2B). Follow-up digital subtraction angiography 17 days after the initial surgery showed the disappearance of the varix and drainage route to the left SMCV remained; however, a new drainage route of the cavernous DAVF draining into the superior sigmoid sinus appeared (Fig.2C, D). In the second direct surgery, the outflow vein was occluded at the exact point where the SMCV and the SSS drain from the shunt point. No recurrence was observed in the follow-up examination 2 years after surgery (Fig.2E, F) and neurological deficits gradually recovered, he still has very mild sensory aphasia but lives his life in a wheelchair.

Discussion

Most cavernous sinus DAVF have multiple venous drainage patterns with the most common being the reversal of flow into the anterior cavernous sinus and ophthalmic vein, followed by that into the inferior petrosal sinus[6]. Cavernous sinus DAVF with a shunt point at the lateral wall only is very rare and only four cases have been reported to date to the best of our knowledge[2-5] (Table). Surgical clipping was selected for all four cases, including the present case, and endovascular treatment was initially attempted in some cases[2, 3]. Inui et al. used glue

to embolize multiple feeders via the left middle meningeal artery[2]. In the case reported by Kim et al., the ILT of the left ICA only fed diffusely and trans-arterial embolization via the ILT of the ICA was not accomplished due to the difficulty of the approach. Most cases of cavernous sinus DAVF develop eye symptoms (pulsating exophthalmos, conjunctival chemosis, and tinnitus)[1]. However, none of four patients with DAVF in the lateral wall of cavernous sinus had eye symptoms because they did not have a drainage root to the superior orbital vein and inferior orbital vein. Three of these four patients had only one or a few drainage roots to the SMCV with high venous pressure and a ruptured varix.

The ILT of the ICA frequently originates from the C4 portion of the ICA and has three branches, with the anterior branch supplying the cavernous area[7]. Cases of DAVF fed by only the ILT of the ICA and treated by a trans-arterial approach[8, 9] or trans-venous approach[10] have been reported. However, in some cases of DAVF fed by the ILT of the ICA, catheters were unable to reach the shunt point via the ILT of the internal cerebral artery because it was very narrow, bent, and winding. Xu et al. showed that an arterial approach may be selected as the primary treatment plan when the blood-supplying artery and the fistula are relatively singular and when the microcatheter may easily reach close to the fistula through the artery[11]. Although the present patient had a single feeder of the ILT of the ICA, it was tortuous and catheters were unable to reach the shunt point; therefore, we performed craniotomy[12].

The appearance of a new drainage root from the cavernous sinus has been reported in cases of cavernous sinus DAVF treated by microsurgery or the trans-arterial approach. Xu et al. showed the recurrence of a new drainer to the anterior condylar confluence in one out of eight cases of cavernous sinus DAVF treated by the trans-arterial approach[11]. Zhang et al. demonstrated that cavernous sinus DAVF was not completely obligated in two out of eight cases treated by the trans-arterial approach[12]. Kuwayama et al. described clipping of the distal draining vein, which may have resulted in the opening of other small draining channels that were not observed on angiography[13]. In the present case, we directly occluded venous drainage with a clip at the distal side apart from the shunt point, which may have contributed to the appearance of a cryptic drainage root. A temporal subcortical hematoma may compress other draining veins and it appeared after removal of the hematoma.

We encountered a rare case of cavernous sinus DAVF with a shunting point on the lateral wall that was ultimately treated with craniotomy and the direct disconnection of venous drainage. The first direct occlusion of the draining vein via craniotomy at the distal portion of the single draining vein was not sufficient and, thus, additional direct occlusion was required. A careful follow-up is important for avoiding the recurrence of shunt flows even after the treatment of cavernous sinus DAVF without a complicated vasculature.

Conflicts of Interest: The authors report there are no competing interests to declare.

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Abbreviation List

CT: computed tomography
ECA: external carotid artery
GCS: Glasgow coma scale
DAVF: dural arteriovenous fistula
ICA: internal carotid artery
ILT: inferolateral trunk
SMCV: superficial middle cerebral vein

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