

**Recurrence of the Cavernous Sinus Dural Arteriovenous Fistula at Adjacent Sinuses**

**Following Repeated Transvenous Embolizations: Case Report and Literature Review**

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## **Abstract**

We present a unique case of a cavernous sinus (CS) dural arteriovenous fistula (DAVF), which recurred at adjacent sinuses following repeated transvenous embolizations (TVEs).

A 68-year-old woman presented progressive left conjunctival chemosis and diplopia. Cerebral angiography revealed a left CS DAVF, which was completely obliterated by TVE via the left inferior petrosal sinus (IPS). Two years later, the DAVF recurred in the left IPS, and in the left sigmoid sinus (SS) three years after the initial treatment in spite of second TVE. Moreover, the left SS and the left internal jugular vein, which had been previously stenotic, have been occluded. The third TVE resulted in the complete obliteration of the SS DAVF.

CS DAVFs may recur at adjacent sinuses even after complete obliteration by TVE. Careful follow-up is necessary to check for the recurrence of DAVFs especially in cases with venous flow changes, such as sinus occlusion, following endovascular treatments.

*Key words:* new dural arteriovenous fistula, recurrence, transvenous embolization, venous flow changes

## **INTRODUCTION**

Dural arteriovenous fistulas (DAVFs) comprise 10-15% of intracranial arteriovenous malformations.<sup>1</sup> Among them, multiple DAVFs have an incidence of 7%.<sup>2</sup> Although a DAVF is generally thought to be an acquired lesion, its pathogenesis has not yet been established. The treatment strategy is based on angiographic features including feeding arteries and venous drainage, location of the fistula, severity of presenting symptoms, and patients' condition.<sup>3</sup> At present, transvenous embolization (TVE) has been widely accepted as a useful treatment for DAVFs.<sup>4</sup> Anatomical obliteration rates with TVE have been reported to range from 63% to 87.5%, and the clinical cure rates from 83% to 96%.<sup>5-10</sup> However, some reports have demonstrated an appearance of a new DAVF following endovascular treatment.<sup>4, 11-16</sup>

We herein present a case of a cavernous sinus (CS) DAVF which recurred in the ipsilateral inferior petrosal sinus (IPS) and then in the sigmoid sinus (SS) in spite of repeated TVEs, and discuss the mechanism of a recurrence of the DAVF.

## **CASE REPORT**

A 68-year-old woman was admitted to our hospital because of a 6-month history of progressive left conjunctival chemosis and diplopia. The patient did not have any history of head injury, intracranial surgery, or intake of female hormones. Cerebral angiography and

magnetic resonance (MR) 3-dimensional spoiled gradient-recalled acquisition (3D-SPGR) images showed a left CS DAVF, which was fed by the bilateral internal and the external carotid arteries (Barrow type D; Fig. 1A-C). It drained into the left superior ophthalmic vein (SOV) and fairly into the left paravertebral venous plexus via the left IPS. Under general anesthesia, TVE was performed from the right femoral vein. A 6-French guiding catheter (Guider; Boston Scientific) was introduced to the left internal jugular vein, and a microcatheter (Navigator IVa; Kaneka medics) was inserted via the left IPS. The whole compartments of the CS were embolized following selective occlusion of the SOV using Guglielmi detachable coils (GDC), interlocking detachable coils, and fibered platinum coils (Vortex). Total number of coils was 22. The DAVF was completely obliterated without changing venous flow patterns (Fig. 1D-F). The patient's clinical symptoms were markedly improved, and was discharged without any neurological deficits.

Two years after the initial treatment, the patient presented progressive left tinnitus. Cerebral angiography and MR 3D-SPGR images showed a recurrence of the DAVF in the left IPS (Fig. 2A-C). She underwent a second TVE because of progressive symptoms. The left IPS was embolized using GDC (total number; 7), and the DAVF disappeared completely (Fig. 2D). At the end of the procedure, the venous flow through the left TS-SS was found to be much slower than before (Fig. 2E).

Three years after the first TVE, the patient again presented progressive left tinnitus. Cerebral angiography showed a recurrence of the DAVF in the left SS (Fig. 3A and B) without reflux of cortical veins (Cognard type IIa). Moreover, the left SS and the left jugular vein, which had been previously stenotic have been occluded (Fig. 3C). A third TVE was undertaken for the left SS DAVF via the right internal jugular vein using detachable coils (GDC and Trufill DCS), which resulted in a complete obliteration (Fig. 3D). There has been no recurrence of the DAVF during two-year follow-up after the third treatment.

## **DISCUSSION**

A variety of modalities have been applied to treat DAVFs, including endovascular procedures,<sup>9, 17, 18</sup> direct sinus packing,<sup>19</sup> surgical interruption of the draining veins,<sup>19</sup> gamma knife surgery,<sup>20</sup> and combinations of these treatments.<sup>21, 22</sup> Among them, TVE has been widely accepted as an effective treatment.<sup>5-10, 23</sup> However, seven cases demonstrating a new DAVF following endovascular treatments have been reported (Table).<sup>4, 11-16</sup> In these DAVFs, new lesions occurred in either the ipsilateral TS-SS or the jugular vein. TVE was performed in four cases, TVE with transarterial embolization (TAE) in two, and TAE in one, respectively. Catheterization to the IPS was performed in six cases. It is noteworthy that sinus occlusion after the treatment occurred in two cases. Nakagawa et al. showed a case of a CS DAVF

which shifted to the TS-SS after partial embolization of the SS combined with ligation of the internal jugular vein,<sup>15</sup> suggesting that the venous flow change could induce a new DAVF. Friedman et al. reported a case of posttraumatic DAVF with a fatal progression following multimodal treatments, partially because the initial treatment failed to completely obliterate the fistula.<sup>24</sup> Our case is so unique that the CS DAVF recurred gradually and posteriorly following endovascular treatments.

The mechanisms of newly formed DAVFs following endovascular treatments remains unclear. There have been several theories reported, including venous hypertension,<sup>25-27</sup> a microcatheter and/or microguidewire manipulation,<sup>4</sup> maturation of an angiographically occult DAVFs,<sup>16</sup> and sinus thrombosis.<sup>4</sup> Recent studies have focused on the molecular mechanisms for the pathoetiology, and some clinical series have demonstrated that high levels of angiogenic growth factors, such as the vascular endothelial growth factor and the basic fibroblast growth factor, might be involved in the formation of DAVFs.<sup>27, 28</sup>

In the present case, additional fistulas may have developed in response to humoral factors produced by either the endovascular manipulations and/or the presence of the occult fistula. Changes in the arterial and/or venous flow patterns, or development of sinus thrombosis after the treatment could affect new DAVFs formation. Decreased local arterial pressure may keep the dural arteriovenous shunts dormant,<sup>27, 28</sup> while the increased pressure after endovascular

treatments could induce a new fistula. In our case, the left CS DAVF drained into the ipsilateral paravertebral venous plexus. Venous hypertension due to stenotic change of the left internal jugular vein could have affected the recurrence of the DAVF. In addition, the endovascular manipulation might have changed venous flow patterns. Considering the recurrence in the left IPS, the fistulas might therefore be incompletely embolized during the first TVE despite the angiographically complete obliteration. Because there is little evidence regarding the pathogenesis for the recurrence of DAVFs, a further accumulation of cases is necessary. We therefore insist that careful follow-up is mandatory to check for the recurrence of CS DAVFs especially in cases with venous flow changes, such as sinus occlusion, following endovascular treatments.

In conclusion, we presented a case of a CS DAVF recurring posteriorly even after the complete obliteration by TVE. Changes in venous flow patterns following endovascular treatments would be a risk factor for the recurrence of DAVFs.

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## Figure Legends

**Table:** The characteristics of the reported cases presenting with a new DAVF following endovascular treatment of the CS DAVF

**Figure 1:** Lateral view of both left carotid angiography (A) and external carotid angiography (B), showed a left cavernous sinus (CS) dural arteriovenous fistula (DAVF) (black arrow), draining into the left superior ophthalmic vein and into the left paravertebral venous plexus via the left inferior petrosal sinus. Axial images of magnetic resonance 3-dimensional spoiled gradient-recalled acquisition (C) show the shunt at the posterior part of the left CS. The DAVF completely disappeared after transvenous embolization (D and F) without changing venous flow pattern (F).

**Figure 2:** Two years after the initial treatment, follow-up left carotid angiography (A; lateral view), external carotid angiography (B; lateral view) and an axial image of magnetic resonance 3-dimensional spoiled gradient-recalled acquisition (C) show a recurrence of the dural arteriovenous fistula (DAVF) (black arrow) in the left inferior petrosal sinus. The DAVF completely disappeared after the second transvenous embolization (D and F). The venous flow through the left TS-SS is much slower than that at the first TVE (E).

**Figure 3:** Three years after the initial treatment, lateral view of both left carotid

angiography (A) and external carotid angiography (B) show a new dural arteriovenous fistula (DAVF) (black arrow) in the left sigmoid sinus. The left sigmoid sinus and the jugular vein are occluded (C). The DAVF completely disappeared after the third transvenous embolization (D).

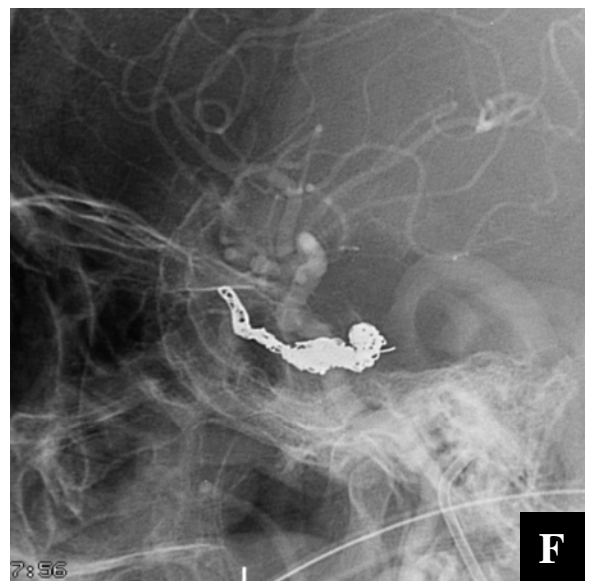
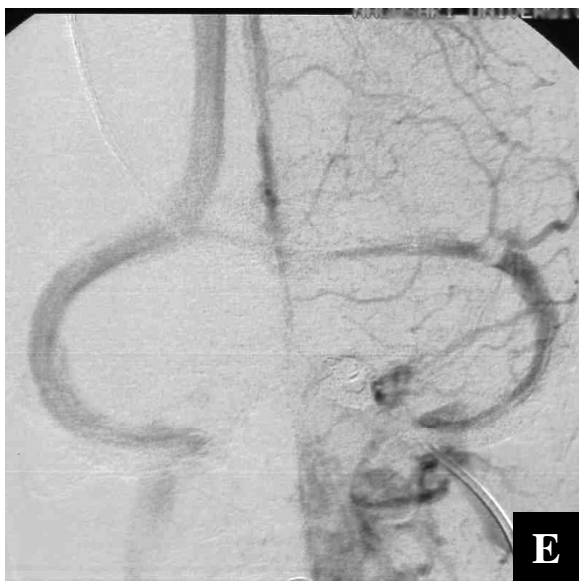
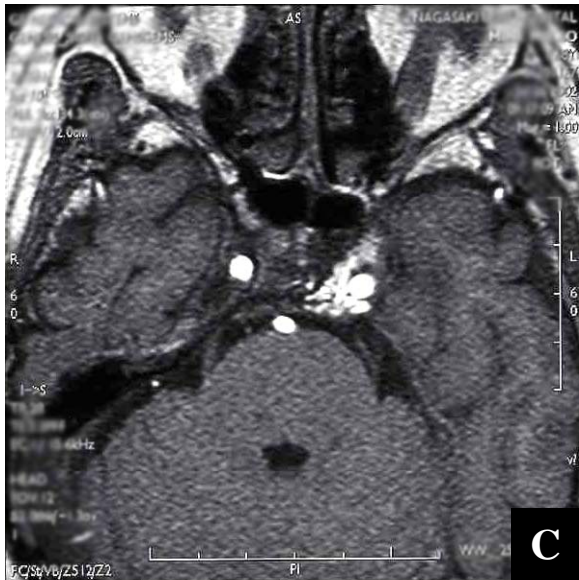
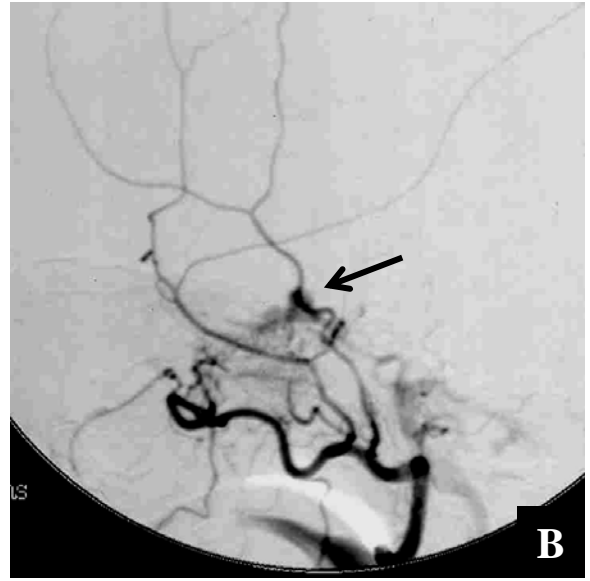


Fig. 1

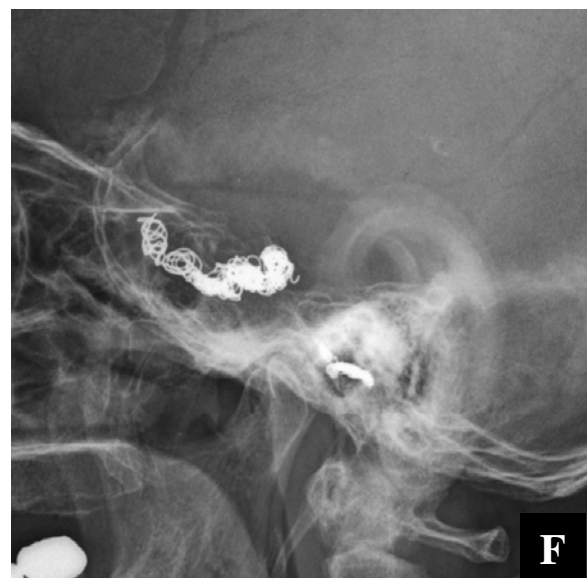
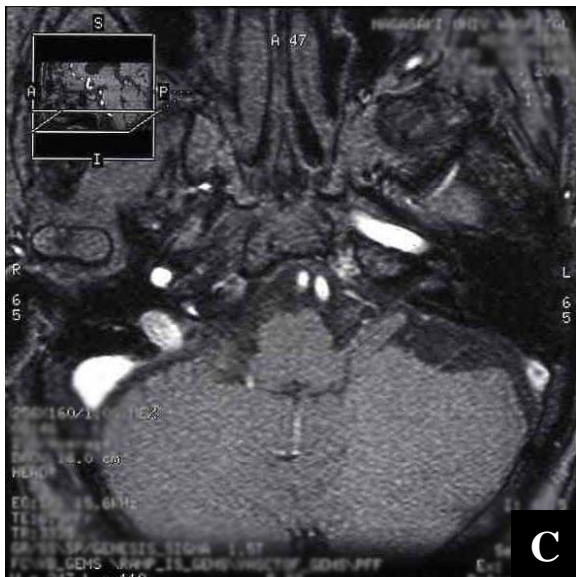
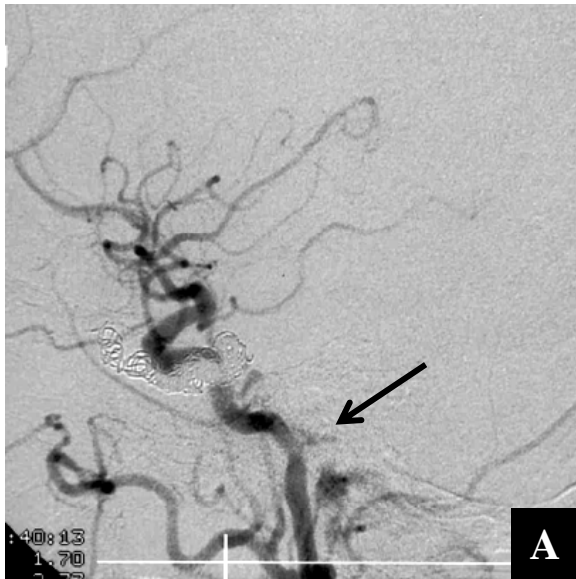


Fig. 2



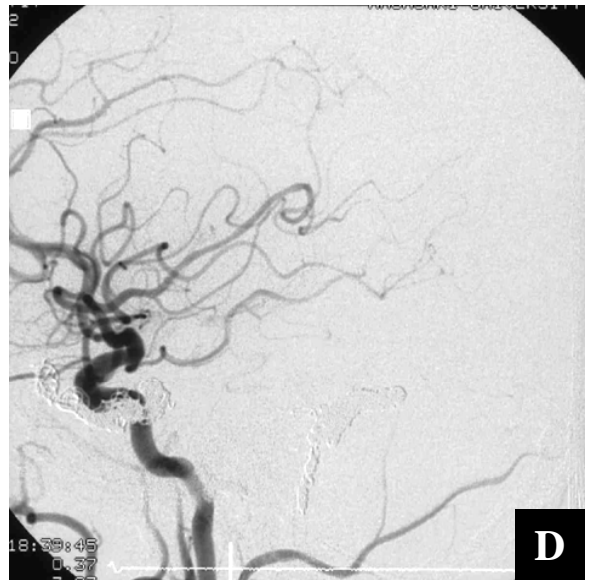
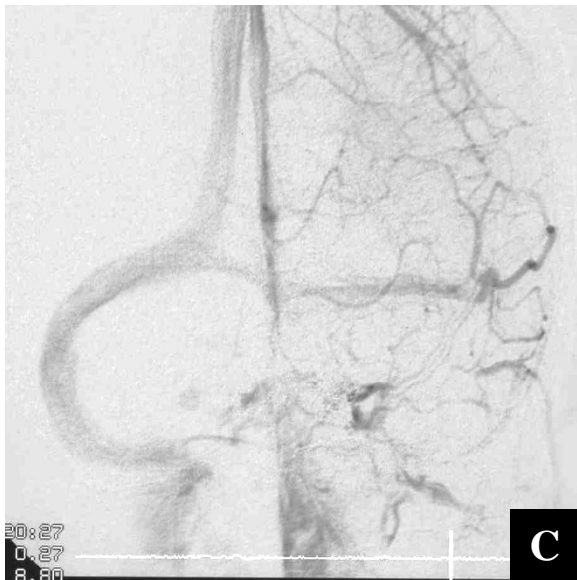
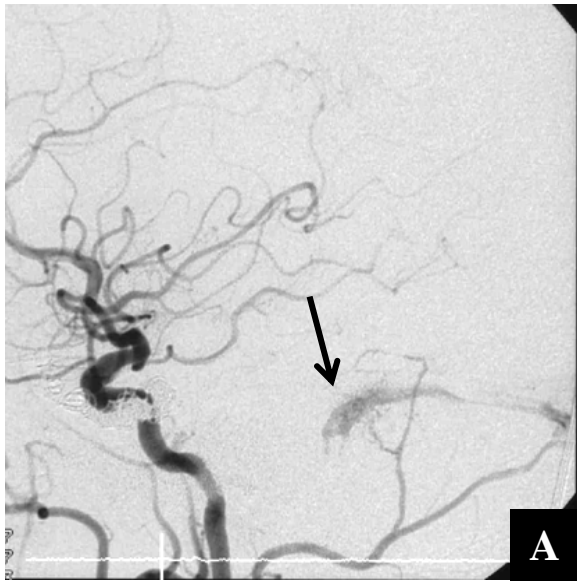


Fig. 3

**Table. The characteristics of the reported cases presenting with a new DAVF following endovascular treatment of the CS DAVF**

No.	Authors (year)	Age, sex	Original location	Drainage	Treatment	Occlusion	Catheterization to the IPS	Interval (months)	Location of new DAVF	Change of venous flow pattern
1	Nakagawa et al. (1992) <sup>19)</sup>	43, F	Left CS	Bilateral SOV, IPS	TAE+TVE (via SOV)	Incomplete	+	4m	Left TS-SS	Occlusion of the left IJV
2	Yamashita et al. (1993) <sup>27)</sup>	54, F	Left CS	Left SOV and left IPS	TAE+TVE (via IPS)	Complete	+	12m	Left SS	—
3	Makiuchi et al. (1998) <sup>17)</sup>	43, M	Left CS	Left SOV	TVE (via SOV)	Complete	+	6m	Left SS	Not shown
4	Kubota et al. (1999) <sup>13)</sup>	43, F	Left CS	Left SOV	TVE (via SOV)	Complete	+	4m	Left jugular vein	Not shown
5	Kawaguchi et al. (1999) <sup>10)</sup>	72, F	Left CS	Left sphenoparietal sinus, left SOV, and left IPS	TVE (via IPS)	Complete	+	30m	Left TS	Occlusion of the left SS
6	Kiyosue et al. (2002) <sup>11)</sup>	66, F	Left CS	Left SOV	TVE (via SOV)	Complete	+	5m	Left jugular vein	—
7	Gupta et al. (2005) <sup>7)</sup>	59, F	Bilateral CS	Left SOV and left IPS	TAE (through fistula)	Complete	—	4m	Left SS	—
8	Present case	68, F	Left CS	Left SOV and left IPS	TVE (via IPS)	Complete	+	24m→ 36m	Left IPS→ Left TS-SS	Occlusion of the left SS and IJV

DAVF, dural arteriovenous fistula; CS, cavernous sinus; SOV, superior ophthalmic vein; IPS, inferior petrosal sinus; TAE, transarterial embolization; TVE, transvenous embolization; SS, sigmoid sinus; TS, transverse sinus; IJV, internal jugular vein.