

# **Brainstem Congestion Caused by Direct Carotid-Cavernous Fistula**

## **—Case Report—**

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### **Abstract**

A 41-year-old woman presented with tinnitus in the left ear and headache, followed by diplopia and pain in the left cheek. Angiography showed a left high-flow direct carotid-cavernous fistula (CCF), causing steal of the blood flow from the internal carotid artery into the cavernous sinus. A few days later, she rapidly developed right hemiparesis, dysarthria, and ocular conjugate deviation to the right, and became somnolent. Angiography at that time revealed occlusion of the superior petrosal sinus, causing engorgement of the veins in the surrounding brainstem. The CCF was completely embolized with interlocking detachable coils. Her consciousness disturbance and ophthalmoparesis dramatically improved within a few days, and the right hemiparesis and dysarthria gradually resolved. Magnetic resonance (MR) imaging after the treatment showed small pontine hemorrhage and perifocal edema but no ischemic lesions in the cerebral hemisphere. Re-evaluation of the MR imaging with gadolinium taken on admission demonstrated engorged veins in the brainstem parenchyma, which corresponded to the hemorrhagic lesion in the brainstem. Brainstem congestion caused by direct CCF is very rare, but it can be life-threatening. Good outcome can be expected if the CCF is completely occluded before congestive hemorrhage occurs.

Key words: carotid-cavernous fistula, venous congestion, brainstem

### **Introduction**

Life-threatening complications associated with carotid-cavernous fistulas (CCFs) include intracerebral hemorrhage, subarachnoid hemorrhage, epistaxis, and cerebral ischemia due to steal of blood flow by the CCF. We describe a case of brainstem congestion due to a direct-type CCF, which is an extremely rare complication.

### **Case Report**

A 41-year-old woman complained of tinnitus in the left ear and headache. Otolaryngological examination and magnetic resonance (MR) imaging in a general hospital revealed no abnormalities. Five days later, she developed diplopia and pain in the left cheek, and was admitted to our hospital. She had

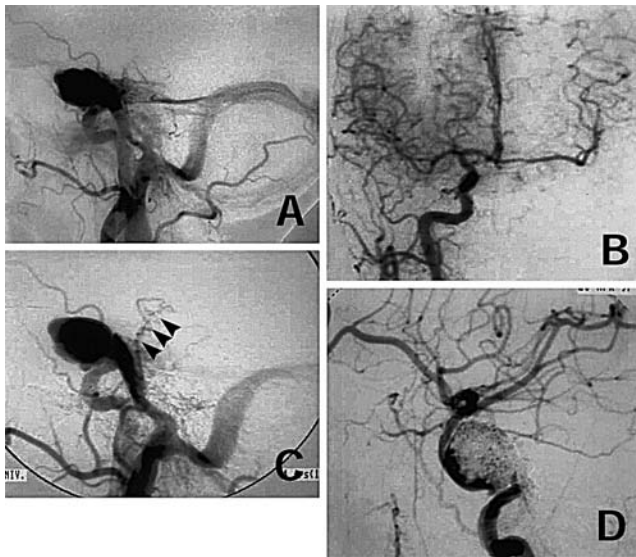
no history of head trauma.

Neurological examination revealed a bruit in the left orbit, left abducens nerve paralysis, and hypesthesia in the first and second divisions of the left trigeminal nerve. There was no chemosis or proptosis. MR angiography showed a disordered signal void surrounding an extension of the cavernous sinus. Angiography detected a left high-flow direct CCF, which caused steal of the blood flow from the internal carotid artery (ICA) into the superior petrosal sinus (SPS) and inferior petrosal sinus (IPS), resulting in cut off of the flow of the left middle cerebral artery (MCA) (Fig. 1A). The MCA was supplied by cross flow from the contralateral side through the anterior communicating artery (Fig. 1B). Single photon emission computed tomography with technetium-99m-hexamethylpropyleneamine oxime showed preservation of the cerebral blood flow of the left cerebral hemisphere.

Four days after admission, she rapidly developed right hemiparesis, dysarthria, and ocular conjugate

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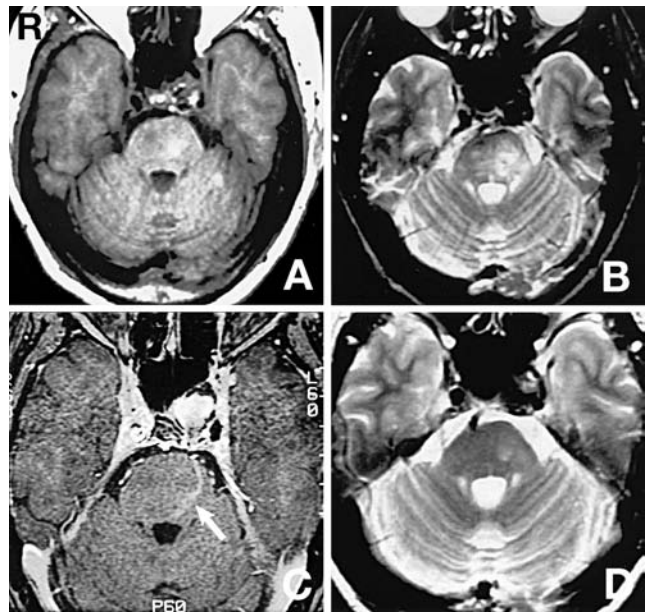
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**Fig. 1** A: Left carotid angiogram on admission showing a high-flow direct carotid-cavernous fistula (CCF) causing steal of the flow from the internal carotid artery (ICA) into the cavernous sinus, and no opacification of the middle cerebral artery (MCA). B: Right carotid angiogram on admission revealing opacification of the left MCA via the anterior communicating artery. C: Left carotid angiogram just before treatment demonstrating occlusion of the superior petrosal sinus, increased flow in the inferior petrosal sinus, and engorgement of the veins in the prepontine and interpeduncular cisterns (arrowheads). D: Left carotid angiogram after embolization showing obliteration of the CCF and complete opacification of the ICA and MCA.

deviation to the right, and became somnolent. We at first suspected that ischemia of the left cerebral hemisphere had developed due to the flow steal from the left ICA into CCF. However, computed tomography (CT) revealed no ischemic lesions in the left cerebral hemisphere. We could not accurately evaluate the brainstem lesions with CT because of bone artifacts. Next day, the CCF was embolized using 30 interlocking detachable coils. Left carotid angiography just before the procedure revealed that the flow dynamics of the CCF were little changed. However, spontaneous occlusion of the SPS had occurred, resulting in increased flow in the IPS and engorgement of the veins in the prepontine and interpeduncular cisterns (Fig. 1C).

Immediately after embolization, angiography showed that the CCF was completely obliterated and



**Fig. 2** A, B: T<sub>1</sub>-weighted (A) and T<sub>2</sub>-weighted (B) magnetic resonance (MR) images one week after the treatment showing small pontine hemorrhage and perifocal edema. C: Re-evaluation of the T<sub>1</sub>-weighted MR image with gadolinium-diethylenetriaminepenta-acetic acid taken on admission demonstrating engorgement of the perforating vein within the pontine parenchyma coinciding with the subsequent hemorrhage and edema (arrow). D: T<sub>2</sub>-weighted MR image 4 weeks after the treatment revealing marked reduction of the perifocal edema.

both the ICA and MCA were completely opacified (Fig. 1D). The left orbital bruit disappeared. Her consciousness disturbance and ophthalmoparesis dramatically improved within a few days, and the right hemiparesis and dysarthria gradually resolved. MR imaging one week after the treatment showed small pontine hemorrhage and perifocal edema but no ischemic lesions in the cerebral hemisphere (Fig. 2A, B).

Re-evaluation of the MR imaging with gadolinium-diethylenetriaminepenta-acetic acid (Gd-DTPA) taken on admission identified engorged veins in the brainstem parenchyma, which corresponded to the subsequent hemorrhagic and edematous lesion in the brainstem (Fig. 2C). Four weeks after the treatment, the right hemiparesis and dysarthria were almost completely resolved. MR imaging showed reduction of the brainstem edema (Fig. 2D). Three months later, she returned to her former office work.

**Table 1 Brainstem congestion caused by direct carotid-cavernous fistula**

Case No.	Author (Year)	Age, Sex	Location	Symptoms	Angiography		MR imaging with Gd-DTPA	Treatment	Outcome
					Occlusion of SPS	Engorged veins in the surrounding brainstem			
1	Turner et al. (1983) <sup>7)</sup>	81, F	midbrain	hemiparesis, comatose	NM	+	NT	—	death
2	Teng et al. (1991) <sup>6)</sup>	36, M	midbrain, thalamus	unconsciousness, hemiparesis	+	dilation of proximal SPS	NT	transarterial balloon embolization	no neurological signs
3		26, M	midbrain	numbness of the right side of the body and face	+	+	NT	transarterial balloon and coil embolization	no neurological signs
4	Present case	49, F	pons	hemiparesis, dysarthria, somnolence	+	+	engorged veins in pontine parenchyma	transarterial coil embolization	no neurological signs

Gd-DTPA: gadolinium-diethylenetriaminepenta-acetic acid, MR: magnetic resonance, NM: not mentioned, NT: not tested, SPS: superior petrosal sinus.

## Discussion

This patient presented with a high-flow direct CCF associated with pontine hemorrhage and edema. We first suspected that the flow steal from the left ICA into the cavernous sinus<sup>3)</sup> had caused ischemia of the left cerebral hemisphere when the patient developed hemiparesis and dysarthria, and subsequently became somnolent. However, she showed ocular conjugate deviation to the right, which can be caused by lesions in the right side of the pons, but not by lesions in the left cerebral hemisphere. Moreover, neither CT before the treatment nor MR imaging after the treatment had detected any ischemic lesions in the left hemisphere. MR imaging one week after embolization of the CCF detected small hemorrhage and perifocal edema in the brainstem (Fig. 2A, B). MR imaging with Gd-DTPA had previously showed engorged veins in the pontine parenchyma, which corresponded to the subsequent hemorrhage and edema (Fig. 2C). Angiography just before embolization had demonstrated spontaneous occlusion of the SPS, which is an essential part of the venous drainage system from the brainstem via the petrosal veins. Occlusion of the SPS had resulted in backward flow into and congestion of the veins in the prepontine and interpeduncular cisterns (Fig. 1C). Therefore, the small hemorrhage probably resulted from brainstem venous congestion due to the CCF.

Brainstem hemorrhage may occur after intravascular embolization or incomplete embolization of direct CCFs.<sup>2)</sup> However, the treatment did not cause hemorrhage in the present case because the critical

symptoms presented before and improved soon after the treatment.

Brainstem congestion caused by direct CCF has been reported in three cases, which all involved congestion in the midbrain (Table 1).<sup>6,7)</sup> In contrast, the present case had pontine hemorrhage associated with direct CCF. The important factors in the development of brainstem congestion are changes in the drainage pattern of the SPS and engorged veins in the surrounding brainstem. Occlusion of the SPS was found in three cases including ours, and dilated veins related to brainstem congestion were found in all four cases. MR imaging with Gd-DTPA clearly showed the engorged perforating veins causing the hemorrhage in the present case, so is very useful for detecting venous congestion.

CCF associated with a high risk of intracerebral venous hemorrhage may appear as parenchymal vermicular enhancement of brain vessels on CT, and as a pattern of dilated and tortuous cerebral veins on angiography.<sup>1)</sup> Our case demonstrated all these findings in the brainstem. Five cases of pontine congestion have been caused by indirect dural CCF.<sup>4,5,8)</sup> Nonopacification of the SPS was demonstrated in four of these cases as well as in some cases of direct CCFs. Abnormal angiographical staining was seen at the pons in two cases<sup>8)</sup> and unusual pontine enhancement on MR imaging, demarcated by the pontine raphe, in three cases,<sup>4,5)</sup> but these characteristics were not seen in cases of direct CCFs. In contrast, no engorged veins were found in the surrounding brainstem in any case of pontine congestion due to dural CCF. Such abnormal staining may result from abnormal communication of the multi-

ple dural branches in the nearby brainstem, which reflects a different pathogenesis to that caused by direct CCF.

Changes occurring in the venous drainage during and after embolization require special attention, and incomplete occlusion must be avoided because of the possibility of malignant venous congestion such as brainstem hemorrhage rather than resolution of the CCF. Good outcome can be expected if the CCF is completely occluded before brainstem hemorrhage occurs.

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