Hemispheric Laminar Necrosis as a Complication of Traumatic Carotid-Cavernous Sinus Fistula

—Case Report—

Masayasu KATO*,**, Yuka IKEGAME*,**, Izumi TOYODA*, Shinji OGURA*,
Hideomi KITAJIMA**, Shinichi YOSHIMURA**, and Toru IWAMA**

*Advanced Critical Care Center, Gifu University Hospital, Gifu;
**Department of Neurosurgery, Gifu University Graduate School of Medicine, Gifu

Abstract

A 43-year-old woman presented with a very rare case of hemispheric laminar necrosis as a complication of traumatic carotid-cavernous sinus fistula (CCF). The patient suffered head injury and extensive burns following a car accident. Oral intubation was performed under sedation. When sedation was discontinued 17 days after injury, the patient demonstrated left hemiparesis. Magnetic resonance imaging showed laminar necrosis affecting the right cerebral hemisphere. Angiography revealed a right high-flow direct CCF. Transarterial embolization of the fistula using a detachable balloon achieved complete occlusion of the fistula. However, the left hemiparesis persisted following this intervention. Traumatic CCF may be missed in patients with disturbed consciousness, so clinicians should not overlook possibility of the triad of symptoms of CCF in patients with head injury.

Key words: carotid-cavernous sinus fistula,   laminar necrosis,   head injury

Introduction

Carotid-cavernous sinus fistula (CCF) is an abnormal communication between the carotid artery and the cavernous sinus manifesting as a classical triad of symptoms consisting of chemosis, proptosis, and bruit. In general, the prognosis for CCF is good. Life-threatening complications such as intracerebral hemorrhage, massive epistaxis, and cerebral ischemia are rare. Here, we report a very rare case of hemispheric laminar necrosis as a complication of traumatic CCF.

Case Report

A 43-year-old woman suffered head injury in a car accident, in addition to extensive burns due to the car catching fire. On admission to our hospital, the patient had a Glasgow Coma Scale score of 14 (E3 V5 M6), and demonstrated no motor weakness. Computed tomography (CT) performed 1 hour after the accident revealed no abnormalities. The patient had burns involving 40% of her total body surface area. Oral intubation was performed and the patient was kept sedated in our hospital intensive care unit. Several debridement and skin grafting operations were performed under sedation, during which the patient's mean blood pressure remained above 80 mmHg and arterial oxygen saturation remained above 98%. Whole body edema including bilateral chemosis was noted following extensive intravenous fluid infusion, but gradually improved except for right chemosis (Fig. 1A). Sedation was discontinued 17 days after admission, and the patient's consciousness gradually improved. However, the patient then demonstrated left hemiparesis.

Magnetic resonance (MR) imaging showed abnormal appearance of the cortex and basal ganglia of the right cerebral hemisphere, dilated bilateral superior ophthalmic veins (SOVs), and abnormal flow voids in the bilateral cavernous sinuses (Fig. 2). Angiography showed a right high-flow direct CCF with complete absence of filling of the right internal carotid artery (ICA) above the fistula (Fig. 3A). The
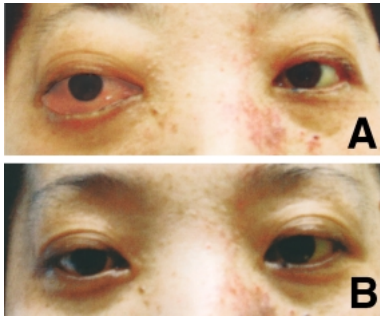
Fig. 1 Photographs of the patient, (A) before treatment for the carotid-cavernous sinus fistula showing right chemosis and proptosis, and (B) after treatment showing improved chemosis and proptosis.
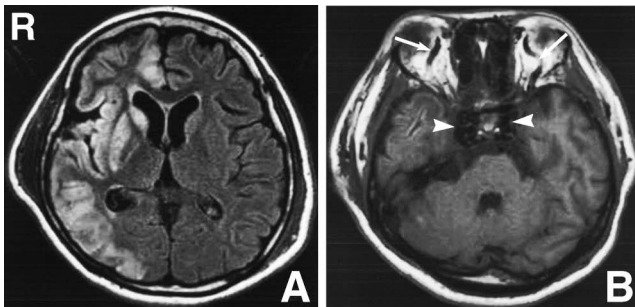

Fig. 2 A: Fluid-attenuated inversion recovery magnetic resonance image showing hyperintense areas in the cerebral cortex, caudate nucleus, and putamen of the right hemisphere. B: T_1-weighted magnetic resonance image showing dilated bilateral superior ophthalmic veins (*arrows*) and abnormal flow voids in the bilateral cavernous sinuses (*arrowheads*).
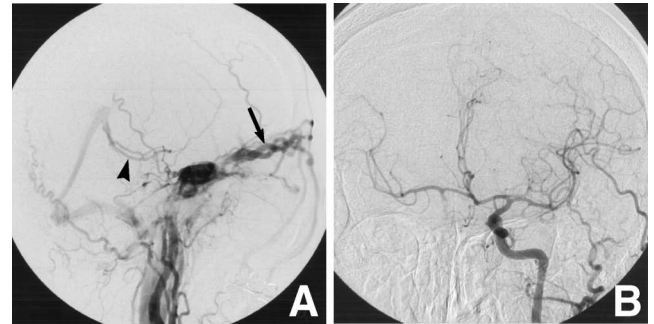

Fig. 3 A: Lateral right carotid angiogram showing a right high-flow direct carotid-cavernous sinus fistula with complete absence of filling of the right internal carotid artery above the fistula, and venous drainage into the superior ophthalmic vein (*arrow*) and the basal vein of Rosenthal (*arrowhead*). B: Anteroposterior left carotid angiogram showing the right middle cerebral artery supplied from the contralateral side through the anterior communicating artery.
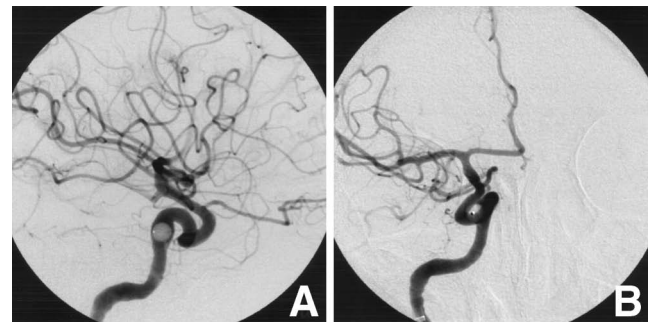

Fig. 4 Right carotid angiograms showing complete obliteration of the carotid-cavernous sinus fistula and the opacified right internal carotid artery. A: lateral view, B: anteroposterior view.

right middle cerebral artery (MCA) was supplied by cross flow from the contralateral side through the anterior communicating artery (Fig. 3B). The fistula drained into the SOV and the basal vein of Rosenthal (Fig. 3A). We suspected that the laminar necrosis in the right cerebral hemisphere was due to the CCF.

Transarterial embolization of the fistula using a detachable balloon resulted in complete occlusion. Immediately after embolization, both the ICA and the MCA were completely opacified (Fig. 4). The patient's chemosis and proptosis improved within a few days (Fig. 1B). She was discharged with residual left hemiparesis 2 months after admission.

Discussion

CCFs can be classified angiographically into types A to D.[2] Type A, the most severe type of CCF, is a direct connection between the ICA and the cavernous sinus. Type A occasionally causes severe complications such as intracerebral hemorrhage, massive epistaxis, decreased visual acuity, and cerebral ischemia.[7] The 3 reported cases of severe cerebral ischemia caused by traumatic CCF were all type A (Table 1).[9,10,18] Angiography revealed a direct CCF with total steal, appearing as complete absence of filling of ICA above the fistula, in two cases.[10,18]

CCFs that show complete angiographical "steal"

Table 1   Cases of severe cerebral ischemia caused by carotid-cavernous sinus fistula (CCF)

Author (Year)	Cause of injury	Initial symptom	Barrow's classifica-tion[2]	Finding of angiography	Treatment	Outcome
Iida et al. (1995)[10]	traffic accident	unknown due to coma	type A	CCF with total steal	—	death
Iida et al. (2002)[9]	falling down	unknown due to sedation	type A	good filling of the cerebral artery	transvenous coil embolization	lt hemiparesis
Wang et al. (2004)[18]	falling down	aphasia, hemiparesis	type A	CCF with total steal	transarterial balloon embolization	hesitant speech, mild rt hemiparesis
Present case	traffic accident	unknown due to sedation	type A	CCF with total steal	transarterial balloon embolization	severe lt hemiparesis

Total steal: complete absence of filling of the internal carotid artery above the fistula.

do not always induce cerebral ischemia to the extent of causing neurological deficits. Seven of 54 patients with traumatic CCFs had total steal,[5] but none of these seven patients had neurological symptoms. One case of traumatic CCF with total steal was followed by cerebral infarction in the area of the MCA, but no collateral flow was present across the anterior communicating artery, so total steal with poor collateral development probably caused the cerebral infarction.[18] A similar case was followed by a fatal cerebral infarction.[10] Post-mortem examination revealed no thrombus or obstructive change in the ICA, so that total steal of arterial blood probably caused the cerebral infarction, similar to complete obstruction of the ICA.[10]

In our case, MR imaging demonstrated cerebral hemispheric laminar necrosis. Laminar necrosis is usually seen in anoxic encephalopathy.[1] However, the patient was managed in the intensive care unit, and arterial oxygen saturation remained above 98% during her hospitalization. Angiography detected CCF with total steal, which drained into the basal vein of Rosenthal. Angiography showed no evidence of carotid arterial dissection or arterial stenosis after embolization of the CCF. This unusual venous drainage may cause deep venous hypertension.[6] We speculate that the reduced blood flow secondary to the steal phenomenon of the ICA and increased deep venous pressure due to the CCF led to severe cerebral hemispheric ischemia, and that this state continued while the patient sedated, leading to laminar necrosis.

The incidence of traumatic CCFs is only 0.2% to 0.3% among all cases of head or facial injury.[10] Detection of traumatic CCF depends on recognizing the classic triad of symptoms. We did not recognize the chemosis as a symptom of CCF, and could not detect cerebral ischemia because our patient was

comatose, so the diagnosis of CCF was delayed and laminar necrosis occurred. Therefore, other methods must be applied to identify CCF in comatose patients. Elevated jugular venous oxygen saturation (SjO$_2$) established the early diagnosis of a CCF in a patient with severe head injury.[3] Another case had similar clinical features.[12] Therefore, SjO$_2$ monitoring may be useful for detecting CCF in ventilated or sedated patients. Continuous monitoring of electroencephalography (EEG) and near-infrared spectroscopy (NIRS) could be safely possible in the intensive care unit.[15,16] EEG monitoring may be useful for indicating outcomes in patients with traumatic brain injury.[16] NIRS may be useful for evaluating brain ischemia.[15] The diagnosis of CCF depends primarily on angiographic findings, but other less invasive methods, such as MR imaging, CT, and transcranial Doppler ultrasonography may be used to screen for CCFs.[4,8,10,14] Venous flow signal in the SOV and cavernous sinus on MR imaging or abnormal venous flow of the cavernous sinus on MR angiography are frequently useful for identifying CCF.[8,11] Recently, MR angiography source images have allowed the diagnosis of CCF without SOV enlargement.[8,14] There was a possibility of causing hemorrhagic infarction by the reperfusion after embolization in this case. The early MR parenchymal enhancement, which is related to damage to the blood-brain barrier, may be a good predictor of symptomatic hemorrhagic infarction in patients with acute ischemic stroke.[17]

The risk of cerebral ischemia is difficult to evaluate based only by the findings of angiography. CCF with direct shunt (type A) should be immediately treated following diagnosis.[13] Review of angiographic data from 155 patients with CCF identified various angiographic features associated with risk of morbidity and mortality, including presence of a

pseudoaneurysm, large varix of the cavernous sinus, venous drainage to cortical veins, and thrombosis of venous outflow pathways distant from the fistula.[7] Our present case report suggests that traumatic CCF may be missed in patients with disturbed consciousness, so clinicians should not overlook possibility of the triad of symptoms of CCF in patients with head injury. Continuous monitoring of SjO$_2$, EEG, or NIRS is necessary for comatose patients. The presence of total steal detected by angiography is not always associated with severe cerebral ischemia, but, given the risk to the patient, early treatment of CCF with total steal is recommended.

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Address reprint requests to: Masayasu Kato, M.D., Department of Neurosurgery, Takayama Red Cross Hospital, 3–11 Tenman–cho, Takayama, Gifu 506–8550, Japan.
e-mail: masayasu.kato@nifty.ne.jp