Brainstem Edema: An Unusual Complication of Carotid Cavernous Fistula

Michael Mu Huo Teng,[1] Tsuen Chang,[2] David Hung-Chi Pan,[3] Chen-Nen Chang,[4] Chun-I Huang,[3] Wan-Yuo Guo,[2] Chi-Chang Chen,[2] Rong-Guey Pang,[3] and Liang-Shong Lee[3]

The complications of carotid cavernous fistula include (1) intracerebral hematoma, subarachnoid hemorrhage, intraorbital hemorrhage, epistaxis, and otorrhagia [1–4]; (2) blurred vision or loss of vision caused by secondary glaucoma, intraorbital hemorrhage, or other causes [1, 5]; (3) cerebral ischemia due to steal of blood by the fistula [1]; (4) progressive proptosis [1, 5]; (5) increased intracranial pressure [1]; and (6) limitation of ocular movements [1, 5]. Recently, we encountered two cases of carotid cavernous fistula with the unusual complication of brainstem edema. Both patients had direct-type carotid cavernous fistulas, which occurred after head injury.

Case Reports

Case 1

A 36-year-old man had a left facial bone fracture followed by open reduction 2 months before admission. Seventeen days later he was shown to have a left carotid cavernous fistula (CCF) (Fig. 1A). Matas' test (manual compression of one common carotid artery to predict the tolerance of carotid occlusion) was performed on the left side, and the patient developed aphasia, right hemiparesis, and loss of consciousness in less than 30 sec. These symptoms resolved completely in about 1 min. Matas' test often produces a false-positive result in the presence of a CCF because of retrograde steal from the intracranial vessels into the fistula. Therefore, right internal carotid angiography was performed with cross compression of the left common carotid artery and demonstrated inadequate collateral flow from the right carotid artery. The Jaeger-Hamby procedure (a rarely used procedure for trapping of the internal carotid artery with muscle embolization of the fistula) was planned and the patient underwent a left external carotid–internal carotid artery bypass as the first step of surgical treatment on the 38th day after trauma. Prolonged unconsciousness and right hemiplegia were noted after surgery.

A CT scan showed an intracerebral hematoma in the left temporoparietal region and a small area of low density in the left thalamus (Fig. 1B). Temporal craniectomy was performed immediately to evacuate the hematoma. The patient gradually regained consciousness and muscle strength over a period of 1 week, but mild motor aphasia remained. He was transferred for embolization 19 days after bypass surgery.

Examination at that time revealed proptosis and chemosis of the left eye and aphasia. Hemiplegia was still present. A CT scan on the same day showed (1) enlargement of the left superior ophthalmic vein and left cavernous sinus compatible with carotid cavernous fistula; (2) a low-density area in the left temporoparietal region due to hematoma evacuation; and (3) low-density change with swelling in the left midbrain, cerebral peduncle, and thalamus (Fig. 1C). Enhancement was noted in the low-density area in the left brainstem on the postcontrast CT scan. An angiogram 21 days after bypass surgery showed occlusion of venous drainage to the superior ophthalmic vein, superior petrosal sinus, and inferior petrosal sinus; and dilatation of the proximal superior petrosal sinus (Fig. 1D). Embolization was performed on the same day. The fistula was completely obliterated with a detachable balloon and the blood flow in the left internal carotid artery was preserved (Fig. 1E). Hemiplegia and motor aphasia disappeared gradually after embolization. Six days after embolization, a CT scan showed resolution of brainstem edema (Fig. 1E). The patient returned to work without any neurologic or ocular symptoms 10 months after embolization.

Case 2

A 26-year-old man who had a head injury 3 months before admission presented with progressive protrusion of the left eye, bilateral orbital bruits, and conjunctival congestion for 2 months. An angiogram showed a direct-type carotid cavernous fistula on the left side (Fig. 2A). Because a detachable balloon failed to enter the fistula from the femoral approach, embolization was performed by direct puncture of the superior ophthalmic vein through the orbit under guidance of real-time digital subtraction angiography. Five steel coils were implanted in the cavernous sinus.

After embolization, the fistula became smaller, and the remaining venous drainage was limited to the superior petrosal sinus, the inferior petrosal sinus, and the basal vein of Rosenthal (Fig. 2B). On the fifth day after embolization the patient experienced numbness of the right side of the body and face. A CT scan showed slight midbrain edema, more on the left side. On the 10th day, dilatation of both pupils,
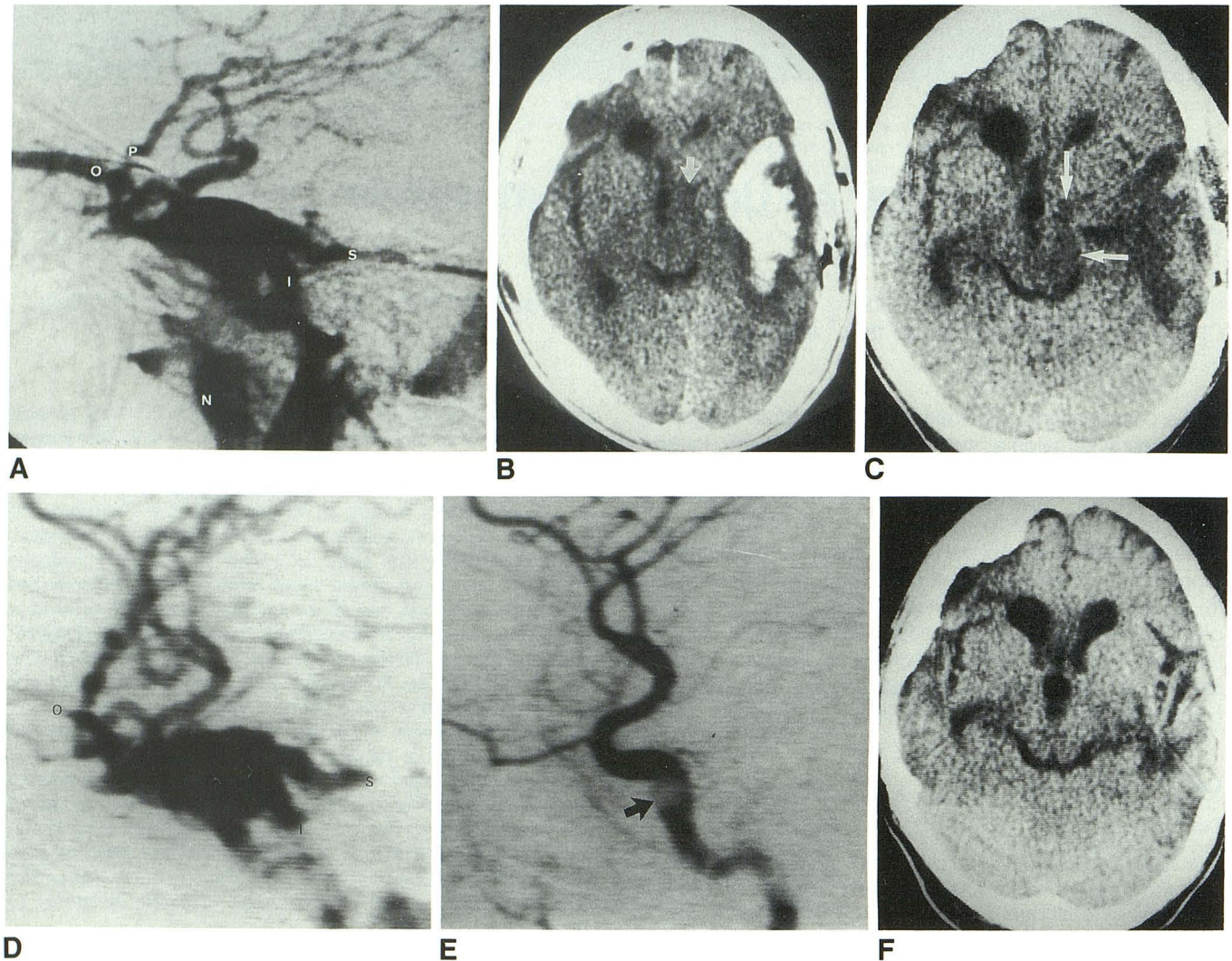
Fig. 1.—Case 1.

A, Arteriogram obtained before bypass surgery. A direct-type carotid cavernous fistula is noted with venous drainage to the superior ophthalmic vein (O), sphenoparietal sinus (P), venous plexus in the nasopharynx (N), and superior (S) and inferior (I) petrosal sinuses.

B, CT scan on the day of external carotid–internal carotid artery bypass. High-density hematoma is noted in left temporal region. Brainstem is normal in density and shape. A small area of low density is noted in left thalamus (*arrow*).

C, CT scan on 19th day after bypass surgery. There is edema and swelling of left side of brainstem and thalamus (*arrows*).

D, Arteriogram obtained before embolization. Venous drainage to superior ophthalmic vein (O), and superior (S) and inferior (I) petrosal sinuses are not as patent as before. Venous plexus in region of nasopharynx is no longer seen. Venous drainage of fistula is redirected into the brain, with dilatation of the proximal superior petrosal sinus (S).

E, Arteriogram obtained after embolization. There is complete obliteration of the fistula and preservation of blood flow in the internal carotid artery. The balloon, which is actually located lateral to the carotid artery in the cavernous sinus, is barely seen (*arrow*).

F, CT scan on sixth day after embolization. Edema and swelling have resolved. The C-shaped quadrigeminal cistern shows no external compression.

gradual weakness of both extremities, and decreased level of consciousness were noted. A CT scan on that day showed extensive edema of the midbrain and the thalamus (Fig. 2D). An angiogram on the 11th day showed (1) the venous drainages to the inferior petrosal sinus and superior petrosal sinus were occluded and (2) all residual fistula drained to the brain via markedly dilated venous pedicles (Fig. 2C). We decided that the fistula had to be occluded completely because the brainstem edema was thought to be the result of venous drainage of the fistula. The second embolization was attempted, initially with a detachable balloon by femoral approach, but again the balloon would not enter the fistula. Because the patient tolerated the test occlusion for more than 15 min, we occluded the internal carotid artery and the fistula with balloons and a steel coil. A follow-up CT

study showed reduction of the brain edema 5 days after the second embolization (Fig. 2E). Complete resolution of the edema was noted on a CT study done 2 months later (Fig. 2F). The patient regained consciousness 20 days after the second embolization and returned to his work 2 years after the head injury without neurologic signs or ocular symptoms.

Discussion

Intracranial hemorrhage, either intracerebral or subarachnoid, is a well known complication of carotid cavernous fistula. Patients with cortical venous drainage have a higher risk of
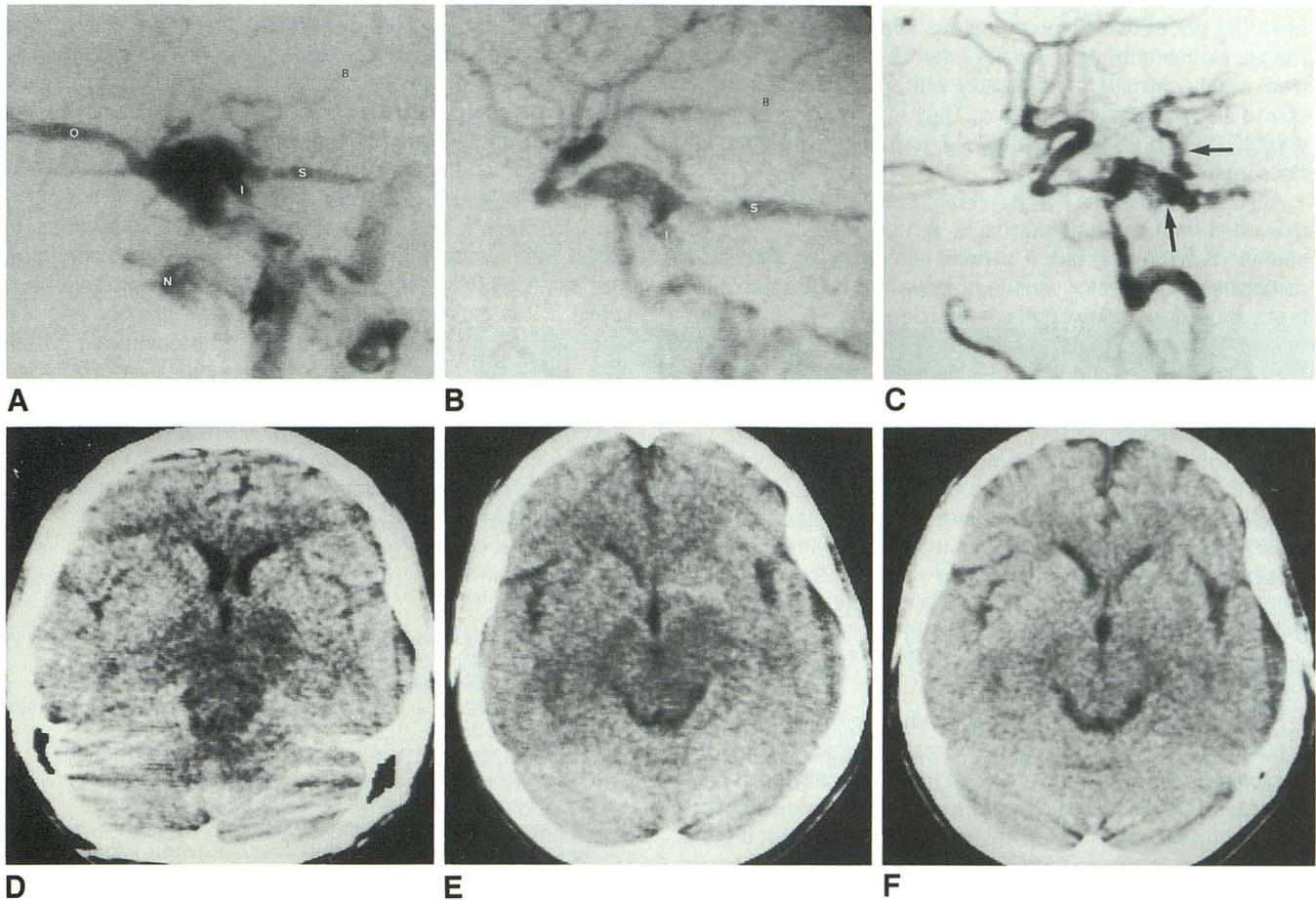
Fig. 2.—Case 2.

A, Arteriogram obtained before embolization. A direct-type carotid cavernous fistula drains to the superior ophthalmic vein (O), venous plexus in the nasopharynx (N), basal vein of Rosenthal (B), and superior (S) and inferior (I) petrosal sinuses.

B, Arteriogram obtained immediately after first embolization. Fistula is smaller in size; anteroinferior compartment of cavernous sinus has been obliterated; and venous drainage of the remaining fistula is to the basal vein (B) and the superior (S) and inferior (I) petrosal sinuses.

C, Arteriogram obtained on 11th day after first embolization. Venous drainage to the superior and inferior petrosal sinus has occluded spontaneously. The remaining venous drainage is to the brain via the markedly dilated venous pedicles (*arrows*).

D, Plain CT scan on 10th day shows extensive edema and swelling over the midbrain, cerebral peduncle, and thalamus.

E, Plain CT scan on fifth day after second embolization shows obvious diminution of the midbrain and thalamic edema.

F, Plain CT scan 2 months after second embolization shows complete resolution of midbrain and thalamic edema.

intracranial hemorrhage [1]. The intracerebral hemorrhage in our first case may have been a complication of carotid cavernous fistula because of the presence of cortical venous drainage through the sphenoparietal sinus (Fig. 1A). However, it could also have been a complication of the external carotid–internal carotid artery bypass procedure.

The possible causes of brainstem edema in our two cases are (1) infarction resulting from arterial insufficiency, venous hypertension, or venous infarct; and (2) white matter edema surrounding the hematoma.

Stealing of blood in a carotid cavernous fistula usually occurs in the carotid territory on the ipsilateral side. Thus, the resulting arterial insufficiency or infarction from stealing of blood by the fistula usually occurs in the carotid territory on the same side as the fistula. Edema or infarction of the brainstem caused by steal occurs when there is occlusion of the internal carotid artery above the origin of the posterior communicating artery on the side of the fistula. In our cases

it is unlikely that brainstem edema was the result of arterial infarction caused by steal of blood from the vertebrobasilar system because vertebral angiograms showed nonopacification of the fistulas.

White matter edema may be perifocal edema surrounding a hematoma. Our second case had no intracranial hematoma. In the first case, the brainstem edema was unlikely to be perifocal edema surrounding a hematoma because (1) perifocal edema is usually contiguous to the hematoma and a temporoparietal hematoma is unlikely to have perifocal edema extending to the brainstem; and (2) perifocal edema shows no enhancement, but our first case showed enhancement on the postcontrast CT. Therefore, the edema in both cases was not likely to have been perifocal edema surrounding intracerebral hematoma.

Venous infarcts, hemorrhagic or nonhemorrhagic, can be observed in cerebral thrombophlebitis or dural arteriovenous fistulas draining into cerebral veins [6]. According to Chiras

et al. [6], about two-thirds of venous infarcts are nonhemorrhagic. Nonhemorrhagic venous infarcts usually show localized, poorly delimited hypodensity with mass effect probably due to localized cerebral edema. They may or may not show enhancement on a postcontrast CT scan. Nonenhancing venous infarcts are rare [6].

Although brainstem edema in both cases was similar to that seen with a nonhemorrhagic venous infarct, their presentations were different. A venous infarct usually manifests as occlusion of a major venous outflow of a large area of the brain, thus often carrying a grave prognosis. The brainstem edema in our cases was a more reversible process because both patients recovered completely after their fistulas were obliterated, suggesting that venous hypertension was the cause of the edema.

The blood pressure of draining veins of a carotid cavernous fistula is usually much higher than in venous infarct. Therefore, it is possible for cases of carotid cavernous fistula to develop a CT appearance similar to that of venous infarct. While intracerebral hemorrhage is a well known complication of carotid cavernous fistulas, a focal low-density change of the brain with mass effect (brain edema) similar to that of a nonhemorrhagic venous infarct has rarely been described in carotid cavernous fistulas.

In both our cases, hemodynamic changes were noted to accompany brainstem edema. Venous drainage to the ophthalmic vein, the superior petrosal sinus, and the inferior petrosal sinus in the first case were spontaneously occluded, and the drainage of the fistula was redirected to the brain parenchyma. In the second case, spontaneous occlusion of the inferior petrosal sinus and the superior petrosal sinus was noted days after the first embolization. Thus, the remaining fistula all drained to the brainstem and basal ganglia and resulted in marked venous hypertension in these focal areas. From the above observations, it appears that the reason why brain edema similar to venous infarct has rarely been seen in carotid cavernous fistulas is probably because there is good extracranial venous drainage in most cases. However, when a hemodynamic change occurs spontaneously or after a therapeutic procedure, the CT appearance is that of a low-density change with mass effect (brain edema), similar to a nonhemorrhagic infarct or an intracerebral hemorrhage, and is probably due to focal venous hypertension in brain parenchyma.

Detachable balloons were first introduced by Serbinenko in 1974 [7] and have become the treatment of choice for a direct-type carotid cavernous fistula [8–11]. Transvenous embolization of the cavernous sinus also has been reported in the treatment of carotid cavernous fistula [12–15]. Because of anatomic variation in the cavernous sinus [16] and the high flow in the fistula, complete obliteration of a direct-type carotid cavernous fistula by transvenous embolization may be difficult [13–15]. To preserve the flow in the internal carotid artery after failing to pass a detachable balloon into the fistula from a femoral approach, we performed direct puncture of the superior ophthalmic vein for embolization in our second case because of a single prior success in obliterating an indirect carotid cavernous fistula by this approach. Unfortunately, the fistula persisted, even after implanted steel coils resulted in compression and narrowing of the cavernous internal carotid artery.

In conclusion, our findings indicate that, in addition to intracranial hemorrhage, brain edema similar to nonhemorrhagic venous infarct may occur in cases of carotid cavernous fistula if the extracranial venous drainage is poor and the major venous drainage is directed to the focal brain parenchyma. The most likely cause of this edema is venous hypertension. In treating a carotid cavernous fistula, the goal should be complete obliteration of the fistula. Any residual fistula left draining into the brain risks the development of intracranial edema similar to a nonhemorrhagic venous infarct or an intracranial hemorrhage.

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