

Contralateral extensive cerebral hemorrhagic venous infarction caused by retrograde venous reflux into the opposite basal vein of Rosenthal in posttraumatic carotid-cavernous fistula: A case report and literature review

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Abstract

We describe a patient with traumatic carotid-cavernous fistula (CCF), subsequently developing contralateral extensive hemorrhagic venous infarction from retrograde venous reflux into the opposite basal vein of Rosenthal. A 54-year-old woman was involved in a motor vehicle accident and sustained severe traumatic brain injury. Two months later, she developed bilateral proptosis and audible bruit. Magnetic resonance imaging (MRI) and magnetic resonance angiography (MRA) of the brain demonstrated the right direct CCF. Fluid-attenuated inversion recovery (FLAIR) images showed a small hyperintense area at the left basal ganglia. Ten days later, she developed right-sided grade 2/5 hemiparesis, facial upper motor neuron weakness, and cognitive impairment. Follow-up MRI showed significant progression of hyperintensities involving the left-sided centrum semiovale, basal ganglia, thalamus, midbrain, pons, cerebellum, basal frontal, temporal lobes, especially subcortical white matter on FLAIR images, and multiple hypointense foci of hemorrhagic component on T2*-weighted gradient-echo images, representing hemorrhagic venous infarction. While waiting for embolization, she rapidly developed right hemiplegia and aphasia, and became somnolent. Under general anesthesia, emergency endovascular treatment was performed successfully to obliterate the fistula without surgical intervention. Five months after endovascular treatment, MRI and MRA confirmed no residual fistula and revealed nearly complete resolution of abnormal increased signal intensity. In the present case, the factors related to the presence of this rare condition were absence of the ipsilateral basal vein of Rosenthal (BVR), occlusion of posterior segment of the contralateral superior petrosal sinus, and a developed uncal vein with hypoplastic second and third segments of the contralateral BVR.

Keywords

Venous infarction, venous congestion, traumatic carotid-cavernous fistula, direct carotid-cavernous fistula, basal vein of Rosenthal

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Introduction

A direct carotid-cavernous fistula (CCF), that is, Barrow type A, is defined by direct high-flow connection between the cavernous segment of the internal carotid artery (ICA) and the cavernous sinus (CS).¹ Direct type A CCF has been classified into traumatic, including iatrogenic, or spontaneous fistula. The traumatic CCF, the most common type, usually occurs from a motor vehicle accident with closed traumatic brain injury associated with a basilar skull fracture.^{2,3} The spontaneous CCF usually results from rupture of a pre-existing intracavernous carotid aneurysm, and may rarely be associated with intrinsic weakness of

the vessel wall in some diseases, such as Ehlers–Danlos type IV and fibromuscular dysplasia.⁴

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The most common clinical manifestations, that is, pulsating proptosis, chemosis, and an audible bruit, depend on the venous drainage. Due to drainage through intercavernous sinuses across the midline, the ocular symptoms may occur on the opposite side to the fistula or bilaterally.⁵ Rarely, aggressive neurologic symptoms, especially deterioration of consciousness, may also develop due to cerebral ischemia and/or hemorrhage.^{6,7}

Cerebral ischemia in traumatic or direct CCF may result from arterial or venous cause. The arterial infarction from stealing of blood flow by direct CCF usually occurs in the ipsilateral carotid territory.⁸ The steal phenomenon can cause life-threatening or fatal complications when collateral circulation from the circle of Willis is insufficient.⁹ On the other hand, venous congestion from retrograde cortical or deep venous reflux in the direct CCFs in the area of the venous territories, resulting in cerebral hemorrhage or less common venous infarction.^{10,11} Intracerebral hemorrhage from direct CCF may be considered rare, with only 1% incidence from the study of Lewis et al.² Intracerebral hemorrhagic venous infarction in direct CCF is much rarer than spontaneous hemorrhage.¹²

We describe a case of traumatic CCF with contralateral extensive cerebral hemorrhagic venous infarction due to retrograde venous reflux into the opposite basal vein of Rosenthal (BVR), which is an extremely rare complication. We also review literature of abnormal appearance in computerized tomography (CT) scan or magnetic resonance imaging (MRI) of brain edema or venous congestion with or without hemorrhagic changes from direct CCFs.

Case report

A 54-year-old woman was involved in a motor vehicle accident and sustained head trauma resulting in unconsciousness for a few hours. In the time after regaining consciousness, she had bleeding from the nostrils and ears without neurological deficits. A CT scan of the head showed acute thin subdural hematomas along both sides of the falx cerebri, skull fracture of the right parietal bone and multiple fractures of the skull base and facial bone, including bilateral zygomatic arches, bilateral lateral orbital walls, superior and lateral wall of the right sphenoid sinus, and lateral wall of the left maxillary sinus. Pedicle screws fixation was performed for correction of unstable thoracolumbar fractures. She gradually recovered without neurological deficits and was discharged home 10 days later. Two months later, she developed redness of both eyes, bilateral proptosis in association with chemosis in both eyes, bilateral audible bruit, and binocular horizontal diplopia. She went back to check with the ophthalmologist in the previous hospital, and was sent to our institute. Ophthalmic examination revealed bilateral proptosis more prominent on the right side, chemosis with conjunctival injection of both eyes, and normal findings on

pupillary examination. Visual acuity was 20/50 in the right eye and 20/40 in the left. There was a limitation to abduction in the right eye. A bruit was audible over both eyes and in the right temporal region. She had no muscle weakness or sensory loss during physical examination. Cerebral angiography and simultaneously performing the endovascular treatment were scheduled in the next three weeks. MRI and magnetic resonance angiography (MRA) of the brain (Magnetom Skyra 3T; Siemens, Erlangen, Germany) with a complete sequence of images obtained before hospital discharge demonstrated the right direct CCF located at the posterior genu of the cavernous carotid artery. Fluid-attenuated inversion recovery (FLAIR) images showed a small hyperintense area at the left putamen, caudate nucleus, and internal capsule, indicating edema, congestion, or early venous infarction (Figure 1(a)). Time-of-flight (TOF) MRA indicated an enlarged left BVR on the opposite side to the fistula (Figure 2(a)). T2-weighted images also revealed the abnormal flow void corresponded to venous anastomoses of the left peduncular vein, BVR, and deep middle cerebral vein (DMCV), respectively (Figure 3(a)). Susceptibility-weighted imaging (SWI) demonstrated hyperintensity in the left BVR, representing arterialized blood flow of this draining vein (Figure 3(b)). Ten days later after staying at home, she developed right-sided grade 2/5 hemiparesis, facial upper motor neuron weakness, and cognitive impairment. She went back again to the local hospital and was sent to obtain MRI of the brain (Magnetom Symphony 1.5T; Siemens, Erlangen, Germany) for evaluation of these problems. The findings demonstrated significant progression of hyperintensities involving the left-sided centrum semiovale, basal ganglia, thalamus, midbrain, pons, cerebellum, basal frontal, temporal lobes, especially subcortical white matter on FLAIR images (Figure 1(b) and Figure 4(a)), an enlarged left BVR and perimesencephalic veins on TOF-MRA (Figure 2(b)), and multiple hypointense foci of hemorrhagic component on T2*-weighted gradient-echo images, representing hemorrhagic venous infarction. The disabled patient was transferred to our institute 10 days later. While waiting for embolization, she rapidly developed right hemiplegia and aphasia, and became somnolent. A CT scan of the brain obtained before treatment showed progression of hemorrhagic venous infarction at the left-sided basal ganglia, thalamus, brainstem, cerebellum, frontal, and temporal lobes with a small acute subcortical hemorrhage at the left temporal lobe, resulting in diffuse brain swelling and midline shift (Figure 5(a)). Emergency endovascular treatment was performed by using a biplane flat panel detector angiographic system (Axiom Artis dBA; Siemens, Erlangen, Germany) under general anesthesia. Cerebral angiography revealed direct CCF from the inferomedial aspect of the posterior genu of the right cavernous carotid artery. Multiple venous drainages included right superior ophthalmic vein (SOV), right inferior petrosal sinus

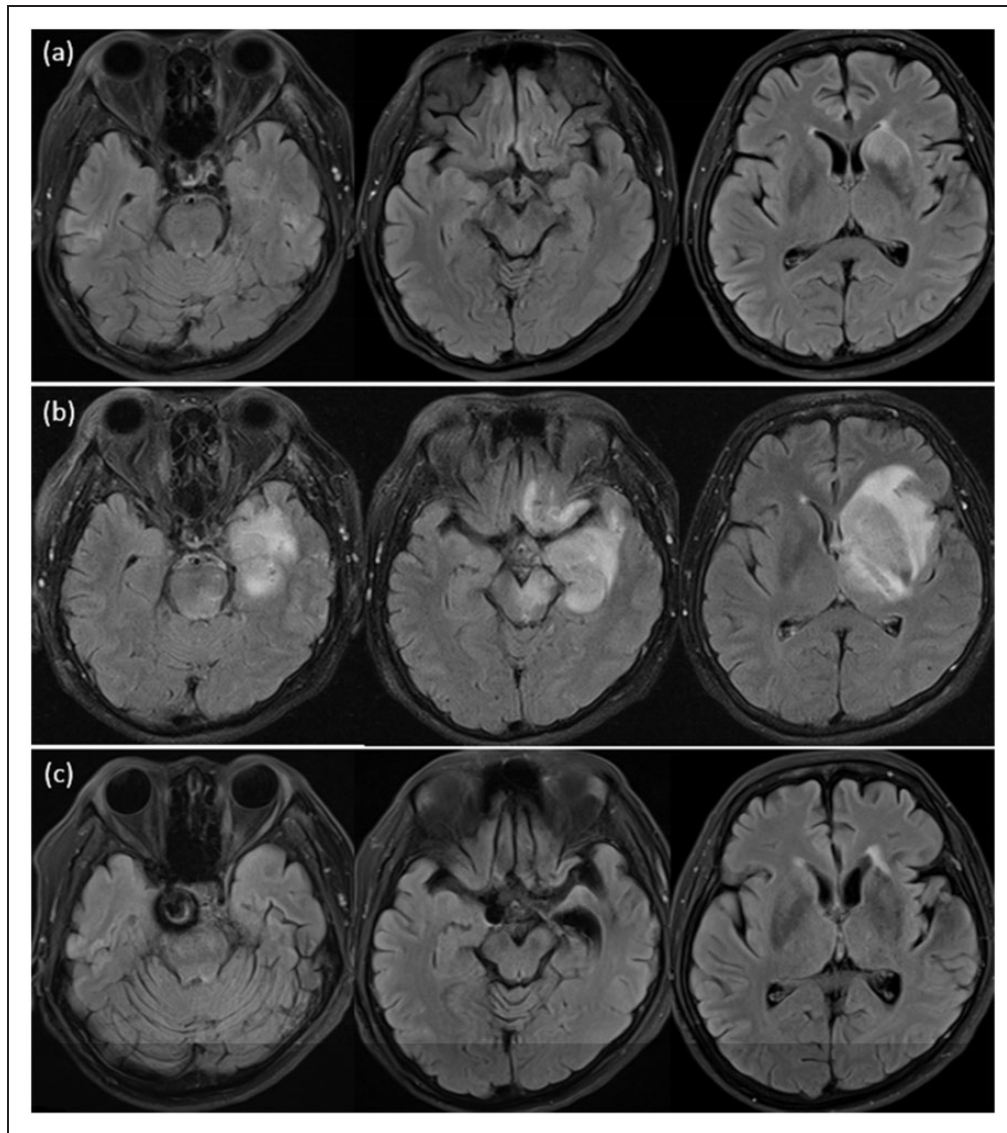


Figure 1. Sequential axial fluid-attenuated inversion recovery (FLAIR) images at level of pons to basal ganglia (BG) obtained at two months after the accident (a), ten days later (b), and five months after embolization (c) demonstrated rapid progression from a small hyperdense area at left BG to extensive hyperintensity involving the left side of BG, thalamus, basal frontal lobe, temporal lobe, midbrain, and pons, representing extensive venous infarction in a period of ten days. Five months after endovascular treatment, FLAIR images confirmed nearly complete resolution of abnormal increased signal intensity.

(IPS), and basilar venous plexus, and posterior intercavernous sinus draining into the left CS. The subsequent venous drainages from the left CS ran anteriorly into the left SOV, posterolaterally into residual segment of the proximal superior petrosal sinus (SPS) with subsequent draining into the left superior petrosal vein (SPV) entering further into the veins of cerebellar hemisphere, posteriorly into bridging vein with draining into the left transverse pontine vein (TPV) also opening into left cerebellar veins, and superiorly into the dilated left uncal vein connecting with the left BVR. From anastomosis of the left uncal vein and the BVR, the drainages then ran anteriorly into the residual dilated vein of the first segment of the left BVR, and posterosuperiorly to the great vein of Galen through multiple hypoplastic portions of the

second and third segments of BVR. There was antegrade flow of the right ICA filling into the right anterior, and middle cerebral territories (Figure 4(b) and Figure 6(a) to (d)). Another retrograde venous drainage detected by angiographic CT in maximum intensity projection (MIP) reformatted images revealed venous reflux from the right SPS running into the right TPV through the right SPV, and then turning around anteromedially through the anterior and lateral pontomesencephalic veins up to the right peduncular vein crossing the midline via the interpeduncular or posterior communicating vein to the left peduncular vein connecting posteriorly into the left perimesencephalic vein and terminating to the dilated left BVR joining with the route from the left uncal vein (Figure 7). Transarterial embolization with one detachable

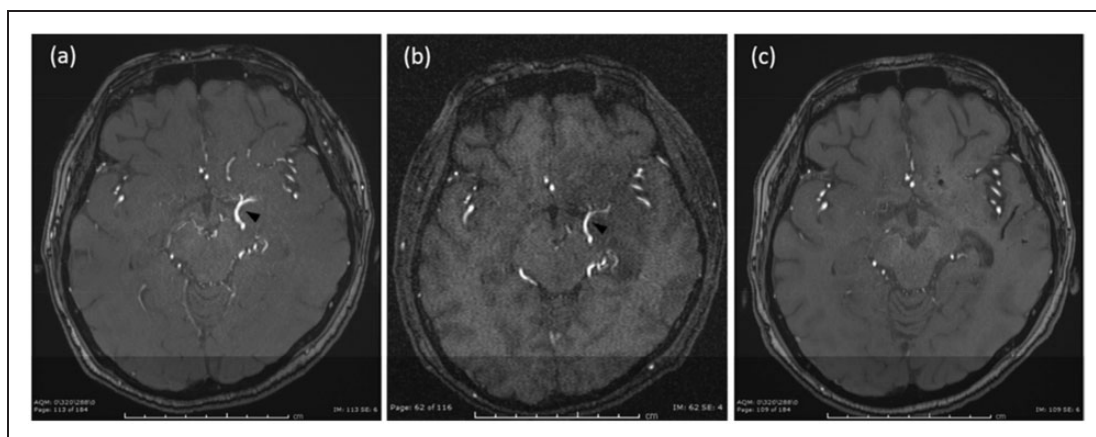


Figure 2. Axial time-of-flight magnetic resonance angiography obtained at two months after the accident (a), ten days later before embolization (b), and five months after embolization (c) confirmed the disappearance of an enlarged left basal vein of Rosenthal (arrow heads) after obliteration of the fistula.

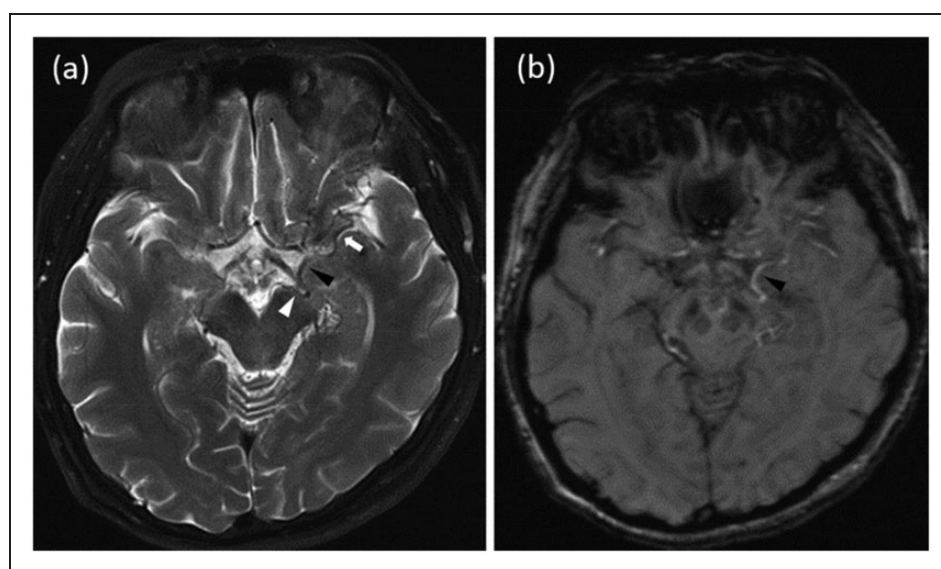


Figure 3. (a) Axial T2-weighted magnetic resonance imaging revealed the flow voids of venous anastomoses of the left peduncular vein (white arrow head), the left basal vein of Rosenthal (BVR) (black arrow head), and the left deep middle cerebral vein (white arrow). (b) Axial susceptibility-weighted imaging demonstrates hyperintensity in the left BVR (black arrow head), representing arterIALIZED blood flow of this draining vein.

balloon (Goldbal; Balt Extrusion, Montmorency, France) was unable to complete obliteration of the fistula. Thus, this balloon was placed to close only posterior drainage (Figure 8(a) and (b)). Unfortunately, occlusion of the right ICA occurred after inflation of another balloon (Figure 8(c)). Even though the left internal carotid and vertebral showed collateral circulation of the right anterior cerebral artery via anterior communicating artery from the left ICA and the right middle cerebral artery via the right posterior communicating artery from the right posterior cerebral artery (Figure 6(e) and (f)), these findings could not guarantee that the collateral circulation was adequate. Transvenous embolization was subsequently performed via the left IPS by using the fibered interlock detachable coils (Boston Scientific

Corporation, Natick, MA, USA) densely packing in the proximal dilated right SOV and posterior part of the right CS (Figure 8(f)). A control right ICA injection immediately after embolization confirmed complete obliteration of the fistula with preservation of the flow in the parent artery (Figure 8(d) and (e)). Following occlusion of the fistula, a bruit entirely disappeared, and ocular symptoms significantly improved. Immediate assessment of the patient revealed improvement of consciousness and muscle power. We decided to use medication for reduction brain swelling instead of performing decompressive surgery.

Follow-up sequential CT scans of the brain demonstrated gradual resolution of extensive hemorrhagic venous infarction until nearly complete disappearance

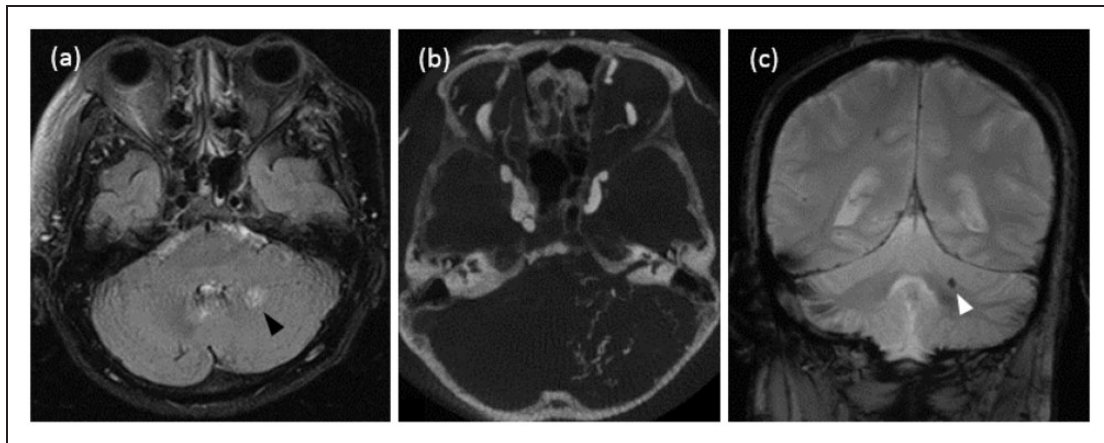


Figure 4. Before embolization, axial fluid-attenuated inversion recovery (FLAIR) MR image (a) and axial maximum intensity projection (MIP) reformatted image of angiographic CT (b) at the level of cerebellum revealed multiple left-sided dilated cerebellar veins and a small hyperintensity (black arrow head) compatible with edema or venous congestion at left deep cerebellar white matter. Five months after embolization, coronal T2*-weighted gradient-echo image (c) shows a small hypointense foci of previous hemorrhage (white arrow head).

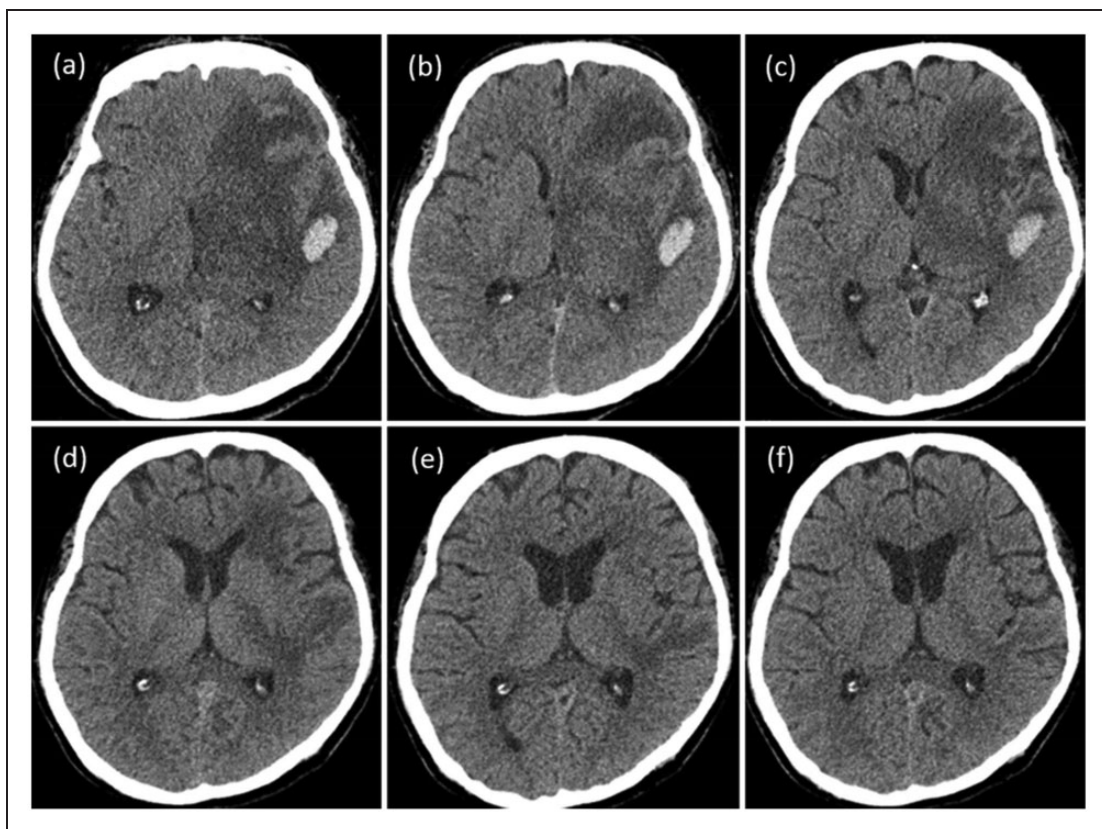


Figure 5. Axial computerized tomography (CT) scan obtained before embolization (a) showed extensive parenchymal edema at the left side of the basal ganglia, thalamus, frontal, and temporal lobes with a small acute subcortical hemorrhage at the left temporal lobe, resulting in diffuse brain swelling and midline shift. Axial CT scans obtained at one day (b), four days (c), two weeks (d), three weeks (e), and one month (f) following embolization revealed that extensive hemorrhagic venous infarction gradually resolved until nearly complete resolution after obliteration of the fistula.

within one month (Figure 5(b) to (f)). MRI and MRA of the brain obtained at five months after endovascular treatment confirmed no evidence flow-related enhancement or early enhancement of the right CS on either three-dimensional TOF-MRA or contrast-enhanced

MRA (Figure 9). FLAIR images revealed nearly complete resolution of abnormal increased signal intensity of the left side of basal ganglia, thalamus, basal frontal lobe, temporal lobe, midbrain, and pons (Figure 1(c)). TOF-MRA also confirmed the disappearance of an

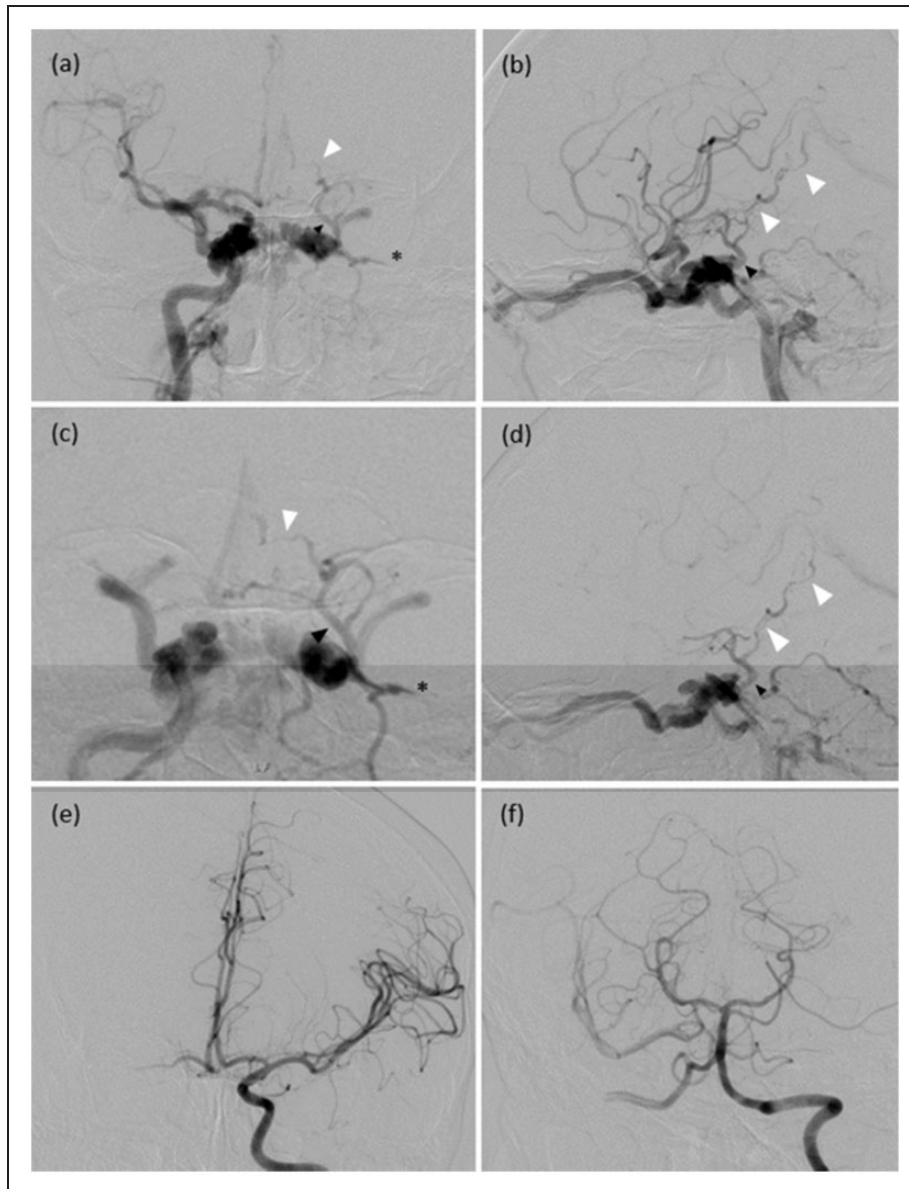


Figure 6. Anteroposterior ((a) and (c)) and lateral ((b) and (d)) views in early ((a) and (b)) and late ((c) and (d)) arterial phases of the right internal carotid artery (ICA) injection demonstrate antegrade flow in the right ICA distal to the direct carotid-cavernous fistula with retrograde drainage into the right superior ophthalmic vein (SOV), the right inferior petrosal sinus, basilar venous plexus, and crossing to the left cavernous sinus (CS) via the posterior intercavernous sinus. The left CS further drains into the left SOV and the left basal vein of Rosenthal (BVR) via the left uncal vein (black arrow heads), running superiorly into the great vein of Galen and anteriorly to the residual dilated vein of the first segment of the left BVR. There were hypoplastic portions (white arrow heads) at the second and third segments of the left BVR. Another route of retrograde venous reflux opens into proximal part of the left superior petrosal sinus, where the residual part was occluded (asterisks), then turns medially to the left superior petrosal vein draining into left cerebellar veins. Anteroposterior views of the left ICA (e) and left vertebral artery (f) injections reveal collateral circulation of the right anterior cerebral artery via anterior communicating artery from the left ICA and the right middle cerebral artery via the right posterior communicating artery from the right posterior cerebral artery.

enlarged left BVR after obliteration of the fistula (Figure 2(c)). T2*-weighted gradient-echo images showed multiple small hypointense foci of an old hemorrhage at the right parietal white matter and left deep cerebellar white matter (Figure 4(c)). At six months' follow-up, ophthalmic examination revealed complete resolution of proptosis, chemosis, and diplopia without secondary glaucoma. Visual acuity with correction was 20/25 in the right eye, and 20/20 in the left. Nine months after embolization, the patient had recovered

completely from aphasia. The time to nearly complete recovery of the right-sided weakness was two years. She is able to independently perform activities of her daily living. However, she still has mild cognitive impairment.

Discussion

Since 1978, Ambler et al.¹³ demonstrated unusual serial angiographic and neurological findings of right atypical

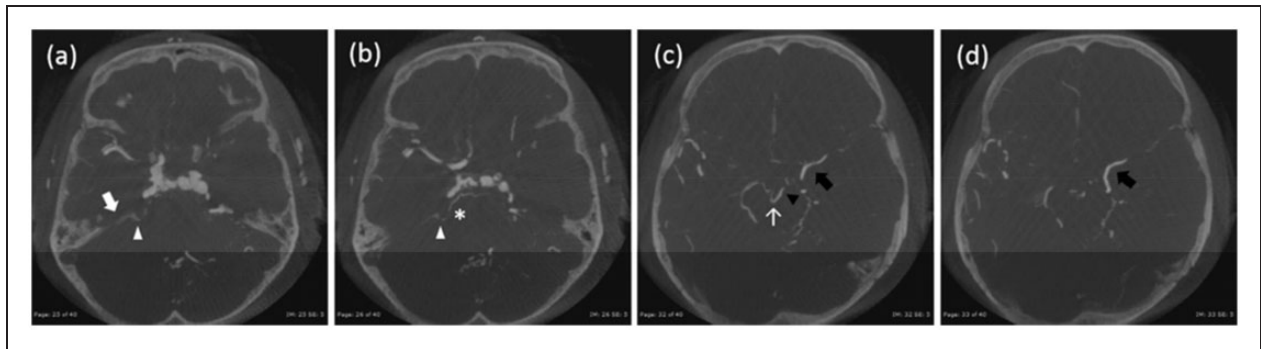


Figure 7. Axial maximum intensity projection reformatted images of angiographic computerized tomography at the level of pons (a) and (b) to midbrain; (c) and (d) reveal another retrograde venous reflux from the right superior petrosal sinus (thick white arrow) running into the right transverse pontine vein (white asterisk) via the left superior petrosal vein (white arrow heads), and then turning around anteromedially via the anterior pontomesencephalic veins (not shown) up to the right peduncular vein crossing the midline via the interpeduncular or posterior communicating vein (thin white arrow) to the left peduncular vein (black arrow head) terminating at the dilated left basal vein of Rosenthal (thick black arrows) on the opposite site.

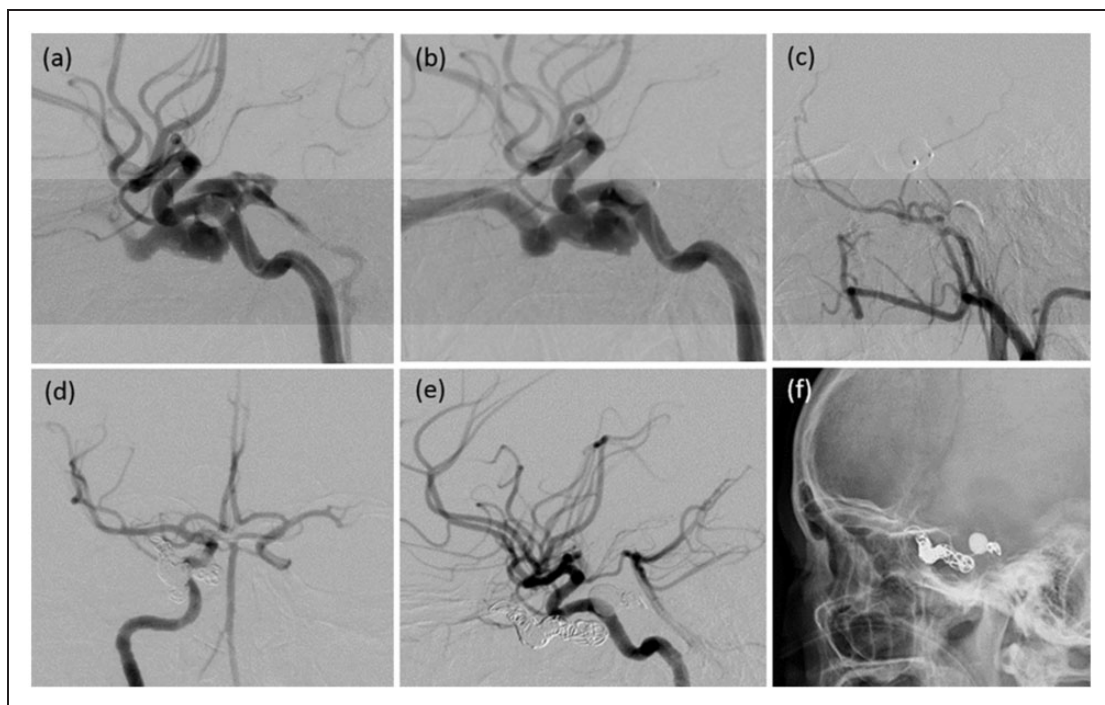


Figure 8. Lateral views of the right internal carotid artery (ICA) injections (a) and (b) reveal incomplete closure of anterior and posterior drainages of the traumatic carotid-cavernous fistula with only one detachable balloon via transarterial approach. Therefore, only the posterior drainage was closed initially, and another balloon was delivered to close the residual drainage. Lateral view of the right common carotid artery injection (c) shows sacrifice of the right ICA after inflation of another balloon. Then, transvenous approach was performed using fibered coils for occlusion of anterior drainage. Anteroposterior (d) and lateral (e) views of the right ICA injections confirm complete obliteration of the fistula with preservation of flow in the parent artery. Lateral radiograph of the skull (f) demonstrates the balloon and pack of deposited coils.

traumatic CCF in middle-aged woman with bilateral CCFs. There was progressive high pressure reflux into the sphenoparietal sinus, middle cerebral vein, and BVR, as well as diminished intracerebral arterial opacification. They believed that progressive intracranial arterial insufficiency accompanied by simultaneous venous hypertension may cause delayed deterioration in four months and death. From gross and microscopic examination, right parasylvian, orbital

gyri, and rostral pons, especially subcortical white matter, were swollen and boggy with elliptical and petechial hemorrhage associated with profoundly hypertrophied pial veins. In our opinion, these findings may correspond with extensive hemorrhagic venous infarction as in the current case.

Five years later, Vaghi et al.¹⁴ reported unusual appearances, that is, hyperdense lesion with perifocal hypodensity and mass effect of frontoparietal area, on

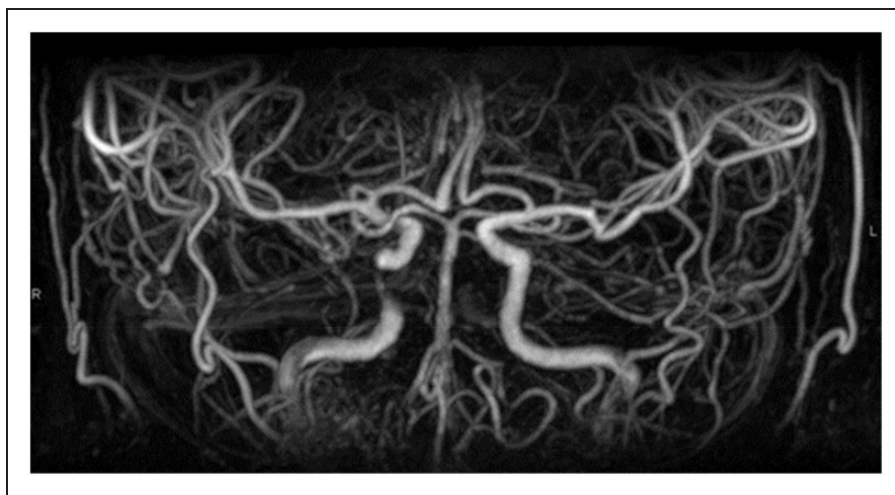


Figure 9. Anteroposterior view of three-dimensional time-of-flight magnetic resonance angiography obtained five months after embolization confirmed no residual right carotid-cavernous fistula.

a CT scan in a case of traumatic CCF. They speculated that these findings could be hemorrhagic venous infarction due to coincidence between the gradual regressions of the CT findings with the angiographic disappearance of the dilated veins after complete occlusion of the direct fistula by the detachable balloon.

We reviewed the literature of cerebral venous infarction or edema caused by venous congestion from direct CCFs. The data, including the sex, age, cause and side of fistula, location and side of brain edema, sinuses and/or veins involved, presence of hemorrhage, methods of treatment, and outcome of patients are described in Table 1.^{6,8,14-23} Including our one case, there were 14 cases (7 females and 7 males) with a median age of 48 years (range 24-88 years). Out of 14 fistulas, 8 were the result of traumatic causes, 5 spontaneous, and 1 iatrogenic. In one case of spontaneous CCFs an underlying connective tissue disorder was suspected.¹⁸ There were 6 fistulas located on the right side, and 8 on the left. Brain edema occurred on the same side in 9 cases, bilateral or diffuse on brainstem in 4, and contralateral in only 1 (i.e. our case). There were extensive edema (involving cerebral cortex, basal ganglia, thalamus, brain stem, and/or cerebellum) in 3 cases, including our case; only cerebral cortex in 2; brain stem and thalamus in 2; only brain stem in 4; only cerebellum in 1; brain stem, cerebellum and spinal cord in 1; and brain stem and spinal cord in 1. Venous infarction or edema presumably related to the BVR occurred in 6 cases, DMCV and/or superficial middle cerebral vein (SMCV) in 2, veins of brain stem in 6, uncal vein in 4, veins of cerebellum in 4, and veins of spinal cord in 3. Occlusion or stenosis of posterior segment of the SPS was found in 6 cases, including our case. Three of these 6 cases had no stenosis or occlusion of the SPS on prior angiography.^{8,16} There was no filling of the SPS in 5 cases. No apparent anterior venous drainage, that is, SOV, was observed in 2 cases.^{19,20}

Hemorrhagic transformation occurred in 8 cases, including our case.

After the onset of symptoms of direct CCFs, aggressive symptoms can develop within a few days, weeks, months, or years. Aggressive symptoms included headache, dysarthria or aphasia, hemiparesis or hemiplegia, hemiparesthesia, tetraparesis, cerebellar ataxia, cognitive impairment, respiratory insufficiency, and deterioration of consciousness. Three patients developed aggressive symptoms after incomplete treatment by embolization or surgical bypass. For almost all patients the symptoms finally developed into life-threatening conditions.

Hemorrhagic venous infarction is the infarction accompanied by hemorrhagic transformation. It may be secondary to increased venous pressure or venous congestion from the redirection of the venous outflow through the valveless veins or sinuses after some conditions, for example, sacrificing of some draining veins, thrombosis of the CS or draining veins, or stenosis of venous outlet.^{8,23,24}

Kamio et al.²³ demonstrated that cerebellar hemorrhage with surrounding edema resulted from direct CCF after performing maxillary surgery involving sacrifice of ipsilateral facial vein and pterygoid plexus. Sacrifice of these veins may alter the venous drainage and increase venous pressure in infratentorial veins. In addition, Aldea et al.²⁰ reported spontaneous direct CCF from a ruptured large cavernous carotid artery aneurysm blocking ipsilateral anterior venous drainage, producing rerouted retrograde flow into SPS, leading to hemorrhagic venous infarction of brainstem and cerebellum. Similarly, Herrera et al.¹⁹ proposed that spontaneous thrombosis of the anterior drainage with subsequent development of reflux into the peripontomesencephalic and perimedullary veins could explain progressive tetraparesis and respiratory insufficiency.

From knowledge of the embryologic processes, the SPS develops primarily as a drainage route from the

Table 1. Literature review of cerebral venous infarction or edema caused by venous congestion from direct carotid-cavernous fistulas.

Authors	Sex/Age	Cause	Fistula side	Edema side	Location	Involved sinuses/veins	Hemorrhage	Endovascular treatment	Surgery	Outcome
Vaghi, 1983 ¹⁴	M/54	Trauma	R	R	FL, PL	SPS, ^a SMCV, DMCV	Yes	Balloon	No	GR
Kasama, ^b 1990 ¹⁵	F/74	Spontaneous	L	L	TL	N/A	Yes	Balloon	No	N/A
Teng, 1991 ⁸	M/36	Trauma	L	L	MB, T	SPS, ^c IPS, BVR	Yes (TL)	Balloon (3rd)	Bypass (1st) ^d DC/EH (2nd)	GR
Murata, 2003 ¹⁶	M/26	Trauma	L	R, L (diffuse)	MB, T	SPS, ^c IPS, UV, BVR	No	Coils (V) (1st) ^d Sacrifice (2nd)	No	GR
Ohshima, ^b 2006 ¹⁷	F/41	Spontaneous	L	L	P	SPS, ^c ATPV	Yes	Coils (A)	No	GR
	F/88	Spontaneous	L	L	FL, TL, BG, T, MB, P	N/A	Yes	N/A	N/A	N/A
Bussière, 2009 ¹⁸	M/39	Spontaneous ^e	L	R, L (diffuse)	P, ME	SPS, ^a Ped.V, APMV, AMV, CV, ASV	No	Balloon	No	GR
Herrera, 2011 ¹⁹	M/27	Trauma	R	R, L (dorsal)	MB, P, ME, SC	SPS, ^c PPMV, PMV (No SOV)	No	Coils (A)	No	PR
Aldea, 2011 ²⁰	F/55	Spontaneous	L	L	P, CE, SC	SPS, CV, ASV (No SOV)	Yes	Sacrifice by balloons (A) (1st)	DC/EH (2nd)	GR
Ract, 2014 ²¹	F/45	Trauma	R	R	TL, BG, MB, P, CP	SPS, ^a UV, BVR, ISV	No	Sacrifice by coils (A)	No	GR
Yu, 2015 ⁶	M/51	Trauma	R	R	P	SPS, ^a TPV, APMV, BVR	No	Balloon (A)	No	GR
Ryu, 2017 ²²	M24	Trauma	R	R, L (ventral)	MB	SPS, ^a BVR, UV, IPed.V	No	Coils (V) (1st) ^d Proximal trapping (3rd)	Bypass/ distal trapping (2nd)	GR
Kamio, 2017 ²³	F/63	Iatrogenic	L	L	CE	SPS, ^c PV, CV	Yes	Sacrifice (A) (V)	No	GR VL (L)
Present study, 2018	F/54	Trauma	R	L	FL, TL, BG, T, MB, P, CE	SPS, ^c PV, UV, DMCV, Ped.V, IPed.V, BVR, ISV, CV	Yes	Balloon (A) Coils (V)	No	GR

^aNo filling of the SPS on angiography.^bDetails from only English abstract.^cStenosis or occlusion of posterior segment of the SPS.^dIncomplete treatment.^eUnderlying connective tissue abnormality.

A: arterial approach; AMV: anterior medullary veins; APMV: anterior pontomesencephalic vein; ASV: anterior spinal vein; ATPV: anterior transpontine vein; BG: basal ganglia; BVR: basal vein of Rosenthal; CE: cerebellum; CP: cerebellar peduncle; CV: cerebellar veins; DC: decompressive craniectomy; DMCV: deep middle cerebral vein; EH: evacuation of hematoma; F: female; FL: frontal lobe; GR: good recovery; IPed.V: interpeduncular vein; IPS: inferior petrosal sinus; ISV: inferior striate veins; L: left; M: male; MB: midbrain; ME: medulla; N/A: data not available; P: tons; Ped.V: peduncular vein; PL: parietal lobe; PMV: perimedullary vein; PPMV: peripontomesencephalic vein; PR: partial recovery; PV: petrosal vein; R: right; SC: spinal cord; SMCV: superficial middle cerebral vein; SOV: superior ophthalmic vein; SPS: superior petrosal sinus; T: thalamus; TL: temporal lobe; TPV: transverse pontine vein; UV: uncus vein; V: venous approach; VL: visual loss; 1st: first procedure; 2nd: second procedure; 3rd: third procedure.

petrosal vein, collecting blood from the anterior cerebellar and brainstem venous drainage systems.²⁵ Therefore, the SPS provides an important route for infratentorial venous drainage from the brainstem and cerebellum via the petrosal vein.²³ SPS was divided into anterior and posterior segments.²⁶ Hypoplasia or occlusion of this sinus may promote venous congestion of the venous system in surrounding brainstem, leading to edema and/or hemorrhage of brainstem and/or cerebellum.^{8,16,19,23}

Importantly, incomplete closure of direct CCF should be avoided due to the possibility of the rerouting of venous drainage into cortical or deep venous systems, leading to cerebral edema and/or hemorrhage, rather than resolution of the fistula. Further complete obliteration of the residual fistula or drainage should be achieved when possible.^{8,22}

To understand the possible mechanisms of cerebral hemorrhagic venous infarction from the CCF, it is important to know the essential anatomy of the BVR, basal cerebral venous drainage of the CS, basal cranial venous sinus, and bridging veins communicating between the CS and the brain.

Based on a venous study by Huang,²⁷ the BVR was divided into three segments, that is, anterior, middle, and posterior. The first segment (striate segment), passing posteromedially above the uncus to reach the anterior part of the cerebral peduncle, is formed mainly by the union of the DMCV and its insular tributaries, the inferior striate veins, the olfactory vein, the orbitofrontal vein, and the anterior cerebral vein. The second segment (peduncular segment), running further posterolaterally around the most lateral portion of the cerebral peduncle and turning posteromedially to the lateral mesencephalic sulcus, is formed mainly by the union of the peduncular vein, the inferior thalamic vein, the inferior ventricular veins, and the uncal vein. Last, the third segment (posterior mesencephalic segment), joining the great vein of Galen or the posterior portion of the internal cerebral vein by running posteromedially and superiorly, is formed mainly by the union of the lateral mesencephalic vein (LMV), the lateral atrial vein, the medial occipitotemporal vein, the posterior longitudinal hippocampal vein, and the posterior thalamic vein.

During late embryogenesis after regression of the tentorial sinus, the BVR was formed by four groups of afferent veins described by Terbrugge and Lasjaunias.²⁸ First, the telencephalic group receives drainage from the olfactory vein, the inferior frontal vein, the anterior communicating vein, inferior striate veins, and the uncal vein. Second, the diencephalic group carries blood from the hypothalamic vein, the hippocampal vein, the inferior thalamic vein, the inferior ventricular vein, the inferior choroidal vein, the posterior communicating vein, the peduncular vein, the anterior pontomesencephalic vein (APMV), and the infratemporal vein. Third, the tegmental group drains blood from the LMV, and the posterior mesencephalic

vein. Last, the tectal group collects blood from the tectal vein, the tectogeniculate vein, and the precentral vein.

The venous drainage from the brain mainly drains into the CS through bridging veins including the SMCV, SPV, and bridging vein from the brainstem.²⁹ The petrosal vein is a bridging vein connecting and receiving venous blood from veins of cerebellum and brainstem by connection with the third segment of the BVR through the LMV and with the APMV through the TPV. The bridging veins from the posterior part of the CS may connect to the medial APMV or TPV.^{20,30-32}

The inferior striate veins drain a large area above the anterior perforated substance including the putamen, caudate nucleus, and internal capsule.³³ In the present case, the presence of abnormal hyperintensity signal on FLAIR images of the putamen, caudate nucleus, and internal capsule represent radiographic evidence of brain edema resulting from venous congestion of tributaries of the inferior striate veins draining into the BVR.

To the best of our knowledge, our patient is the first report of a posttraumatic CCF creating contralateral cerebral hemorrhagic venous infarction. Lau et al.³⁴ reported an atypical case of the left spontaneous direct CCF causing venous hypertension in the right SMCV from cross-flow through intercavernous sinus, resulting in contralateral intracerebral hematoma. Interestingly, this fistula resolved spontaneously without intervention. Similarly, hemorrhagic venous infarction in the current case occurred opposite to the direct fistula through posterior intercavernous sinus and also the posterior communicating vein. An anastomotic venous circle of veins was described by Cullen et al.³⁵ This venous circle is formed by the anterior and posterior communicating veins, linking bilateral BVRs across the midline, representing a route of contralateral venous drainage. It may become an essential route for the absence of segments of the BVR or high-flow arteriovenous shunts accessing the deep venous system. Our case may be the suitable example of this speculation due to the absence of the right BVR in high-flow traumatic CCF, resulting in redirection of venous drainage to the contralateral BVR.

Fortunately, our patient has regained almost a complete recovery, both clinically and radiologically, from the life-threatening condition after performing endovascular treatment without surgical intervention, teaching us to know about the natural history of this very rare consequence of traumatic CCF. However, it is imperative to obliterate the fistula before hemorrhagic transformation when brain edema caused by venous congestion from direct traumatic CCF is early diagnosed. Early diagnosis can be achieved by FLAIR and SWI images.

Abnormal hyperintensity within the venous structures on the SWI images represents arterialized blood flow in the veins from arteriovenous shunting in vascular malformations of the brain or from cortical venous

reflux in dural arteriovenous fistulas (DAVFs).^{36,37} However, we also found abnormal hyperintensity and flow void of the left BVR on TOF and T2-weighted images, respectively.

The BVR and veins around brainstem are often difficult to identify on routine angiography. Using angiographic CT with a flat panel detector, these small veins can be visible in multiplanar reformations and MIP reformatted images as shown in the present case.

Based on the secondary longitudinal anastomoses between embryonic veins (i.e. the deep telencephalic vein, ventral and dorsal diencephalic vein, and mesencephalic vein) by Padget,²⁵ Suzuki et al.³⁸ classified variations of the BVR using three-dimensional CT angiography into five drainage pathways including draining to the great vein of Galen, to the CS or sphenoparietal sinus, to the SPS through the LMV, to the SPV through the peduncular vein, and finally to the transverse or straight sinus through the primitive tentorial sinus. In the present case, the BVR not only drains into the great vein of Galen, but also connects the CS through the dilated uncal vein. By the axial angiographic CT in MIP reformatted images, we found another anastomosis to the peduncular vein, which was rarely seen in the study of Suzuki et al.³⁸

Compared with low-flow fistulas, CS DAVFs (CSDAVFs) may also rarely manifest as venous infarction and/or intracerebral hemorrhage.^{7,30} Miyamoto et al.³⁰ found that angiographic risk factors were cortical venous drainage only, ectasia of the draining vein, agenesis of the second and third segment of the BVR, and thrombosis of the SOV, distal segment of the SPS, and distal cortical vein drainage. From study of anatomical variations of the uncal vein in normal and CSDAVFs by Ide et al.,⁷ the uncal vein is a small cerebral vein communicated between the CS and BVR. It may terminate into the CS or SMCV. The connection between the first and second segments of the BVR is usually hypoplastic when there is a well-developed uncal vein. The results of our literature search found that there was a dilated uncal vein in four cases, including our case.^{8,21,22} However, a hypoplastic segment of the BVR was shown on angiography only in our case and Ract et al.'s case.²¹ Shimada et al.²⁶ studied visualization and hemodynamic features of the SPS in normal and CSDAVFs. The SPS frequently has segmentally hypoplastic features on angiography. A higher risk of venous reflux into the cerebellopontine veins occurred in CSDAVFs associated with hypoplastic posterior segment of the SPS. Similarly, our case and another five cases from our review had occlusion or hypoplastic posterior segment of the SPS, resulting in edema of brainstem, cerebellum, and/or spinal cord.^{8,16,19,23} Three of five cases were confirmed spontaneous thrombosis of the SPS because prior angiography revealed the patent SPS.^{8,16} The SPS was not identifiable in another five cases, and probably thrombosed.^{6,8,14,21,22}

Interestingly, Chan et al.¹² reviewed brainstem dysfunction with venous congestion and/or hemorrhage

caused by CCF (i.e. 9 direct and 13 indirect fistulas). Most direct CCFs had engorged veins in surrounding brainstem on angiography, while most indirect fistulas had venous congestion without engorged veins. Four cases had brainstem hemorrhage caused by direct fistulas.

Endovascular treatment currently has been the therapy of choice for traumatic CCFs, and consists of a transarterial and transvenous approach. A venous approach, via mainly IPS or ophthalmic vein, may be used in case of unsuccessful transarterial embolization or previous ligation of the ICA. Emergency treatment is indicated in the setting of progressive visual loss, epistaxis from pseudoaneurysm extended into sphenoid sinus, rapid aggravation of ophthalmic symptoms from migration of a treated previous balloon, and unconscious patients with exclusion of other intracranial lesions.⁵ Emergency treatment with balloon embolization followed by immediate removal of hematoma has been recommended for a life-threatening condition in traumatic CCF with intracerebral venous hemorrhage.¹⁰ Without previous closure of the high-flow fistula, uncontrollable hemorrhage may occur and lead to disastrous results.^{11,22} Therefore, cerebral edema or infarction due to venous congestion in direct CCF should be considered as urgency or emergency conditions, and requires prompt endovascular treatment. From the present review, endovascular treatments were performed through arterial and/or venous routes with balloon and/or coils in the majority of these cases. Four CCFs needed to be sacrificed or trapped. Only two cases required decompressive craniectomy with removal of a blood clot. Most patients had good recovery except two patients had no data available. One case had partial recovery, and another case still has lost vision of the left eye.^{19,23}

Generally, the impairment of venous drainage from thrombosis of the cerebral deep venous system located within the core of the brain, that is, the internal cerebral veins, the BVR, or the great vein of Galen, may lead to irreversible hemorrhagic venous infarction, mainly in cerebral white matter.³⁹ In contrast, most patients in the present review have returned to normal after obliteration of the fistula, representing that hemorrhagic or non-hemorrhagic venous infarction caused by venous hypertension from direct CCF was a more reversible process.

To the best of our knowledge, our case is probably the most extensive venous infarction, involving frontal lobe, temporal lobe, basal ganglia, thalamus, brainstem, and cerebellum. Only two cases of extensive cerebral edema and/or hemorrhagic transformation from direct CCF have been previously reported. Ohshima et al.¹⁷ reported extensive hemorrhagic venous infarction, involving left frontal lobe, temporal lobe, basal ganglia, thalamus, midbrain, and pons, associated with left spontaneous CCF. They speculated that these findings possibly associated with hypovolemia and perioperative infection during biopsy of the left ethmoidal sinus. Ract et al.²¹ also reported a patient

with a life-threatening traumatic CCF associated with extensive edema of ipsilateral temporal lobe, basal ganglia, and brainstem caused by venous congestion of an anatomical variation of the BVR. They speculated that venous infarction-related factors included large-sized fistula, a hypoplastic segment of the BVR with developed uncal vein, and a hypoplastic or thrombosed SPS. Similarly, our case also had a hypoplastic segment of the BVR, dilated uncal vein, and a hypoplastic or thrombosed SPS. However, the fistula in our case was small, located opposite the side of the brain, and developed more extensive venous infarction with hemorrhagic transformation.

Aldea et al.²⁰ suggested using anticoagulant drugs in order to avoid massive thrombosis of the venous drainage, causing a progressive worsening of the neurological status, after the occlusion of the high-flow CCF. Despite no anticoagulation for prevention of the ongoing venous thrombosis following the obliteration of the CCF, this condition did not occur in our case. However, this management remains controversial, and there are still no evidence-based guidelines.

Conclusion

We reported an extremely rare case of contralateral extensive hemorrhagic venous infarction caused by a traumatic CCF related to retrograde venous reflux into the opposite BVR. The cause of this condition may relate to absence of the ipsilateral BVR, occlusion of posterior segment of the contralateral SPS, and a developed uncal vein with hypoplastic second and third segments of the contralateral BVR. Neurologically and radiologically, the extensive hemorrhagic venous infarction has nearly completely resolved following obliteration of the fistula by endovascular treatment without surgical intervention. Cerebral edema or infarction due to venous congestion in direct CCF should be considered as urgency or emergency conditions, and requires prompt endovascular treatment with complete obliteration of the fistula.

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