Carotid-Cavernous Fistula Associated with an Intracranial Lesion Caused by Cortical Venous Reflux

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Summary

Digital subtraction angiography (DSA) and magnetic resonance imaging (MRI) findings in 20 patients with carotid-cavernous fistula (CCF; 3 direct CCFs and 17 indirect CCFs) were retrospectively reviewed to evaluate venous drainage patterns that may cause intracerebral haemorrhage or venous congestion of the brain parenchyma.

We evaluated the relationship between cortical venous reflux and abnormal signal intensity of the brain parenchyma on MRI. Cortical venous reflux was identified on DSA in 12 of 20 patients (60.0%) into the superficial middle cerebral vein (SMCV; $n=4$), the uncal vein ($n=2$), the petrosal vein ($n=2$), the lateral mesencephalic vein (LMCV; $n=1$), the anterior pontomesencephalic vein (APMV; $n=1$), both the APMV and the petrosal vein ($n=1$) and both the uncal vein and the SMCV ($n=1$). Features of venous congestion, such as tortuous and engorged veins, focal staining and delayed appearance of the veins, were demonstrated along the region of cortical venous reflux in the venous phase of internal carotid or vertebral arteriography in six of 20 patients (30.0%). These findings were not observed in the eight CCF patients who did not demonstrate cortical venous reflux. MRI revealed abnormal signal intensity of the brain parenchyma along the region with cortical ve-

nous reflux in four of 20 indirect CCF patients (20%). Of these four patients, one presented with putaminal haemorrhage, while the other three presented with hyperintensity of the pons, the middle cerebellar peduncle or both on T2-weighted images, reflecting venous congestion. The venous drainage routes were obliterated except for cortical venous reflux in these four patients and the patients without abnormal signal intensity on MRI had other patent venous outlets in addition to cortical venous reflux.

CCF is commonly associated with cortical venous reflux. The obliteration or stenosis of venous drainage routes causes a converging venous outflow that develops into cortical venous reflux and results in venous congestion of the brain parenchyma or intracerebral haemorrhage. Hyperintensity of brain parenchyma along the region of cortical venous reflux on T2-weighted images reflects venous congestion and is the crucial finding that indicates concentration of venous drainage into cortical venous reflux.

Introduction

Carotid-cavernous fistula (CCF) is angiographically classified into direct CCF and indirect CCF1). Direct CCF occurs secondary to laceration of the internal carotid siphon caused by head injury or rupture of an internal carotid

Figure 1 Case 5. 73-year-old female with indirect CCF. Non-contrast CT (A) demonstrates left putaminal haemorrhage. Hypodense area is seen on the left frontal operculum in addition to perifocal edema. Axial T2-weighted image (B) demonstrates a mildly hyperintense area corresponding to the putaminal haemorrhage. An area of hyperintensity is apparent around the hematoma and on the left frontal operculum. Left putaminal haemorrhage shows mildly hyperintensity on axial T1-weighted image (C). The hypointense area is also seen on the left frontal operculum. Early venous phase of right common arteriogram in a frontal projection (D) shows DAVF of the right cavernous sinus with cortical venous reflux into the left SMCV (black arrow) via the intercavernous sinus and the left cavernous sinus. Although bilateral SOVs are also venous drainage routes, they are obliterated at the extraconic portion (white arrows). An anastomotic vein (double black arrows) with the SMCV and the superior sagittal sinus is small in caliber. Right external arteriogram in a frontal projection (E) immediately following TVE. Left SMCV (black arrow) and SOV (double black arrows) were embolized at the junction with the left cavernous sinus. Following TVE, the CCF drains into the reopened left IPS (white arrow).

aneurysm^{1,2}. Indirect CCF is abnormal communication between the dural branches of the internal or external carotid arteries and the cavernous sinus^{1,2}. Indirect CCF is a synonym with cavernous sinus dural arteriovenous fistula (DAVF). Although these are the different entities, both usually present with the same symptoms such as ophthalmopathy or pulsatile tinnitus and rarely cause intracerebral haemorrhage or venous infarction¹⁻⁵. Symptoms are dependent on the direction of venous outflow of the

CCF¹⁻⁵. In this study we evaluated the brain parenchymal lesions caused by the cortical venous reflux of CCF.

Methods

Subjects comprised 20 consecutive patients with angiographically proven CCF. They consist of 3 direct CCF patients (one male and two females, age range 33-51 years, mean age 44.6 years) and 17 indirect CCF patients (three ma-

les and 14 females, age range 48-77 years, mean age 65.3 years). All CCF patients were symptomatic, and complained of ophthalmopathy or pulsatile tinnitus.

All direct CCFs occurred after head injury. Onset in one indirect CCF patient involved intracerebral haemorrhage caused by cortical venous reflux into the superficial middle cerebral vein (SMCV). All CCF patients underwent diagnostic digital subtraction angiography (DSA) and embolotherapy following admission to our institute. Two patients with direct CCF underwent transcatheter intra arterial embolization (TAE) using detachable balloons and remaining 18 patients with direct or indirect CCF underwent tranvenous embolization (TVE) using microcoils.

Magnetic resonance imaging (MRI) was performed on a 1.5 T superconducting unit with a standard head coil within one week prior to embolization and again one-two weeks after embolization. Routine T1-weighted imaging before and after the intravenous administration of gadopentetate dimeglumine (Gd-DTPA) and routine T2-weighted fast spin-echo imaging was obtained with the following parameters: for T1-weighted imaging, TR/TE, 380-420/9-12; and for T2-weighted imaging, TR/TE, 4000/101-106. All images were obtained with a field of view of 18 or 22 cm and a 192 x 256 matrix. Section thickness was 3 or 4 mm. Presence or absence of cortical venous reflux and related abnormal signal intensity of the brain parenchyma were evaluated by DSA and MRI. Venous congestion was also evaluated in the region of the cortical venous reflux in the venous phase of internal carotid or vertebral arteriography.

Two patients with cortical venous reflux with hyperintensity of the brain stem on T2-weighted images and another with normal features on MRI underwent single photon emission computed tomography (SPECT) using I-123 n-isopropyl-p-iodoamphetamine (IMP) or Tc-99m-hexamethylene propyleneamine oxime (HMPAO) before and after TVE.

Results

Indirect CCF Group

DSA identified cortical venous reflux in ten of 17 indirect CCF patients (63%), and drained into the SMCV (n=2), the uncal vein (n=2), the petrosal vein (n=2), the lateral mesencephalic

vein (LMCV; n=1), the anterior pontomesencephalic vein (APMV; n=1), both the APMV and the petrosal vein (n=1) and both the uncal vein and the SMCV (n=1; table 1). In four patients MRI identified abnormal signal intensity of the brain parenchyma in the region of cortical venous reflux, including the region from the basal ganglia to the frontal operculum (n=1), the pons (n=1) and the region from the pons to the middle cerebellar peduncle (n=2). In the patient with intracerebral haemorrhage, the fistula was situated on the right cavernous sinus and the venous outflow drained into the left SMCV via the intercavernous sinus (figure 1). Other venous drainages into the bilateral superior ophthalmic veins (SOVs) were demonstrated but they were occluded at the extraconic portion. The anastomotic vein of the left SMCV with the superior sagittal sinus had been small in caliber and the venous outflow concentrated into the left SMCV and its tributaries. In this patient, CT revealed putaminal haemorrhage, while MRI demonstrated hyperintensity on both the putamen and the frontal operculum on T2-weighted images.

In the three patients with hyperintensity of the pons or the middle cerebellar peduncle on T2-weighted images, cortical venous reflux was identified into the petrosal vein, the APMV, or both (figure 2). Two of these patients showed ipsilateral contrast enhancement of the pons demarcated by the pontine raphe, and another revealed no contrast enhancement of the lesion on contrast-enhanced T1-weighted images. Hyperintensity on T2-weighted images and enhancement on the contrast study disappeared or decreased in size on the follow-up MRI after TVE in all three patients.

Features of venous congestion such as tortuous and engorged veins, focal staining and delayed appearance of the veins were demonstrated on the region of cortical venous reflux in the venous phase of the internal carotid or vertebral arteriography in six of ten patients (60.0%). These findings were not shown in seven CCFs without cortical venous reflux. In the three patients with hyperintensity of the pons or the middle cerebellar peduncle on T2-weighted images by MRI, delayed enhancement was identified on the region of cortical venous reflux in the venous phase of vertebral arteriography (figure 2D).

In these patients, because the focal staining continued through the late venous phase on the

Figure 2 Case 3. 77-year-old female with indirect CCF, A-E: before TVE, F-I: after TVE. Axial T2-weighted image (A) shows hyperintensity of the left middle cerebellar peduncle and serpentine flow voids on the left cerebellar hemisphere. On post-contrast axial T1-weighted image (B), no enhancement is seen in the left middle cerebellar peduncle. Dilated cortical veins are enhanced on the left cerebellar hemisphere. Left external arteriogram in a lateral projection (C) demonstrates left-sided CCF with cortical venous reflux into the left APMV (white arrow) and the petrosal vein (black arrow) via the SPS. The spinal venous drainage (double white arrows) is shown via the lateral medullary vein. Left SOV is demonstrated as another venous drainage but is obliterated at the extraconic portion (double black arrows). Venous phase of the left vertebral arteriogram in a Townes projection (D) demonstrates diffuse staining on the left cerebellar hemisphere and left side of the brain stem. The veins are poorly demonstrated within the stained region. I-123 IMP SPECT before TVE (E) shows decreased CBF of the left cerebellar hemisphere (arrows). Left common arteriogram in a lateral projection 3 months after TVE (F) demonstrates disappearance of CCF. Venous phase of left vertebral arteriogram in a Townes projection 3 months after TVE (G) shows marked decrease of the staining and normalization of venous features. I-123 IMP SPECT 1 week after TVE (H) shows increased CBF of the left cerebellar hemisphere (arrow) compared with the previous study (E). Axial T2-weighted image 1 week after TVE (I) shows decreased hyperintensity of the left cerebellar peduncle. The serpentine flow voids on the left cerebellar hemisphere disappeared.

Table 1 MRI, DSA and SPECT findings of CCF with cortical venous reflux.

Case	CCF Type	MRI		DSA			SPECT CBF
		T2-hyper	CE	CVR	other drainages	venous congestion	
1	indirect	pons-MCP	positive	petrosal v.	ips SOV(occluded)	positive	decreased
2	indirect	pons	positive	APMV	blt SOVs	positive	not done
3	indirect	MCP	negative	petrosal v.,APMV	ips SOV(occluded)	positive	decreased
4	indirect	none	negative	petrosal v.	ips SOV,ips IPS	negative	not done
5	indirect	putamen	negative	SMCV	blt SOVs(occluded)	positive	not done
6	indirect	none	negative	SMCV	blt SOVs,contra IPS	positive	not done
7	indirect	none	negative	SMCV,uncal v.	ips IPS	positive	decreased
8	indirect	none	negative	uncal v.	blt SOVs	negative	not done
9	indirect	none	negative	uncal v.	ips SOV	negative	not done
10	indirect	none	negative	LMCV	blt SOVs,ips IPS	negative	not done
11	direct	none	negative	SMCV	blt SOVs,blt IPSes	obscure	decreased
12	direct	none	negative	SMCV	blt SOVs,blt IPSes	obscure	not done

MRI, magnetic resonance imaging; DSA, digital subtraction angiography; SPECT, single photon emission computed tomography; CCF, carotid-cavernous fistula; T2-hyper, hyper signal intensity on T2-weighted image; CE, contrast enhancement of the brain parenchyma; CVR, cortical venous reflux; CBF, cerebral blood flow; MCP, middle cerebellar peduncle; petrosal v., petrosal vein; APMV, anterior pontomesencephalic vein; SMCV, superficial middle cerebral vein; uncal v., uncal vein; LMCV, lateral mesencephalic vein; ips, ipsilateral; SOV, superior ophthalmic vein; blt, bilateral, IPS, inferior petrosal sinus; contra, contralateral.

ipsilateral cerebellar hemisphere and the brain stem, identification of the veins was difficult in the staining lesion. Angiography demonstrated improvement in venous congestion after TVE in these patients (figure 2G).

I-123 IMP or Tc-99m HMPAO SPECT was performed in the three patients with cortical venous reflux into the petrosal vein, the APMV and the SMCV, respectively. They revealed decreased cerebral blood flow (CBF) on the region of cortical venous reflux, that improved after TVE in all three patients (figure 2E,H).

All 17 patients underwent TVE using interlocking detachable coils (IDCs) or fibered platinum coils (FPCs) via the inferior petrosal sinus (IPS) or the facial vein by femoral approach or SOV puncture. Obliteration of CCF was obtained in 16 patients. Although CCF was located in the right cavernous sinus in the patient with left-sided intracerebral haemorrhage due to cortical venous reflux into the left SMCV, catheterization was not achieved into the right cavernous sinus because of the right IPS

thrombosis. To prevent cortical venous reflux into the haemorrhagic portion, the micro-catheter was navigated via the left IPS and palliative embolization was accomplished at the junction of the left cavernous sinus with the left SMCV and the left SOV (figure 1).

Direct CCF Group

DSA identified cortical venous reflux into the SMCV in two of three direct CCF patients (66.7%; table 1). MRI demonstrated no abnormal signal intensity on the brain parenchyma along the cortical venous reflux in both patients. Angiographic evaluation of veins was difficult on ipsilateral internal carotid arteriography because of insufficient vessel opacification of contrast medium by steal phenomenon.

Tc-99m HMPAO SPECT was performed on the patient with cortical venous reflux into the SMCV and decreased CBF was identified on the ipsilateral cerebral hemisphere, that improved after TVE.

CCF disappeared in all patients and symp-

toms resolved following embolization. One direct CCF patient underwent TAE at the fistulous portion of the internal carotid artery with a fetal type of the posterior communicating artery after the ipsilateral external carotid-internal carotid artery bypass because of hypoplasia of both anterior communicating artery and proximal portion of the ipsilateral posterior cerebral artery.

Discussion

Haemorrhagic stroke or venous congestion is sometimes associated with DAVFs^{4,9}. Awad et Al.⁷ described that retrograde leptomeningeal venous drainage (cortical venous reflux), variceal or aneurysmal venous dilatations, and galenic drainage were significant factors predisposing to aggressive neurological presentations. These complications are more common in DAVF of the transverse-sigmoid sinus or the tentorium than DAVF of the cavernous sinus^{6,8}. This is probably because the cavernous sinus communicates with numerous venous sinuses or veins, such as the SOV, the inferior ophthalmic vein, the SMCV, the venous sinus of the lesser sphenoid wing (the sphenoparietal sinus), the uncal vein, the pterygoid plexus, the superior petrosal sinus (SPS), the IPS and the contralateral cavernous sinus via the intercavernous sinus, shunt flow from the DAVF does not easily converge into the cortical vein and will readily diverge⁹. This logic is also applied to direct CCF. CCF usually presents with ophthalmopathy or pulsatile tinnitus and rarely cause haemorrhage or venous congestion of the brain parenchyma due to cortical venous reflux^{1-5,9-12}.

Willinsky et Al.⁸ described cortical venous reflux in 27% of patients with indirect CCF, compared with 42% and 100% of those with transverse sinus and tentorial DAVFs, respectively. In our study, cortical venous reflux was seen in ten of 17 patients (63%) with indirect CCF. The reason for the higher occurrence in our study compared with that of Willinsky et Al.⁸ is unclear. One reason may be that the number of patients in our study is relatively small. Angiographical evidence of venous congestion was seen in six of ten indirect CCF patients (60%) with cortical venous reflux in our study. Willinsky et Al.⁸ used the term "pseudophlebitic pattern" (PPP) to describe findings such as tortuous and engorged veins identified

on the venous phase of brain circulation in patients with venous congestion related to an intracranial DAVF. Willinsky et Al. observed PPP was seen in 46 of 57 patients (81%) with cortical venous reflux, and ten patients with PPP showed brain parenchymal hyperintensity on T2-weighted images. In our study, four of six patients with PPP revealed abnormal signal intensity of the brain parenchyma. Of the four patients with PPP, one presented with intracerebral haemorrhage and the other with venous congestion. Although two patients with PPP exhibited normal signal intensity of the brain parenchyma with MRI, one of them demonstrated decreased CBF in the region of cortical venous reflux on I-123 IMP SPECT.

In the group with abnormal signal intensity on MRI, a common angiographic finding was the convergence of the shunt flow of CCF into the cortical vein due to the obliteration of other venous drainage routes. Two patients with both PPP and no abnormal signal intensity had other patent venous outlets in addition to cortical venous reflux.

Several previous studies^{10,11} referred to the MR findings of the brain parenchymal venous congestion caused by cortical venous reflux in CCFs. Most of the cases revealed enlargement and hyperintensity of the brain stem or the cerebellar hemisphere on T2-weighted images. Previous studies refer to haemorrhagic complication of CCFs caused by cortical venous reflux into the sylvian vein or the vein of lateral recess of the 4th ventricle^{2,9,12}.

In these cases, DSA demonstrated CCF with cortical venous reflux and the obliteration or stenosis of other venous drainage routes as with our cases. DAVFs frequently cause thrombosis of the venous drainage routes and a change in the direction of venous drainage, both resulting in cortical venous reflux^{4-8,13}. CCFs with cortical venous reflux are considered to have a high risk of bleeding or venous congestion^{2,4-8}. An association of these conditions with PPP indicates a high risk pattern⁸. Brain parenchymal hyperintensity in the region of cortical venous reflux on T2-weighted images is the clearest indicator of a concentration of venous drainage into cortical venous reflux. These findings improved following TVE, indicating that the condition is reversible. Urgent treatment is therefore required to obliterate the fistula as venous congestion may lead to venous infarction or haemorrhage^{2,11,14}.

Conclusions

CCFs are commonly associated with cortical venous reflux.

The obliteration or stenosis of venous drainage routes causes a converging venous outflow into cortical venous reflux and results in

venous congestion of the brain parenchyma or intracerebral haemorrhage. Brain parenchymal hyperintensity in the region of cortical venous reflux on T2-weighted images reflects venous congestion and is clear indicator of a concentration of venous drainage into cortical venous reflux.

References

- 1 Barrow DL, Spector RH et Al: Classification and treatment of spontaneous carotid-cavernous fistulas. *J Neurosurg* 62: 248-256, 1985.
- 2 Halbach V, Hieshima GB et Al: Carotid cavernous fistulae: indications for urgent treatment. *Am J Neuroradiol* 8: 627-633, 1987.
- 3 Newton TH, Hoyt WF: Dural arteriovenous shunts in the region of the cavernous sinus. *Neuroradiology* 1: 71-81, 1970.
- 4 Davies MA, Brugge K et Al: The natural history and management of intracranial dural arteriovenous fistulae part 2: aggressive lesions. *Interventional Neuroradiology* 3: 303-311, 1997.
- 5 Satomi J, Satoh K et Al: Angiographic changes in venous drainage of cavernous sinus dural arteriovenous fistulae after palliative transarterial embolization of observational management: a proposed stage classification. *Neurosurgery* 56: 494-502, 2005.
- 6 Malik GM, Pearce JE et Al: Dural arteriovenous malformations and intracranial haemorrhage. *Neurosurgery* 15: 332-339, 1984.
- 7 Awad IA, Little JR et Al: Intracranial dural arteriovenous malformations: factors predisposing to an aggressive neurological course. *J Neurosurg* 72: 839-850, 1990.
- 8 Willinsky R, Goyal M et Al: Tortuous, engorged pial veins in intracranial dural arteriovenous fistulas: correlations with presentation, location, and MR findings in 122 patients. *Am J Neuroradiol* 20: 1031-1036, 1999.
- 9 Kuwayama N, Endo S et Al: Surgical transvenous embolization of a cortically draining carotid cavernous fistula via a vein of the sylvian fissure. *Am J Neuroradiol* 19: 1329-1332, 1998.
- 10 Uchino A, Kato A et Al: Pontine venous congestion caused by dural carotid-cavernous fistula: report of two cases. *Eur Radiol* 7: 405-408, 1997.
- 11 Takahashi S, Tomura N et Al: Dural arteriovenous fistula of the cavernous sinus with venous congestion of the brain stem: report of two cases. *Am J Neuroradiol* 20: 886-888, 1999.
- 12 Satoh K, Satomi J et Al: Cerebellar haemorrhage caused by dural arteriovenous fistula: a review of five cases. *J Neurosurg* 94: 422-426, 2001.
- 13 Cognard C, Houdart E et Al: Long-term changes in intracranial dural arteriovenous fistulae leading to worsening in the type of venous drainage. *Neuroradiology* 39: 59-66, 1997.
- 14 Zeidman SM, Monsein LH et Al: Reversibility of white matter changes and dementia after treatment of dural fistulas. *Am J Neuroradiol* 16: 1060-1063, 1995.

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