Acta Neurochir (Wien) (2007) 149: 675–680
DOI 10.1007/s00701-007-1229-7
Printed in The Netherlands

Acta Neurochirurgica

Clinical Article

Bilateral traumatic carotid-cavernous fistulae: Strategies for endovascular treatment

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Received November 23, 2006; accepted April 24, 2007; published online June 11, 2007

Summary

Background. Most traumatic carotid-cavernous fistula/e (TCCF) are unilateral, and simultaneous bilateral TCCF are uncommon. The purpose of this study was to evaluate the angiographic architecture of bilateral TCCF and report our experience with their endovascular management.

Method. Over 15 years, 252 consecutive patients with TCCF were referred to our institute for endovascular treatment. Bilateral TCCF occurred in 5 men and 2 women with a mean age of 31 years. The angiographic architectures of bilateral TCCF were evaluated with cerebral angiography. All patients underwent a single session of transarterial embolisation by using various permanent embolic materials and were followed up clinically or with angiography for a mean of 22 months (range 9–36 months).

Findings. All patients presented with neuro-ophthalmic symptoms and signs. No new instances of cerebrovascular ischemia or intracranial haematoma resulted from bilateral TCCF. All fistulae were associated with partial arterial steal and were successfully occluded by using a detachable balloon and/or a detachable coil with or without a liquid adhesive. Of 14 TCCF, 9 were completely obliterated with preserved flow of the internal carotid artery (ICA). In the other 5 fistulae, the ICA had to be sacrificed to achieve occlusion because the anatomy of the fistula was complex. All fistula related symptoms resolved immediately or gradually during clinical follow up. No clinically significant procedure related neurological complications or recurrent fistulae were observed.

Conclusions. All bilateral TCCF were associated with a partial arterial steal phenomenon. Single session endovascular treatment using various embolic materials was effective in managing these high-flow fistulae. In all patients, it was possible to preserve one or both ICAs.

Keywords: Endovascular treatment; balloon or coil embolisation; haemodynamics; bilateral traumatic carotid-cavernous fistulae.

Introduction

A traumatic carotid-cavernous fistula (TCCF) is a high-flow fistula caused by a tear in the cavernous portion of the internal carotid artery (ICA) after head injury. The tear creates an abnormal direct communication between the high-pressure carotid arterial system and the low-pressure cavernous-sinus venous system. Such tears are usually single and unilateral with a 2–5 mm fistular track [12]. On rare occasions, a second, separate tear occurs in the same ICA [18].

As a consequence of their high-flow shunting characteristics, most TCCF result in reverse flow to the superior ophthalmic vein and superficial middle cerebral vein, with rapid shunting to the inferior petrosal sinus and the pterygoid vein. This flow may cause neuro-ophthalmic symptoms, cranial nerve palsy, or occasionally, intracerebral haemorrhage [8]. TCCF have variable effects on

cerebrovascular haemodynamics because of arterial steal and shunting of flow to the venous system. In theory, bilateral TCCF would be expected to cause a greater effect on cerebrovascular haemodynamics than unilateral lesions. In addition, endovascular management is more challenging with bilateral TCCF because the risk of sacrificing the ICAs on both sides is likely to cause cerebral hypoperfusion.

The purpose of this study was to evaluate the angiographic architecture of bilateral TCCF and to report our experience with their endovascular management using various embolic materials.

Materials and methods

We identified 252 consecutive patients with TCCF referred to our institute for endovascular treatment between January 1990 and December 2005. Seven patients (2.8%) had bilateral TCCF after being involved in motor vehicle accidents. There were 5 men and 2 women with a mean age of 31 years (range 19–44 years). These patients were referred for endovascular treatment because of TCCF-related neuro-ophthalmic symptoms and signs, such as chemosis ($n=7$), bruit ($n=6$), blurred vision ($n=5$), proptosis ($n=3$), and/or ptosis ($n=1$).

Cranio-facial computed tomographic scans were obtained in all patients to exclude cerebral and facial injuries which may have occurred at the time of head trauma. Such injuries (Table 1), included intracerebral haematoma ($n=6$), subdural haematoma ($n=5$), epidural haematoma ($n=2$), subarachnoid haemorrhage ($n=1$), and craniofacial fracture ($n=3$). Six patients underwent surgical removal of the intracranial haematomas before being referred for embolisation, because of haematomas causing mass effect on intracranial structures and all these patients suffered various permanent neurological sequelae. One female patient with a subdural haematoma received conservative treatment and recovered well without any neurological disturbance. Endovascular treatment was performed 3–15 weeks (mean 10 weeks) after the traumatic event occurred. During this interval, the patient's clinical progression such as new symptoms related to ischaemia or intracranial haematoma were carefully monitored.

Prior to embolisation, four-vessel cerebral angiography including an occlusion test were performed with either local ($n=5$) or general ($n=2$) anaesthesia to delineate the locations of the fistulae, evaluate the presence of arterial steal, pattern of venous drainage and collateral circulation, other traumatic cerebrovascular lesions and the patient's tolerance of ICA occlusion. In 5 patients, the balloon occlusion test was selected to assess the effect of sacrificing the ICA, while two patients underwent a venous phase filling test under general anesthesia for this purpose because of their poor mentation.

Following angiography, transarterial embolisation was performed under local ($n=2$) or general ($n=5$) anaesthesia depending on the patient's neurological status and tolerance of the procedure. Systemic heparinisation was achieved and the activated clotting times were maintained at about twice the baseline level. A 6F–8F guiding catheter was positioned in the ICA. In all patients, use of a latex detachable balloon (Goldvalve; Nycomed Ingenor, Paris, France) was selected as first-line treatment. The balloon was coaxially advanced into

Table 1. *Demographics and clinical outcomes* in 7 patients with bilateral TCCF*

Patient no./ sex/age (years)	Clinical manifestations	Craniofacial injury	Embolic materials	ICA preservation	Follow-up (months)
1/F/21	Bruit, chemosis, proptosis	ICH, SDH, facial fractures	R and L balloon	Bilateral	32
2/M/23	Blurred vision, chemosis, proptosis	ICH, EDH, skull-base and facial fractures, MAVF	R coil and NBCA; L balloon and coil	L	25
3/M/19	Bruit, chemosis	ICH, EDH, MAVF	R balloon and coil; L balloon, coil, and NBCA	R	13
4/M/46	Blurred vision, bruit, chemosis	ICH, SDH	R and L balloon and coil	R	12
5/M/28	Blurred vision, bruit, chemosis, ptosis	ICH, SDH, SAH	R balloon and coil; L balloon	L	36
6/M/44	Blurred vision, bruit, chemosis	ICH, SDH, frontal fracture	R balloon; L balloon and coil	R	24
7/F/39	Blurred vision, bruit, chemosis, proptosis	SDH	R and L coil	Bilateral	9

EDH Epidural haematoma; *F* female; *ICH* intracerebral haematoma; *L* left; *M* male; *MAVF* meningeal arterio-venous fistula; *R* right; *SAH* subarachnoid haemorrhage; *SDH* subdural haematoma.

* All patients had bilateral, partial arterial steal due to bilateral TCCF. Fistular occlusion was achieved in all patients.

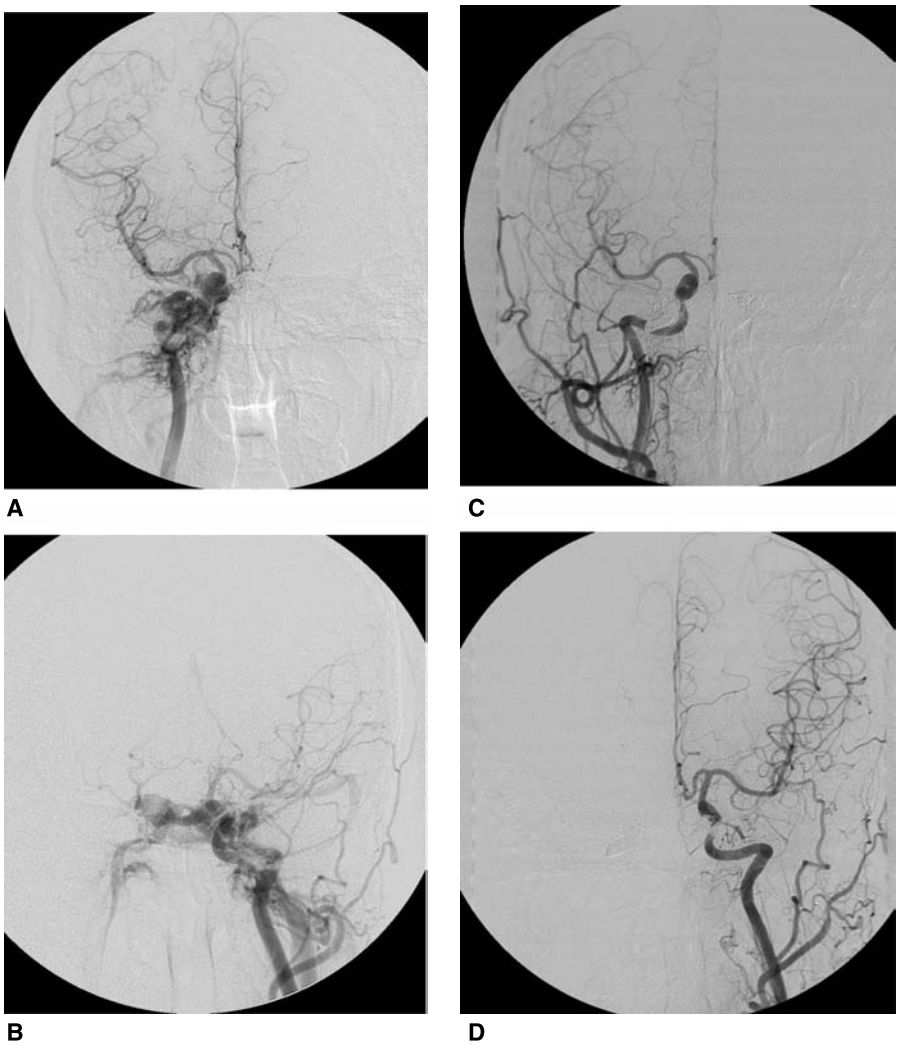

Fig. 1. (A, B) A 39-year-old woman with bilateral TCCF (patient number 7). Bilateral carotid angiograms revealing the TCCF associated with partial arterial steal. (C, D) The TCCF are totally occluded with detachable coils with preservation of flow in the ICAs. Detachable balloons failed to negotiate the cavernous sinuses because of a small fistular tract on the right side and because of repeated puncture of balloon on the left side

the cavernous sinus to occlude the fistulae after it was inflated.

The initial results demonstrated that 4 TCCF were successfully occluded with the detachable balloon, with preserved flow in the ICA. In 7 fistulae, the balloon failed to occlude the defect because the tear was large or because of transection of the ICA. In 3, the balloons could not be navigated into the cavernous sinus owing to a small fistular track ($n = 1$) or because of repeated puncture of the detachable balloon by spicules from a skull-base fracture ($n = 2$).

To further treat these 10 fistulae, platinum ($n = 2$) or detachable ($n = 8$) coils (Target Therapeutics, Freemont, CA, USA) were deployed into the cavernous sinus through the microcatheter. Complete closure was achieved in 5 TCCF (Fig. 1), but large, residual fistular flow remained in the other 5, and preservation of the ICA seemed unlikely because of complicated fistular anatomy (e.g., a large tear or transection of the ICA). These 5 fistulae were eventually occluded by using detachable balloons, coils, and/or an infusion of a 60% mixture of *N*-butyl-2-cyanoacrylate (NBCA, Melsungen AG, Melsungen, Germany) and iodised oil (Lipiodol) into the ICA and fistulae (Fig. 2).

An angiogram was obtained immediately after embolisation to ascertain occlusion of the fistulae and the patency of the ICA. All patients underwent clinical ophthalmological and neurological follow-up for a mean of 22 months (range 9–36 months). Three patients underwent follow-up angiography ($n = 2$) or magnetic resonance angiography ($n = 1$) at 4 days, 1 week, and 6 months after embolisation.

Results

Partial arterial steal and opacification of the intracranial branches of the ICA were depicted in all 14 fistulae (Figs. 1 and 2). None of the fistulae were associated with

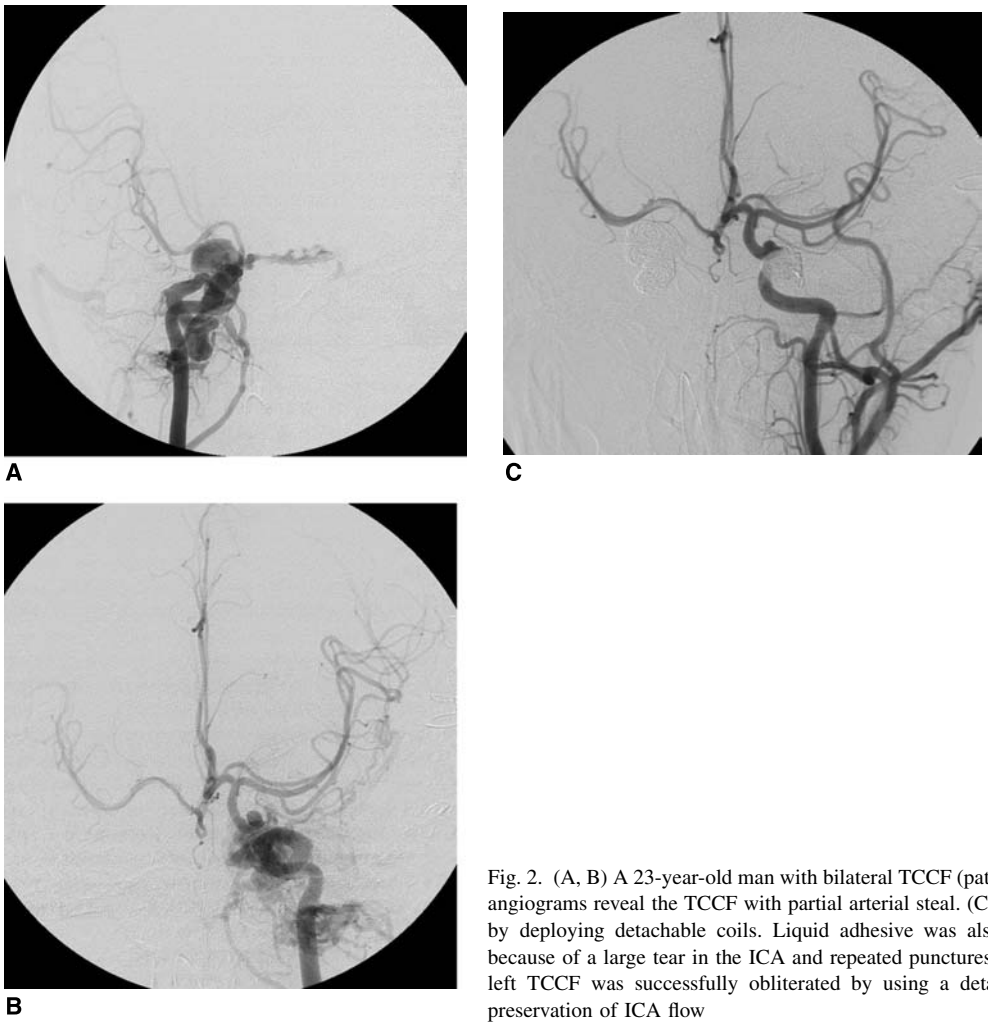

Fig. 2. (A, B) A 23-year-old man with bilateral TCCF (patient number 2). Bilateral carotid angiograms reveal the TCCF with partial arterial steal. (C) The right TCCF was occluded by deploying detachable coils. Liquid adhesive was also used in the fistula and ICA because of a large tear in the ICA and repeated punctures of the detachable balloon. The left TCCF was successfully obliterated by using a detachable balloon and coil, with preservation of ICA flow

complete arterial steal of the ICA blood flow. Associated middle meningeal arterio-venous fistulae were found in 2 patients. We found no evidence of new-onset ischaemia or intracranial haematoma related to the angiographic architecture of the bilateral TCCF owing to arterial steal or venous hypertension between the time of trauma and the start of endovascular treatment. Prior to embolisation, all patients tolerated the ICA occlusion test for each fistula. Table 1 summarises the results and follow-up data for the series.

Post-embolisation angiography demonstrated complete closure of 9 fistulae with preservation of the ICA (Fig. 1). In 5 fistulae, the ICA had to be occluded to achieve angiographic cure because of complicated fistular anatomy, such as a large tear (Fig. 2A and C) or transection of the ICA.

One patient had an asymptomatic pseudo-aneurysm in the cavernous ICA. Two patients had temporary ptosis, which completely resolved within 6 months. No clinically significant complications or recurrences were observed in any patient.

All fistula-related clinical manifestations resolved gradually and completely. The patients were discharged home or transferred to another hospital in stable neurological condition 3–10 days after embolisation. Delayed angiography was not performed in most patients because resolution of their clinical symptoms was evident and clinical follow-up showed no recurrence of their symptoms or signs.

Discussion

Bilateral carotid-cavernous fistulae are uncommon and occur in approximately 1–2% of patients with this condition [3, 9, 17]. Most bilateral TCCF are associated with severe head trauma [1, 6, 10] and/or systemic injury [4]. These patients commonly succumb to the trauma before they are referred for endovascular treatment. Symptoms and signs of bilateral TCCF are the same as those of unilateral lesions but usually mild by comparison [10]. Bilateral TCCF do not necessarily produce simultaneous, bilateral clinical manifestations because there are differ-

ences in the size and haemodynamics of the fistulae [19]. Bilateral chemosis and proptosis do not necessarily indicate bilateral TCCF because an inter-cavernous channel may make it possible for a unilateral fistula to cause bilateral or only contralateral symptoms.

In a typical untreated, unilateral TCCF, related intracranial complications (e.g., ischaemia, intracranial haematoma) are uncommon despite notable shunting of the ICA flow to the venous system. The probable reason is that the cerebral collateral circulation is competent and that most of the shunted flow is diverted to the superior ophthalmic vein and/or inferior petrosal sinus and drains into the dural sinuses. In theory, bilateral TCCF should worsen arterial steal, venous hypertension, changes in cerebrovascular haemodynamics and the stress on the vertebro-basilar system compared with a unilateral TCCF. However, we did not find any fistula-related intracranial complications in our patients with bilateral TCCF. This was presumed to reflect the fact that the simultaneous occurrence of complete arterial steal in both fistulae and of poor collateral channels from the contralateral anterior or ipsilateral posterior circulation was a rarity. In addition, both fistulae may have shared the same venous drainage and may have connected both cavernous sinuses through the coronal vein and thereby, the shunted flows may have been directed against each other, diminishing the arterial steal and shunted flow and ameliorating the clinical manifestations. This phenomenon was verified in that all fistulae were associated with partial arterial steal though the fistular tracts were large (Fig. 2A). Previous single case reports have also noted this finding [1, 4, 6, 11, 16, 19].

Spontaneous thrombosis of high-flow bilateral TCCF is extremely rare. Alkhani *et al.* [1] reported a patient in whom bilateral TCCF spontaneously resolved and transsellar inter-carotid vascular communications developed. Total resolution of bilateral TCCF after one lesion is treated with balloon occlusion of the ICA is also reported [10]. However, most untreated TCCF are associated with decreased visual acuity, ocular necrosis, and cranial nerve palsy. Moreover, life-threatening intracranial haemorrhage or epistaxis occurs in 3–9.3% of patients [6, 8]. Therefore, aggressive endovascular management of TCCF is generally recommended.

Bilateral TCCF may fill the contralateral cavernous sinuses by means of the coronal vein and achieve a cerebrovascular haemodynamic balance. Treatment of one fistula with blockage of the shunted flow may substantially change the haemodynamics of the contralateral TCCF. For example, arterial insufficiency or venous hypertension ipsilateral to the larger of 2 TCCF might increase if the contralateral, small TCCF is embolised first because of a sudden loss of venous flow and pressure in the opposing inter-cavernous sinus. In addition, compromised flow in the ICA due to therapeutic occlusion may sufficiently increase intravascular pressure on the contralateral side to expand the shunt, particularly in patients treated in different sessions. Therefore, treatment of bilateral fistulae in different sessions may aggravate or potentiate symptoms of the untreated lesion. Gaston *et al.* [6] described a patient who died from spontaneous cerebral haemorrhage 48 h after a transarterial balloon was used to occlude a contralateral fistula with preservation of the ICA. Another report described fatal intracranial haemorrhage after surgical occlusion of a contralateral ICA and fistula [4]. We prefer to treat both fistulae in the same session if the first fistula was uneventfully occluded usually by embolizing the relatively severe and large fistula and treating the associated arterial steal first. However, if the first fistular anatomy is complex or difficult to approach and the procedure is long and associated with potential risk, treatment of the second fistula at another session is more proper.

Another important issue is the potential risk of hypoperfusion with endovascular occlusion of bilateral TCCF and sacrifice of the ICA. Recent refinements in the techniques for detachable balloons, detachable coils, and liquid adhesive alone or in combination have increased the rate at which ICA flow is preserved [15]. In some instances, the ICA must be sacrificed to obtain an angiographic cure. However, occlusion of both ICAs to treat bilateral TCCF may greatly stress the vertebrobasilar system and pose a high risk of cerebral hypoperfusion, as reflected in the mortality rate of 19% [2]. In our series, both ICAs were preserved in 2 patients, and 1 ICA was preserved in 5. Therefore, we believe that preservation of blood flow in one or both ICAs is always feasible with endovascular treatment because we rarely encountered complicated fistular anatomy in both ICAs that required their simultaneous occlusion. However, if occlusion of bilateral ICAs is the only way to treat the fistulae before endovascular treatment, extracranial-to-intracranial bypass surgery should be considered.

The best endovascular approach for TCCF is still the use of a detachable balloon by means of the arterial route. Single instances of successful balloon occlusion of bilateral TCCF with ICA preservation have been reported [11, 16]. However, in patients with complicated anatomy (e.g., tortuous ICA, small fistular tract, small venous compartment or intimal flap), navigation of the balloon

into the cavernous sinus to seal the fistula is difficult. In our series, detachable balloons failed to occlude the fistulae of 10 TCCF because bony fragments repeatedly punctured the balloon ($n = 2$), because the fistular track was small ($n = 1$), or because fistulae persisted after embolisation with detachable balloons ($n = 6$). In these fistulae, a microcoil was selected as an alternative. Coils are most effective for treating low-flow carotid-cavernous fistulae 2–3 mm in diameter [5, 7, 13].

NBCA is a liquid adhesive and permanent embolising agent widely used to treat intracranial arteriovenous malformations. The advantage of NBCA is its good penetration, easy delivery through a microcatheter, and rapid polymerisation and thrombolisation after infusion. Results were promising when NBCA was used as an embolic agent to supplement transarterial embolisation of TCCF [14]. We used NBCA to enhance fistular thrombosis only after balloons and/or coils failed to totally occlude a fistula. In high-flow fistulae, highly concentrated or even pure NBCA may be necessary to achieve rapid polymerisation and to prevent migration of the polymers to supraclinoid ICA or ophthalmic artery.

In conclusion, bilateral TCCF were uncommon, and most were associated with severe head injuries, intracranial haematomas, and partial arterial steal. Cerebrovascular ischaemia or haematoma such as arterial steal or venous hypertension, was uncommon in bilateral TCCF. Single session endovascular treatment was effective in managing these high-flow fistulae. In all patients, occlusion of the fistulae with preservation of one or both ICAs was feasible by using a combination of embolic materials.

Acknowledgements

Sources of support: This work was supported in part by a grant from the Taipei Veterans General Hospital (TVGH-C1-144) and from the National Science Council (NSC95-2314-B-075-115).

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Comment

This is an outstanding series – 252 CC fistulae over 15 years. Of these 7 patients had bilateral fistula and these are the core of the article. All these fistulae were traumatic.

In 4 fistulae, a detachable balloon was used, in 5 fistulae coiling has been successful, and in 5 fistulae parent artery occlusion has been necessary. The authors enjoyed 0 morbidity/mortality rate, all symptoms disappeared.

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