Traumatic Carotid–Cavernous Sinus Fistula Associated with an Intradural Pseudoaneurysm: A Case Report

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Komiyama M, Yasui T, Yagura H, Fu Y, Nagata Y. Traumatic carotid–cavernous sinus fistula associated with an intradural pseudoaneurysm: a case report. Surg. Neurol 1991;36:126–32.

A case of traumatic carotid–cavernous sinus fistula (CCF) associated with an intradural pseudoaneurysm is reported. A 42-year-old man developed traumatic CCF after severe head trauma. Cerebral angiography demonstrated a direct CCF associated with an intradural pseudoaneurysm at the C2 portion. Transarterial balloon embolization of the CCF caused severe subarachnoid hemorrhage. A CCF with an intradural pseudoaneurysm is life-threatening and requires emergency treatment. However, balloon occlusion in such cases is contraindicated because of possible rupture of a pseudoaneurysm. Trapping or a direct surgical approach is the treatment of choice.

KEY WORDS: Balloon embolization; Pseudoaneurysm; Subarachnoid hemorrhage; Traumatic carotid–cavernous fistula

Spontaneous resolution of traumatic carotid–cavernous sinus fistulas (CCFs) is very rare, so active treatment is required, with transvascular embolization being the treatment of choice [3,5,6,10,18]. The goal of therapy is to occlude the fistula while preserving the carotid flow.

Intracranial hemorrhage associated with a CCF is relatively rare and, in most cases, has been due to spontaneous intracerebral hemorrhage [1,4,9,19,20]. Recently, we encountered a case of traumatic CCF associated with an intradural pseudoaneurysm. Balloon occlusion of the fistula preserving the carotid flow caused severe subarachnoid hemorrhage. In this report, we discuss the therapeutic indications for such cases.

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Received October 29, 1990; accepted January 15, 1991.

Case Report

A 42-year-old man was transferred to our clinic immediately after receiving a severe head injury in an automobile accident.

On admission, he was stuporous, with a score of 7 on the Glasgow Coma Scale. Moderate left hemiparesis was noted. He had a scalp laceration in the right frontal region through which a small amount of brain tissue was protruding. A skull film showed an extensive bilateral frontobasal fracture. Computed tomography (CT) scan demonstrated acute hydrocephalus and a moderate subarachnoid hemorrhage (Figure 1).

Emergency Operation for the Open Fracture

An emergency operation for the open frontobasal fracture was carried out. There was a large, dilated red vein on the right cortical surface of the frontal lobe but no indications as to whether it was pathological. Although the dura mater seemed to be almost completely closed, the frontal skull base was not extensively explored. A ventricular drainage tube was inserted into the left frontal horn for acute hydrocephalus. Postoperatively, the patient's neurological condition was unchanged.

Postoperative Course and Cerebral Angiography

In the following week, the patient's neurological condition gradually improved. However, his right eye became edematous and injection was noted. Bruits synchronous to his heart beat were audible over his right eye. Cerebral angiography was carried out on the sixth posttraumatic day, which revealed a direct CCF at the right cavernous sinus and a small right frontal arteriovenous malformation (AVM) (Figure 2). The exact location of the fistula was not visible on the angiogram, but the image strongly suggested that it was at the C2 portion of the internal carotid artery. The CCF drained into the right superior ophthalmic vein and bilateral inferior petrosal veins, and there was no associated cortical drainage. The main feed-

0090-3019/91/$3.50

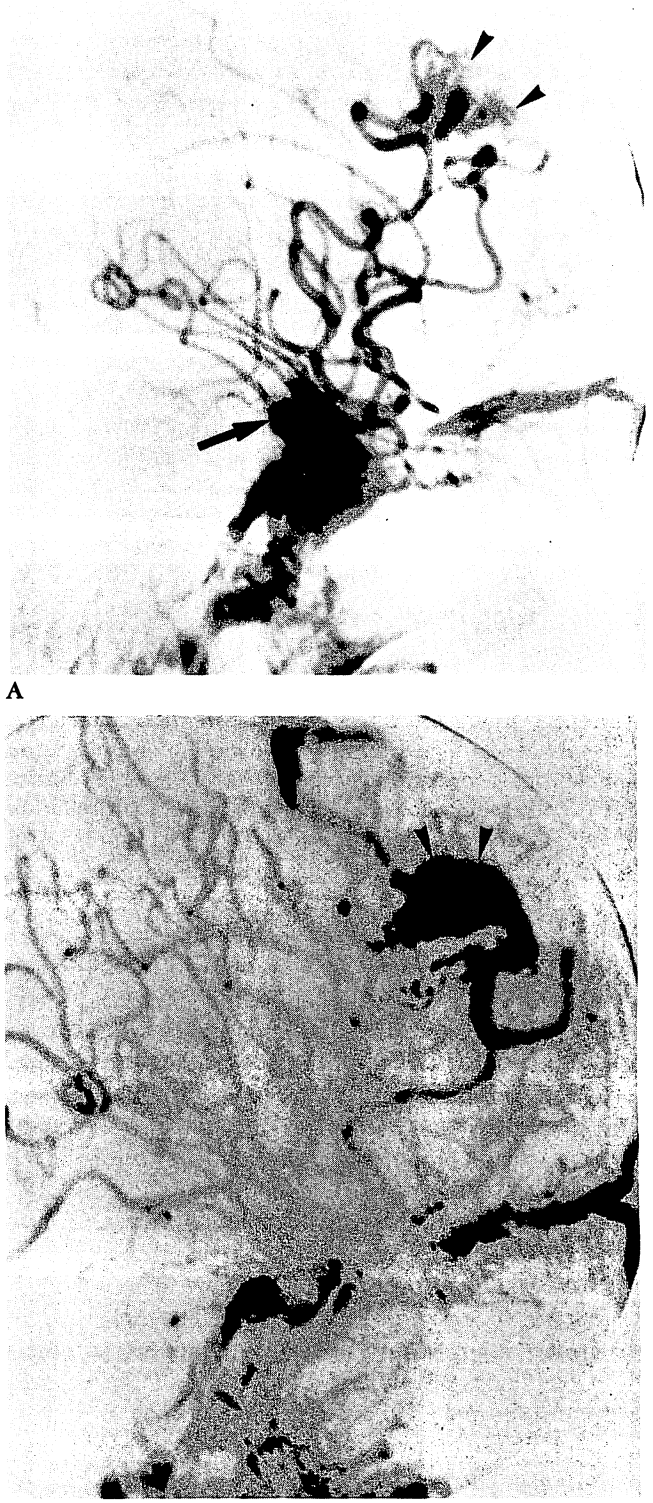
Figure 1. *A CT scan immediately after severe head trauma shows a remarkable subarachnoid hemorrhage and depressed fracture of the frontal bone. A small air bubble can also be seen (arrow).*

ing arteries of the frontal AVM were two branches from the right middle cerebral artery, and drainage was through three cortical veins–one connected to the cavernous sinus and the remaining two to the superior sagittal sinus, one of which was recognized during the emergency operation.

Embolization of the Traumatic Carotid–Cavernous Sinus Fistula

The absence of a localized subarachnoid hemorrhage or hematoma suggested that the AVM was still silent and that the accident probably was responsible for the subarachnoid hemorrhage. Thus, therapy was first directed solely to the traumatic CCF.

On the 10th posttraumatic day, transarterial embolization of the CCF was attempted. Through a transfemoral route with the patient under local anesthesia, a guiding catheter (8 Fr) was introduced into the right internal carotid artery. A #17 latex Debrun Goldvalve balloon (Ingenor Medical System, Paris, France) was mounted to a tracker-18 catheter with an extended tip (Target



A

B

Figure 2. *Right carotid injection (***A,** *early arterial phase;* **B,** *late arterial phase) demonstrates a direct CCF associated with an intradural pseudoaneurysm (arrow). Drainage is to the superior ophthalmic vein and to the inferior petrosal sinus. A right frontal arteriovenous malformation (arrowheads) is also visible. The feeding arteries are two branches of the middle cerebral artery. The draining routes are to the superior sagittal sinus and the cavernous sinus through the cortical veins.*

Therapeutics, San Jose, Calif.), and a microguidewire was placed in the catheter using a method similar to that described by Nelson [14]. The actual location of the fistula was at the C2 portion of the carotid artery. The balloon attached to the tracker catheter was easily navigated into the fistula site (Figure 3 *A* and *B*). Although we did not realize at this moment that the microcatheter was navigated to the fistula through an intradural pseudoaneurysm, it was thought dangerous to overdistend the wall of the fistula with balloons. Using the roadmap technique, balloons were inflated with a minimum volume of diluted contrast medium to obtain almost total occlusion, intentionally leaving a small fistulous shunt. The balloons were detached by gentle retraction of the tracker catheter. The fistula was so large that two balloons were required to occlude it. Postembolization angiography with right carotid injection showed a minimal fistulous connection between the internal carotid artery and cavernous sinus (Figure 3 *C* and *D*). No conversion of the drainage pathways was noted. The patient's neurological state was stable during these procedures, and bruits over the right eye were no longer audible.

Proximal Balloon Occlusion of the Internal Carotid Artery

About 1.5 hours later, the patient suddenly became comatose and the ventricular tube drained fresh blood. A CT scan showed a dense subarachnoid hemorrhage and ventricular hemorrhage (Figure 4). The patient was immediately brought back to the "angio" suite. Right carotid angiography demonstrated complete occlusion of the CCF with preservation of the carotid flow, without any demonstrable pseudoaneurysm (Figure 5). Although angiography did not disclose any abnormality except for the AVM, it was thought that a pseudoaneurysm at the C2 portion had ruptured. Even with the poor collateral circulation, we thought it necessary to sacrifice the right carotid flow to avoid rebleeding. It was thought best to trap the fistula using two balloons, but to avoid compromising the flow of the right anterior choroidal artery, proximal balloon occlusion using two #19 latex Debrun Goldvalve balloons at the C4 and C5 portions was carried out (Figure 6).

On the subsequent day, CT scan demonstrated a large infarction in the right hemisphere, and the patient remained comatose. Angiography carried out 1.5 months later showed an aneurysmal pouch at the C2 portion but no CCF was observed (Figure 7). The AVM showed no change, but the right ophthalmic artery contributed as an anastomosis from the external carotid system to the internal carotid system.

Dural Plasty for Cerebrospinal Fluid Leakage and Total Removal of the AVM

About 2 months later, the patient developed rhinorrhea and pneumocephalus. Dural plasty of the right frontal base and total removal of the AVM were carried out. Intraoperatively, dural and bony defects were noted at the right frontal base, which was covered with a fascia lata graft. The AVM seemed not to have bled before. The distal portion of the carotid artery was not explored. At the 5-month follow-up, the patient could respond to simple commands but was bedridden with left hemiplegia.

Discussion

An endovascular approach is now accepted as the treatment of choice for traumatic CCFs [3,5,6,10,18]. The ultimate goal of this treatment regimen is to occlude the fistula completely while maintaining a patent carotid flow.

Hemorrhagic complications associated with CCFs, excluding epistaxis and otorrhagia, have been rare. Most reported cases were spontaneous intracerebral hematomas due to increased pressure of the cortical veins draining the CCFs [1,4,9,19,20], and there are few reports of subarachnoid hemorrhage associated with CCFs [7,9,13,17].

In the large series of traumatic CCFs reported by Debrun et al [5], only one among the 54 cases had a fistula located at "the anterior ascending intercavernous segment" of the internal carotid artery, but there were no cases of intradural location. Since the balloon catheter reached the CCF through the C2 portion of the internal carotid artery and late angiography showed the aneurysmal pouch at this location, the CCF was associated with an intradural pseudoaneurysm. Similar true aneurysms are found at this location, such as the congenital berry aneurysm, with a partially intradural origin and mainly intracavernous fundus [15].

From our experience, it should be stressed that the exact location (including whether the location of the fistula is intradural or extradural) and size of the fistula should be clarified before embolization. Vertebral injection with manual carotid compression [11] and internal carotid injection using a double balloon catheter [2] have both been reported to be useful methods. It should also be clarified whether there is a pseudoaneurysm or a cavernous sinus varix (a dilated cavernous sinus extending into the subarachnoid space), both of which are life-threatening and require emergency treatment [9]. Angiographical differentiation between a pseudoaneurysm and a cavernous sinus varix, however, may be difficult or impossible [9]. Overinflation of a balloon in a pseudoaneurysm or cavernous sinus varix can easily

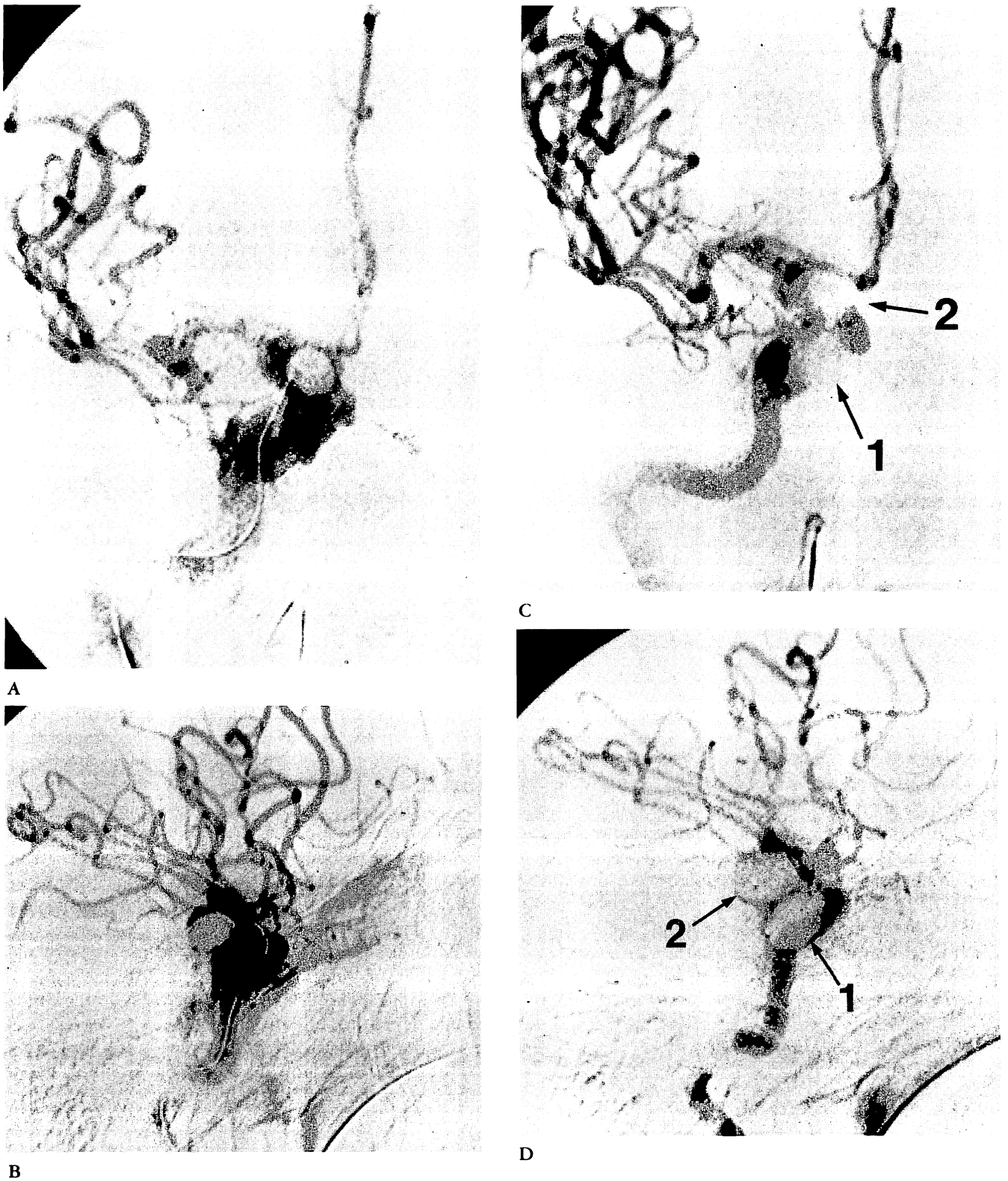


Figure 3. *Right carotid injection during balloon embolization (***A,** *anteroposterior view;* **B,** *lateral view). A latex detachable balloon is attached to the tip of a tracker-18 catheter. The first balloon is navigated into the fistula through a pseudoaneurysm at the C2 portion of the carotid artery. Right carotid injection immediately after two-balloon embolization (***C,** *anteroposterior view;* **D,** *lateral view). The first balloon (1) is located caudal to the second balloon (2). A faint fistulous shunt can be observed, but neither conversion of the drainage routes nor cortical drainage is seen.*

Figure 4. *A CT scan 1.5 hours after balloon embolization of the CCF. Severe subarachnoid hemorrhage is demonstrated. One contrast-filled balloon is also visible (arrow).*

Figure 5. *Right carotid injection carried out immediately after the severe subarachnoid hemorrhage shows complete disappearance of the traumatic CCF with preservation of the carotid flow.*

A

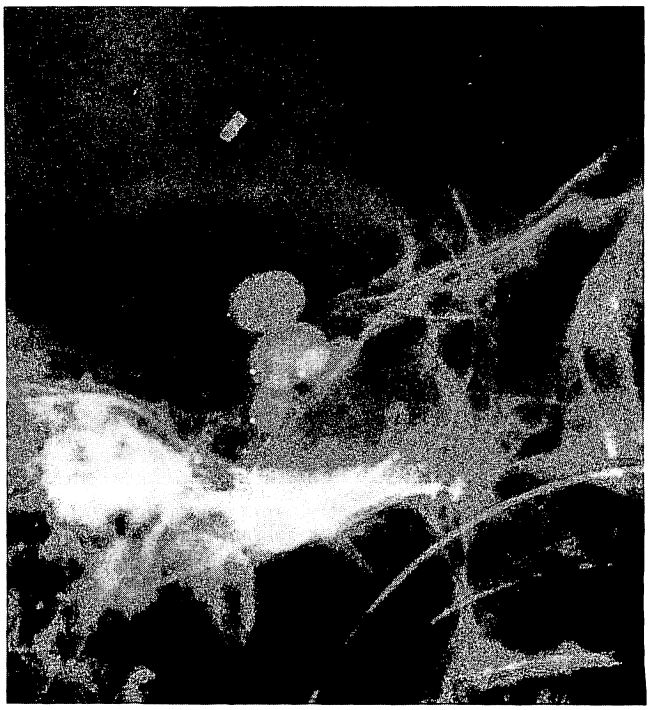


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Figure 6. *Plain skull x-ray films (***A,** *anteroposterior view;* **B,** *lateral view) after proximal balloon occlusion of the internal carotid artery show the two large balloons used for occlusion of the CCF and the two small balloons at the C4 and C5 portions of the carotid artery used for proximal occlusion of the carotid artery. The tip of the drainage tube to the left frontal horn is also seen.*

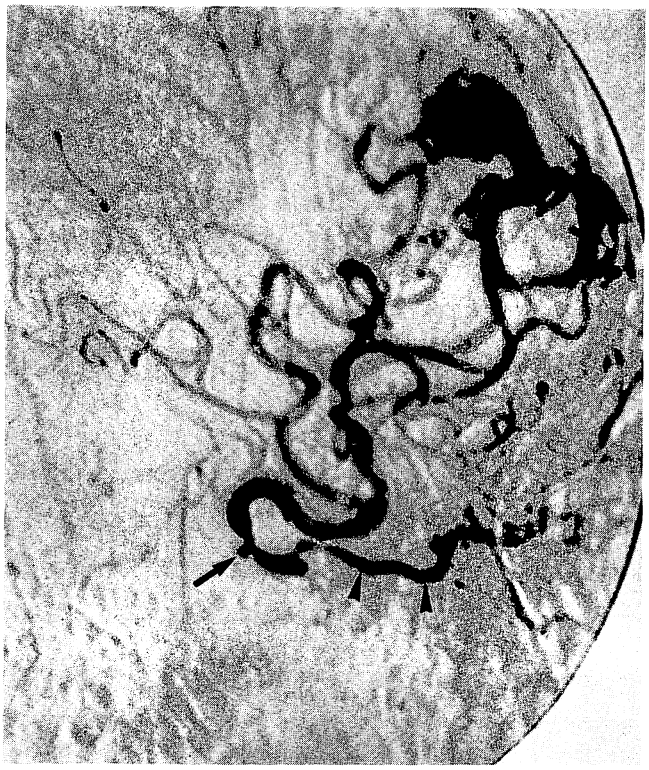

Figure 7. *Right common carotid injection carried out 1.5 months later demonstrates occlusion of the internal carotid artery and collateral circulation through the ophthalmic artery (arrowheads) from the external carotid system to the internal carotid artery. An aneurysmal pouch at the posterior aspect of the C2 carotid artery can be seen (arrow).*

cause a rupture, theoretically resulting in fatal subarachnoid hemorrhage. Although there was a possibility that transvenous balloon embolization would have been better for a pseudoaneurysm than transarterial embolization because of reduced risk of tearing the carotid arterial wall, either therapeutic modality could cause a fatal subarachnoid hemorrhage. Trapping should be considered the treatment of choice when the patient can tolerate carotid occlusion. Although we did not carry out test occlusion of the carotid artery using a balloon catheter, this technique itself is rather easy and should be performed. When the patient cannot tolerate occlusion, direct surgery or an extracranial–intracranial bypass followed by trapping is the treatment of choice.

Direct surgical obliteration of a direct CCF, which was pioneered by Parkinson [16], is now possible because of recent advances in microsurgery and improved understanding of cavernous sinus anatomy [8,12,13]. However, it is still difficult to occlude a pseudoaneurysm without compromising the carotid flow. Van Dellen [21] reported that when a pseudoaneurysm of the carotid artery is in the cavernous sinus, it is not amenable to direct clipping and is better treated by trapping. Debrun et al [6] concluded that surgery for a traumatic CCF should be limited to those cases in which the internal carotid artery cannot be preserved or the internal carotid artery has been previously ligated. However, if there is a chance that the CCF is associated with an intradural pseudoaneurysm or cavernous sinus varix, trapping or direct surgery should be considered as the treatment of choice rather than endovascular treatment.

In conclusion, the treatment of choice for traumatic CCFs in most cases is endovascular occlusion of the fistula. However, a CCF associated with an intradural pseudoaneurysm is life-threatening and requires emergency treatment. It should be treated by trapping or by direct surgical treatment.

References

1. Ambler MW, Moon AC, Sturner WQ. Bilateral carotid–cavernous fistulae of mixed types with unusual radiological and neuropathological findings. J Neurosurg 1978;48:117–24.
2. Berenstein A, Kricheff II. Balloon catheters for investigating carotid cavernous fistulas. Radiology 1979;132:762–4.
3. Berenstein A, Kricheff II, Ransohoff J. Carotid–cavernous fistulas: intraarterial treatment. AJNR 1980;1:449–57.
4. d'Angelo VA, Monte V, Scialfa G, Fiumara E, Scotti G. Intracerebral venous hemorrhage in "high-risk" carotid–cavernous fistula. Surg Neurol 1988;30:387–90.
5. Debrun G, Lacour P, Vinuela F, Fox A, Drake CG, Caron JP. Treatment of 54 traumatic carotid–cavernous fistulas. J Neurosurg 1981;55:678–92.
6. Debrun GM, Nauta HJ, Miller NR, Drake CG, Heros RC, Ahn HS. Combining the detachable balloon technique and surgery in imaging carotid cavernous fistulae. Surg Neurol 1989;32:3–10.
7. Dohrmann PJ, Batjer HH, Samson D, Suss RA. Recurrent subarachnoid hemorrhage complicating a traumatic carotid–cavernous fistula. Neurosurgery 1985;17:480–3.
8. Dolenc V. Direct microsurgical repair of intracavernous vascular lesions. J Neurosurg 1983;58:824–31.
9. Halbach VV, Hieshima GB, Higashida RT, Reicher M. Carotid cavernous fistulae: indications for urgent treatment. AJNR 1987;8:627–33.
10. Halbach VV, Higashida RT, Hieshima GB, Hardin CW, Yang PJ. Transvenous embolization of direct carotid cavernous fistulas. AJNR 1988;9:741–7.
11. Huber P. A technical contribution to the exact angiographic localization of carotid cavernous fistulas. Neuroradiology 1976;10:239–41.
12. Isamat F, Ferrer E, Twose J. Direct intracavernous obliteration of high-flow carotid–cavernous fistulas. J Neurosurg 1986; 65:770–5.
13. Mullan S. Treatment of carotid–cavernous fistulas by cavernous sinus occlusion. J Neurosurg 1979;50:131–44.
14. Nelson M. A versatile, steerable, flow-guided catheter for delivery of detachable balloons. AJNR 1990;11:657–8.
15. Nutik S. Carotid paraclinoid aneurysms with intradural origin and intracavernous location. J Neurosurg 1978;48:526–33.
16. Parkinson D. Carotid cavernous fistula: direct repair with preservation of the carotid artery. Technical note. J Neurosurg 1973;38:99–106.

17. Sedzimir CB, Occleshaw JV. Treatment of carotid–cavernous fistula by muscle embolization and Jaeger's maneuver. J Neurosurg 1967;27:309–14.

18. Serbinenko FA. Balloon catheterization and occlusion of major cerebral vessels. J Neurosurg 1974;41:125–45.

19. Tanaka A, Fukushima T, Tomonaga M. Intracerebral hematomas in cases of dural arteriovenous malformation and carotid–cavernous fistula. Surg Neurol 1986;25:557–62.

20. Turner DM, Vangilder JC, Mojtahedi S, Pierson EW. Spontaneous intracerebral hematoma in carotid–cavernous fistula. Report of three cases. J Neurosurg 1983;59:680–6.

21. Van Dellen JR. Intracavernous traumatic aneurysms. Surg Neurol 1980;13:203–7.