The Endovascular Treatment of Traumatic Cavernous Sinus Arteriovenous Fistulas: A Single-center Experience

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Objective: The treatment for traumatic carotid cavernous fistula (TCCF) that occurred after head trauma is evaluated based on our experience.

Methods: The clinical characteristics, treatments, and outcomes were retrospectively evaluated in six patients with TCCF that we treated between April 2012 and July 2015.

Results: The mean age of the patients, consisting of five males and one female, was 45.5 ± 16.8 years. The head trauma was acute subdural hematoma (ASDH) in one patient, acute epidural hematoma (AEDH) in one, brain contusion in two patients, and skull fracture in three patients. The patients exhibited bulbar conjunctival congestion, exophthalmos, and disturbance of ocular movement mean of 2.2 ± 1.8 months (2 days and 5 months) after injury, and the one patient who suffered rupture of pseudoaneurysm showed arterial nasal bleeding. The presence of fistula in the cavernous portion of the internal carotid artery was confirmed by cerebral angiography, and the mean maximum diameter of the fistula, measured by 3D DSA, was 9.6 ± 3.2 mm (2.4–19.9 mm). Endovascular procedures were completed in all patients, but among those with a large fistula, parent artery occlusion (PAO) was selected for three patients with ischemic tolerance, and transvenous embolization (TVE) of the cavernous sinus was selected for two patients with no tolerance. In a patient with a small and simple fistula, percutaneous transluminal angioplasty (PTA) was performed with covered stent placement. The shunt disappeared, and neurological symptoms were resolved within 1 month, in all patients. The postoperative course during a mean follow-up period of 21.0 ± 13.5 months was uneventful without recurrence.

Conclusion: Satisfactory outcomes could be achieved by endovascular treatment for TCCF by selecting an appropriate method for each patient.

Keywords ► traumatic carotid cavernous fistula, parent artery occlusion, transvenous embolization

Introduction

Traumatic carotid cavernous fistula (TCCF), a fistula formed between the internal carotid artery and cavernous sinus due to

trauma, accounts for about 4% of traumatic cerebral vascular injuries.¹⁾ There are also reports that intracranial endovascular treatment is highly effective for TCCF, with the fistula closure rate reaching 88%,²⁾ and consistently effective treatment is required. In this study, we evaluated the clinical characteristics, treatments, and outcomes in six patients who underwent endovascular surgery for TCCF at our department.

Subjects and Methods

We treated six patients with TCCF between April 2012 and September 2015. This series of patients was retrospectively evaluated regarding the following factors: sex, age, outcome of injury, time from injury to the onset, symptoms, intracranial angiographic findings, particularly, the fistula diameter, treatments, adverse events associated with the procedure, and outcome during the follow-up period.

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Results

The information concerning the six patients is summarized in **Table 1**. The male/female ratio of the patients was 5:1, and their mean age at treatment was 45.5 ± 16.8 years (22–72 years). The head injuries, which were caused by a traffic accident in five patients and a fall in one patient, were acute subdural hematoma (ASDH) in one patient, acute epidural hematoma (AEDH) in one patient, brain contusion in two patients, and skull fracture in three patients. Symptoms appeared a mean of 2.0 ± 1.8 months (2 days and 5 months) after head injury, and they were bulbar conjunctival congestion/exophthalmos in six patients and pulsatile nasal bleeding in one patient. Cerebral angiography showed a fistula in the cavernous portion of the internal carotid artery that drained into the cavernous sinus. The maximum diameter of the fistula was measured using a workstation (Syngo; Siemens, Munich, Germany) after 3D rotation DSA from the internal carotid artery. When the shunt flow is high, and the structure of the fistula was unclear, the retrograde blood flow from the vertebral artery or contralateral internal carotid artery was analyzed by visualizing it with manual compression of the affected internal carotid artery, and the mean maximum diameter of the fistulas was determined as 9.6 ± 3.3 mm (2.4–19.9 mm). As the therapeutic strategy for patients with a large fistula diameter, parent artery occlusion (PAO) was selected when there was ischemic tolerance on the balloon occlusion test (BOT) with occlusion of the internal carotid artery on the proximal side of the fistula, and transvenous embolization (TVE) was selected when there was no ischemic tolerance. Endovascular treatment was performed under general anesthesia in all patients. One of the three patients who underwent PAO had received TVE a total of two times, which had resulted in a reduction but persistence of the blood flow through the fistula. Re-evaluation of the findings on preoperative BOT showed an increase in the blood flow from the contralateral internal carotid artery via the anterior communicating artery due to occlusion of the affected internal carotid artery on the proximal side of the fistula, and a false-positive result due to the intracranial steal phenomenon was suggested. Therefore, while this patient was eventually treated by PAO, no ischemic symptom appeared. Complete cure could be achieved by TVE in two patients, and the fistula diameter was 10 mm or less in both (7.5 mm in Case 4, 5.3 mm in Case 5). The percutaneous transluminal angioplasty (PTA) was performed with covered stent placement in Case 6, who had a smallest fistula diameter of 2.4 mm and showed poor

Table 1 The characteristics, treatments, and outcome of the patients of the direct carotid-cavernous fistula caused by trauma

Case	Age (years)	Sex	Trauma	Symptom	Onset to symptom	Fistula size	IVR①	IVR②	IVR③	Complication	Outcome	F/U
1	30	M	Contusion, skull frac.	Ptosis, blindness	1	19.9	PAO			NA	GR	24
2	42	M	Contusion	Exophthalmos, chemosis	1	12.0	TVE	TVE	PAO	NA	GR	8
3	22	M	AEDH	Exophthalmos, chemosis, epistaxia	1	10.4	PAO			NA	GR	18
4	72	M	ASDH	Exophthalmos, chemosis	5	7.5	TVE			NA	MD	4
5	59	F	Orbital bone frac.	Exophthalmos, chemosis	4	5.3	TVE			NA	GR	35
6	48	M	Skull frac.	Exophthalmos, chemosis	0.1	2.4	PTAS			NA	GR	3

AEDH: acute epidural hematoma; ASDH: acute subdural hematoma; IVR: interventional radiology; PAO: parent artery occlusion; PTA: percutaneous transluminal angioplasty; TVE: transvenous embolization

Fig. 1 (A) Nasal endoscopic view; (B and C) preoperative left internal carotid angiogram (B) AP view, (C) lateral view; (D and E) preoperative right vertebral angiogram (D) AP view, (E) lateral view. The nasal endoscope showed the pulsatile hematoma under the nasal mucosa of the left superior concha (A). The left internal carotid artery angiogram showed the direct carotid cavernous fistula in the cavernous position. The arrow point is the fistula of the shunt. The right vertebral artery angiogram showed the retrograde flow from the left PcomA to the fistula under the balloon occlusion of the left internal carotid artery. The arrow point is the fistula of the shunt. AP: anterior-posterior; Lat: lateral; PcomA: posterior communicating artery

ischemic tolerance, after evaluation by the institutional review board.

Regarding the relationship between the fistula diameter and endovascular procedure, the mean fistula diameter was 14.1 ± 4.2 mm in the PAO group and 5.1 ± 2.1 mm in the non-PAO group with a significant difference ($p = 0.04$, Mann-Whitney U-test). Retrospectively, PAO was necessary in all patients with a fistula diameter of 10 mm or larger. After treatment, the shunt disappeared in all patients, and the neurological symptoms observed before surgery were resolved or alleviated within 2 months. None showed recurrence during a follow-up period of 21.0 ± 13.5 months.

Representative cases

Case 3: A 22-year-old man presented with nasal bleeding, bilateral bulbar conjunctival congestion, and left exophthalmos.

The patient fell off a bicycle, hit the left frontal region, and was transported. CT of the head showed a left frontal skull fracture and a left AEDH, and emergency craniotomy was performed on the same day. However, the patient developed massive nasal bleeding 1 month after surgery and was transported to our hospital by ambulance.

The findings on arrival were as follows: a blood pressure of 78/50 mmHg, a heart rate of 110 bpm, pallor, JCSII-10,

bilateral bulbar conjunctival congestion, and left exophthalmos. While nasal bleeding was temporarily arrested, a pulsating hematoma was noted in the mucosa of the superior nasal concha on transnasal endoscopy (**Fig. 1A**).

On left internal carotid artery angiography, a fistula was noted in the cavernous portion of the left internal carotid artery, all blood drained into the shunt, and the distal intracranial artery was not visualized (**Fig. 1B** and **1C**). A 9 Fr Optimo (Asahi Intecc, Aichi, Japan) was placed in the left internal carotid artery, and right vertebral artery angiography was performed while blocking the cervical internal carotid artery. The retrograde blood flow from the posterior communicating artery (PcomA) drained to the venous side through the fistula, and the fistula diameter was 10.4 mm (**Fig. 1D** and **1E**). Since the fistula was large, and symptoms of cerebral ischemia were not induced by blocking the affected internal carotid artery on the proximal side of the fistula, emergency internal carotid artery occlusion using platinum coils was performed under general anesthesia on the same day. First, to preserve the PcomA, we attempted to guide a Scepter XC 4×11 mm (Terumo, Tokyo, Japan) to the origin of the PcomA through a guiding catheter placed in the left internal carotid artery, but it could not be advanced anterogradely through the lesion. Therefore, it was guided retrogradely across the lesion via

Fig. 2 (A) Intraoperative right vertebral angiogram with the balloon occlusion of left internal carotid artery; (B and C) intraoperative X ray; (D and E) postoperative right vertebral angiogram (D) DA image, (E) DSA enlarged image, (F) DSA image; (G) postoperative nasal endoscopic view. The micro-balloon was difficult to pass through the origin of PcomA, so the micro-guide wire was reversally passed through from PcomA to the left internal carotid artery (A and B). The micro-balloon was easily passed through the shunting point along the micro wire (C). The coil embolization of the left internal carotid artery including the fistula was performed under the balloon protection of the left PcomA (D). The shunt was disappeared after the coil embolization (E and F). The nasal hematoma was also disappeared (G). DA: digital angiography; PcomA: posterior communicating artery

the PcomA through a 6 Fr Fubuki placed in the right vertebral artery using Chikai 14 guide wire 200 cm (ASAHI, Intecc, Aichi, Japan) and advanced to the proximal side of the left internal carotid artery (**Fig. 2A** and **2B**). Next, we succeeded in advancing the Scepter XC along the same wire (**Fig. 2C**) and placing it at the origin of the PcomA. By dilating the balloon, the left internal carotid artery including the fistula could be occluded with platinum coils while preserving the PcomA (**Fig. 2D–2F**).

After the procedure, bulbar conjunctival congestion and exophthalmos immediately disappeared, and no recurrence of nasal bleeding was observed (**Fig. 2G**). There were also no symptoms of ischemia, and the patient was discharged to home. No recurrence was noted during a 2-year follow-up period after the procedure.

Case 6: A 48-year-old man presenting with left bulbar conjunctival congestion, left exophthalmos, and pulsatile tinnitus

The patient fell after drinking alcohol and hit the face. CT of the head showed fracture of the lateral side of the left orbital bone, and the patient was admitted for observation,

but he noted pulsatile tinnitus from 2 days after admission. Left exophthalmos and left palpebral conjunctival congestion were noted, and vascular murmur was heard posteriorly to the left auricle and in the left cheek.

On left internal carotid artery angiography, a fistula was detected in the cavernous portion of the left internal carotid artery. The fistula diameter was 2.4 mm. All blood drained into the shunt, and the distal intracranial artery was not visualized (**Fig. 3A** and **3B**). Right vertebral artery angiography showed retrograde filling of the cavernous sinus through the fistula via the PcomA (**Fig. 3C** and **3D**). On BOT in the left internal carotid artery on the proximal side of the fistula, ischemic symptoms appeared in 10 minutes. Brain perfusion study also showed a slight decrease in the blood flow primarily in the left frontal and temporal lobes. Occlusion of the internal carotid artery was impossible due to poor ischemic tolerance. In addition, while the fistula was small, the shunt flow was extremely high. Therefore, it was decided to perform PTA with covered stent placement after evaluation by the institutional review board and with consent from the patient and his family. Under general

Fig. 3 (A and B) Preoperative left internal carotid angiogram (A) AP view; (B) lateral view. (C and D) Preoperative right vertebral angiogram (C) AP view; (D) lateral view. The left internal carotid artery angiogram showed the direct carotid cavernous fistula in the cavernous position. The arrow point is the fistula of the shunt. The right vertebral artery angiogram showed the retrograde flow from the left PcomA to the fistula. The arrow point is the fistula of the shunt. AP: anterior-posterior; Lat: lateral

anesthesia, a guiding catheter was navigated to the left internal carotid artery, and, after the true lumen of the internal carotid artery was secured with an Aguru Support 0.014 inch 300 cm (Boston Scientific, Marlborough, MA, USA), a GRAFTMASTER RX 4 × 16 mm (Abbott, Chicago, IL, USA) was guided to the fistula, where it was dilated and deployed at 15 atmospheres (**Fig. 4A** and **4B**). Since affixation was partially inadequate, a Gateway 3.5 × 8 mm (Stryker, Kalamazoo, MI, USA) was dilated at 8 atmospheres (**Fig. 4C**), resulting in the disappearance of the shunt and restoration of anterograde blood flow in the left internal carotid artery (**Fig. 4D** and **4E**). Ophthalmic symptoms were immediately relieved with disappearance of pulsatile tinnitus.

Discussion

TCCF is a rare disorder that accounts for 0.17%–1.01% of traumatic brain injuries, and it is frequently caused by skull fracture near the internal carotid artery. The internal carotid artery is fixed by the dural ring between the lacerated foramen and anterior clinoid process, and, if it is stretched by

head trauma, fistula is formed due to concentration of external force on the fixed part.¹⁾

Symptoms of TCCF include blepharoptosis (72%–98%), bulbar conjunctival congestion (55%–100%), vascular murmur (71%–80%), and headache (25%–84%), and they may be accompanied by ophthalmic symptoms such as diplopia (88%), blurred vision, and ophthalmoplegia, and all symptoms show rapid exacerbation.³⁾

The optimal treatment for TCCF is closure of the fistula with preservation of the internal carotid artery. Shunt occlusion with a balloon catheter was reported in the early 1970s, and closing of fistula with a detachable balloon became the first-line treatment since the late 1970s. Thereafter, as manufacturing of detachable balloons was discontinued, treatment using detachable coils has been performed, but cure has become increasingly difficult. If the fistula is large, there is a risk of protrusion of coils toward the internal carotid artery and incomplete occlusion, but tight packing may cause mass effect to the cavernous sinus.⁴⁾

Presently, various techniques and devices are applied to close fistulas, including balloon-assisted coil packing,⁵⁾ covered stents,⁶⁾ flow diverting stents,⁷⁾ and liquid embolic agents such as Onyx,⁴⁾ but it is necessary to accurately determine the site of fistula in the internal carotid artery, measure the fistula diameter, and select the procedure and device accordingly.⁸⁾ There has not been a report evaluating the relationship between the vascular structure or diameter of fistulas in the internal carotid artery and the procedures selected for their treatment, probably because of the difficulty in evaluating the vascular structure and diameter of fistulas by common cerebral angiography. At our facility, we have examined the structure and measured the diameter of the fistula by 3D rotation imaging from the target internal carotid artery using Artis Q System (Siemens). Particularly, when the fistula was large, and the contrast agent did not reach the intracranial vessels, 3D rotation imaging using a cross flow by contralateral internal carotid artery or vertebral artery angiography was useful. Regarding methods for accurate imaging of fistulas, Tsuji et al. reported that better understanding of the vascular anatomy and more accurate measurement of the fistula diameter were possible by intentionally creating a cross flow from the vertebral artery while occluding the proximal side of the internal carotid artery with a balloon guiding catheter.⁹⁾ In addition, it is desirable to select an even more appropriate therapeutic procedure by more accurately determining the site and size of the fistula including

Fig. 4 (A) Preoperative right vertebral angiogram; (B) intraoperative right vertebral angiogram; (C) intraoperative X ray; (D and E) postoperative left internal carotid angiogram (D) AP view; (E) lateral view. The stiff micro-guide wire (Aguru support; Boston Scientific, Marlborough, MA, USA) was carefully passed through to the distal part of the left internal carotid artery. The covered stent (GRAFTMASTER RX; Abbott, Chicago, IL, USA) was navigated and deployed at the shunting point (A and B). After the deployment of the stent, the angioplasty using the micro-balloon (Gateway; Stryker, Kalamazoo, MI, USA) was added (C). The shunt was disappeared (D and E). AP: anterior-posterior

its spatial relationship with surrounding skull base fracture by the use of new modalities including cone-beam CT.¹⁰ However, if the internal carotid artery is blocked on the proximal side of the fistula for preoperative BOT, the intracranial blood flow of the contralateral internal carotid artery via the anterior communicating artery or of the vertebrobasilar artery system via the posterior communicating artery may be reduced due to the steal phenomenon. Since the possibility of a false-positive result was suggested as in Case 6, it is considered optimal to perform BOT at the fistula. Concerning the evaluation of TCCF and BOT, in our patients, fistulas with a small diameter could be treated by transvenous coil embolization or PTA with preservation of the internal carotid artery, but those with a diameter of 10 mm or greater could not be cured by TVE and eventually required PAO. When a covered stent is used as in this study, fistulas can be closed while preserving the parent artery, but, for this, anatomical understanding including the site and size of the fistula by accurate diagnostic imaging is necessary. The postoperative course was uneventful in all our patients, and the symptoms were resolved within 2 months. Although the number of patients is limited, our results are considered to contribute to the evaluation of the therapeutic strategy for TCCF.

Conclusion

We consider that the strategy of endovascular treatment for TCCF should be evaluated according to the size of the fistula. For this, accurate determination of the size and site of the fistula is important, and the selection of the procedure appropriate for each lesion is necessary.

Disclosure Statement

There are no conflicts of interest to disclose regarding this paper.

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