Case Report

Combined endovascular and microsurgical management of complex traumatic carotid-cavernous fistula: Three case reports

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ABSTRACT

Background: With the evolution of the endovascular devices, the management of endovascular interventions has become the current standard therapy for traumatic carotid-cavernous fistula (TCCF). However, only endovascular treatment may not be feasible in some patients with atypical TCCF.

Case Description: We described three complex TCCFs that could not be managed by conventional endovascular methods. The first patient had recurrent TCCF previously treated by muscle embolization and ligation of affected carotid arteries 23 years ago. Another two patients had TCCFs association with large pseudoaneurysm within the sphenoid sinus. In each patient, the fistula was successfully closed by trapping procedure using a combination of endovascular and surgical treatment.

Conclusion: To reduce costs of treatment, trapping operation by combining surgical and endovascular treatment may be considered as an alternative option for complex TCCF which has some features including chronic stage, preexisting carotid artery ligation, or association with large venous pouch of the cavernous sinus or sphenoid sinus pseudoaneurysm.

Keywords: Chronic recurrent fistula, Direct carotid-cavernous fistula, Sphenoid sinus pseudoaneurysm, Trapping procedure, Traumatic carotid-cavernous fistula

INTRODUCTION

Direct carotid-cavernous fistulas (CCFs) are high-flow shunts directly communicated between the internal carotid artery (ICA) and the cavernous sinus, categorizing as Type A by Barrow classification of CCFs. The most common etiology of direct CCFs is trauma, while spontaneous CCFs usually arise from spontaneous rupture of an aneurysm of the cavernous segment of ICA. Spontaneous resolution of direct CCFs is extremely rare. The most common clinical symptoms, that is, pulsating proptosis, chemosis, and/or audible bruit, depend on the venous drainage. Rarely, aggressive neurological manifestations, especially deterioration of consciousness, may also develop due to cerebral ischemia and/or hemorrhage.
There are two basic forms of therapy for traumatic carotid-cavernous fistula (TCCF) including obliteration of the fistula while maintaining patency of the ICA and sacrifice of the affected ICA. Most TCCF can be treated by endovascular approach with preservation of the parent artery, but in a small number of fistulas cannot be completely occluded without parent vessel sacrifice. In addition, only endovascular approach was not feasible in some cases. The goal of successful treatment of complex TCCF continues to be a major challenge for the neurosurgeon. We described three complex TCCFs that could not be managed by conventional endovascular methods. In each instance, the fistula was, successfully, closed by trapping procedure using a combination of endovascular and surgical treatment.

CASE DESCRIPTION

Case 1

A 64-year-old left-handed woman was admitted to the local hospital due to seizure with transient loss of consciousness. She complained of headache and mild cognitive impairment for the past 1 year. A computed tomography (CT) scan of the head showed markedly dilatation of the left CS, sphenoparietal sinus, and cortical veins along left cerebral hemisphere. There was some venous wall calcification [Figure 1]. Cranial magnetic resonance imaging, magnetic resonance angiography, and magnetic resonance venogram demonstrated left CCF with extensive cortical venous reflux [Figure 2]. Her medical history showed an accident by a fall from a height of about 4 m followed by loss of consciousness and suffered subsequently from right hemiparesis 25 years ago. She was treated by physiotherapy and admitted in the local hospital for 1 month. Two years after an accident, she developed left proptosis and underwent muscle embolization with ligation of ipsilateral neck vessels from another tertiary hospital. Her proptosis completely resolved, and she had no symptoms until the past 1 year. The patient was transferred to our institute for further investigation and proper treatment. Physical examination revealed the previous surgical scar at the left side of her neck [Figure 3]. Visual acuity was 20/50 in the right eye and 20/100 in the left eye. She had no proptosis.

Cerebral angiography was performed before treatment. The left common carotid artery (CCA) injection with 3D rotational angiography showed no the existence of the external carotid artery (ECA), severe stenosis of the proximal ICA, and long segment of multiple small vascular channels, representing the recanalization of the left occluded ICA through vasa vasorum, at C3 to C4 vertebral level. The previous surgical clip is noted [Figures 4a-d]. There was a direct high-flow fistula between the cavernous segment of the left ICA and the CS with retrograde venous drainage into markedly dilated left sphenoparietal sinus, superficial middle cerebral vein (SMCV), superior sagittal sinus through the vein of Trolard, and the left transverse sinus through the vein of Labbé [Figures 4e-i]. Good intracranial collateral circulation of the left cerebral hemisphere from the contralateral ICA and vertebral artery (VA) through enlarged and patent anterior communicating artery (ACoA) and left posterior communicating artery (PCoA) was observed.

Under general anesthesia and heparinization, transarterial detachable balloon embolization was attempted, but the detachable balloon catheter could not be advanced into the fistula through ACoA from the right ICA, and PCoA from the VA. Then, the microcatheter was navigated into the fistula through ACoA from the right ICA. The largest GDC coil was attempted to place into the fistula, but the retrograde shunt into the fistula was so great that the coils mass was unable and kept migrating into the large venous pouch. Subsequently, the microcatheter was advanced further into the petrous segment of the left ICA proximal to the fistula which was occluded with Axium Detachable coil (Medtronic, Minneapolis, Minnesota, USA) [Figures 4j-l]. Then transarterial coil embolization was attempted at left supracavernous ICA, but it...
On the following day, the patient underwent the left frontotemporal craniotomy. Anterior clinoidectomy was subsequently performed, because it was difficult to place the clip on the ICA due to the large arterialized venous pouch. The clinoidal segment of the left ICA was clipped by the fenestrated clip encircling the left optic nerve [Figure 5]. In addition, the left ophthalmic artery (OA) was clipped by another clip. Three hours after surgery, the patient developed generalized tonic-clonic seizure. Cranial CT scan was obtained immediately and demonstrated abnormal hyper density within dilated cortical veins along left cerebral hemisphere, probably representing partially thrombosis of the cerebral veins [Figure 6]. The seizure was controlled by antiepileptic drugs and the patient was discharged home 1 week later. Postoperative course of the patient was uneventful.

One month after surgery, cerebral angiography was obtained and revealed the remaining of severe stenosis of the proximal ICA with associated vasa vasorum and reduction in shunt flow and size of the dilated draining vein along the left cerebral hemisphere [Figures 7a-d]. In addition,
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Figure 4: Patient 1. (a) Anteroposterior (AP) and (b) lateral views with (c, d) 3D rotational angiography of the left common carotid artery injection show no the existence of the external carotid artery, severe stenosis of the proximal internal carotid artery (ICA), and long segment of multiple small vascular channels, representing the recanalization of the left ICA through vasa vasorum, at the level of C3–C4. Previous surgical clip (arrow) is noted. AP views of the (e) left and (i) right ICAs injections and lateral views of (f) the left ICA and the left vertebral artery injections in (g) arterial and (h) venous phases demonstrate the left direct carotid-cavernous fistula (arrowheads) with retrograde drainage into markedly engorged sphenoparietal sinus and superficial middle cerebral veins. Following the occlusion of the left petrous ICA with coils, (j) AP and (k) lateral views of the left ICA injection reveal significant reduction of shunt flow. (l) AP view of unsubtracted image illustrates the entire coil mesh.

The fistula received additional supply from the contralateral ICA and left VA [Figures 7e-h]. Prowler plus microcatheter (Codman Neurovascular, Raynham, MA, USA) was used over the Agility Steerable guidewire (0.016 soft, Codman Neurovascular, Raynham, MA, USA) that was successfully advanced into the fistula through the residual lumen of the left ICA. Embolization was performed with multiple fibered interlocking detachable coils (Interlock-35, Boston Scientific, Natick, MA). Post embolization angiography showed nearly obliteration of the fistula [Figures 7i-I]. Post embolization course of the patient was uneventful. Cranial CT scan, obtained 5 days after the second embolization, revealed abnormal diffuse hyperdensity within the left sphenoparietal sinus, SMCV, the vein of Trolard, and the vein
of Labbé, probably representing ongoing venous thrombosis [Figure 8]. Follow-up cerebral angiography, obtained 1 year after the second embolization, confirmed no recurrence of the fistula [Figure 9]. The patient had recovered completely from neurological deficits. She remained well on follow-up 9 years after combined surgical and endovascular treatment and had few episodes of seizure requiring the continuation of antiepileptic drug. Final cranial CT scan revealed encephalomalacia of the left cerebral hemisphere with multiple scattered ring calcifications [Figure 10].

Case 2

A 14-year-old right-handed boy sustained a severe head injury following a motorcycle accident with multiple organ injuries, including skull base fracture, right traumatic eye injury leading to blindness, left tympanic membrane perforation, and close fracture both bones right forearm. He was admitted to a local hospital and discharged home 3 weeks later. One month later, he developed right proptosis, bilateral audible bruit, and binocular horizontal diplopia. He went back to the previous local hospital and obtained cranial CT scan with contrast injection. The CT scan showed fractures at right-sided planum sphenoidale and superior wall of the sphenoid sinus, enlarged right superior ophthalmic vein (SOV), dilatation of bilateral CS, and large pseudoaneurysm within sphenoid sinus [Figures 11a-c]. These findings were consistent with right TCCF, and the patient was transferred to our institute.

On ophthalmic examination, the right eye had proptosis, chemosis, lateral rectus palsy, fixed dilated pupil, and no light perception. Visual acuity was 20/20 in the left eye.

Cerebral angiography was performed and revealed direct high-flow fistula between clinoidal segment of the right ICA
Figure 6: Patient 1. (a) axial, (b) sagittal, and (c) coronal images of noncontrast cranial computed tomography scan, obtained 3 h after surgery, reveal abnormal hyperdensity within dilated cortical veins along left cerebral hemisphere, probably representing partially thrombosis of the cerebral veins.

Figure 7: Patient 1. Cerebral angiography obtained 1 month after surgery. (a-d) Anteroposterior (AP) and lateral views of the left common carotid artery injection reveals the remaining of severe stenosis of the proximal internal carotid artery (ICA) with associated vasa vasorum and reduction in shunt flow and size of the dilated draining vein along the left cerebral hemisphere. (e) AP view of the right ICA injection demonstrates the clival branch (arrowhead) from the right meningohypophyseal trunk supplying the fistula. AP views of the (f) right and (g) left vertebral arteries (VAs) and (h) lateral view of the left VA injections show the reconstitution from the muscular branch (arrows) of the left VA feeding the fistula. After embolization with coils at the fistulous part, (i) AP and (j) lateral views of the left CCA and (k) AP and (l) lateral views of the left VA confirm nearly complete obliteration of the fistula.
and dilated right CS with a large pseudoaneurysm arising from medial aspect of the right clinoidal ICA and extending inferiorly into sphenoid sinus. The right ICA terminated in the fistula, and there was no filling of the distal carotid artery. The large pseudoaneurysm located just proximal to the site of CCF. Multiple venous drainages included right SOV, right SMCV, right inferior petrosal sinus (IPS), right basal vein of Rosenthal to the vein of Galen, and posterior intercavernous sinus draining into the left CS. The subsequent venous drainages from the left CS ran anteriorly into the left SOV, and posteroinferiorly into the left IPS [Figures 11d-e]. The right ECA injection demonstrated retrograde flow through the right OA through meningo-OA anastomosis, supplying the fistula [Figure 11f]. The fistula also fed by the clival branch from the left meningohypophyseal trunk (MHT) of the left ICA. There was good intracranial collateral circulation through the circle of Willis with patent ACoA and right PCoA to right hemisphere [Figures 11g-i]. We decided to treat this patient using sacrifice the right ICA.

Under general anesthesia and heparinization, transarterial detachable balloon embolization was attempted to place to balloon across the orifice of the fistula, but the balloon always migrated into the pseudoaneurysm. It was impossible to place a balloon in a stable position across the orifice of the fistula, and we were unable to navigate either a balloon

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**Figure 8:** Patient 1. (a) axial, (b) sagittal, and (c) coronal images of noncontrast cranial computed tomography scan, obtained 5 days after second embolization with coils, reveal abnormal hyperdensity within dilated cortical veins along left cerebral hemisphere, probably representing ongoing venous thrombosis.

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**Figure 9:** Patient 1. Cerebral angiography obtained 1 year after the second embolization. (a) Anteroposterior (AP) and (b) lateral views of the left common carotid artery, (c) AP view of the right internal carotid artery, (d) AP view of the right external carotid artery, and (e) AP and (f) lateral views of the left vertebral artery injections confirm no recurrence of the fistula.
or microcatheter beyond the fistula to allow distal control. Therefore, the right ICA was occluded with detachable balloons at two points including just proximal to the fistula and at the cervical portion of the right ICA. The right CCA injection confirmed complete occlusion of the right ICA with intradural ICA opacification through retrograde filling of the right meningo-OA anastomosis [Figure 12]. The VA injection demonstrated the remaining of retrograde filling of the intracranial portion of the right ICA and the fistula through the PCoA. The decision was made to proceed with surgery by clipping of the right supraclinoid ICA distal to the fistula.

On the following day, the patient underwent the right frontotemporal craniotomy. The right optic nerve and the ophthalmic segment of the right ICA were identified under microscope [Figures 13a and b]. Anterior clinoidectomy was subsequently performed, because we planned to place the clip proximal to right OA. However, the right OA could not be identified due to large venous pouch. Therefore, the aneurysmal clip was applied on the ophthalmic segment of right ICA. After clipping the right supraclinoid ICA, the right arterialized sylvian veins changed in color from arterialized reddish color to purple [Figures 13c and d]. Postoperative course was uneventful.

Follow-up cerebral angiography, obtained 2 weeks after surgery, revealed the small residual fistula supplied by the right OA reconstituted from the right middle meningeal artery and the clival branch from the left MHT with draining into the right SOV, IPS, cortical, and deep venous systems. The left vertebral angiography filled the right middle cerebral artery (MCA) through the right PCoA and the left ICA filled the right anterior cerebral artery (ACA) through ACoA. In addition, maximum intensity projection reformatted images of angiographic CT illustrated the disappearance of the pseudoaneurysm within the sphenoid sinus. Transvenous embolization was subsequently carried out through the right IPS using fibered interlocking detachable coils (Interlock-35, Boston Scientific, Natick, MA) densely packing in the proximal right SOV and within the CS [Figure 14]. He had resolution of proptosis and audible bruit with gradual improvement of the right lateral rectus palsy. Cerebral angiography, obtained 1 year after the second embolization, confirmed no recurrence of the fistula [Figure 15].

Case 3

A 16-year-old right-handed man was involved in a motorcycle accident and sustained a severe head injury. Cranial CT scan showed acute subdural hematoma at right frontotemporal region and multiple fractures affecting the right frontal bone, orbital bone, zygoma, skull base, ethmoid bone, and sphenoid bone with bone spicule within the sphenoid sinus [Figure 16a]. The patient underwent emergency craniotomy with removal of blood clot and

![Figure 10: Patient 1. Cranial computed tomography scan obtained 9 years after treatments. (a-f) Sequential axial images of noncontrast cranial computed tomography scan reveal encephalomalacia of the left cerebral hemisphere with multiple scattered ring calcifications.](image)
open reduction and internal fixation of frontal and orbital bones at the local hospital. The following day, he regained consciousness and was blind in the right eye. Few days later, he developed right proptosis and audible bruit on the right side. The patient was transferred to our institute for further investigation and management. On ophthalmic examination, the right eye had proptosis, chemosis, ptosis, total ophthalmoplegia, fixed dilated pupil, and no light perception. Visual acuity was 20/20 in the left eye. The patient had right-sided weakness (muscle strength 4/5).
Cerebral angiography was carried out and disclosed direct high-flow fistula from medial tear of the clinoidal segment of the right carotid artery draining into dilated right CS with a large lobulated out-pouching pseudoaneurysm protruding through into the right sphenoid sinus. The pseudoaneurysm arising from medial aspect of the right clinoidal ICA and extending inferiorly into sphenoid sinus [Figures 16b and c]. The ECA injection showed no supply to the fistula. There was no antegrade flow filling the right ACA and MCA. There was good intracranial collateral circulation through the circle of Willis with patent ACoA and PCoA, respectively. The decision was made to proceed with distal trapping by clipping of the right supraclinoid ICA. On the same day, the patient was moved to operating room and underwent the right frontotemporal craniotomy. The right optic nerve and the ophthalmic segment of the right ICA were identified under microscope [Figure 17a]. Without anterior clinoidectomy, the right OA could identify, and side blending clip was then applied on the ICA proximal to the OA [Figure 17b]. The patient made an uneventful recovery.

Follow-up cerebral angiography, obtained 1 month after surgery, confirmed complete obliteration of the fistula and associated pseudoaneurysm [Figure 18]. The proptosis, chemosis, and audible bruit subsided within 1 month. The ophthalmoplegia was no longer present at 1 year follow-up. At 2 years after treatment, CT angiography was carried out and revealed no recurrence of the fistula [Figure 19].

DISCUSSION

The evolution of the various strategies used to treat TCCF

Over the past 200 years, the treatments of high-flow TCCF have been developed with four main principles including the obstruction of the flow in the arterial afferent to the fistula such as ligation of the carotid artery, the occlusion of the venous exits of the CS such as ligation of the SOV, the promotion of thrombosis formation within the CS, and direct closure at the fistula site.

In 1809, the first treatment strategy for TCCF was the ligation of CCA. The ligation of vessel has been developed in various forms including ligation of ICA, ECA, or orbital vein. In case neither ligation of the affected ICA nor SOV leads to a cure, ligation of the opposite carotid artery may be performed with very cautious. Patients harboring TCCF may develop severe cerebral ischemia following ligation of the affected carotid artery despite angiographic studies indicating excellent collateral potential from the opposite carotid system. The carotid compression test should be performed before every ligation procedure to prevent cerebral infarction after the ligation of the carotid artery.

The rate of cure with the ligation of cervical vessels was <50%. Due to collateral recruitment through the circle of Willis to supply the supraclinoid segment of ICA following carotid artery ligation, trapping procedure by adding intracranial clipping of ICA distal to the fistula has been developed. The OA can be sacrificed during surgery without loss of vision in many cases due to adequate collateral filling from branches of the ECA supplying the central retinal artery. However, some
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Figure 14: Patient 2. (a) Axial view of cranial computed tomography (CT) bone window scan demonstrates the surgical clip near the right anterior clinoid process. Sagittal maximum intensity projection reformatted images of angiographic CT (b) before and (c) after surgery reveal the disappearance of the pseudoaneurysm within the sphenoid sinus. Lateral views of (d) arterial and (e) venous phases of the right external carotid artery injection reveal the residual fistula fed by the right ophthalmic artery (asterisk) reconstituted from the right middle meningeal artery. (f) AP view of the left internal carotid artery injection demonstrates the clival branch (arrow) from the left meningohypophyseal trunk supplying the fistula. (g) AP and (h) lateral views of the left vertebral artery injection show good collateral supply of the right cerebral hemisphere from the vertebrobasilar system via the right posterior communicating artery. (i) Lateral view of unsubtracted image illustrates the entire coil mesh after embolization.

difficulty may be encountered in exposing the OA during surgery, because it usually arises from the ICA beneath the optic nerve.\textsuperscript{[16]} Based on the review of surgically treated cases of direct CCFs by Hamby,\textsuperscript{[22]} proximal occlusion of ICA was more effective than ligation of the CCA, but neither of these methods was a...
satisfactory treatment for direct CCF. Simultaneous intracranial and extracranial occlusions of the affected carotid artery have given far better results than primary occlusions of the carotid artery. None of these patients have suffered neurological deficits and none has lost vision in the involved eye. Nevertheless, some authors found that there was high percentage of patients
Gurdjian introduced a strip of muscle about 20 cm long and 0.5 cm in thickness into the affected ICA and to plug the artery as high as possible by pushing the muscle toward the cranial cavity with an appropriate probe for complete cure of a direct CCF. They speculated that the ligation of the carotid artery may cause the formation of blood clot which will eventually obliterate the fistula in the CS. With his technique, a much shorter distance is left for further clotting to obstruct the fistula in the ICA. In 1937, Browder described the successful treatment of TCCF by open surgery with direct occlusion of an arteriovenous fistula with strips of muscle following the ligation of the affected ICA. In 1965, Parkinson described the first case of successful direct surgical approach to the cavernous segment of the right ICA for packing the sinus with muscle under hypothermia and cardiac arrest. Almost two decades later, Dolenc proposed the technique of direct microsurgical repair of intracavernous vascular lesions, including CCF, without hypothermia, and cardiac arrest based on a thorough anatomical knowledge of the CS and advances in microsurgery. However, this technique has not been widely used due to the risk of damaging the cranial nerves and requiring a highly skilled surgeon.

Before an era of advanced embolization technique, Hamby proposed a definitive operation by clipping the intracranial carotid, followed by embolization of the fistulous carotid segment with a strip of muscle and occlusion of the extracranial common, internal, and external carotid arteries. Even though it is difficult to do, the OA should be clipped if possible. In 1974, the introduction of balloon catheter and detachable balloon by Serbinenko has tremendously changed the treatment of high-flow CCFs. Subsequently, Debrun et al. developed detachable balloon with their technique and treated many TCCF by inserting through the affected carotid artery into the fistulous sac. In 1979, Mullan recommended treatment of CCFs using thrombogenic techniques by open surgery and retrograde packing of the CS through the ophthalmic vein and/or superior petrosal

with postoperative visual deterioration, cerebral ischemia, or catastrophic complication following surgical treatment, including carotid artery ligation or trapping procedure. Furthermore, only 50% of patients having trapping operations were completely cured. In 1931, the new idea to attack on a TCCF was proposed by inserting an embolus of muscle into the ICA which would be propelled by the blood flow to the fistulous opening (i.e., Brooks method). Subsequently, Lang and Bucy reported successful treatment of TCCF by the Brooks method. They described a case, in which embolization of muscle alone has proved to be adequate treatment for TCCF and suggested that this method should be considered as the primary treatment. Another adaptation of the Brooks technique,
exits with thrombogenic materials, inducing thrombosis within the sinus. He could preserve carotid artery patency in most patients with no visual or ischemic complications after procedures.

Since 1982, there were several reports of complications of detachable balloon catheter technique in the treatment of TCCFs such as inadvertent balloon detachment, the balloon rupture, early deflation, or shrinkage of the balloon, allowing the fistula to reopen, production of cranial nerve palsies when multiple balloons are used. After the withdrawal of detachable balloon from the U.S. markets leading to the lack of availability of detachable balloon, both transarterial and transvenous coil embolization were adapted with adjunctive techniques of transarterial balloon or stent protection of the parent vessel. Furthermore, covered stent and flow diverter stent have become an alternative option. Due to high cost, long procedure time, requiring the long-term follow-up outcome, and unfamiliarity for interventionists of these devices, however, detachable balloon embolization remains the treatment of choice in many countries, including our country. At present, the goal of treatment for TCCF is to obliterate the fistula completely while maintaining a patent carotid flow.

Recurrent TCCF following muscle embolization, ICA ligation, or combination

The incidence of recurrent TCCF following the carotid artery ligation remains unknown. Patients harboring recurrent TCCF usually presented with ocular symptoms from dilated affected SOV. The supply to the recurrent TCCF from the recanalized previously ligated ICA or hypertrophy of the vasa vasorum is extremely rare. Garcia-Cervigon et al. reported a case of recurrent TCCF 10 years after treatment by placement of the Fogarty catheter in the affected ICA at the level of the fistula followed by surgical ligation of the CCA. The fistula was fed by the distal portion of the affected ICA through PCoA and meningeal branches of the ECA opacified through the ascending cervical and occipital arteries. The venous drainage was almost exclusively cortical veins. Similarly, recurrent TCCF in our first case was supplied by multiple feeders coming from ACoA, PCoA, and the recanalized ICA with hypertrophic vasa vasorum. The venous drainage was exclusively cortical draining veins.

In the past three decades, the use of muscle embolization, trapping, and/or cervical carotid artery ligation remained to be utilized in our country due to extenuating circumstances such as poor socioeconomic status. The greatest factor for the failure of the muscle embolization is the inability to select an appropriate embolus sized precisely to fit an orifice of the fistula. In our first case, the patient underwent previous muscle embolization with ligations of internal and external carotid arteries. The embolus of muscle may be too small and pass through the orifice of the fistula, but it could occlude the affected ophthalmic draining vein leading to the resolving of proptosis. However, the fistula remained persist and the draining veins may reroute into the sphenoparietal sinus, SMCV, and cortical veins.

In case of recurrent TCCF with previously ligated ICA, the fistula can be closed through different routes including transarterial through ACoA from the contralateral ICA or PCoA from the vertebrobasilar system, or transvenous route through SOV or IPS, direct surgical exposure of the CS, and direct puncture or exposure through the remnant of the affected cervical ICA above the occlusive site. Barrow et al. suggested avoiding direct carotid puncture for balloon embolization due to requiring a large introducer sheath under heparinization, risking of an expanding hematoma formation, and leading to upper airway obstruction and acute respiratory insufficiency. In addition, direct surgical approach to the CS, especially in case of chronic recurrent TCCF, may lead to high morbidity and mortality result.

In our first case, we first attempted embolization with detachable balloon through ACoA and PCoA, but it was difficult to navigate the balloon catheter into the fistula due to relatively stiff device. Therefore, we used the microcatheter navigating through ACoA from the contralateral ICA for embolization with coils. Unfortunately, the retrograde shunt into the fistula was so great that the coils mass was unable and kept migrating into the venous pouch. Then, we decided to trap this fistula with coiling proximal and distal to the fistula. However, it was impossible to safely pack the supraclinoid ICA after packing the petrous segment ICA. The fistula showed inaccessible through draining venous systems. Consequently, we chose intracranial clipping of the supraclinoid ICA.

Chronic recurrent or occult TCCF after trapping operation

The presence of a long-standing TCCF, which was sustained more than 20 years, is extremely rare. In our first case, without other neurological symptoms and follow-up cerebral angiography, TCCF was sustained for more than 20 years until her seizure developed. The enormously draining veins in our first case may represent the presence of a long-standing TCCF.

O’Reilly et al. reported a case of TCCF occurred 16 years after a Hamby procedure, clipping of supraclinoid ICA, muscle embolization, and ligation of cervical carotid arteries, presenting with subarachnoid hemorrhage. The fistula was supplied by meningeal collateral arteries and drained exclusively into deep venous system with aneurysmal dilatation, source of hemorrhage. There was no drainage into SOV and IPS. They predicted that there could be other
patients who had combined arterial trapping with muscle embolization of the fistula resulting in an occult fistula with the risk of an intracranial hemorrhage. Similarly, SOV and IPS were not dilated in our first case, and there was ectasia of the cortical veins with aneurysmal dilatation. Fortunately, no intracranial hemorrhage, probably resulting in severe neurological sequelae, occurred in our first case.

The fistula may persist or recur if the cavernous segment of ICA is not completely thrombosed following the trapping procedure. Failure of trapping operation for TCCF may cause by the collateral supply, including the ophthalmic system, ECA, and contralateral MHT, reconstituting the trapped cavernous segment and supplying the fistula. Interestingly, Kapp et al. reported a case of gunshot injury with the persistent TCCF for 12 years previously treated by many methods including intracranial and extracranial ligation of the ICA with muscle embolization of the fistula, ligation of ECA and CCA, direct surgical exposure with packing ICA with muscle, ligation of supracleidoid carotid branches and OA, and neck exploration with muscle embolization. The persistent fistula was supplied by muscular branches of the VA entered the affected ICA near the base of the skull. In our first case, the fistula also had an additional supply from muscular branches of the VA into the fistula, which is extremely rare.

**TCCF in association with extensive venous congestive encephalopathy (EVCE)**

In cerebral vascular lesions such as brain arteriovenous malformations and dural arteriovenous fistulas, EVCE is condition when too much blood drained into the venous systems involving extensive cerebral veins. The progression of EVCE can be divided into acute and chronic stages. This condition often indicates a more aggressive natural history. The common clinical manifestations, the consequence of venous congestion, usually consist of headache, cognitive impairment, seizure, focal deficits, and/or hemorrhage. Progressive cognitive impairment is often in chronic stage. At chronic stage, tortuous, dilated, and engorged veins with calcification could be seen on imaging findings. Prompt treatment is warranted, and complete elimination is often difficult. Therefore, staged treatment may be chosen. Aggressive treatment is effective and results in an acceptable prognosis. In our first case, the patient’s symptoms, course of the disease, and imaging findings were probably consistent with chronic EVCE. Staged and aggressive treatments may suitable for this patient.

**Concomitant TCCF and pseudoaneurysm within the sphenoid sinus**

Patients harboring the traumatic pseudoaneurysm within the sphenoid sinus usually exhibit the classic triad of unilateral blindness, orbital fracture, and massive epistaxis. Sphenoid sinus pseudoaneurysm originates from the cavernous segment of ICA caused by traumatic vessel injury and located in the sphenoid sinus. Sphenoid sinus pseudoaneurysms is classified into two types including with and without CCF depending on the site of the injury. TCCF concomitant with sphenoid sinus pseudoaneurysm has rarely been reported. Most patients suffered multiple skull fractures, especially the lateral wall of the sphenoid bone, adjoining the sphenoid sinus and ethmoid sinus. This entity may present either massive epistaxis, a life-threatening clinical situation, or ophthalmic symptoms of venous engorgement, including proptosis, chemosis, bruit, and vision impairment. Early recognition is essential to prevent the development of severe massive hemorrhage from epistaxis. Fortunately, we recognized sphenoid sinus pseudoaneurysm in our case 2 and 3 before the development of massive epistaxis.

The management of a TCCF associated with traumatic aneurysm in sphenoid sinus can be challenging. The inflation of a balloon within pseudoaneurysm association with TCCF can cause a rupture, resulting in fatal massive intracranial hemorrhage. Isamat et al. described a case of TCCF combined with a large pseudoaneurysm. They tried to preserve the ICA circulation and treated this fistula by open surgery and direct intracavernous obliteration with muscle and fibrin sealant. The fistula was closed, but a large pseudoaneurysm remained persist following by the ligation of the cervical ICA. Van Dellen suggested that the false traumatic aneurysm of the carotid artery in the CS should be treated by trapping procedure instead of direct clipping which had high morbidity and mortality.

Recently, Cho et al. demonstrated successful treatment of TCCF accompanied by a giant pseudoaneurysm in the sphenoid sinus by endovascular treatment with intra-arterial approach using multiple coils and an ethylene-vinyl alcohol (EVOH) copolymer-based liquid embolic agent injection while protecting the injured site of the ICA with a balloon catheter. However, this technique required much higher cost than our technique. In addition, delayed migration of an EVOH copolymer-based liquid embolic agent may occur following embolization of TCCF.

**Trapping procedure by combined endovascular and surgical approach for TCCF**

The management of CCF is variable and depends on the anatomy of the fistula. Surgical assistance for TCCF should be limited to those case when the affected ICA cannot be preserved, or when the affected ICA has been ligated previously. The CS can be reached through different approaches including transarterial or transvenous route, and direct surgical exposure. Therefore, each technique may be considered depending on hemodynamic...
profile, the angioarchitecture of the fistula, feeding arteries, and venous draining veins.

Combined endovascular and surgical treatments for concurrent of a TCCF and sphenoid sinus pseudoaneurysm have rarely been reported. At present, Ghorbani et al.\[19\] reported a case of TCCF with an associated giant traumatic aneurysm in the sphenoid sinus presented with impaired vision due to progressive mass effect of the aneurysm. Using endovascular treatment with coiling, the sac of the aneurysm was occluded partially, while the cavernous segment of the affected ICA and fistula was occluded entirely. Subsequently, the patient underwent endoscopic transnasal removal of the thrombosed aneurysm for relieving the mass effect from bilateral optic nerves and chiasma with partial improvement of the initial visual symptoms.

Another technique of endovascular trapping of TCCF by Coley et al.,\[6\] they described the successful treatment of large high-flow TCCFs using a combination of proximal balloon occlusion and distal coiling of the parent vessel. The distal coil embolization was achieved by following retrograde catheterization of the distal parent vessel through the contralateral ICA or ipsilateral VA. They also suggested that if it had proved impossible to gain access to the distal carotid through the collateral circulation, surgical approach would have been required.

For sacrifice the affected ICA with balloon, the balloon should be inflated against the opening of the fistula.\[17\] The second balloon should be placed below the first one for preventing from the first balloon becoming a distal embolus when it deflates prematurely.\[17\] Debrun et al.,\[12\] reported three patients harboring TCCF with incomplete closure or reopening of the fistula due to balloon migration or deflation at the fistula site, while the affected ICA remained occluded by a proximal balloon. These patients were treated by intracranial ligation of the ICA associated with ligation of the OA when the ICA could not be clipped below it. The fistula was cured in all three cases. Similarly, we could not place the balloon against the opening of the fistula in our second and third cases harboring TCCF associated with large pseudoaneurysm within the sphenoid sinus. Therefore, we decided to trap the cavernous segment ICA by proximal balloon occlusion and intracranial clipping of the ICA distal to the fistula.

Follow-up cerebral angiography is essential to verify a cure of TCCF after the trapping procedure. Following trapping procedure for TCCF, the recurrent or residual fistula can be reached through different approaches including direct surgical approach, direct percutaneous transorbital approach, or transvenous approach through SOV or IPS.\[12,13,47,53\] In our first case, we could fortunately advance the microcatheter through the recanalized previously ligated ICA during the second embolization and pack coils into the residual fistula, which reduced in size and flow after clipping of the intracranial ICA.

Teng et al.,\[47\] proposed an alternative way to treat a recurrent TCCF after a trapping procedure. The patient underwent ten operations for recurrent TCCF over the past 16 years. They performed direct puncture of the CS through a transorbital approach through the superior orbital fissure for embolization with coils and glue with successful result. However, the patient had subconjunctival hemorrhage for several days and ptosis for about 2 months.

The development of indirect CCF following the treatment of TCCF

Yoshino et al.,\[34\] reported the development of indirect CCF or cavernous dural arteriovenous fistula following trapping procedure for TCCF. They speculated that angiogenic factors, expressing during healing of the lacerated CS wall, and venous hypertension attributed to direct CCF may increase the risk of the development of the fistula supplying by dural branches. However, there was preexisting dural supply to TCCF before trapping in this case report. In addition, Terada et al.,\[48\] reported the de novo dural arteriovenous fistula appearing 5 months after complete closure of TCCF by transarterial embolization with detachable balloon. The dural supply to TCCF did not be mentioned before the treatment. There was a pseudoaneurysm due to the deflated balloon. They suspected that long-term venous hypertension in TCCF and CS thrombosis following balloon embolization may be causal factors of this dual fistula in the affected CS. Furthermore, Lv et al.,\[47\] reported the development of indirect CCF following treatment of TCCF by covered stent. Ideally, the treatment of TCCF should include occlusion at the site of the fistula. In our opinion, indirect CCF may develop in case of incomplete closure of TCCF after treatment, especially in fistula with pre-existing dural supply before treatment, as shown in our first and second cases.

The venous approach may be indicated when the fistula has been previously trapped but is still function.\[12\] Including our second case, most previous case reports of the development of indirect CCF after treatment of TCCFs were successfully obliterated by transvenous embolization with coils through IPS.\[48,54\]

CONCLUSION

The treatment of TCCF has evolved from surgery to endovascular management using detachable balloon or coils through arterial or venous approach. The endovascular method currently is the treatment of choice for TCCF due to its ability to preserve the carotid artery and flexibility in treatment strategy with various approaches to the fistula. However, surgical assistance may remain useful for some atypical TCCFs.
To reduce costs of treatment, trapping operation by combining surgical and endovascular treatment may be considered as an alternative option for complex TCCF which has some characteristics including chronic stage, preexisting carotid artery ligation, or association with large venous pouch of the cavernous sinus or sphenoid sinus pseudoaneurysm.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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