Post-traumatic persistent trigeminal artery-cavernous fistula: a case report and review of literature

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Persistent trigeminal artery (PTA) is a rare abnormal vascular anastomosis connecting the internal carotid artery (ICA) with the vertebrobasilar artery. The incidence is 0.1%-0.6% in adult cerebral angiography.1,2 We have recently treated a rare case of post-traumatic PTA-cavernous fistula. In this study, we discussed the mechanisms and treatment of this disease by reviewing literature.

CASE REPORT

General data of patient
A 28-year-old man had no history of diseases or major trauma, but 10 months ago before admission, he was injured in the head and suffered from bleeding in the ear and nose in a traffic accident. Head CT indicated “multiple brain contusions, subarachnoid hemorrhage, and ethmoid and transsphenoidal visible blood”. He received conservative treatment for one month in another hospital and discharged with recovery. One month ago, he had symptoms of swelling, pain, vision declining, and double vision in the left eye, and the symptoms were getting worse with time passing by. Physical examination showed conjunctiva congestion and edema in the left eye, vision being 0.3, congestion and edema in the retina and optic nerve head found by fundus examination, limited abduction of the eye ball but without intracranial noises. Eye color Doppler reported as follows: “Significantly widened vein is observed, up to 9 mm in width in the left superior ophthalmic vein, and pulsed wave Doppler (PW) measurement indicates the spectrum of artery with blood-like venous flow”. Angiogram of the left ICA (Fig. 1A) showed that the blood in the left cavernous sinus in the early image was supplied by the PTA via drainage from the left enlarged superior ophthalmic vein, but no obvious drainage from the inferior petrosal sinus and the cortical veins. Imaging of the left external carotid artery (ECA, Fig. 1B) showed that multiple branches of accessory meningeal artery and foramen rotundum artery also contributed to the blood supply, but with less blood flow. Angiogram of the left vertebral artery (VA, Fig. 1C) showed that the basilar artery supplied blood to the left cavernous sinus through PTA and the development of the posterior communicating artery was abnormal, while the development of the vertebrobasilar system was normal. Brain angiography did not show aneurysms or any other abnormalities in the other vascular blood vessels.

Surgical procedures
Under anesthesia with neuroleptic agents (5 mg Droperidol and 0.1 mg Fentanyl) and local anesthesia (5 ml 1% Lidocaine), a 6F guiding catheter was placed in the left common carotid artery (CCA), then an Echelon-10 microcatheter carrying silver-speed-10 microguidewire was ultra-elected into the trigeminal artery. We tried repeatedly to put the microguidewire and microcatheter through the fistula into the left cavernous sinus, but failed, because the fistula was too small (as confirmed by microcatheter imaging). Therefore, we put a 2 mm×2 cm Sapphire NXT coil to occlude both the trigeminal artery and the fistula. Angiogram of the left ICA showed that the fistula had completely been occluded and no image of the cavernous sinus was displayed. The guiding catheter was introduced into the head end of the left ECA, the microcatheter was introduced into the accessory meningeal artery, and 0.4 ml of Onyx glue was injected at a slow speed but steadily (with the rate of 0.1 ml per minute) under roadmap imaging. During the injection, we stopped injecting twice (1 minute each) to allow the full diffusion of the Onyx glue into the small branches of
the accessory meningeal artery to the fistula. And then we withdrew the microcatheter to the meningeal artery trunk and slowly injected 0.3 ml of Onyx glue again. After injection, we pulled out the microcatheter. We also put the same model of the microcatheter with a microguidewire to the foramen rotundum artery and slowly injected 0.4 ml of Onyx glue.

**Results**

Angiogram of the left CCA and left VA after embolization (Figs. 2A and 2B) showed that the previous fistula, which was supplied by the trigeminal artery, the accessory meningeal artery and the rotundum artery, was completely occluded, and that no image of the cavernous sinus was showed up. Angiogram of the left ICA (Fig. 2C) showed the image of the left cavernous sinus faintly. The potential anastomosis branches between the inferolateral trunk of the cavernous sinus of the left ICA and the foramen rotundum artery were opened and fed the left cavernous (no image showing before surgery), but the blood flow was very small. And no image of drainage vein was showed up. We expected that self-thrombosis might occur in the future and so did nothing with it temporally.

The patient was discharged 5 days after surgery with significantly improved swelling and pain in the left eye, improved vision to 0.5, and mitigated diplopia. The left CCA of the patient was kept oppression after discharge. The patient was followed up for one year and the symptoms and signs completely disappeared. However, the patient refused to receive the examination of digital subtract angiography (DSA) again.

**DISCUSSION**

PTA represents a rare vascular anastomosis between the carotid and basilar arterial systems. In the 3-14-mm embryonic stage, the trigeminal artery normally exists and functions to provide blood flow to the primitive hindbrain. During subsequent embryonic development, the trigeminal artery involutes and disappears with the formation of the basilar system, including the circle of Willis and the vertebrobasilar arteries. Some may remain and form PTAs.  

PTA originates from the carotid cavernous sinus. It starts in the cavernous sinus and gets confluence to the basilar artery between the cerebellar superior and inferior arteries. PTA easily forms aneurysms and ruptures because of the vessel wall defects in development. If the head end of PTA near the cavernous sinus within the ICA ruptures, it will cause the formation of the trigeminal cavernous fistula (TCF), which, but, is rarely seen in clinic. Enomoto et al first reported a case of TCF caused by rupture of the aneurysm at the connection of ICA and PTA. In the follow-up reports, some had aneurysms but some did not have. Most cases were spontaneous. As for the mechanism of spontaneous TCF without aneurysm, Oka et al thought that although no aneurysm was found in some spontaneous TCF, aneurysms might exist as well, because ruptured aneurysms were hard to be found by imaging in the cavernous sinus due to the high blood flow. Another possible explanation is that the vascular wall of the trigeminal arteries easily ruptures due to defects in development. 

Post-traumatic TCF is very rare. In the reported cases, all patients showed carotid-cavernous fistula (CCF) symptoms within a relatively short period of time after trauma and belonging to Barrow type B or type A. McKenzie et al speculated that the mechanism of this disease may be associated with injury-induced rupture of PTA in the cavernous sinus. Flandroy et al reported a case caused by rupture of post-traumatic pseudoaneurysm.

This is the first case report of CCF of Barrow type D, in which CCF symptoms appeared 10 months after head injury. This case is extremely rare. The possible mechanisms may be as follows. Head trauma and fracture of the skull base may cause a small tear and a small amount of bleeding in the PTA in cavernous sinus, which will consequently result in thrombus in the crack. Sometimes later, after thrombus reopens, it becomes a pseudoaneurysm. Rupture of the pseudoaneurysm might cause TCF. We did not find aneurysm by imaging before, during and after surgery. The reason might be that rupture of aneurysm may make it difficult to be found in the cavernous sinus or that aneurysm is too small to be seen. Alternatively, fracture of the skull base damages the PTA and makes its vessel wall so fragile that spontaneous rupture of blood vessels might occur at any time and cause TCF. Initially, it forms CCF of Barrow type B. After a period of time, it becomes complicated CCF of Barrow type D with multiple fistulas because of reopening of the original potential anastomosis among accessory meningeal artery, foramen...
rotundum artery and the cavernous sinus, which is caused by the siphon role of high blood flow within the cavernous sinus or high-pressure in the cavernous sinus.

When making strategies to treat TCF, we should consider Lizuka’s classification:\textsuperscript{10} the naive type, rear circulatory system dependent on PTA (Since PTA is the main blood supply of the rear circulatory system, it cannot be occluded, otherwise it will cause cerebral infarction), and the mature type, rear circulatory system independent on PTA (Since PTA does not play a significant role in intracranial blood supply, it can be occluded and will not cause obvious cerebral ischemia during the treatment).

Endovascular treatment is the first choice to treat TCF either through the transarterial approach or through the transvenous approach or both.\textsuperscript{2} McKenzie et al\textsuperscript{7} thought that, like a typical CCF, balloon embolization had the lowest risk and should be the first choice as treatment method was concerned. Accurate embolization of fistula with electrolysis-coil through the artery had the same good results.\textsuperscript{11} It has been reported that, if the fistula is too small to pass through the cavernous sinus when treating TCF with the mature type of PTA, the embolization of the fistula together with the trigeminal artery could be used successfully in treatment of TCF.\textsuperscript{9}

**Fig.1.** Diagnostic angiographic study before embolization. A: Left internal carotid arteriogram showing opacification of the distended left cavernous sinus supplied by the PTA via drainage from the left enlarged superior ophthalmic vein; B: Left EC arteriogram showing multiple branches of accessory meningeal artery and foramen rotundum artery also attributing to the blood supply to distended left cavernous sinus; C: Left vertebral arteriogram (the left carotid artery kept oppression) showing an enlarged left PTA with posterior to anterior flow through the dilated left cavernous sinus to the superior ophthalmic vein.

**Fig.2.** Angiographic study after embolization. A: Left carotid arteriogram showing the previous fistula supplied by the trigeminal artery, the accessory meningeal artery and the rotundum artery being completely occluded, with no opacification of the cavernous sinus; B: Left vertebral arteriogram showing the previous fistula supplied by the trigeminal artery, the accessory meningeal artery and the rotundum artery being completely occluded, with no opacification of the cavernous sinus; C: Left internal carotid arteriogram showing the potential anastomosis branches between the inferolateral trunk of the cavernous sinus of the left ICA and the foramen rotundum artery reopening and feeding the left cavernous (no image before surgery), but blood flow being very small.
Oka et al thought that transvenous embolization should be chosen only when the orifice of fistula is too small to prevent balloon or microcatheter from entering the fistula. He reported one case of spontaneous TCF with high flow. Embolization was performed with coils passing through the vein into the cavernous sinus owing to no suitable balloon. Fistula disappeared after two weeks as shown by angiography, although embolization of the cavernous sinus was not complete, which may be due to thrombosis in the cavernous sinus induced by the coils. Chan et al improved the treatment method by treating the TCF with transvenous approach. They believed that embolization with transarterial approach or both the transarterial and the transvenous approaches should be performed in the case of trigeminal artery-aneurysms.

In our case, imaging showed that both the anterior and posterior circulatory systems were well developed and independent. The blood circulation of the posterior circulatory system did not depend on PTA. Therefore, it belonged to the mature type, in which PTA could be occluded. Since the trigeminal artery was not large enough to use a balloon, we initially tried to perform complete embolization of the cavernous sinus and the fistula using coils passing through the left ICA-the trigeminal artery-fistula-left cavernous pathway. However, during the surgery, we found that the fistula was too small to insert the microguidewire and microcatheter into the left cavernous sinus. We tried many times, but failed to pass through the fistula. This small fistula might be caused by rupture of small branches of the trigeminal artery in the cavernous sinus. Therefore, we placed a coil in the fistula of the trigeminal artery to block both the fistula and the trigeminal artery and used Onyx glue to block the fistula of the accessory meningeal artery and the foramen rotundum artery. Angiography after embolization showed that the fistulas of the original trigeminal artery, the accessory meningeal artery and the foramen rotundum artery were occluded. The surgery was successful and the clinical symptoms were significantly improved since then. However, angiogram of the left ICA showed that a new extreme small fistula was fed by reopened potential anastomosis artery between the inferolateral trunk of the cavernous sinus of the ICA and the foramen rotundum artery (no image before surgery). As the anastomosed artery was very small and the blood flow into the cavernous sinus was very little, we expected that self-thrombosis might occur in the future, which might be accelerated by oppressing the left CCA. Thus we did not process it temporarily. After one-year tracking, we verified that the treatment was very successful and the symptoms and signs of the patient completely disappeared. This result suggested that the CCF might be self-healing. Therefore, we suggest that tracking the patient and oppressing the carotid artery should be considered when treating very low flow CCF or high flow CCF that turns into very low flow CCF after endovascular treatment. Some researchers reported that the self-healing rate of Barrow type D-CCF could reach 70.9% when it becomes the Barrow type B-CCF after embolization.

Additionally, we suggest that standard angiography should include VA because a PTA may not be shown in the carotid artery imaging. It is necessary to estimate whether the trigeminal-cavernous fistula is completely occluded through angiogram of the left ICA, ECA and VA after embolization, and a very small remnant fistula or a new fistula can be found after angiography.

In conclusion, we suggest the following strategies that balloon or coil embolization should be the first choice to treat high flow TCF with a single blood supply but with no combined aneurysms. If the fistula is too small, embolization of the PTA and the fistula with balloon or coil should be considered for treating the mature type PTA. If failing in the transarterial embolization, we should choose the transvenous approach to perform embolization of cavernous sinus using coil, balloon or Onyx glue. For the treatment of Barrow type D-CCF, we should choose embolization treatment via the transvenous or/and transarterial approaches.

REFERENCES


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