Persistent primitive trigeminal artery–cavernous sinus fistula with intracerebral hemorrhage: endovascular treatment using detachable coils in a transarterial double-catheter technique

Case report and review of the literature

KOJI TOKUNAGA, M.D., KENJI SUGIU, M.D., MASAHIRO KAMEDA, M.D., KYOJI SAKAI, M.D., KAORU TERASAKA, M.D., TORU HIGASHI, M.D., AND ISAO DATE, M.D.

Department of Neurological Surgery, Okayama University Graduate School of Medicine and Dentistry, Okayama; and Department of Neurosurgery, Himeji Central Hospital, Himeji, Japan

Intracerebral hemorrhage occurred in this 61-year-old woman with preexisting diplopia and proptosis. Results of angiography demonstrated a persistent primitive trigeminal artery (PPTA)–cavernous sinus fistula with cortical venous reflux. Two microcatheters were introduced transarterially through the PPTA into the two draining pathways in the cavernous sinus. Coils were delivered in both pathways simultaneously to prevent further venous overload on either path. The fistula was successfully occluded without complication while the PPTA was preserved. The authors describe this double-catheter technique for coil embolization of a fistula and review the literature concerning PPTA–cavernous sinus fistulas.

KEY WORDS • persistent primitive trigeminal artery • intracerebral hemorrhage • cavernous sinus fistula • coil embolization • endovascular therapy

A PPTA is the most frequently discovered persistent primitive communication between the cavernous ICA and the BA and has little or no clinical significance in most cases. In fact, this anomaly appears on only 0.1 to 0.3% of cerebral angiograms. The PPTA originates proximal to the meningohypophyseal trunk, coursing inferior and then superior to the abducent nerve, passes through or lateral to the dorsum sellae, and enters the BA between the superior cerebellar and anteroinferior cerebellar arteries.

We report on the case of a patient with a PPTA–cavernous sinus fistula causing ICH. Use of a transarterial double-catheter technique that did not aggravate hemodynamic stress during embolization successfully occluded the fistula. We report the details of this case and review the literature.

Case Report

History and Examination. This 61-year-old woman was admitted to our hospital with the sudden onset of disturbed consciousness, severe right hemiparesis, and motor-dominant aphasia. She had a history of minor head trauma from 20 and 25 years previously. She reported having tinnitus, diplopia, and left ptosis 5, 3, and 2 years before admission, respectively.

At the time of admission, the patient had an audible pulse-synchronous bruit around the left orbit. An initial computerized tomography scan exhibited a hematoma with perifocal edema in the left mesial temporal lobe and basal ganglia. FIG. 1. Computerized tomography scan obtained on admission, demonstrating a hematoma with perifocal edema in the left mesial temporal lobe and basal ganglia.

Abbreviations used in this paper: BA = basilar artery; CCF = carotid–cavernous fistula; ICA = internal carotid artery; ICH = intracerebral hemorrhage; PPTA = persistent primitive trigeminal artery.
artery angiogram revealed an abnormal artery that arose from the upper BA, passed laterally and then anteriorly, and finally connected to the fistula (Fig. 3). These angiographically demonstrated features served to confirm the clinical diagnosis of a PPTA–cavernous sinus fistula.

Operation. The patient underwent endovascular treatment of the fistula via the transarterial route. After administering a local anesthetic agent, a No. 7 French catheter was placed in the left ICA. Through this guiding catheter, two Rapid-transit microcatheters (Cordis Endovascular Systems, Inc., Miami Lakes, FL) with two markers were advanced into the PPTA past the fistula. One of the catheters was placed at the posterior portion of the sinus connecting with the basal vein of Rosenthal; the other was positioned at the anterior portion draining into the sylvian vein. A fibered Guglielmi detachable coil (Target Therapeutics, Fremont, CA) having a 4-mm diameter was situated in the posterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Simultaneously, an interlocking detachable coil 20 cm long and 7 mm in diameter was detached at the anterior portion. Additional coils were then delivered from both microcatheters. In total, three Guglielmi coils, two interlocking coils, and six fibered microcoils were placed in the affected cavernous sinus to occlude the fistula.

Postoperative Course. Control angiograms demonstrated that the fistula had disappeared and that blood flow was restored in the PPTA from the left ICA to the BA (Fig. 5 arrow). Postoperatively, the patient’s proptosis and diplopia resolved immediately. She was discharged with mild right hemiparesis and dysphasia after 2 months’ rehabilitation following endovascular treatment.

Discussion

Thirteen patients having a PPTA–cavernous sinus fistula have been described in the literature. Clinical symptoms, such as decreased visual function, cranial nerve palsies, progressive proptosis, and tinnitus, do not differ from those caused by a direct high-flow CCF. Intracerebral and subarachnoid hemorrhage can occur in some patients with direct CCFs. Halbach and colleagues10 reported on four cases (3.1%) of intracerebral hematoma from among a series of 127 patients with a direct CCF. The patients in these cases had cortical venous drainage from the fistula that correlated with the site of the hemorrhage. In our patient, the first in the literature to have a PPTA–cavernous sinus fistula with an ICH, the varix on the drainage pathway was believed to be the site of hemorrhage into the mesial temporal lobe and basal ganglia.

Persistent primitive trigeminal artery–cavernous sinus fistulas occur not only at the junction of the ICA and the PPTA,5,12 but also on the PPTA itself,11 as happened in our patient. They can develop spontaneously or be caused by trauma. Enomoto, et al.,6 reported the first case of a PPTA–cavernous sinus fistula, which developed after rupture of a primitive trigeminal artery aneurysm. Guglielmi and associates12 also reported two cases in which a ruptured aneurysm on the PPTA might have been responsible for producing the fistula. It is not always easy, however, to discriminate angiographically a preexisting aneurysm from a pseudoaneurysm or a dilated compartment in the cavernous sinus after a fistula develops. Some authors have asserted that structural fragility of the PPTA itself may be responsible for vessel

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**Fig. 2.** Lateral views of left ICA angiograms obtained during early (left) and late (right) phase, showing an abnormal artery emerging from C-5 (arrow) as well as a fistulous connection with the left cavernous sinus. A varix on the posterior drainage (arrowhead) was believed to be the rupture point that had caused the patient’s hematoma.

**Fig. 3.** Right vertebral artery angiogram (anteroposterior view) exhibiting an abnormal artery arising from the upper BA and connecting to the fistula.

**Fig. 4.** A lateral fluoroscopic image showing a fibered Guglielmi detachable coil (arrow) and an interlocking detachable coil delivered from microcatheters situated posteriorly and anteriorly, respectively.
Double-catheter technique for PPTA–cavernous sinus fistula

rupture either with or without trauma.\textsuperscript{11,12} Although the fistula in our patient was supposed to have occurred spontaneously without severe head injury, her previous minor head trauma might have contributed to a weakened vessel wall.

Balloon occlusion of the fistula, which immediately interrupts the arteriovenous shunt, is the treatment of choice for direct CCFs. Five groups of authors have reported the successful treatment of PPTA–cavernous sinus fistulas with balloon occlusion,\textsuperscript{6,8,11,13} and the PPTA was preserved in two cases.\textsuperscript{11,13} In recent studies, Bernstein, et al.,\textsuperscript{2} and Oka, et al.,\textsuperscript{1} demonstrated the usefulness of transvenous coil embolization for PPTA–cavernous sinus fistulas. In our patient, navigating a balloon through the fistula was difficult because of the tortuosity and the small caliber of the PPTA. We did not use a transvenous approach because the inferior petrosal sinus was occluded. Instead, we introduced two microcatheters transarterially into the PPTA and subsequently into the cavernous sinus through the fistulous portion of the PPTA. One of the microcatheters was placed at the posterior portion of the cavernous sinus adjacent to the varix draining into the basal vein of Rosenthal; the other was positioned at the anterior portion draining into the superficial sylvian vein. Motoyama and associates\textsuperscript{14} described a case of ICH after transvenous coil embolization of a high-flow CCF. They believed that the hemorrhage was caused by venous hypertension from cortical reflux following an incomplete occlusion. Simluoto and colleagues\textsuperscript{19} suggest using great care not to redirect the flow into the remaining venous pathways during coil embolization of direct CCFs and thereby aggravating ocular symptoms or hemorrhage. In our patient, placement of coils proximal to the branching point of draining pathways by using a single microcatheter might have occluded the arteriovenous shunt more simply, but we were not sure whether this maneuver was possible. Given that the posterior portion draining into the basal vein was considered responsible for the patient’s hemorrhage, we chose to occlude this portion completely. Because selective occlusion of the posterior portion might have induced further venous stress in the territory of the superficial sylvian vein, thus resulting in additional hemorrhage, both the anterior and posterior portions were occluded simultaneously by using this double-catheter technique.

Conclusions

We described the first case of a PPTA–cavernous sinus fistula with ICH. Transarterial coil embolization was performed instead of balloon embolization, which seemed difficult because of the tortuosity of the PPTA. Introducing two microcatheters into the cavernous sinus prevented further venous overload during embolization.

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References


Address reprint requests to: Koji Tokunaga, M.D., Department of Neurological Surgery, Okayama University Graduate School of Medicine and Dentistry, 2-5-1 Shikata-cho, Okayama 700-8558, Japan. email: kojitoku@md.okayama-u.ac.jp.