Online February 14, 2023
Journal of Neuroendovascular Therapy 2023; 17: 88–92 DOI: 10.5797/jnet.cr.2022-0058

Case Report

A Case of Iatrogenic Dural Artery–Pial Vein Shunt Treated with Endovascular Treatment

Hideaki Shigematsu, Kazuma Yokota, Akihiro Hirayama, and Takatoshi Sorimachi

Objective: A few cases of postsurgical iatrogenic arteriovenous shunts have been reported, with the arterial blood flow directly entering the pial veins. Herein, we reported a patient with a dural artery–pial vein shunt found 1 year after aneurysmal clipping.

Case Presentation: A 64-year-old male presented with generalized convulsion 1 year after cerebral aneurysmal clipping. A CT showed intracerebral hemorrhage in the temporo-occipital cortex and a dural artery–pial vein shunt in proximity to the previous craniotomy center. The arterial blood flow from the deep temporal artery, the middle meningeal artery, and the anterior auricular branch of the superficial temporal artery shunted into the superficial middle cerebral vein, with evident cortical venous reflux. Embolization was performed with n-butyl-2-cyanoacrylate and completely occluded the shunt. The patient was discharged without neurological deficits.

Conclusion: Endovascular liquid embolization may be an effective treatment for iatrogenic dural artery–pial vein shunt.

Keywords ▶ transarterial embolization, iatrogenic arteriovenous shunts, n-butyl-2-cyanoacrylate

Introduction

The occasional occurrence of iatrogenic arteriovenous (AV) shunts after craniotomy or burr hole surgery has been reported; most of these shunts were dural arteriovenous fistulas (AVFs), with the arterial blood flow entering a dural vein. A few cases of arteriovenous shunt, in which the arterial blood flow directly entered the pial vein, have been reported.[1–5] We describe the case of a patient with a dural artery–pial vein shunt presented as intracerebral hemorrhage occurring after craniotomy, which was successfully treated with endovascular therapy using a liquid embolic material.

Department of Neurosurgery, Tokai University School of Medicine, Isehara, Kanagawa, Japan

Received: September 1, 2022; Accepted: January 17, 2023
Corresponding author: Hideaki Shigematsu. Department of Neurosurgery, Tokai University School of Medicine, 143, Shimokasuya, Isehara, Kanagawa 259-1193, Japan
Email: shige@is.icc.u-tokai.ac.jp

Case Presentation

A 64-year-old male, who had undergone clipping for a ruptured right middle cerebral artery aneurysm 1 year previously at our hospital, presented with general convulsions. He suffered from a transient postictal left hemiparesis and fully recovered the next day. A CT scan showed a hematoma in the right temporo-occipital cortex (**Fig. 1A**). An MRA revealed an anomalous vessel structure in the region of the right Sylvian fissure (**Fig. 1B**). An angiography was performed the next day to obtain the definitive diagnosis. The right external carotid artery (ECA) angiogram showed a dural artery–pial vein shunt in proximity to the previous craniotomy center, supplied by the anterior deep temporal artery (ADTA), the middle meningeal artery (MMA), and the anterior auricular branch of the superficial temporal artery (**Fig. 2A**). On the 3D DSA images superimposed to the skull image, both the ADTA and the anterior auricular artery could be seen entering the intracranial space through the bone edge of the craniotomy site at the temporal base and running on the dural surface to the shunt point (**Fig. 2B**). The right MMA selective angiogram showed that the feeder arteries were combined and flowed directly into the superficial middle cerebral vein (SMCV) (**Fig. 2C** and **2D**). The connection between the SMCV and the cavernous sinus was occluded, and the venous drainage refluxed through

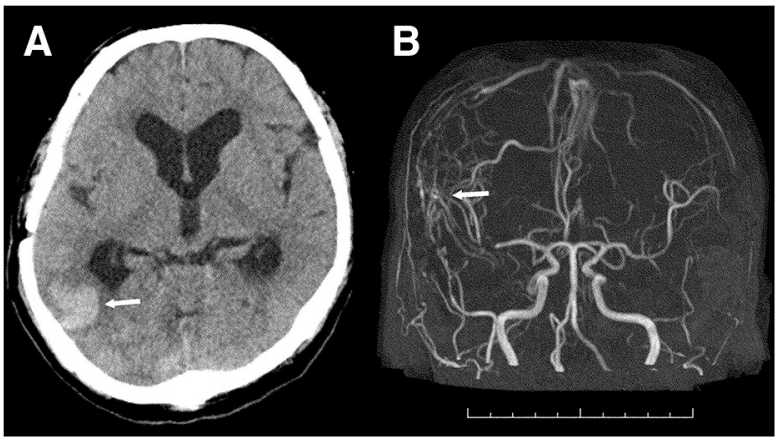

Fig. 1 (**A**) A CT at admission showing an intracerebral hemorrhage in the temporo-occipital cortex (arrow). (**B**) An MRA showing an anomalous vessel structure around the Sylvian fissure area (arrow).

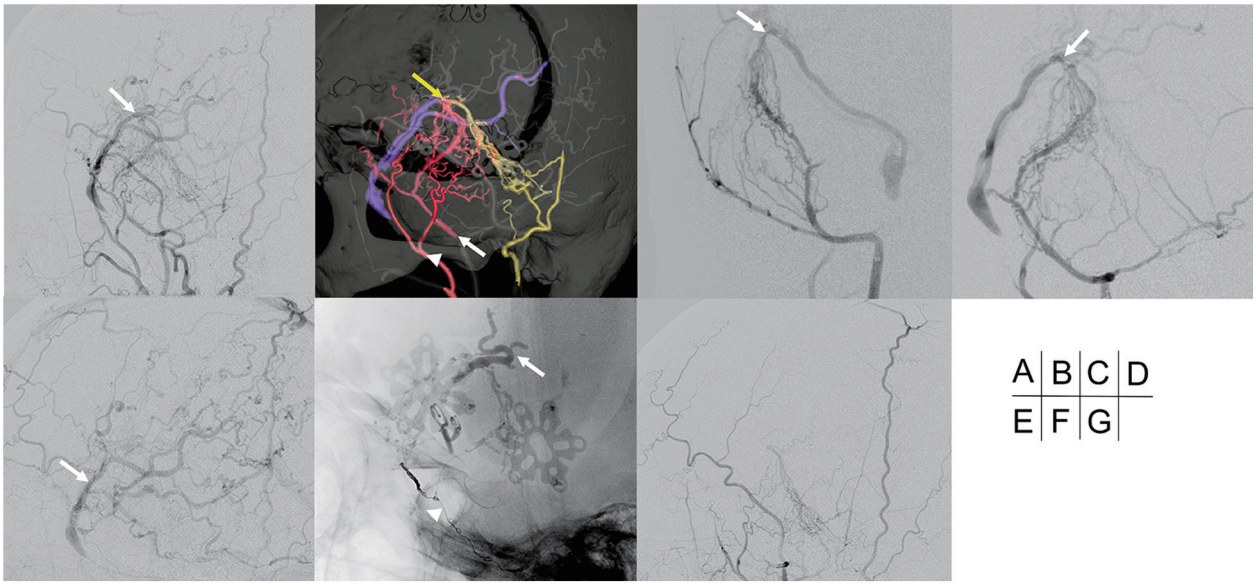

Fig. 2 (**A**) The right ECA angiogram (lateral view) showing an AV shunt (arrow). (**B**) A 3D DSA with a skull image demonstrating the feeding arteries, which are the deep temporal artery (red, arrowhead), the middle meningeal artery (MMA) (red, arrow), and the anterior auricular branch of the superficial temporal artery (yellow). These merge into the shunt point (yellow arrow) and drain into the SMCV. The deep temporal artery and the anterior auricular branch go from the subcutaneous layer into the intracranial space through the bone edge at the skull base side. (**C**) The right MMA selective angiogram (frontal view) showing an AV shunt (arrow). (**D**) The MMA angiogram (lateral view) showing an AV shunt (arrow). The collected feeders are shunting directly into the SMCV. (**E**) The late arterial phase of the right external carotid angiogram (lateral view) showing the arterial blood flow into the SMCV (arrow) and reflux into the cortical veins. (**F**) A fluoroscopy showing penetration of NBCA into the shunt point including the venous outlet (arrow). A microcatheter was placed in proximity to the shunt point through a loosely packed coil (arrowhead). (**G**) A right external carotid angiogram after the embolization demonstrating the complete disappearance of the dural artery–pial vein shunt. AV: arteriovenous; ECA: external carotid artery; MMA: middle meningeal artery; NBCA: n-butyl-2-cyanoacrylate; SMCV: superficial middle cerebral vein

the cortical veins into the basal vein of Rosenthal and the superficial cortical veins in the parietal area (**Fig. 2E**). The right internal carotid artery (ICA) angiogram showed that the venous outlet in the right ICA territory mainly flowed into the superior and inferior sagittal sinus instead of the SMCV. Moreover, the veins in the parietal and temporal lobes presented a pseudophlebitic appearance. The right ECA maximum intensity projection (MIP) coronal image showed that the collected feeders from the anterior auricular branch shunted directly into the SMCV (**Fig. 3A**–**3C**). A 3D DSA showed that the feeders from the anterior auricular branch and the MMA were flowing directly into different sites of the SMCV. These findings led to the diagnosis of a dural artery–pial vein shunt. We decided to perform endovascular treatment with n-butyl-2-cyanoacrylate (NBCA) first; if the NBCA were unsuccessful, we planned a switch to craniotomy to dissect the draining vein.

Embolization was performed under general anesthesia on the same day of the diagnostic angiography. Occlusion of the small feeders from the ADTA was initially planned

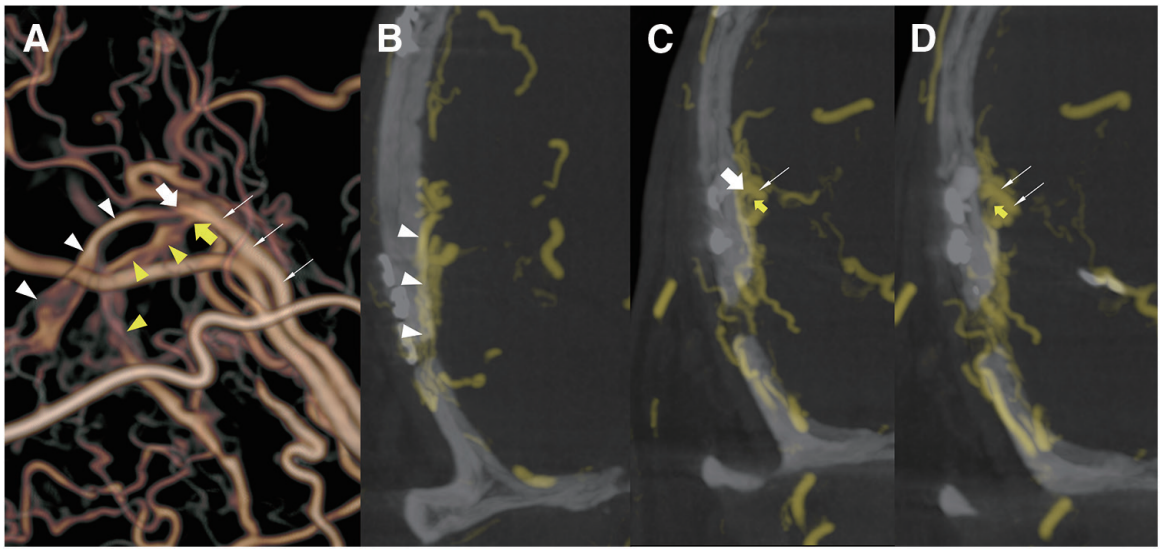

Fig. 3 **(A)** A 3D DSA showing the shunt feeders: the anterior auricular branch of the superficial temporal artery (white arrowheads) is flowing directly into the SMCV (thin white arrows), and the MMA (yellow arrowheads) is also flowing directly into the SMCV (thin white arrows). The white and yellow thick arrows indicate the respective shunt points. **(B)** The coronal image of the right ECA MIP showing feeders from the anterior auricular branch (white arrowheads). **(C)** The right ECA MIP coronal image showing the AV shunt point of the feeders from the anterior auricular branch (white arrow) and feeders from the MMA (yellow arrow) on the SMCV. **(D)** The right ECA MIP coronal image showing the SMCV (thin white arrows) and shunt point of the feeders from the MMA (yellow arrow) on the SMCV. AV: arteriovenous; ECA: external carotid artery; MIP: maximum intensity projection; MMA: middle meningeal artery; SMCV: superficial middle cerebral vein

to facilitate the penetration of NBCA into the venous outlet from the main feeder (MMA). A microcatheter (Magic 1.5 FM; BALT, Montmorency, France) was placed into the distal ADTA using a microguidewire (0.008 Chikai 200 cm; Asahi Intecc, Aichi, Japan) through a distal access catheter (4.2 Fr Fubuki catheter; Asahi Intecc) via a guiding catheter (6 Fr Fubuki 90 cm; Asahi Intecc) located in the right ECA. The distal branch of the ADTA was occluded using 0.2 mL of 33% NBCA. Then, a microcatheter (Magic 1.2 FM; BALT) was introduced into one of the two anterior branches of the MMA. A branch MMA was occluded using 33% NBCA. After that, a small coil (SMART Coil WAVE Extrasoft 1.5 mm × 3 cm; Penumbra, Alameda, CA, USA) was loosely placed proximally in another anterior branch of the MMA to prevent NBCA proximal reflux. A microcatheter (Magic 1.2 FM) was introduced in proximity to the shunt point through the loosely placed coil. The shunt point and venous outlets were occluded using 33% NBCA (**Fig. 2F**). The shunt completely disappeared on the final cerebral angiogram (**Fig. 2G**). The patient was discharged 1 month later with no neurological deficits.

Discussion

Five cases of iatrogenic AV shunt, in which arterial blood flow directly entered the pial vein, have been reported (**Table 1**).[1–5] The average age of the 6 patients, including the present patient, was 40.2 years. The previous surgeries were clipping for a cerebral aneurysm in three cases, superficial temporal artery–middle cerebral artery bypass for moyamoya disease in two, and lobectomy for intractable convulsions in one. Two patients presented with intracranial hemorrhage, whereas in the other four, the dural artery–pial vein shunt was found on follow-up neuroimaging studies. The interval between the initial surgery and the diagnosis of the AV shunt was 1 year or less, except for the hemorrhagic cases, with an interval of 20 years. As for the treatment in the cases previously reported, feeder occlusion was performed with surgery in two patients, including additional Gamma-Knife treatment in one[1,5] and endovascular feeder occlusion using absorbable gelatin sponge powder in one,[2] whereas conservative treatment was adopted in two cases,[3,4] in which the superficial temporal artery, previously used for bypass surgery, supplied the shunt. Three of the five patients previously described were diagnosed with AVFs,[1,3,4] one with mixed pial and dural AVF,[5] and one with dural AVF.[2] This case is the first report of iatrogenic dural artery–pial vein shunt. Selective angiogram and MIP images are important for the differential diagnosis between dural AVF and the dural artery–pial vein shunts. With regard to the endovascular treatment, the occlusion of the shunt point must fully extend to the venous

Table 1 Cases of iatrogenic AV shunts, in which arterial blood flow directly went into the pial vein

Study	Age/ sex	Previous surgery	Duration from surgery to onset	Symptom	Feeder	Treatment	Shunt outcome
Vadivelu et al.[2]	42/F	Clipping of ruptured MCA aneurysm	4 days	Asymptomatic	MMA	Embolization (absorbable gelatin sponge)	Disappearance
Feroze et al.[4]	51/F	STA–MCA bypass for moyamoya disease	6 months	Asymptomatic	STA (bypass graft)	Conservative	Remaining
Pabaney et al.[1]	62/M	Temporal lobectomy for epilepsy	20 years	SAH, IVH	MMA	Surgery	Disappearance
Doi et al.[5]	45/M	Clipping of unruptured MCA aneurysm	1 year	Asymptomatic	MMA	Surgery	Disappearance
Peeters et al.[3]	50/n.a.	STA–MCA bypass for moyamoya disease	1 year	Asymptomatic	MMA, MCA	Conservative	Remaining
Present case	64/M	Clipping of ruptured MCA aneurysm	1 year	ICH	MMA, ADTA, AAA	Embolization (NBCA)	Disappearance

AAA: anterior auricular artery; ADTA: anterior deep temporal artery; AV: arteriovenous; ICH: intracranial hemorrhage; IVH: intraventricular hemorrhage; MCA: middle cerebral artery; MMA: middle meningeal artery; NBCA: n-butyl-2-cyanoacrylate; SAH: subarachnoid hemorrhage; STA: superficial temporal artery

component for a complete cure. To the best of our knowledge, this report also describes the first iatrogenic dural artery–pial vein shunt treated with endovascular shunt occlusion using a liquid embolic material. In the present case, loose packing with a coil in the proximal feeder was helpful to prevent NBCA reflux into the proximal feeder and facilitate the penetration of the embolic material into the venous side.

No obvious mechanism for dural artery–pial vein shunt occurrence has been reported. In the previous case reports of iatrogenic dural AV shunt, two possible mechanisms have been suggested: one was a venous pressure change during the surgery, which could open latent AV channels in the dura mater,[6] and the other was the attachment of arteries in the muscles and/or the scalp to the dura mater after surgery facilitating the shunt occurrence.[7] In the present case, a 3D DSA with a skull image demonstrated that the feeding arteries penetrated the intracranial space through the basal side of the craniotomy edge. On the temporal base side, the dura mater was not incised during the previous surgery; therefore, the feeding arteries probably run on the surface of the dura mater toward the shunt point at the center of the previous craniotomy. At the shunt point, the blood flow from the feeding arteries entered the pial vein through the dura mater. The possibility of the opening of occult AV channels between the dura mater and the pial vein was eliminated, because no connection of the dura mater and the pial vein was observed during surgery. Therefore, the dural artery–pial vein connection must have been developed postoperatively. We did not use artificial materials during the craniotomy, including absorbable gelatin sponge, cellulose fiber material, or fibrin glue, which could have induced an inflammatory reaction, in the shunt area. Although inflammation induced by the subarachnoid hemorrhage might have been related to the presence of the dural artery–pial vein shunt, the exact mechanism of shunt formation remains unknown.

In this case, the localization of the shunt point was evaluated on an angiogram with a bone image, which could not depict the dura mater for itself. Then, whether the shunt point was located on a vein surface or the dura mater could not be differentiated clearly. A definitive diagnosis of dural artery–pial vein shunt based on current angiographic imaging studies was difficult to obtain, though at least the shunt point was found on the surface of the SMCV in this case. Therefore, further case accumulation of this disease and verification of shunt location should be needed.

Conclusion

We report a case of iatrogenic dural artery–pial vein shunt presenting intracerebral hemorrhage 1 year after aneurysmal clipping surgery. Iatrogenic shunts should be considered one

of the differential diagnoses in a case of intracranial hemorrhage after craniotomy. Endovascular embolization using liquid material is effective in treating these iatrogenic dural artery–pial vein shunts.

Disclosure Statement

The authors declare no conflicts of interest.

References

1) Pabaney AH, Robin AM, Basheer A, et al. Surgical management of dural arteriovenous fistula after craniotomy: case report and review of literature. *World Neurosurg* 2016; 89: 731.e7–731.e11.
2) Vadivelu S, Xin X, Loven T, et al. Iatrogenic dural arteriovenous fistula and aneurysmal subarachnoid hemorrhage. *Neurosurg Focus* 2012; 32: E1.
3) Peeters SM, Colby GP, Guivatchian E, et al. Spontaneous resolution of dural and pial arteriovenous fistulae arising after superficial temporal artery to middle cerebral artery bypass for moyamoya disease. *World Neurosurg* 2020; 142: 404–407.
4) Feroze AH, Kushkuley J, Choudhri O, et al. Development of arteriovenous fistula after revascularization bypass for Moyamoya disease: case report. *Neurosurgery* 2015; 11(Suppl 2): E202–E206.
5) Doi K, Otani N, Hayashi M, et al. Mixed pial and dural arteriovenous fistula after craniotomy: case report and literature review. *Br J Neurosurg* 2019 Oct 9. [Epub ahead of print]
6) Ugrinovski J, Vrcakovski M, Lozance K. Dural arteriovenous malformation secondary to meningioma removal. *Br J Neurosurg* 1989; 3: 603–607.
7) Nabors MW, Azzam CJ, Albanna FJ, et al. Delayed postoperative dural arteriovenous malformations. Report of two cases. *J Neurosurg* 1987; 66: 768–772.