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Ruptured brainstem arteriovenous malformation associated with a thalamoperforating artery aneurysm arising from the P1 segment of the right posterior cerebral artery: illustrative case

Yoshihito Hirata, MD,[1] Masakazu Okawa, MD, PhD,[1] Akira Ishii, MD, PhD,[1] Yu Abekura, MD, PhD,[1] Hisae Mori, MD, PhD,[2] Takayuki Kikuchi, MD, PhD,[1] Yukihiro Yamao, MD, PhD,[1] Susumu Miyamoto, MD, PhD,[1] and Yoshiki Arakawa, MD, PhD[1]

[1]Department of Neurosurgery, Faculty of Medicine, Kyoto University, Kyoto, Japan; and [2]Department of Neurosurgery, National Cerebral and Cardiovascular Center Hospital, Osaka, Japan

BACKGROUND Cerebral aneurysms of perforating arteries are rare and can be difficult to detect on computed tomography angiography (CTA) and digital subtraction angiography. Treatment is challenging and associated with a significant risk of morbidity. Endovascular treatment of a thalamoperforating artery (TPA) aneurysm within the midbrain has not previously been reported.

OBSERVATIONS A 13-year-old girl with no previous medical history presented with unconsciousness and anisocoria. Head computed tomography showed a right midbrain hemorrhage. CTA showed a midbrain arteriovenous malformation fed by a TPA aneurysm arising from the P1 segment of the right posterior cerebral artery. The feeder had a small distal aneurysm, which increased in size over time. Endovascular embolization was then performed.

LESSONS Cerebral aneurysms of perforating arteries are rare and can be difficult to treat. This is the first report of the endovascular treatment of a TPA aneurysm within the midbrain. Understanding the individual patient's brainstem perforator anatomy and the associated blood flow is essential before occluding a TPA aneurysm to avoid causing ischemia or infarction. Arteriovenous malformation embolization within the brainstem should be avoided because of interperforator anastomoses.

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KEYWORDS arteriovenous malformation; cerebral aneurysm; endovascular treatment

Cerebral aneurysms of perforating arteries are rare and can be difficult to detect on computed tomography angiography (CTA) and digital subtraction angiography. Treatment is challenging and associated with a significant risk of morbidity.[1] We report a patient with a brainstem arteriovenous malformation (AVM) associated with a ruptured thalamoperforating artery (TPA) aneurysm, which increased in size over time and was treated with endovascular feeder embolization using a coil and glue.

Illustrative Case

A 13-year-old girl with no previous medical history presented to our hospital with unconsciousness and anisocoria. Head computed tomography showed a right midbrain hemorrhage with extension into the ventricles and obstructive hydrocephalus (Fig. 1A). CTA revealed a midbrain AVM fed by a TPA aneurysm arising from the P1 segment of the right posterior cerebral artery (PCA; Fig. 1B). The distal portion of the TPA feeder was mildly dilated. This small aneurysm was considered the source of the hemorrhage (Fig. 1C–F). The AVM nidus was small and located within the right midbrain near the red nucleus. Venous drainage was into both basal veins (Spetzler-Martin grade III). The patient was treated with rapid placement of an external ventricular drain into the right lateral ventricle and aggressive control of hypertension. Although no rehemorrhage occurred, the distal feeder aneurysm increased in

ABBREVIATIONS AVM = arteriovenous malformation; BA = basilar artery; CTA = computed tomography angiography; PCA = posterior cerebral artery; PComA = posterior communicating artery; TPA = thalamoperforating artery.

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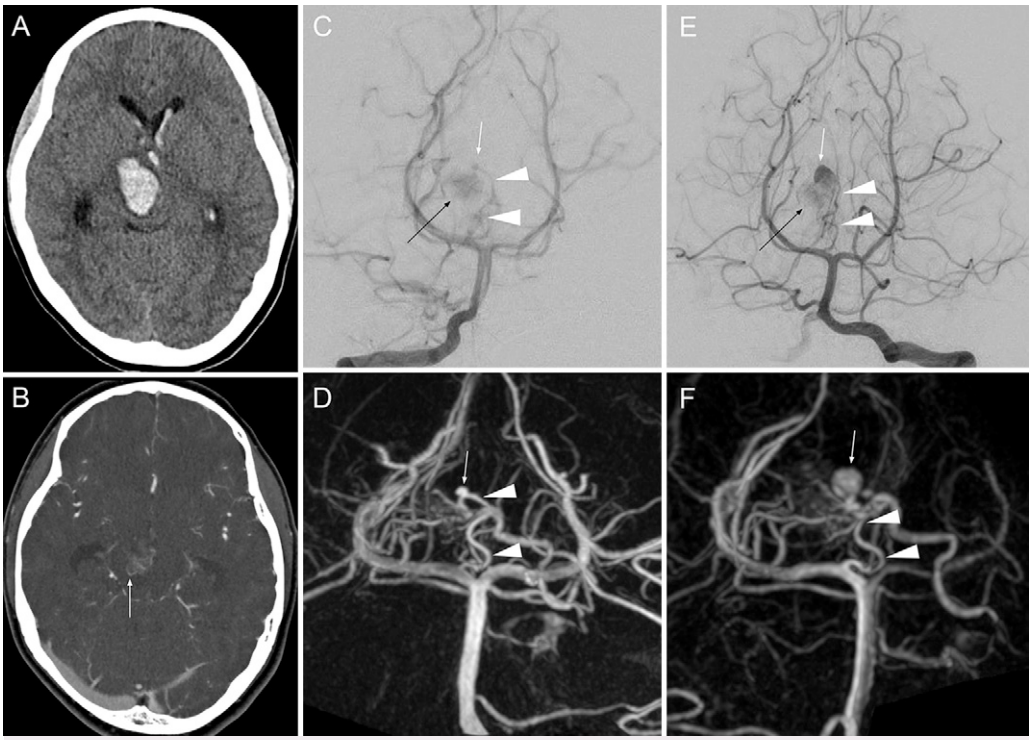

FIG. 1. A: Noncontrast head computed tomography (CT) shows midbrain hemorrhage with intraventricular extension. **B:** Contrast-enhanced head CT shows a suspected AVM nidus (*white arrow*). **C and D:** Angiographic studies on day 1 showed an AVM (*black arrow*) fed by a right TPA (*white arrowheads*) with a small distal aneurysm (*white arrows*). **E and F:** Studies on day 29 showed the AVM (*black arrow*), feeder (*white arrowheads*), and increased aneurysm size (*white arrows*).

size over time (6 mm on day 29). Endovascular embolization was performed on day 34.

After the induction of general anesthesia, a 6-Fr Roadmaster (Goodman) was placed in the left vertebral artery. A Guidepost 130 distal access catheter (Tokai Medical Products) was placed in the basilar artery (BA) through a Marathon microcatheter (Medtronic) and a Chikai 10 microguidewire (Asahi Intecc). We tried to navigate the microcatheter into the TPA, but a severe bend in the TPA just distal to its origin precluded the BA approach. Then, a 6-Fr Launcher (Medtronic) was placed in the petrous portion of the right internal carotid artery. A Guidepost was placed just proximal to the bifurcation of the right posterior communicating artery (PComA). A Marathon microcatheter with a Chikai 10 microguidewire was navigated into the TPA just proximal to the aneurysm through the PComA (Fig. 2A–D).

To prevent distal migration of the glue cast, the aneurysm was first coiled with a Barricade complex 3.0 mm $\times$ 4.0 cm coil (Blockade Medical; Fig. 2E). The second coil could not be inserted into the aneurysm. Then, 33% diluted *n*-butyl 2-cyanoacrylate 0.07 ml was applied proximal to the aneurysm (Fig. 2F). Left vertebral angiography after the procedure revealed the feeder occlusion (Fig. 2G).

Diffusion-weighted magnetic resonance imaging after the procedure showed no evidence of cerebral infarction (Fig. 2H). Stereotactic AVM radiosurgery was performed later. At the 6-month follow-up, the patient had recovered completely except for a mild right oculomotor palsy and mild hemiparesis.

Patient Informed Consent

The necessary patient informed consent was obtained in this study.

Discussion

Observations

Perforating artery aneurysms may arise from branches of the BA around the midbrain and anterior to the pons.[1–4] They are occasionally observed in patients with moyamoya disease, who develop collateral circulation via perforating branches; however, aneurysms within the brainstem have not previously been reported.[3] To the best of our knowledge, this is the first report of the endovascular treatment of a TPA aneurysm within the midbrain.

Several previous case reports and series have discussed the treatment of perforating artery aneurysms.[1–8] Many patients were treated conservatively, and, in most cases, the aneurysms resolved at between 6 weeks and 16 months of follow-up. Because the natural history of perforating artery aneurysms remains unclear, conservative treatment may be a feasible option. However, rebleeding can occur during the acute or subacute phase, which may result in permanent neurological deficit.[4] During the acute phase, our patient received conservative treatment because of the potential spontaneous resolution of the aneurysm and the risk of perforator infarction associated with endovascular intervention. Nevertheless, the aneurysm exhibited progressive enlargement over time. Given that the risk of

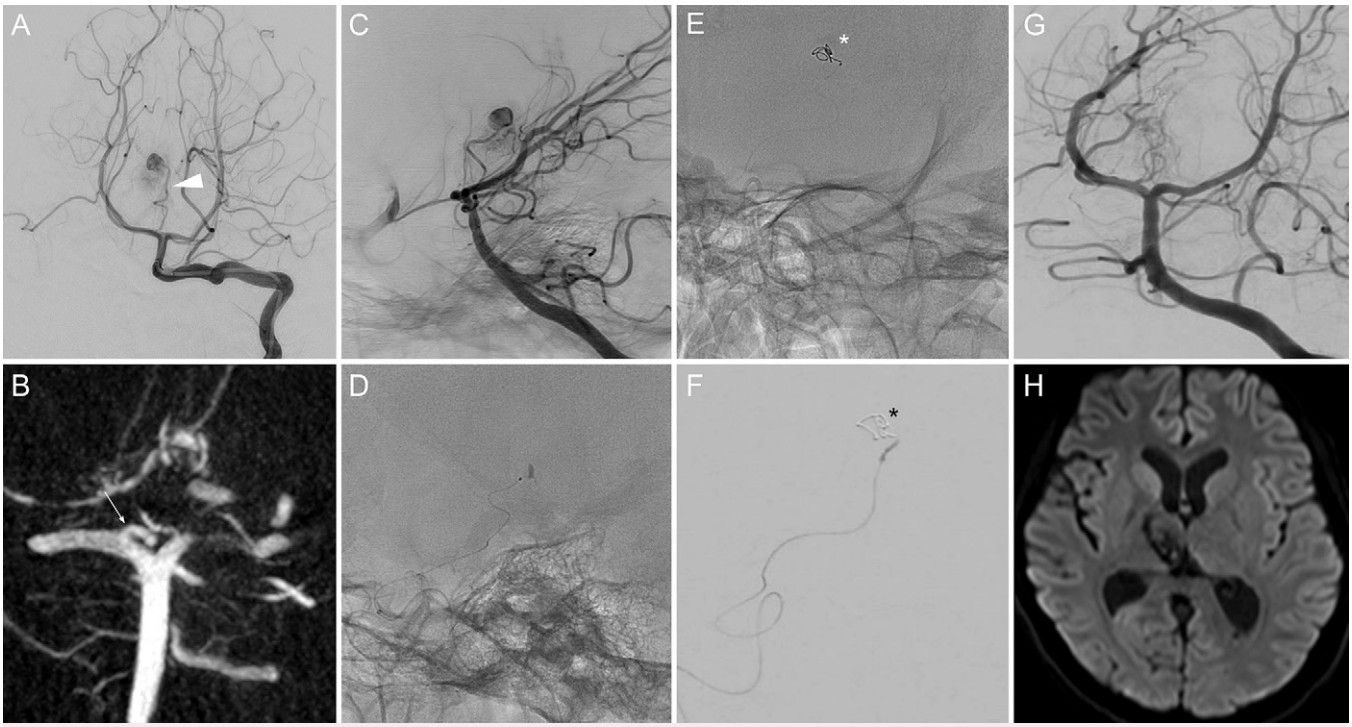

FIG. 2. A: Anteroposterior left vertebral angiography (VAG) during endovascular treatment shows the right TPA feeder (*white arrowhead*). **B:** Coronal CTA shows the severe medial bend in the feeding artery (*white arrow*) that precluded navigation of the microcatheter via the BA. **C:** Lateral view of VAG with carotid compression shows the right PComA. **D:** Lateral view of VAG shows the microcatheter just proximal to the aneurysm. **E and F:** Right oblique views of VAG show aneurysm coiling (*white asterisk*) and glue application (*black asterisk*). **G:** Right oblique view of left VAG after embolization shows disappearance of the aneurysm and persistence of the nidus. **H:** Diffusion-weighted imaging after the procedure shows no cerebral infarction.

rupture appeared to surpass the risk of treatment, we decided to perform endovascular therapy during the subacute phase.

Perforating artery aneurysms may not be detected on initial angiography. In almost half of cases, two or more angiographic studies are needed to detect the aneurysm.[1,2,4,6–8] When a perforating artery aneurysm is suspected and no aneurysm is detected, repeat angiography should be considered to prevent delayed diagnosis and treatment.

Treatment of TPA aneurysms is challenging because a perforator infarction can be fatal or neurologically devastating. TPA aneurysms most frequently arise from the P1 segment of the PCA, which is the segment located between the basilar bifurcation and the origin of the PComA.[9] The TPAs enter the posterior perforated substance between the cerebral peduncles and the mamillary bodies and supply the midbrain and diencephalon. The midbrain perforators have an average diameter of 255 μm and supply the central and paramedian midbrain.[9,10] They are very thin, located deep within the brain, and supply vital structures; therefore, aneurysms associated with them are extremely difficult to treat.

We considered several technical aspects when treating our patient. The first was whether occluding the parent TPA would result in infarction. Variations in the TPAs have been classified in several studies.[9,11–13] Lang and Brunner[11] described four types: type 1, multiple branches arising from each side; type 2, a single branch or couple of branches on one side and multiple branches on the other; type 3, a thick branch on each side; and type 4, one large stem on one side supplying both sides. The artery of Percheron is known as a single TPA trunk that supplies both sides of the paramedian thalamus and rostral midbrain.[14] Type 4 or Percheron-type TPAs should not be occluded, because bilateral infarction may result. In our patient, TPAs arose from both P1 segments, which suggested that the risk of bilateral infarction was relatively low.

Lee et al.[15] reported an anterior TPA aneurysm related to bilateral internal carotid artery occlusion, which was treated with endovascular glue embolization of the parent artery. The procedure was performed under local anesthesia to enable temporary occlusion testing. During temporary occlusion of the anterior TPA, the patient did not develop any neurological deterioration, and embolization using a detachable coil and 20% diluted glue was completed. We performed our procedure with the patient under general anesthesia owing to the patient's young age, so temporary occlusion testing was not performed. However, local anesthesia should be considered in most patients to enable temporary occlusion testing and minimize the risk of infarction. Neurophysiological monitoring can also be helpful to minimize the risk of infarction and should be considered in some cases. In our case, however, because the aneurysm was located in the brainstem, neurophysiological monitoring could not be performed.

The second aspect we considered was whether the glue material would migrate into other perforators. Approximately 79% of TPAs anastomose with other perforators arising from the BA, superior cerebellar artery, and PComA.[16,17] Therefore, the risk of distal glue migration into other perforators and consequent brainstem infarction was present. To prevent this, we coiled the aneurysm before glue embolization.

Third, we were unable to navigate the microcatheter to the aneurysm via the vertebral artery because of a severe medial bend at the TPA origin. Few previous studies have commented on the angle of the TPA at its origin. However, although the PCA runs around the brainstem in an outward direction, the TPA often courses inward to enter the posterior perforated substance.[9,10,13,16] Consequently, branching can exhibit a steep trajectory. Therefore, approaching the TPA from the posterior circulation can be challenging. Although the left PComA was not developed in our patient, catheter guidance via the right PComA was possible. This route can serve as an alternative for catheter navigation into P1 perforators (Video 1).

VIDEO 1. Clip showing intraoperative digital subtraction angiography. The microcatheter could not be navigated via the BA because of the severe medial bend of the thalamoperforating feeder artery at its origin. A microcatheter was guided via the right PComA to reach the aneurysm. The aneurysm was first coiled. Then, *n*-butyl 2-cyanoacrylate was applied proximal to the aneurysm. Anteroposterior view of left VAG after the procedure showed disappearance of the aneurysm. Click here to view.

Lessons

Perforating artery aneurysms are rare and can be difficult to treat. Those arising in the posterior circulation may require intervention, although spontaneous remission is possible. To the best of our knowledge, this is the first report of the endovascular treatment of a TPA aneurysm within the midbrain. Understanding the individual patient's brainstem perforator anatomy and the associated blood flow is essential before occluding a TPA to avoid causing ischemia or an infarction. AVM nidus embolization within the brainstem should be avoided because of interperforator anastomoses.

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Author Contributions

Conception and design: Okawa, Hirata, Ishii, Miyamoto. Acquisition of data: Okawa, Hirata. Analysis and interpretation of data: Hirata, Abekura. Drafting the article: Hirata, Kikuchi. Critically revising the article: Okawa, Kikuchi, Miyamoto. Reviewed submitted version of manuscript: Okawa, Mori, Kikuchi, Yamao, Miyamoto, Arakawa. Approved the final version of the manuscript on behalf of all authors: Okawa. Administrative/technical/material support: Okawa, Ishii. Study supervision: Miyamoto, Arakawa.

Supplemental Information

Videos

Video 1. https://vimeo.com/854375220.

Correspondence

Masakazu Okawa: Kyoto University, Kyoto, Japan. okawam@kuhp.kyoto-u.ac.jp.