

Short-segment Internal Trapping for Symptomatic Thrombosed Large Fusiform Vertebral Artery Aneurysms (Bird's Nest Trapping): A Technical Note

Masahiro NISHIHORI,¹ Takashi IZUMI,¹ Tetsuya TSUKADA,¹
Asuka Elisabeth KROPP,¹ Kenji UDA,¹ Kinya YOKOYAMA,¹
Yoshio ARAKI,¹ and Toshihiko WAKABAYASHI¹

¹*Department of Neurosurgery, Nagoya University Graduate School of Medicine,
Nagoya, Aichi, Japan*

Abstract

Internal trapping with coils is an established treatment of symptomatic large non-branching thrombosed fusiform vertebral artery aneurysms (VAA). However, when perforators arise near the aneurysm neck, parent artery occlusion has a high risk of causing medullary infarction. As an alternative treatment, we performed short-segment internal trapping of the artery using n-butyl-2-cyanoacrylate (NBCA) and coils (bird's nest trapping). Before treatment, perianeurysmal perforators are carefully detected using high-resolution three-dimensional rotational angiography (3DRA). Double microcatheters are advanced to the distal portion of the aneurysm through a balloon guiding catheter where coils are deployed without tight packing. Then, NBCA is injected into the coil mass, taking care to preserve perforators and significant branches. The same maneuver is repeated in the proximal portion of the aneurysm. Coil placement is avoided within the middle of the aneurysm; however, if necessary, only a small number of coils are placed to prevent worsening of mass effect. Two quinquagenarian males presented with a large thrombosed fusiform VAA that caused symptoms due to mass effect. In each case, perforators arose from the parent artery and short-segment internal trapping with NBCA and coils was performed. Symptoms improved after treatment and follow-up imaging confirmed aneurysm shrinkage with no long-time recurrence. In symptomatic large fusiform VAAs where the distance from the lesion to important perforators is extremely short, internal trapping using a combination of NBCA and coils can be more useful than conventional internal trapping.

Keywords: NBCA, internal trapping, vertebral artery aneurysm, endovascular treatment, thrombosed aneurysm

Introduction

Partially thrombosed aneurysms are often large and contain organized intraluminal thrombus that can grow gradually over time. Although treatment of asymptomatic cases remains controversial, symptomatic ones may require treatment. Large partially thrombosed aneurysms are one of the most difficult diseases to treat in neurosurgery, as their treatment

often requires techniques other than conventional clipping, such as thrombectomy with clip reconstruction or bypass with parent artery occlusion.^{1–3)} Due to the confined space of the posterior fossa, the surgical approaches to posterior fossa aneurysms require the knowledge and skills of skull base surgery, and operation time and surgical risks may increase.¹⁾ However, in thrombosed aneurysms with high surgical risk, endovascular treatment may not be generally indicated from the perspective of recurrence. Internal trapping with coils has been established as the treatment of choice for symptomatic non-branching thrombosed large fusiform vertebral artery aneurysms (VAAs).^{2–6)} Flow diverter stenting (FDS) is also an option, but this technique

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is associated with higher rates of ischemic complications, morbidity, and mortality compared to anterior circulation aneurysms.⁷⁾

Before high-resolution angiography was available and widely used, length of trapped segment and/or posterior inferior cerebellar artery (PICA) location were associated with medullary infarction.^{8,9)} However, in recent years, improvements in angiographic spatial resolution have made it possible to avoid unnecessary obstruction of medullary perforators. When perforators or arterial branches arise in the vicinity of the aneurysm, an alternative solution is required, as internal trapping may lead to infarction in the brain supplied by the perforators. This is because a conventional internal trapping with coil alone requires coil embolization over a sufficiently long distance to include a portion of the normal segment in addition to the abnormal segment. In addition, internal trapping for a short distance with a few coils alone might cause incomplete aneurysm occlusion when the distance between the aneurysm and perforators is extremely short. In contrast, if a large number of coils are placed in the abnormal segment to complete trapping, the mass effect may worsen. In this report, we describe an advanced internal trapping technique that can achieve short-segment internal trapping while maintaining patency of the perforators using n-butyl-2-cyanoacrylate (NBCA) and platinum coils. In Japan, the use of liquid embolic NBCA for endovascular treatment is considered off-label but can be used with written informed consent; ONYX (Covidien, Irvine, CA, USA) can be used for arteriovenous malformations and dural arteriovenous fistulas but not for aneurysms. We present two patients with a symptomatic large thrombosed VAA who were treated using this new technique.

Methods

The identification of perianeurysmal perforators is essential prior to trapping. Modern angiography can now reveal perforators that we have never assessed before. Currently, we use bi-planar angiography that incorporates three-dimensional rotational angiography (3DRA) and multiplanar reconstruction (MPR) imaging to interpret vessel anatomy in detail. Both 3D-volume rendering (VR) images and MPR images are carefully evaluated to determine the presence of minute vessels. In contrast to computed tomography angiography (CTA) and magnetic resonance imaging (MRI), this method can show the positional relationship between nearby perforators and the thrombosed portion of the aneurysm.

Concept and aim of the bird's nest trapping

The aim of treatment of thrombosed aneurysms is as follows: (1) reduction of mass effect, (2) suppression of aneurysm growth, and (3) prevention of rupture. The schema of a large thrombosed aneurysm and peripheral branches is shown in Fig. 1A. If a sufficient number of coils to achieve complete occlusion were used, mass effect from the treated aneurysm would be exacerbated (Fig. 1B). On the other hand, if only a small number of coils were used, incomplete occlusion would result and almost all treatment aims would not be achieved (Fig. 1C). To preserve the patency of perforators and branches in the distal and proximal neck respectively, complete trapping with a few coils alone would be expected to be difficult. Therefore, as shown in Fig. 1D, we hypothesized that NBCA and coils in combination could achieve complete trapping in a short distance while avoiding ischemia of the perforators and minimizing mass effect from treatment. In addition, if craniotomy and aneurysm resection were required in the future due to aneurysm regrowth or recurrence, a smaller coil mass is easier to manipulate and resect. The concept of this technique is similar to how a bird's nest is comprised of both liquids and solids, hence the name "bird's nest trapping." For postoperative management, we elected to administer anticoagulant therapy longer than usual to prevent thromboembolic complications associated with NBCA.

Technical details of "bird's nest trapping"

A balloon guiding catheter is mandatory to perform flow control. After a diagnostic catheter is advanced into the contralateral VA, proximal flow control to prevent migration or scattering of glue and thrombus is achieved by inflating the guiding balloon. Next, systemic heparin is administered and double microcatheters are advanced to the distal portion of the aneurysm. Under proximal flow control, a few platinum coils are deployed while paying careful attention to the patency of nearby perforators. Then, distal contrast dye flow is evaluated by micro digital subtraction angiography (DSA). If the contrast agent flows distally, the tip of the microcatheter is moved to the proximal side and micro-DSA is repeated to finely adjust the microcatheter tip to a position approximately one-third distal to the center of the coil mass. It is important to restrict the number of coils to the extent that glue behavior remains visible. After adjusting the position of the microcatheter under flow arrest, diluted NBCA (25–33% concentration) is slowly injected into the coil mass with several short pauses to spread the glue throughout the parent artery. After the NBCA injection is completed, the microcatheter is withdrawn and

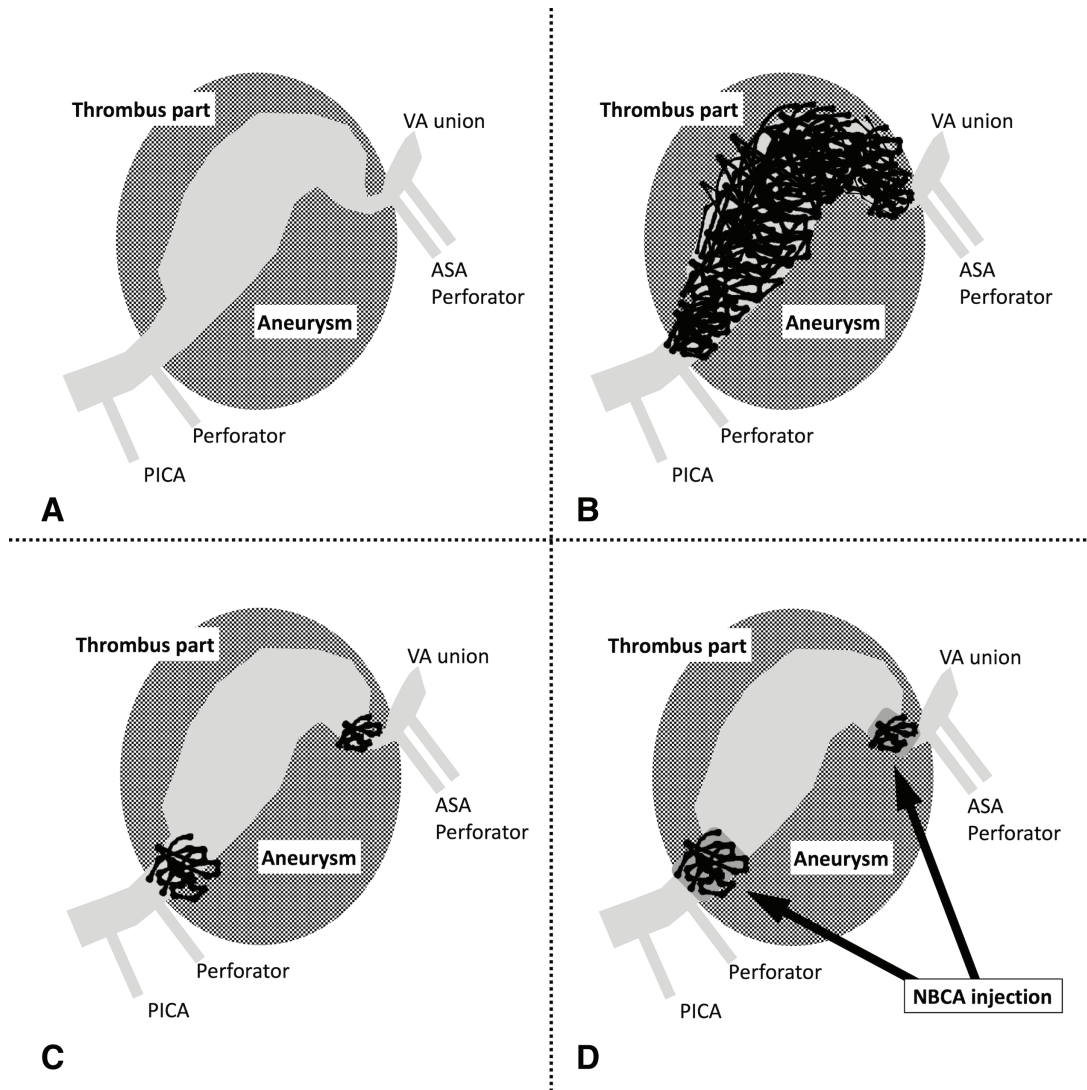


Fig. 1 A schema showing the concept of “bird's nest trapping” and other methods. (A) A thrombosed aneurysm schema with an extremely short distance between the aneurysm and the origin of the perforators is shown. (B) If coiling alone is used for tight packing, aiming for complete occlusion, the mass effect may worsen. (C) A high possibility of incomplete occlusion and recurrence may result when only a small number of coils are used. (D) A schema of “bird's nest trapping” is shown. This method prevents worsening mass effect by reducing the number of coils in the aneurysm while allowing complete occlusion by adding NBCA to a small number of coils. NBCA: n-butyl-2-cyanoacrylate.

contralateral vertebral angiography (VAG) is performed to confirm complete occlusion of the distal portion. For the proximal portion, the same procedure is repeated while preserving the perforators.

Results

We treated two patients with a symptomatic large thrombosed fusiform VAA using this technique. In each patient, the distance between the perforators and aneurysm was extremely short, necessitating usage of this technique. After treatment, the

aneurysms shrank without significant worsening of mass effect and symptoms improved. No major complications occurred. Both patients were discharged with a modified Rankin Scale (mRS) score of 1, and follow-up imaging confirmed aneurysm shrinkage without recurrence for over 5 years.

Case 1 is a 53-year-old man with no significant past medical history who presented with Wallenberg syndrome and a giant right fusiform VAA. On CTA, the aneurysm was partially thrombosed with a diameter of approximately 34 mm (Fig. 2A). T2-weighted imaging (T2WI) showed a mass lesion compressing

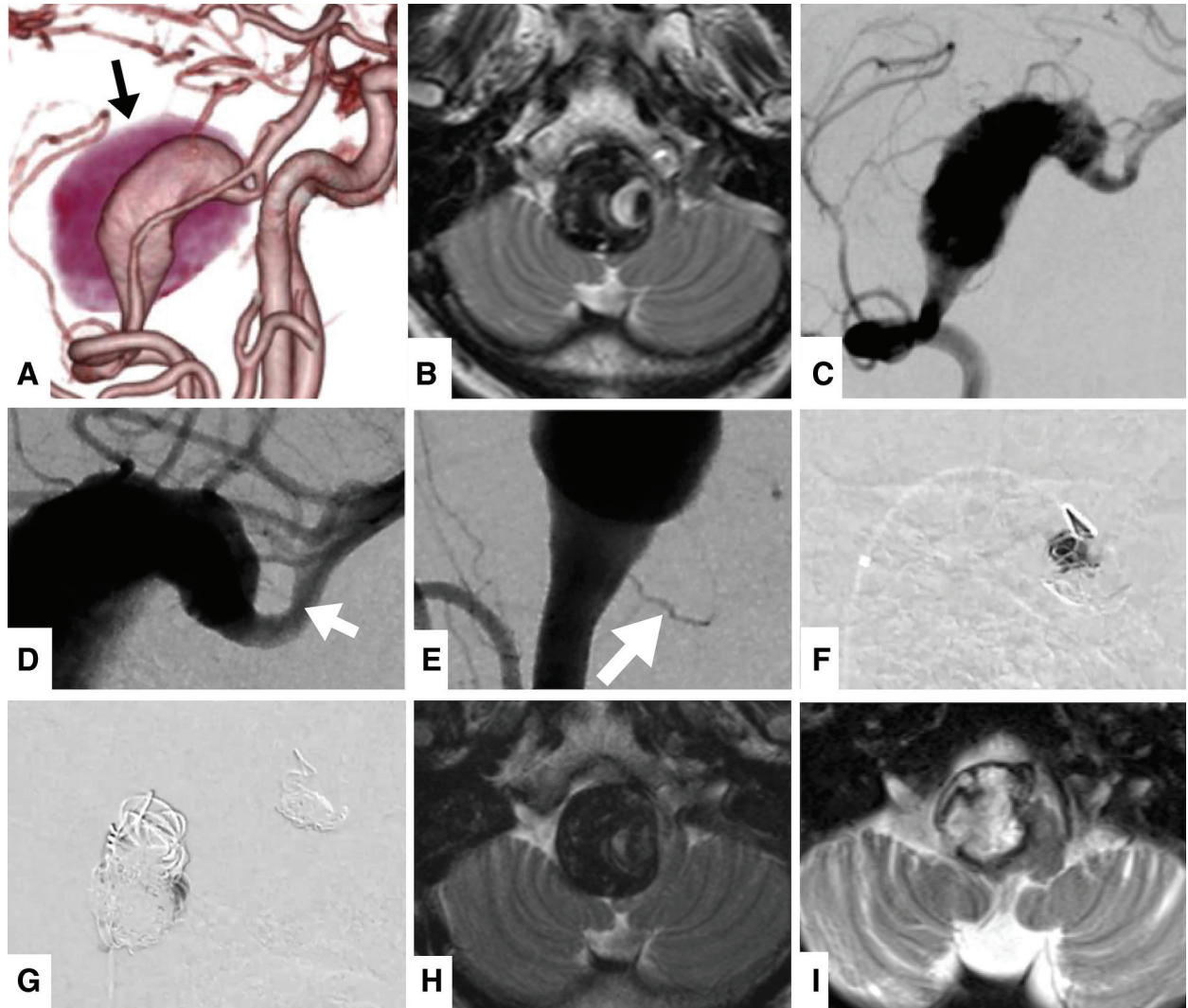


Fig. 2 Preoperative, treatment, and postoperative images of Case 1. (A) VR CTA imaging. The black arrow indicates the thrombosed portion of the aneurysm. (B) T2WI shows a 34 mm × 33 mm aneurysm compressing the medulla oblongata. (C) Lateral view of DSA. (D) Magnified DSA view of the distal aneurysm. The white arrow indicates perianeurysmal perforators. The distance from their origin to the aneurysm is approximately 2 mm. (E) Magnified DSA view of the proximal aneurysm. The white arrow indicates a perforator feeding the medulla oblongata. (F) DSA imaging showing NBCA injection after several coils were placed in the distal aneurysm. NBCA extends widely throughout the blood vessels. (G) DSA imaging showing NBCA injection after several coils were placed in the proximal aneurysm. (H) T2WI immediately after aneurysm trapping showed no obvious signal change around the aneurysm. (I) T2WI 6 years after treatment showed aneurysm shrinkage (22 mm × 24 mm). CTA: computed tomography, DSA: digital subtraction angiography, NBCA: n-butyl-2-cyanoacrylate, T2WI: T2-weighted imaging, VR: volume rendering.

the medulla (Fig. 2B). DSA and 3DRA remodeling imaging showed the anterior spinal artery (ASA) arising from the distal portion of the aneurysm as well as several perforators arising from the distal and proximal portions (Figs. 2C, 2D, and 2E). The distance between the ASA and thrombosed aneurysm was very short, approximately 2 mm, which precluded trapping with coils alone. Direct clipping or trapping was considered very difficult to perform due to the

size of the aneurysm. Therefore, we elected to perform short-segment internal trapping using the “bird’s nest trapping” described above.

A 7-F balloon guiding catheter was placed in the ipsilateral VA and complete occlusion of both ends of the parent artery was achieved using coils and NBCA under proximal flow control as described above (Figs. 2F and 2G). Adequate patency of the perforators and ASA was preserved. Heparin was

continuously administered for 24 hours after the procedure to prevent excessive thrombus formation. Although the patient's symptoms temporarily mildly deteriorated after the procedure, no new ischemic lesions, worsening of perianeurysmal edema or aneurysm enlargement were found (Fig. 2H). Symptoms gradually improved and the patient is now independent. Follow-up MRI has shown aneurysm shrinkage (Fig. 2I).

Case 2 is a 51-year-old man who presented with subacute paraparesis of the lower limb and a right VAA (diameter, approximately 18 mm). CTA showed partial thrombosis of the aneurysm. T2WI showed medullary compression (Figs. 3A, 3B, and 3C). DSA and 3DRA remodeling imaging demonstrated only one medullary perforator arising from the proximal neck (Figs. 3D and 3E) that needed to be preserved. Figure 3F is the schema showing the positional relationship between the thrombosed aneurysm and perianeurysmal perforator. We elected to treat the aneurysm using the "bird's nest trapping." A 7-F balloon guiding catheter was placed and double catheters were navigated to the distal portion of the aneurysm. Under proximal flow control, two coils were deployed into the stenotic parent artery and two into the aneurysm (Fig. 3G). Once contrast dye flow was confirmed by micro-DSA, we slowly injected NBCA (25% dilution) into the coil mass (Fig. 3H). Paraparesis gradually improved over 2 weeks and the patient was able to walk independently again. Follow-up MRI has shown aneurysm shrinkage (Fig. 3I).

Discussion

In the two cases presented here, we treated a VAA with short-segment internal trapping using NBCA and platinum coils, which preserved patency of medullary perforators and achieved good clinical outcomes. Both aneurysms shrank in size and both patients experienced improvement in symptoms, showing that trapping of symptomatic large VAAs is safe and feasible. Safe aneurysm occlusion depends on preservation of the perforators and nearby branches. Although the "bird's nest trapping" described here requires slightly advanced skills, it can preserve perforator patency without worsening mass effect and achieve short-segment internal trapping, similar to trapping using surgical clips. Although endovascular trapping using a combination of coils and NBCA or ONYX has been previously reported for various cerebrovascular aneurysms,^{10–15} the techniques used differ from ours. In the previous cases, a liquid material was used in combination to improve the occlusion rate of large, pseudo-, or distal cerebral aneurysms, if simple coil

embolization was insufficient to achieve adequate occlusion and could lead to recurrence and bleeding. The novelty of this method is that we were able to complete internal trapping within a relatively short distance and to inject NBCA safely under flow control using a balloon guiding catheter while preventing migration of glue into the perforators.

Essential components of the "bird's nest trapping" are use of a balloon guiding catheter and the slow infusion of diluted NBCA throughout the entire vessel. If the VA has a small diameter that does not allow placement of the balloon guiding catheter, the technique may be abandoned and trapping performed with platinum coils alone. In addition, visibility of the NBCA infusion must be considered early during coil deployment; if too many coils are deployed, it will be impossible to confirm adequate spread of NBCA along the short axis of the vessel. Therefore, it is surprisingly important to only use a few coils. Moreover, to maintain the NBCA around the coil mass and permit adequate visibility during infusion, we recommend a 25–33% dilution of NBCA. As regards procedure-related risks, there may be difficulty with microcatheter removal and following coil migration. It is presumed that long-time injection of a high concentration diluted NBCA may lead to difficulties in decannulation. However, because a low-concentration diluted NBCA was administered for a short time, that is, up to a total of 30 seconds, removal of the microcatheters was smooth in both cases.

This technique can be difficult to apply safely, particularly in cases where visualization of the perforators is insufficient because of the angiography resolution or the vascular architecture itself. Identifying the perianeurysmal perforators is crucial for applying the "bird's nest trapping"; if not correctly identified, the liquid embolization material can easily migrate into them. Furthermore, if the PICA arises from the aneurysm itself, trapping should not be performed and hybrid surgery with bypass considered. Considering the difficulty of the procedure and potential ischemic complications from trapping, we believe that the "bird's nest trapping" should only be indicated for symptomatic fusiform VAAs. For asymptomatic large fusiform VAAs, stent-assisted coiling (SAC) or FDS preserving blood flow is the treatment of choice, as reported before.¹⁶ Based on an analysis of 18 patients with large thrombosed fusiform VAA at our institution, ischemic complications occurred in three out of six patients (50%) treated with internal trapping, as compared with 2 out of 12 (17%) treated with SAC. In patients with asymptomatic aneurysms, there were no recurrences after SAC.

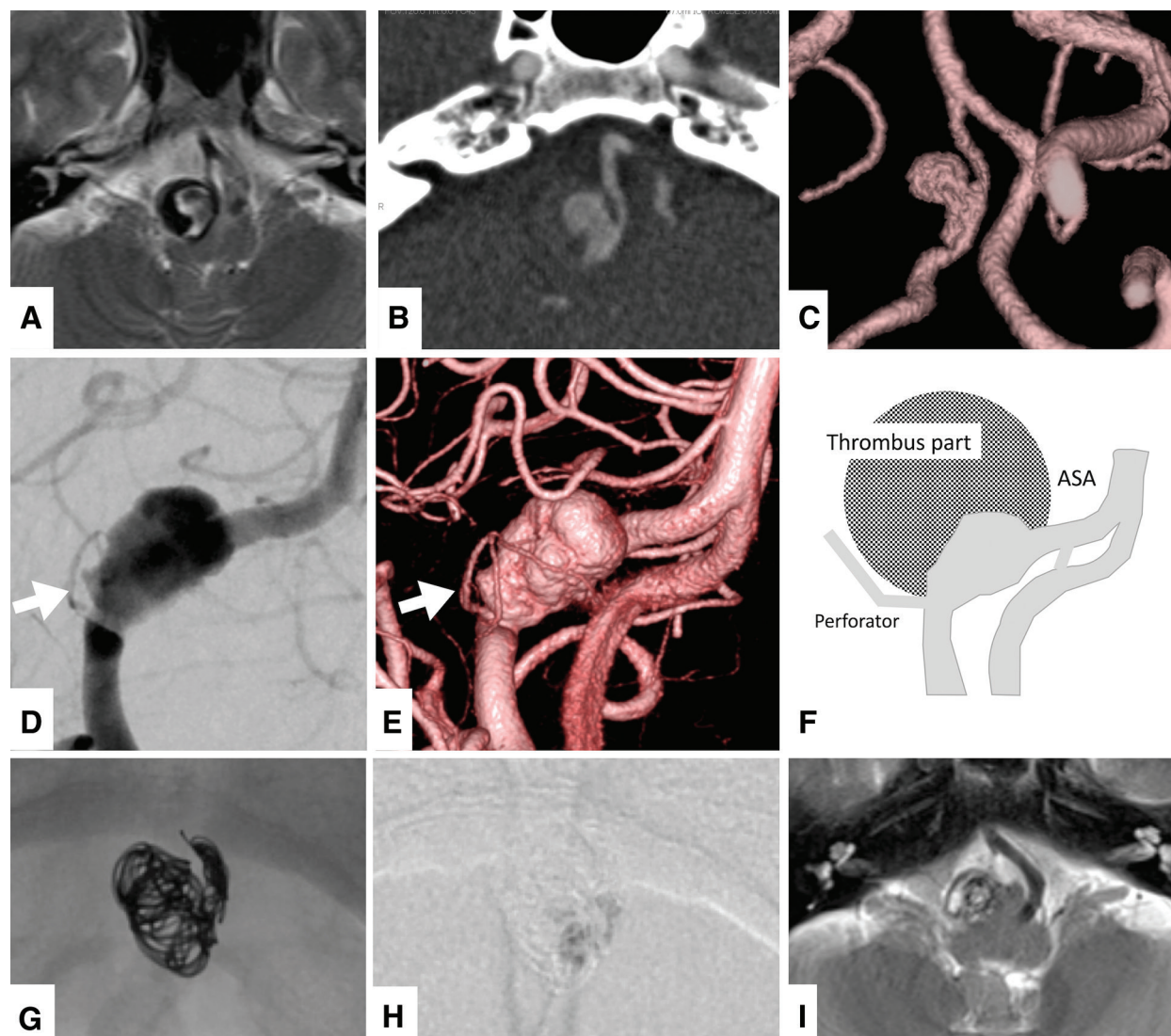


Fig. 3 Preoperative, treatment, and postoperative images of Case 2. (A) T2WI showed medullary compression from a 18 mm × 16 mm aneurysm and medially located contralateral vertebral artery. (B) Axial CTA imaging showed partial thrombosis of the aneurysm. (C) VR CTA. (D) Oblique view of digital subtraction angiography and (E) 3D-rotational angiography VR imaging at the same angle. A perforator feeding the medulla oblongata arises from the proximal aneurysm (white arrow). (F) Schema showing the positional relationship between the thrombosed aneurysm and perianeurysmal perforator. (G) Occlusion was easily achieved with two coils deployed into the stenotic distal parent artery and two coils into the aneurysm. (H) NBCA was injected into the proximal end of the coil mass to complete trapping. (I) T2WI 5 years after treatment showed aneurysm shrinkage (13 mm × 10 mm). 3D: three-dimensional, CTA: computed tomography angiography, NBCA: N-butyl-2-cyanoacrylate, T2WI: T2-weighted imaging, VR: volume rendering.

The symptoms in our two cases were Wallenberg syndrome in the first and lower limb paraparesis in the second. Although Wallenberg syndrome occurring due to VA pathology is straightforward, paraparesis is not. Possible cerebrovascular causes of paraparesis include ruptured anterior communicating or anterior cerebral artery aneurysms. Paraparesis has also been reported in a few cases of ruptured VA-PICA aneurysm,¹⁷⁾ but not in association with unruptured

posterior circulation aneurysms. In ruptured VA-PICA aneurysms, the cause of paraplegia from an anatomical point of view is damage of the superficial descending pyramidal tract fibers. The fibers innervating the lower limbs are located in the outer surface layer and are more susceptible to injury from extramedullary compression, whereas the upper limb fibers are located deeper. In our second case, we speculated that the medulla was being compressed

from both sides, by the VAA and the medially located contralateral VA. The most pertinent factor in the development of neurological disorders in patients with giant posterior circulation aneurysms is aneurysm volume, not extent of lateral protrusion or degree of brainstem compression.^{18,19)} Although brainstem compression is rare, it was clearly causing symptoms in our second case, as the paraplegia improved after treatment. For symptomatic aneurysms, internal trapping with large amounts of coils as in Fig. 1B is controversial. In an aneurysm with a relatively large thrombosed part, it is unlikely for the mass effect to be worsened by filling with a large number of coils. Conversely, in treating the symptomatic aneurysm with a small or no thrombosed part, the mass effect is likely to worsen. According to a multivariate analysis of multimodality treatment for unruptured internal carotid artery aneurysms that presented with visual field deficits caused by mass effects over the anterior optic pathway, only endovascular coiling significantly prevented improvement in the visual field compared with other treatments such as bypass and/or trapping.²⁰⁾ These results clearly indicate that embolization with large numbers of coils in symptomatic large aneurysms exacerbates the mass effect on the surrounding nerves. As per the merit of this “bird’s nest trapping,” in addition to the advantage of avoiding the worsening mass effect, resection is more likely to be easy when there is recurrence and regrowth after internal trapping.

Iihara et al. previously reported excellent results after multimodality treatment of partially thrombosed large or giant posterior circulation aneurysms; they also presented a management strategy and mentioned that endovascular trapping may be effective for aneurysms at non-branching sites in conclusion. Because of the comparative rarity of large posterior circulation thrombosed aneurysms, this strategy has been very useful until now. In this series, all six cases of non-branching VA aneurysms were treated using parent artery occlusion. Five cases underwent endovascular trapping and one case (a non-branching large VAA) underwent surgical occlusion.²⁾ Although there may have been selection bias, improvement in mRS score occurred in almost all cases and moderate shrinkage occurred in two of the four cases for which imaging follow-up was available. More recently, flow diverter stenting has been used as an alternative treatment option; however, this technique is controversial. In a 2018 meta-analysis, Kiyofuji et al. analyzed 131 aneurysms and reported outcomes of posterior circulation non-saccular aneurysms treated with FDS.⁷⁾ VAAs had a better outcome than other locations, although larger aneurysms showed

a considerably higher rate of ischemic complications compared to anterior circulation aneurysms. Overall morbidity and mortality were 26% and 21%, respectively. Most of the morbidity was described to be associated with thromboembolic complications. An updated subgroup analysis of the results of flow re-direction endoluminal device treatment of posterior circulation aneurysms has recently been reported. The most common aneurysm type was saccular and favorable safety results were shown, but thromboembolic complications were more frequent in non-saccular aneurysms.²¹⁾ The therapeutic results of mid/distal-basilar and holobasilar aneurysm involving AICA showed the worst neurological outcome.²²⁾ Conversely, if holobasilar aneurysms or aneurysms with a large amount of thrombus were not involved, the treatment outcome was reported to be excellent.²³⁾ In light of these findings, the two present cases are not good indications for FDS. Thus, it is important to identify significant treatment risk factors and optimize patient selection. Although only two cases have been studied in this report, symptomatic thrombosed fusiform vertebral aneurysm is a rare disease, and there is a need to share our experience in treating such patients.

Conclusions

In patients with symptomatic thrombosed non-branching large VAAs, if the distance from the aneurysm to the perforators is extremely short, internal trapping using a combination of coils and NBCA is a treatment option and can be more useful than conventional internal trapping. Continued investigation and analysis of further cases and long-term follow-up data are needed.

Conflicts of Interest Disclosure

The authors have no conflict of interest to disclose.

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Corresponding author: Masahiro Nishihori, MD
 Department of Neurosurgery, Nagoya University of
 Graduate School of Medicine, 65 Tsurumaicho,
 Showa-ku, Nagoya, Aichi 466-8550, Japan.
e-mail: nishihori@med.nagoya-u.ac.jp