



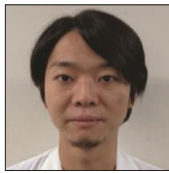
Case Report

Acute surgery for a case of superior vermian arteriovenous malformation producing raised venous pressure coexisting with basilar-superior cerebellar artery aneurysm presenting subarachnoid hemorrhage; Case report

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ABSTRACT

Background: Superior vermian subtype of arteriovenous malformation (AVM) coexisting with proximal feeder aneurysm on basilar-superior cerebellar artery (BA-SCA) junction is an extremely rare situation. We experienced a case of this rare entity presenting with subarachnoid hemorrhage (SAH), and herein, introduce the outline and clinical features of this experience together with the actual surgical video.

Case Description: A 54-year-old man SAH patient with severe headache, disturbance of consciousness, and left oculomotor palsy was urgently admitted to our hospital. Imaging examination demonstrated superior vermian AVM with BA-SCA aneurysm, and both lesions were treated through two different approaches (left pterional craniotomy in conjunction with zygomectomy, and left posterior interhemispheric occipital transtentorial approach) in acute phase of SAH. Both lesions were completely disappeared postoperatively and the patient's postoperative course was favorable, without symptomatic cerebral vasospasm. Although slight oculomotor palsy remained, the patient recovered well and was transferred to a rehabilitation hospital for further improvement.

Conclusion: In the cases of AVM coexisting with proximal feeder aneurysm, presenting with SAH, disorders of intracranial venous return associated with an AVM can be a vital hindrance to managing cerebral vasospasm; therefore, treating both lesions in the acute phase may lead to good outcomes.

Keywords: Arteriovenous malformation, Basilar artery aneurysm, Clipping, Occipital transtentorial approach, Subarachnoid hemorrhage

INTRODUCTION

Cerebellar arteriovenous malformation (AVM) has a relatively higher rupture rate than supratentorial AVM, and cerebellar AVM is difficult to treat because of its location surrounded by vital anatomical structures.^[5,9] Among the types of cerebellar AVM, the superior vermian subtype accounts for only 3%^[5,15] of all cases, and cerebellar AVM coexisting with flow-related or intranidal aneurysm accounts for 20% of all cerebellar AVM^[15] in other words, superior vermian AVM coexisting with a feeder aneurysm is extremely rare.

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In this study, we experienced an excellent outcome in a case of superior vermian AVM coexisting with a basilar-superior cerebellar aneurysm (BA-SCA) presenting with subarachnoid hemorrhage (SAH). In this case, we successfully performed radical two-stage operation for both lesions in the acute phase. In this report, we introduce the outline and clinical features of this experience, and present our methodology for completing safe surgery together with the actual surgical video.

CLINICAL PRESENTATION

A 54-year-old man presenting with abrupt severe headache, disturbance of consciousness with Glasgow coma scale (GCS) of 12 (E3V4M5), and left oculomotor palsy (ptosis of 3 mm, moderate exotropia, and anisocoria of 2 mm) was urgently transported and admitted to our hospital.

Computed tomography (CT) revealed diffuse SAH, particularly thickened at the right ambient to quadrigeminal cisterns [Figure 1], and subsequent digital subtraction angiography (DSA) revealed an AVM located at the superior vermis of the cerebellum, with a left BA-SCA aneurysm [Figures 2 and 3].

The AVM was mainly fed by two branches from the ipsilateral SCA in addition to the contralateral SCA and ipsilateral posterior inferior cerebellar artery (PICA) [Figure 2a and b]. These vessels drained through the superior vermian vein into the vein of Galen, and then the venous flow entered the confluence of the sinuses through the

straight sinus, which was slightly refluxed to the superior sagittal sinus [Figure 2c]. No other noticeable feeders and drainers were observed. The maximum diameter of the AVM was 28 mm, and the anterior-superior surface of the nidus was exposed in the precerebellar and quadrigeminal cisterns, which were widened by the dilated and tortuous feeding arteries associated with the AVM. We diagnosed a superior vermian AVM with Spetzler–Martin Grade II.

The aneurysm was located at the origin of the SCA, the main feeder of the AVM, and the dome and neck width were 10.5 mm and 4.1 mm, respectively. The aneurysmal neck and apex of the dome were located 5.0 mm and 12.1 mm above the clinoid line (connecting the anterior and posterior clinoid process), respectively [Figure 2d].

We diagnosed SAH, World Federation of Neurological Surgeons Grading System for Subarachnoid Hemorrhage (WFNS) Grade 3. Imaging findings made it difficult to determine whether the aneurysm or AVM was the rupture site; however, the absence of intracranial hemorrhage (ICH) or intraventricular hemorrhage (IVH) and the presence of left oculomotor palsy suggested the possibility of aneurysmal rupture more than AVM rupture, and we prioritized treatment using aneurysmal clipping.

Considering the position of the neck and apex of the dome, we added zygomectomy in addition to standard frontotemporal craniotomy [Figure 3]. The sphenoid ridge and greater wing were shaved, and the superior-lateral wall of the orbit was flattened to the depth of the meningo-orbital

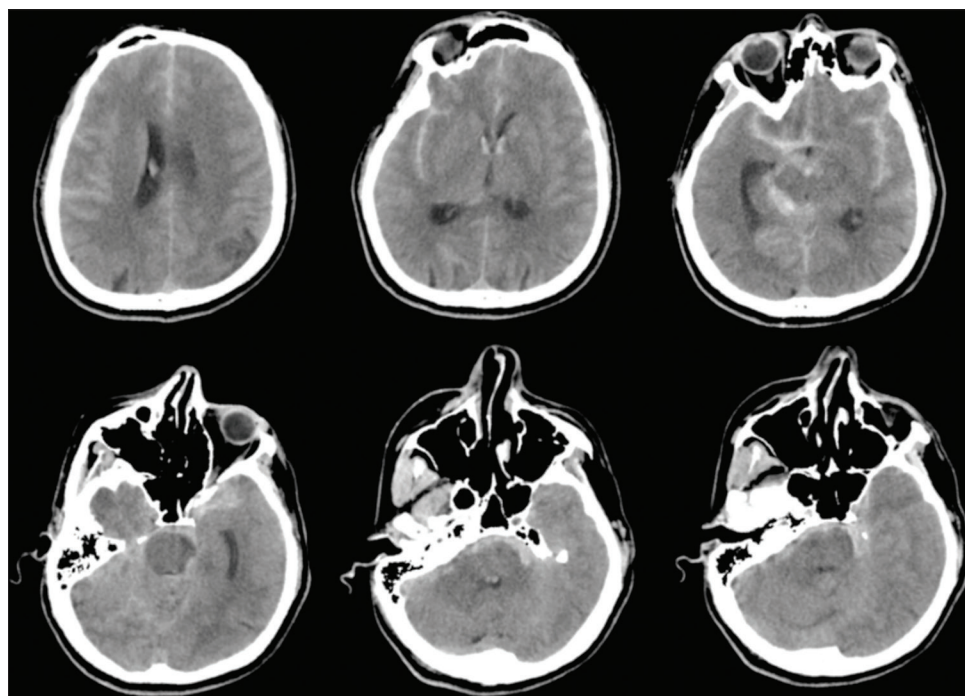


Figure 1: Initial CT on admission revealed diffuse SAH, particularly thickened at the right ambient to quadrigeminal cisterns.

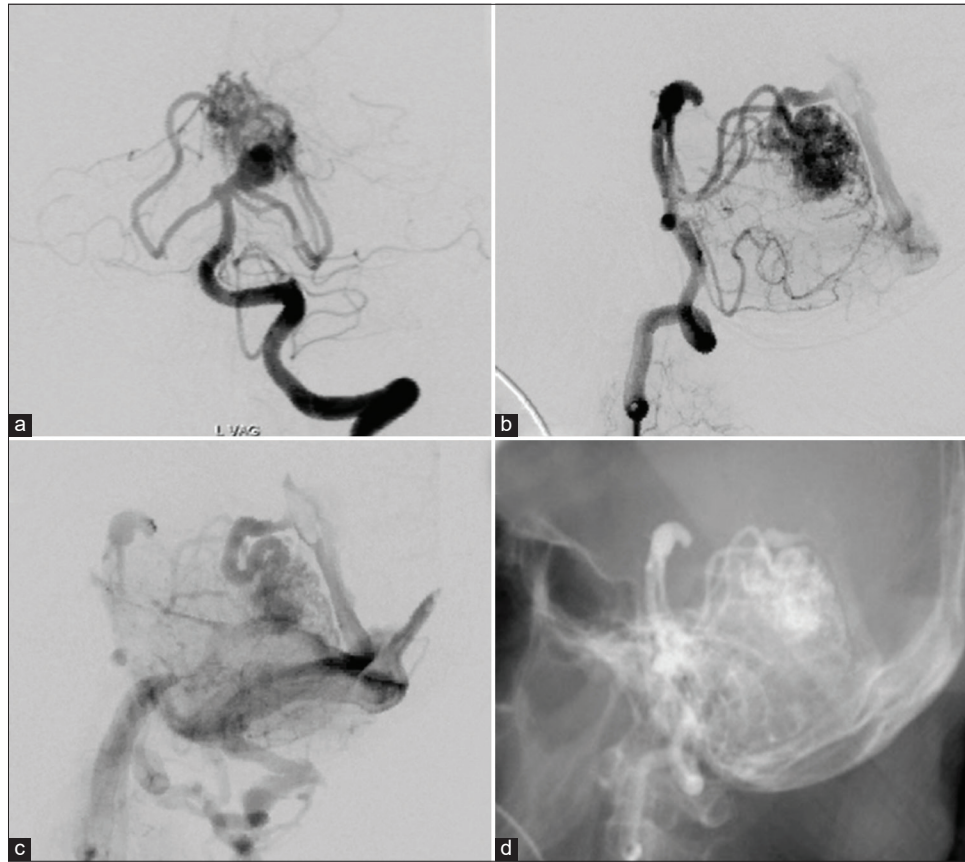


Figure 2: Left vertebral angiogram of initial DSA. (a) Anteroposterior view (b) lateral view (c) venous phase of lateral view (d) lateral view with bone.

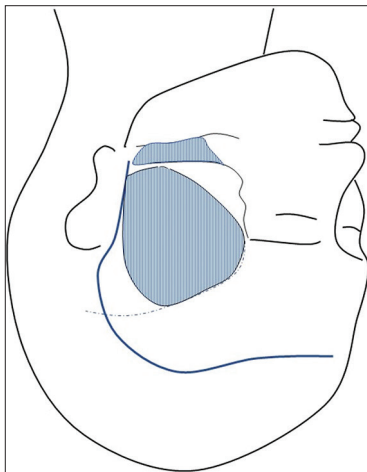


Figure 3: The schema demonstrating the range of the pterional craniotomy and zygomectomy of aneurysmal clipping.

band using a high-speed drill [Figure 4a]. Dissecting the deep middle cerebral veins and anterior temporal arteries from the temporal surface enabled gentle posterior retraction of the temporal pole through the space under these vessels, and this step created a surgical field looking up into the

retrocarotid space from caudolaterally [Figure 4b]. The aneurysm was clipped through this widened retrocarotid space [Figure 4c] while fully observing the surrounding vital structures; however, an obvious rupture point with a fibrin plug could not be detected because of the higher location of the aneurysmal apex. Moreover, intracranial pressure was quite high intraoperatively, which suggested that increased venous pressure in the deep venous system associated with the AVM might be causing the disorder in the supratentorial venous return. Indeed, the ventricular and cisternal drains inserted during the initial surgery showed a pressure of ≥ 150 mm H₂O continuously after the surgery, which could vitally hinder managing cerebral vasospasm after SAH. In addition to these findings, postoperative MRI on day 5 demonstrated no apparent brain contusion due to the initial surgery and no cerebral vasospasm [Figure 5]; therefore, we subsequently removed the AVM on day 8 to survive the latter half of the risk period for cerebral vasospasm.

The patient was positioned in left-sided lateral recumbency, and a left rectangular occipital craniotomy along the venous angle made by the superior sagittal sinus and transverse sinus was made [Figure 6]. By opening the quadrigeminal cistern and draining the cerebrospinal fluid, the left

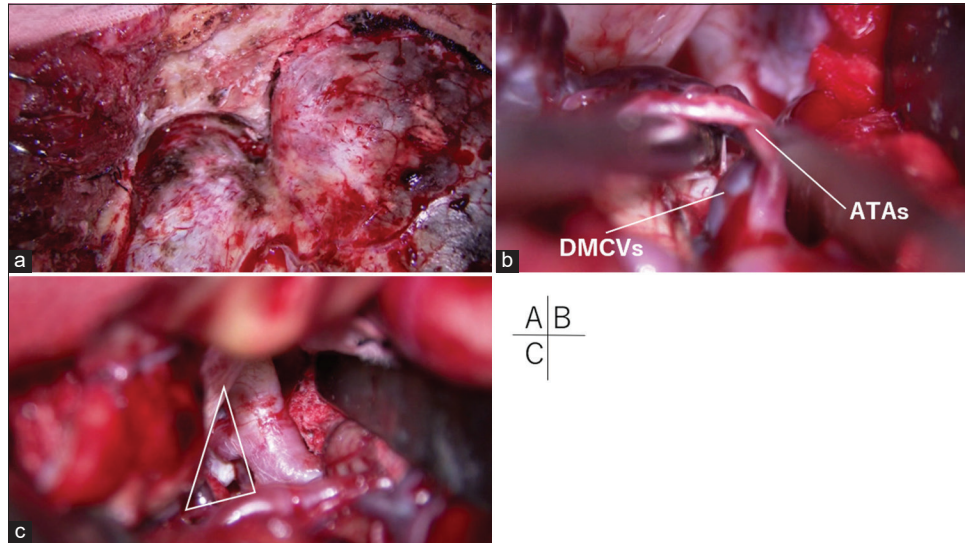


Figure 4: (a) The superior-lateral wall of the orbit was flattened to the depth of the meningo-orbital band. (b) Dissecting the deep middle cerebral veins (DMCVs) and anterior temporal arteries (ATAs) from the temporal surface enabled gentle posterior retraction of the temporal pole through the space under these vessels. (c) The aneurysm was clipped through widened retrocarotid space (white triangle).

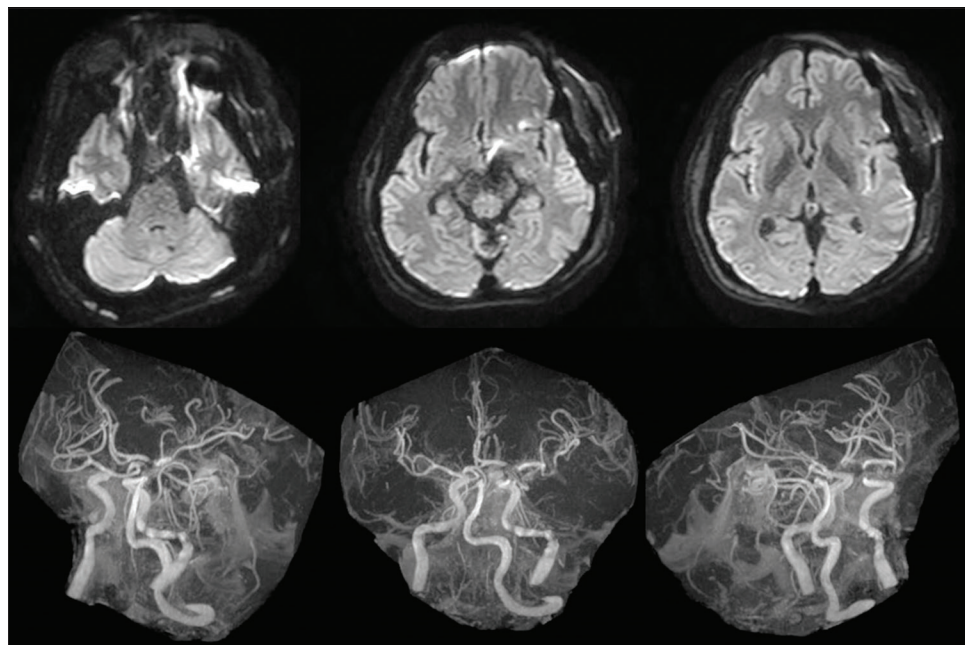


Figure 5: DWI-MRI and MRA on day 5 demonstrated no conspicuous brain damage and cerebral vasospasm.

occipital lobe naturally relaxed. Next, we made a tentorial incision parallel to the straight sinus beginning from the tentorial incisura and advancing posteriorly toward the transverse sinus by coagulating the venous oozing from the tentorial sinus [Figure 7a]. In the quadrigeminal cistern, the bilateral SCA feeders were detected and cross-clamped as close to the nidus as possible to decrease intranidal pressure [Figure 7b]. Next, as appropriate, relatively thick feeders were skeletonized, coagulated, and cut after reducing the arterial pressure using microclips [Figure 7c],

and the boundary plane was created by coagulating and aspirating the surrounding gliosis layer using a low-voltage bipolar device. At the posterior-superior boundary of the nidus, the main feeder from the PICA was coagulated and divided [Figure 7d], and the nidus was elevated anteriorly. Relatively robust feeders flowing into the nidus near the main drainer were fully skeletonized, coagulated, and cut while preserving the passing arteries [Figure 7e]. Finally, the main drainer was ligated, and the nidus was totally removed [Figure 7f].

Postoperatively, the pressure in the ventricular drain promptly decreased to <50 mm H₂O, and the patient recovered, with improved consciousness to GCS E4V5M6. There were no conspicuous brain infarctions and contusions on postoperative imaging, and the aneurysm and the nidus were completely disappeared in postoperative DSA [Figure 8]. The patient's postoperative course was favorable, without symptomatic cerebral vasospasm. Although approximately 1 mm of ptosis remained, the exotropia and anisocoria resolved completely. The patient was transferred to a rehabilitation hospital, with a modified Rankin scale score of 2.

You can see the actual surgical video on electronic supplementary material [Videos 1 and 2].

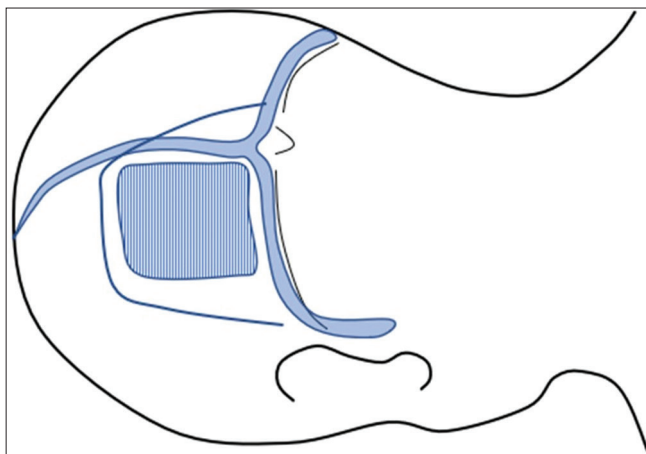
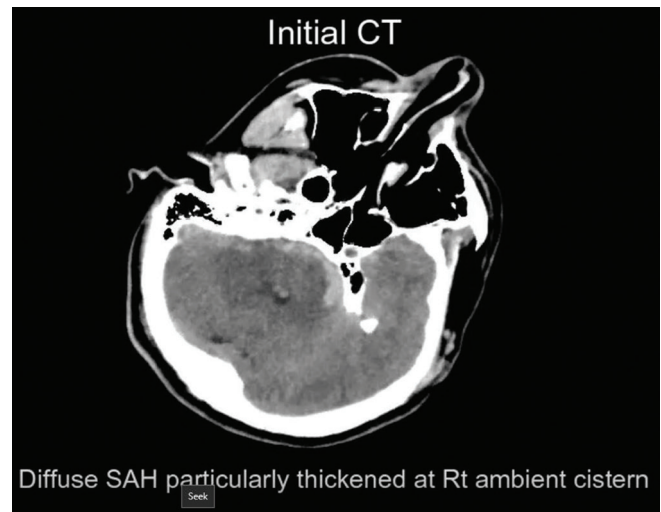


Figure 6: The schema demonstrating the range of the craniotomy of occipital tentorial approach.

DISCUSSION

The bleeding frequency in cases of AVM coexisting with an aneurysm is higher than that of AVM alone (1.7% vs. 7% per year, respectively),^[4] and in particular, posterior fossa AVM is more likely to present with a hemorrhagic onset^[5,15]. When bleeding occurs in an AVM coexisting with an aneurysm, it is essential to first identify the source of the bleeding. In cases of posterior fossa AVM coexisting with an aneurysm presenting with hemorrhage, intranidal aneurysm has a higher risk of rupture than AVM. In contrast, flow-related aneurysms are



Video 1: This video demonstrates the clipping part of the actual surgical procedure for a case of superior vermian arteriovenous malformation coexisting with basilar-superior cerebellar artery aneurysm.

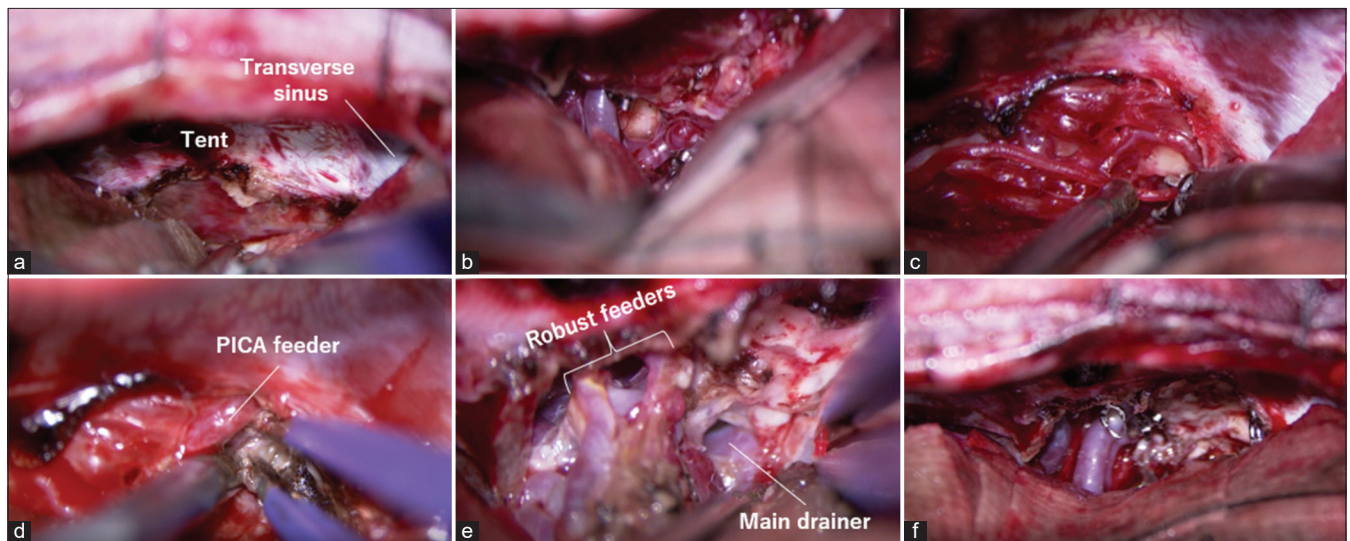
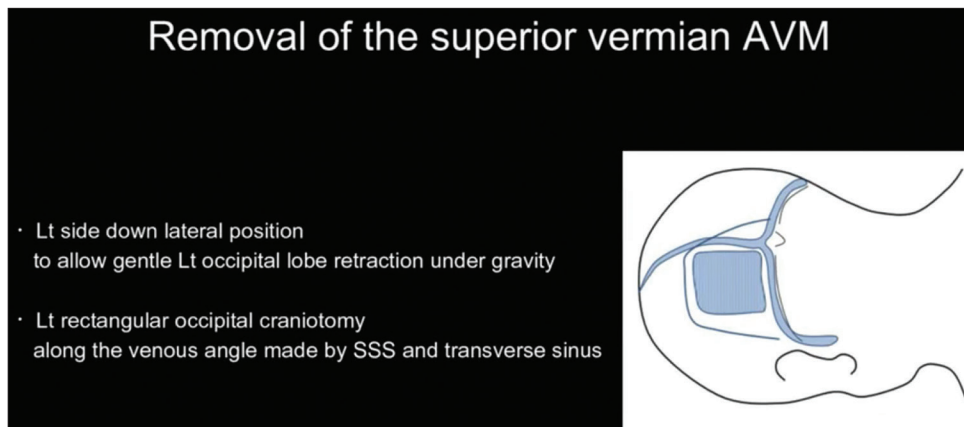


Figure 7: (a) Tentorial incision parallel to the straight sinus was advanced posteriorly. (b) The quadrigeminal cistern was opened and the main feeders of superior cerebellar artery were clipped preliminary. (c) Relatively thick feeders were separated after reducing the arterial pressure using microclips. (d) The posterior inferior cerebellar artery (PICA) feeder was skeletonized. (e) Robust feeders were fully skeletonized, coagulated, and cut while preserving the passing arteries. (f) The nidus was removed.



Video 2: This video demonstrates the AVM removal part of the actual surgical procedure for a case of superior vermian arteriovenous malformation coexisting with basilar-superior cerebellar artery aneurysm.

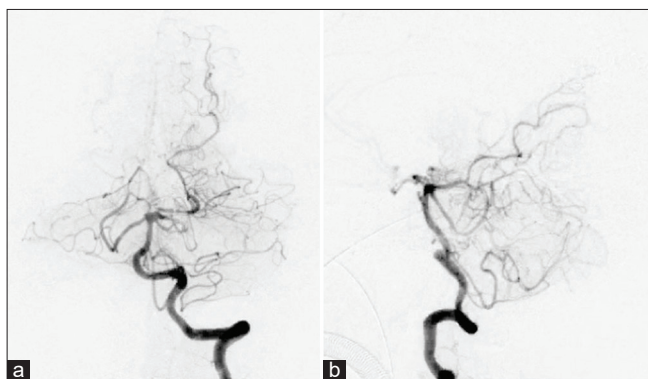


Figure 8: The aneurysm and the nidus were completely disappeared on postoperative DSA. (a) Anteroposterior view (b) lateral view.

less likely to rupture than AVMs. The CT findings at onset are useful because pure SAH suggests the possibility of bleeding from the aneurysm, and conversely, SAH accompanied by ICH or IVH suggests likely bleeding from the AVM.^[14,15] In our case, the CT findings showed pure SAH without ICH or IVH, and left oculomotor palsy supported aneurysmal rupture. However, the subarachnoid clots particularly thickened in the right ambient to quadrigeminal cisterns, and the flow-related aneurysm in the branch of the inflowing left SCA suggested AVM rupture.

In cases of AVM coexisting with an aneurysm, strategies emphasizing aneurysmal treatment tend to be adopted first considering that if the AVM is treated first, the increased hemodynamic stress to the flow-related aneurysm may result in rupture.

Redekop *et al.* subdivided flow-related aneurysms into proximal feeder aneurysms existing in the proximal end of the feeding artery (circle of Willis, middle cerebral artery, and vertebrobasilar artery, etc.) and distal feeder aneurysms existing on the trunk of the feeding artery. However, the authors reported that the former has a very low probability

of disappearing after AVM treatment (4%), while the latter is likely to regress after AVM treatment (80%).^[14] In our case, if we prioritized treating the AVM, the possibility of spontaneous regression of the aneurysm was quite low. Instead, the aneurysm had to be treated simultaneously because the hemodynamic stress to the aneurysm could increase after AVM treatment, resulting in aneurysmal rupture. For these reasons, we prioritized aneurysmal treatment.

Regarding treating the AVM, ruptured aneurysms have a higher risk of re-rupture and worsening.^[8] In contrast, the risk of re-rupture of an AVM is relatively low, and recovery of neurological function is good compared with aneurysmal SAH and hypertensive ICH; therefore, some argue that acute treatment of ruptured AVMs is unnecessary.^[6,12] In contrast, the efficacy of acute treatment of ruptured AVMs is described in recent reports,^[7,13] and in particular, Pavesi *et al.* recommended active surgical treatment in the hyperacute phase only for AVMs with a Spetzler–Martin grade of I or II.^[13]

In our case, we could not confirm aneurysmal rupture during the initial surgery, and the disorder of the venous return due to the increased venous pressure in the straight sinus associated with the AVM caused increased intracranial pressure; thus, the existence of the AVM was a vital hindrance to managing the SAH. Therefore, we determined that it was desirable to treat the AVM as soon as possible.

Regarding the surgical approach to each lesion, it is generally desirable to treat through a single approach that enables observing both lesions in the same surgical field. To treat the aneurysm, we considered a pterional approach and a subtemporal transtentorial approach, and only the latter allowed access to both lesions. However, in aneurysmal clipping using the subtemporal transtentorial approach, there was a risk of obstruction to the vein of Labbe because of retraction of the temporal lobe of the dominant hemisphere.

As for the AVM, the subtemporal transtentorial approach was suitable for early processing of the ipsilateral SCA, but it was a little too proximal and lateral to expose the contralateral and dorsal surfaces of the AVM and was inappropriate as an approach to the lesion located in the retrocollicular midline region.^[11,16,17] For these reasons, we selected the pterional approach for the BA-SCA aneurysm. In the actual surgery, the aneurysm was located relatively high and the exposure was still limited even after zygomectomy; therefore, adding extradural anterior clinoidectomy to mobilize the internal carotid artery and oculomotor nerve may have improved the visualization of the aneurysm.

Regarding the approach to the AVM, we considered the suboccipital supracerebellar infratentorial approach and the posterior interhemispheric occipital transtentorial approach (OTA). The AVM in the current case was fed by multiple SCA branches in the precerebellar or quadrigeminal cistern in front of the anterior surface of the cerebellum, and early processing of these arteries was essential to decrease intranidal pressure. However, this space might be hidden by the dilated superior vermian vein or AVM nidus itself in the supracerebellar approach. Furthermore, the increased intranidal pressure and impaired perfusion of the draining vein due to the downward retraction of the swollen cerebellum increased the risk of causing intraoperative AVM rupture;^[16,18] therefore, we selected OTA. OTA is the most appropriate approach to offer a broad vertical view of the anterior-superior and anteromedial aspect of the vermis and cerebellar hemisphere, and it is rarely necessary to sacrifice vital neurovascular structures. Preoperative embolization for AVM with Spetzler–Martin Grade II helps to relieve intraoperative nidus pressure by occluding deep-seated and high-flow feeders, which are difficult to identify intraoperatively.^[20] However, in our case, main feeders were considered easy to identify and process through the OTA, as mentioned above; therefore, preoperative embolization was not necessary.

Although there are various opinions regarding the patient's position, we prefer to have the affected sided down, with the patient in lateral recumbency, to allow natural and gentle retraction of the occipital lobe under gravity and to prevent postoperative occipital lobe contusion causing hemianopsia.^[2,16,19] It is also important to prevent occipital lobe damage due to contact between the dural edge and the occipital lobe by not expanding the range of the dural incision to the outside. In this approach, bleeding from the tentorial sinus may limit posterior extension of the tentorial incision, resulting in difficulty processing the posterior feeders from the PICA, or more external SCA or anterior inferior cerebellar artery feeders. In addition, if the AVM body greatly extends contralaterally beyond the midline or exhibits caudal dilatation to the cerebellar tonsils or the fourth ventricle, using only this approach may limit exposure.^[3,16,19] In such

cases, it is necessary to consider combined approaches and concomitant preoperative feeder embolization.^[1,10]

CONCLUSION

We reported an excellent outcome in a case of superior vermian AVM coexisting with a BA-SCA proximal feeder aneurysm successfully treated by a two-stage operation in the acute phase of SAH.

In particular, disorders of intracranial venous return associated with an AVM can be a vital hindrance to managing cerebral vasospasm, and treating both lesions in the acute phase may lead to good outcomes.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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