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Case Report

Double LVIS Jr. Stenting of a Ruptured Proximal Anterior Inferior Cerebellar Artery Aneurysm: A Case Report

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Objective: We report a case of ruptured aneurysm at the anterior pontine segment of the anterior inferior cerebellar artery (AICA) which re-ruptured after stent placement and was treated by overlapping stenting.
Case Presentation: A 53-year-old woman presented with headache. CT demonstrated subarachnoid hemorrhage. DSA revealed no evident source of bleeding. On day 10, she complained of sudden headache and CT demonstrated re-bleeding. On repeated DSA, an aneurysm at the anterior pontine segment of the right AICA was found. An LVIS Jr. stent was deployed at the right AICA including the aneurysm. On postoperative day 23, the aneurysm ruptured again. Another LVIS Jr. stent was deployed at the same area. On day 56, she was discharged home without neurological deficit.
Conclusion: Intracranial aneurysms not indicated for coil embolization or parent artery occlusion are difficult to treat. Overlapping stenting may be a treatment option for such aneurysms.

Keywords ▶ anterior inferior cerebellar artery, subarachnoid hemorrhage, stenting, LVIS Jr.

Introduction

Aneurysms developing in the peripheral anterior inferior cerebellar artery (AICA) are rare, accounting for approximately 0.1% of all cerebral aneurysm cases.[1)] Endovascular treatment of aneurysms in this region has been occasionally reported, but there has been no case report of treatment with stent placement alone. We report a patient with an AICA anterior pontine segment aneurysm that developed upon subarachnoid hemorrhage. It was treated by placing a single LVIS Jr., but it re-ruptured and subsequent rupture was prevented by overlapping stenting. We report the case with a literature review.

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Case Presentation

Patient: A 53-year-old woman.
Family history: Not contributory.
Medical history: Not contributory.
Present illness: Right occipital headache developed after the patient became aware of hearing impairment during bathing, and she visited the emergency service of our hospital.
Neurological findings on the first examination: The patient complained of mild headache rated Japan Coma Scale 0 and Glasgow Coma Scale 15, but no symptom of neurological deficit was clearly observed.
Neuroradiological findings: On head CT, Fisher grading scale 3 subarachnoid hemorrhage was noted in the right premedullary cistern over the right ambient cistern and right Sylvian fissure (**Fig. 1A** and **1B**). Head MRI and emergency DSA were performed, but no source of bleeding was observed. On MRA, the left vertebral artery (VA) was not visualized through the neck region (**Fig. 1C**).

Based on the above, the patient was diagnosed with Hunt and Kosnik grade I, World Federation of Neurosurgical Societies grade I subarachnoid hemorrhage of unknown source of bleeding, and resting and blood pressure control at a systolic blood pressure lower than 130 mmHg were performed. Re-examination by cerebral angiography was

Fig. 1 (**A** and **B**) CT on admission shows subarachnoid hemorrhage in the right premedullary cistern, ambient cistern, and Sylvian fissure. (**C**) MRA on admission shows no evident source of bleeding.

performed on day 6 after the onset, but the source of bleeding was unclear. On day 10 after the onset, headache sharply aggravated and a small volume of subarachnoid hemorrhage newly developed centering in the right premedullary cistern on head CT (**Fig. 2A**). DSA was repeated and an aneurysm was present in the non-vascular bifurcation region of the anterior pontine segment of the right AICA. The right posterior inferior cerebellar artery (PICA) was lost and the right AICA perfused the right PICA region (**Fig. 2B–2D**). The aneurysm diameter was 0.9 mm and the AICA diameter was 1.4 mm in the proximal region of the aneurysm.

Based on the above, the patient was diagnosed with subarachnoid hemorrhage induced by rupture of the AICA anterior pontine segment aneurysm. Surgery was performed after sufficiently explaining the following conditions and receiving consent to these: Parent artery occlusion will be performed if the aneurysm ruptures during surgery and hemostasis is difficult, and decompressive craniotomy may be necessary when cerebral infarction and brain swelling are severe.

Findings of the first surgery: Aspirin at 300 mg and clopidogrel at 300 mg were orally administered 30 minutes before entering the operating room. Under general anesthesia, A 5Fr FUBUKI Dilator Kit (Asahi Intecc, Aichi, Japan) was inserted into the right brachial artery and a 3.4Fr Tactics (Technocrat, Aichi, Japan) was placed in V4 at the 5th cervical vertebra level of the right VA. To advance a Headway17 (MicroVention/Terumo, Aliso Viejo, CA, USA) from the basilar artery to the right AICA, the micro guidewire crossing the lesion was pulled while adding resistance, which made the Headway17 jump to the right AICA and rapidly increased the blood pressure. The blood pressure was reduced and the LVIS Jr. 2.5 × 13 mm (MicroVention/Terumo) was quickly placed centering on the aneurysm. Bleeding from the aneurysm was noted on imaging performed immediately after placement (**Fig. 3A**), but heparin was not neutralized. When images were acquired after waiting for a few minutes, bleeding from the aneurysm had stopped and the aneurysm was smaller than before surgery and difficult to see (**Fig. 3B**); therefore, the procedure was completed.

Course after the first surgery: After surgery, bed rest and blood pressure control at a systolic blood pressure below 130 mmHg were continued. Aspirin and clopidogrel were orally administered at 100 mg/day and 75 mg/day, respectively, from the day following surgery. DSA was performed on day 33 after onset. The aneurysm was visualized, although it was smaller than before surgery (**Fig. 3C**). On day 36 after onset, headache became stronger during showering and repeat bleeding in the right premedullary cistern was noted on head CT (**Fig. 4A**).

Findings of the second surgery: Overlapping stenting using an LVIS Jr. 2.5 × 13 mm was applied in the same region using the same approach and device as in the first surgery. The aneurysm became no longer visualized immediately after placement (**Fig. 4B**). Spasm of the AICA proximal to the stent placement region occurred, but it improved after waiting for 5 minutes on imaging.

Course after the second surgery: The bed rest level expanded in a stepwise manner after surgery. On DSA performed on day 49 after onset, the aneurysm was not visualized (**Fig. 4C**). On day 53 after onset, symptoms of neurological deficit were absent and the patient was discharged to home. After discharge, oral administration of 100 mg/day of aspirin and 75 mg/day of clopidogrel was continued, and oral clopidogrel administration was discontinued 1 year after the first surgery. At 20 months after onset, the patient was independent

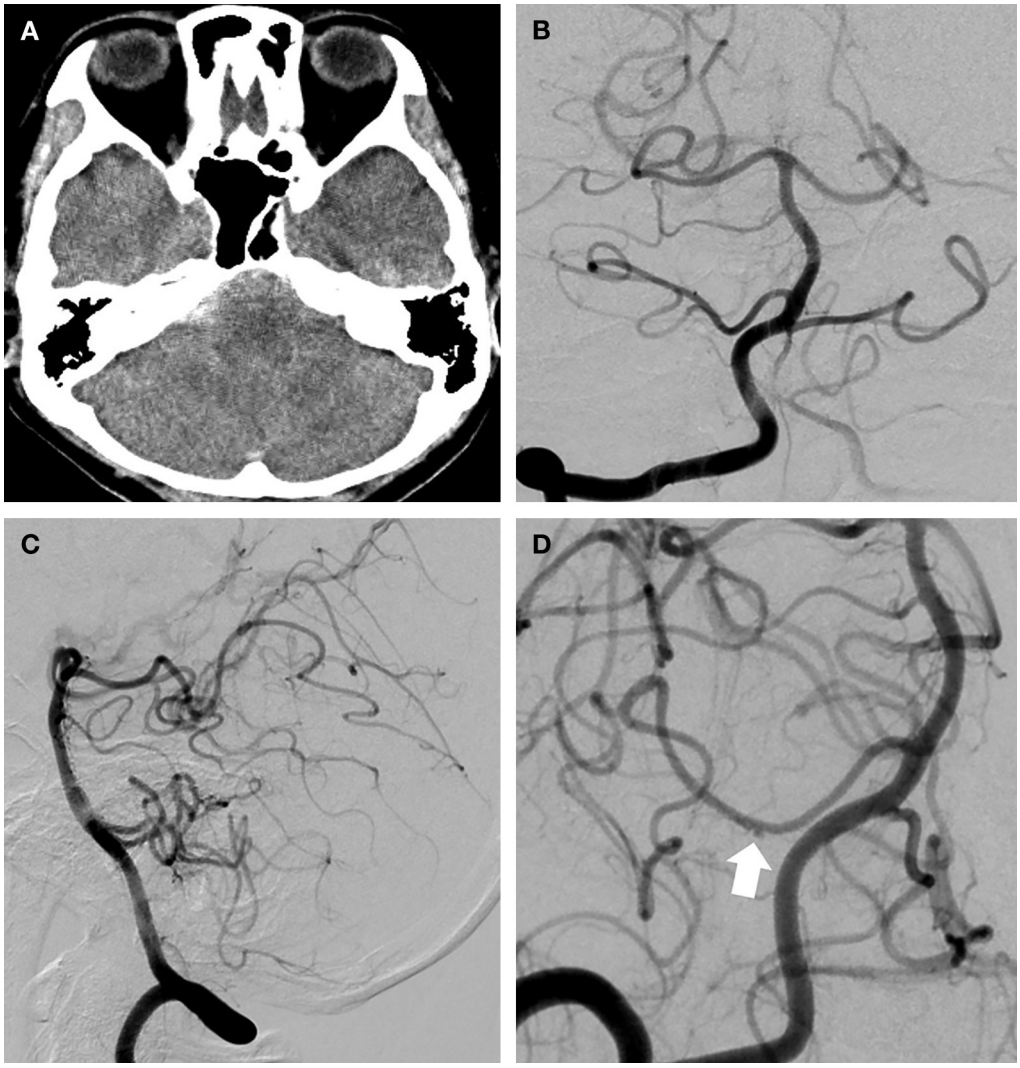

Fig. 2 CT (**A**) and right vertebral angiography (**B–D**) just after re-bleeding. (**A**) CT shows new subarachnoid hemorrhage in the right premedullary cistern. (**B** and **C**) Anteroposterior and lateral views show the absence of the right PICA. (**D**) Oblique view shows a saccular aneurysm (arrow) in the anterior pontine segment of the right AICA. AICA: anterior inferior cerebellar artery; PICA: posterior inferior cerebellar artery

in daily life without an adverse event and had returned to their workplace (mRS0). No new abnormality was noted on head MRI and visualization of the AICA peripheral to the stent placement was favorable on MRA (**Fig. 4D**).

Discussion

The frequency of aneurysm development in the peripheral AICA is 0.03%–0.5%, being rare, and 80% of these cases develop upon subarachnoid hemorrhage.[2,3] Gi et al.[4] anatomically classified the development site into six segments and observed the highest frequency (72%) of aneurysm formation in the vicinity of the metal loop. The aneurysm in the present patient was located in the anterior pontine segment from the origin of the AICA over the region immediately before bifurcation to the lateral and medial branches. To our knowledge, 10 cases of aneurysms in this region have been reported, including our patient.[5,6] All 10 cases were in females, the age was 15–85 years old (mean: 59.6 years old), and the disease developed upon subarachnoid hemorrhage in 8 (80%). The aneurysm developed in a bending region other than the bifurcation in all cases. Regarding the cause of development in a bending region in the posterior cranial fossa including the AICA, it was noted that a blood vessel gradually formed from the embryonic reticular structure, the bending region corresponded to the part which lost the branch in this step, and the vascular wall of this region was congenitally vulnerable.[7] The ipsilateral PICA was defective

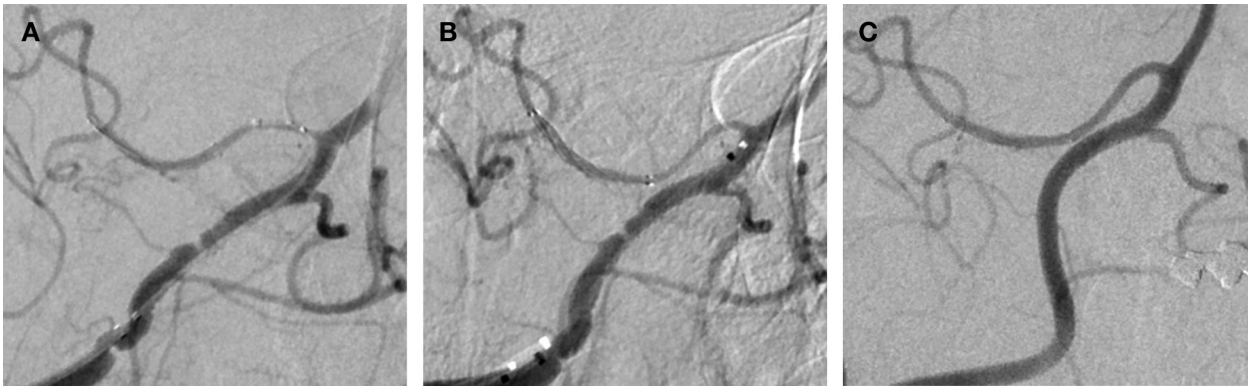

Fig. 3 Oblique view of the right vertebral angiography. (**A**) Just after stent placement, the aneurysm is bleeding. (**B**) A few minutes after stent placement, the aneurysm stops bleeding. (**C**) Twenty days after stent placement, the aneurysm is smaller than before surgery.

(eight cases) or hypoplastic (two cases),[5,6] suggesting that accompanying hemodynamic stress on the AICA was involved in the development and rupture of the aneurysms. Five of the 10 cases of AICA anterior pontine segment aneurysm were diagnosed as dissecting aneurysm.[5,6] In our patient, the disease was unable to be diagnosed as dissecting aneurysm because the macroscopic and pathological findings were unclear due to neuroendovascular treatment and the neuroradiological findings, but it developed at a non-bifurcation site, suggesting dissecting aneurysm. For treatment of AICA anterior pontine segment aneurysms, endovascular treatment was performed in 7 of the 10 cases, and parent artery occlusion and coil embolization were performed in 2 and 4, respectively.[5,6] When extensive cerebral infarction due to parent artery occlusion is of concern, trapping after occipital artery-AICA anastomosis is a treatment option, but the surgical approach to the proximal AICA is relatively difficult and bypass is also difficult because it is applied in a deep region, especially in the acute phase. In this patient, the right PICA was defective and the region perfused by the right AICA was wide, suggesting that extensive cerebral infarction of the right AICA induced by parent artery occlusion is likely. Moreover, the aneurysm was hemispherical with a major axis of 0.9 mm, being small; therefore, coil embolization was judged as inapplicable and treatment by stent placement alone was performed. To our knowledge, no case of peripheral AICA aneurysm treated by stent placement alone has been reported.

When both parent artery occlusion and coil embolization are inapplicable, flow diverter stent placement is investigated expecting a flow diverter effect,[8] but the indication in Japan is 'large or giant wide neck-type intracranial aneurysms located in the petrous part of the internal carotid artery (ICA) over the superior hypophyseal region' and indicated cases are limited because it cannot be used in the acute phase of rupture. Treatment by placing two or more stents in the parent artery of an aneurysm expecting the flow diverter effect is mainly performed for vertebral arterial dissecting aneurysms and internal carotid arterial blood blister-like aneurysms. Chung et al.[9] reported nine cases of vertebral arterial dissecting aneurysm in which three stents were placed and complete regression was achieved in 88.9%. They reviewed 44 and 11 cases of vertebral arterial dissecting aneurysm treated by placing two and three stents, respectively, for 55 cases in total, and reported that complete regression was acquired in 61.8%. Fang et al.[10] placed 2, 3, and 4 stents in 9, 5, and 1 case of internal carotid arterial blood blister-like aneurysm, respectively, and complete occlusion of the aneurysm was acquired in the six cases with three- and four-stent placement, but recurrence developed in three (33%) of the nine cases with two-stent placement. Based on these reports, the metal coverage increases as more stents are placed, leading to a higher aneurysm occlusion rate.

The metal coverage of a flow diverter stent is 30%–35%, whereas those of stents used to assist coil embolization of cerebral aneurysms are as follows: LVIS, 23% and Enterprise, 8%, being low. The metal coverage of LVIS is higher than that of Enterprise and when 2 LVIS stents were placed by overlying, the wall shear stress and flow velocity in the aneurysm decreased by 63.9% and 46.1%, respectively, exhibiting a stronger effect than those (51.1% and 37.9%, respectively) of placing one flow diverter stent (PIPELINE).[11] Zhang et al.[12] placed two and three LVIS stents in 12 and 1 case of ruptured internal carotid arterial blood blister-like aneurysm, respectively, and observed the features of recurrence in four cases (30.8%), but it was spontaneously occluded by withdrawal of antiplatelet agents in two and

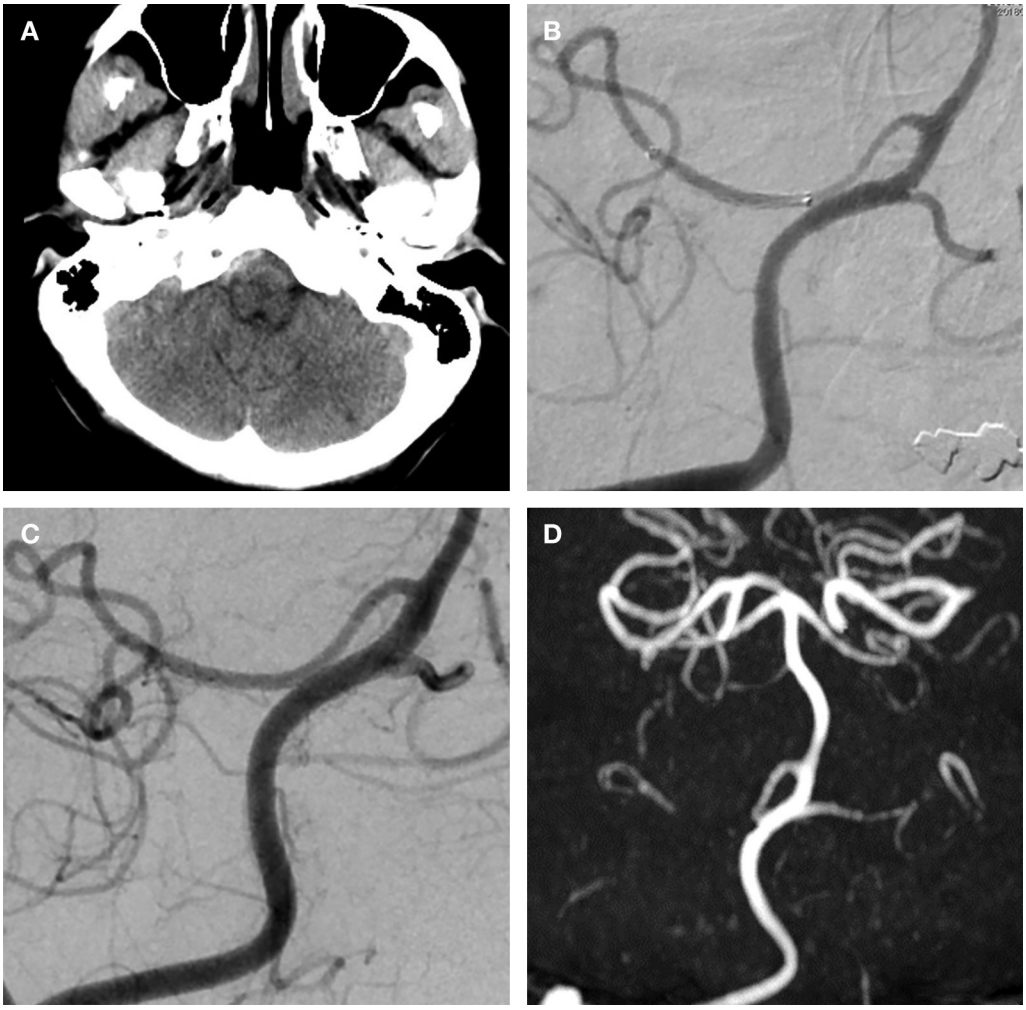

Fig. 4 (**A**) Thirty-six days after onset, CT shows new subarachnoid hemorrhage in the right premedullary cistern. Oblique view of the right vertebral angiography. (**B**) Just after overlapping stenting, the aneurysm disappeared. (**C**) Ten days after overlapping stenting, the aneurysm did not recur. (**D**) Twenty months after onset, MRA confirmed that the right AICA is patent. AICA: anterior inferior cerebellar artery

occlusion was acquired by additional LVIS placement in one case, achieving a favorable outcome with a final occlusion rate of 92.3% (12/13). Yan et al.[13] treated 15 aneurysms (ICA: 5, VA: 5, basilar artery [BA]: 2, middle carotid artery [MCA]: 1, PcoA: 1, Acho A: 1) by overlapping stenting using 2 LVIS in 10 patients. Coil embolization was concomitantly applied to 13 aneurysms and 2 were treated by stenting alone. On cerebral angiography performed after 3–8 months, complete occlusion was acquired in 86.7% (13/15) and the parent artery was patent in all cases. Santillan et al.[14] reported 35 cases of cerebral aneurysm with a parent arterial diameter of 0.9–2.5 mm (mean: 2.2 mm) treated by coil embolization using an LVIS Jr., in which perioperative in-stent thrombus was formed in 4 (11.4%). Y-stents were placed in three of the four cases, and they discussed that an increase in the amount of metal increases the possibility of thrombotic complications. In the present patient, the AICA with a 1.4-mm diameter was treated by overlapping stenting using 2 LVIS Jr stents. As the AICA diameter was homogeneous and the curve was mild in the stent placement region, close adherence of the stent to the vascular wall was relatively favorable and this may have been a cause of the absence of thrombotic complications.

At our facility, systolic blood pressure is controlled below 130 mmHg during bed rest in patients with subarachnoid hemorrhage of unknown source of bleeding. Cerebral angiography is performed targeting days 7 and 14 after the onset, and the rest is gradually withdrawn when the source of bleeding is still unclear on day 14. In this patient, re-rupture occurred on day 10 after onset and the presence of the right proximal AICA aneurysm was clarified by cerebral angiography. On right vertebral arteriography, bending and

meandering of the distal V2 over V3 were strong, and the angle between the basilar artery and right AICA was sharp. As microcatheter operation around the lesion was expected to be difficult for the first surgery, a 3.4Fr Tactics was used as an intermediate catheter and the operability was favorable. A Synchrosoft2 was advanced crossing the lesion to advance a Headway17, but it was caught in the origin of the AICA. When the wire was pulled while adding resistance by pushing the Headway17, the Headway17 jumped into the AICA and the aneurysm ruptured. The AICA rapidly became linear and distorted the aneurysm, which was presumed to be the cause of rupture. For the stent, an LVIS Jr. was selected because the metal coverage is high and it is indicated for blood vessels thinner than those for which an LVIS is indicated. When one 2.5 × 13 mm LVIS Jr. was placed and imaged after waiting for a few minutes, no extravasation was observed, and the aneurysm became small and unclear. As the bleeding volume was small and it stopped within a short time, the parent artery was not occluded. Overlapping stenting was considered at this point, but it was not performed considering the risk of re-rupture aggravating the condition due to re-advancing the Headway17 pulled back to the basilar artery into the Rt. AICA and the risk of thrombotic complication because rupture occurred in the lesion cross process. The procedure was completed and the course was followed. On DSA on postoperative day 20, the aneurysm was smaller than before surgery and the contour was unclear. The rest level gradually expanded, but repeated bleeding was noted. Parent artery occlusion was to be performed if rupture occurred again during the second surgery and hemostasis was difficult, but intraoperative rupture did not occur and overlapping placement of a 2.5 × 13 mm LVIS Jr. was possible in the same region as that in the first surgery employing the same procedure. As the AICA diameter to receive stenting was 1.4 mm, being thin, only one LVIS Jr. was first placed, but for sufficient flow diverter effects, placing two or more stents was necessary. In each case, it is necessary to investigate placing two or more stents by trade-off between thrombotic complications and prevention of hemostasis. In this patient, aneurysm rupture occurred during the first surgery and spasm of the right AICA developed during the second surgery. Guiding of a microcatheter to the AICA and application of endovascular procedures to the peripheral AICA lesion were difficult because bending of the V3 VA segment was strong, the angle of the right AICA bifurcation from the basilar artery was sharp, and the AICA vascular system was thin. It may be important to prevent loading pressure on the AICA vascular wall by improving operability using an intermediate catheter and performing precise operations through sufficiently investigating the device.

For intracranial aneurysms for which it is difficult to directly apply coil embolization, those expected to develop serious complications due to parent artery occlusion, and for which a direct approach is difficult, such as that observed in this patient, hemostatic effects by stent placement alone may be expected and this may be a treatment option.

Conclusion

An AICA anterior pontine segment aneurysm that developed upon subarachnoid hemorrhage re-ruptured when only one LVIS Jr. was placed, but hemostatic effects were acquired by overlapping stenting. For cerebral aneurysms for which coil embolization, parent artery occlusion, and direct surgery are difficult to apply, this method may be a useful treatment option.

Disclosure Statement

The authors declare no conflict of interest.

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