Online May 21, 2022
Journal of Neuroendovascular Therapy 2022; 16: 439–445 DOI: 10.5797/jnet.oa.2021-0096

Original Article

Case Series of Endovascular Therapy for Blood Blister-Like Aneurysm in Acute Phase

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Objective: Blood blister-like aneurysms (BBAs) of the internal carotid artery are highly challenging to treat due to their variable morphology and tendency for rupture and regrowth. Here, we report a single-institution experience of endovascular therapy (EVT) for BBA treatment.

Methods: We retrospectively reviewed patients with ruptured BBA from 2006 to 2019. All patients in whom BBA was treated with EVT were included. Patients' aneurysmal characteristics, progression status, aneurysm occlusion on follow-up angiography, and modified Rankin Scale (mRS) score were recorded.

Results: A total of 11 patients (5 women and 6 men) with the mean age of 46 ± 10 years were included in this study. As initial treatment, 9 patients were treated with stent-assisted coiling (SAC). Immediate angiographic results showed that 2 cases were body filling, 4 were neck remnant, and 3 were complete obliteration. Perioperative ischemic complications were not observed. On postoperative day 1, 2 patients suffered from rerupture, and their prognoses were poor. Retreatments were performed in 5 patients. Parent artery occlusion (PAO) was performed in 6 patients including 2 initial treatments and 4 retreatments. Symptomatic infarction developed in 2 patients. In 3 patients, bypass in combination with PAO was performed. Clinical data revealed discharge mRS scores of 0–2 and 3–6 in 4 and 7 patients, respectively.

Conclusion: SAC is effective for the management of BBA. Careful follow-up and response are necessary after treatment with SAC.

Keywords ▶ blood blister-like aneurysm, endovascular therapy, stent-assisted coiling, parent artery occlusion, overlapping stent

Introduction

Intracranial blood blister-like aneurysms (BBAs) account for 0.5%–2.0% of ruptured intracranial aneurysms.[1] These aneurysms are located most frequently on nonbranching sites in the supraclinoid segment of the internal carotid artery (ICA). They are often small in size with a fragile wall and broad, poorly defined neck. BBAs are believed to represent a specific type of dissection or pseudoaneurysm.[2,3] These lesions have a very high risk of early recurrence and postoperative rebleeding.[4] Considering their fragile state and difficult morphology, BBAs prove challenging to manage either surgically or endovascularly. Endovascular interventional techniques evolved as an effective treatment for BBA.[5,6] Various endovascular approaches have been reported, but optimal management remains controversial. We present our experience in treating BBAs with endovascular treatment.

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Received: October 28, 2021; Accepted: April 11, 2022
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Materials and Methods

Patient selection

We retrospectively reviewed patients with ruptured BBAs who were treated from 2006 to 2019. All patients with BBAs who were treated with endovascular therapy (EVT) in the acute phase (within 3 days after subarachnoid hemorrhage [SAH]) were included. The diagnostic standards

for BBA included a location site in the supraclinoid portion of the ICA, no relationship with branch vessels, and a wide neck. The clinical conditions of the patients were evaluated using the Hunt and Kosnik (H&K) grading system.[7] Functional outcomes were assessed using the modified Rankin Scale (mRS)[8] at discharge. The degree of aneurysmal occlusion was classified into 3 groups: complete obliteration (CO), neck remnant (NR), and body filling (BF). We also collected background characteristics, including age, sex, and aneurysm size.

This study was conducted in accordance with the principles of the Declaration of Helsinki and approved by the institutional review board (approval number: 2021-148). The requirement for informed consent was waived for this study, as all patient identifiers were removed, and the data were collected retrospectively.

Treatment

Basically, stent-assisted coiling (SAC) was the first-choice treatment for BBA in the acute phase; before 2010; i.e., before the neck bridge stent was approved, SAC with coronary stent or parent artery occlusion (PAO) was performed depending on the case. Moreover, an extracranial ventricular drainage or spinal drainage was placed before the endovascular procedure depending on the case. Interventional procedures were performed within 3 days after SAH.

All treatments were performed under general anesthesia. Heparin was administered intravenously, followed by a saline flush. Activated clotting time was maintained at 250 to 300 seconds during the procedure. An 8 Fr balloon guiding catheter was placed in the cervical ICA via the common femoral approach. The first microcatheter, which was used for stenting, was utilized to the middle cerebral artery. A second microcatheter, which was used for coiling, was placed into the BBA sac or nearly at the neck of the BBA. Soft coils were selected and carefully placed into the BBA or around the neck of the BBA using the semi-jailing technique. Coil embolization was then performed as compactly as possible to include the front of the BBA with gradual stent deployment. If needed, more coils were added using the transcell technique. Before treatment, a loading dose of aspirin and clopidogrel (300 mg each) was administered through a nasal tube to the patients. After treatment, conventional dual antiplatelet regimen was continued in doses of 100 mg/day aspirin and 75 mg/day clopidogrel. In PAO, segmental ICA trapping was performed under proximal occlusion with an 8-Fr balloon guide catheter. The standard method is to occlude the aneurysm and parent artery with coils.

Screening follow-up was performed with simple X-ray or MRI. Angiographic follow-up was performed within 10 days after the operation. DSA was repeated if recurrence was observed. If significant recurrence of aneurysms was observed, coil embolization, placement of stents, or PAO with or without bypass was considered. Coil embolization was considered as the first choice if enough space was available to insert coils. Recently, overlapping stents have been considered as the first choice. However, in cases of hemodynamic tolerability or second retreatment, we considered PAO.

Results

Patient demographic characteristics, aneurysm size and location, and functional outcomes are summarized in **Table 1**. A total of 11 patients (5 women [45%] and 6 men [55%]) with a mean age of 46 ± 10 years were included in this study. The preoperative H&K grades were I, II, III, and IV in 1 (9%), 2 (18%), 3 (27%), and 5 (45%) patients, respectively. The mean aneurysm neck size was 3.9 ± 1.5 mm, and the mean aneurysm height was 2.0 ± 0.6 mm. Nine patients (82%) were treated with SAC, whereas 2 (18%) were treated with PAO on the initial treatment.

The postoperative course for 9 patients treated with SAC is summarized in **Table 1**. A driver stent (Medtronic, Minneapolis, MN, USA) was used in 3 patients (27%), Enterprise (Johnson & Johnson, Miami, FL, USA) in 4 (36%), and LVIS (MicroVention–Terumo, Aliso Viejo, CA, USA) in 2 patients (18%). Before 2010, a driver stent was used when we could not use neck bridge stents. In case 9, the coil could not be deployed; thus, the patient was treated with overlapping stents with LVIS.

Immediate angiographic results showed that 2 cases (18%) were BF, 4 (36%) were NR, and 3 (27%) were CO. Perioperative ischemic complications were not observed. Two patients (18%) suffered from rerupture on postoperative day 1, and their prognoses were poor. A representative reruptured case is shown in **Fig. 1**. Regrowth of the lesion was observed on angiographic follow-up in 6 patients (55%). The median period from the initial treatment to regrowth was 14 days (range, 1–24 days). Retreatments (coil embolization in 2 cases, PAO in 2 cases, and overlapping stents with LVIS stent in 1 case) were performed in 5 patients. Recurrence occurred in 2 patients treated with coil embolization as

Table 1 Basic characteristics and postoperative course of patients with blood blister aneurysms treated by EVT

Case	Sex	Age	H&K Grade	Aneurysm Size (mm)	EVT	Stent	Result	Rerupture	Regrowth	First retreatment (days from initial treatment)	Second retreatment (days from initial treatment)	mRS
1	M	45	3	1.8	SAC	Driver	NR	-	+	Coiling (41)	PAO (72)	0
2	F	56	2	2.1	SAC	Driver	CO	-	-	–	–	3
3	M	33	3	1.4	SAC	Driver	CO	-	+	PAO (23)	–	2
4	F	56	4	1.9	SAC	Enterprise	BF	-	+	–	–	4
5	M	32	3	1.7	SAC	Enterprise	CO	+	+	PAO (2)	–	6
6	F	48	2	3.2	SAC	Enterprise	NR	+	NA	–	–	6
7	M	56	4	3	SAC	Enterprise	BF	-	+	Coiling (39)	PAO (54)	4
8	F	54	1	1.7	SAC	LVIS	NR	-	+	Stent (16)	–	0
9	M	36	4	1.6	SAC	LVIS × 2	NR	-	-	–	–	6
10	M	54	4	2.3	PAO	–	CO	-	-	–	–	6
11	F	39	4	1.4	PAO	–	CO	-	-	–	–	5

+: yes; -: no; BF: body filling; CO: complete obliteration; EVT: endovascular therapy; F: female; H&K: Hunt & Kosnik; M: male; mRS: modified Rankin Scale; NA: not applicable; NR: neck remnant; PAO: parent artery occlusion; SAC: stent-assisted coiling

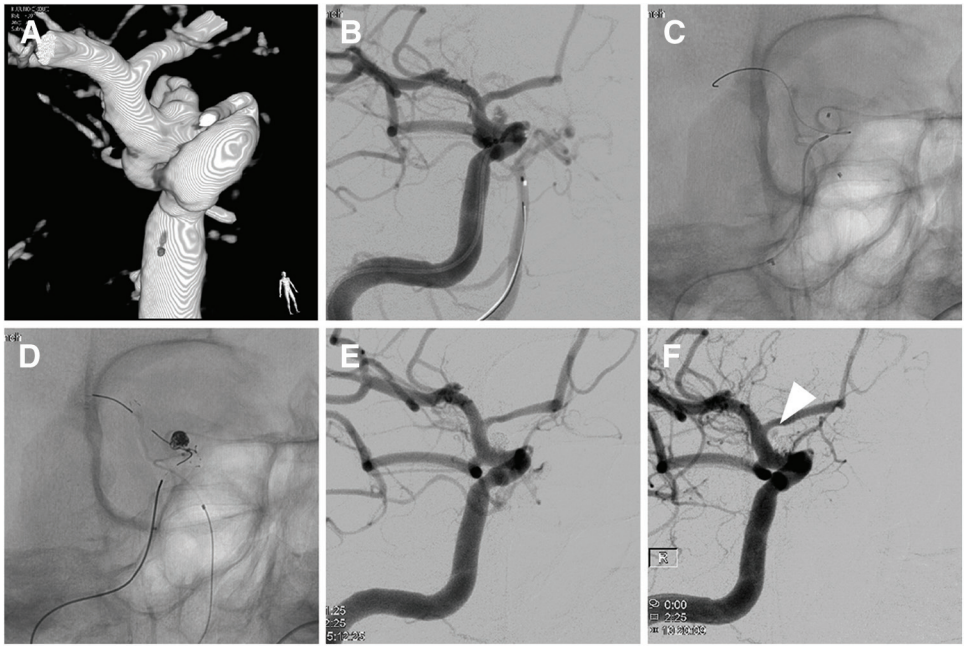

Fig. 1 Case 6 involved a 48-year-old woman with H&K grade 2 SAHs, which reruptured after SAC. (**A**) 3D reconstruction of the left ICA revealed a blood blister aneurysm. (**B**) Left ICA and right vertebral artery angiography. (**C**) A microcatheter for coil embolization was utilized via the posterior communicating artery. A microcatheter for stenting was utilized via the left ICA. (**D**) A microcatheter was jailed in the aneurysm with an Enterprise stent using the semi-jailing technique. (**E**) Immediate angiography showed CO. (**F**) Angiography after rerupture on postoperative day 1 showing a newly contrasted lesion on the side of the coil mass (arrowhead). CO: complete obliteration; H&K: Hunt and Kosnik; ICA: internal carotid artery; SAC: stent-assisted coiling; SAH: subarachnoid hemorrhage

the first retreatment; they were treated with PAO in the second retreatment. Two patients who were treated with overlapping stents did not experience rerupture or regrowth. A representative case is shown in **Fig. 2**.

As an initial treatment in the acute phase of SAH, PAO was performed in 2 patients. One patient (case 11) of severe cerebral vasospasm was difficult to manage, which resulted in a poor prognosis. Moreover, one reruptured case (case 5) had severe primary brain damage due to SAH,

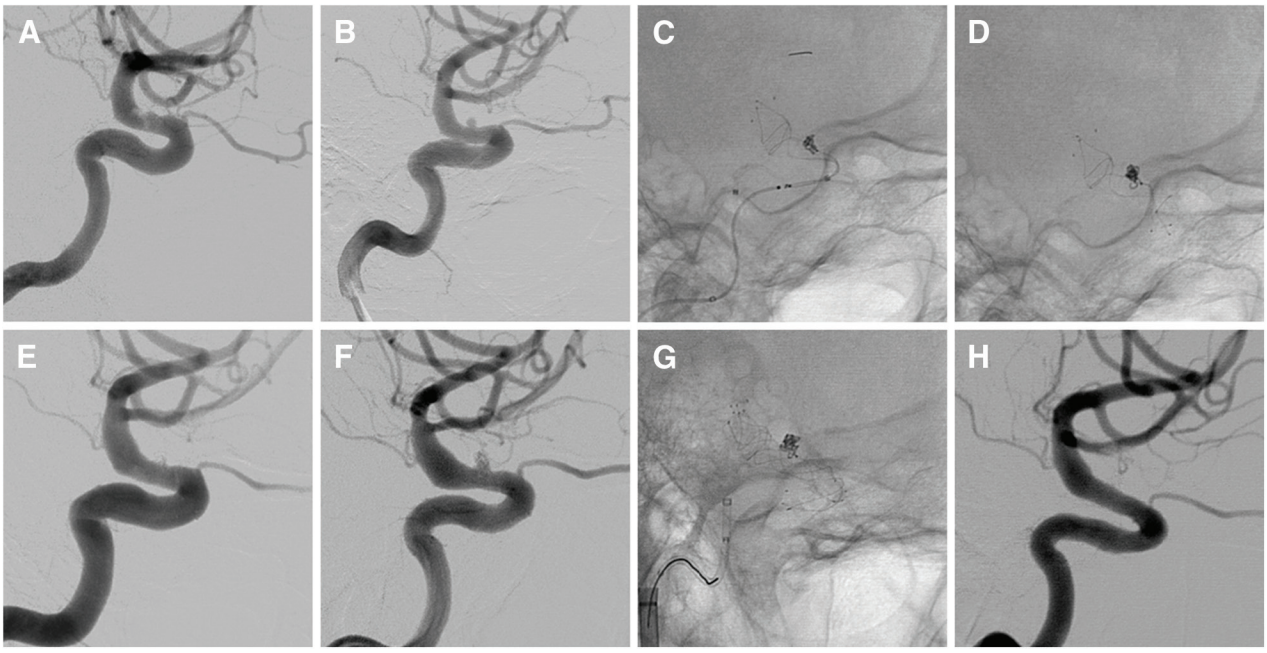

Fig. 2 Case 8 involved a 54-year-old woman with H&K grade 2 SAHs treated with overlapping stent. (**A**) 3D reconstruction image of initial angiography showing an irregular bulge at the anterior wall of the right ICA. (**B**) Follow-up angiography at the 3rd postoperative day revealing an enlargement of the irregular bulging portion. (**C**) During the operation, a microcatheter was jailed in the sac neck with an LVIS stent using the semi-jailing technique. (**D**) Soft coils were inserted, and the stent was deployed to increase the metal coverage density of the sac neck. (**E**) Postoperative angiogram showing NR. (F) Follow-up angiography on the 13th postoperative day revealing a regrowth of the aneurysm. (**G**) Additional stent deployment was performed using a second LVIS stent. (**H**) Follow-up angiogram on the 3rd month postoperative showing CO. H&K: Hunt and Kosnik; ICA: internal carotid artery; NR: neck remnant; SAH: subarachnoid hemorrhage

which resulted in poor prognosis. PAO was performed in 3 patients as the first or second retreatment after the acute phase of SAH. Two patients underwent single superficial temporal artery to middle cerebral artery (STA–MCA) bypass, whereas 1 patient underwent double STA–MCA bypass in combination with PAO. Symptomatic infarction occurred in 1 patient.

Mean duration of admission was 36 days (range: 6–106 days). Clinical data revealed discharge mRS scores of 0–2 and 3–6 in 3 patients (27%) and 8 patients (73%), respectively. Out of 9 patients treated with SAC, 3 patients (33%) had discharge mRS scores of 0–2, whereas 6 patients (67%) had discharge mRS scores of 3–6.

Discussion

This study presents case series of BBAs treated with EVT in which rerupture occurred in 22% of cases treated with SAC and 56% were retreated with coil embolization, PAO, or overlapping stent. The case treated with overlapping LVIS stent showed good embolization results. Furthermore, 17% of patients treated with PAO had symptomatic stroke.

Endovascular treatment techniques include PAO, coil embolization, balloon-assisted coiling, SAC, overlapping stents, and flow-diverter stents. Although several different techniques have been proposed by leading centers across the world, no consensus has been reached on the treatment that could reduce mortality due to poor clinical outcomes.[9,10]

Deconstructive occlusion of the parent artery is considered effective for the prevention of postoperative growth or rebleeding of the aneurysm.[11] However, even in patients with excellent collateral circulation, inherent hypoperfusion caused by decreased cerebral blood flow or vasospasm in the acute phase may be present. In this study, 1 out of 6 patients treated with PAO experienced symptomatic infarction and 1 patient suffered from vasospasm. PAO can be an option in cases where reconstructive therapy is difficult, but the indications should be carefully evaluated, especially in the acute phase of SAH. Bypass surgery can be one option for PAO.[12] Favorable outcomes have been reported with high-flow bypass and trapping; however, the technique is not easy.[13]

Restricted reports suggest that coil embolization with or without the balloon-assisted technique could be an

effective treatment for selective saccular-shaped BBAs.[14] However, coil embolization is potentially hazardous and should not be considered for the treatment of BBAs because of the pathogenesis of these aneurysms. With the application of stents, endovascular reconstructive therapy has gradually become the dominant treatment method for ruptured BBAs.

SAC has been reported to be an effective alternative treatment option. The placement of coils into the saccular portion of BBAs increases the risk of aneurysm perforation. Therefore, the smaller and softer coils are favorable for insertion, and the semi-jailing technique constrains the coil mass and allows denser packing without premature rupture.[15] Previous reports utilized low-porosity neck bridge stents such as Enterprise or Neuroform (Stryker, Kalamazoo, MI, USA) stents, which may have resulted in the need for more stents to increase the metal coverage area. LVIS stents have higher metal coverage than laser-cut stents. Zhu et al.[16] reported that the LVIS stent was less likely to result in recanalization than non-LVIS stents. However, no significant difference was observed in the complication rate of both groups. In addition, applying the bulging technique with the LVIS stent may increase metal coverage in the sac neck.[17]

Single SAC often cannot provide enough flow diversion effect, leading to regrowth or rerupture.[18] In our cases, rerupture occurred in 2 cases treated with single SAC. Overlapping stents can also provide a flow diversion effect, which reduce the amount of intra-aneurysmal flow and shear stress on the aneurysmal wall.[19] A multicenter study showed that BBAs treated with multiple stents resulted in a higher obliteration rate, lower recurrence rate, and lower perioperative hemorrhagic risk.[20] Some reports showed that the use of overlapping stent without coiling achieved complete healing. It is thus an option in cases where coil implantation is difficult.[21,22]

For acute SAH, the use of antiplatelet medications concomitant with the use of stents is a major concern. A dual antiplatelet regimen to avoid thrombotic complications is needed when patients are treated with SAC. Symptomatic cerebral infarction due to SAC was not experienced; however, rebleeding after SAC occurred in 2 patients. A multicenter study showed that the perioperative ischemic and hemorrhagic morbidity rates of patients treated with 1 and 2 stents were 3.9% versus 10.4% and 9.8% versus 2.6%, respectively.[20] Individualized platelet function assessment and precise antiplatelet treatment may be maintained to balance the risks of hemorrhagic and ischemic complications.[23]

Retreatments were performed in 5 patients treated with SAC as the first intervention. Retreatments included coil embolization, PAO, and overlapping stents. Although coil embolization can possibly cause regrowth, it is an effective method to overcome the acute phase. PAO has poor results as an acute treatment; however, it can be effective after the acute phase when performed in combination with bypass in patients who have poor collateral flow. As mentioned above, the overlapping stent has been reported to have good results and seems to be an effective method.

Retreatments (coil embolization in 2 cases, PAO in 2 cases, and overlapping stents with LVIS stent in 1 case) were performed in 5 patients. Recurrence occurred in 2 patients who were treated with coil embolization as the first retreatment; they were treated with PAO in the second retreatment.

The application of flow diverter stents and covered stents has gradually increased in recent years.[24,25] A systematic review found that 72% of complete occlusion can be achieved using flow diverter stents.[26] However, flow diverters have unignorable reports of a low initial occlusion rate and early rebleeding.[27] Another ongoing concern is the off-label application of neck bridge stents or flow diverter stents for ruptured aneurysm.

The main limitations of this study are its small sample size, single-center retrospective design, and lack of statistical analysis. In addition, the study was not standardized or randomized. Over the long period included in this study, endovascular devices have changed significantly. In fact, several different types of stents were used in this study. The results of this study do not directly represent the current results of SAC. Future studies involving quantitative analyses could provide more detailed and conclusive information.

Conclusion

In this small series, our data support the use of SAC as an effective treatment for BBA. After SAC for BBA, careful follow-up is necessary, with sufficient attention to recurrence and rerupture. To determine the best treatment option for BBA, more comparison studies are needed.

Funding

This study was supported by grants from JSPS KAKENHI (18K08996 and 18H02913).

Disclosure Statement

All authors have no conflicts of interest to declare.

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