

Cerebral vasospasm resulted in “stent shortening” after pipeline assisted coil embolization for blood blister aneurysms

Ting Wang, MD, PhD^a, Seidu A. Richard, MD, PhD^{a,c}, Junrao Li, MD, PhD^a, He Jiao, MD, PhD^b, Changwei Zhang, MD, PhD^{a,*} , Chaohua Wang, MD, PhD^a, Sen Lin, MD, PhD^a, Xiaodong Xie, MD, PhD^a, Chao You, MD, PhD^a

Abstract

Blood blister aneurysms (BBAs) are intracranial arterial lesions appearing in nonbranching sites of the supraclinoid internal carotid artery as well as the basilar artery. Endovascular treatment of BBAs is still not well established because of the rarity of these lesions. We report incidences of BBAs with associated vasospasms. Treatment of the BBAs and associated vasospasms with single Pipeline Flex embolization device (PLED) assisted coiling resulted in seemingly shortening of the PLEDs in 3 patients.

A retrospective analysis of patients with BBAs who were treated with single PLED assisted coiling from July 2018 to October 2019 was conducted. Patients' ethnic and medical records, aneurysmal characteristics, intraoperative-rupture, cerebral vasospasm (CVS), postprocedure contrast filling, follow-up imaging, and results were analyzed. Neurological examination at baseline and outcome based on modified Rankin scale (mRS) at discharge as well as follow-ups were also documented.

Six patients consisting of 5 females and 1 male, with a mean age of 48.3 years (range from 34–67) were identified during our analysis. All the BBAs were located in nonbranching site of supraclinoid segment of internal carotid artery with a mean neck width of 4.5 mm and mean aneurysm size of 4.23 mm. PLED assisted coiling's were performed in all of them. CVS was observed in 3 patients while 1 patient had an intraoperative-rupture of the BBA. Postprocedure contrast filling was still present in 1 patient. All the patients had good outcomes with discharge and follow-up mRS scores ≤ 2 except 1 patient with mRS score = 3 with aphasia.

PLED assisted endovascular coiling is very safe and efficient in treating patients with BBAs. Resolution of CVSs after treatment made the PLEDs seemingly shorter. Furthermore, a combination of arterial lengthening after gaining their tensile strength back and radical expansion of the PLED could account for the shortening.

Abbreviations: BBAs = blood blister aneurysms, CT = computed tomography, CVS = cerebral vasospasm, DSA = digital subtraction angiography, ICA = internal carotid artery, mRS = modified Rankin scale, PLED = Pipeline Flex embolization device, PPCF = postprocedure contrast filling, ROC = Raymond–Roy occlusion classification, SAH = subarachnoid hemorrhages, TCR = trigeminocardiac reflex.

Keywords: blood blister aneurysms, cerebral vasospasm, coils, endovascular, Pipeline Flex embolization device, surgery

1. Introduction

Blood blister aneurysms (BBAs) are intracranial arterial lesions appearing in nonbranching sites of the supraclinoid internal carotid artery (ICA) as well as the basilar artery.^[1–3] BBAs constitutes about 0.6% to 1.7% of all ICA aneurysms and also accounts for about 6.6% of all ruptured ICA aneurysms.^[3–5]

Aneurysmal subarachnoid hemorrhages (SAH) accounts for about 1% to 2% of all intracranial SAHs although they are relatively rare.^[2,6] They are often associated with subadventitial dissections leading to focal wall defect deficient in internal elastic lamina as well as media and the arterial gap is only enclosed by adventitia as well as thin fibrinous tissue.^[1,7–9]

Editor: Maya Saranathan.

This work was financially supported by National Nature Science Foundation of China (No: 81701174).

The authors have no conflicts of interests to disclose.

The datasets generated during and/or analyzed during the current study are not publicly available, but are available from the corresponding author on reasonable request.

^a Department of Neurosurgery, West China Hospital, Sichuan University, 37 Guo Xue Xiang Street, Chengdu, P. R. China, ^b Department of Radiology, West China Hospital, Sichuan University, 37 Guo Xue Xiang Street, Chengdu, P. R. China, ^c Department of Medicine, Princefield University, P. O. Box MA 128, Ho-Volta Region, Ghana.

* Correspondence: Changwei Zhang, Department of Neurosurgery, Postgraduate Training Centre, West China Hospital, Sichuan University, 37 Guo Xue Xiang Road, Chengdu, Sichuan 610041. P. R. China (e-mail: 68646075@qq.com).

Copyright © 2021 the Author(s). Published by Wolters Kluwer Health, Inc.

This is an open access article distributed under the terms of the Creative Commons Attribution-Non Commercial License 4.0 (CCBY-NC), where it is permissible to download, share, remix, transform, and build upon the work provided it is properly cited. The work cannot be used commercially without permission from the journal.

How to cite this article: Wang T, Richard SA, Li J, Jiao H, Zhang C, Wang C, Lin S, Xie X, You C. Cerebral vasospasm resulted in “stent shortening” after pipeline assisted coil embolization for blood blister aneurysms. *Medicine* 2021;100:33(e26971).

Received: 15 April 2020 / Received in final form: 18 June 2021 / Accepted: 1 August 2021

<http://dx.doi.org/10.1097/MD.00000000000026971>

Surgical treatment of BBAs is particularly challenging because they are pseudoaneurysms with thin and fragile walls as well as absence of a definable necks.^[2,10] They are highly prone to intraoperative rupture as well as postoperative hemorrhage, leading to major morbidities as well as mortality from surgical intervention.^[4,5,11,12] Endovascular treatment with balloons, coils, stents as well as stent-assisted coil embolization have proven to be safe and efficient alternative treatment options.^[2,3,10] Nevertheless, challenges are still encountered with endovascular treatment because of the small size as well as unfavorable shape of BBAs.^[3] A very rare complication associated with surgical or endovascular therapy for aneurysm is the trigeminocardiac reflex (TCR).^[13,14]

Coil embolization has been used as treatment for BBAs with resultant complications such as intraprocedural rupture and early interval recurrence of the BBAs with SAH.^[2,15,16] Currently, stent-assisted coil embolization has proven to be effective for stable occlusion of BBAs.^[2,17] Nevertheless, endovascular treatment of BBAs is still not well established because of the rarity of these lesions. Our retrospective analysis demonstrated safety and efficiency of stent-assisted coil-embolization technique in management of BBAs. Nevertheless, we observed incidences of BBAs associated cerebral vasospasms (CVSs). We further observed that, this treatment option also relieved BBA associated CVSs resulting in seemingly shortening of the Pipeline Flex embolization devices (PLEDs).

2. Materials and methods

2.1. Methods

This retrospective study was approved by our Institutional Research Review Board. All the patients as well as their relatives involved in this study were dually informed about our intention to involve them in a study and they totally consented to the use of their information. A written informed consent was signed by all the patients.

2.2. Blood blister aneurysms diagnostic criteria

- 1) Typical clinical presentation of SAH.
- 2) Computed tomography (CT) confirmed SAH.
- 3) The aneurysm located in nonbranching site of supra-clinoidal segment ICA on digital subtraction angiography (DSA) imaging.
- 4) Accompanied with stenosis or dilation of parent artery on DSA.
- 5) Aneurysmal shape changes obvious on 2 different angiograms in a short time.
- 6) CVS was obvious in pre- or intraoperation imaging.
- 7) Poor contrast filling in aneurysmal sac.

Criteria 1 to 3 were constant while criteria 4 to 7 must be met at least once.

2.3. Patients

Patients with BBAs who were treated with single PLED (Medtronic Neurovascular, Irvine, CA) assisted coiling from July 2018 to October 2019 were included in this study. All patients in this study were embolized with single PLED and coils. In all patients, we used 20% (20 mL contrast and 80 mL normal saline mixture) dilute contrast enhanced high-resolution Xper-CT (Philips, Best, The Netherlands) to monitor the length and shortening of PLED. Patients' ethnic and medical records,

aneurysmal characteristics, intraoperative-rupture, CVS, post-procedure contrast filling (PPCF), follow-up imaging, and result were documented and analyzed. Neurological examination at baseline and outcome based on modified Rankin scale (mRS) at discharge as well as follow-ups were also documented and analyzed. The Bouthillier classification of ICA segments was utilized to establish the location of the BBAs. The Raymond–Roy occlusion classification (ROC) was used to classify aneurysmal occlusion into 3 groups: complete occlusion (ROC1), neck remnant (ROC2), as well as residual aneurysm (ROC3).

2.4. Antiplatelet medication regimen

All patients were given a loading dose of 300 mg of aspirin and 225 mg of clopidogrel at least 2 hours prior to their procedures. Per our protocol, dual-antiplatelet therapy was maintained up to 6 months after the procedures. Aspirin alone was continued for an extra 6 months after the dual-antiplatelet therapy. The total duration of aspirin therapy after the endovascular procedure was 12 months.

2.5. Selection of pipeline flex embolization device

We used 3D reconstructed images to determine the dimensions of the parent arteries as well as the dimensions of the aneurysms. Subsequently we used the FD-20 software (Philips, Best, The Netherlands) to determine the size of the PLED for each patient according to the dimensions of the parent arteries.

2.6. Endovascular treatment

All patients underwent DSA prior to their endovascular procedures. We utilized a triaxial system consisting of 9F short femoral sheaths, 7F 90 cm long sheaths and 5F Navien intermediate catheters to assess the petrous/cavernous segments of the ICAs. Normal saline was continuously infused to prevent any ischemic events after femoral sheaths were placed successfully. The 7F long sheaths were advanced into cervical segments of the ICAs to finish the frontal views, lateral views and three-dimensional angiographies of parent arteries. After positioning the Marksman microcatheters (Medtronic Neurovascular, Irvine, CA), echelon10 microcatheters (Medtronic Neurovascular, Irvine, CA) were advanced into aneurysmal sacs between the 7F long sheaths and 5F Navien catheters. PLEDs were then positioned and initial partial coiling's of the aneurysms done. Subsequently, the PLEDs were half released to protect parent arteries and to assist in the final coiling's of the BBAs.

PLEDs were finally fully deployed after complete coiling of the BBAs. Immediate postdeployments and coiling's, working projections of coiling's, frontal and lateral views were obtained to assess PPCFs. Subsequently, 20% dilute contrast enhanced Xper-CTs were performed to determine the location of the distal and proximal end of the PLEDs. Arteries with vasospasms were noted intraoperatively. Angiographies were used to determine the lengths of the entire C6 or C7 segments as well as shortening of the PLEDs and coils in-situ.

2.7. Follow-up angiography

All patients were scheduled for DSA follow-ups, and average angiography follow-ups time was 3.5 months (range from 2–6 months). Frontal and lateral angiographies and 20% dilute contrast enhanced Xper-CTs were performed in all cases to assess

Table 1

Patient demographics, aneurysmal characteristics, intra-procedure complications, and follow-up results.

No	Hunt-Hess grade	Fisher grade	TOSA	Ethnicity	Hypertension	Diagnostic criteria	Coil	PLED size	Location	Neck (mm)	Aneurysm		Intraoperative rupture	PPCF	mRS at discharge	Image FU (month)	Image result	mRS at FU	Stent shortening
											mean size (mm)	CVS							
1	II	II	18 h	Han	N	1 to 3, 4	Y	4.75 × 35	L-C6	6.3	7.3	N	N	Y	1	6	ROC3	0	N
2	III	II	24 h	Tibetan	N	1 to 3, 5, 8	Y	3.75 × 20	R-C7	3.2	4.65	N	Y	N	2	3	ROC1	0	N
3	III	IV	8 h	Han	N	1 to 3, 7	Y	4.75 × 20	L-C7	5.1	4.25	Y*	N	N	2	9	ROC3 ^a	0	Y
4	II	IV	30 h	Han	N	1 to 3, 5, 6	Y	3.25 × 30	L-C6	3.2	2.1	Y	N	N	1	6	ROC1	0	Y
5	II	IV	8 h	Han	Y	1 to 3, 6	Y	3.0 × 18	L-C7	6.9	4.9	Y	N	N	3	2	ROC1	2	Y
6	II	IV	18 h	Tibetan	Y	1 to 3, 4, 8	Y	5.0 × 20	R-C7	2.2	2.2	N	N	N	1	2	ROC1	0	N

CVS = cerebral vasospasm, CT = computed tomography, DSA = digital subtraction angiography, FU = follow-up, ICA = internal carotid artery, L = left, mRS = modified Rankin scale, N = no, PA = parent artery, PLED = Pipeline Flex embolization device, PPCF = postprocedure contrast filling, ROC = Raymond-Roy occlusion classification, R = right, SAH = subarachnoid hemorrhage, TOSA = time of onset of symptom to admission, Y = yes.

Diagnostic criteria.

- 1) Typical clinical presentation of SAH.
- 2) CT confirmed SAH.
- 3) The aneurysm located in nonbranching site of supra-clinoid segment of ICA in DSA.
- 4) Accompanied with stenosis or dilation of PA in DSA.
- 5) Aneurysm shape changed obviously in 2 different angiograms in a short time.
- 6) CVS was obvious in pre or intraoperation image.
- 7) Poor contrast filling in aneurysmal sac.

Criteria 1 to 3 were constant while criteria 4 to 8 must meet at least once.

^arecurrence of the BBA.

aneurysm occlusion, recurrence, CVS, and stent shortening prior to completion of the procedures. On follow-ups, angiographies were again used to determine the “shortening” of the PLEDs and

coils in-situ with no vasospasms. The relative position of distal end of PLEDs and the anterior choroidal arteries were used detect “shortening”.

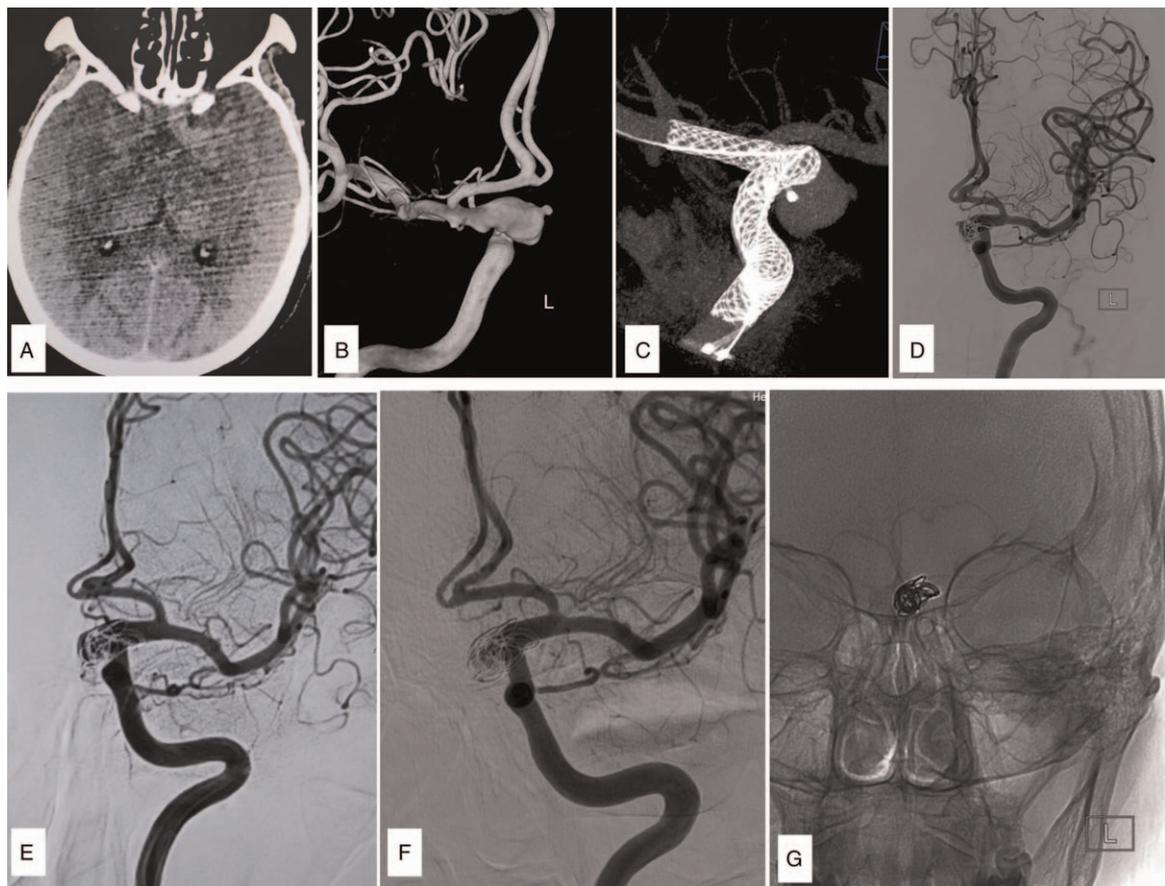


Figure 1. (case 1) A shows SAH in CT. B shows an aneurysm located in the nonbranching sites of ophthalmic segment of left ICA and dilation of the parent artery. C and D show PLED and coils in-situ which the daughter sac was still present on immediate postprocedure imaging. E to G are follow-up images. E shows disappearance of the daughter sac at discharge. F and G are 6-month follow-up images showing opacification of aneurysmal sac with coils in-situ. CT = computed tomography, ICA = internal carotid artery, PLED = Pipeline Flex embolization device, SAH = subarachnoid hemorrhage.

3. Results

3.1. Baseline characteristics

Six patients consisting of 5 females and 1 male, with a mean age of 48.3 years (range from 34–67) were identified during our analysis. The time of onset of symptoms to admission ranged from 8 hours to 30 hours. Our evaluation identified 4 patients with Hunt–Hess II while 2 had Hunt–Hess III. On CT imaging, Fisher grade IV was established in 4 patients while 2 patients had Fisher grade II. Four patients were Han Chinese while 2 were Tibetans. Hypertension was established in 2 patients (Case 5, Fig. 5; Case 6, Fig. 6) while 4 were nonhypertensive. The diagnostic criteria per patient is as shown in Table 1. In all patient, criteria 1 to 3 was met while criteria 4 to 8 were optional.

3.2. Blood blister aneurysms characteristics

All the BBA were located in the nonbranching sites of the supraclinoid segments of ICAs with a mean neck width of 4.5 mm and mean aneurysm size of 4.23 mm (Table 1). Four of the lesions were located in the C7 segments while 2 were located in the C6 segments. On the other hand, 4 BBAs were located at left side while 2 were located at right side (Table 1).

3.3. Operative outcome

PLEDs assisted endovascular coiling's were successfully performed in all of them. CVSs were observed in 3 patients (Case 3,

Fig. 3; Case 4, Fig. 4; Case 5, Fig. 5). The CVSs were severe in 2 patients and mild in 1 patient (Y*). Intraoperative-rupture of the BBA occurred in 1 patient (Case 2, Fig. 2). After the embolization procedures, PPCF was still present in 1 patient (Case 1, Fig. 1) while in 5 patients', the aneurysms occluded completely. At discharge, 3 patients had mRS score of 1, 2 patients had score of 2, while 1 patient had a score of 3 with aphasia after the operation (Table 1).

3.4. Clinical and image follow-up

Five patients had mRS score of 0 on last clinical follow-ups while 1 patient had a score of 2 with unresolved aphasia. Complete obliteration (ROC1) was observed in 4 patients while ROC3 was observed in 2 patients (Table 1). The patient with ROC3* also had a recurrence of the BBA on 9 months follow-up and was managed conservatively (Case 3, Fig. 3). Furthermore, aneurysmal sac contrast filling was still present in 1 patient (ROC3) during 6 months follow-up DSA evaluation. The CVSs resolved in 3 patients during follow-ups. The entire C6 or C7 segments were relatively much longer after the resolutions of the vasospasms (Case 3, Fig. 3; Case 4, Fig. 4; Case 5, Fig. 5). Nevertheless, stent shortenings were observed in these 3 patients. It was obvious that, the “stent shortenings” were associated with the resolutions of the CVSs and the lengthening of the arteries which seemly looks like the stents had shorten. Moreover, the stent sizes and lengths were still same on follow-up imaging's.

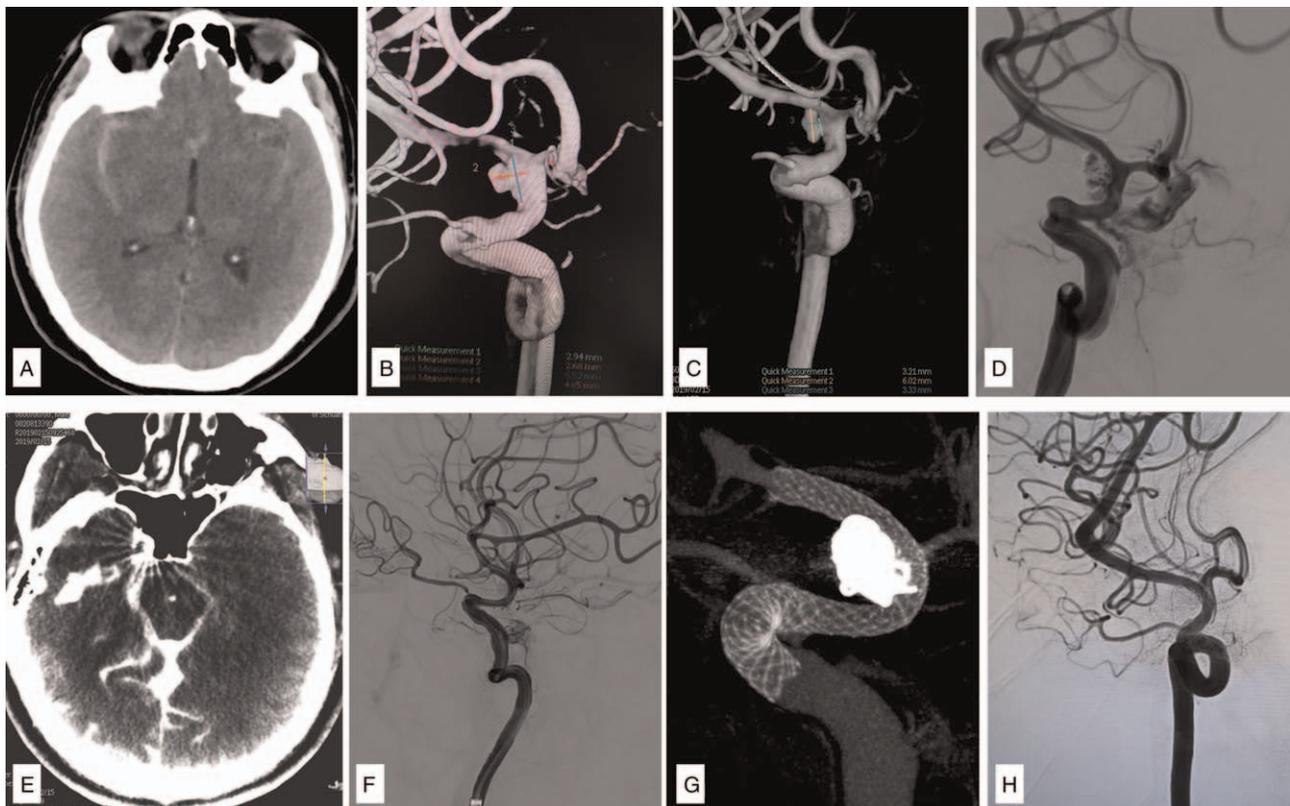


Figure 2. (case 2) A to C are preoperation CT and DSA images. A: shows SAH in CT, B and C are angiographs within an interval of 2 days, in which aneurysm growth was observed. D and E show intraoperative rupture during coiling of the BBA. E: is an intraoperative CT confirming contrast in subarachnoid space. F and G are postoperation images showing no further bleeding. G: shows the PLED and coils clearly. H is 3-month follow-up image showing no recurrence of the BBA. BBA = blood blister aneurysm, CT = computed tomography, DSA = digital subtraction angiography, PLED = Pipeline Flex embolization device, SAH = subarachnoid hemorrhages.

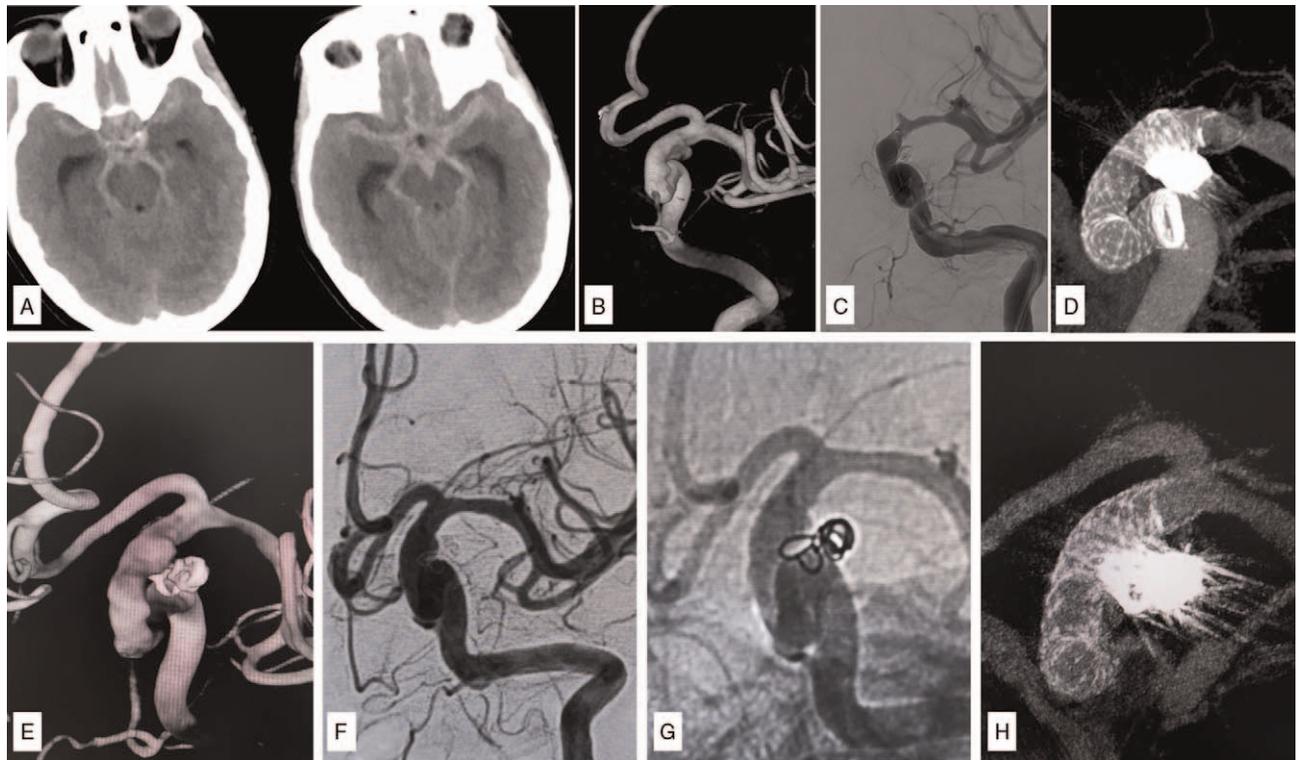


Figure 3. (case 3) A and B are preoperative images. A: displays diffused SAH with Fisher grade IV. B shows the aneurysm and poor contrast filling in aneurysmal sac. C and D are postoperation images. C: shows mild CVS with complete coiling of the aneurysm. D: shows the PLED and coils clearly. E to H are 2-month follow-up images showing recurrence (ROC3*) the BBA with compacted and dislocated coils. Furthermore, the distal end of the PLED seem "shortened" with resolution of CVS compared to C and D. BBA = blood blister aneurysm, CVS = cerebral vasospasm, PLED = Pipeline Flex embolization device, ROC = Raymond–Roy occlusion classification, SAH = subarachnoid hemorrhages.

The associated CVSs influenced the choice of PLED sizes for these 3 patients. Nevertheless, we cannot rule out the fact that the shortenings could be due to radical expansions of the devices.

4. Discussion

BBAs are small, blister-like, wide neck intracranial arterial lesions often found on the nonbranching section of the supraclinoid ICA as well as the BA. They are often associated with acute SAH with malignant possibilities.^[2,5,9,12] We identified 6 patients with BBAs consisting of 5 females and 1 male, with a mean age of 48.3 years (range from 34–67) during our analysis. All the BBAs were located in the nonbranching sites of the supra-clinoidal segments of the ICAs with a mean neck width of 4.5 mm and mean aneurysm size of 4.23 mm. In 4 of patients, the lesions were located in the C7 segments while in 2 patients the lesions were located in the C6 segments. On the other hand, 4 BBAs were located at left side while 2 were located at right side. Several studies have demonstrated that, arteriosclerosis with resultant ulceration as well as hypertension are the major mechanisms of BBA pathogenesis.^[3,5,9,18] Hypertension was established in 2 patients while 4 were nonhypertensive. Nevertheless, we did not observe arteriosclerosis or any such signs in all our patients.

The time of onset of symptoms to admission ranged from 8 hours to 30 hours and typical clinical manifestation was SAH which was established with CT scan. Thus, CT was the initial radiology evaluation modality for our BBA patients. Identification BBAs on angiography is often problematic because they often mimic other tiny intracranial lesions and can be ignored,

mistaken for artifacts or focal atheromatous anomalies, or utterly ignored because of overlap of vessel curvatures.^[2,12,19] Nevertheless, repeated angiographies often demonstrate luminal alterations or out pouching of these aneurysms. The most cardinal radiological diagnostic modalities in our study were CT scan and DSA. CT scan identified the initial associated SAH while DSA established the definitive diagnosis of BBAs.

The mRS is a 6-point disability scale with achievable scores ranging from 0 to 5.^[20] An extra separate score of 6 is frequently added for patients who expire. It is a very reliable and easy neurology assessment record for evaluating critical sick patients.^[20] Meckel et al^[2] observed a good functional outcome with mRS 0 to 2 in 10 out of 13 patients (76.9%) at discharge from the hospital and in 12 out of 13 patients (92.3%) at midterm follow-up of 6 to 27 months. Peschillo et al,^[21] in a meta-analysis involving 199 patients with BBAs who underwent endovascular treatment with embolization, observed good outcomes with mRS 0 to 2. They revealed that, 86.4% of patient treated with primary stenting recorded mRS 0 to 2, 85.2% with stent-assisted coiling recorded mRS 0 to 2 while 82.2% with flow diversions recorded mRS 0 to 2.^[21]

At discharge, 3 of our patients had mRS score of 1, 2 patients had score of 2, while 1 patient had a score of 3 with aphasia after the operation. On last clinical follow-up before our analysis, 5 patients had mRS score of 0 while 1 patient had a score of 2 with unresolved aphasia. Peschillo et al^[22] revealed that Hunt–Hess grading in their meta-analysis involving 334 patients treated with microsurgery and endovascular treatment options were associated with good outcomes. Our evaluation identified 4 patients with

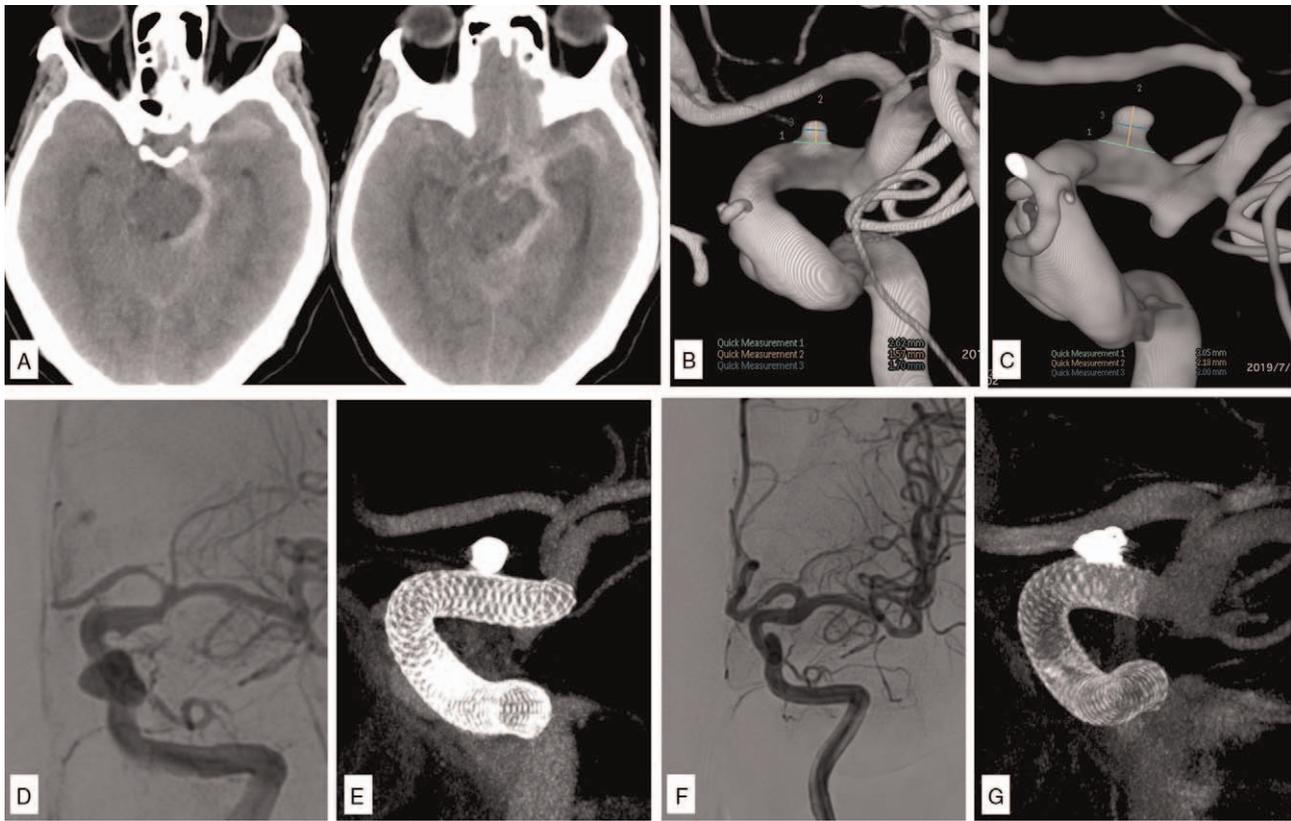


Figure 4. (case 4) A to C are preoperation images. A: show diffused SAH with Fisher grade IV. B and C are angiographs performed in 2 days intervals, showing aneurysm shape and size changes. D and E are postoperation images. D: shows complete obliteration of aneurysm and CVS. E: shows the relative position of distal end of PLED and the AchA. F and G are 6-month follow-up images. F: shows no recurrence of aneurysm and resolution of the CVS which made the PLED seemly shorter. G: shows the relative position of the distal end of PLED and the AchA have been changed. AchA = anterior choroidal artery, CVS = cerebral vasospasm, PLED = Pipeline Flex embolization device, SAH = subarachnoid hemorrhages.

Hunt–Hess II while 2 had Hunt–Hess III. Singh et al in a review indicated that, the overall outcome was most dependent on initial Hunt–Hess or Fisher grades rather than a specific therapy.^[3] On CT imaging, Fisher grade IV was established in 4 patients while 2 patients had Fisher grade II.

Surgical clipping alone may be insufficient to treat BBAs because the arterial segments the lesions originates are mostly circumferentially, making them unfit for clip application.^[9,23] In most cases, clipping often results in iatrogenic stenosis of the ICAs because some part of the parent arteries are often included in the clip configurations.^[2] Stenosis may result in flow-limiting cerebral ischemia as well as eventual infarction.^[2] Apart from direct clipping, other surgical techniques, like clipping on wrapping material, parallel clipping under hypotension, ICA trapping bypass surgery, as well as direct staple clip reconstruction of the artery, have been tried.^[9,10,12,24]

Several studies have demonstrated that, endovascular techniques with or without stent assisted coil embolization is challenging due to the small size of BBAs. Furthermore, BBAs lacks well-defined saccular component as well as necks to allow the introduction of coils safely.^[1,4,25,26] Implantation of coils into the saccular component of BBAs is usually a dangerous maneuver and may result in perforation or re-hemorrhage.^[1,27] We successfully implanted PLEDs in all our patients and assisted endovascular coiling performed. We achieved complete aneurysm occlusion (ROC1) in 4 of them. Nevertheless, in 2 patients

we observed residual aneurysm (ROC3). One of the patients with residual aneurysm (ROC3*) had a recurrence of the BBA.

Meckel et al^[2] observed immediate complete occlusion of a BBA in 4 out of 13 patients (30.8%). They did not observe delayed recanalization or recurrence of the BBAs in the 4 patients on follow-up angiographies. Furthermore, they observed residual aneurysmal neck or dog ears in 4 out of 13 and residual filling of the aneurysmal body in 4 out of 13, on posttreatment angiograms. Nevertheless, they observed recurrence or regrowth in 1 of their patient 3 weeks after the procedure which was retreated with coils.^[2] We observed postprocedural contrast filling in only 1 patient out of the 6 patients we treated with PLED assisted endovascular coiling's. Kim et al^[17] observed immediate recurrence of a BBA they treated with stent-assisted coil embolization. They retreated the patient using the stent-within-a-stent technique which provided extra support of the fragile aneurysm neck as well as reduced flow impingement as well as accelerated aneurysm thrombosis and healing.^[17] We detected recurrence or regrowth (ROC3*) of the BBA on 2 months follow-up and the patient was managed conservatively.

Meckel et al^[2] indicated that, if the BBA architecture is even more unfavorable with a neck /dome ratio of <2, a single stent may only aid in holding the coils in place but not deliver adequate extra flow-diversion to cover a very wide aneurysm neck. They further indicated that, double-stent implantation/high-attenuation meshed stents with extra coiling or even primary parent

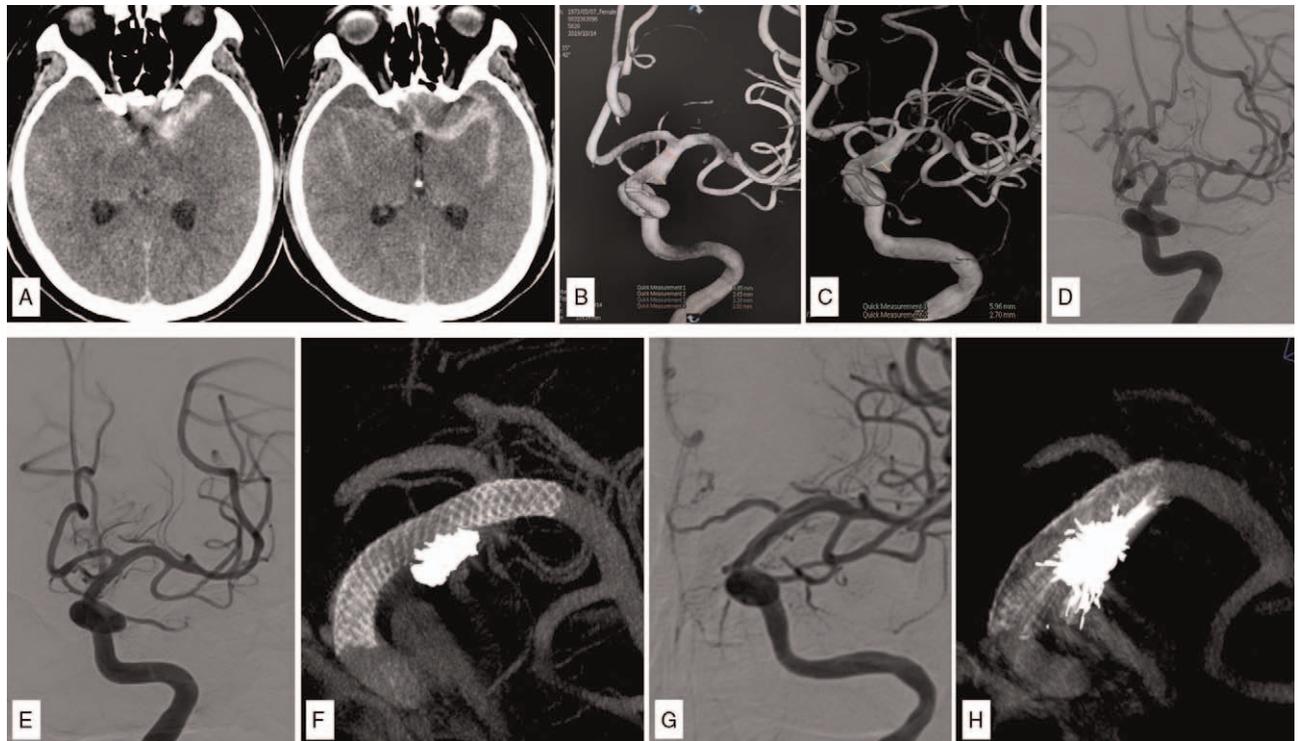


Figure 5. (case 5) A to D are preoperation images. A: shows diffused SAH. B and C show a broad neck aneurysm while C: shows CVS. D is a working projection which displays the aneurysm and CVS well. E and F are postop images. E: shows complete obliteration of aneurysm on immediate postoperative angiography. F: shows the relative position of the distal end of the PLED and ACA. G and H are 2-month follow-up images. G: shows no recurrence of aneurysm and resolution of the CVS which made the PLED seemly shorter. H: shows the seemly short PLED clearly. ACA = anterior cerebral artery, CVS = cerebral vasospasm, PLED = Pipeline Flex embolization device, SAH = subarachnoid hemorrhages.

artery occlusion in very long-segmental lesions should be adapted.^[2] Intraoperatively, after well positioning of PLED, partially coiling of the aneurysm was done and PLEDs were half released to protect the parent arteries as well as assist in further coiling. When coiling's were completed, PLEDs were deployed fully to cover the aneurysmal defects or necks. Russin et al^[28] observed an overall intraoperative rupture risk of about 28% in prediagnosed patients and about 57% in undiagnosed patients with BBAs undergoing surgical treatment. Intraoperative-rupture of the BBA occurred in 1 patient with stent assisted coil embolization therapy.

Chalouhi et al^[29] reported spontaneous delayed migration or shortening of the PLEDs after the treatment of saccular aneurysms. In one of their cases, they indicated that, the PLED moved distally due to shortening because it was perhaps well anchored distally than it was proximally.^[29] It's the PLED that shorten or the normalization of the vascular architecture that seemly make the PLED shorter? We are very sure that, the vascular architecture changes during the formation of the aneurysm on the branching or nonbranching artery. This architectural change often results in shortening of the parent artery harboring the aneurysm. After treating the aneurysms with PLED assisted coiling's, the shorten vessels regain their normal tensile strengths and lengthens making the implanted PLEDs seemly look shorter.

Outpouching of the vessel will contract both proximal and distal ends making the vessel shorter than its original length. It will be very difficult if not impossible for an implanted device to shorten. However, it is proven that, vessels contract and relax. Nevertheless, we cannot rule out the fact that the shortening could be due to radical expansions of the devices which resulted

in the shortenings. In all patients, we use utilized 20% (20mL contrast and 80mL normal saline mixture) diluted contrast enhanced high-resolution Xper-CT to monitor the lengthening of the disease arteries and observed the seemly shorting of PLEDs.

Another very rare complication associated with surgical or endovascular therapy for aneurysm is the TCR.^[13,14] Although the pathophysiological mechanism of TCR is still a matter of debate, sympathetic overactivity, local catecholamine excess, parasympathetic stimulation as well as concurrent stimulation of both branches of the autonomic nervous system have been implicated.^[13,14] Also, increases ICP as a result of SAH has been implicated.^[14] TCR usually resolves after the stimuli that triggered it was removed.^[13,14] Also, 1 mg of atropine and 50 mg of ephedrine were capable of resolving the first case of TCR reported in literature.^[13] None of our patients experienced this complication during and after treatments. However, we advocate that, patients who undergo this treatment modality be followed for a long period of time.

5. Conclusion

PLED assisted endovascular coiling is a very safe and efficient treatment option for patients with BBAs. In our cases with CVSS, the associated CVSSs influenced the selections of PLED sizes. We therefore advocate that, in patients with BBAs with associated CVSSs, the PLED sizes should be a little bigger than the normal calculated sizes to allow for arterial lengthening and wall apposition after resolution of the CVSSs. A combination of arterial lengthening after gaining their tensile strength back and radical expansion of the PLEDs could account for the shortenings.

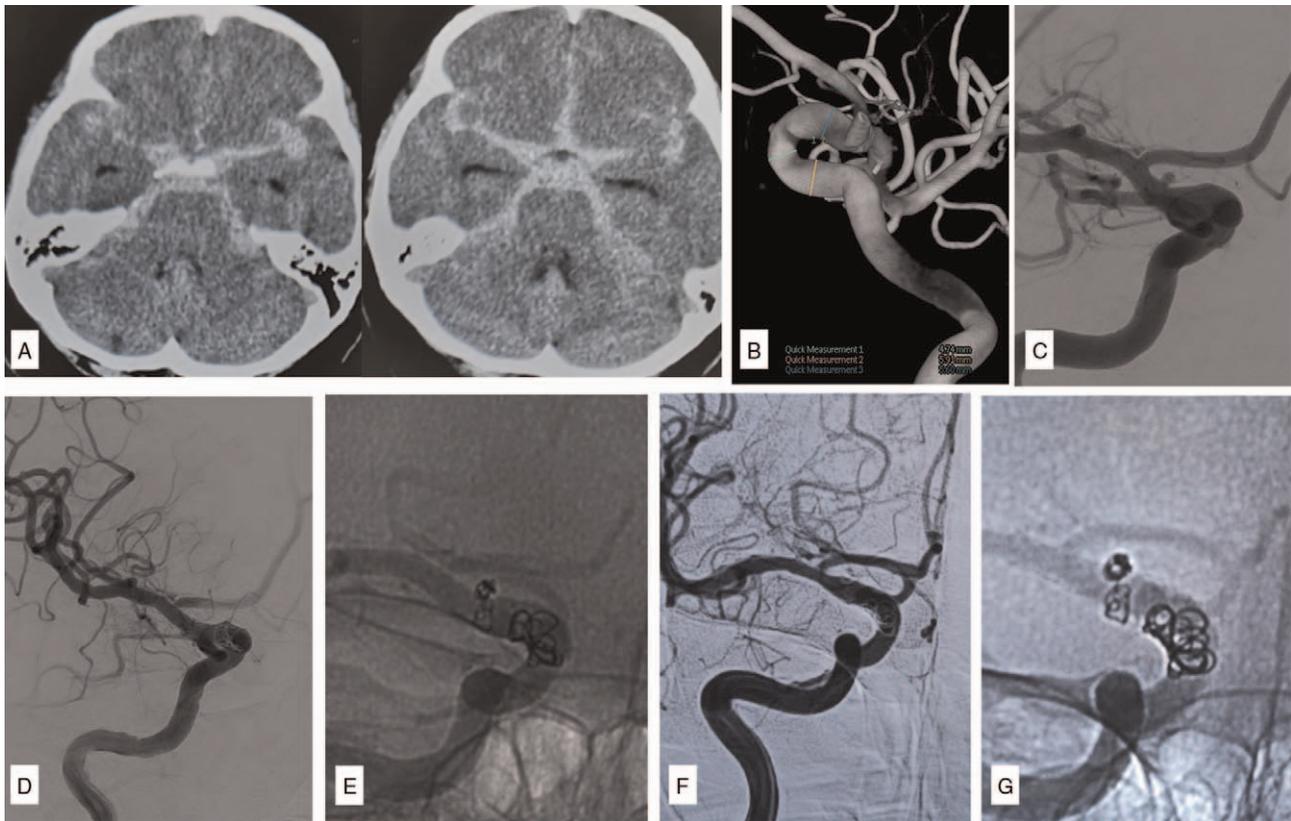


Figure 6. (case 6) A to C are preop images. A: shows diffused SAH. B and C show dilation of the parent artery and the BBA. D and E are postoperative images, showing complete obliteration of the BBA. F and G are 2-month follow-up images. F: shows complete occlusion of the BBA. G displays the PLED and coils. BBA = blood blister aneurysm, PLED = Pipeline Flex embolization device, SAH = subarachnoid hemorrhages.

Author contributions

All the author contributed to conception, design, acquisition of data, analysis, and interpretation of data. Initial drafting of the article was by SAR & TW but all authors contributed in revising it critically for important intellectual content. All authors approved the final version to be published and agreed to be accountable for all aspects of the work including accuracy and integrity of any part of the work.

Conceptualization: Ting Wang, Seidu A Richard, Junrao Li, He Jiao, Zhang Changwei, Chaohua Wang, Sen Lin, Xiaodong Xie, Chao You.

Data curation: Ting Wang, Seidu A Richard, Junrao Li, He Jiao, Zhang Changwei, Chaohua Wang, Sen Lin, Xiaodong Xie, Chao You.

Formal analysis: Ting Wang, Seidu A Richard, Junrao Li, He Jiao, Zhang Changwei, Chaohua Wang, Sen Lin, Xiaodong Xie, Chao You.

Funding acquisition: Zhang Changwei.

Methodology: Ting Wang, Seidu A Richard, Junrao Li, He Jiao, Zhang Changwei, Chaohua Wang, Sen Lin, Xiaodong Xie, Chao You.

Resources: Zhang Changwei, Chaohua Wang, Xiaodong Xie.

Supervision: Zhang Changwei, Xiaodong Xie.

Writing – original draft: Ting Wang, Seidu A Richard.

Writing – review & editing: Ting Wang, Seidu A Richard, Junrao Li, He Jiao, Zhang Changwei, Chaohua Wang, Sen Lin, Xiaodong Xie, Chao You.

References

- [1] Rouchaud A, Brinjikji W, Cloft H, Kallmes DF. Endovascular treatment of ruptured blister-like aneurysms: a systematic review and meta-analysis with focus on deconstructive versus reconstructive and flow-diverter treatments. *Am J Neuroradiol* 2015;36:2331–9.
- [2] Meckel S, Singh T, Undren P, et al. Endovascular treatment using predominantly stent-assisted coil embolization and antiplatelet and anticoagulation management of ruptured blood blister-like aneurysms. *Am J Neuroradiol* 2011;32:764–71.
- [3] Singh P, Patel A, Mack WJ. Blister aneurysms: decision-making when there's no perfect answer. *Endovasc Today* 2018;18:80.
- [4] Meling TR, Sorteberg A, Bakke SJ, Slettebø H, Hernesniemi J, Sorteberg W. Blood blister-like aneurysms of the internal carotid artery trunk causing subarachnoid hemorrhage: treatment and outcome. *J Neurosurg* 2008;108:662–71.
- [5] Abe M, Tabuchi K, Yokoyama H, Uchino A. Blood blisterlike aneurysms of the internal carotid artery. *J Neurosurg* 1998;89:419–24.
- [6] Nerva JD, Morton RP, Levitt MR, et al. Pipeline embolization device as primary treatment for blister aneurysms and iatrogenic pseudoaneurysms of the internal carotid artery. *J Neurointerv Surg* 2015;7:210–6.
- [7] Gonzalez AM, Narata AP, Yilmaz H, et al. Blood blister-like aneurysms: single center experience and systematic literature review. *Eur J Radiol* 2014;83:197–205.
- [8] Lee J-W, Choi H-G, Jung J-Y, Huh S-K, Lee K-C. Surgical strategies for ruptured blister-like aneurysms arising from the internal carotid artery: a clinical analysis of 18 consecutive patients. *Acta Neurochir* 2009;151:125.
- [9] Ogawa A, Suzuki M, Ogasawara K. Aneurysms at nonbranching sites in the supraclinoid portion of the internal carotid artery: internal carotid artery trunk aneurysms. *Neurosurgery* 2000;47:578–86.
- [10] Ren Y, Liu L, Sun H, et al. Microsurgical versus endovascular treatments for blood-blister aneurysms of the internal carotid artery: a retrospective study of 83 patients in a single center. *World Neurosurg* 2018;109:e615–24.

- [11] Nakagawa F, Kobayashi S, Takemae T, Sugita K. Aneurysms protruding from the dorsal wall of the internal carotid artery. *J Neurosurg* 1986;65:303–8.
- [12] Sim SY, Shin YS, Cho KG, et al. Blood blister-like aneurysms at nonbranching sites of the internal carotid artery. *J Neurosurg* 2006;105:400–5.
- [13] Spiriev T, Kondoff S, Schaller B. Trigemino-cardiac reflex during temporary clipping in aneurysmal surgery: first description. *J Neurosurg Anesthesiol* 2011;23:271–2.
- [14] Spiriev T, Kondoff S, Schaller B. Cardiovascular changes after subarachnoid hemorrhage initiated by the trigeminocardiac reflex—first description of a case series. *J Neurosurg Anesthesiol* 2011;23:379–80.
- [15] Park JH, Sung PI, Han DH, et al. Endovascular treatment of blood blister-like aneurysms of the internal carotid artery. *J Neurosurg* 2007;106:812–9.
- [16] Chen J, Zhang K. Endovascular treatment of blood blister-like aneurysms of the internal carotid artery. *Int J Cerebrovasc Dis* 2018;26:599–604.
- [17] Kim BM, Chung EC, Park SI, Choi CS, Won YS. Treatment of blood blister-like aneurysm of the internal carotid artery with stent-assisted coil embolization followed by stent-within-a-stent technique: case report. *J Neurosurg* 2007;107:1211–3.
- [18] Ishikawa T, Nakamura N, Houkin K, Nomura M. Pathological consideration of a “blister-like” aneurysm at the superior wall of the internal carotid artery: case report. *Neurosurgery* 1997;40:403–6.
- [19] Gaughen J, Raghavan P, Jensen M, Hasan D, Pfeffer A, Evans A. Utility of CT angiography in the identification and characterization of supraclinoid internal carotid artery blister aneurysms. *Am J Neuroradiol* 2010;31:640–4.
- [20] Olavarría VV, Brunser A, Cabral N, et al. The distribution of the modified Rankin scale scores change according to eligibility criteria in acute ischemic stroke trials: a consideration for sample size calculations when using ordinal regression analysis. *Contemp Clin Trials Commun* 2017;5:133–6.
- [21] Peschillo S, Cannizzaro D, Caporlingua A, Missori P. A systematic review and meta-analysis of treatment and outcome of blister-like aneurysms. *Am J Neuroradiol* 2016;37:856–61.
- [22] Peschillo S, Miscusi M, Caporlingua A, et al. Blister-like aneurysms in atypical locations: a single-center experience and comprehensive literature review. *World Neurosurg* 2015;84:1070–9.
- [23] Ding D, Starke RM, Hope A, Brew S. Flow-diverting stent-assisted coil embolization of a ruptured internal carotid artery blister aneurysm with the pipeline flex embolization device. *J Neurosci Rural Pract* 2017;8:664.
- [24] Yanagisawa T, Mizoi K, Sugawara T, et al. Direct repair of a blisterlike aneurysm on the internal carotid artery with vascular closure staple clips. *J Neurosurg* 2004;100:146–9.
- [25] Ahn JY, Cho JH, Jung JY, Lee BH, Yoon PH. Blister-like aneurysms of the supraclinoid internal carotid artery: challenging endovascular treatment with stent-assisted coiling. *J Clin Neurosci* 2008;15:1058–61.
- [26] Lim YC, Kim BM, Suh SH, et al. Reconstructive treatment of ruptured blood blister-like aneurysms with stent and coil. *Neurosurgery* 2013;73:480–8.
- [27] Walsh KM, Moskowitz SI, Hui FK, Spiotta AM. Multiple overlapping stents as monotherapy in the treatment of ‘blister’ pseudoaneurysms arising from the supraclinoid internal carotid artery: a single institution series and review of the literature. *J Neurointerv Surg* 2014;6:184–94.
- [28] Russin JJ, Kramer DR, Thomas D, et al. The importance of preoperative diagnosis of blister aneurysms. *J Clin Neurosci* 2015;22:1408–12.
- [29] Chalouhi N, Tjoumakaris S, Gonzalez L, et al. Spontaneous delayed migration/shortening of the pipeline embolization device: report of 5 cases. *Am J Neuroradiol* 2013;34:2326–30.