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Blister aneurysms of the internal carotid artery: Surgical treatment and management outcome from a single center experience

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ABSTRACT

Objective: Blood-blister aneurysms (BBAs) of the internal carotid artery (ICA) are rare entities, but clinically important cause of subarachnoid hemorrhage (SAH). Several surgical and endovascular strategies have been attempted for these heterogeneous lesions. In this study, the authors analyzed the treatment strategy and outcomes in a series of cases of ICA blister aneurysms treated microsurgically.

Patients and methods: We retrospectively reviewed 15 consecutive cases of patients harboring ruptured BBAs, microsurgically treated at our institution between 2014 and 2018. We performed an analysis of the clinical and surgical aspects, as well as post-operative angiograms and outcomes.

Results: Fifteen patients were identified; 9 (60%) were female. The mean age of presentation was 43,8 years. Most patients presented in good clinical conditions (Hunt-Hess 1–3 = 86%). The most common Fisher grade at presentation was 3 (60% of cases). All patients underwent digital subtraction angiography (DSA), revealing broad-based aneurysms at non-branching sites on the dorsal wall of the ICA. Intraoperatively, BBAs were confirmed in all cases. The lesions were approached through pterional (11–73%) or lateral supraorbital (4–27%) craniotomy. Direct clipping was performed in all but one lesion, in which case the clip-wrapping technique was used. Final angiographic control revealed complete occlusion in 14 cases. One patient required reoperation due to residual aneurysm filling. At discharge, a good outcome (Glasgow Outcome Scale [GOS] 4 or 5) was observed in 12 (80%) patients. Three patients were discharged with a GOS of 3.

Conclusion: Blood-blister-type aneurysms are rare and challenging lesions. Preoperative knowledge and careful surgical planning can prevent poor clinical outcomes. Surgical treatment remains an effective and safe option in this context.

1. Introduction

Blood-blister aneurysms (BBA) from Internal carotid artery (ICA) are a subgroup of small intracranial aneurysms with extremely thin and fragile walls and a poorly defined broad neck [1–3]. These lesions usually appear on the dorsal wall of the internal carotid artery (ICA) [1]. They are a rare entity and represent 0.3%–1% of intracranial aneurysms and 0.9%–6.5% of ICA aneurysms. BBAs lead to a higher morbidity and mortality rates when compared to ruptured ICA saccular aneurysms [4,7]. Most saccular aneurysms arise at the branching points of the ICA [8,9], whereas BBAs usually originate directly from the vessel wall, unrelated to arterial branches [10]. These aneurysms were first described by Sundt and Murphey [11] in 1969 and later studied in more detail by Nakagawa et al. [3] in 1986 with the development of microsurgery.

The preoperative diagnosis of these lesions can be challenging [12]. Digital subtraction angiography (DSA) visualizes the lumen of the aneurysm instead of its wall. Thereafter, it is impossible to determine the aneurysmal tissue that must be incorporated into the microsurgical reconstruction [13]. Regarding preoperative planning, ICA dorsal wall aneurysms should be considered as a BBA until proven otherwise.

Despite decades of advances in microsurgical and endovascular techniques, BBAs remain difficult to manage without a well-established and reliable management strategy. Their reduced size, broad base and

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thin or nonexistent walls make their conventional treatment with microsurgical clipping or endovascular techniques difficult. Currently, there is no general consensus on the safest method of treatment for BBAs [5,14].

In the present study, we analyzed the clinical characteristics and surgical results in a series of 15 consecutive cases of patients harboring BBAs in non-branching ICA sites in order to obtain a better understanding of the clinical nature and optimal treatment for these aneurysms.

2. Patients and methods

The study consists of a series of cases diagnosed and treated as Blister-type aneurysms during the period from 2014 to 2019 at the Neurosurgery center of the Hospital da Restauração - PE. This retrospective study was approved by our institutional review board, and informed patient consent was not required. Medical records, radiological examinations and surgical videos were analyzed retrospectively. Blood blister aneurysms (BBAs) were defined by abnormal focal arterial dilatation in the presence of SAH, related to non-branching points in the dorsal wall of the ICA. Morphological findings were confirmed by angiographic study and direct intraoperative observation.

The diagnostic criterion applied was aneurysm at a non-branching site of the supraclinoid internal carotid on angiography, most often in its dorsal wall. In our series, we included only patients with ruptured BBAs who met these inclusion criteria, as confirmed by operative findings. Any patients with unruptured aneurisms, or located at branching sites of the ICA, such as carotid-ophthalmic, ICA bifurcation, cavernous portion of ICA, IC-superior hypophyseal artery were excluded.

The pre and post-operative assessment were performed using the Hunt-Hess (HH) and the Glasgow Outcome Scale (GOS) at discharge, respectively. All pre-operative cranial computed tomography (CT) were analyzed and the grade of subarachnoid hemorrhage (SAH) was evaluated using the Fisher scale. Data such as sex and age were also measured. All patients were treated microsurgically by direct clipping or clip-wrapping. The surgical approach and the definitive method of treatment were also considered in the analysis. Postoperative angiography was performed in all cases in order to confirm the aneurysm exclusion and also analyze any degree of parental vessel stenosis.

3. Results

Overall 15 consecutive patients harboring BBAs in the supraclinoid segment of the ICA were diagnosed between 2014 and 2018 in this single-center series. We observed a prevalence of females (9 cases), with a mean age of 43.8 years (ranges from 24 to 61 years). The mean follow-up was 23.4 months (Table 1). All aneurysms were located at non-branching points of the dorsal wall of the supraclinoid ICA.

Most patients presented in good clinical conditions (2 HH grade 1, 8 HH grade 2 and 3 HH grade 3). Two patients presented in HH grade 4 and there was no patient who presented in HH grade 5 in our series.

Table 1
Most prevalent clinical and radiological characteristics of patients undergoing clipping and the clip-wrapping in this series; HH- Hunt-Hess scale; GOS - Glasgow Outcome Scale.

Variable	Result
Female gender	60.0%
Mean age	43.8 years
Mean HH	2,33
HH 1-3	86%
Fisher grade III	60%
Mean GOS on discharge	4,13

Table 2
: Clinical and radiological characteristics of the patients submitted to clipping and to the clip-wrapping in this series; HH- Hunt-Hess scale; GOS - Glasgow Outcome Scale.

	Age	Gender	Fisher	Hunt-Hess	Treatment	GOS on discharge
1	50	F	3	2	Clipping	4
2	61	M	3	1	Clipping	4
3	43	F	3	2	Clipping	4
4	51	F	3	3	Clipping	4
5	47	F	3	3	Clipping	5
6	54	M	3	2	Clip Wrapping	3
7	42	F	1	2	Clipping	4
8	38	M	3	1	Clipping	5
9	33	F	1	3	Clipping	4
10	48	F	3	2	Clipping	5
11	34	M	4	4	Clipping	3
12	40	F	4	2	Clipping	5
13	47	F	3	4	Clipping	3
14	24	M	4	2	Clipping	5
15	46	M	2	2	Clipping	4

Admission cranial CT scan showed Fisher 3 bleeding pattern in most of the patients (9 cases – 60%), whereas Fisher grade 1 was observed in 2 patients (13%). SAH associated with intraventricular hemorrhage (Fisher grade 4) was seen in 2 (13%) additional patients. Endovascular treatment was not considered as the first alternative in any of the cases and all patients were treated microsurgically, usually between the 6th and 14th day post-SAH (mean 8,7 days post-SAH). Three patients were operated more than 14 days after SAH (16, 18 and 24 days after SAH). The lesions were approached through a pterional (73%) or lateral supraorbital (27%) craniotomies. Anterior clinoidectomy was performed in 8 cases (53%) in order to obtain proximal control. All lesions were treated by direct clipping except in one case in which the clip-wrapping technique was performed (Table 2). Intraoperative aneurysm rupture occurred in only two cases and did not cause post-operative neurological deficits. Post-operative DSA revealed total obliteration in 14 of the 15 aneurysms (93% of total occlusion). One patient needed reoperation for treatment of residual aneurysm.

Most patients achieved a good postoperative clinical result (GOS 4 and 5 in 80% of the cases). Three (20%) patients were discharged with significant disability (GOS 3). No surgical mortality was observed in our series. Two patients developed surgical site infections and needed reoperation and antibiotics for treatment and one patient developed a cerebrospinal fluid (CSF) fistula, requiring surgical intervention for correction.

4. Illustrative case

Patient 3 is a 43-year-old female presented with the sudden onset of severe headache, nausea, vomiting, without disturbance in consciousness. Physical exam revealed mild meningism, but no focal neurological deficits. Her condition was graded as Hunt and Hess II. Cranial CT scan revealed diffuse SAH without intraventricular hemorrhage (Fisher grade 3 bleeding) – Fig. 1. DSA revealed the presence of a broad base aneurysm at a non-branching site of the supraclinoidal ICA dorsal wall (Fig. 2 and 3). A left pterional craniotomy was performed and intraoperative confirmation of a BBA was achieved (Fig. 4). An angled clip was applied parallel to the carotid artery, deliberately including a healthy wall portion of the artery, causing a minor stenosis of the parent artery (Fig. 5). Two reinforcement clips were applied for occlusion of residual segments of the aneurysm (Fig. 6). Intraoperative Doppler ultrasound probe showed excellent flow on ICA distal to the aneurysm. Postoperative DSA confirmed aneurysm occlusion without significant ICA stenosis (Figs. 7 and 8). The patient was discharged without neurological deficits (GOS 5) on post-hemorrhage day 16.

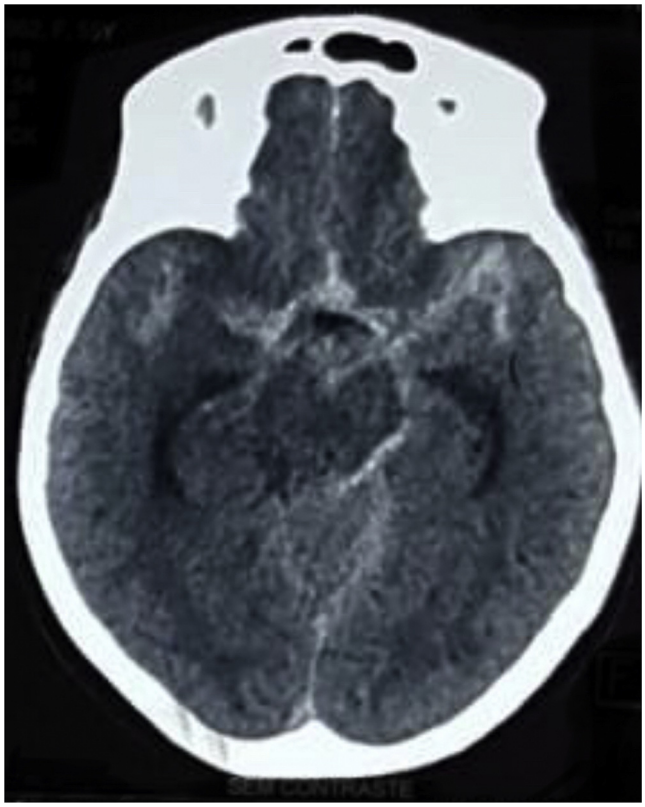

Fig. 1. Preoperative CT scan showing a Subarachnoid Hemorrhage (SHA) Fisher III.

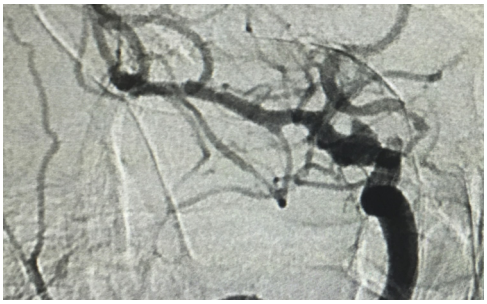

Fig. 2. Preoperative 3D angiographic reconstruction in oblique orientation demonstrated a left ICA blister aneurysm with antero-medial projection.

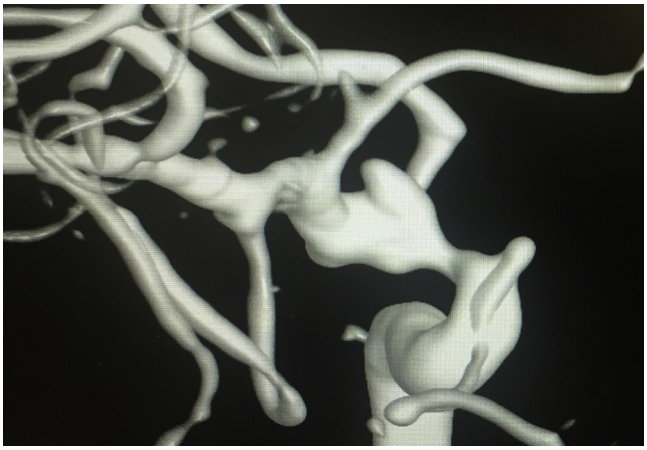

Fig. 3. Preoperative 3D angiographic reconstruction in oblique orientation demonstrated a left ICA blister aneurysm with antero-medial projection.

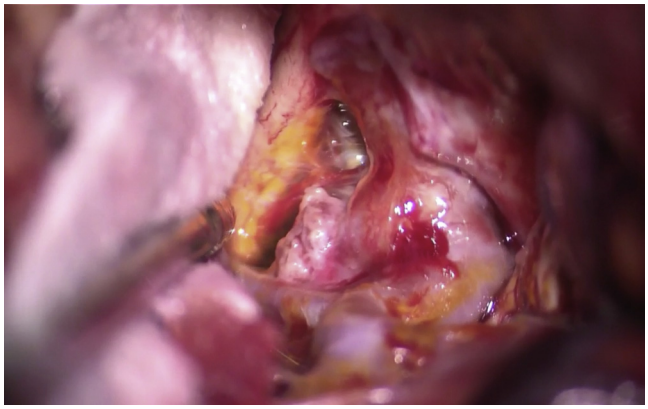

Fig. 4. The aneurysm was dissected without intraoperative rupture.

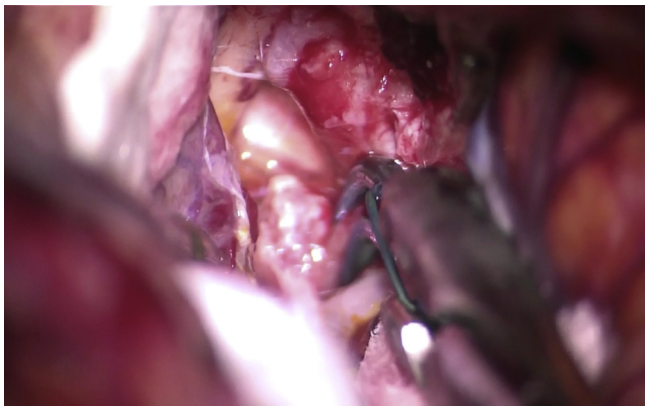

Fig. 5. Initial clipping was performed with angled clip.

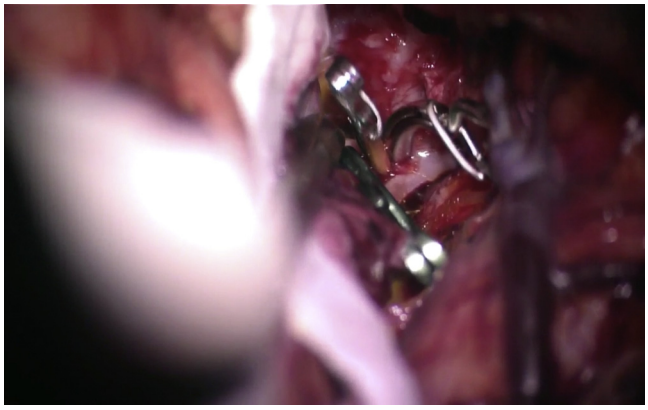

Fig. 6. Complementation of the aneurysm occlusion with clipping of its residual neck.

5. Discussion

Blister-like aneurysms are rare lesions. They present several morphological aspects which makes its treatment a real challenge. Its irregular broad base, its thin and fragile walls and its small sizes are characteristics that impose difficulty for any therapeutic method. Conservative treatment, however, is associated with high mortality rates and intervention is mandatory for these lesions [15]. Pre-operative recognition of BBA can be difficult, but is extremely important for surgical planning. Any supraclinoidal ICA dorsal wall aneurysm should be considered as a BBA until proven otherwise and microsurgical treatment should be performed as soon as possible [16–18]. Indo et al. analyzed retrospectively more than 1600 angiograms and concluded

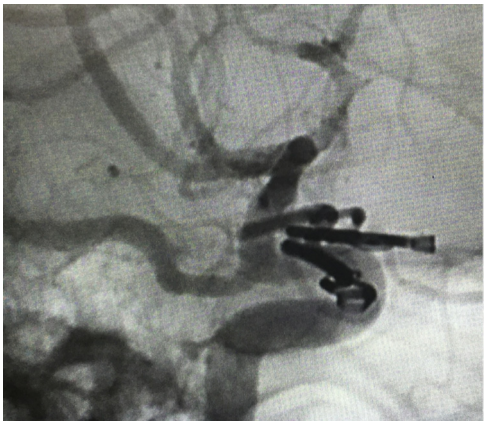

Fig. 7. Aneurysm was successfully excluded from the arterial circulation, as visualized in the postoperative angiography.

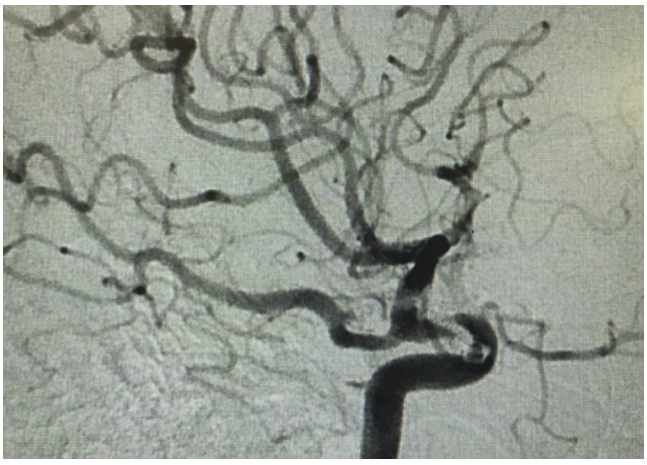

Fig. 8. Complete obliteration was achieved, as seen in postoperative angiography.

that saccular aneurysms of the ICA dorsal wall were significantly more common in patients with an anomalous origin of the ophthalmic artery, whereas BBAs occurred associated with normal ophthalmic arteries [19]. This angiographic feature should always be observed and may help preoperative planning. In all of our 15 cases, the ophthalmic artery had a normal origin.

According to the classification recently proposed by Bojanowski et al. [20], there are four distinct morphological subtypes of BBA: Classic (type I) – in which case there is only a small bulge on the ICA wall, with no neck; berry type (type II) – which resembles saccular aneurysms, but with extremely thin and friable walls; longitudinal (type III) – with a “neck”/dissection involving a longer segment of the ICA wall (larger than the vessel diameter); and circumferential (type IV) – in which case the lesion involves almost all or all the circumference of the ICA. In our series, type 2 aneurysms where recognized in most cases (7 patients), followed by type 1 BBAs (5 patients). Type 3 was found in 2 additional cases and type 4, in only 1 patient.

6. Microsurgical treatment

The microsurgical techniques for BBAs treatment include arterial wall reconstruction by clipping, [21,22] wrapping [1,22], trapping with or without revascularization [14,23–25], and primary suture repair [26]. The various therapeutic approaches reflects the uncertainty about which method will yield the best results [27,29].

In this series of cases, we had a prevalence of female (60%), in consonance with the literature, [3] as well as higher incidence around the fifth decade of life (mean age of 43.8 years). Microsurgical direct clipping [30,10] was the first alternative for all patients. Most patients were treated in the acute phase (before the 14th day post-SAH). Those treated after this period were due to delay in referrals. All lesions were approached through pterional or lateral supraorbital craniotomies, with no need for more elaborated skull base approaches. Usually, pterional approaches were used for patients with more extensive subarachnoid bleeding and/or associated hydrocephalus, whereas lateral supraorbital craniotomies were performed for low grade patients. Intraoperatively, carefull and wide Sylvian fissure dissection was performed in all of the cases. It is important to avoid frontal lobe retraction at all costs, because the aneurysm "dome" is frequently adhered to the orbital gyri and elevating the frontal lobe may precipitate intraoperative rupture. If needed, brain relaxation can be obtained by draining CSF by opening Liliequist membrane lateral to ICA (in the carotid-oculomotor space).

Obtaining proximal and distal control is a critical step when dealing with all aneurysms, but is particularly important for BBA. Distal control is usually easily obtained by Sylvian fissure splitting and visualization of the ICA bifurcation, but some BBA may be proximal in the ICA, making proximal control more difficult. In approximately half of our cases, anterior clinoidectomy was necessary in order to obtain proximal control. Cervical ICA exposure may also be performed for such purpose. After proximal and distal control are achieved, aneurysm dissection and clipping can be performed. In our series, 14 aneurysms were directly clipped. It is important to apply the clip parallel to the ICA and to incorporate a healthy wall portion of the ICA into the clipping, which may lead to a small, hemodynamically insignificant, ICA stenosis. In our series, 1 patient had a circumferential (type 4) BBA, precluding direct clipping. This patient was treated with the clip-wrapping technique, in which case the BBA and adjacent vascular wall are all reinforced with a Gore-Tex or a cotton band followed by the application of a clip to firmly attach the wrap around the vessel and induce subtle narrowing the parent vessel lumen [31,32]. Although suboptimal, clip-wrapping promotes the reinforcement of the carotid wall as a whole and offer some immediate protection for rebleeding and lesion progression [33,34], as previously demonstrated by Germanò et. al [35], is a valid option for treatment of aneurysms not easily clippable nor coilable and may yield favorable long-term results [22,36].

When trapping is considered as a method of treatment, a balloon occlusion test, to evaluate collateral arterial flow and the patency of distal ICA, should be performed before surgery. If the collateral circulation of the distal ICA is sufficient, trapping of BBA must have good outcomes. On the other hand, if the collateral circulation is insufficient, extracranial-intracranial (EC-IC) bypass may be required. This complex method is associated with high risks and complications [23]. Kamijo et al. [37] reported 7 consecutive patients with ruptured BBAs who were treated with trapping and EC-IC bypass. They showed that this surgical strategy might result in a favorable outcome in the treatment of BBAs.

Direct clipping of a BBA may result in intraoperative aneurysm rupture, bleeding, and laceration of the vessel wall [3,38]. In this series, we had only 2 cases (13%) of intraoperative rupture associated with BBA manipulation. Successful clipping was obtained in both cases without significant stenosis in the parent vessel. We believe 2 points are critical to avoid intraoperative rupture: strictly avoiding the use of brain retractors and absolute no frontal lobe retraction. Additionally, the aneurysm should only be manipulated after proximal and distal control are achieved. Despite careful manipulation of the vessel and clip placement, focal avulsions and tears on the ICA may occur during surgery for BBA. Depending on the size of the tear on the ICA, one may treat it by primary vessel repair, with clipping of part of the vessel circumference, inducing some degree of stenosis or by direct suture. Larger avulsion may be treated by trapping with or without bypass [14,23,24].

Post-operative angiographic control was performed in all cases, revealing complete exclusion in 14 cases. In one patient, post-operative

DSA showed persistent aneurysm filling and reoperation for clip repositioning was needed and performed successfully, resulting in complete obliteration of the lesion. There were no cases of significant ICA stenosis. Eighty percent of our patients achieved good clinical results (GOS 4 and 5). In the same way, other surgical series report the same good results with primary clipping as the preferred method [12,22,39,40,7]. Three patients presented significant neurological deficits (GOS 3) at discharge, all 3 cases due to delayed cerebral ischemia secondary to SAH.

7. Endovascular treatment

Several authors have used endovascular treatment as the primary management of blister aneurysms, [6,4,41] however there are poor results compared to the microsurgical treatment of these lesions. The endovascular therapy currently consists of basically 3 main options: endovascular ICA occlusion [41,42] stenting with or without coiling [6,4,41–43], and flow diverters [44,40,45],

Although several endovascular dispositive have been tried in recent years for the treatment of blister-type aneurysms, we do not advocate the use of this technique as the first line treatment, as it has been associated with procedural complications such as re-hemorrhage, ischemic complications, vision loss and death [46]. Aneurysm morphology is unfavorable for conventional coiling, and flow diverters often have delayed aneurysm obliteration with high rebleeding rate mainly in the first two weeks [47]. Also the necessity of dual antiplatelet therapy in the setting of subarachnoid hemorrhage may increase the risk of rebleeding and/or complications related to treatment of hydrocephalus or intracranial pressure monitoring [26]. Despite the recent trend of treating BBAs with flow diverters, we believe that with a properly trained, experienced cerebrovascular team, microsurgery should be considered as the first therapeutic option. Endovascular treatment may be an appropriate option for elderly patients or those with prohibitive comorbidities, and should be performed preferentially after the 14th day post-SAH.

8. Limitation of the study

Obvious limitations of our study are directly associated with retrospective data analysis and with the rarity of the disease being discussed, containing a relatively small number of patients.

9. Conclusion

BBAs are rare and challenging lesions. Through careful preoperative recognition and planning, these aneurysms can be safely treated by microsurgery. Direct clipping can be performed in most of the cases, affording high rates of early aneurysm obliteration and good clinical outcomes. With an experienced cerebrovascular surgery team, microsurgical management should remain the first alternative for the treatment of BBAs.

Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j.clineuro.2019.05.006.

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