Blood blister–like aneurysms at nonbranching sites of the internal carotid artery

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Object. The clinical features of blood blister–like aneurysms (BBAs) that arise at nonbranching sites of the internal carotid artery (ICA) differ from those of saccular aneurysms. In this study, the authors attempt to describe optimal treatments for BBAs, which have yet to be clearly established.

Methods. Ten of 483 patients with aneurysmal subarachnoid hemorrhage who had been seen at the authors’ institution between March 2001 and June 2005 had intraoperatively confirmed BBAs at nonbranching sites of the ICA. All ten patients were women between the ages of 37 and 64 years (mean age 49.3 years); five had a history of hypertension. The BBAs were localized to the right side of the ICA in seven cases. All patients were successfully treated; clipping was undertaken in six, clipping combined with wrapping in three, and trapping in one. These methods were used in conjunction with various other surgical techniques such as brain relaxation by draining cerebrospinal fluid, anterior clinidectomy, exposing the cervical ICA, gentle subpial dissection (for aneurysms that adhered to the frontal lobe), complete trapping of the ICA before clipping, and protecting the brain. Clip slippage occurred at the end of dural closing in two cases; the aneurysm was completely obliterated using multiple clips combined with ICA stenosis in one of these cases and ICA trapping with good collateral flow in the other. An excellent clinical outcome was achieved in eight patients, whereas two patients were disabled from massive vasospasm. The authors retrospectively reviewed radiological and surgical data in all cases to determine which treatment methods produced a favorable outcome.

Conclusions. Blood blister–like aneurysms located at nonbranching sites of the ICA are difficult to treat. Preoperative awareness and careful consideration of these lesions during surgery can prevent poor clinical outcomes.

Key Words • aneurysm • subarachnoid hemorrhage • blood blister–like aneurysm • internal carotid artery

Aneauysms located at the anterior aspect of nonbranching sites in the supraclinoid ICA were described first by Sundt and Murphey. Such aneurysm locations have also been referred to as the “dorsal,” “distal medial,” “superior,” and “anterior walls” of the ICA or ICA trunk. These lesions are classified into two types: saccular aneurysms and BBAs. The latter are thin-walled, broad-based aneurysms that lack an identifiable neck. They are fragile and can rupture during microsurgery and cause postoperative rebleeding more frequently than saccular aneurysms. Even though BBAs and saccular aneurysms occur within the same anatomical regions, these two lesion types are distinct. The natural history of the BBA has not been clarified because this type of lesion is rare. Furthermore, there is confusion regarding the nomenclature and classification of BBAs based on characteristics such as anatomical location and configuration. Although several surgical strategies are available to treat BBAs, the safest treatment modality is still a matter of conjecture. In the present study, we analyzed the clinical characteristics and surgical outcomes in a series of BBAs at nonbranching sites of the ICA to obtain a better understanding of the clinical nature of, and the optimal treatment for, these aneurysms.

Clinical Material and Methods

Initially, we reviewed the data and charts for 483 patients who had undergone surgery for aneurysmal SAH between March 2001 and June 2005. From this population, we selected those patients with aneurysms at nonbranching sites of the supraclinoid portion of the ICA. We excluded 124 patients with aneurysms adjacent to the opthalmic, superior hypophyseal, posterior communicating, or anterior choroidal artery. Thirteen patients harbored aneurysms along the anterior aspect of nonbranching sites with no relation to the ICA branches. Of these, three patients with saccular-type aneurysms were excluded from this study. The remaining 10 patients had BBAs that were confirmed during surgery. These patients constituted 2.1 and 7.3% of all patients with ruptured aneurysms and ruptured ICA aneurysms, respectively.

Various methods were used to perform an ipsilateral craniotomy in all patients, including cervical ICA exposure.
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and anterior clinoidectomy for proximal control of the ICA. The Hunt and Hess system of classification was used to rate the preoperative condition of patients. The Glasgow Outcome Scale was used to evaluate the postoperative condition of patients at the time of the final clinical follow-up evaluation. Postoperative angiography was performed to determine the surgical outcome.

Results

A summary of patient characteristics (age, sex, history of hypertension, aneurysm site, and intra- and postoperative results) are presented in Table 1. All 10 patients were women with a mean age of 49.3 years (range 37–64 years) who had presented with an SAH, which was caused by the rupture of a BBA having a Fisher grade of 2 in two patients, Grade 3 in seven patients, and Grade 4 in one patient. Three patients had multiple aneurysms, which were located within the ipsilateral anterior choroidal artery in one patient (Case 2), at the basilar tip in another (Case 3), and at the ipsilateral ventral wall of the ICA in the third (Case 7). The BBA was localized to the right or left side of the ICA in seven and three cases, respectively. Five patients had a history of hypertension, but all were free from other risk factors such as smoking, alcohol consumption, familial connective tissue disorders, and use of medications such as oral contraceptives.

The maximum diameter of the BBAs as measured on angiograms ranged from 2 to 8 mm. In seven cases, the BBA was located at the anterior aspect of the dorsal wall of the ICA; in the other cases, the lesion was located at the anterolateral (two cases) or anteromedial aspect (one case). A second angiography study was performed in one patient (Case 9) in whom a BBA was strongly suspected; results of follow-up angiography performed 1 week later revealed that the suspected aneurysm had increased in size.

All BBAs were surgically accessed via the pterional approach, except for one case in which we used the eyebrow approach (Case 8). Clip application with or without wrapping was performed in combination with various additional surgical procedures, including lumbar CSF drainage (six cases), removal of the anterior clinoid process (five cases), and preparation of the cervical ICA for proximal control (three cases). The sylvian fissure was opened before accessing the carotid and chiasmatic cisterns in nine cases; in one patient (Case 8) in whom the chiasmatic cistern had been dissected before opening the sylvian fissure, the latter procedure resulted in premature rupture of the BBA. In most anterior and anteromedial BBAs, frontal lobe retraction was delayed as long as possible because of adhesion to the aneurysm dome. Subpial dissection of the frontal lobe was performed at the final stage immediately before applying an aneurysm clip, after obtaining sufficient space for temporary clipping of the ICA. Direct surgical clip application was possible in six cases. In three cases, we applied multiple clips (mean 1.9 clips per aneurysm, range 1–3 clips) onto wrapping material (cellulose fabric) that covered the entire circumference of the ICA. The ICA was trapped in one patient (Case 10) who had experienced rebleeding after clipping because the aneurysm had been separated from the parent artery with the clip during dural closure. Tearing of the aneurysm neck during dissection occurred in five patients (Cases 1, 4, 7, 8, and 10). In two patients (Cases 7 and 8) in whom neck clipping was impossible because of a lacerated lesion neck, we applied clips onto wrapping. Two patients (Cases 2 and 10) with postoperative rebleeding after clipping survived without any focal neurological deficits. These favorable outcomes were likely the result of the clips slipping off during dural closure; therefore, bleeding from the ICA wall defect could be controlled immediately.

Results of follow-up angiography in all patients revealed complete obliteration of all aneurysms with or without stenosis of the parent artery. The follow-up angiogram obtained in the patient in Case 10, who had undergone trapping, revealed good collateral flow through the ACoA and ipsilateral PCoA. All patients underwent regular follow-up evaluations in an outpatient ward, and the mean follow-up period was 27.2 months (range 5–49 months). An excellent clinical outcome was achieved in eight patients, whereas two patients (Cases 4 and 8) were disabled as a result of massive vasospasms.

Illustrative Cases

Case 8

History and Examination. This 45-year-old woman was admitted to our hospital in a drowsy state. A CT scan revealed a large hemorrhagic clot extending throughout the basal, interpeduncular, and ambient cisterns and the bilateral sylvian fissures. Four-vessel angiography results showed a small aneurysm on the anterior wall of the ICA (Fig. 1A).

Operation. An ipsilateral craniotomy was performed via the eyebrow approach. An extradural anterior clinoidectomy was performed to obtain sufficient space for proximal control. After incision of the dura mater, the frontal lobe was retracted, and the chiasmatic cistern was opened to drain CSF. During this procedure, the aneurysm ruptured. The remainder of the cistern was dissected under continuous suction, and compression was applied with a cellulose cotton sheet (Bemsheet; Kawamoto, Osaka, Japan) to reduce bleeding. Finally, the ICA was exposed. There was a hole in the anterior aspect of the ICA that corresponded to the site at which the BBA had been torn, leaving only a fragile remnant of the aneurysm neck. Because direct clamping of the neck appeared to be too dangerous, the aneurysm was wrapped with cellulose fabric and constricted via intentional ICA stenosis by using two right-angled clips with temporary trapping of the ICA. Intraoperative Doppler ultrasonography findings revealed that there was good blood flow distal to the clipped ICA.

Postoperative Course. Nine days later, the patient exhibited left hemiparesis with gradual mental deterioration but no new lesions on a subsequent CT scan. An immediate follow-up angiogram revealed moderate right ICA stenosis attributable to the two clips. Blood flow distal to the stenosed portion was comparatively well maintained (Fig. 1B) despite mild vasospasm. There was moderate vasospasm on the contralateral side. Papaverine (150 mg) infused bilaterally into the ICA caused a small increase in blood flow (Fig. 1C), which was insufficient to maintain adequate circulation in the left side. Cerebral infarction within the left frontoparietal area was visualized on a CT scan obtained a few
days later. The patient was discharged with profound right-sided weakness, and she died of sepsis of gastrointestinal origin within 1 year.

**Case 10**

**History and Examination.** This 47-year-old woman was admitted to our hospital with severe headache and a change in her mental status. A CT scan revealed a thick clot throughout the basal region, and 3D reconstruction of the intracranial vessels revealed a tiny focal bulging of the anterior wall of the right ICA. Angiography findings demonstrated a funnel-shaped aneurysm dilation of the ICA at the contralateral side of the PCoA (Fig. 2A), which is typical of BBAs. It was agreed that direct clipping might tear the fragile aneurysm neck and that wrapping the entire circumference of the ICA was impossible given the presence of the PCoA on the other side of the aneurysm. There was sufficient collateral flow from the contralateral ICA through the ACoA on compression of the ipsilateral ICA; blood flow from the PCoA was also good. Surgical clipping was planned, and ICA trapping would be used if the aneurysm neck was torn or if clipping was impossible.

**Operation.** A craniotomy via the ipsilateral pterional approach was performed with cervical exposure of the ICA. The sylvian fissure was dissected widely, and sufficient CSF was drained to prevent excessive retraction of the frontal lobe, which might have adhered to the aneurysm. The lesion was exposed through subpial dissection of the frontal lobe (Fig. 2B). Before the final clipping, we attempted to wrap the aneurysm with fabric cellulose, which caused it to rupture. The cervical ICA was clamped immediately, and then the lesion was directly clipped. The aneurysm was believed to have been clipped completely at that time (Fig. 2C). After closing the dura, we observed massive bleeding through a small opening in the dura, which led to brain swelling. With the cervical ICA clamped, an emergency partial frontal lobectomy was performed to facilitate assessment of the ICA. There was active bleeding from a hole in the anteromedial wall of the ICA, and the aneurysm clip was displaced. Because there was no neck to clip, the ICA was trapped. The first clip was placed proximal to the aneurysm, and the distal clip was placed obliquely proximal to the origin of the PCoA (Fig. 2D and E) so that blood flow from the PCoA to the distal ICA would be preserved while trapping the ICA.

**Postoperative Course.** Follow-up angiography findings obtained 2 weeks later revealed good collateral circulation from the ipsilateral PCoA and ACoA in both the anteroposterior and lateral views and complete obliteration of the aneurysm (Fig. 2F). The patient recovered without any focal neurological deficits and was discharged 4 weeks later.

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**TABLE 1**

**Clinical summary of 10 patients with BBAs of the ICA***

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Age (yrs)</th>
<th>Sex</th>
<th>Preop Hypertension</th>
<th>Side of Aneurysm</th>
<th>Intraop Rupture</th>
<th>Postop Rupture</th>
<th>Op Method</th>
<th>Clinical Outcome</th>
<th>Follow Up (mos)</th>
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<td>yes</td>
<td>clipping &amp; wrapping</td>
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<td>44</td>
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<tr>
<td>3</td>
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<tr>
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<td>clipping &amp; wrapping</td>
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<td>no</td>
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<td>yes</td>
<td>yes</td>
<td>ICA trapping</td>
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* H & H = Hunt and Hess.
† Dead.
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Discussion

Clinical Features of BBAs

Lesions arising within nonbranching sites of the supraclinoid ICA have an incidence from 0.3 to 1.7% for intracranial aneurysms and from 0.9 to 6.5% for ICA aneurysms. Saccular aneurysms can occur at nonbranching sites along the anterior aspects of the supraclinoid segment of the ICA, but BBAs with fragile walls and necks represent the most common types of lesions found in this area. In addition to their anatomical location, BBAs have several unique clinical and pathological features. Most saccular aneurysms arise within the angle formed by the parent artery and a significant arterial branch, whereas BBAs usually originate directly from the vessel wall, without any relationship to the arterial branches. Results of an autopsy indicated that BBAs are focal wall defects covered by a thin layer of fibrous tissue and adventitia and lack the usual collagenous layer. Therefore, these aneurysms have a high risk of premature rupture during surgery and are associated with the development of large lacerations on the parent vessel during microsurgery, both of which may result in a poor outcome.

It has also been reported that BBAs occur predominantly on the right side and in females. Frequently, these lesions are associated with hypertension, arteriosclerosis, and ICA dissection. In our study, more than one half of the patients had BBAs on the right side and a history of hypertension, findings that resemble those of others. Almost all patients had atherosclerotic parent arteries adjacent to the BBAs, but there was no relation to ICA dissection in those in the present study.

We cannot explain why BBAs occur more frequently in female persons. Nonetheless, all of the patients in our study were middle-aged women and were relatively young compared with the general population of patients with aneurysmal SAH. Even though they were relatively young, all had severe atherosclerosis of the ICA, which was confirmed during surgery. Therefore, atherosclerosis of the ICA may be a major factor in the pathogenesis of BBA. Not only focal wall defects associated with arteriosclerosis-induced ulceration and penetration of the internal elastic lamina, but also high female susceptibility under certain conditions may play important roles in the development of BBAs.

Hypertension has been associated with intra- and postoperative aneurysm rupture, which was borne out in the present study—four of five patients with ruptured BBAs also exhibited hypertension. We surmise that neck tearing occurs more frequently in hypertensive patients because of a difference in the stiffness of the atherosclerotic parent artery adjacent to the ICA wall at which the BBA is located. Presumably, abrupt histological transition from the sclerotic ICA wall to the fragile aneurysm neck is more likely to result in laceration of the wall because of clip torsion.

Radiological Characteristics of BBAs

All patients included in our series had presented with SAH. The anterior location and small size of the BBAs mean that anteroposterior- and lateral-view angiography studies provide less complete visualization of these lesions;

Fig. 2. Case 10. A: Preoperative angiogram, right oblique view, revealing a focal aneurysm dilation on the anterior wall of the right ICA (arrowhead). B–E: Photographs obtained during surgery, showing the treatment course. The lesion had a very thin, fragile neck and a dome (B), but direct clipping was attempted first (C). Unable to clip the aneurysm neck after clip displacement, we performed ICA trapping. The first clip was placed proximal to the ICA, whereas the distal clip was placed obliquely proximal to the origin of the PCoA (D) to preserve blood flow from the PCoA to the distal ICA while trapping the ICA. The dotted line indicates the trajectory used in the trapping (E) while avoiding the PCoA (arrowhead). F: Postoperative angiograms demonstrating good collateral flow to the middle cerebral artery territories distal to the trapped ICA.
therefore, we recommend performing rotational angiography in cases of suspected BBA. Special attention is required in oblique-view angiography studies, preferably with the Waters angle, to avoid missing these lesions. The widespread availability of 3D CT angiography and meticulous evaluation of 3D scans allow BBAs to be more easily detected.

Blood blister–like aneurysms reportedly exhibit rapid growth and changes in shape, factors demonstrated in one patient (Case 9) in the present study. Therefore, even if a patient with a dense SAH and no evidence of perimesencephalic nonaneurysmal SAH has no aneurysms according to angiography studies, a BBA cannot be excluded until after a careful review of conventional, 3D CT, and follow-up angiograms.

If angiography studies reveal a lesion that might be a BBA, it is preferable to confirm collateral flow from the posterior circulation and the contralateral side via the PCoA and ACoA. This strategy will allow one to determine whether the lesion can be trapped if it cannot be clipped because of a possible laceration of the neck. If trapping is considered, it may be useful to perform preoperatively a balloon occlusion test for collateral flow. In addition, the external carotid artery should be examined in case it is necessary to perform bypass surgery. Furthermore, the location of the aneurysm in relation to the PCoA and anterior choroidal artery should be determined, because wrapping the full circumference of the ICA or applying an encircling clip may be difficult if these arteries are located on either side of the aneurysm. In cases in which the lesion is trapped opposite the PCoA, we suggest that the distal clip be applied obliquely from the other side of the PCoA to its immediately proximal portion, as illustrated in the patient in Case 10. This method makes it possible to completely block back-flow from the PCoA to the aneurysm and maintain blood flow from the posterior circulation to the distal branches of the trapped artery.

**Surgical Outcomes Associated With BBAs**

Blood blister–like aneurysms are particularly dangerous lesions because they frequently rupture during and after surgery. The challenge for surgeons is to determine the best treatment (clipping, clipping onto wrapping, or trapping). To date, applying clips onto wrapping of various materials has been proposed as the best method for the treatment of BBAs; ICA trapping, clipping, and wrapping alone are associated with much worse outcomes. Note, however, that surgeons are often unwilling to wrap BBAs because these lesions are extremely fragile. Moreover, complete wrapping or applying an encircling clip is sometimes impossible without sacrificing branches of the ICA or perforating vessels.

Direct clipping of a BBA is straightforward, but this method often leads to rupture of the lesion at its fragile neck, even if sophisticated parallel clipping is performed. Moreover, if clip blades do not catch the wall of the parent artery, rebleeding and aneurysm regrowth will occur. To avoid this result, clipping combined with ICA stenosis has been performed. However, applying a clip that intentionally narrows the ICA reportedly results in severe ischemic complications. Trapping is another option in the treatment of BBAs. However, poor collateral circulation distal to the trapped ICA leads to an extremely poor outcome. Recently, it was reported that microsutures could be used to directly repair lacerations of the parent ICA. Nonetheless, this method requires considerable time and skill. In recent years, endovascular approaches for BBAs, such as coil embolization, have been attempted in cases of pseudoaneurysm formation. Theoretically, however, coil embolization might be very dangerous in treating a BBA given the lesion’s wide neck and hemispheric dome with a very weak fibrous layer lacking collagenous tissue.

To overcome the aforementioned difficulties and to improve outcomes, it is crucial to be aware of the presence of BBAs before surgery and to prepare thoroughly for all possible methods of treatment. First, one must perform an anatomic review to determine the best location for trapping and predict the results of this procedure. Next, one must prepare for all potential surgical treatments, including clipping, clipping onto wrapping, trapping, bypass, and direct suturing. All surgical equipment must be prepared before opening the dura. Such equipment includes wrapping material, microsurgical suture material, and all types of clips (such as the encircling clip graft, also known as the Sundt clip, which is known to be effective in treating aneurysms with neck tearing or laceration of the ICA).

Surgeons should consider each of the methods available for exposing the ICA (including anterior clinoidectomy and cervical exposure), particularly those for temporary proximal occlusion and ICA pressure control. Blood blister–like aneurysms often adhere to the frontal lobe in an anterior and anteromedial direction, whereas they often adhere to the temporal lobe in an anterolateral direction. Therefore, brain retraction should be minimized, although sufficient CSF should be drained via a wide sylvian dissection. In addition, gentle subpial dissection of the brain may be warranted in cases with adherent lesions.

Clips should be applied while pressure within the ICA is low, and the clip blades should be situated parallel to the parent artery and should catch the arterial wall beyond the lesion. These principles apply to both direct clipping and clipping onto wrapping. If this method is not used, clip distortion may occur toward the end of the operation.

According to the literature, postoperative bleeding leads to death in more than 90% of cases. In the present study, however, two patients who suffered a postoperative rupture had an excellent outcome and resumed normal life activities without any neurological deficits, despite the fact that clip slippage caused the aneurysm in each to tear completely. This good result occurred because the lesions ruptured postoperatively, before dural closure; therefore, we were able immediately to treat the rupture. Based on this experience, we surmise that postoperative rupture of the aneurysm may have occurred as a result of clip torsion or slippage, which may have caused the frontal or temporal lobe to return to its original position after closing the dura. Perhaps elevated blood pressure led to the same result. Thus, we recommend confirming the stability of clips with induced blood pressure elevation and repeated irrigation. This step should be performed before closing the dura, given that postoperative rebleeding is more difficult to detect and treat after the dura is closed.
Lessons and Further Considerations in the Treatment of BBAs

A review of our data on BBAs has revealed insights into the specific characteristics of these lesions and the techniques that can be used to treat them. Are direct clipping and clipping onto wrapping really safe? How can we distinguish which aneurysm can be safely clipped during surgery? These questions are impossible to answer based on preoperative angiography studies. Therefore, it is necessary to prepare for all of the methods that can be used to obliterate BBAs, including clipping and the other aforementioned techniques. Of clinical note, BBAs are prone to rupture during microdissection, and the preparation of the cervical ICA is very important. Moreover, collateral flow through the ACoA and PCoA should be examined before surgery in all suspected cases of BBA. This step is particularly important because complete obliteration of the BBA may cause significant stenosis of the parent artery. In the present study, we did not perform a balloon occlusion test, which requires additional procedure time and heparinization. Furthermore, this test sometimes causes an ischemic complication. Nonetheless, in selected cases in which collateral circulation is poor and the lesion needs trapping, we must perform a balloon occlusion test and consider bypass surgery. In these cases, a superficial temporal artery must be preserved for possible use. If required, trapping should be performed while saving the PCoA collateral flow, if possible, via oblique clip placement. Despite the availability of effective surgical techniques for treating BBAs, preparative awareness of these lesions is the factor that most significantly determines surgical outcome. To prevent a poor outcome, preparative examination of BBAs should be combined with various surgical techniques such as preparation of the cervical ICA, gentle pial dissection (for adherent BBAs), complete trapping of the ICA before clipping, and good brain protection (if trapping is prolonged).

Conclusions

The clinical characteristics of BBAs are distinct from those of saccular aneurysms. Blood blister–like aneurysms are extremely dangerous lesions that often rupture during or after surgery. The outcomes in patients with BBAs can be improved by obtaining a thorough awareness of these lesions before surgery. An angiographic analysis of the BBA’s location as well as an examination of the collateral flow should be performed before surgery. In addition, one should prepare for all possible surgical strategies, including clipping, wrapping, and trapping.

References


Manuscript received August 31, 2005. Accepted in final form March 1, 2006.

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