Acute extracranial-intracranial bypass using a radial artery graft along with trapping of a ruptured blood blister–like aneurysm of the internal carotid artery

Clinical article
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Object. Fragile aneurysm walls and poorly defined necks render the surgical treatment of blood blister–like aneurysms (BBAs) located at nonbranching sites of the supraclinoid internal carotid artery extremely challenging. Such aneurysms have a remarkable tendency to rupture during surgery, especially during the acute period. The authors describe the clinical course of patients with subarachnoid hemorrhage (SAH) caused by BBA rupture and emphasize the value of internal carotid artery trapping combined with high-flow extracranial-intracranial (trapping/EC-IC) bypass during the acute period following SAH.

Methods. The authors analyzed the clinical records of 7 consecutive female patients with a mean age of 61 years (range 51–77 years) who had been treated between January 2006 and December 2008 at their institute.

Results. All 7 patients presented with SAHs corresponding to Fisher Grade 3 and World Federation of Neurosurgical Societies Grades II, III, IV, and V in 3, 1, 2, and 1 patient, respectively. Surgery was postponed in the 3 patients, including 1 in whom the trapping/EC-IC bypass procedure was performed during the chronic period. Two of the 3 patients in whom surgery was postponed experienced preoperative rebleeding, and repeated angiography revealed remarkable enlargement of the aneurysm; both of these patients died before surgery could be performed. The remaining 4 patients underwent trapping/EC-IC bypass during the acute period following SAH. The outcome was excellent (Glasgow Outcome Scale Scores 5), and postoperative angiography demonstrated complete obliteration of the BBA as well as good graft patency in all 5 patients who underwent trapping/EC-IC bypass. Intraoperative bleeding from the BBAs never occurred in any of these 5 patients.

Conclusions. Ruptured BBAs were successfully treated with a trapping/EC-IC bypass during the acute SAH period. This surgical strategy for treating BBAs during the acute period might be a promising option for these rare but high-risk lesions. (DOI: 10.3171/2009.10.JNS09970)

Key Words • intraoperative bleeding • aneurysm rupture • subarachnoid hemorrhage

Abbreviations used in this paper: BBA = blood blister–like aneurysm; DS = digital subtraction; EC-IC = extracranial-intracranial; GOS = Glasgow Outcome Scale; ICA = internal carotid artery; RA = radial artery; SAH = subarachnoid hemorrhage; SWS = stent-within-a-stent; trapping/EC-IC bypass = trapping of the ICA aneurysm combined with high-flow EC-IC bypass; WFNS = World Federation of Neurosurgical Societies.

Blood blister–like aneurysms located at nonbranching sites of the supraclinoid internal carotid artery are characterized by extremely thin, fragile walls, poorly defined necks, and a high tendency to rupture during surgery, especially during the acute SAH period. Direct clipping of BBAs is usually very difficult and such attempts frequently result in aneurysm avulsion and ICA laceration. Thus, multiple surgical strategies have been tried, including parallel clip placement, encircling clip placement, vascular closure staple clipping, clip placement on wrapping, and ICA trapping. Recent endovascular approaches, such as primary coil embolization with or without adjunctive devices such as stents and balloons, have also been used for BBAs. Regardless of whether the modality is surgical or endovascular, many of these procedures pose a high risk of intra- or postoperative rebleeding, an association with increased peri-procedural morbidity and mortality rates, and a poor outcome. Here, we emphasize the value of trapping an ICA aneurysm combined with high-flow EC-IC bypass (trapping/EC-IC bypass) during the acute period following SAH in patients with BBAs.
Methods

We referred to the institutional review board of Saitama Medical Center for approval of this study, and they documented that this type of study did not need to be examined by them.

Table 1 lists a summary of characteristics in 7 female patients (mean age 61 years, range 51–77 years) with ruptured BBAs who presented at our institution between January 1, 2006, and December 31, 2008. The patients were neurologically examined on admission, and their SAHs were classified as WFNS Grades I to V. The extent and distribution of SAH on admission CT scans were evaluated according to Fisher grading criteria. A diagnosis of BBA was confirmed on DS angiograms showing a small and slightly elevated hemispheric bleb or irregular protrusion located at the nonbranching sites of the supraclinoid ICA. Surgery was delayed in 3 patients (Cases 1–3) to avoid intraoperative rupture and additional damage to already fragile brains as a result of the SAH. The other 4 patients underwent early surgery as described below.

Neurosurgical staff together with neuroradiologists and neurologists evaluated the postoperative condition of each patient based on GOS scores at discharge and 6 and 12 months thereafter. Obliteration of the aneurysm and patency of the arterial bypass graft was confirmed on postoperative angiography.

Surgical Strategy

An EC-IC bypass was performed using an RA graft interposed between the external carotid artery at the cervical portion and the M2 segment, followed by surgical ICA trapping. During the first part of this 3-step procedure, the cervical skin was linearly incised along the anterior edge of the sternocleidomastoid muscle to identify and expose the ipsilateral cervical carotid bifurcation. An RA graft ~18 cm long was simultaneously harvested by another team. Following an ipsilateral frontotemporal craniotomy, the ipsilateral cervical carotid bifurcation was temporarily occluded to avoid intraoperative bleeding from the aneurysm, and the sylvian fissure was opened to provide a sufficiently large operative field. Thereafter, the frontal lobe was carefully elevated to facilitate the approach to the ICA. The aneurysm was trapped by placing a clip on the ICA just proximal and distal to the aneurysm.

Results

Table 1 summarizes the clinical features of the 7 patients with Fisher Grade 3 SAH caused by BBA rupture. On admission, 3, 1, 2, and 1 patient had initial WFNS Grades II, III, IV, and V SAH, respectively. These cases made up 2.9% of all ruptured intracranial aneurysms and 9.1% of all ruptured ICA aneurysms that had been treated at our facility between January 1, 2006, and December 31, 2008. All BBAs were located at the anterior wall of the supraclinoid portion of the ICA and were not associated with the arterial branch. Five aneurysms were located on the left side (Cases 1 and 3–6) and 2 on the right (Cases 2 and 7). Multiple aneurysms were located at the bilateral ICA–ophthalmic artery segment, the ipsilateral posterior communicating artery, and the ipsilateral anterior cerebral artery in the patients in Cases 3, 4, and 6, respectively.

Surgery was delayed in the patients in Cases 1–3. The patient in Case 1 underwent trapping/EC-IC bypass during the chronic stage. In the patients in Cases 2 and 3 rebleeding developed preoperatively as well as during the subacute period following SAH, and repeated angiography revealed remarkable enlargement of the aneurysm. Both of these patients died of rebleeding during the chronic period following SAH, before the planned surgery.

The patients in Cases 4–7 were treated with trapping/EC-IC bypass during the acute period following SAH. No intraoperative bleeding was derived from the BBAs in any of the 5 patients who underwent trapping/EC-IC bypass. Postoperative angiography demonstrated obliteration of the aneurysm and good arterial graft patency in all 5 patients.

### Table 1: Summary of characteristics in 7 patients with BBAs of the ICA*

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Age (yrs)</th>
<th>Initial WFNS Grade</th>
<th>Aneurysm Location</th>
<th>Aneurysm Size (height x length)</th>
<th>Treatment Schedule</th>
<th>Preop WFNS Grade</th>
<th>Clinical Course (days after SAH)</th>
<th>Vasospasm</th>
<th>GOS Score/FU</th>
<th>Mos</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>77</td>
<td>III</td>
<td>lt</td>
<td>4 x 3</td>
<td>postponed</td>
<td>I</td>
<td>T&amp;B (21)</td>
<td>severe</td>
<td>5/37</td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>55</td>
<td>II</td>
<td>rt</td>
<td>3 x 3</td>
<td>postponed</td>
<td>rebleeding (10), death (13)</td>
<td>moderate</td>
<td>1</td>
<td></td>
<td></td>
</tr>
<tr>
<td>3</td>
<td>51</td>
<td>IV</td>
<td>lt</td>
<td>1 x 2</td>
<td>postponed</td>
<td>rebleeding (7), death (10)</td>
<td>mild</td>
<td>1</td>
<td></td>
<td></td>
</tr>
<tr>
<td>4</td>
<td>70</td>
<td>II</td>
<td>lt</td>
<td>3 x 2</td>
<td>acute</td>
<td>II</td>
<td>T&amp;B (1)</td>
<td>none</td>
<td>5/14</td>
<td></td>
</tr>
<tr>
<td>5</td>
<td>51</td>
<td>V</td>
<td>lt</td>
<td>5 x 3</td>
<td>acute</td>
<td>IV</td>
<td>T&amp;B (2)</td>
<td>severe</td>
<td>5/9</td>
<td></td>
</tr>
<tr>
<td>6</td>
<td>70</td>
<td>II</td>
<td>lt</td>
<td>5 x 3</td>
<td>acute</td>
<td>II</td>
<td>T&amp;B (1)</td>
<td>none</td>
<td>5/8</td>
<td></td>
</tr>
<tr>
<td>7</td>
<td>53</td>
<td>IV</td>
<td>rt</td>
<td>1 x 2</td>
<td>acute</td>
<td>IV</td>
<td>T&amp;B (0)</td>
<td>mild</td>
<td>5/6</td>
<td></td>
</tr>
</tbody>
</table>

* FU = follow-up; T&B = aneurysm trapping and bypass.
Although symptomatic vasospasm developed in the patients in Cases 5 and 7, our conventional treatment for vasospasm, which has been described in detail elsewhere, avoided delayed ischemic or permanent neurological deficits in these 2 patients. The outcome was excellent (GOS Score 5) in all 5 patients who underwent trapping/EC-IC bypass. The patient in Case 5 is described below as a representative case.

Illustrative Case

Case 5

History and Examination. This 51-year-old woman suddenly lost consciousness and was comatose (WFNS Grade V SAH) on admission. A CT scan showed diffuse SAH and a large hematoma in the left frontal lobe (Fisher Grade 3). Cerebral angiography revealed a 5 × 3-mm aneurysm at nonbranching sites in the supraclinoid portion of the left ICA (Fig. 1). The WFNS grade improved from V to IV on the following day, when moderate weakness in the right side and a clear response to painful stimuli were evident.

Operation. Two days after the initial hemorrhage, the patient underwent trapping/EC-IC bypass, and the aneurysm was surgically trapped without intraoperative bleeding. A blood blister–like bulge together with purplish discoloration of the left ICA was evident during surgery (Fig. 2).

Postoperative Course. Four days after the hemorrhage, the patient responded well to verbal commands but remained aphasic and moderately hemiparetic on the right side. The right side further weakened, and a consciousness disturbance developed 1 week later. Immediate postoperative angiography demonstrated that the bypass was patent and that the aneurysm was successfully trapped, although the left middle cerebral artery was severely vasospastic (Fig. 3). No new lesions were evident on a subsequent CT scan. Our conventional version of hypervolemia-hypertension-hemodilution therapy led to a complete recovery (GOS Score 5) for this patient and discharge to home within 1 month of admission.

Discussion

Blood blister–like aneurysms account for 0.4–2.7% of ruptured intracranial aneurysms and 0.9–9.4% of ruptured ICA aneurysms. Because these aneurysms have extremely thin, fragile walls and poorly defined necks, they are very difficult to treat with either surgical clipping or endovascular coiling. Direct clip application for BBAs frequently causes premature rupture, laceration of the lesion, and parent artery stenosis or occlusion. Although surgical approaches include parallel clip placement, encircling clip placement, vascular closure staple clipping, and clip placement on wrapping, many of these strategies are also associated with high morbidity and mortality rates because of intraoperative rupture and postoperative bleeding.

Endovascular approaches, such as primary coil embolization with or without stent assistance, SWS techniques, and covered-stent techniques, have been described. However, primary coil embolization is often insufficient to prevent rebleeding or BBA regrowth. Lee et al have recently achieved good results in 8 of 9 patients with...
BBAs treated using reconstructive endovascular surgery with ICA preservation via SWS or covered-stent techniques. Note, however, that an SWS procedure may not prevent BBA rebleeding or regrowth, despite the use of at least 2 overlapping stents. Furthermore, the covered-stent technique may carry a risk of additional damage to an already fragile BBA and ICA wall. In the study by Lee and colleagues, the ICA ruptured and led to the intraoperative death of 1 of 3 patients who had undergone covered-stent procedures. Moreover, because the long-term effects remain questionable, these strategies cannot be considered as a first option for definitive treatment.

In our series, 5 of 7 patients with BBAs underwent trapping/EC-IC bypass. Theoretically, aneurysm trapping should prevent rebleeding and regrowth over the long term. In a cadaveric study Ishikawa et al.4 have found that these lesions are focal wall defects covered with thin fibrous tissue and that they are not true aneurysms. Furthermore, arterial dissection might be the pathogenesis of some BBAs according to numerous other reports.1,3,4,10,12,13 In Japan, based on nationwide surveillance, Satoh et al.15 have recently found that 97 (50.2%) of 193 patients who had undergone surgical interventions actually harbored a dissecting type of aneurysm. If dissection is a presumed pathogenesis of BBAs, trapping would be the only possible and effective surgical option. Data in the present study demonstrated an excellent outcome without BBA rebleeding or regrowth in all 5 patients who underwent aneurysm trapping.

Note, however, that permanent ICA occlusion may result in very poor outcomes when collateral flow is insufficient. Meling et al.10 have reported that all 7 patients in whom the ICA was sacrificed had poor outcomes because of cerebral infarcts. Moreover, severe infarction invariably arose because of vasospasm even though 4 of their patients had sufficient collateral flow according to preoperative angiography. These authors concluded that sacrificing the ICA within 48 hours of an SAH leads to a very poor outcome, even in patients with adequate collateral capacity according to preoperative angiography. This phenomenon may be led by vasospasm-induced compromise of the cerebral collateral vessels. Based on these observations, Meling et al. have postulated that a high-flow EC-IC bypass before the onset of vasospasm would prevent vasospasm-induced cerebral infarcts and subsequent death associated with ICA sacrifice in the acute SAH period. We trapped aneurysms on the ICA and performed concomitant bypass surgery based on this theory. A high-flow EC-IC bypass using an RA graft was chosen to provide extensive cerebral perfusion. None of the 5 patients in our series had ischemic complications immediately after permanent ICA occlusion without EC-IC bypass. From the viewpoint of postoperative preservation of the cerebral circulation, the necessity for combined bypass surgery should be judged based on the results of balloon test occlusion. Whether such procedures could be tolerated under the likelihood of cerebral vasospasm is doubtful, however, even though some patients might tolerate acute ICA occlusion without bypass immediately after surgery as a result of the test occlusion. This line of reasoning renders EC-IC bypass indispensable in patients undergoing ICA sacrifice to treat BBAs during the acute period as well as during subsequent periods.

Baskaya et al.2 have described a series of 4 patients with BBAs who underwent EC-IC bypass and surgical or endovascular trapping during the chronic stage. Three patients had a good outcome, whereas 1 died after preoperative rebleeding developed. The walls of BBAs are extremely thin and fragile, which renders them vulnerable to rupture before surgery. Among our series, preoperative rebleeding occurred and the BBA rapidly enlarged in 2 of the 3 patients in whom surgery was delayed; both of these patients (Cases 2 and 3) eventually died. Because rapid BBA enlargement has also been documented by many others,1,3,9,11,12,16,20 the susceptibility to bleeding and rapid enlargement of these aneurysms represent indications for an early surgical approach. However, BBAs have a marked tendency to rupture during surgery, especially during the acute period,5,16,18,20 and the postoperative outcome is significantly worse after early surgery compared with that following delayed surgery.10,15 To prevent premature rupture, careful and minimal retraction of the brain, gentle subpial brain dissection around the aneurysm, and temporary occlusion of the cervical ICA are advocated.7,8,12,18 Note, however, that these procedures might be insufficient and temporary occlusion of the cervical ICA might be possible for only short periods.
Early surgery for ruptured blood blister–like aneurysms

Temporary occlusion of the cervical ICA after a high-flow EC-IC bypass might minimize intraaneurysmal pressure for long enough to enable careful and gentle manipulation, thus minimizing the risk of intraoperative rupture. None of the 4 patients in our series experienced intraoperative bleeding from the aneurysm after trapping/EC-IC bypass performed during the acute period.

The reported outcomes in most surgical studies are poor because of a high incidence of pre- and postoperative rebleeding, intraoperative premature rupture, and ischemic complications following ICA sacrifice.1,4,10,12,15 Accordingly, we believe that trapping/EC-IC bypass during the acute SAH period could be considered the definitive treatment for ruptured BBAs. Our experience indicates that adjusting the timing of this type of surgery from the chronic to the acute period has benefits for patients with such lesions.

Conclusions

To the best of our knowledge, the present report is the first to document the value of trapping/EC-IC bypass for treating ruptured BBAs during the acute period. Although our series includes only a few patients and a larger population is essential for further assessment, we believe that this surgical strategy represents a promising option for treating such rare but high-risk aneurysms.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author contributions to the study and manuscript preparation include the following. Conception and design: K Kamijo. Acquisition of data: K Kamijo. Analysis and interpretation of data: K Kamijo. Drafting the article: K Kamijo. Critically revising the article: K Kamijo, T Matsu. Final approval of the article: T Matsu. Study supervision: T Matsu.

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