Occult rupture of a giant vertebral artery aneurysm following proximal occlusion and intrasaccular thrombosis

Case report

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✔ The authors describe a unique clinicopathological phenomenon in a patient who presented with an unruptured giant vertebral artery aneurysm and who underwent endovascular proximal occlusion of the parent artery followed, several days later, by surgical trapping of the aneurysm after delayed subarachnoid hemorrhage (SAH). The intraoperative finding of a thrombus extruding from the wall of the aneurysm at a site remote from the origin of the SAH underscores the possibility that occult rupture of an aneurysmal sac can occur in patients with thrombosed giant aneurysms.

KEY WORDS • giant aneurysm • vertebral artery • ruptured aneurysm • subarachnoid hemorrhage

TRADITIONALLY, aneurysm rupture is thought to occur via a warning leak or frank hemorrhage and is associated with a classic clinical picture of SAH, namely a sudden, severe headache with or without collapse or neurological deficit.[3,12] We report on a woman who presented with an unruptured giant VA aneurysm that was initially treated endovascularly by proximal parent artery occlusion, and who subsequently experienced two clinical deteriorations: the first manifested as delayed hemiplegia from presumed aneurysm swelling after extensive induced intrasaccular thrombosis, and the second manifested as delayed SAH for which she underwent emergency surgical treatment. Strikingly, during surgery we found a mass of thrombus extruding from the disrupted aneurysm sac at a site distinctly remote from the origin of the SAH. This previously undescribed clinicopathological phenomenon indicates that occult rupture of the thrombosed giant aneurysm had occurred in the days prior to the SAH and emphasizes the hazards associated with incomplete exclusion of the aneurysm from the circulation.

Case Report

History. This 27-year-old right-handed woman presented to our hospital with a 6-week history of increasing up-

per extremity weakness and incoordination, unsteadiness of gait, dysphagia for bulky foods, intermittent cough, and episodes of limb paresthesias and spasms. During the 48 hours before her presentation, she had experienced relatively rapid progression of weakness and incoordination in all extremities, some neck stiffness, right occipital headache, a weakening cough, and shortness of breath at rest. Her medical history was unremarkable except for benign orthostatic hypotension. She had no history of acute-onset, severe headache and no family history of aneurysm or SAH.

Initial Clinical Evaluation. The patient was evaluated in our emergency room and was found to be alert and stable hemodynamically, with a Glasgow Coma Scale[20] score of 15. Results of her cranial nerve examination included nystagmus on left gaze, absent gag reflex, and mild sternocleidomastoid muscle weakness bilaterally. There was moderate neck stiffness, mild unsteadiness of gait, proximal and distal weakness in all extremities, hypertonia of her lower extremities, widespread marked hyperreflexia, bilateral positive Babinski sign, and unsustained ankle clonus on the right side. A CT scan demonstrated a well-defined 3-cm lesion located at the level of the medulla and pons that was associated with mild obstructive hydrocephalus. There was no radiological evidence for SAH. A nonthrombosed 3-cm giant aneurysm situated anterior to the medulla was demonstrated on MR imaging and MR angiography (Fig. 1).

Further Evaluation. The patient was admitted to the hospital (Day 0) for further evaluation and treatment. Four-vessel cerebral angiography performed on Day 1 revealed

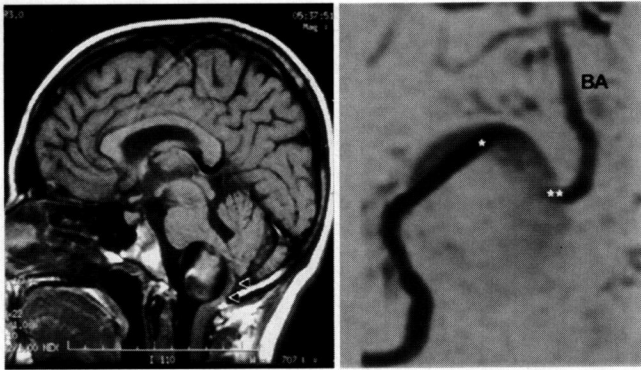# Occult rupture of a giant aneurysm following proximal occlusion

FIG. 1. Diagnostic MR studies obtained at the time of admission (Day 0). *Left:* Noncontrast T$_1$-weighted midline sagittal MR image demonstrating complex flow pattern and flow void in the aneurysm. Note the marked compression and posterior displacement of the medulla *(arrowheads)*. *Right:* Time-of-flight MR angiogram revealing entry *(single asterisk)* and exit points *(double asterisk)* of radiographically normal portions of the right VA relative to the aneurysmal portion.

FIG. 2. Right VA angiograms (Day 1). Anteroposterior *(left)* and lateral views *(right)* demonstrating a 3 × 3 × 3.3–cm saccular aneurysm arising from the distal right VA. There was no evidence for the presence of intraaneurysm thrombus. The aneurysm orifice was estimated to be approximately 5 to 10 mm wide; there was no angiographically definable neck. The right posterior inferior cerebellar artery (not demonstrated in these images) was found to originate directly from the aneurysm.

a 3 × 3 × 3.3–cm nonthrombosed saccular aneurysm arising from the distal right VA that was 5 to 10 mm proximal to the origin of the BA (Fig. 2). Widely patent PCoAs were demonstrated; however, the left VA did not contribute to the intracranial circulation. Because of the size and location of the aneurysm, its wide base arising from the sole vertebrobasilar artery trunk, and the presence of large PCoAs, endovascular proximal parent artery occlusion was selected as the initial treatment. A provision was made, however, for the possibility of surgical repair of the aneurysm with or without hypothermic circulatory arrest and cardiopulmonary bypass if trial balloon occlusion of the right VA failed in this patient.

Endovascular Treatment. The patient tolerated several minutes of right VA balloon occlusion on Day 2 (Fig. 3), after which a detachable balloon catheter system was advanced through the right VA to the C1–2 level. The balloon was inflated for 35 minutes, resulting in extracranial right VA occlusion. The patient remained neurologically unchanged throughout the procedure. As expected, there was excellent retrograde filling of the BA through large bilateral PCoAs, and some antegrade filling of the intracranial right VA through right external carotid artery and VA cervical muscle branches. The latter were believed to be desirable to minimize the risks associated with occlusion of perforating arteries proximal to and at the site of the aneurysm. The balloon was therefore detached at this level, after which four platinum microcoils were advanced and deposited in the right VA immediately proximal to the detached balloon. Apart from brief, transient nystagmus and dysarthria immediately following the procedure, the patient remained stable neurologically and was transferred to the intensive care unit for observation. She was maintained on heparin infusions for 3 days to minimize the risk of clot propagation from the aneurysm into the BA trunk.

Clinical Course After Proximal Occlusion. The first signs of clinical deterioration in the patient following proximal VA occlusion became evident on Day 7, approxim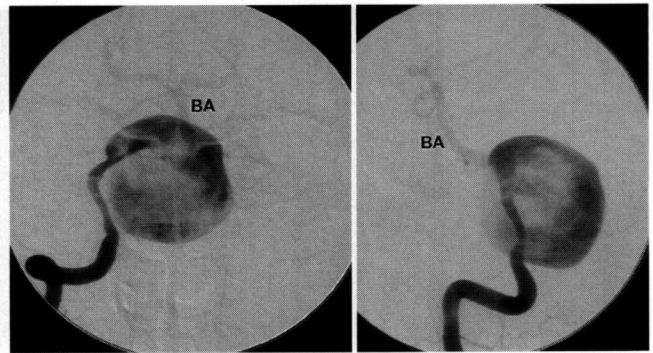ately 5 days after VA occlusion and 2 days after discontinuation of heparin infusion. At this time, moderate left-sided hemiparesis was apparent, as were sinus bradycardia (40 beats/minute) and postural hypotension; there were no other clinical changes. By Day 8, the left-sided hemiparesis had progressed and the patient reported worsening dysphagia. A head CT scan with addition of contrast demonstrated extensive new thrombosis that filled approximately 90% of the aneurysm sac, with residual contrast agent in the superior aspect of the aneurysm. The patient was started on high-dose intravenous steroid medications, which had a good effect in reducing the left hemiplegia over the subsequent 24 hours. The patient remained neurologically and hemodynamically stable for 48 hours more, and on Day 11 MR imaging (Fig. 4 *left*) and MR angiography (Fig. 4 *center*) were performed to assess the status of the aneurysm; extensive intrasaccular thrombosis with a persistent vascular channel were noted. On the evening of Day 12 while at rest, the patient experienced acute, severe cervicothoracic pain radiating up to her occipital and frontal regions, along with neck stiffness, nausea, and dyspnea. Neurological examination revealed no new focal deficit, but SAH was suspected. A CT scan revealed a small hemorrhage in the fourth ventricle and perimesencephalic cisterns (Fig. 4 *right*). The patient was classified as Grade II according to the World Federation of Neurosurgical Societies system.[6] We decided to operate the following morning, with provisions made for cardiopulmonary bypass, profound hypothermia, and circulatory arrest.

Surgical Treatment. Twelve hours after her SAH (that is, on Day 13), the patient underwent right retromastoid suboccipital craniectomy and first cervical hemilaminectomy for thrombectomy and trapping of the aneurysm. Strikingly, during exposure of the sac a large mass of subacute thrombus was noted on the mediolateral surface of the aneurysm, extruding from a site of focal disruption in the thin aneurysm wall (Fig. 5). Although the entry point of the right VA was identified in the upper third of the aneurysm laterally, its exit point medially could not be readily identified. In the course of dissecting this region, acute,
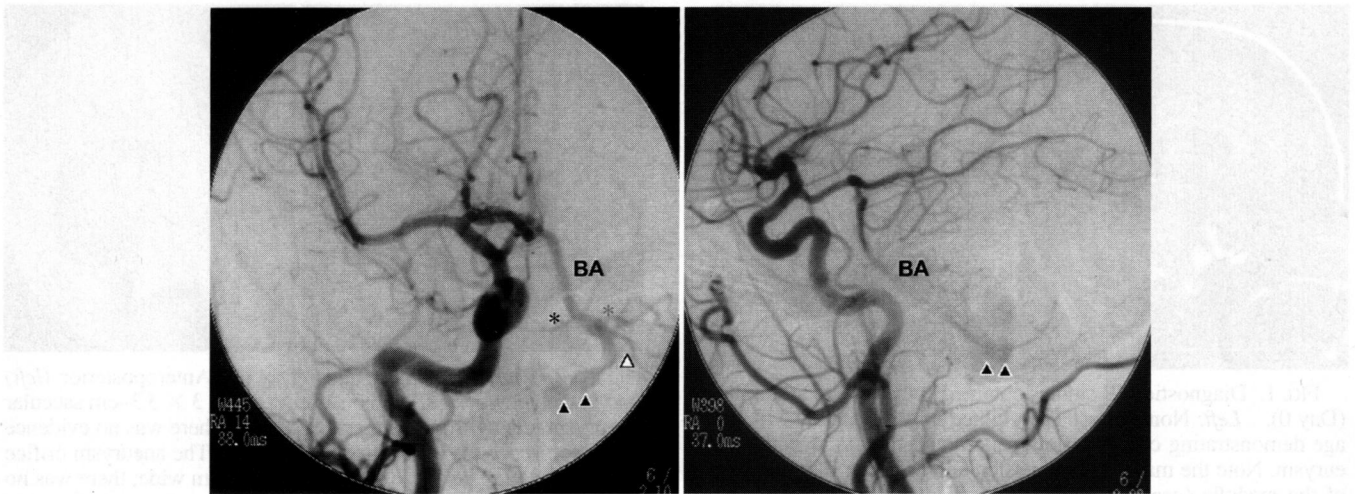
FIG. 3. Right carotid artery angiograms (Day 2). Anteroposterior *(left)* and lateral views *(right)* views obtained during trial balloon occlusion of the right VA at the C1–2 level demonstrating retrograde flow along the BA with faint retrograde opacification of the superior aspect of the aneurysm *(black arrowheads)*. Right *(black asterisk)* and left *(gray asterisk)* anterior inferior cerebellar arteries, and a diminutive left VA *(white arrowhead)* are also apparent.

brisk bleeding occurred from the superior pole of the aneurysm, which was suspected to be the site of the SAH. Bleeding was controlled and the cardiac surgery team proceeded with thoracotomy and initiation of cardiopulmonary bypass and hypothermia. With the patient's body temperature cooled to 17°C, circulatory arrest was required for 15 minutes for further dissection and definitive treatment of the aneurysm. During exploration while the patient was in circulatory arrest, the site of hemorrhage was confirmed at the superior pole of the aneurysm near the exit of the distal right VA. This was distinctly remote from the previously noted site of sac disruption and thrombus extrusion. A thorough evacuation of the remainder of the intraaneurysmal thrombus was accomplished after trapping and restoration of the circulation.

Postoperative Course. The patient was discharged from the hospital 1 month after her operation. Her only deficits (all right-sided) were tongue, vocal cord, and shoulder weakness, and dysphagia that required temporary nasogastric feeding tube placement. She experienced no sensory loss and no impairment of cognitive function. Her 3-month follow-up MR studies (Fig. 6) demonstrated findings consistent with aneurysm obliteration and retrograde filling of the BA. At 2.5 years after her operation, the patient has made a complete neurological recovery, and leads an active and normal life.

Discussion
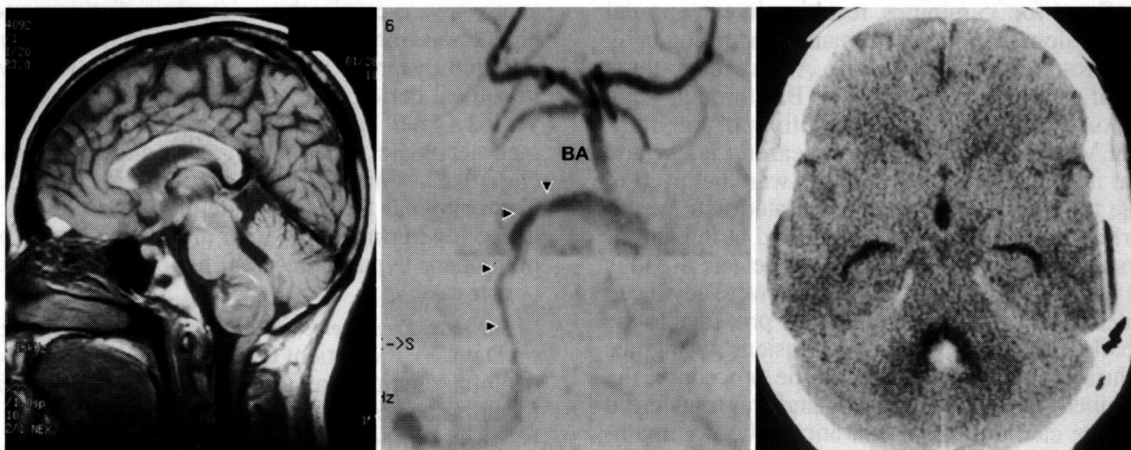
The unique phenomenon reported in this case was the

FIG. 4. Thrombosis (Day 8) and rupture (Day 12) of aneurysm following proximal occlusion (Day 2) of the right VA. *Left:* Noncontrast T_1-weighted midline sagittal image demonstrating (compared with Fig. 1 *left*) extensive thrombus filling the aneurysmal sac except at its superior aspect. *Center:* Phase-contrast MR angiogram demonstrating a residual channel of blood flow along the lateral and superior aspect of the aneurysm *(arrowheads)*. *Right:* Noncontrast axial CT scan demonstrating extension of high-attenuation hemorrhage into the fourth ventricle and over the tentorium. The ventricular size is normal.
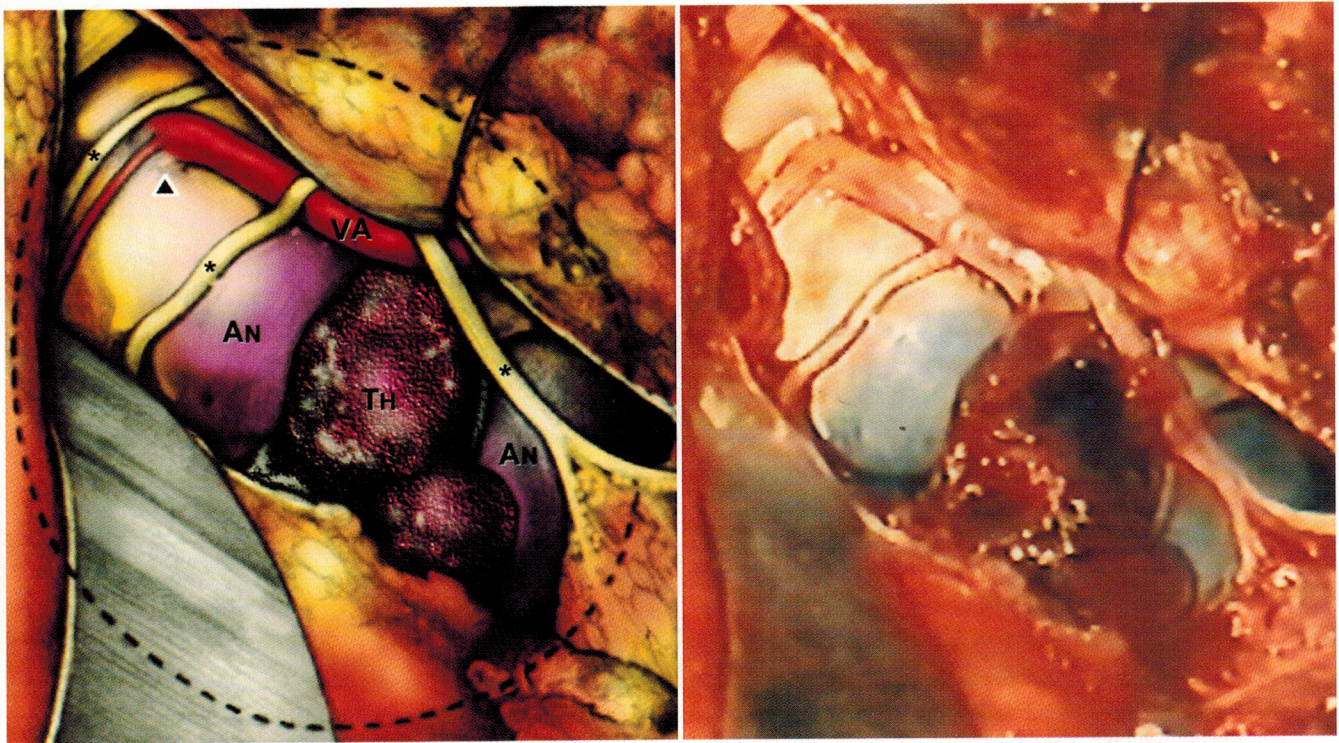
FIG. 5. Views of the surgical field (Day 13). Artistic rendering *(left)* of the intraoperative photograph *(right)*, showing the aneurysm (AN) with its extruded thrombus (TH), as viewed following right retromastoid suboccipital craniectomy. Also shown is the right VA up to its point of entry distally *(arrowhead)* into the aneurysm. The *asterisk* denotes right lower cranial nerves X and XI, whereas the *broken line* depicts the extent of unexposed aneurysm sac.

silent disruption of the aneurysm wall after extensive intrasaccular thrombosis, which resulted in partial extrusion of the clot in the absence of frank SAH. Furthermore, the actual SAH occurred several days later from a separate site in the disrupted aneurysm. We speculate based on the clinical course, neurological studies, and intraoperative findings that the occult aneurysm disruption was due to a combination of expansion of the aneurysm sac and alteration in local hemodynamic stress that occurred in conjunction with the extensive thrombosis.

Giant Aneurysm Thrombosis

Up to 75% of giant intracranial aneurysms present clinically with mass effect.[3] Thrombosis is known to occur spontaneously in approximately 50% of giant aneurysms,[1,21] and is the therapeutic goal of intraaneurysm embolization[14,18] or proximal parent artery occlusion.[2,5,10,11] Intrasaccular thrombosis in these lesions may cause clinical sequelae due to compression of the local structures or impairment of blood flow in segmental or distal branches of the artery or surface parenchymal vessels.[13,15,23] Also, extensive intraaneurysm thrombosis is known not to preclude aneurysm rupture[12] or, in certain cases, aneurysm growth. Although our patient presented with a nonthrombosed giant aneurysm, extensive intrasaccular thrombosis occurred following endovascular treatment and within 48 hours of cessation of heparin infusion. Neurological deterioration that occurred at this time was attributed to increased brainstem compression by the less compliant and perhaps expanded aneurysm mass.

Mechanisms of Growth and Rupture of Thrombosed Giant Aneurysms

The process of thrombosis can itself cause physical expansion of an aneurysm sac by converting its contained fluid phase (circulating blood) into a solid phase composed of a lattice of cellular and extracellular products.[16,23] If thrombosis is complete, clot retraction may eventually occur, in which an aneurysm undergoes involution caused by progressive fibrosis with reduction of the compressive mass.[18] In the event of incomplete thrombosis, however, aneurysm growth and rupture can occur, which may be fatal.[8,13,17–19,22] Incomplete thrombosis leading to growth and rupture of the sac may be facilitated by the presence of collateral arteries feeding a residual lumen, recanalization of the thrombus, and/or recurrent intramural hemorrhages from a vascularized aneurysmal wall.[1,10,11,15,22] In aneurysms that have been treated by proximal parent artery occlusion, however, another particularly important mechanism may account for growth and rupture. This process, associated with morphological conversion of an aneurysm from lateral to terminal,[4] involves exaggerated hemodynamic stress at the junction between retrograde filling and the intrasaccular thrombus, akin to a pulsatile water-hammer effect at this point.[4,7,8] In our patient, extensive but incomplete thrombosis with preservation of some antegrade flow via muscular VA collateral vessels distal to C1–2 was in accordance with the preoperative plan. Unfortunately, in this case and as has been reported previously by others,[9,18,19,21] the presence of such a channel probably contributed to aneurysm rupture. We suspect that there was in-
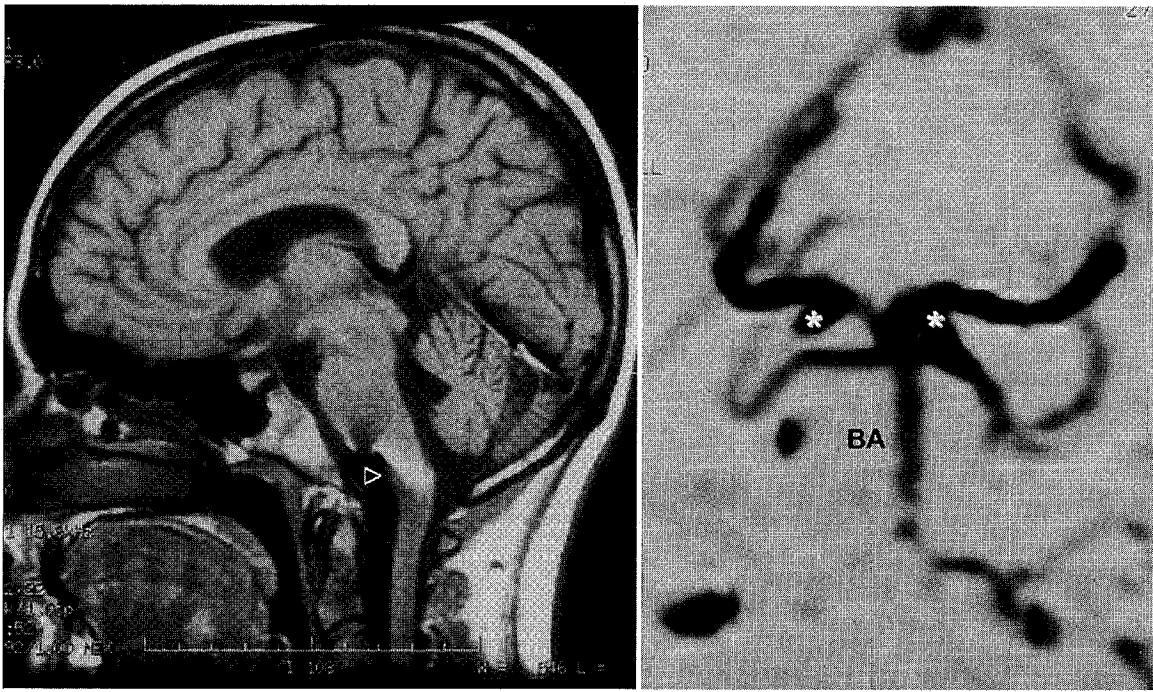
FIG. 6. Three-month follow-up MR studies. *Left:* Noncontrast T₁-weighted midline sagittal MR image obtained following trapping and excision of the aneurysm, demonstrating (compared with Fig. 1 *left*) resolution of the associated mass effect on the brainstem. Some clip artifact is apparent *(arrowhead)*. *Right:* Time-of-flight MR angiogram demonstrating flow in the BA and successful exclusion of the aneurysm. *Asterisks* denote the PCoAs.

creased hemodynamic stress caused by opposing retrograde filling of the aneurysm from the BA that contributed to a water-hammer effect at the superior pole of the aneurysm, which was the eventual site of the SAH.

Occult Rupture

Although our patient experienced only one episode of SAH preoperatively, the observation of a mass of subacute thrombus extruding from the aneurysm sac at its mediolateral portion was evidence of its previous occult disruption. Notably, this site was remote from the origin of the SAH as identified intraoperatively. We suspect that silent rupture of the sac occurred at some time between Days 4 and 12, and although this disruption may or may not have coincided with the clinical signs of exaggerated neurological deficit, it was certainly occult in terms of the absence of clinical or radiological evidence for SAH.

Conclusions

We describe for the first time a patient in whom rupture of an extensively but incompletely thrombosed aneurysm occurred multifocally, including one clinically occult episode. This clinicopathological phenomenon provides evidence that in thrombosed giant aneurysms, rupture of the sac is not necessarily associated with clinical signs of warning leak or frank SAH. Although our young patient made a complete recovery after the staged treatments, her case emphasizes the need to eliminate these treacherous lesions as completely as possible. This leads us to recommend definitive treatment consisting of exclusion of the aneurysm from the circulation either by clipping or surgi-

cal or endovascular trapping. If endovascular treatment is preferred, complete trapping of the aneurysm should be the goal rather than preservation of partial flow. Our concept of preserving antegrade flow through the aneurysm via proximal collateral vessels seems unwise if the collateral vessels for retrograde flow are robust, at least in cases of suspected thin-walled saccular aneurysms (especially in young patients). The preservation of some antegrade flow is more appropriate in those patients with marginal retrograde collateral vessels or suspected thick-walled atherosclerotic fusiform aneurysms.

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