Surgical Intervention for Angiographically Unexplained Subarachnoid Hemorrhage

—Report of Three Cases—

Nobuyuki TAKAI, Isamu EZUKA, Takatoshi SORIMACHI
and Takashi KUMAGAI

Department of Neurological Surgery, Niigata Rosai Hospital, Joetsu, Niigata

Abstract

We report three patients with subarachnoid hemorrhage in which no causative lesion was detected angiographically. Surgical intervention revealed a ruptured aneurysm in all three cases, which were clipped successfully. Although the prognosis for a patient with subarachnoid hemorrhage of unknown cause is good, there still remain long-lasting unpleasant complaints probably arising from psychogenic factors. We especially recommend exploratory craniotomy when subarachnoid hemorrhage is located anterior to the circle of Willis, accompanied by thick and obviously lateralizing subarachnoid clots on computed tomography and/or rebleeding in the same anatomical location, even if no causative lesions can be found angiographically.

Key words: subarachnoid hemorrhage, surgical management, thrombosed aneurysm, unknown etiology

Introduction

Subarachnoid hemorrhage (SAH) of unknown origin accounts for 5–20% of all SAH.³¹⁾ Previous discussion of the prognostic factors for SAH of unknown origin has concluded that if a technically adequate and complete angiography is performed in the first place, a repeat study is unnecessary.^{3,4,9–11,16)} Because of the good prognosis, operative findings and indications have been little discussed.^{6,17,23,24)}

We report the operative findings of three patients with SAH of unidentified origin angiographically and discuss treatment policy for such cases.

Case Reports

Case 1: A 48-year-old hypertensive male was well until May 21, 1991, when he suffered sudden onset of severe headache. Returning home he lost consciousness for 1 hour and was admitted to our

hospital within 2 hours of the ictus. Computed tomographic (CT) scans revealed SAH with dominant distribution of clots in the left Sylvian fissure (Fig. 1). A ruptured aneurysm of the left middle cerebral artery (MCA) was suspected from the distribution of the clots. His clinical status was grade 3 on the Hunt and Hess scale. On the same day, panangiography revealed a small right MCA aneurysm, but the left carotid and vertebral angiograms were normal (Figs. 2 and 3).

Operation on day 0 *via* the right pterional approach found no evidence of rupture of the right MCA aneurysm. Immediately after clipping of the aneurysm, *via* the left pterional approach the thick clots within the left Sylvian fissure were removed and subarachnoid drainage emplaced. A small thrombosed aneurysm with thin-walled neck was found at the left M₁–M₂ junction and was clipped.

Postoperatively, his mental activity was diminished for several days. He was discharged on June

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Authors' present addresses: T. Sorimachi, M.D., Department of Neurosurgery, Brain Research Institute, Niigata University, Niigata, Japan; T. Kumagai, M.D., Department of Neurosurgery, Mito Saiseikai General Hospital, Mito, Japan.

Fig. 1 Case 1. CT scans, showing subarachnoid clots spreading to the left Sylvian fissure and basal cistern, distributed predominantly within the Sylvian fissure.

Fig. 4 Case 2. CT scans, showing subarachnoid clots spreading to the basal, interpeduncular, and bilateral Sylvian cisterns, distributed predominantly within the right Sylvian fissure.

28. He is now well without neurological deficits.

Case 2: A 59-year-old hypertensive male suffered sudden onset of severe headache when he went to the bathroom in the early morning of November 6, 1988. He was admitted to our hospital on the same day. Immediate CT scans showed SAH with dominant distribution of clots in the basal cistern and the right Sylvian fissure (Fig. 4). A ruptured MCA or internal carotid artery (ICA) aneurysm was suspected (Hunt and Hess grade 3). However, cerebral panangiography on the following day discovered no aneurysm (Fig. 5).

The arachnoid membrane over the Sylvian fissure was dissected extensively *via* the right pterional approach to examine the intra-Sylvian portion of the MCA, ICA, and anterior communicating artery, but no abnormality was found. When the ICA was gently retracted laterally to remove clots between the ICA and optic nerve, arterial blood spouted out. The M_1 ,

A_1 , and ICA were temporarily clipped, and the entire surface of the ICA investigated. A miliary aneurysm was found on the rear surface of the ICA independent of both the posterior communicating and anterior choroidal arteries. This aneurysm was clipped with a Sugita fenestrated clip.

His postoperative course was uneventful until November 15, when he became semicomatose with left hemiplegia. CT scans showed marked cerebral edema in the right hemisphere and multiple infarcts in bilateral hemispheres. He became bedridden with akinetic mutism and died from multiple organ failure on January 19, 1989.

Case 3: A 75-year-old hypertensive female was well until March 27, 1985, when she suffered sudden onset of severe headache. She presented complaining of persistent headache on April 1. CT scans showed high-density spots within the left Sylvian fissure (Fig. 6 left). The diagnosis was SAH due to a ruptured

Fig. 2 Case 1. Right carotid angiogram, anteroposterior view, demonstrating a small aneurysm in the M_1 portion.

Fig. 3 Case 1. Left carotid angiograms are normal. *left*: Anteroposterior view, *right*: oblique view.

Fig. 5 Case 2. Right carotid angiograms are normal. *left*: Anteroposterior view, *right*: oblique view.

Fig. 6 Case 3. *left*: CT scan, showing high-density spots in the posterior portion of the left Sylvian cistern. *right*: CT scan, showing localized clots within the left Sylvian fissure.

Fig. 7 Case 3. Left carotid angiograms are normal. *left*: Anteroposterior view, *right*: lateral view.

MCA aneurysm (Hunt and Hess grade 2). She suffered transient unconsciousness when rebleeding occurred on April 3. CT scans demonstrated localized clots in the left Sylvian fissure (Fig. 6 *right*). On the next day, direct bilateral carotid and left

transbrachial vertebral angiograms found no aneurysm (Fig. 7).

Operation *via* the left pterional approach clipped a thrombosed small aneurysm located at the junction of the M₂ branches. Her postoperative course was uneventful and she was discharged without neurological deficits.

Discussion

Rebleeding is rare in SAH of unknown origin compared to aneurysmal SAH, less than 9.1% in the former, but 50% per year within 6 months and 3% per year after 6 months in the latter.^{7,10,12,15,16,18,19} The long-term prognosis is also distinctly favorable in contrast to that of aneurysmal SAH. Therefore, surgical management is contraindicated.

There are several possibilities for the pathogenesis of SAH of unknown cause: rupture of the fine ventriculostriate and thalamoperforating vessels¹⁾; bleeding from a small aneurysm followed by spontaneous thrombosis^{5,6,24,26,28,29)}; bleeding from a microaneurysm (<2–3 mm in diameter)^{13,14,23)}; perimesencephalic nonaneurysmal SAH²¹⁾; hemorrhagic necrosis of a pituitary adenoma²⁾; and rupture of dilated veins of dural arteriovenous malformations.³²⁾

Sakai *et al.*²³⁾ emphasized the necessity of surgical intervention for SAH with an unknown cause because of such pathogeneses. Previous reports found either a thin portion or a small protrusion of the arterial wall in four patients,²³⁾ or aneurysms including two thrombosed small aneurysms,⁶⁾ two non-thrombosed aneurysms (5 and 6 mm),¹⁷⁾ and one thrombosed and one non-thrombosed small aneurysm and two other aneurysms.²⁴⁾ All our cases had ruptured aneurysms: two thrombosed small and one miliary aneurysms. Ruptured aneurysms were surgically confirmed in 11 of 16 reported cases despite the negative angiographic findings. Prognostic factors for SAH due to a ruptured aneurysm include the degree of hemorrhage and other factors such as mural arterial hemorrhage and genetic factors.²⁷⁾ Patients with SAH of unknown cause tend to present with a better clinical condition than those with aneurysmal rupture.^{7,12,18)} However, the origin of SAH of unknown cause may be a ruptured aneurysm, and poor prognosis may be predicted in cases with massive subarachnoid clots.^{8,20,30)}

We decided to operate on our three patients in the acute stage despite negative angiographic findings because of the dangers of severe vasospasm, considering the dense clots revealed by CT, and rebleeding. The craniotomy in Case 3 was performed because of

clots distributed in the same anatomical location on CT scans after rebleeding on day 7. As rebleeding had occurred, a second rebleeding was probable when thrombolysis began. Surgery in Case 1 was intended to remove dense clots, and revealed the thrombosed aneurysm with a thin-walled neck. A new aneurysm is likely to form from a weak neck even if the aneurysm is thrombosed. The outcome for Case 2 was miserable due to severe vasospasm, which may have been aggravated by excessive manipulation providing mechanical stimuli to arteries because the aneurysm was difficult to access.

Despite the good prognosis for SAH of unknown cause, unpleasant symptoms such as frequent headache, vertigo, and increased fatigue were experienced in 21–50% of patients.^{3,7,16,22,25)} The highest incidence of recurrent headache was 74%.¹⁸⁾

Based on our experience, we recommend exploratory craniotomy for angiographically unexplained SAH because: 1) subarachnoid clots frequently associated with SAH can be removed; 2) rebleeding can be prevented, if the causative lesions are detected; 3) unpleasant symptoms probably arising from psychogenic factors may be reduced by diminished anxiety about unsolved pathogenesis; and 4) less anxiety for the attending surgeon and physician. However, excessive manipulation of arteries should be avoided. We especially recommend exploratory craniotomy when SAH is located anterior to the circle of Willis, accompanied by thick and obviously lateralizing subarachnoid clots on CT scans and/or rebleeding in the same anatomical location, even if no causative lesions can be found angiographically.

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Address reprint requests to: N. Takai, M.D., Department of Neurological Surgery, Niigata Rosai Hospital, 1-7-12 Toun-cho, Joetsu, Niigata 942, Japan.