# Late Recurrence of Subarachnoid Hemorrhage Associated with Aneurysmal Regrowth Originating from the Anterior Communicating Artery after Neck Clipping

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**Abstract:** While complete neck clipping of ruptured cerebral aneurysms prevents rerupture, recurrent subarachnoid hemorrhage (SAH) due to the rupture of an aneurysm at the same sites of the previous aneurysm may occur, even if no residual neck was detected during and after neck clipping. We present the cases of 3 patients with recurrent SAH due to regrowth after neck clipping of aneurysms at the anterior communicating artery (AComA). Two of the recurrent AComA aneurysms, accessed *via* the pterional approach, arose from the arterial wall adjacent to the previous necks; the third was a de novo aneurysm. Because their growth was in a posterior or superior direction, we posit that a portion of the neck was inadvertently not clipped during the first operation, and thus, flow in the AComA and perforators to the hypothalamus was maintained. Our findings suggest that unrecognized residual necks, vascular wall fragility around the earlier aneurysm, and hemodynamic changes following neck clipping contributed to their recurrence of SAH. Therefore, we recommend follow-up angiographic study to confirm complete neck clipping, especially in patients in whom the pterional approach was used.

Keywords: Cerebral aneurysms, recurrence, subarachnoid hemorrhage, neck clipping, anterior communicating artery.

## INTRODUCTION

Complete neck clipping of cerebral aneurysms is thought to prevent aneurysmal regrowth or rerupture. Recurrent subarachnoid hemorrhage (SAH) due to the rupture of aneurysms treated by neck clipping [1-5] can be classified as either early (within 1 month) or late recurring (more than 1 year after the clipping surgery) [4, 6]. Late recurrence is reportedly associated with de novo aneurysms, and regrowth of a treated aneurysm [1, 5, 7]. Although the mechanisms underlying this type of recurrent SAH have been studied, few topographical analyses have been conducted because the number of cases of these aneurysms is small. Here, we present 3 additional patients with recurrent SAH due to the regrowth of aneurysms at the anterior communicating artery (AComA) after neck clipping. We hypothesize that AcomA aneurysms impede blood flow both in the parent artery, and in perforators to the hypothalamus, and that neck clipping of these aneurysms is anatomically complex.

## **CASE PRESENTATION**

#### Case 1

This 44-year-old woman was admitted immediately after the sudden onset of headache. Computed tomography (CT) revealed SAH in the interhemispheric and basal cisterns (Fig. 1A). She manifested Hunt and Kosnik (H&K) grade I,

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and was alert and without neurological abnormalities. Angiography revealed a small aneurysm at the A1-2 junction (Fig. **1B**). It was obliterated with a Sugita clip, and clipping was thought to be complete (Fig. **1C**). She was discharged without any neurological deficits. Eight years later, she was admitted again; she was comatose and manifested recurrent SAH (Fig. **1D**) with massive intraventricular hemorrhage (Fig. **1E**). Angiography showed that another aneurysm had developed, again in a superior-posterior direction; it was adjacent to the previously clipped aneurysm (Fig. **1F**). Neck clipping was not performed because medical treatment yielded no neurological improvements.

#### Case 2

This 65-year-old woman presented with severe headache. CT showed diffuse SAH in the interhemispheric and basal cisterns (Fig. 2A). She was alert with an H&K grade of II, and there were no neurological findings. Angiography revealed multiple aneurysms at the AComA and both middle cerebral arteries (MCAs) (Fig. 2B). Her SAH was thought to be attributable to the AComA aneurysm, whose neck was thought to have been clipped completely with 2 Sugita clips (Fig. 2C). The 2 unruptured aneurysms at the left MCA were also obliterated. One year later, she was admitted again; she was comatose and had recurrent SAH (Fig. 2D). Although there was no angiographic evidence of aneurysm (Fig. 2E), angiographs obtained 2 weeks later showed an AcomA aneurysm growing in a superior-posterior direction (Fig. 2F). Its neck was clipped immediately, and another Sugita clip was applied. Sacrifice of perforators into the hypothalamus re-

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**Fig. (1). (A)** A CT scan following the first attack clearly showed SAH at the interhemispheric and basal cisterns. (**B**) Angiography revealed a small aneurysm at the A1-2 junction projecting in the anterior-superior direction (arrow). (**C**) Postoperative angiogram demonstrated the disappearance of the ruptured aneurysm. A CT scan after the second attack also revealed SAH at interhemispheric cistern (**D**) and intraventricular hemorrhage (**E**). (**F**) Angiogram showing a recurrent aneurysm between the previously placed clip and the A1-2 junction; it projects in the superior direction (arrow). (**G**) Schematic drawing of the operative field accessed *via* the pterional approach on the left side. The aneurysm projects in the anterior-superior direction. We posit that the recurrent aneurysm developed from the posterior portion of the left A1-2 junction. A1 and A2; A1 and A2 segments of the anterior cerebral artery, AComA; anterior communicating artery.

sulted in a small infarction. Two months later, she was transferred to the rehabilitation center, with severe disability.

## Case 3

This 52-year-old woman was admitted at another hospital for headache and vomiting. CT showed SAH in the suprachiasmatic and anterior interhemispheric cisterns (Fig. **3A**). She was alert without any neurological abnormalities; her H&K grade was II. The neck of a ruptured AComA aneurysm, projecting anterior-inferiorly, was clipped using a Sugita straight clip (Fig. **3B**, **C**). She recovered well and was discharged. Eight years later, she was referred to our hospital because of sudden-onset headache and vomiting. CT revealed only intraventricular hemorrhage in the right lateral ventricles (Fig. **3D**). Angiography demonstrated an AComA aneurysm with a superior-posterior projection (Fig. **3E**, **F**). The neck of the newly developed aneurysm was completely separate from the clip applied at the first operation, and the de novo AComA aneurysm was successfully clipped, with-



Fig. (2). (A) A CT scan performed following the first attack revealed diffuse SAH in the interhemispheric and basal cisterns. (B) Left carotid angiogram showed multiple aneurysms at the AComA and MCA. The AComA aneurysm projected in the inferior-anterior direction (arrow). (C) Postoperative angiogram demonstrating the disappearance of the ruptured AComA aneurysm. (D) A CT scan performed after the second attack revealed SAH in the interhemispheric and carotid cisterns. (E) Left carotid angiogram. There is no clear evidence of a recurrent or de novo aneurysm. (F) Left carotid angiogram obtained 2 weeks after the second SAH showed that the AComA aneurysm had grown in the posterior-superior direction (arrow). (G) Schematic drawing of the operative field accessed *via* the interhemispheric approach. A new aneurysm developed at the posterior portion of the AComA, and projected in the superior-posterior direction. A1 and A2; A1 and A2 segments of the anterior cerebral artery, AComA; anterior communicating artery.

out occluding the hypothalamic arteries. A ventriculoperitoneal shunt was placed 3 weeks after the second operation. She was discharged and remained asymptomatic in the course of a 7-year follow-up.

## DISCUSSION

Recurrent SAHs at the same site of previously ruptured and clipped aneurysms has been reported, even in the absence of any residual neck, and possible underlying mecha-

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**Fig. (3). (A)** CT scan performed after the first attack showed iso-density in the suprachiasmatic and interhemispheric cisterns. (**B**, **C**) Left carotid angiogram demonstrated a ruptured aneurysm at the AComA with anterior-inferior projection (arrow) (B; anterior-posterior view, C; lateral view). (**D**) A CT scan performed after the second attack revealed only intraventricular hemorrhage in the third ventricle and the bilateral anterior horns of the lateral ventricles. (**E**, **F**) Angiogram obtained at SAH recurrence demonstrated an AComA aneurysm with dorsal posterior projection (arrow) (E; anterior-posterior view, F; lateral view). (**G**) Schematic drawing of the operative field accessed *via* the pterional approach on the left side. At the first operation, the clip was placed parallel to the anterior communicating artery. As there was no regrowth of the previous aneurysm, the recurrent SAH was attributed to a de novo aneurysm that projected in the posterior-superior direction. A1 and A2; A1 and A2 segments of the anterior cerebral artery, AComA; anterior communicating artery.

nisms have been described previously [1-5, 7]. Vascular topographical factors, including the fragility of the parent artery [8], difficulties in ensuring the complete obliteration of aneurysms, and hemodynamic stress should be considered in such cases [4, 9, 10]. Table 1 summarizes the clinical features of 7 previously reported cases, along with our 3 patients, who had SAH recurrence due to the regrowth of an AComA aneurysm after neck clipping. The previous reports involved 5 men and 2 women, who ranged in age from 33-53 years (mean, 42.7 years) at the first SAH. Neck clipping was judged to be

 Table 1.
 Characteristics of 10 Patients with Late-recurring Subarachnoid Hemorrhage Due to Regrowth of an Aneurysm at the Anterior Communicating Artery after Neck Clipping of the Previous Reports, and Our Cases

No.	Age/Sex (1 <sup>st</sup> attack)	H&K	Clipping	Outcome	Interval (years)	Procedure (2 <sup>nd</sup> attack)	Outcome	References
1.	44 / M	*	incomplete	GR	3	clip	GR	21
2.	43 / M	*	incomplete	GR	8	clip	D	21
3.	33 / M	*	incomplete	GR	8	clip	D	21
4.	41 / M	*	complete	GR	13	clip	MD	21
5.	53 / F	II	complete	GR	8	clip	MD	1
6.	50 / F	*	*	GR	17	clip	poor	20
7.	35 / M	*	complete	GR	12	clip	*	19
8.	44 / F	Ι	complete	GR	8	-	VS	our case
9.	65 / F	II	complete	GR	1	clip	SD	our case
10.	63 / F	II	complete	GR	11	clip	GR	our case

GR; good recovery,

I; interhemispheric approach,

MD; moderately disabled,

Ope App; operative approach, P; pterional approach,

SD; severely disabled,

VS; vegetative state.

\*: No data available

complete in 3 and incomplete in 3 cases (unknown in one). All 7 patients harbored a single AComA aneurysm not only at the time of the first SAH, but also the recurrent SAH. All of these patients recovered well after the first SAH. However, after the second bleeding episode, the clinical outcome was good in only 1 patient; 2 survived with moderate disability, 1 was severely disabled, and 2 patients died (unknown outcome in 1 patient). Similarly, the clinical outcome was unsatisfactory in 2 out of our 3 patients after the second SAH. The interval between the first and second SAH in the previously reported cases was 3-17 years (mean, 11.0 years); H&K grade, operative approach, and clips applied were not described [1-4].

In operated patients with late SAH recurrence, 3 factors have been proposed; de novo aneurysms [11,12], regrowth of a residual neck due to incomplete initial neck clipping of the previously ruptured aneurysm [13,14], and changes in the normal structure of the parent artery adjacent to the clipped neck due to chronic hemodynamic stress [4, 9, 10]. Additionally, recurrent SAH at the site of a previously treated aneurysm may be due to breaking or slipping of the applied clip [15,16], fragility of the parent artery adjacent to the aneurysm [8], vessel injury due to repeated temporary clipping [17], or the presence of a residual neck due to incomplete clipping [13,14].

The Cerebral Aneurysm Rerupture After Treatment (CARAT) study compared the rates of recurrent hemorrhage in patients with ruptured aneurysms, in which treated initially with coil embolization or surgical clipping, although this was not a randomized trial [18]. Bleeding from the aneu-

rysm more than 1 year after treatment occurred in 1 patient treated with coiling, but in none treated with clipping. During a follow-up period, the annual rates of recurrent bleeding after the first year were 0.11% and 0% for treatment with coiling and clipping, respectively. The authors concluded that SAH recurrence after complete aneurysm clipping is extremely unlikely. However, if there is incomplete clipping, continued surveillance is warranted.

In the Retreatment of Ruptured Cerebral Aneurysms in Patients Randomized by Coiling or Clipping in the International Subarachnoid Aneurysm Trial (ISAT), a follow-up study of the patients was performed in order to compare the frequency, timing, and consequences of aneurysm recurrence [19]. After neurosurgical clipping, 30 (2.9%) patients were retreated early, and 9 (0.85%) late, 3 (0.3%) after rebleeding, and 6 (0.6%) without. The mean time to retreatment was 5.7 months, while the mean interval between primary treatment and rebleeding was only 2.3 months (range, 2-3 months).

In the Incidence of Recurrent Subarachnoid Hemorrhage After Clipping for Ruptured Intracranial Aneurysms study, out of 752 patients with a total of 6016 follow-up years (mean follow-up, 8.0 years), 18 had recurrence [20]. In the first 10 years after the initial SAH, the cumulative incidence of recurrent SAH was 3.2%. In the 18 patients with a recurrent SAH after successful initial treatment, 19 recurrent aneurysms were found; 13 patients had an aneurysm in a new location, 3 patients had an aneurysm located at the clip site from the previous operation, and 1 patient had both an aneurysm located at a new location and an aneurysm located at the previous clip site. In this study, out of the 4 patients with an aneurysm at the clip site, all were classified as possible regrowths.

In our series, the recurrent AComA aneurysms all projected in the superior-posterior direction. We posit that in cases 1 and 2, a residual neck was allowed to remain, in order not to obliterate the perforators that maintain blood flow to the hypothalamus, and thus, clipping was incomplete at the posterior portion of the AComA, the part at the greatest depth when the pterional approach is used (Figs. **1G** and **2G**). In case 3, the second AComA aneurysm developed at a site separate from the previous neck, and was considered a de novo aneurysm (Fig. **3G**). We posit that local hemodynamic changes in blood flow from the dorsal posterior direction induced by the applied clip, along with local fragility of the arterial wall were factors underlying the development of the new aneurysm [10].

Earlier reports provided no details regarding the operative approach used at the second operation. In our 3 patients the pterional approach was applied for neck clipping after the first SAH. As all of the recurrent AComA aneurysms projected in the superior-posterior direction, we speculate that during the first operation using the pterional approach, the superior-posterior portion of the AcomA masked a part of the aneurysmal neck, resulting in incomplete neck clipping.

The operative approaches used to address the recurrent AComA aneurysms in our patients were as follows. In case 2, the recurrent aneurysm projecting superiorly and posteriorly was approached *via* the interhemispheric route in order to avoid interference of the existing clip in the operative field, which would have rendered additional clip application difficult. In case 3, the recurrent aneurysm also projected in the superior-posterior direction. The pterional approach was appropriate in this case, because the aneurysm was new and the existing clip had been applied on the anterior-inferior surface of the AComA and thus did not hinder application of future clips (Fig. **3G**). Nonetheless, the interhemispheric approach, which yields a wide operative field and allows for inspection of the entire aneurysm, would have been appropriate in this case.

The recurrent SAH due to bleeding from the aneurysm projecting in the posterior-superior direction may have reached the hypothalamus and resulted in the perforation of the ventricles, leading to deterioration in the patients' neurological status. In patients with previous SAH, the risk for SAH recurrence is 2-3 times greater than in the general population [12, 21, 22]. Therefore, we recommend follow-up angiographic study in patients who undergo the pterional approach for aneurysmal neck clipping, even if clipping is believed to have been complete. In patients whose recurrent AComA aneurysm projects in the posterior-superior direction, we recommend that the interhemispheric approach be used, as it yields a wide operative view that is not affected by the presence of the previously placed clip.

## ABBREVIATIONS

A(	ComA	=	Anterior	communicating	g artery
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CT = Computed tomography scan

SAH = Subarachnoid hemorrhage

#### ACKNOWLEDGEMENT

None Declared.

## **CONFLICT OF INTEREST**

The author(s) confirm that this article content has no conflicts of interest.

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Received: April 16, 2012

Revised: June 26, 2012

Accepted: June 28, 2012

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