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Case Report

Rebleeding from clipped aneurysm after 35 years: Report of 2 cases

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Received: 18 June 15 Accepted: 24 June 15 Published: 10 August 15

This article may be cited as:

Ishida A, Matsuo S, Asakuno K, Nemoto A, Niimura K, Yoshimoto H, et al. Rebleeding from clipped aneurysm after 35 years: Report of 2 cases. Surg Neurol Int 2015;6:134. http://surgicalneurologyint.com/Rebleeding-from-clipped-aneurysm-after-35-years:-Report-of-2-cases/

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Abstract

Background: A successfully applied clip for a ruptured aneurysm keeps the aneurysm's neck closed, preventing rerupture throughout the patient's life. Unfortunately, rebleeding from a clipped aneurysm does occur, but the likelihood declines with time. Since relatively old people suffer from subarachnoid hemorrhage, they die from diseases other than rebleeding, such as cancer. Therefore, rebleeding from a clipped aneurysm after two decades is quite rare.

Case Description: Here, we report 2 cases of rerupture after an extremely long time since the initial clipping. In both cases, the old clip was removed, and the regrown gourd-shaped aneurysm was successfully obliterated. The clips in both cases were submitted to their manufacturers and inspected thoroughly. They were found to be second-generation, stainless steel clips, and were almost intact, even keeping their closing forces. In both cases, the clip existed on the surface of the newly made dome, and the previous dome completely disappeared.

Conclusions: We experienced 2 cases of rebleeding from the clipped aneurysm after 35 years. In one of the cases, the clip was a Yasargil second generation stainless steel clip that retained its mechanical properties and surface elemental composition *in vivo* for a long time. These cases should be informative as they show extremely long-term course of a clip applied for a ruptured aneurysm.

Key Words: Long-term follow-up, rebleeding, subarachnoid hemorrhage

Access this article online Website: www.surgicalneurologyint.com DOI: 10.4103/2152-7806.162552 Quick Response Code:

INTRODUCTION

Complete neck clipping for ruptured aneurysms has been the most important procedure in modern neurosurgical history. Until recent dramatic improvement of endovascular obliteration, clipping was the dominant treatment for subarachnoid hemorrhage (SAH), being performed in a large number of cases. SAH caused

by aneurysmal rupture had its peak in their 50's and 60's, and survivors usually died because of cancer or cardiovascular disease. For all that, intracranial aneurysms develop during life and survivors of SAH are at risk for a recurrence. [1,9] In patients with SAH, new aneurysms can develop at a new location (*de novo*) or at the clip site (regrowth). The recurrence usually occurs at a new location, and rebleeding from the clip site is less likely

to cause SAH as time passes.^[1,5] There are a number of reports of SAH caused by rebleeding from the clip site, but those occurred within the first decade after the initial clipping.^[1,2,6,9,10] Because of the aging population of some developed countries, long-term follow-up is becoming a great concern.

Here, we describe in detail the cases of rerupture from the treated aneurysm after 35 years. We also describe the regrown aneurysm and the retrieved clips, which were studied thoroughly by their manufacturers.

CASE REPORTS

Case 1

History and examination

A 75-year-old man had SAH due to left middle cerebral artery aneurysmal rupture and clipping surgery 35 years ago. It appeared that he was doing well afterward; however, he collapsed, and he was then transferred to a nearby hospital. It was determined that he had suffered a second SAH due to a rupture of the original aneurysm [Figure 1a]. At the hospital, he underwent three-dimensional computed tomography (CT) angiography (CTA) for the SAH, revealing a gourd-shaped aneurysm with the old clip attached at its dome [Figure 1b and c]. His family asked for a surgical treatment, and the patient was transferred to our hospital.

Operation

Fronto-temporal craniotomy was performed as in the previous operation. After careful removal of sylvian fissure hematoma, the old clip was identified [Figure 1d]. With a very cautious dissection, the aneurysm appeared in full view. The dome head was buried in the temporal

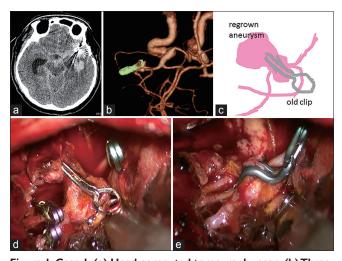


Figure 1: Case 1. (a) Head computed tomography scan. (b) Threedimensional computed tomography angiography showing a gourdshaped regrown aneurysm under the clip (implanted for 35 years) at left middle cerebral artery bifurcation. (c) Schematic image of the regrown aneurysm. (d) Intraoperative photograph showing the old clip with cotton clothes on the regrown aneurysm. (e) A crankshaped clip was applied along the closure line

lobe, causing serious temporal intracranial hemorrhage. Dissection of cotton clothes stuck to the aneurysm was attempted, but that would have caused a premature rupture, and the attempt was abandoned. After trapping the parent arteries, the clip was carefully removed from the dome. The large dome was then cut out, and a crank-shaped clip was applied to make a closure line [Figure 1e].

Postoperative course

The postoperative recovery was uneventful, but intracerebral hematoma caused irreversible brain damage. Even after ventriculoperitonial shunt, the patient has been bedridden since the SAH occurred.

Characteristics of the retrieved clip

The clip was found to be a Yasargil Phynox aneurysm clip (Aesculap AG, Tuttlingen, Germany). It was a second-generation product (code number, FD742), and there was no severe damage or any corrosion [Figure 2a]. X-ray photoelectron spectroscopy (XRP) revealed that it was made of stainless steel, and the constituent of the metal was not changed after extremely long-term implantation [Figure 2b]. Black spots on its surface were speculated to be a residue of organic products [Figure 2c and d]. The closing force was 98 g, which was only 12 g less than the standard force.

Case 2

History and examination

A 72-year-old woman had SAH, and a ruptured anterior communicating artery aneurysm clipped 35 years ago and appeared to have fully recovered afterward. The

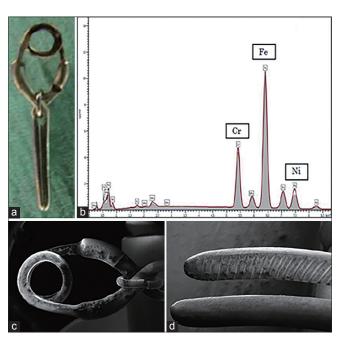


Figure 2: (a) Photographs depicting the retrieved Yasargil clips (FD742). (b) Graphs revealing the profile of the retrieved clip analyzed using X-ray photoelectron spectroscopy. (c and d) Scanning electron micrographs of the retrieved clip

patient presented with a severe headache. CT scanning showed SAH in the interhemispheric fissure, indicating rerupture of the previous aneurysm [Figure 3a]. Subsequent digital subtraction angiography revealed a gourd-shaped aneurysm with the old clip attached at its dome [Figure 3b and c]. Because of the complicated shape, surgical treatment was favored over endovascular coiling.

Operation

Skin incision and craniotomy were performed, as they had been 35 years prior. By removing and washing the hematoma, the interhemispheric fissure was easily opened, and the clip head appeared. Clearing the hematoma away from the clip, the aneurysm appeared in full view [Figure 3d]. When the old clip was removed, premature rupture occurred but it was kept under control. Three straight clips were used to obliterate the aneurysm completely [Figure 3e].

Postoperative course

The patient awoke from the surgery without neurological defects but with minor disturbance of cognitive function, and then returned to normal daily life. The postoperative and follow-up CTA showed no evidence of a residual aneurysm.

Characteristics of the retrieved clip

The clip was determined to be a product of Mizuho Ikakogyo Co., Ltd., (Tokyo, Japan). It was identified as a Sugita clip (Sugita No. 3, 07-940-03). Without XRP spectroscopy, it was concluded that it was Elgiloy (cobalt-chromium alloy) because it was the company's only product 35 years ago, at the time of original implantation. There was no severe damage or any corrosion of the clip [Figure 4a and b]. Careful

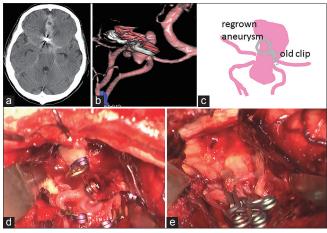


Figure 3: Case 2. (a) Head computed tomography scan. (b) Three-dimensional angiography showing a guard-shaped regrown aneurysm under the clip (implanted for 35 years) at anterior communicating artery. (c) Schematic image of the regrown aneurysm. (d) Intraoperative photograph showing the old clip on the regrown aneurysm. (e) Three straight clips were applied to obliterate the aneurysm completely

observation of the clip surface with scanning electron microscope revealed no abnormality [Figure 4c and d]. The closing force was 129 g, which was within the range of the standard force.

DISCUSSION

Published data on long-term morbidity and mortality of survivors of SAH are still limited. Long-term causes of death in survivors of SAH were largely unrelated to the SAH and were mainly other factors (mostly cancer and cardiovascular disease). [5] Patients were 40 times more likely to die from another cause than from the treated aneurysm. [5]

Previous studies have used varying methods and criteria in studying and reporting long-term mortality rates of SAH survivors. The largest study to date reported that there were 13 lethal rebleeds from the clipped saccular intracranial aneurysms out of 1440 cases at the median times of 4.9 years. [1] A recent long-term follow-up of patients in the international subarachnoid aneurysm trial cohort showed that there was no case of rebleeding from the clipped aneurysm after 10 years. [5] These data suggest that the neck remnant grows and rebleeds within approximately 5 years; otherwise, it is stabilized without rerupture. Reports of rebleeding from the clipped aneurysm occurred within two decades, after which there were no reports.

Even after operations performed by experienced neurosurgeons, neck remnants are found in nearly 10% of cases. [3] Since the first such operations were performed 35 years ago and at different hospitals, neither operative pictures nor postoperative angiogram was available. The findings at the second surgery indicating that the aneurysm clips still possessed an intact closing force would seem to imply that there was a significant residual aneurysm at the time of the first operation. There are no surgical findings presented from the first surgeries. There would seem to be little evidence to support the statement of the authors that "there must have been a

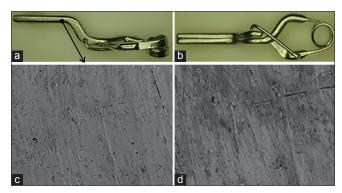


Figure 4: (a and b) Photographs depicting the retrieved Sugita clip (No. 3, 07-940-03). (c and d) Scanning electron micrographs of the retrieved clip (left,×350, right, ×750)

neck remnant, but it would have been really small and grown at an extremely slow pace."

Kashimura *et al.* emphasize that removal of previously placed clips is necessary when reoperation is performed for cerebral aneurysms that have reruptured a long time after their initial clipping.^[2] The authors also provided several tips for removing the stuck clip.^[2] In the 2 cases examined in this paper, the clips were also stuck to the tissue and embedded cotton. The clip was somehow freed and removed, but premature rupture occurred in the latter case. In the angiography, the old clip appeared not to have played any role in hemostasis, but it actually did. Surprisingly, the clip was necessary even after 35 years.

The first-generation Yasargil aneurysm clip was made in 1968 from implant steel with a half spring coil and no jaw guidance. The second-generation clip was introduced in 1970 with a 1.5 spring coil and safety ring to optimize closing force and stability. The Yasargil aneurysm clip made from Phynox increased magnetic resonance imaging safety again in 1983 (third-generation). The previous study demonstrated that the third-generation Yasargil aneurysm clips retain their mechanical properties and surface elemental composition in vivo for a long period. [7] Because the Yasagil clip from case 1 had a 35-year history, the second-generation clip was used. Despite previous negative reports on stainless steel clips, [4,8] aneurysm clips retained their mechanical properties in vivo for a long period in these 2 cases. Those who had clipping surgery before the third generation clip was introduced have since aged. Though it is quite rare, we might face a similar case because of the extended longevity.

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