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Case Report

Delayed aneurysm rupture in a patient treated with flow redirection endoluminal device: A case report and literature review

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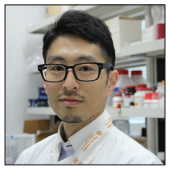

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ABSTRACT

Background: Delayed aneurysm rupture after flow-redirection endoluminal device (FRED) implantation is rare. We report a case of internal carotid-cavernous fistula (CCF) caused by a delayed aneurysm rupture of the cavernous portion of the internal carotid artery (ICA) after FRED implantation.

Case Description: A 75-year-old woman had a gradually enlarging aneurysm at the C4 portion of the left ICA. We performed FRED implantation for the same. The FRED implantation procedure was smooth and the FRED expanded well and attached to the vessel wall. Five days after surgery, the patient developed a strong headache, hyperemia of the left eye, and disturbance of the left eye movement. Magnetic resonance imaging and angiography revealed a left CCF with cortical venous reflux. We performed transarterial aneurysmal and transvenous cavernous sinus embolization. Postoperatively, angiography showed no fistula and complete occlusion of the aneurysm; however, minor eye movement disorder persisted.

Conclusion: To prevent the development of delayed aneurysm rupture in patients treated with FRED, preoperative consideration of whether to add coil embolization is important, even if the aneurysm is located in the C4 portion of the ICA and there is no risk of subarachnoid hemorrhage, including the size of aneurysm. In this report, we have tried to alert surgeons regarding the risk of delayed aneurysm rupture due to FRED implantation.

Keywords: Carotid cavernous fistula, Delayed aneurysm, Flow-redirection endoluminal device, Internal carotid artery

INTRODUCTION

Balloon- or stent-assisted coil embolization is a known endovascular treatment of wide-necked aneurysms. However, these treatments are limited by associated aneurysm recurrence rates.[7] The use of a flow diverter (FD), such as a pipeline embolization device (PED) (Medtronic, Minnesota, USA), is a favorable treatment for these aneurysms with high rates of complete occlusion.[6] Similarly, the use of a flow-redirection endoluminal device (FRED) (Microvention-Terumo, California, USA), an FD consisting of two layers, is also safe with a high rate of complete occlusion.[13,18] However, in the treatment of aneurysms with FDs, delayed aneurysm rupture — defined as rupture of the target aneurysm after the deployment of the FDs — is a fatal complication, with a reported occurrence of 0.3–4%.[1,4] A risk factor for delayed aneurysm

rupture is large or giant aneurysms.[12] However, few cases of PED implantation for relatively small aneurysms resulted in delayed aneurysm rupture,[9] but none of these cases had FRED implantation. Herein, we report a case of delayed aneurysm rupture that occurred after FRED placement.

Written informed consent to publish this report was obtained from the patient before the submission process.

Case

A 75-year-old woman (height: 148 cm, weight: 55 kg) was diagnosed with a gradually enlarging left internal carotid artery (ICA) aneurysm by magnetic resonance imaging (MRI). There were no neurological dysfunctions and no preexisting conditions. The aneurysm was a saccular aneurysm at the C4 portion of the ICA with a size of 9.4 mm × 9.1 mm, a neck of 8.0 mm, and a depth of 6.4 mm (aspect ratio of 0.8) [Figures 1a and b]. Antiplatelet therapy with 100 mg of aspirin and 75 mg of clopidogrel was initiated, and surgery was performed 29 days thereafter. Platelet aggregation capacity was measured by transmission method 1 day before the surgery. Since aspirin showed slightly low aggregation ability, 100 mg of cilostazol was additionally administered the day before and on the day of the endovascular surgery.

A 6Fr Axcelguide (Medikit, Tokyo, Japan) was placed at the left ICA. Headway 27 (Microvention-Terumo, California, USA) and CHIKAI 14 (ASAHI INTECC, Aichi, Japan) were inserted into SOFIASELECT (Microvention-Terumo, California, USA) and guided to the C1 portion of the ICA. We placed the FRED (5.0 mm × 26 mm) from the origin of the ophthalmic artery to the C5 portion of the ICA to cover the neck of the intracranial aneurysm. The FRED expanded well. Post-dilatation was not performed as cone-beam computed tomography showed that the stent fitted well into the artery [Figure 1c]. A comparison of pre and postoperative angiograms showed that the contrast did not enter into the inflow zone of the aneurysm in the arterial phase [Figures 2a and b]. In the capillary phase, there were areas of contrast stagnation [Figures 2c and d]. Coil embolization was not performed as there was no risk of subarachnoid hemorrhage due to the location of the aneurysm at the C4 portion of the ICA.

After the intervention, in addition to continuing oral aspirin 100 mg and clopidogrel 75 mg, argatroban 60 mg was administered over 24 h. There were no obvious findings of neurological deficits. The patient was discharged 3 days after the surgery. However, 2 days after discharge, the patient came back to our hospital with a strong headache. Conjunctival hyperemia of the left eye as well as left oculomotor nerve and trochlear nerve palsy was noted. MRI showed the occurrence of a left internal carotid-cavernous fistula (CCF; Figure 3a) due to rupture of the treated aneurysm. Cerebral angiography showed venous reflux into the superior ophthalmic vein (SOV) and superficial middle cerebral vein [Figure 3b]. Therefore, to prevent intracranial hemorrhage, endovascular treatment was performed.

First, transarterial coil embolization of the ruptured aneurysm was attempted with the transcell technique. A 6Fr Axcelguide (Medikit Tokyo, Japan) was placed at the left ICA. We tried to advance the CHIKAI 14 and Headway Duo (Microvention-Terumo, California, USA) into the aneurysm through the stent mesh using a 4.2Fr FUBUKI (ASAHI INTECC, Aichi, Japan) as an intermediate catheter, which was pushed strongly against the stent. However, the Headway Duo was never advanced through the stent mesh along the CHIKAI 14, which easily went into the aneurysm. Thereafter, the CHIKAI 10 and Marathon (Medtronic, California, USA) were advanced into the aneurysm through the stent mesh after several attempts. Because only iED coils (Kaneka Medics, Kanagawa, Japan) smaller than 4 mm in size were able to pass through the Marathon, the aneurysm was not packed completely due to compartment formation,

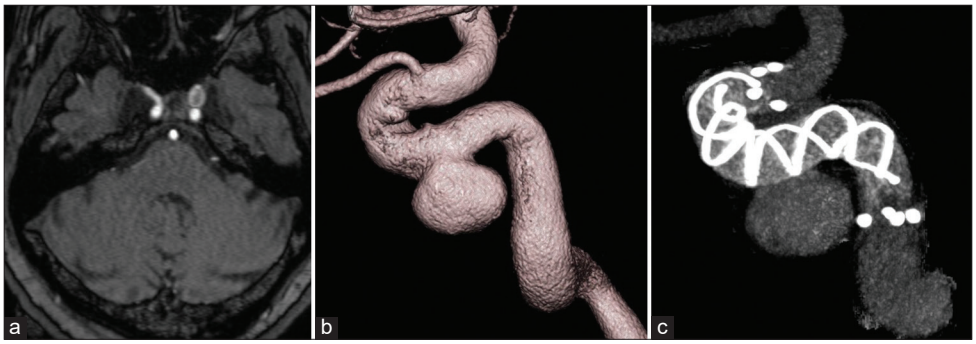

Figure 1: (a) Magnetic resonance angiography showing an unruptured aneurysm at the cavernous portion of the left internal carotid artery. (b) 3D rotated angiography showing a saccular aneurysm at the C4 portion, with a size of 9.4 mm × 9.1 mm, a neck of 8.0 mm, and a depth of 6.9 mm (aspect ratio of 0.8). (c) Cone-beam computed tomography performed after the procedure shows a stent fitted well into the artery.

and the shunt flow was not stopped. Therefore, additional embolization with 50% n-butyl-2-cyanoacrylate was performed. However, the CCF appeared to persist on the left ICA angiography. For this reason, transvenous embolization was performed. A 7-Fr Guider (Stryker, Michigan, USA) was placed to the left inferior petrosal sinus. Headway Duo and a 45° pre-shaped Excelsior SL-10 (Stryker, Michigan, USA) were placed through the cavernous sinus and the SOV, respectively. Coil embolization of the SOV and cavernous sinus was performed. Although the shunt remained after the procedure, cortical venous reflux disappeared [Figure 3c].

After the intervention, the patient continued to take aspirin 100 mg and clopidogrel 75 mg. The conjunctival hyperemia and trochlear nerve palsy improved, although the oculomotor nerve palsy persisted. A head MRI showed the disappearance of shunt flow. The patient was discharged 12 days after the intervention with slight left oculomotor nerve palsy (modified Rankin Scale 1); however, the symptoms improved 6 months after the intervention. The angiogram showed complete occlusion of the aneurysm and CCF, and 1 year after the surgery, only a slight eye movement disorder remained. The cerebral angiograms showed no lesion recurrence.

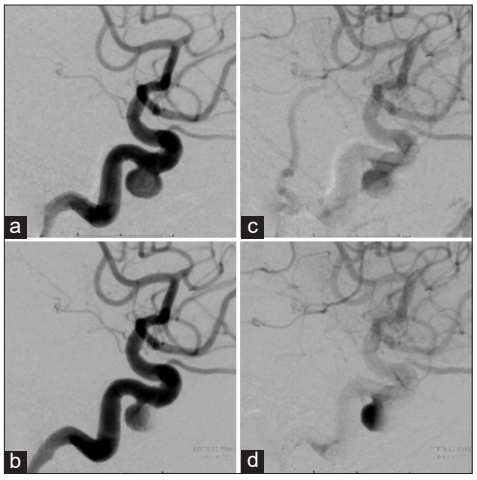

Figure 2: (a) Left internal carotid arteriography before the procedure in the arterial phase. (b) Arterial phase after the procedure showing partial areas with no inflow of contrast that were present within the aneurysm. (c) Capillary phase before procedure showing no stagnation of blood flow within the aneurysm. (d) Capillary phase after the procedure showing stagnation within the aneurysm.

DISCUSSION

We reported a case of delayed aneurysm rupture that occurred after FRED placement despite the aneurysm having a low risk of delayed rupture. We attempted to alert the surgeons to the risk of delayed aneurysm rupture with FRED implantation to avoid a worse prognosis resulting from rupture. The FRED, which has been in use since 2012, has been reported to be effective and safe.[5,15-17] However, there is a report of a poor outcome of delayed rupture of large intracranial aneurysm with FRED implantation.[17] Rouchaud *et al*.[18] reported poor prognosis of delayed aneurysm ruptures, with 81.3% experiencing death or poor neurological outcome, furthermore, Hou *et al*.[9] reported that 9.5% of patients survived regardless of subsequent treatment. We believe that this report of potentially serious complications serves as a reminder for surgeons in the field.

According to our literature review, six cases of delayed rupture of aneurysms caused by the placement of a FRED have been reported.[11,14,15,17] Based on these previous reports, the incidence of delayed rupture of intracranial aneurysm after placement of a FRED is believed to be 0.3–1.3%. Including our case, seven cases of delayed rupture of aneurysm after placement of a FRED have been reported to date [Table 1]. In all cases, the delayed aneurysm rupture occurred within 5–233 days (median 35 days) postoperatively. Further

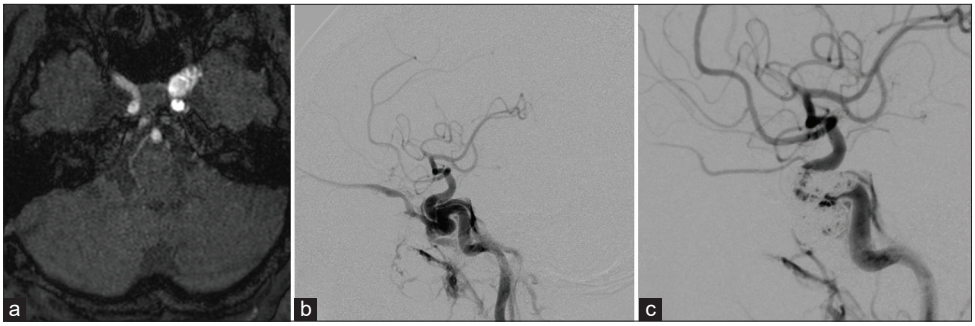

Figure 3: (a) Magnetic resonance angiography showing left internal carotid-cavernous fistula. (b) Left carotid angiography showing reflux into the superior ophthalmic vein and superficial middle cerebral vein. (c) Left carotid angiography after the procedure shows little flow to the cavernous and inferior petrosal sinuses and no cortical reflux. Therefore, we decided to complete the transvenous embolization.

Table 1: Cases of delayed aneurysm rupture after FRED implantation reported to date.

Case	Ruptured or Unruptured	Location	Morphology	Size (mm)	Coil Assistance	Interval to Rupture (Days)	Further Treatment	mRS	Reference
1	Unruptured	Basilar trunk	Dissecting	>25	With coil	35	NA/NM	6	Piano *et al.*
2	Unruptured	Supraclinoid IC	Saccular	20	No coil	21	NA/NM	5	Pierot *et al.*
3	NA/NM	NA/NM	NA/NM	NA/NM	NA/NM	233	NA/NM	6	Killer-
4	NA/NM	NA/NM	NA/NM	NA/NM	NA/NM	28	NA/NM	6	Oberpfalzer *et al.*
5	Unruptured	Supraclinoid IC	Saccular	17	No coil	76	Additional FD implantation	0	McDougall *et al.*
6	Unruptured	IC-PC	Saccular	16	No coil	18	Additional FD implantation, ventricular drainage	0	
7	Unruptured	Cavernous sinus	Saccular	9.4	No coil	5	Coil embolization (TAE, TVE)	2	Present case

IC: Internal carotid artery, NA/NM: Not applicable/not mentioned; PC: posterior communicating artery; TAE: Transarterial embolization, TVE: Transvenous embolization, mRS: modified Rankin Scale, FD: Flow diverter

treatment was performed in three cases, including our case. In two cases, additional FD implantation was performed.

Regarding the Silk FD with single-layer braided design, the risk factors for delayed aneurysm rupture include large or giant aneurysms, symptomatic aneurysms, aspect ratio >1.6, and jet blood flow in the aneurysm.[12] However, the risk factors associated with delayed aneurysm rupture of a FRED have not yet been identified. Hou *et al*.[9] reported six cases of bleeding in PED implanted cases with aneurysms smaller than 10 mm; delayed aneurysm rupture in FRED implantation was not observed. No reports of delayed aneurysm rupture smaller than 10 mm were noted in review articles that reported only FRED implantation cases.[15,16] In the present case, we performed FRED placement for an aneurysm in the cavernous sinus of the ICA with an aspect ratio of 0.8. In addition, after placement of the FRED, jet blood flow was not suspected on imaging. Despite the limited risk factors for delayed aneurysm rupture, the risk of it occurring must always be kept in mind.

Some causes of delayed aneurysm rupture after FD implantation have been reported to be due to increased pressure in the aneurysm due to rapid thrombosis,[3] weakening of the aneurysm wall due to inflammation caused by thrombus in the aneurysm,[12] migration of the FD into the aneurysm, and damage to the arterial wall during the procedure.[2] In this case, there was no migration of the FRED into the aneurysm or damage to the arterial wall. Therefore, the cause of the delayed aneurysm rupture may have been rapid thrombosis or the inflammation of the aneurysm. It is unclear whether rapid thrombosis occurred immediately after implantation. However, Figure 2 suggests the possibility that the thrombosis occurred during the 5 days before the rupture.

For the prevention of delayed aneurysm rupture, there is no definite consensus on whether FD placement should be combined with coil embolization of the aneurysm or additional FD placement. With regard to additional coil embolization, Kulcsar *et al*.[12] suggested that the addition of coils prevents rapid thrombosis and unstable thrombus formation, resulting in the prevention of delayed aneurysm rupture. On the contrary, in a computational fluid dynamics study, Hassan *et al*.[8] reported that additional FD placement raises the intra-aneurysmal pressure and jet blood flow occurs. Therefore, they stated that the situation could lead to delayed rupture of the aneurysm. In the clinical setting, Hou *et al*.[9] reported that 38.7% of patients with delayed aneurysm rupture had multiple FD implants. Although additional FD implantation promotes thrombosis and increases the rate of complete embolization,[10] it is still unclear whether it lowers the incidence of delayed aneurysm rupture. In this case, the aneurysm was located at the extradural segment; therefore, we did not perform coil embolization. We believe that coil embolization should be considered before the FRED placement if jet blood flow is observed, due to the difficulty in placing a microcatheter in the aneurysm using the transcell technique, due to the small cell size.

The patient developed oculomotor palsy after sinus-packing. Zhang *et al*.[20] recently described that 2.4% of the patients who received sinus-packing for dural arteriovenous fistulas of the cavernous sinus suffer from transient oculomotor dysfunction. In addition, Schaaf *et al*.[19] noted that coil embolization of the aneurysm itself, rather than sinus

packing, improved 96% of cases, including the diplopia that was present preoperatively. Considering these reports, the occurrence of oculomotor nerve palsy within this treatment series was relatively low. Additional coil embolization, including sinus-packing after delayed aneurysm rupture, was considered acceptable.

CONCLUSION

We reported a case of delayed aneurysm rupture that occurred after FRED implantation to alert surgeons about the possibility of this complication. The number of cases of FRED implantation is expected to increase in the future due to its ease of use and extended indications. However, even if FRED implantation is safer and the rupture rate of its aneurysm is lower than other options, delayed aneurysm rupture might still occur, as shown in this case. Careful follow-up, such as platelet aggregation evaluation and preoperative consideration on whether to add coil embolization, may help to prevent the development of delayed aneurysm rupture for patients treated with a FRED. However, it should be noted that the main limitation of this study is that only a single case was reported, and thus a mechanism could not be determined; therefore, further studies with more cases are needed.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest

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