CASE REPORT

Recurrence of a totally occluded aneurysm after treatment with a pipeline embolization device

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SUMMARY

Treatment with a pipeline embolization device (PED) is widely accepted as an excellent option for patients harboring giant, wide neck, saccular intracranial aneurysms. It has been shown to be safe and very effective. We describe a previously unreported case of a large middle cerebral artery aneurysm, related to the carotid tip, that was treated with a PED and coils. The 6 month follow-up angiogram demonstrated complete occlusion of the aneurysm. However, the 9 month angiogram revealed recurrent filling of the aneurysm sac. To our knowledge, this is the first report to describe recanalization of an aneurysm totally occluded on the control angiography after treatment with a PED. Unanswered questions include the mechanism of recanalization and the need for additional control angiograms and MRI following a previous examination revealing total aneurysm occlusion.

BACKGROUND

Flow diverter devices have emerged as an excellent treatment alternative for complex intracranial aneurysms. Their use has increased based on safety and effectiveness studies.¹⁻⁴ In addition to the advantages of this endoluminal device, flow diverter stents are notable for durable and definitive results, which has overcome the recanalization phenomenon.¹⁻³

The processes underlying successful aneurysm occlusion with the pipeline embolization device (PED) are well known, resulting from intra-aneurysmal stasis leading to thrombosis within the aneurysmal sac, and neointimal overgrowth of the construct.¹ Some mechanisms of aneurysm persistence are proposed.⁵ However, several clinical trials have demonstrated that once the aneurysm is completely occluded, no recanalization is observed.²⁻⁴

We present a patient harboring a large, wide neck, side wall aneurysm, adjacent to the internal carotid artery bifurcation, who experienced recurrent filling of the sac after angiography at 6 months showed complete occlusion of the aneurysm. To our knowledge, this is the first reported case of an aneurysm treated with a PED, totally occluded at the first angiographic follow-up, which on a subsequent angiogram revealed recurrent filling of the aneurysm, introducing the concept of recanalization in flow diversion.

CASE PRESENTATION

A 73-year-old woman presented with a 4 month history of intermittent headaches. She had no history of smoking, but had systemic hypertension. The neurological examination was normal.

INVESTIGATIONS

MRI revealed a left, well circumscribed, round, uniformly enhancing lesion at the internal carotid bifurcation region. There was no edema surrounding the lesion. An angio CT showed a 21 mm wide necked aneurysm, located at the origin of the middle cerebral artery, very close to the internal carotid artery bifurcation. The angiogram confirmed that the tip of the internal carotid artery was spared (figure 1).

TREATMENT

Endovascular treatment was performed because of the large volume of the aneurysm. Simple coiling was not possible because of the broad neck. Stent or balloon assisted coiling could possibly cause a mass effect because of the high volume of coils necessary to occlude the aneurysm, and the chance

Figure 1 (A) MRI shows a left, well circumscribed, round lesion at the internal carotid bifurcation region. (B, C) An angio CT reveals a large aneurysm, located at the origin of the middle cerebral artery, close to the internal carotid artery (ICA) bifurcation. (D) The angiogram demonstrates that the ICA bifurcation is not involved.

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Figure 2 (A) Immediate control angiography showing deployment of the pipeline embolization device (PED) plus coils. (B) The 6 month control angiogram reveals total occlusion of the aneurysm. (C, D) Note the PED totally covering the neck of the aneurysm.

of recanalization would not be negligible. Thus it was planned to treat the aneurysm with a flow diverter device from the middle cerebral artery to the internal carotid artery. Considering the large diameter of the aneurysm, and the risk of rupture before complete occlusion was achieved, it was decided to use a small amount of coils in order to promote faster occlusion without causing a mass effect.

Figure 3 (A, B) The 9 month MRI demonstrates edema surrounding the aneurysm. (C) Late angiography shows recanalization of the previously totally occluded aneurysm. (D) Note the pipeline embolization device is located at the same position, covering the neck.

The patient was asked to take clopidogrel (75 mg) and aspirin (100mg) for 5 days prior to the treatment, and for 6 months afterwards. Under general anesthesia, access to the aneurysm was performed in a standard fashion.

A PED 4.0×25 mm (Medtronic, Irvine, California, USA) was deployed from the M1 to the internal carotid artery, covering the aneurysm neck. Contrast stagnation was observed inside the aneurysm. Through a jailed microcatheter, 10 Target coils (Stryker Neurovascular, Fremont, California, USA) were implanted inside the sac. Immediate angiography revealed subtotal aneurysm occlusion, but inadequate stent to vessel wall apposition in the internal carotid artery. A 4×10 mm Transform C balloon (Stryker Neurovascular) was used to perform angioplasty and achieve optimal flow diverter apposition (figure 2).

OUTCOME AND FOLLOW-UP

The patient had an uneventful recovery. She had one follow-up angiogram at 6 months which showed complete aneurysm occlusion, without neointimal hyperplasia. At that time, clopidogrel was discontinued and the patient was maintained on aspirin.

However, 9 months after treatment, the patient presented with right sided hemiballism. MRI revealed a small area of frontal edema, adjacent to the aneurysm. Diffusion weighted imaging did not show any ischemic lesion. Due to this unusual presentation, angiography was performed and demonstrated construct patency but recurrent filling of the aneurysm sac (figure 3).

The patient improved spontaneously. She did not present with any other involuntary movement. Given that the patient was currently asymptomatic, and the residual aneurysm filling was small, aspirin was discontinued and no other treatment was proposed. Follow-up MRI and angiogram will be performed 1 year after treatment.

DISCUSSION

In an increasing number of cerebrovascular centers, the PED has become a firstline option for unruptured, large, sidewall cerebral aneurysms. Compared with traditional embolization techniques, flow diverters have proved to be at least as safe, and provide adequate and stable occlusion in the long term.¹⁻³

Contrary to other traditional treatment modalities, the longer the follow-up, the higher the probability of total aneurysm occlusion after treatment with flow diverters.¹ Lang *et al*⁵ reported continuous growth of an aneurysm treated with a flow diverter. It is not certain that the aneurysm was totally occluded before it started increasing in volume. Follow-up evaluation was done with angio CT, which is not the gold standard for assessing flow diverters.

Iihara *et al*⁶ described a patient harboring a partially thrombosed giant aneurysm of the vertebral artery. Endovascular trapping had been performed. Although there was no angiographically demonstrated evidence of filling, the aneurysm continued to enlarge. MRI demonstrated residual filling of the sac. The patient underwent craniotomy. Marked development of the vasa vasorum on the occluded segment was observed. Therefore, a totally occluded aneurysm may present with continuous growth via the vasa vasorum.⁷ That is why MRI is very important to evaluate the efficacy of treatment. Aneurysm volume reduction should be expected. On the other hand, angiography evaluates only the lumen of the parent vessel.

The PED is the most studied flow diverter device, and no recanalization of a previously angiographically occluded aneurysm has been reported.^{2,3} However, in 2016, Zhang reported

Learning points

- ▶ Flow diverters are widely accepted as the first choice to treat sidewall complex aneurysms.
- ▶ Flow diverter devices are notable for durable and definitive results.
- ▶ This is the first report to describe recanalization after treatment with a pipeline embolization device.
- ▶ The mechanism of recanalization is not well known.
- ▶ Long term follow-up is mandatory after treatment with flow diverters.

recurrence of a completely occluded aneurysm treated with a Tubridge flow diverter.⁸

As the aneurysm in our case was not located at a bifurcation, and it had a very wide neck, involving the parent artery circumferentially, dissection is a possible etiology. In a recent review, complete occlusion rate at the latest follow-up of dissecting aneurysms treated with a PED was approximately 70%.⁹ Considering that these aneurysms are characterized by a dynamic behavior, careful follow-up is mandatory.

Several mechanisms are related to flow diverter treatment failure, including inadequate aneurysm coverage, pre-existing stents, and endoleak.⁵ None of these mechanisms can explain the recanalization in our case. Another mechanism that could explain the recurrent filling in our case could be branch vessel runoff. However, a lenticulostriate artery arising from the M1 aneurysm was not identified. Thrombus organization inside the aneurysm sac, until neointimal formation occurs and totally covers the aneurysm neck, may play a role in the recanalization phenomenon.

Considering the low risk of rupture in our case, we decided to wait and perform another angiogram, expecting late occlusion of the aneurysm. Although an alternative option would be retreatment by deploying another PED, it is not clear if multiple PEDs would increase the occlusion rate.¹⁰

Although the PED is one of the most studied devices in interventional neuroradiology, long term follow-up is mandatory to allow stronger conclusions to be drawn about durability. Whether or not additional angiography is necessary after a previous angiogram has shown complete aneurysm occlusion remains unclear. MRI should be considered as part of the follow-up protocol.

Contributors FPT helped to conceive the study, and drafted and approved the manuscript. ACU helped to conceive the study and the study design, helped with revision of the manuscript, and gave final approval of the manuscript. MTR and LHC-A participated in data acquisition, editing of the figures, revision of the manuscript, and final approval of the manuscript. DGA conceived the study, revised the manuscript critically, and approved the final manuscript.

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