CASE REPORT

Spontaneous regression of intracranial aneurysm following remote ruptured aneurysm treatment with pipeline stent assisted coiling

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SUMMARY

Spontaneous aneurysm regression is a rare phenomenon. We present the interesting case of a 54-year-old woman who was admitted with a Hunt/Hess grade IV, Fisher grade III subarachnoid hemorrhage and multiple intracranial aneurysms. She was treated with coiling of the largest paraclinoid aneurysm and placement of a flow diverting pipeline embolization device that covered all internal carotid artery (ICA) aneurysms. A follow-up angiogram at 6 months showed remodeling of the ICA with complete obliteration of all treated aneurysms. A distant, untreated, right frontal M2 aneurysm regressed spontaneously, after the flow was diverted away from it with the stent. The literature is reviewed, and potential pathophysiological mechanisms leading to aneurysm regression are discussed.

BACKGROUND

The introduction of flow diverting stents has allowed minimally invasive treatment of large proximal internal carotid artery (ICA) aneurysms that were traditionally treated with extensive skull base approaches.¹

Thrombosis of traumatic pseudoaneurysms and ruptured or unruptured intracranial aneurysms has been described in the past.² However, spontaneous unruptured aneurysm regression is a very rare phenomenon,^{3 4} which may mimic an ischemic stroke.⁵

We present a case of spontaneous regression of a distant untreated right frontal M2 aneurysm, after the flow was diverted away from it with a pipeline embolization device (PED) that was placed to treat a ruptured large right paraclinoid ICA aneurysm.

CASE PRESENTATION

A 54-year-old woman was admitted with a Hunt/Hess grade IV subarachnoid hemorrhage (figure 1). A cerebral angiogram revealed a 22 mm right paraclinoid aneurysm without distinct neck, involving nearly the entire vessel wall, in addition to 4.7 mm right paraophthalmic, 3.2 mm right dorsal ICA wall, 4.4 mm right frontal M2, and 2.4 mm left middle cerebral artery (MCA) bifurcation aneurysms (figure 2).

TREATMENT

She was loaded with 650 mg of aspirin and 600 mg of clopidogrel. VerifyNow testing 8 h later demonstrated that the patient was resistant to clopidogrel, and was loaded with 180 mg of ticagrelor

overnight. Repeat VerifyNow testing in the morning prior to intervention showed an adequate response. After general anesthesia was induced, the patient was fully heparinized. A Neuron Max-088 guide catheter was placed in the right cervical ICA. A Navien-72 catheter was telescoped into the cavernous ICA. The large aneurysm was crossed with a Headway-27 microcatheter over a Synchro-2 soft and a 'buddy' Synchro-10 wire. There was a very early MCA bifurcation. The microcatheter was navigated into the proximal right temporal M2, away from the frontal M2 aneurysm. The large aneurysm was catheterized with an Echelon-10 microcatheter (figure 3). A 3.75 mm×35 mm PED was deployed from the proximal mid temporal M2 to the cavernous ICA. The PED covered all three right ICA aneurysms. As some of the aneurysms treated were past the superior hypophyseal segment, this PED use was considered off-label in the USA. The aneurysm was then subtotally coiled with Target-360 coils. Immediate post procedure angiographic runs showed adequate protection of the dome of the largest aneurysm and some intra-aneurysmal contrast stasis (figure 4). The patient recovered well from the subarachnoid hemorrhage, and she was discharged on aspirin and ticagrelor.

Figure 1 Non-contrast CT scan demonstrating diffuse subarachnoid hemorrhage and dome of the large paraclinoid aneurysm (arrow).

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Figure 2 Diagnostic cerebral angiogram, anteroposterior (A) and oblique (B) views, right internal carotid artery injection, demonstrating large paraclinoid, paraophthalmic, and dorsal wall aneurysms and laterally pointing 4 mm right frontal M2 aneurysm (arrow).

OUTCOME AND FOLLOW-UP

The patient returned for an angiogram 6 months later. She was neurologically intact and had returned to work 3 months prior. The right paraclinoid, paraophthalmic, and dorsal ICA wall aneurysms were completely occluded. Interestingly, the right frontal M2 aneurysm had regressed significantly (figure 5). The right A1 thrombosed, and both A2s filled from the left. The left MCA bifurcation aneurysm remained unchanged. Ticagrelor was stopped, and she was kept on lifelong aspirin.

DISCUSSION

Flow diverting stents are used to cover large cavernous and proximal supraclinoid aneurysms with a wide neck.⁶ Alteration of flow within and around the aneurysm inflow zone leads to gradual thrombosis and atrophy of the aneurysm, while flow into the parent vessel and perforating branches is preserved.⁷ It is believed that the PED acts as a scaffold over which endothelial cells can grow, eventually incorporating the PED into the wall of the parent artery, while the ostia of vascular branches remain open.⁸

The PED is used as a sole device for definitive treatment of certain aneurysms.⁹ There are reports describing the migration

of the device, which may happen in a delayed fashion.¹⁰ We decided to use the longest possible PED, in order to prevent complications associated with possible foreshortening of the device and avoid placement of more than one stent.

Only a few reports have described the placement of a PED in conjunction with coils.^{6 11} A recent paper by Nossek *et al* described their experience of treating 27 patients with 28 aneurysms with pipeline stent-assisted coiling. They reported no complications associated with the coil mass or placement of the PED. This method might allow for early thrombosis and impending size reduction of the aneurysm. Furthermore, a partially dense coil mass may lessen mass effect on the surrounding structures and may reduce the chance of in-stent thrombosis, as experienced by Siddiqui *et al*.^{11 12} Therefore, we decided to partially coil the aneurysm in addition to covering its neck with a flow diverting stent.

The most fascinating finding in our case was the spontaneous regression of the untreated right frontal M2 aneurysm. Although natural thrombosis of intracranial aneurysms has been described in the past,¹³ their spontaneous angiographic regression is a rare occurrence. Disruption of the internal elastic lamina, atherosclerotic alteration of the aneurysmal wall, focal

Figure 3 Cerebral angiogram, anteroposterior (A) and lateral (B) views, right internal carotid artery (RICA) injection, during endovascular treatment showing microcatheters advanced into the paraclinoid aneurysm for coil placement and temporal M2 branch for stent placement. A jet of contrast is seen filling the paraclinoid aneurysm dome. The paraophthalmic and dorsal wall RICA and right frontal M2 aneurysms are depicted.

Figure 4 Skull X-ray, anteroposterior (A) and lateral (B) views, demonstrating coil mass, contrast stagnation within the paraclinoid aneurysm, and stent placement (proximal and distal ends of stent indicated by arrows).

inflammatory changes, and disequilibrium between arterial wall integrity and focal hemodynamic stress are some of the factors that lead to aneurysmal formation.¹⁴

Kondziolka *et al*¹⁵ reported regression of flow related aneurysms following resection or embolization of arteriovenous malformations, while aneurysms not directly related to the arteriovenous malformation angioarchitecture may remain stable or grow over time. Li *et al* reported a patient with severe right ICA stenosis and an unruptured anterior communicating artery aneurysm. The carotid stenosis was treated with carotid endarterectomy and “resulted in reduced flow through the left A1”. Restoration of flow after the procedure balanced the stress across the anterior communicating artery and resulted in regression of the aneurysm after 14 months.⁴

Hans *et al*¹⁶ published the case of a patient with a giant cavernous and two smaller supraclinoid ICA aneurysms that were treated with proximal ICA sacrifice, which in turn caused reversal of flow into the supraclinoid ICA and led to spontaneous regression rather than thrombosis of the two distal aneurysms. Chow *et al* described the first case of spontaneous aneurysmal regression following coiling of a concurrent aneurysm. They argued that either the treatment of the proximal aneurysm changed the flow dynamics within the vessel of interest and the smaller distal aneurysm, thus causing it to regress, or the aneurysm thrombosed due to intimal injury from the catheterization attempt.³ We hypothesize that PED placement in a separate vascular branch than the one harboring the untreated aneurysm reduced the tension against the aneurysmal wall and allowed the indirect repair of this flow related aneurysm.

Figure 5 Cerebral angiogram, 6 months post-treatment, anteroposterior (A), oblique (B), and lateral (C) views, right internal carotid artery (RICA) injection, demonstrating no residual or recurrent aneurysm, including regression of the previously noted right frontal M2 aneurysm arrow. Opacification of the A1 segment of the RICA is no longer seen.

Learning points

- ▶ Flow diverting stents are increasingly used with or without coils to achieve parent vascular remodeling and obliteration of difficult aneurysms.
- ▶ Meticulous attention should be given to stent size and length selection when treating aneurysms, in order to counter possible stent migration or shortening.
- ▶ Follow up angiograms should be obtained for evaluation of in-stent stenosis and aneurysmal recanalization or regrowth.
- ▶ While changing the flow dynamics of a proximal vessel may eventually lead to spontaneous regression of a downstream aneurysm, it is important to emphasize that our results are purely anecdotal and should not be generalized. Additional vascular and cross sectional studies are necessary to determine whether this is a safe and durable treatment method.

Contributors All persons who meet authorship criteria are listed as authors, and all authors certify that they have participated sufficiently in the work to take public responsibility for the content, including participation in the concept, design, analysis, writing, or revision of the manuscript. Furthermore, each author certifies that this material has not been and will not be submitted to or published in any other publication before its appearance in the *Journal of NeuroInterventional Surgery*.

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