Optic chiasm compression from mass effect and thrombus formation following unsuccessful treatment of a giant supraclinoid ICA aneurysm with the Pipeline device: open surgical bailout with STA-MCA bypass and parent vessel occlusion

Case report

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The Pipeline Embolization Device (PED) (ev3 Inc.) is now approved by the FDA for use in the US for internal carotid artery (ICA) aneurysms, and are now approved by the FDA specifically for this use. Potential pitfalls, however, have not yet been described in the pediatric neurosurgical literature. The authors report on a 10-year-old boy who presented to the Barrow Neurological Institute after progressive visual decline. He had undergone placement of a total of 7 telescoping PEDs at another facility for a large ICA aneurysm. Residual filling of the aneurysm and significant expansion of intraaneurysmal thrombus with chiasmal compression on admission images were causes for concern. The patient underwent a surgical bailout with a superficial temporal artery–middle cerebral artery bypass, with parent artery occlusion. Postoperative vascular imaging was notable for successful occlusion of the parent vessel, with no evidence of filling of the aneurysm.

Reports on the pitfalls of PEDs in the neurosurgical literature are scarce. To the authors’ knowledge this represents the first paper describing a successful open surgical bailout for residual aneurysmal filling and expansion of thrombus after placement of a PED.

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Key Words • Pipeline Embolization Device • aneurysm • chiasmopathy • Parry-Romberg syndrome • bypass • parent vessel occlusion • optic chiasm • vascular disorders

The Pipeline Embolization Device (PED) (ev3 Inc.) is now approved by the FDA for use in the US for internal carotid artery (ICA) aneurysms. Despite excellent results in a host of series, there are still some disadvantages and caveats when the device is used for the treatment of difficult intracranial aneurysms. We describe a case in which Pipeline embolization resulted in incomplete treatment and intraaneurysmal thrombus formation, causing mass effect on the optic chiasm and necessitating further treatment.

Abbreviations used in this paper: AP = anteroposterior; ICA = internal carotid artery; MCA = middle cerebral artery; MRA = MR angiography; PED = Pipeline Embolization Device; RA = radial artery; STA = superficial temporal artery.

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tion, his vision deteriorated precipitously. Over the course of 1 month, he lost vision in his left eye as well as the temporal hemifield of his right eye (Fig. 3A and B). He was placed on a course of steroids at the other facility, with little improvement in his vision. Magnetic resonance angiography (MRA) studies obtained 2 months later demonstrated continued filling of the aneurysm despite intraaneurysmal thrombus formation, as well as mass effect on the optic chiasm. The patient underwent repeat angiography 5 times, with placement of an additional 6 telescoping PEDs over the course of 4 months. The final construct spanned the cavernous portion of the ICA into the distal left middle cerebral artery (MCA). Follow-up angiograms obtained at 1 month (Fig. 2) and 4 months (Fig. 4) after initial treatment did not show contrast filling of the aneurysm, but at 6 months, MRA studies of the head demonstrated that the aneurysm lumen had progressively accumulated thrombus and was beginning to expand (Fig. 5). Throughout this time, the patient had been maintained on dual-antiplatelet therapy with aspirin and clopidogrel.

Examination. Nine months after treatment, the aneurysm continued to show signs of growth. At this point the patient was referred to our institution for evaluation of open surgical options. On the initial visit, the visual deficit in his left eye was fixed, with only intermittent/unreliable light perception. His right eye visual acuity waxed and waned but eventually stabilized to a fixed temporal field cut (Fig. 3). He had otherwise unremarkable examination results, with intact speech, and cognitive/motor function appropriate for his age. An initial diagnostic angiogram performed at our center showed continued filling of the aneurysm with multiple irregularly shaped portions (Fig. 6 left). Bilateral ophthalmic arteries were noted to be filling. The late arterial phase clearly showed the outline of the thrombosed aneurysm (Fig. 6 right). We concluded that despite repeated treatments with the PED the aneurysm continued to fill, resulting in new layers of thrombus that expanded the aneurysm dome and furthered the mass effect on the optic chiasm (Fig. 7).

Given the risk to vision in his good eye, we recommended treatment with 1) a high-flow bypass from the common carotid artery to the M2 segment of the MCA and 2) clip ligation of the parent vessel, either distal to the aneurysm, proximal to the aneurysm, or both, depending on the findings at the time of surgery. The PED construct had resulted in significant narrowing of the lumen in the distal M1, just proximal to the MCA bifurcation (Fig. 4C). One of our main concerns, in addition to the risk of hypoperfusion of the patient’s dominant hemisphere, was the ability to perfuse these lateral lenticulostriate arteries.

Operation. We performed a left orbitopterional craniotomy and exposed the giant aneurysm in the opticocarotid cistern. The sylvian fissure was adherent and friable; therefore we attempted to limit the dissection to what was necessary, given that the operation was in the dominant hemisphere. We easily visualized the PED construct through the parent vessel (Fig. 8). The falciiform ligament

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Fig. 1. Pretreatment cerebral angiograms obtained in a 10-year-old boy. Anteroposterior (AP) (left) and lateral (right) views are notable for a large, fusiform ICA aneurysm.

Fig. 2. Cerebral AP (A) and lateral (B) angiograms obtained 1 month after PED treatment are notable for absent flow within the aneurysm. Also, lateral (C) and AP (D) scout radiographs of the PED construct were obtained.
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On both sides of the left optic nerve was opened to decompress and detether the left optic nerve from the skull base. Consideration was given to placing an aneurysm clip between the anterior choroidal artery and the posterior communicating artery. However, this would have excluded the anterior choroidal artery from proximal anterograde flow, leaving only flow across the anterior communicating artery and retrograde through the bypass to perfuse this vessel. We therefore opted not to place a clip distally and proceeded with a superficial temporal artery (STA)–MCA bypass, using both the frontal and parietal branches of the MCA as donor vessels. Cervical carotid artery–MCA bypass with a radial artery (RA) donor was considered but was not performed due to the small size of the RA and its short length, which would probably not withstand future growth of the patient.

The first anastomosis was performed to a small (<1 mm) M3 branch on the temporal side of the sylvian fissure. The second anastomosis was performed using the parietal branch sutured to an RA graft in an end-to-end fashion, followed by an end-to-side anastomosis to an M2/M3 vessel at the level of the sylvian fissure. Indocyanine green angiography demonstrated excellent patency in the larger parietal STA–RA–M1/M4 vessel and sluggish flow in the other vessel of the double-barrel bypass. At this point, we decided to perform partial occlusion of the ICA in the cervical region just distal to the bifurcation. Complete occlusion of the cervical ICA was not performed in case there was insufficient flow from the bypass and across the anterior communicating artery to perfuse his left hemisphere adequately. The decision was made to observe the patient for several days, after which a balloon test occlusion with possible complete occlusion of the cervical ICA would be performed.

**Postoperative Course.** When the patient woke up, he was at his neurological baseline, without any speech, motor, or cognitive deficit. He underwent MRA on the 1st postoperative day, which showed no filling of his left ICA distal to the common carotid artery bifurcation and no filling of the aneurysm (Fig. 9). The larger vessel of the STA-MCA bypass filled back to the level of the ICA terminus, with flow visualized in his left A1 (Fig. 9C). Because the vessel had already occluded on its own and the patient was neurologically unchanged, the previously planned endovascular test occlusion was unnecessary.

![Fig. 3. Kinetic visual field testing at 3 months preoperatively in the right eye (A) and left eye (B), and at 6 months postoperatively in the right eye (C) and left eye (D). The results are notable for considerable improvement in visual fields.](image-url)
He was discharged from the hospital without incident on postoperative Day 3. Six weeks after surgery, he had no further change in his vision and remained at his neurological baseline. Ophthalmological visits with detailed kinetic visual field tests at 2 months and 5 months postoperatively were notable for dramatic improvement in his visual fields (Fig. 3C and D). The ophthalmology report at 5 months stated that the “right temporal visual field defect and paracentral scotoma and left inferonasal island both improving since last field.” Because this young patient had already received exposure to a significant amount of radiation as a result of multiple angiograms, we decided to avoid a formal angiogram for diagnostic purposes at discharge and will perform an MRA study in 1 year.

**Discussion**

The open vascular treatment of aneurysms that have failed to respond to endoluminal therapies is not a new phenomenon. This report represents one of the first cases of open surgical salvage of an aneurysm with a PED construct already in place. Flow diverters, including the Silk flow-diverting stent (Balt Extrusion) and the Pipeline device, are relatively new. As the number of successes accumulates, so have some notable shortcomings.

A recent report of 2 cases has shown that the Pipeline device can result in mural destabilization causing delayed, spontaneous aneurysm growth with possible rupture of the treated aneurysm. Others have also reported that thrombosis in an aneurysm treated with flow diversion may not always lead to remodeling but can cause autolysis of the aneurysm wall and delayed rupture; as the authors suggest, the biological behavior of the devices remains to be better understood. Although the cause of this phenomenon is unknown, the timing of aneurysm obliteration, if any, could play an important role in the eventual success of treatment; however, this remains to be proven. There is ongoing research in flow dynamics relating to flow diverters, but this represents an understudied field of research. One such study demonstrated...
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that even reductions in flow velocity do not significantly reduce intraaneurysmal pressure.12

As those who have placed these devices have seen, there is great variability in how quickly aneurysm obliteration occurs. In some cases this can occur almost immediately, whereas others take a much longer period of time. If the obliteration of the aneurysm does not occur fast enough and there is reduction (but not cessation) in flow, there also exists a risk for ingress without adequate egress. This ball-valve mechanism has been shown to result in hemorrhage and death.13 For this reason, some have advocated placement of coils within aneurysms at the same time as PED treatment to aid aneurysm obliteration.14 Thrombus formation within a large aneurysm that has been treated with flow diversion (or even by surgical means) can occur early or late, but as long as the degradation of thrombus is faster than its formation, the aneurysm should not increase in size and result in worsened mass effect.

As is seen in this case, it is also possible to form thrombus within the aneurysm very quickly and cause mass effect. In the aforementioned report from Hampton et al., aneurysm growth resulted in mass effect and edema.5 It is not uncommon for giant aneurysms to cause edema in the surrounding brain and also to have intraluminal nonfilling compartments consisting of thrombus. Such thrombus formation is probably not static but involves ongoing simultaneous deposition and degradation of thrombus material within the aneurysm. In our experience, the natural history of this occurrence is in fact sometimes adversely affected by iatrogenic manipulation, whether surgical ligation or flow diversion. We have previously debulked giant aneurysms internally to reduce the burden of thrombus within the aneurysm and its resultant mass effect. This debulking is sometimes essential, but manipulation of the aneurysm has also exacerbated malignant edema formation in the surrounding brain that can offset the effect of reducing the aneurysm mass. Aneurysm debulking was not performed in this particular patient for fear that it would require further exposure and retraction that might have proven harmful in the dominant hemisphere, especially considering the friability and adherent nature of his sylvian fissure.

Resolution of mass effect and compressive symptoms has been achieved with use of the Pipeline device in difficult aneurysms,15 but in this report the opposite occurred. Rather than alleviating mass effect, previously nonexistent mass effect was induced with flow diverter placement. It is unusual that such a process would occur so quickly after treatment and that significant visual

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**Fig. 6.** The preoperative diagnostic cerebral angiograms obtained 9 months after PED insertion at Barrow Neurological Institute are notable for persistent filling of the ICA aneurysm (left), which is particularly evident in the late arterial phase (right).

**Fig. 7.** Preoperative plain axial CT scan of the head obtained 9 months after PED insertion at the Barrow Neurological Institute is notable for a large, partially thrombosed ICA aneurysm surrounding the pipeline construct.

**Fig. 8.** Intraoperative image of the PED visible through the parent vessel. Used with permission from Barrow Neurological Institute.
disturbance from chiasm compression rapidly occurred. As the compression persisted, the placement of additional overlapping telescoping devices in an attempt to obliterate the aneurysm and eventually shrink the mass was unsuccessful. This ultimately led to the open surgical bypass and cure of the aneurysm.

Conclusions

We demonstrate here the case of a PED construct that caused mass effect and chiasmopathy in a pediatric patient, presumably due to expanding thrombus within an incompletely obliterated mixed fusiform and saccular giant aneurysm. Treatment for the continued aneurysm filling and mass effect was by occlusion of the parent vessel in the neck with a low-flow bypass to the MCA, resulting in considerable improvement in visual examination results over several months. Although the PED has proven to be successful for many aneurysms, the current report illustrates another cautionary case.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author contributions to the study and manuscript preparation include the following. Conception and design: Abla, Albuquerque. Acquisition of data: Abla, Zaidi. Analysis and interpretation of data: Abla, Zaidi. Drafting the article: Abla, Zaidi. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Spetzler. Administrative/technical/material support: all authors. Study supervision: Spetzler, Abla, Zaidi, Crowley, Britz, McDougall.

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