

Compressive optic neuropathy caused by a flow-diverter-occluded-but-still-growing supraclinoid internal carotid aneurysm: illustrative case

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BACKGROUND Flow diverter stenting is an effective treatment for large proximal internal carotid artery (ICA) aneurysms. Cranial neuropathy caused by the mass effect of the aneurysm usually subsides over time. However, a new onset of compressive optic neuropathy after successful flow diverter stenting of a large proximal ICA aneurysm is seldom reported.

OBSERVATIONS A 57-year-old woman had a right supraclinoid ICA aneurysm (approximately 17 mm) on magnetic resonance angiography (MRA) in a health checkup. She received intervention with the Pipeline embolization device. Six months later, she started to experience progressive hemianopia in the left half of the visual field. Nine months after stenting, MRA showed that the aneurysm was growing and causing mass effect, but digital subtraction angiography confirmed that the aneurysm was completely excluded from the circulation. She received a craniotomy for microsurgical decompression of the optic nerve and coagulation shrinkage of the aneurysm. Clipping and thrombectomy were not attempted. Her visual fields recovered gradually. Follow-up MRA showed that the aneurysm also diminished in size.

LESSONS Whether the coagulation technique of the flow-diverter-occluded aneurysm alone is enough to cause satisfactory shrinkage and interaction between the flow diverter and the aneurysmal vasa vasorum/neointima formation should be further examined.

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KEYWORDS case report; flow diverter; optic neuropathy; microsurgery; Pipeline embolization device; supraclinoid aneurysm

Large or giant aneurysms of the internal carotid artery (ICA) have a poor prognosis when left untreated.¹ In the past, ICA trapping (with or without bypass) and endovascular coiling have been considered the standard treatment modalities for these lesions but with high rates of complications and aneurysm recanalization, respectively. Flow diversion treatment emerged as a viable alternative to the traditional ICA trapping and endovascular coiling techniques² and showed promising outcomes for patients with either multiple cranial neuropathies or isolated optic neuropathy.^{3,4} In the literature, however, little is mentioned about

a flow-diverter-occluded large ICA aneurysm still being able to grow and cause compressive optic neuropathy. Here, we present a rare case and discuss treatment concerns.

Illustrative Case

A 57-year-old woman with a medical history of hypertension and uterine cancer status after total abdominal hysterectomy was found to have a right ICA aneurysm (approximately 17 mm) on magnetic resonance angiography (MRA) of the brain in a health checkup

ABBREVIATIONS DSA = digital subtraction angiography; ICA = internal carotid artery; MRA = magnetic resonance angiography.

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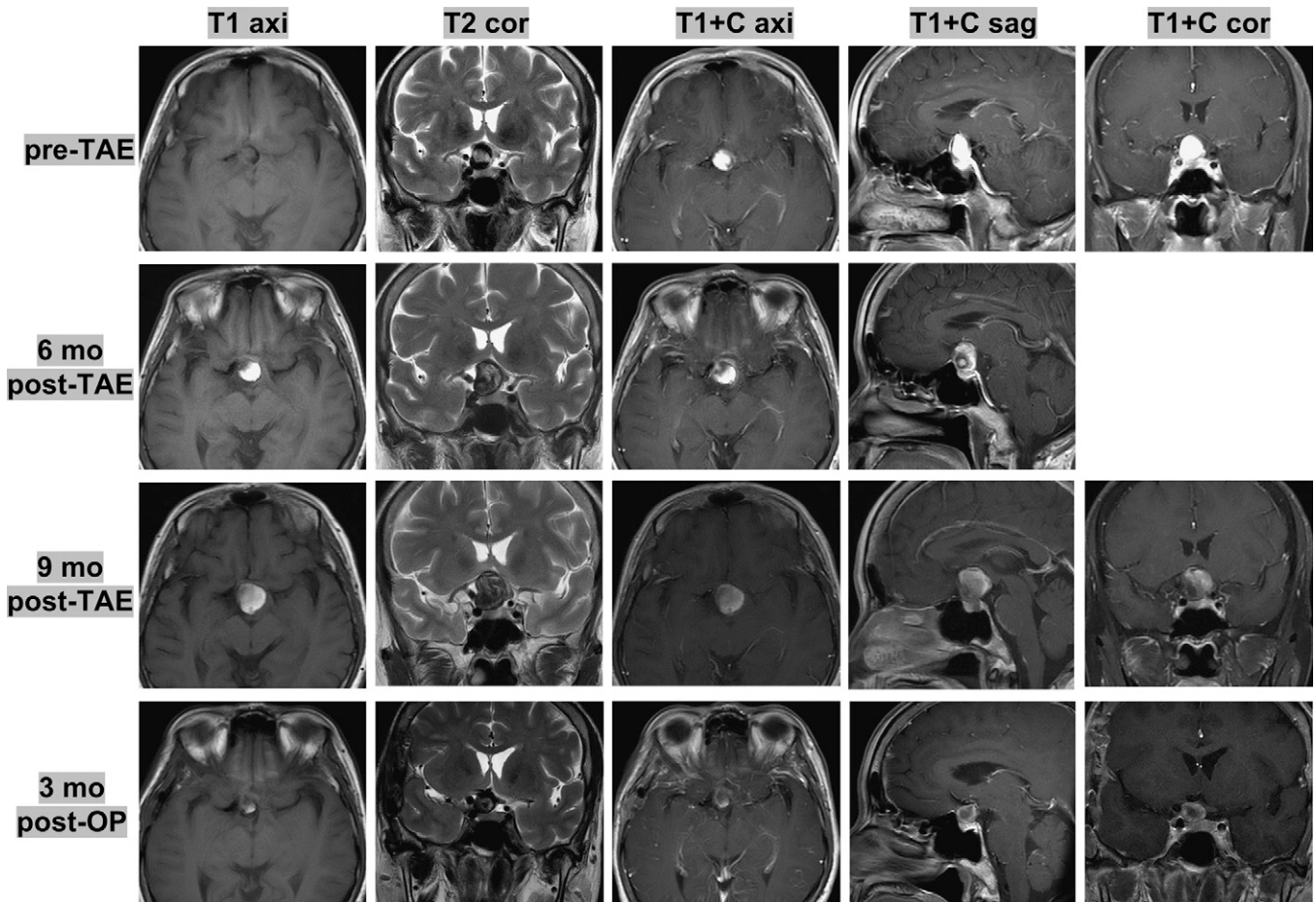


FIG. 1. The sequential changes of a right supraclinoid ICA on brain magnetic resonance imaging. The pretransarterial embolization (pre-TAE) size of the aneurysm was 17 mm (dome) and 6.7 mm (neck). After an uneventful Pipeline embolization device (PED) placement, the aneurysm was still growing (post-TAE rows). Due to worsening compressive optic neuropathy, microsurgical decompression of the optic nerve/chiasm and coagulation shrinkage of the aneurysm were performed. The patient's clinical condition improved, and the size of the aneurysm decreased (post-OP row). axi = axial; C = contrast; cor = coronal; sag = sagittal; TAE = transarterial embolization.

(Fig. 1). She was referred to our neurosurgical outpatient clinic for management. She had no symptoms and signs at that time. After discussion, she chose to receive flow diverter stenting for the aneurysm. Digital subtraction angiography (DSA) of the brain showed a right supraclinoid ICA aneurysm with superomedial projection in favor of a superior hypophyseal artery aneurysm (Fig. 2A–C). The aneurysm neck was approximately 6.7 mm. A flow diverter device (Pipeline embolization device, 4.5 × 20 mm, ev3) was deployed at the right supraclinoid region uneventfully (Fig. 2D–F). The patient was discharged with dual-antiplatelet therapy (aspirin 100 mg every day and clopidogrel 75 mg every day; P2Y12 platelet function not tested) 1 day after treatment. Six months after receiving the flow diverter, however, she started to experience progressive hemianopia in the left half of the visual field. MRA of the brain 6 months after stenting showed partial thrombosis of the aneurysm, approximately 15 mm. The dual-antiplatelet therapy was then discontinued, and only use of aspirin (100 mg every other day) remained in hope that complete thrombosis might lead to a decrease in the aneurysm size and halt the mass effect on the optic nerves and the chiasm. Three months later, the patient had complete

hemianopia in the left-sided visual fields of both eyes; the right-sided visual field of the right eye was also affected (Fig. 3).

Follow-up MRA 9 months after stenting showed complete thrombosis of the aneurysm, but the size of the aneurysm increased to approximately 18 mm (Fig. 1). DSA confirmed that the right supraclinoid aneurysm was completely excluded from the circulation (Fig. 2G–I), but the aneurysm was still growing and causing mass effect. The patient received pulse corticosteroid therapy (methylprednisolone 250 mg every 6 hours for 3 days) for her compressive optic neuropathy, but the steroid treatment was not effective. A half month later, she finally received a craniotomy for microsurgical decompression of the optic nerve and coagulation shrinkage of the aneurysm. We opened the right optic canal, cut the falciform ligament, released the optic nerve sheath, and coagulated the aneurysm via the bipolar forceps to shrink its size. During the operation, severe compression of the optic nerves and the chiasm was observed. The flow diverter could be seen through the ICA (Fig. 4). The patient's visual fields improved after the operation (Fig. 3). MRA of the brain 3 months after the operation showed that the aneurysm had diminished in size (Fig. 1).

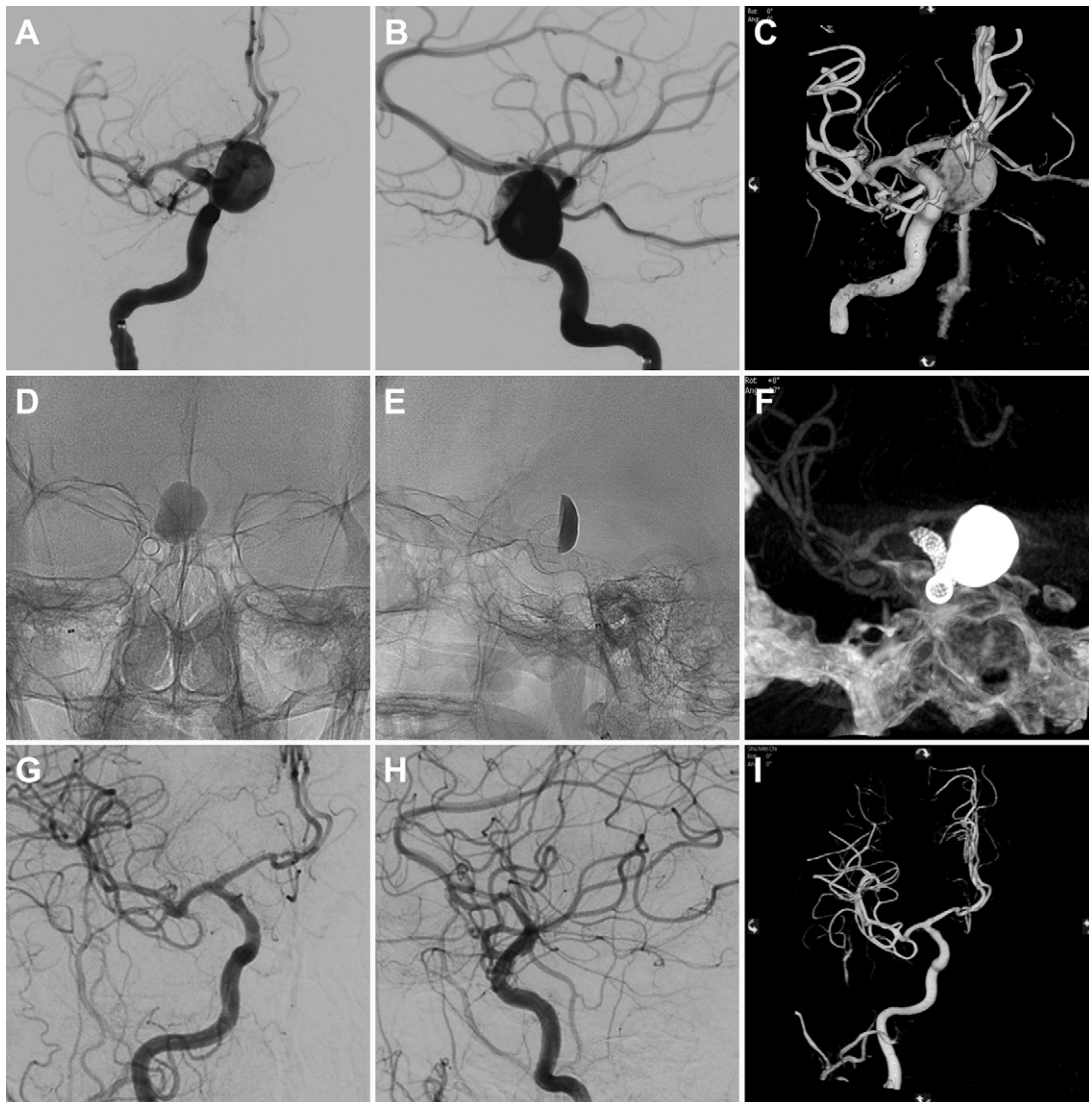


FIG. 2. The sequential changes of a right supraclinoid ICA on DSA. **A–C:** DSA revealed a right supraclinoid ICA aneurysm with superomedial projection in favor of a superior hypophyseal artery aneurysm. The aneurysm neck was approximately 6.7 mm. **D–F:** A flow diverter device (PED, 4.5 × 20 mm) was deployed at the right supraclinoid region, and contrast stagnation inside the aneurysm was noticed after the stenting. **G–I:** The aneurysm was confirmed to be excluded from the circulation on DSA 9 months after stenting.

Discussion

Observations

Cranial nerve compression is a common presenting symptom for these large lesions before treatment. In a multiinstitutional study, flow diversion showed that 60% of patients with isolated optic neuropathy had improvement at 12-month follow-up, placing it slightly behind microsurgery (mean, 68.6%) and ahead of coil embolization (mean, 46.2%) in the literature.³ In the Pipeline for Uncoilable or Failed Aneurysms Study (PUFS), among patients with baseline compressive optic neuropathy, 53% showed any improvement by the 6-month follow-up.⁴ However, a new onset of compressive optic neuropathy after successful flow diverter stenting of a large proximal ICA aneurysm is seldom reported. In our case, the patient had no baseline deficits. Her aneurysm was

excluded by the flow diverter, but its size was still increasing and causing mass effect. Because DSA confirmed exclusion of the ICA aneurysm by the flow diverter (Fig. 2), an additional flow diverter was not helpful. Microsurgical decompression was the last choice we had to treat the problem. We used the Dolenc approach to open the optic canal and achieve optic nerve decompression.⁵ After the extradural anterior clinoidectomy, the distal dural ring, the falciform ligament, and the optic nerve sheath were incised extensively for further optic nerve decompression. During the operation, we found that the aneurysm wall was thick, and the flow diverter was clearly seen through the ICA (Fig. 4). The decision to clip the aneurysm and perform a thrombectomy was not made because we worried that the clips may cause deformation of the flow diverter with subsequent ICA injury or

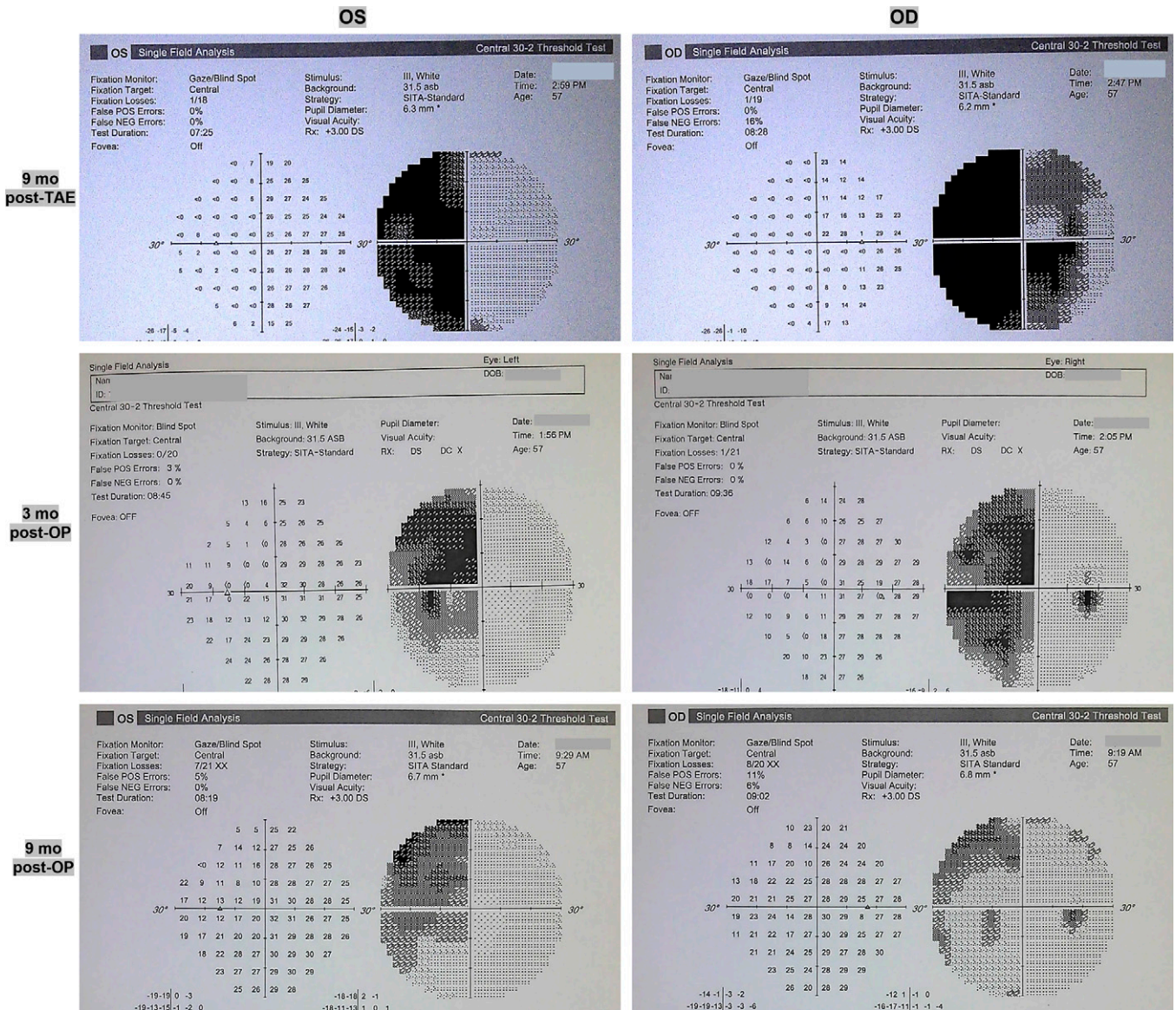


FIG. 3. The sequential examination results of the patient's visual acuity/field. Six months after stenting, the patient started to report progressive hemianopia in the left half of the visual field. The diagnosis was confirmed in a local ophthalmology clinic, and she was referred back to our hospital. Nine months after stenting, the patient had complete hemianopia in the left-sided visual fields of both eyes; the right-sided visual field of the right eye was also affected (post-TAE row). After microsurgical decompression of the optic nerve/chiasm and coagulation shrinkage of the aneurysm, her visual acuity/field improved (post-OP rows). OD = right eye; OS = left eye.

occlusion. To perform thrombectomy without clipping was risky if the endothelium growing on the flow diverter was not strong enough to hold the ICA flow. Hence, we finally decided to coagulate and shrink the aneurysm only, which was a rather conservative measure. We coagulated the surface of the aneurysm and its neck as much as we could by using both straight and angled bipolar forceps, trying to disrupt the vasa vasorum in the aneurysm wall and shrink its size. Fortunately, the patient's visual fields improved after the operation (Fig. 3), and MRA of the brain 3 months after the operation showed that the aneurysm had diminished in size (Fig. 1).

Lessons

Possible pathomechanisms for the continuous size expansion of the aneurysm, which had been excluded by the flow diverter, are (1) the consequences of the reaction of adventitial vasculature (vasa vasorum) and neointima formation to endovascular injury/trauma after the flow diverter placement,^{6,7} (2) intramural hemorrhage and inflammatory reaction,^{8,9} and (3) intrathrombotic vascular channels and subsequent establishment of blood flow between the parent artery and channels.^{10,11} Vasa vasorum are adventitial vessels supplying nutrients/oxygen to the tunica adventitia/media and removing metabolic wastes from the outer

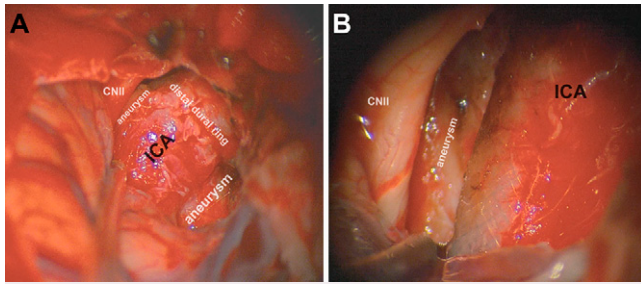


FIG. 4. Intraoperative findings. The Dolenc approach was used to open the optic canal and achieve optic nerve (cranial nerve II [CNII]) decompression. **A:** After the extracranial anterior clinoidectomy, the distal dural ring, the falciform ligament, and the optic nerve sheath were incised extensively for further optic nerve decompression. **B:** We found that the aneurysm wall was thick, and the grid-like flow diverter was clearly seen through the ICA. We coagulated the surface of the aneurysm and its neck as much as we could by using both straight and angled bipolar forceps, trying to disrupt the vasa vasorum in the aneurysm wall and shrink its size.

vessel walls. Adventitial inflammation leads to weakening of the tunica media by the release of proinflammatory factors that invade the media and degrade the extracellular matrix, the elastic lamina of the vascular wall, and the integrity of the vessel lumen, which results in dilation of the vessel and aneurysm formation.^{6,10,12} Neovascularization is also an important factor here, and repeated subadventitial hemorrhages from the new vasa vasorum could play a major role in aneurysm pathogenesis.¹² The vasa vasorum as a potential source of aneurysmal growth should be recognized. However, traditional DSA studies may fail to diagnose enlargement of the aneurysm sac, and high-resolution vessel wall magnetic resonance imaging should be considered.¹³ Another pathomechanism, the water-hammer effect, was postulated to explain the aneurysm growth after intraaneurysmal balloon occlusion,¹⁴ but this embolization material is no longer available in our country.

From previous study, 15% of patients with ICA aneurysm had worsening cranial neuropathy in the short term after placement of a flow diverter, and this number decreased to 4.8% by 12 months, which is important to note for counseling patients if flow diversion is to be used.³ In our case, the deterioration of the patient's visual acuity/field was fast and severe, despite the use of high-dose steroids,¹⁵ which prompted us to call off the wait-and-see approach. Hence, we finally chose the surgical treatment for the compressive optic neuropathy. In recent literature review, despite reports of visual improvement with corticosteroids, optic canal decompression, and medical therapy for indirect traumatic optic neuropathy, the weight of published evidence did not demonstrate consistent benefits for either intervention.¹⁶ Treatment strategies should be customized for each patient.

Microsurgical decompression of the optic nerve could be considered a salvage alternative for a patient with a flow-diverter-excluded-but-still-growing large ICA aneurysm. Whether the coagulation technique of the flow-diverter-occluded aneurysm alone is enough to cause satisfactory shrinkage and the interaction between the flow diverter and the aneurysmal vasa vasorum/neointima formation should be further examined.

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Disclosures

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

Conception and design: Liao, Tsuei, Chen, Cheng, Shen. Acquisition of data: Liao, Tsuei, Fu, Chen. Analysis and interpretation of data: Tsuei, Chen. Drafting the article: Liao, Fu. Critically revising the article: Liao, Cheng, Shen. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Liao. Administrative/technical/material support: Liao, Tsuei, Chen, Cheng, Shen. Study supervision: Liao, Shen.

Supplemental Information

Previous Presentations

The case was presented previously in a webinar of the Asian Congress of Neurological Surgeons, March 9, 2022.

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