



Subarachnoid hemorrhage-negative Terson syndrome after intracranial artery treatment with a flow diverter device

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ABSTRACT

Purpose: To report a case of subarachnoid hemorrhage-negative Terson syndrome following intracranial artery treatment with flow diverter stents.

Observations: A 40-year-old Asian woman presented with floaters in her right eye after treatment of an intracranial aneurysm with flow diverter stents. Vitreous hemorrhage and sub-inner limiting membrane (sub-ILM) hemorrhage were present in her right eye. On fluorescein angiography, contrast perfusion and vascular occlusion were not noted. Magnetic resonance imaging (MRI) did not show any evidence of subarachnoid hemorrhage (SAH). We hypothesize that the bleeding was due to Terson syndrome associated with intracranial treatment with the flow diverter stents. During follow-up, the vitreous hemorrhage and sub-ILM hemorrhage disappeared, and the floaters in her vision improved.

Conclusions and Importance: This is the first reported case of vitreous hemorrhage and sub-ILM hemorrhage that should be considered to be Terson syndrome, after flow diverter stents treatment in the absence of SAH.

1. Introduction

Aneurysm coil embolization is performed as a treatment for large and giant cerebral aneurysms, but the occlusion rate is low, and the recurrence rate is high.¹ In recent years, a device called the flow diverter stent (FD) has been used for large and giant cerebral aneurysms, and its effectiveness and safety profile have been reported.² These devices are associated with potential risks, including ophthalmic complications such as retinal artery occlusion, ischemic optic neuropathy, and eye movement disorders.³

Terson syndrome was reported in the 1900s as a vitreous hemorrhage caused by an intracranial hemorrhage.⁴ It has been reported to occur in 8–19.3% of subarachnoid hemorrhage (SAH) cases.^{5,6} However, intraocular hemorrhage that was thought to result from Terson syndrome in the absence of SAH has also been reported, e.g., cerebral venous sinus thrombosis, treatment for intracranial disease, and viral meningitis.^{7–10}

Here, we present the first case of vitreous hemorrhage (VH) and sub-inner Limiting Membrane (sub-ILM) hemorrhage not associated with the presence of SAH, after cerebral aneurysm treatment with FD.

1.1. Case report

A 40-year-old Asian woman was referred to the neurosurgery service in our hospital to treat bilateral internal carotid artery aneurysms in the cavernous sinus. The left internal carotid artery aneurysm was treated with FD through neurosurgical intervention. The patient made good progress after the procedure, and then proceeded to treatment of the right internal carotid artery aneurysm with FD. The day after her right-sided operation, she noticed floaters in her right field of vision when she woke up. She also described a pain in the back of her right eye. Her past medical history was significant for iron-deficiency anemia (her preoperative and postoperative hemoglobin values were 8.6 g/dl and 8.5 g/dl, respectively). Her medication history comprised of aspirin 300 mg, clopidogrel sulfate 75 mg, and vonoprazan fumarate 10 mg, prior to and following surgery.

On ophthalmic examination, her right visual acuity was 20/16, and her intraocular pressures were 14 mmHg bilaterally. Her anterior ocular segment was normal. VH and retinal hemorrhage were noted in the right eye, but no hemorrhage was found in the left eye (Fig. 1A and B). Fluorescence angiography (FA) confirmed hypofluorescence due to

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blocking by the hemorrhages, but no obvious hyper-fluorescence indicative of neovascularization or vascular occlusion (Fig. 1C). On Spectral Domain Optical Coherence Tomography (SD-OCT), the retinal hemorrhage was shown to be sub-ILM hemorrhage (Fig. 2). Magnetic resonance angiography (MRA) showed that the FD were placed in the aneurysm, and the aneurysms had shrunk accordingly. MRI findings were suspicious for microcerebral infarction, but no obvious SAH was seen. Postoperative headaches were attributed to postoperative nausea and vomiting because no intracranial lesion was seen. The patient was managed with ongoing surveillance, the VH and sub-ILM hemorrhage gradually disappeared, and the floaters disappeared. There was no obvious abnormality on FA seen after the symptoms improved.

2. Discussion

Coil embolization and parent artery occlusion have been used for the treatment for giant cerebral aneurysms, but high recurrence rates are a recognized issue.¹ FD is a mesh stent device with high metal coverage for the treatment of cerebral aneurysms.² It suppresses blood flow in the aneurysm by being placed in the parent artery, and promotes thrombosis to the aneurysm. In the Pipeline for Uncoilable or Failed aneurysms Study (PUFS), 86.8% of aneurysms were completely occluded six months later.² However, ocular complications such as retinal artery embolism (0.9%), diplopia (0.9%), and worsening visual fields (0.9%) were reported in this trial.² When the ophthalmic artery is included in the stent placement site, the frequency of postoperative ophthalmologic complications associated with the use of FD has been reported to be 39.3%.³ Retinal artery occlusion, ischemic optic neuropathy, and eye movement disorders have been reported.¹⁻³ In this case, these complications did not occur, but the patient did experience vitreous hemorrhage and sub-ILM hemorrhage.

Terson syndrome is characterized by intraocular hemorrhage associated with intracranial lesions.⁴ The pathophysiology of Terson syndrome is not fully understood. One leading theory is that it results from increases in subarachnoid pressure around the optic nerve due to a sudden increase in intracranial pressure.¹¹ The increase in pressure causes compression of the central retinal vein, and venous hypertension results in rupture of thin retinal vessels.¹¹ Terson syndrome is typically caused by SAH. However, fundus hemorrhage caused by intracranial lesions or intracranial treatment has been previously reported as Terson syndrome without SAH.⁷⁻¹⁰ Intracranial lesions included cerebral venous sinus thrombosis⁷ and viral meningoencephalitis.⁸ In both cases, the etiology of bleeding was intracranial hypertension. Asahi et al. suggested that increased vascular permeability associated with inflammation contributed to the increase in intracranial pressure.⁸ As for intracranial treatments causing fundus bleeds, Hoving et al. reported postoperative macular hemorrhage as a result of increased intracranial pressure during an endoscopic third ventriculostomy procedure for hydrocephalus.⁹ Gupta et al. reported intraretinal hemorrhage after coil embolization following treatment of an unruptured aneurysm. The authors thought that bleeding was due to increased intracranial pressure or ischemia during the procedure, but they did not find clear evidence of

either on MRI. Thus, the authors speculated that the patient had anti-phospholipid antibody syndrome and that anticoagulants made her prone to easy bleeding.

In this case, the patient became aware of floaters and a headache the day following FD treatment, and sub-ILM hemorrhage and VH were observed on examination. FA did not show a clear source of bleeding. The preoperative aneurysm was located in the cavernous sinus and included the origin of the ophthalmic artery. The aneurysm was so big that four FDs were used during the procedure. We theorize that intracranial hypertension might occur because of the complicated intracranial procedure. Although intracranial pressure was not measured intraoperatively, postoperative headaches may indicate that increased intracranial pressure occurred while surgery was ongoing.

To prevent ischemic complications associated with FD treatment, anticoagulant medications are administered, starting 1 week before surgery to 3 months or more after surgery.² This may have promoted the development of VH and sub-ILM hemorrhage.

The treatment of Terson syndrome is observation, because of the spontaneous recovery of the hemorrhage within a few months. However, vitrectomy is often performed at an early stage to prevent both retinal dysfunction due to hemorrhage or the onset of other lesions, such as epiretinal membranes.¹²⁻¹⁴ In our case, vitrectomy was not performed because the hemorrhage was small, and it was early after cerebral aneurysm surgery. The VH and sub-ILM hemorrhage resolved and the subjective symptoms improved a few days after the procedure. The fundus examination and FA failed to show any bleeding sources, after the patient improved symptomatically.

3. Conclusions

To the best of our knowledge, this report describes the first case of SAH-negative Terson syndrome after cerebral aneurysm treatment using an FD. Our findings suggest that if floaters occur after intracranial artery treatment using FD, there is a possibility of developing Terson syndrome.

Patient consent

Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the identification of the patient.

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Authorship

All authors attest that they meet the current ICMJE criteria for authorship.

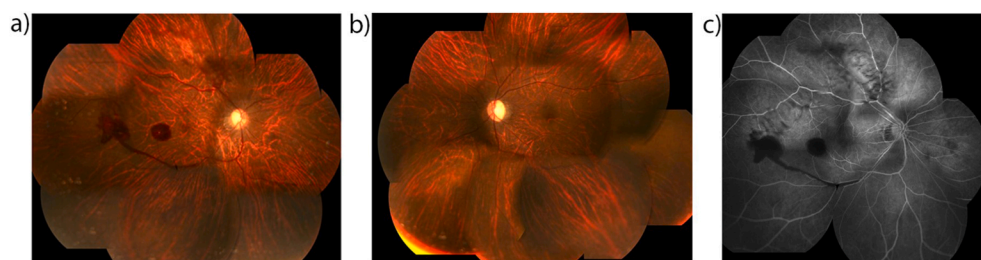


Fig. 1. Imaging on presentation demonstrating ocular hemorrhage of the right eye. A. Fundus photography of the right eye. Vitreous hemorrhage in the wide area and retinal hemorrhage on the temporal side of macula were observed. B. Fundus photography of the left eye. There was no hemorrhage or other abnormalities noted. C. Fluorescence angiography of the right eye. There was hypofluorescence due to blocking by the hemorrhages, but no obvious hyper-fluorescence indicative of neovascularization or vascular occlusion.

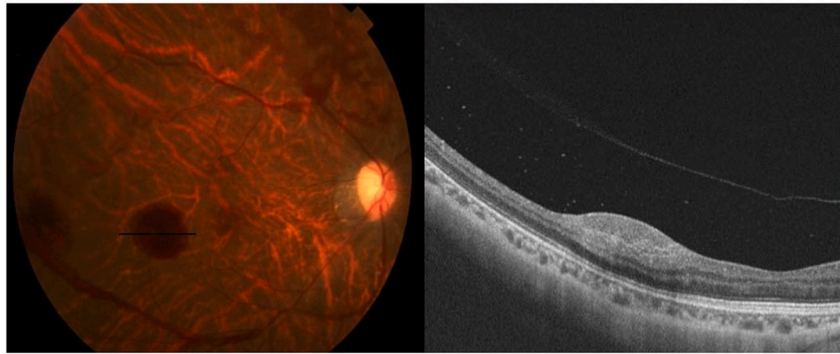


Fig. 2. Spectral domain ocular coherence tomography (SD-OCT) of the right eye. Retinal hemorrhage in the temporal side of the macula was shown to be sub-inner limiting membrane (sub-ILM) hemorrhage.

Conflicts of interest

The following authors have no financial disclosures: TM, SM, SH, MK, ET, JY, TT, KK, and YO.

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