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DOI: 10.1159/000511110	1

DOI: 10.1159/000511110 Received: July 13, 2020 Accepted: August 23, 2020 Published online: July 16, 2021 © 2021 The Author(s). Published by S. Karger AG, Basel www.karger.com/cop This article is licensed under the Creative Commons Attribution-NonCommercial 4.0 International License (CC BY-NC) (http://www.karger.com/Services/OpenAccessLicense). Usage and distribution for commercial purposes requires written permission.

Case Report

A Case of Idiopathic Dense Vitreous Hemorrhage: Suspected Rupture of a Large Retinal Arterial Macroaneurysm on the Optic Disc

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Keywords

Vitreous hemorrhage · Hematoma · Retinal arterial macroaneurysm · Optic disc

Abstract

We report a novel case of vitreous hemorrhage associated with suspected rupture of 2-discdiameter retinal arterial macroaneurysm on the optic disc. A 90-year-old woman presented with blurred vision (sudden onset) in her left eye. Examination of the fundus revealed acute onset vitreous hemorrhage of unknown origin without retinal detachment. She underwent vitrectomy, but after excision of the dense vitreous hemorrhage, a 2-disc-diameter hematoma appeared on the optic disc and was removed promptly. Because the bleeding at the base of the hematoma was of arterial origin and pulsating, the first vitrectomy could not achieve hemostasis. Five days after the first surgery, we performed a second vitrectomy. This revealed a subretinal hemorrhage along the superior and inferior arcade vessels and a macular hole, which was almost completely closed with an inverted internal limiting membrane flap. Unfortunately, the macular hole reopened 41 days after the second surgery. In patients presenting with only a large hematoma on the optic disc, it might be prudent to leave the hematoma. However, this large retinal arterial macroaneurysm was on a rare location on the optic disc, making it doubly difficult for the surgeons to diagnose and choose the best option intraoperatively. The differential diagnosis for dense vitreous hemorrhage of unknown origin should include a large retinal arterial macroaneurysm on the optic disc.

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Introduction

Dense vitreous hemorrhage occurs in conditions such as proliferative diabetic retinopathy, age-related macular degeneration, rhegmatogenous retinal detachment, retinal vein occlusion, retinal arterial macroaneurysms, posterior vitreous detachment, uveitis, trauma, and the Telson syndrome. In such cases, vitrectomy is often used to remove the vitreous hemorrhage and treat the primary disease or cause to avoid recurrence.

Herein, we report a case of dense vitreous hemorrhage with a 2-disc-diameter hematoma on the optic disc. After removing the hematoma, pulsating arterial bleeding caused subretinal hemorrhage and a macular hole.

Case Presentation

A 90-year-old woman was referred to the Osaka University Hospital with acute onset vitreous hemorrhage of unknown origin. She reported a sudden onset of blurred vision in her left eye. On examination, her best-corrected visual acuity was light perception in the left eye and 20/16 in the right eye, and her intraocular pressure was 13.0 mm Hg OD and 12.0 mm Hg OS. Fundus examination of the left eye by B-mode ultrasonography revealed dense vitreous hemorrhage without retinal detachment. A bulge on the optic disc was also detected (Fig. 1a). Additionally, she had a history of hypertension, but her blood pressure was well-controlled with oral medication. Results of preoperative systemic examinations revealed a thoracic aortic aneurysm. A pars plana vitrectomy was performed to treat the dense vitreous hemorrhage in her left eye. After excision of the vitreous hemorrhage, a two-disc-diameter hematoma, which looked encapsulated, appeared on the optic disc and was promptly removed (Fig. 1b). Because the source of the hemorrhage was arterial and pulsating, the first vitrectomy could not achieve hemostasis (Fig. 1c, d). Aside from the optic disc hematoma, other areas of the eve, such as the macula, arcade vessels, and the peripheral retina, showed no lesions. Five days after the first surgery, we performed a second vitrectomy, which revealed subretinal hemorrhage along the superior and inferior arcade vessels and a macular hole (Fig. 2a). To prevent recurrent bleeding, we trimmed the edges of the hematoma on the optic disc to minimize its size. Because the macular hole, caused by the retinal arteriolar aneurysm rupture, was refractory [1, 2], we closed it using the indocyanine green-assisted inverted internal limiting membrane flap technique (Fig. 2b). The macular hole, which was almost closed (Fig. 2c), reopened 41 days after the second vitrectomy (Fig. 3a, c, d). The patient did not wish to undergo further surgeries. The left eye was actively monitored at subsequent follow-up visits.

Discussion

In our case, removal of the hematoma on the optic disc resulted in pulsating arterial bleeding. We could not make a definitive diagnosis, however, and presume with a high probability that the cause of this vitreous hemorrhage was a ruptured arteriolar aneurysm for several reasons. We did not find any underlying issues that could cause vitreous hemorrhage other than the hematoma on the optic disc. The hematoma looked encapsulated with a film on its surface. Additionally, the patient had a history of hypertension, and a thoracic aortic aneurysm was found during the preoperative examination. Arterial macroaneurysms are associated with systemic vascular conditions such as hypertension in about 20% of the cases [3].



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Fig. 1. a B-mode ultrasonographic image of the left eye at the first clinical examination, showing vitreous hemorrhage without retinal detachment. A bulge on the optic disc can be observed. **b** After removal of the vitreous hemorrhage, an encapsulated hematoma can be seen on the optic disc. **c** Immediately after removal of the hematoma, the pulsating arterial bleeding could not be stopped. **d** An attempt to remove the pulsating arterial bleeding resulted in new vitreous hemorrhage.

On that assumption, discontinuing the second surgery without removing the hematoma on the optic disc was judged as the best option because removing the hematoma would have resulted in pulsating arterial bleeding, making hemostasis difficult. However, this case presented 3 obstacles that made diagnosis difficult. First, arterial macroaneurysms on the optic disc are relatively rare, accounting for only 3.7% of all retinal arterial macroaneurysms cases. Acquired retinal arterial macroaneurysms often develop in the posterior pole and are mostly confined to the first 3 orders of retinal arterial bifurcations [3].

The second reason that complicated the diagnosis of the optic disc hematoma as a retinal arterial macroaneurysm was its size. Tezel et al. [4] reported that the mean size of retinal arterial macroaneurysms was about 281.60 \pm 57.28 µm. However, the present retinal arterial macroaneurysm was about 2-disc-diameters in size. One case of a large retinal arterial macroaneurysm at the first bifurcation has been reported [5]; however, there are no reports of a large retinal arterial macroaneurysm on the optic disc. Intraoperatively, the hematoma was red, not pulsatile, and appeared to be filled with coagulants.



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Fig. 2. a Subretinal hemorrhage was seen around the optic disc. To avoid recurrent bleeding, we trimmed the edges of the hematoma on the optic disc to minimize its size. **b** One-third disc-diameter macular hole and preretinal hemorrhage from it are seen (white arrow). Inverted ICG-assisted internal limited membrane flap (arrowhead) technique was performed to close the macular hole. **c** OCT images acquired after the second surgery show that the macular hole was almost completely closed. ICG, indocyanine green; OCT, optical coherence tomographic.

Therefore, the original retinal arterial macroaneurysm might have been smaller, as stated in previous reports [3, 4, 6–8]. However, we estimate that the size of the macroaneurysm in our patient increased gradually because the retinal arterial pressure is higher in the optic disc than in any other part of the eyeball. In fact, a previous report stated that the diameter and area of macroaneurysms correlate significantly with the diameter of the relevant arterial segment [4]. Moreover, Tezel et al. [4] concluded that hemorrhagic macroaneurysms are close to the optic disc and are located on relatively large arterioles. The present case is consistent with this conclusion.

Finally, dense vitreous hemorrhage did not allow us to diagnose its cause preoperatively. All the preoperative clinical information we had was the B-mode ultrasonographic image shown in Figure 1. Retinal arterial macroaneurysms usually undergo spontaneous involution [5, 6, 9, 10]. To the best of our knowledge, a case of dense vitreous hemorrhage that was associated with a rupture of an arterial macroaneurysm on the optic disc has not been reported yet. This made the definitive diagnosis and decision-making during the first surgery very challenging. Complications in this case were subretinal hemorrhage and formation of a macular hole. Generally, arteriolar aneurysms on the optic disc present limited complications [5, 6, 9, 10], and the prognosis is good in most cases [3]. The first

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Fig. 3. a Fundus photography of the left eye 41 days after the second surgery showing subretinal hemorrhage and a macular hole. **b** An OCT image of the optoce disc showing the lesion on the optoc disc, with the residual hematoma (white dased line in **a**). **c** The temporal optic disc lesion remained hypofluorescence throughout the fluorescein angiography examination. **d** An OCT image showing the reopened macular hole 41 days after the second surgery (white arrow in **a**). OCT, optical coherence tomographic.

surgery presented no abnormal findings except the hematoma on the optic disc; however, on excision of the optic disc hematoma, pulsating arterial bleeding occurred from the disc and hemostasis could not be achieved. The surgery was concluded with high intraocular pressure to achieve hemostasis. The second surgery was performed 5 days after the first. The first surgery resulted in subretinal hemorrhage and the formation of a macular hole. We surmised that the optic disc hemorrhage that had developed at the end of the first surgery stopped, coagulated, and remained on the optic disc. The high intraocular pressure prevented further pulsatile, vigorous bleeding from entering the vitreous cavity. Consequently, it flowed under the retina and passed into the fovea, where the adhesion was weak. In addition, removal of the arterial macroaneurysm in the first surgery might have resulted in the formation of a tear in the retinal nerve fiber layer surrounding the optic disc, which might have accelerated the subretinal hemorrhage.

Unfortunately, this case showed an unexpected negative clinical course. We elected to present this case because we believe it must be difficult for many surgeons to diagnose and choose the best option intraoperatively when treating such rare large retinal arterial macroaneurysm on the optic disc.

Conclusion

In patients presenting with only a large hematoma on the optic disc, it might be prudent to leave the hematoma.



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Statement of Ethics

This research complied with the guidelines for human studies and was conducted ethically, following the World Medical Association Declaration of Helsinki. Written informed consent was obtained from the patient for publication of this case report and any accompanying images. The authors have no ethical conflicts to disclose.

Conflict of Interest Statement

The authors have no conflicts of interest to disclose.

Funding Sources

This study received no funding.

Author Contributions

S.T., K.N., H.S., and K.N. participated in drafting the manuscript. S.T., K.N., and H.S. participated in the diagnosis and treatment of the patient.

References

- 1 Sato R, Yasukawa T, Hirano Y, Ogura Y. Early-onset macular holes following ruptured retinal arterial macroaneurysms. Graefes Arch Clin Exp Ophthalmol. 2008;246(12):1779–82.
- 2 Kawaji T, Takasu I, Takano A, Sato T, Hashida M. Internal limiting membrane peeling-repositioning technique for macular hole after macular hemorrhage associated with rupture of retinal arterial macroaneurysm. Retina. 2019;39(Suppl 1):S84–6.
- 3 Palestine AG, Robertson DM, Goldstein BG, Rochester MD. Macroaneurysms of the retinal arteries. Am J Ophthalmol. 1982;93(2):164–71.
- 4 Tezel T, Günalp I, Tezel G. Morphometrical analysis of retinal arterial macroaneurysms. Doc Ophthalmol. 1994;88(2):113–25.
- 5 Ohno-Matsui K, Hayano M, Futagami S, Tokoro T, Ohno S. Spontaneous involution of a large retinal arterial macroaneurysm. Acta Ophthalmol Scand. 2000;78(1):114–7.
- 6 Kaori H, Yoshinori M, Kazuha O, Shuichi Y. Three cases of retinal arterial macroaneurysm on the optic disc. Nichigan Zasshi. 2010;114(9):801–4.
- 7 Ozgonul C, Besirli CG. Arterial macroaneurysm of the optic disc. Am J Ophthalmol Case Rep. 2018;10:279–81.
- 8 Panton RW, Goldberg MF, Farber MD, Panton RW, Goldberg MF, Farber MD. Retinal arterial macroaneurysms: risk factors and natural history. Br J Ophthalmol. 1990;74:595–600.
- 9 Quhill F, Smith JMA, Scotcher SM. Arterial macroaneurysm on the optic disc. Eye. 2004;18(3):321-2.
- 10 Das-Bhaumik RG, Lindfield D, Quinn SM, Charles SJ. Optic disc macroaneurysm in evolution: from incidental finding to branch retinal artery occlusion and spontaneous resolution. Br J Ophthalmol. 2011;95(1):145–55.

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