Hemorrhagic Macular Cysts in Patients with Terson’s Syndrome Observed through an Ophthalmic Surgical Microscope

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Abstract
The characteristics of hemorrhagic macular cysts (HMCs) in patients with Terson’s syndrome, focusing on the vitreoretinal interface observed through an ophthalmic surgical microscope, are described. Between May 2015 and February 2022, 19 eyes (17 patients) with vitreous hemorrhage (VH) occurring after subarachnoid hemorrhage underwent pars plana vitrectomy. After removing dense VH, 2 of 19 eyes had HMCs. In both cases with HMCs, they formed a dome-like shape and were located beneath the internal limiting membrane (ILM), lying beyond the clean posterior precortical vitreous pocket (PPVP) without hemorrhage despite the severe VH. Based on the microsurgical findings, it appears that two types of HMCs consisting of subhyaloid and sub-ILM hemorrhages in Terson’s syndrome may be involved in the impairment of adhesion of the posterior border of the PPVP and the ILM surface of the macula due to micro bleeding, and that the PPVP may prevent the sub-ILM type of HMC from breaking into the subhyaloid space and transforming into the subhyaloid type. In conclusion, the PPVP may play an important role in the formation of HMCs in Terson’s syndrome.
**Introduction**

The association of vitreous hemorrhage (VH) with subarachnoid hemorrhage (SAH) is known as Terson’s syndrome [1]. The most widely accepted mechanism for Terson’s syndrome is that dilatation of the retrobulbar optic nerve due to rapid intracranial pressure mechanically compresses the central retinal vein and that venous hypertension results in a rupture of thin retinal vessels. In approximately one-third of Terson’s syndrome, a premacular accumulation of blood, termed a hemorrhagic macular cyst (HMC), may also be found [2, 3]. Based on the blood’s relation to the internal limiting membrane (ILM), two types of HMCs can be distinguished in Terson’s syndrome [2]. One is a subhyaloid hemorrhage if the blood is located anterior to all layers of the retina, and the other is a sub-ILM hemorrhage if the blood has accumulated under the ILM. HMCs have some characteristic features such as dome-shaped membrane in front of posterior pole on B-scan ultrasonography or a highly reflective band above the premacular hemorrhage, corresponding to the ILM, and an overlying patchy membrane with low optical reflectivity consistent with the subhyaloid on optical coherence tomography (OCT) of the macula. Although several cases of HMC in Terson’s syndrome have been reported, their pathogeneses remain unclear [4–6]. Two cases of HMC with severe VH in Terson’s syndrome, focusing on the vitreoretinal interface observed through an ophthalmic surgical microscope, are reported.

**Case Report/Case Presentation**

Between May 2015 and February 2022, 19 eyes (17 patients) with VH occurring after SAH underwent pars plana vitrectomy (PPV). After removing dense VH, 2 of 19 eyes had HMCs. The procedures used conformed to the tenets of the Declaration of Helsinki and were approved by the local ethics committee (Ethics Committee of Nakamura Memorial Hospital).

**Case One**

A 44-year-old man presented with sudden onset of a severe headache and unconsciousness. After being admitted to our emergency room, he was diagnosed with an SAH resulting from left vertebral artery dissection and immediately underwent stent-assisted coil embolization. After the procedure, he recovered consciousness but had visual loss in both eyes. On examination, best corrected visual acuity (BCVA) was 20/200 in the right eye and counting fingers in the left eye. Fundoscopy of both eyes showed dense VH consistent with Terson’s syndrome. A 25-gauge PPV was performed in the right eye 22 days after the onset of SAH. During the vitrectomy procedure, a clean posterior vitreous space without hemorrhage was observed, and it was confirmed that it could be the posterior precortical vitreous pocket (PPVP) because a bag-shaped space and posterior vitreous cortex dyeing by injection of triamcinolone acetonide were seen under the surgical microscope (supplement video). After induction of posterior vitreous detachment (PVD), brilliant blue G was then used to stain the ILM [7, 8]. A small opening was created in the edge of sub-ILM hemorrhage by a backflush needle. Subsequently, dense white-yellowish blood was removed. Two days after the surgery of the right eye, a 25-gauge PPV was performed in the left eye, and sub-ILM hemorrhage was not observed. Although his BCVA improved to 20/20 in both eyes after surgery, visual distortion continued in the right eye. Spectral-domain OCT showed a wavy shape of the retinal pigment epithelium layer and irregularity of the photoreceptor inner/outer segment junction layer in his right eye, indicating retinal damage from sub-ILM hemorrhage (Fig. 1).
Case Two

The second patient was a 49-year-old man with a diagnosis of SAH resulting from right vertebral artery dissection. He immediately underwent stent-assisted coil embolization. On regaining consciousness, he noticed visual loss in his right eye. On examination, BCVA in the right eye was counting fingers and that in the left eye was 20/20. Fundoscopy of the right eye showed a blurred yellowish-white opacity in the posterior pole with VH (Fig. 2a). Swept-source OCT showed a patchy hyperreflectivity under the vitreoretinal interface, suggesting sub-ILM hemorrhage (Fig. 2b). A 25-gauge PPV was performed in the right eye 76 days after SAH onset. During PPV, the PPVP without hemorrhage and sub-ILM hemorrhage were observed under the surgical microscope (Fig. 3a). The ILM was stained by brilliant blue G after the induction of PVD. A small opening was created in the edge of the ILM overlying the hemorrhage by a backflush needle. Subsequently, dense white-yellowish blood was removed, similar to case one (Fig. 3b). Three days after the surgery, the BCVA in the right eye improved to 20/20, and fundus photography and swept-source OCT showed successful removal of the sub-ILM hemorrhage. His visual distortion improved 2 months after the surgery.

Discussion/Conclusion

Both of the present cases with HMCs were sub-ILM type behind dense VH in Terson’s syndrome. Several authors have reported that the majority of HMCs in Terson’s syndrome are sub-ILM type rather than subhyaloid type [4, 9, 10]. Meier et al. [11] reported that all seven eyes in children with HMC caused by Terson’s syndrome and shaken baby syndrome showed...
sub-ILM hemorrhage, not subhyaloid hemorrhage. In the present 19 eyes of 17 patients with Terson's syndrome who underwent PPV, there was no case with subhyaloid hemorrhage with VH. It is impossible to find the subhyaloid hemorrhage behind dense VH under an ophthalmic surgical microscope. Therefore, it may not have been possible to observe the subhyaloid type of HMC. Furthermore, there might have not been a case with subhyaloid hemorrhage after resorption of VH because the PPV in the present 19 eyes for VH was performed relatively early in the course of Terson's syndrome (mean 63.9 ± 41.1 days).

Several authors suggest that whether a hemorrhage will hydrodissect the ILM and result in a sub-ILM cyst or will break through the ILM and posterior hyaloid face is determined by some factors such as the pressure of the extravasatum and resistance of the ILM [4–6]. Morris et al. [3] reported that blood pooling beneath the ILM in the posterior pole is facilitated by the fact that the ILM thickens here, and its attachment plaques to the retina disappear. The present 2 cases with HMCs did not have PVD, and the dome-shaped HMCs were located beneath the ILM, lying beyond the clean PPVP without hemorrhage. The PPVP, which Kishi...
and Shimizu [12] named since they found the presence of a bursa premacularis, is a liquefied lacuna anterior to the macular area that is physiologically present in the vitreous without PVD. The posterior vitreous cortex, which comprises the posterior border of the PPVP, is more firmly adhered physiologically to the ILM surface of the macula [13]. Thus, it appears that two types of HMCs in Terson’s syndrome may also be involved in the impairment of adhesion of the posterior border of the PPVP and the ILM surface of the macula due to micro bleeding and that the unaffected PPVP may prevent the sub-ILM type of HMC from breaking into the subhyaloid space and transforming into the subhyaloid type.

Weingeist et al. [14] reported that biomicroscopic examination of elevated epiretinal membranes in Terson’s syndrome often showed one or more holes in the surface of the ILM and suggested that these pseudomacular holes represent sites where blood entered the vitreous cavity. In the present 19 eyes, there was a case with a macular hole after removing dense VH during PPV. It is unclear if the patient had HMC, but it was observed during surgery that she had a complete PVD and that the ILM was separating from the neurosensory retina. These findings may suggest that sub-ILM hemorrhage with PVD may produce a micro hole on the HMC because of rupture of the vulnerable ILM surface without firmly adhering to the posterior wall of the PPVP and enter the vitreous cavity. In conclusion, the presence of PPVP may play an important role in the formation of HMCs in Terson’s syndrome.

Statement of Ethics

This study adhered to the Declaration of Helsinki. The patients provided written informed consent for the publication of this case report and any accompanying pictures. This study protocol was reviewed, and the need for approval was waived by Nakamura Memorial Hospital.

Conflict of Interest Statement

The authors have no financial disclosures.

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Author Contributions

Kaori Hanai treated the patients and collected clinical data. Kaori Hanai and Masato Hashimoto wrote the manuscript, and Hirohiko Nakamura revised the manuscript. Kaori Hanai, Masato Hashimoto, and Hirohiko Nakamura approved the final version of the manuscript. The authors agree to be responsible for all aspects of this study.

Data Availability Statement

All data generated or analyzed during this study are included in this article. Further inquiries can be directed to the corresponding author.
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