

Terson syndrome with macular hole: A case report

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Case report

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Abstract

Background Terson's syndrome with macular hole (MH) is rarely seen, and the mechanism of which is not clear. Here we report a case of Terson Syndrome with the inner limiting membrane (ILM) peeled off spontaneously associated with a rare finding: MH. Case presentation This report presents the case of a 36-year-old female patient with aneurysmal subarachnoid hemorrhage (SAH) and Terson syndrome in the right eye was admitted to our hospital with blurred vision in August 2018. Pas plana vitrectomy (PPV) was performed in the right eye, a hyaloid detachment and dyeing of the ILM with indocyanine green (ICG) was assisted. After removal of the vitreous hemorrhage (VH), a full-thickness MH was noted and we also noticed a particular aspect: the ILM was already peeled spontaneously. So we conducted gas tamponade, and face-down positioning after PPV. At two weeks follow-up, spectral domain optical coherence tomography (SD-OCT) confirmed that the MH had closed, while the thickness of nasal retina was 0.137 nm thicker than that in the temporal side. Her best corrected visual acuity (BCVA) was 0.15 in the right eye and 1.0 in the left eye. Conclusions MH is a rarely seen complication of Terson Syndrome. We conferred that the pathogenic mechanisms of this unusual MH may include stretching forces at the ILM-macular interface.

Background

Terson syndrome refers to a condition characterized by intraocular hemorrhage due to subarachnoid hemorrhage (SAH) in association with acutely elevated intracranial pressure^[1], and of which vitreous hemorrhage (VH) is the major symptom^{[2][3]}. In most cases, the hemorrhage is simple and can be removed by timely pas plana vitrectomy (PPV) with immediate improvement of vision^[4]. However the mechanism about VH in Terson Syndrome is still controversial.

We describe the case of a patient with Terson syndrome accompanied by VH in the right eye and in whom MH was found during the surgery of vitrectomy. We also found ILM within the macular region had already been peeled off spontaneously during the surgery. To our knowledge, there are few reports about this association. Here we report a case of Terson Syndrome with the ILM peeled off spontaneously associated with MH.

Case Presentation

A 36-year-old female patient was admitted to our hospital with decreased visual acuity in the right eye. She just had surgery for SAH that occurred 1 month ago due to the rupture of the intracranial aneurysm. Before the SAH, she hadn't noticed any changes to her vision in both eyes. An eye examination was performed by an ophthalmologist; the best corrected visual acuity (BCVA) was hand motion (HM) in the right eye and 1.0 in the left eye (Standard logarithmic visual acuity chart, Chinese Edition). Intraocular pressures and anterior segment examination were unremarkable. Fundus examination showed massive VH in the right eye. A partial posterior vitreous detachment (PVD) and dense VH was confirmed in the right eye by performing ophthalmic B-scan ultrasonography examination (Fig. 1). A head computed

tomography (CT) showed the SAH after aneurysmal rupture (Fig2). Terson syndrome was high in the differential diagnosis.

The patient underwent PPV in the right eye to remove the VH. After removal of the VH, a full-thickness macular hole was noted and the remainder of the retina appeared normal after PPV (Fig. 3 A). In order to peel the ILM, the fluid-air exchange was performed and 0.25% indocyanine green (ICG) was injected slowly under the air (Fig. 3 B), to our surprise, the region ranging from the upper vascular arch to the lower vascular arch had not been stained, while the other part of the posterior pole of the fundus was stained well (Fig. 3 C). That means the ILM had been peeled spontaneously. So we conducted gas tamponade, and face-down positioning after PPV.

At two weeks follow-up, both BCVA and spectral domain optical coherence tomography (SD-OCT) were recorded. Her BCVA was 0.15 in the right eye and 1.0 in the left eye. SD-OCT showed that the MH was closed completely, while the thickness of the nasal retina of the foveal was thicker than that in the temporal side (Fig.4).

Discussion

Terson syndrome was defined as VH secondary to subarachnoid or intracranial hemorrhage^[5]. So far, the mechanism of the blood entrance is not clear^[6]. Two pathogenetic theories^[7] have been accepted for explaining the process of Terson syndrome. One is that the blood from the SAH extended directly into the vitreous space through the intervaginal space around the optic nerve by penetrating the lamina cribrosa of the sclera. The other possible mechanism that has been proposed is that the SAH induces sudden intracranial hypertension, which is transmitted through the optic nerve sheath to the optic nerve head, thereby causing the stasis of the retinal veins and inducing the rupture of the retinal veins or the peripapillary capillaries. In fact both theories may be correct and complement each other^[8].

The clinical classification of the intraocular hemorrhage has been described as hemorrhage of subretinal, retinal, preretinal or subhyaloid, or intravitreal. Morris and colleagues^[10] provided a classification of Terson syndrome which histologically describes the potential location of the hemorrhage: sub-membranous hemorrhage (sub-ILM) and preretinal hemorrhage (between ILM and posterior hyaloid). Sub-ILM hemorrhage can often be observed in Terson syndrome^[10]. [Abed Alnabi W](#)^[11] reported a case that the appearance of perimacular folds associated with rapid accumulation of blood in the sub-ILM space was considered mainly due to the anterior-posterior traction of the ILM-macular. In our case, after removal of the dense VH, we can see clearly the dissection of the ILM and the retina, as was confirmed by staining of ICG during the vitrectomy, we hypothesize on the cause of VH in our case that the amounts of sub-ILM hemorrhage gathered large enough in a short period of time, as consequences, the ILM was torn off abruptly and a large amount of blood spread into the vitreous cavity.

A full thickness MH which was found during the vitrectomy was another rare complication in our case. It has been well accepted that the mechanism of MH is the tangential traction of vitreomacular or ILM-

macular anterior–posterior^[12]. Moreover, a sudden bloody dissection of the ILM may produce the tractional forces responsible for causing the MH. In this case, the ILM was not peeled surgically. So we conferred that the pathogenic mechanisms of this unusual MH may due to the traction of anterior-posterior on the fovea by ILM thickening or peeling.

Another sign that should not be ignored is the OCT which performed post-surgery. The OCT showed that the MH was closed, while the retinal thickness in the nasal side of the macular central foveal was much thicker than that in the temporal side. This may be explained as the traction in the nasal side is stronger than that in the temporal side, by which we infer that hemorrhage under ILM in the nasal side is more than that in the temporal side. This sign seemly supports the first theory that the blood comes from the edge of the optic nerve.

Above all, we infer that the ILM peeled off spontaneously and the MH in our case was due to ILM-macular traction coming from a sudden large amount of blood accumulation under the ILM in a short period of time. For this reason, we are ready to accept the first theory of the previously proposed mechanisms of Terson’s syndrome, who thinks the blood from the subarachnoid hemorrhage extended directly into the vitreous space through the intervaginal space around the optic nerve by penetrating the lamina cribrosa of the sclera. It is difficult to be explained by the second theory, who thinks the blood is from the ruptured retinal veins or the peripapillary capillaries like central retinal vein occlusion (CRVO). As we know flame-shaped retinal hemorrhage occurs in CRVO, only when it accompanies neovascularization, VH happens seldom largely and suddenly.

Conclusions

In conclusion, we shouldn’t ignore that macular hole is also a complication of Terson syndrome, although it is rarely happened. Because of the dense hemorrhage in vitreous, the macula cannot be visualized before surgery; the surgeon ought to discuss the possibility of a risk for repairing an MH with the patient before surgery.

Abbreviations

macular hole	MH
inner limiting membrane	ILM
subarachnoid hemorrhage	SAH
Pas plana vitrectomy	PPV
indocyanine green	ICG
vitreous hemorrhage	VH

spectral domain optical coherence tomography	SD-OCT
best corrected visual acuity	BCVA
hand motion	HM
posterior vitreous detachment	PVD
computed tomography	CT
central retinal vein occlusion	CRVO

Declarations

- **Ethics approval and consent to participate**: Ethical approval was not required as this manuscript presents a case study. It was performed in accordance with the tenets of the Declaration of Helsinki.
- **Consent for publication**: Written informed consent was obtained from the patient for publication of this case report and any accompanying images.
- **Availability of data and material**: All data generated or analysed during this study are included in this published article
- **Competing interests**: The authors declare that they have no competing interests.
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- **Authors' contributions**: HuiQi and LingZuo contributed equally to this case report and they were both major contributors in writing the manuscript. Hongtao Yan contributed to the preparation of the manuscript and figures. YanCheng are responsible for collecting information of the patient. All authors read and approved the final manuscript.
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Figures

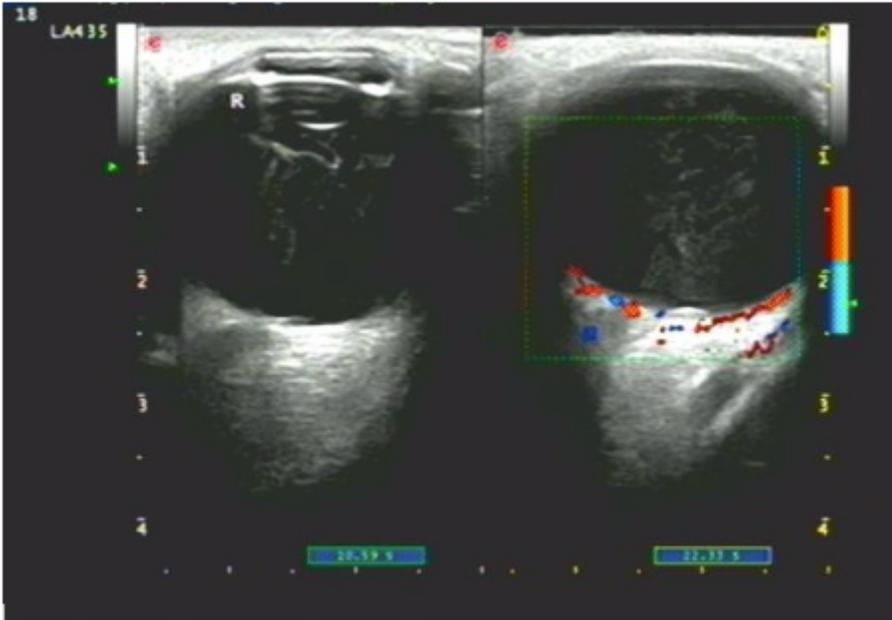


Figure 1

Ultrasonography image of the right eye: A partial PVD and dense VH.

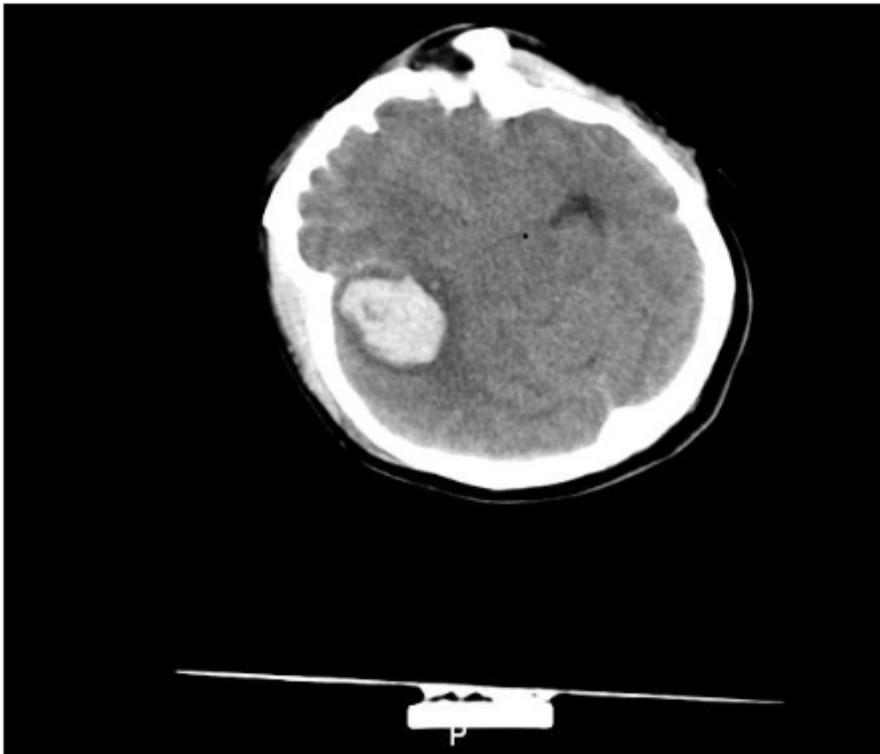


Figure 2

A head CT showed the SAH after aneurysmal rupture.

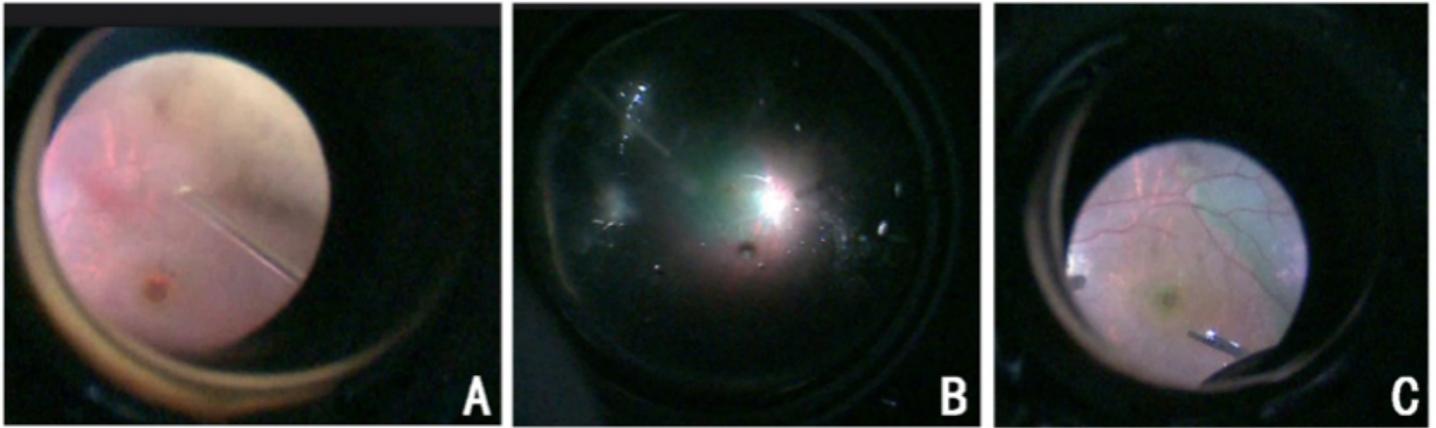


Figure 3

Images of the right eye underwent vitrectomy.

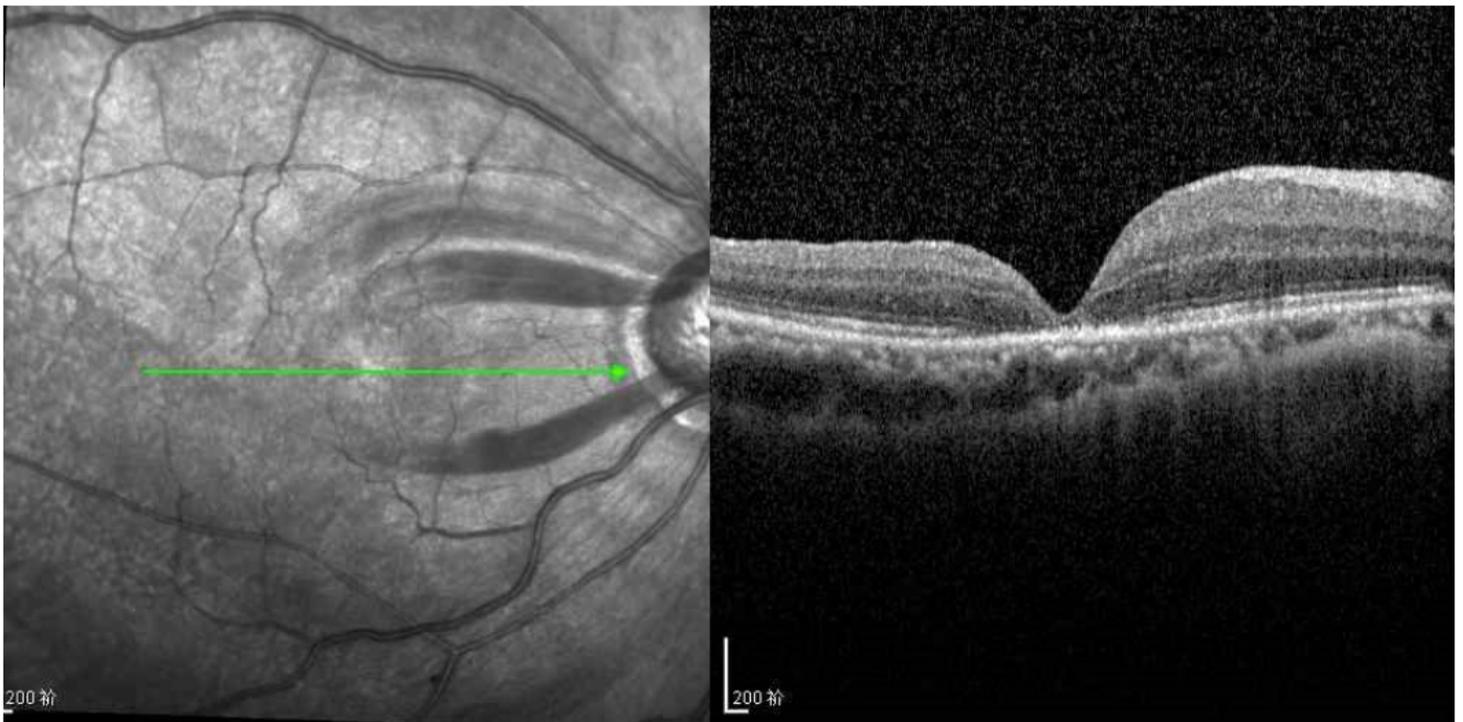


Figure 4

OCT image of the right eye at two weeks after vitrectomy. The MH was completely closed. The retinal thickness in the nasal side of the foveal is 395nm and the temporal side is 258nm.

Supplementary Files

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- [checklist.docx](#)