

# Delayed closure of macular hole secondary to Terson syndrome after vitrectomy

## A case report and literature review

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### Abstract

**Rationale:** Macular hole (MH) is a rare complication of Terson syndrome. Delayed closure of persistent MH after pars plana vitrectomy (PPV) is occasionally reported in literature, none of them is MH secondary to Terson syndrome. We describe a case of MH secondary to Terson syndrome and delayed closure occurred after PPV, and we also study the characteristics of delayed closure of persistent MH by reviewing related literatures.

**Patient concerns:** A 61-year-old man presented with vitreous hemorrhage in right eye following a subarachnoid hemorrhage due to spontaneous rupture of his right vertebral artery dissecting aneurysm. The visual acuity was hand motion in the right eye and 20/30 in the left eye. Fundus examination showed dense and diffuse vitreous hemorrhage in the right eye.

**Diagnoses:** Terson syndrome was diagnosed according to his subarachnoid hemorrhage history and vitreous hemorrhages in right eye.

**Interventions:** PPV combined with phacoemulsification and intraocular lens implantation was performed in his right eye, and internal limiting membrane (ILM) peeling was also performed due to a MH noted during the surgery.

**Outcomes:** One week after PPV, optical coherence tomography (OCT) showed a persistent MH. Without any intervention, the MH became smaller and flattened, with the best corrected visual acuity (BCVA) improved to 30/200 at 1 month after surgery. Six months later, the MH completely closed with BCVA improved to 20/40. According to our literature review, there are 8 cases of the delayed MH closure, which includes idiopathic MH (4 eyes), traumatic MH (2 eyes), and vitreomacular traction (2 eyes). There is no report about delayed closure of MH secondary to Terson syndrome. The times for these delayed closure occurred following PPV were ranged from 1 to 28 months. Holes even with obviously raised edges after PPV may spontaneously close, just like the case presented here.

**Lessons:** Delayed closure of persistent MH after PPV is rarely reported. The significance of this case is to suggest that similar patients should be monitored carefully by OCT, and additional surgery for the MH may be delayed, since delayed closure is possible. The exact mechanisms of delayed closure of persistent MH still need to be clarified.

**Abbreviations:** BCVA = best corrected visual acuity, ILM = internal limiting membrane, MH = macular hole, OCT = optical coherence tomography, PPV = pars plana vitrectomy.

**Keywords:** delayed closure, macular hole, optical coherence tomography, Terson syndrome, vitrectomy

## 1. Introduction

Terson syndrome is recognized as intraocular hemorrhage associated with subarachnoid hemorrhage, intracerebral hemorrhage, or traumatic brain injury, which was first described by

French ophthalmologist Albert Terson in 1900.<sup>[1]</sup> Intraocular hemorrhage may be present in the vitreous, subhyaloid, subretina, or subinternal limiting membrane (ILM). Multiple complications have been reported after Terson syndrome.<sup>[2]</sup> Macular hole (MH) is a rare complication of Terson syndrome. There was only 1 report of 2 patients with Terson syndrome who were found to have MH during pars plana vitrectomy for vitreous hemorrhage.<sup>[3]</sup>

Unclosed MH after vitrectomy is also known as persistent MH,<sup>[4]</sup> and there are few reports on spontaneous closure of this kind of MH. To the best of our knowledge, only 8 eyes of MH delayed closure have been reported so far.<sup>[5–11]</sup> Different names were used in the literatures to describe this kind of closure, such as delayed, late, or spontaneous closure. To distinguish from spontaneous closure of MH without any treatment, we use delayed closure to describe this specific type of spontaneous closure.

Herein, we report a MH secondary to Terson syndrome in which delayed closure was occurred within 6 months following vitrectomy. Meanwhile, we review literature about delayed closure of MH after vitrectomy to demonstrate the characteristics and possible mechanisms of the phenomenon of delayed closure.

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## 2. Case report

A 61-year-old man presented with vitreous hemorrhages in his right eye for 4 months. The patient developed a subarachnoid hemorrhage due to spontaneous rupture of his right vertebral artery dissecting aneurysm on February 5, 2017. As a result of the event, the patient experienced multiple neurological deficits including motor, language, and cognitive difficulties. The patient successively received external ventricular drainage, stent-assisted coiling embolization, and ventriculoperitoneal shunt in the neurosurgery center and recovered gradually, with some remaining mild language and motor deficits. Since he recovered consciousness 4 months later, the patient had been complaining of decreased vision in his right eye and visited our center on June 7, 2017. There was no history of diabetes, hypertension, or systemic vascular disease. There was no other ocular history of note. This study was approved by the Ethics Committee of the First Affiliated Hospital, College of Medicine, Zhejiang University, and was performed in accordance with the tenets set forth in the Declaration of Helsinki. The patient has agreed to publish this case and informed consent has been obtained.

At initial ophthalmological examination, slit-lamp biomicroscopy, intraocular pressure measurement, fundus examination, and ocular ultrasound were performed. The visual acuity was hand motion in the right eye and 20/30 in the left eye. The anterior segment examination was unremarkable except for the presence of anterior subcapsular cortical and nuclear lens opacities that were bilateral and symmetrical, intraocular pressure in both eyes was within normal limits. Fundus examination showed dense and diffuse vitreous hemorrhage in the right eye and unremarkable in left fundus. Ocular ultrasound showed vitreous hemorrhage in the right eye with no evidence of retinal detachment and posterior vitreous detachment. Terson syndrome was diagnosed according to his subarachnoid hemorrhage history and vitreous hemorrhages in the right eye.

One week later, the patient underwent 23G pars plana vitrectomy combined with phacoemulsification and intraocular lens implantation in his right eye. After the removal of the vitreous hemorrhage, a full-thickness MH and intact dome-shaped ILM were noted. Internal limiting membrane peeling and air tamponade were performed; expansile gas was not used due to the fact that expansile gases have not been obtainable in China in recent years. At 1 week after vitrectomy, the intravitreal air had been absorbed, and his best corrected visual acuity (BCVA) was 20/200 in the right eye, and optical coherence tomography (OCT) scan showed a persistent MH with obviously elevated retinal cuff and distinct intraretinal cysts (Fig. 1A). An additional surgery with air or silicon oil tamponade for the unclosed MH was proposed, but the patient refused the suggestion due to his physical status.

At 1 month postoperative follow-up, the BCVA improved to 30/200 in the right eye and OCT scan showed the MH became smaller with both base diameter and minimum diameter reduction and flattened with fewer intraretinal cysts compared with 1 week follow-up (Fig. 1B). Six months later, the hole had closed spontaneously and the BCVA improved to 20/40 (Fig. 1C).

## 3. Literature review

A systematic literature search was conducted in PubMed on articles in English using the following search terms: “Delayed closure or late closure or spontaneous closure” and “macular hole” and “vitrectomy”. After selections, only 7 articles<sup>[5-11]</sup> reported the phenomenon of MH delayed closure after

vitrectomy. We collected the information of each eye in these articles, which were shown in Table 1. Of these 8 eyes, the preoperative diagnosis included idiopathic MH (4 eyes), traumatic MH (2 eyes), and vitreomacular traction (2 eyes). There is no report about delayed closure of MH secondary to Terson syndrome. The times for these delayed closures occurred following vitrectomy were ranged from 1 to 28 months. The morphological characteristics of these unclosed MH were described in Table 1.

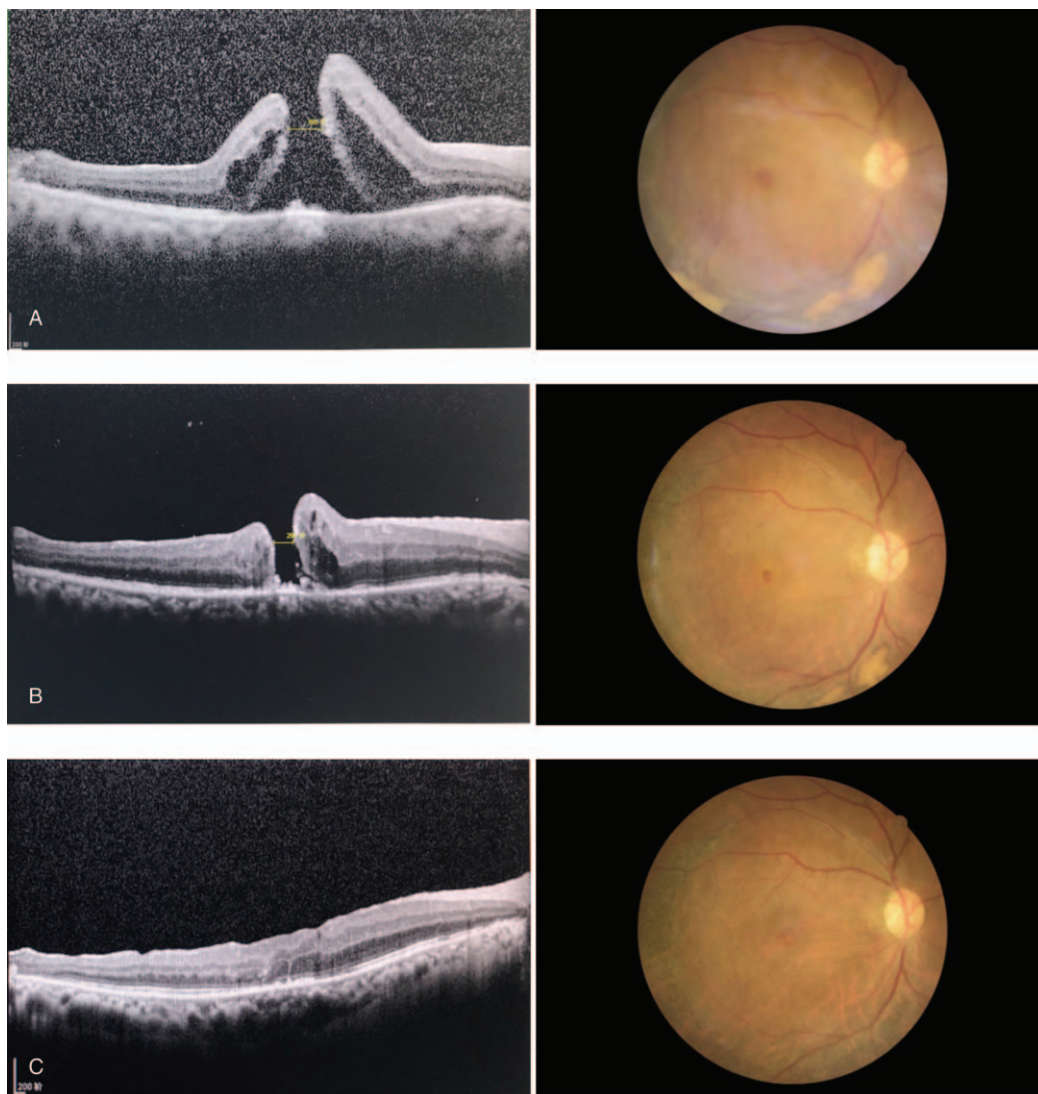
## 4. Discussion

Terson syndrome encompasses any intraocular hemorrhage associated with intracranial hemorrhage and elevated intracranial pressures. Intraocular hemorrhage following Terson syndrome is more commonly present in the sub-ILM and subretina. Macular hole as a rare complication of Terson syndrome has been reported occasionally.<sup>[3]</sup> Nevertheless, MH secondary to some similar intraocular hemorrhagic diseases, such as retinal arterial macroaneurysm, valsalva retinopathy, and postoperative hypotony, have also been reported.<sup>[12-14]</sup> Sagara et al<sup>[12]</sup> reviewed clinical records for 56 eyes with rupture of a retinal arterial macroaneurysm and found that macular holes occurred in 7 eyes (12.5%). Their findings suggest that the presence of subretinal and sub-ILM hemorrhages after the rupture of a retinal arterial macroaneurysm are risk factors for MH formation.<sup>[12]</sup>

In this case, the exact cause of the MH is unclear. Several mechanisms may involve in its formation. First, the presence of subretinal and sub-ILM hemorrhages in this case may elevate hydrodynamic pressure in the sub-ILM or subretinal space and weaken the fovea.<sup>[12]</sup> Second, because posterior vitreous detachment did not occur in this case, traction force on the fovea which was induced by contraction of the vitreous cortex may be a correlative factor for the MH formation. Third, premacular hemorrhage may create a tangential tractional force over the fovea to promote MH formation.<sup>[15]</sup> Last, the macular hole might be iatrogenic, caused by removing the vitreous hemorrhage. However, due to the intact ILM during surgery, the possibility of an iatrogenic macular hole is low.

Due to the patient's physical status, he was unable to maintain a prone head position in the postoperative period. The MH still persisted after the intravitreal air had been absorbed. Interestingly, however, delayed closure of the MH occurred 6 months after surgery. The delayed closure of persistent MH after PPV is quite a rare occurrence. Delayed closure has been reported to occur in idiopathic MH (4 eyes),<sup>[6-8,11]</sup> traumatic MH (2 eyes),<sup>[10,11]</sup> and vitreomacular traction (2 eyes).<sup>[5,9]</sup> To our knowledge, there are no papers describing the phenomenon of delayed closure occurring in the macular hole secondary to Terson syndrome.

According to our review of the delayed macular hole closure in 8 cases, it is found that there are 3 types of unclosed macular holes: type M, type U, and type W (Table 1). Type M macular holes were presented in 3 cases<sup>[5-7]</sup>, it is characterized by a wide basal diameter, which is much larger than the minimum diameter. The edge of the hole is obviously raised with a large number of intraretinal cysts, which may be related to the softness and elasticity of retina at the edge of the hole after ILM peeling. Such kinds of holes usually go through a process of flattening the hole edge and reducing the hole size to become a type U macular hole before being closed. The hole closure process in our case is completely consistent with these changes. On the other hand, type



**Figure 1.** Delayed closure of a macular hole secondary to Terson syndrome after vitrectomy. A, One week after surgery. OCT shows an MH surrounded by a fluid cuff with perifoveal cysts, the minimum diameter was 565  $\mu\text{m}$ . Fundus photographs show a full-thickness MH and subretinal hemorrhages inferior to the temporal-inferior arcade. B, One month after surgery. OCT shows that the MH became smaller with the minimum diameter reduced to 290  $\mu\text{m}$  and fewer intraretinal cysts. Fundus photographs show a smaller MH. C, Six months after surgery. OCT shows the disappearance of the MH and cysts, although a small interruption in the ellipsoid layer and the external limiting membrane can be appreciated. Fundus photographs show the MH was closed and subretinal hemorrhages were absorbed. MH = macular hole, OCT = optical coherence tomography.

U or type W macular hole is often smaller in size and no obvious morphological changes are observed before closure.

The exact mechanism of the delayed MH closure is still unclear. Chen et al<sup>[16]</sup> found that spontaneous closure of traumatic macular holes was associated with smaller minimum diameters of macular holes and the absence of intraretinal cysts. In this case, the initial postoperative state of MH is a type M with wide base diameter and a large number of intraretinal cysts. Nevertheless, the MH became smaller and less intraretinal cysts after type M change to type U. Type U was conformed to those characteristics. Based on the present case, we speculate that complete detachment of the posterior hyaloid in the vitrectomy and the ILM dome-shaped separation had fully released the tractional force around the macular hole. It was perhaps for this reason that 1 month after surgery, the edge of the MH reattached to the retinal pigment epithelium and the MH became smaller.

After MH become smaller, ILM peeling induces glial cell proliferation across the hole<sup>[17]</sup> and small macular defects, may allow easy migration of glial cells.<sup>[16]</sup> These mechanisms may help the delayed closure occurred in this case.

## 5. Conclusions

Terson syndrome combined with sub-ILM hemorrhage is not uncommon, but the complication of MH is still rarely reported. The exact formation mechanism of this kind of MH is not well understood and delayed closure of the MH in Terson syndrome after vitrectomy has never been reported. The dome-shaped ILM, which separated simultaneously during sub-ILM hemorrhage, may be a favorable factor for the spontaneous closure of the MH. The mechanism for delayed closure of long-term or chronic MH remains to be confirmed by further research. The significance of

**Table 1**  
Data of patients in the 7 articles.

Author*	Gender	Age	Diagnosis	ILM-peeling	Tamponade	Morphological transformation of persistent MH	Diameter of MH (um)	Duration before closure	VA after PPV	Final VA after closure
Rutul P <sup>[5]</sup>	Male	70	VMT	No	No	M U close	1414 (base)	28 m	FC	20/80
Christiane I <sup>[6]</sup>	Male	74	IMH	Yes	C2F6	M U close	2561 (base)	9 m	20/150	20/60
Peter D <sup>[7]</sup>	Male	73	IMH	Yes	C2F6	M close	NA	1 m	20/200	20/30
Filiz A <sup>[8]</sup>	Female	57	IMH	No	C3F8	U close	NA	5 m	NA	20/100
Dominik O <sup>[9]</sup>	Female	78	VMT Outer LMH	Yes	Air	U close	NA	1 m	20/250	20/40
Pukhraj R <sup>[10]</sup>	Male	42	TMH	Yes	C3F8	U close	NA	7 m	20/125	20/80
Pukhraj R <sup>[11]</sup>	Male	38	TMH	Yes	Gas	W close	164	7 m	20/400	20/200
	Female	67	IMH	Yes	Gas	W close	218	7 m	20/200	20/120



Type M (The hole edge is obviously raised with surrounding intraretinal cysts and the base diameter is much longer than the minimum diameter).



Type U (The hole edge attach to the underlying retinal pigment epithelial layer without hole edge thinning and the base diameter is similar to the minimum diameter).



Type W (The hole edge is thinned and attached to the underlying retinal pigment epithelial layer).

IMH=idiopathic macular hole, TMH=traumatic macular hole, VA=visual acuity, VMT=vitreomacular traction.

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this case is to suggest that similar patients should be monitored carefully by OCT and the additional surgery for the MH may be delayed, since delayed closure is possible.

### Author contributions

**Conceptualization:** Ye Shen.

**Data curation:** Wen Sun.

**Project administration:** Yan Sheng.

**Resources:** Ye Shen.

**Writing – review & editing:** Yan Sheng.

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