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Case report

Macular pucker formation after macular hole surgery with inverted internal limiting membrane flap technique and silicone oil tamponade

Akira Hirata [a,b,*], Akiomi Takano [c], Takahiro Kawaji [d], Kei-ichiro Nakamura [b]

[a] Hayashi Eye Hospital, Fukuoka, Japan
[b] Division of Microscopic and Developmental Anatomy, Department of Anatomy, Kurume University School of Medicine, Kurume, Japan
[c] Takano Eye Clinic, Kumamoto, Japan
[d] Sato Eye Clinic, Kumamoto, Japan

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ABSTRACT

Purpose: The inverted internal limiting membrane (ILM) technique was recently introduced for refractory macular hole. Here, we evaluate a case of macular pucker formation after macular hole surgery using the inverted ILM flap technique and silicone oil tamponade. After undergoing vitrectomy combined with ILM removal, the patient had a good visual prognosis.

Observations: A 49-year-old male with macular hole affecting both of his eyes underwent vitrectomies. Three months after the first surgery in his right eye, macular pucker formation was observed in the macula, which was associated with the ILM flap used to cover the macular hole. After peeling the ILM, the macula returned to a normal contour and visual acuity improved. Examination of the removed ILM revealed macrophage-like cells containing silicone oil particles that were responsible for the ILM contraction.

Conclusions and Importance: When using the inverted ILM flap technique and silicone oil, macular pucker may occur after macular hole surgery. Peeling of the ILM flap restored the macular shape and did not reopen the macular hole, thereby improving visual acuity. Thus, silicone oil should be used with caution when performing macular hole surgery with the ILM flap technique.

1. Introduction

Recently, inverting the internal limiting membrane (ILM) onto the macular hole for refractory macular hole repair has been reported, with this method considered to contribute to the improvement of the macular hole closure rate.[1] Since the introduction of the procedure, various inverted ILM techniques have been proposed and shown to be useful.[2–5] However, as far as the literature is searched, there are no reports on postoperative complications associated with the ILM flap.

We report a case of macular pucker due to contraction of the ILM flap that occurred following surgery in a case where silicone oil tamponade was used after the creation of an ILM flap that was placed over the macular hole. The subsequent peeling of the contracted ILM flap improved both the macular shape and visual acuity.

2. Case report

A 49-year-old male, who had been suffering from a visual disturbance in his left eye for at least three years and in his right eye for six months, was referred to Takano Eye Clinic for treatment of both eyes in February 2018. There were no apparent systemic or family histories. Best-corrected visual acuity (BCVA) was 20/100 in his right eye and counting fingers in his left eye. Slit lamp examination showed mild cataract in both eyes. A conventional fundus examination indicated a macular hole in his right eye and bullous retinal detachment in his left eye (Fig. 1A and B). Optical coherence tomography (OCT) showed stage 3 macular hole in his right eye and macular hole associated retinal detachment in his left eye (Fig. 1C and D). The axial length of his right eye was 22.12 mm and that of his left eye was 23.55 mm. After discussing the available treatment options, the patient decided to undergo surgeries in both eyes. After obtaining informed consent from the patient, a vitrectomy combined with cataract surgery in his left eye was performed on February 13, 2018. After phacoemulsification and aspiration, an intraocular lens was implanted. Subsequently, a pars plana vitrectomy using a 25-gauge trocar system was then performed. The retinal detachment extended to the equator due to the macular hole,

* Corresponding author. Hayashi Eye Hospital, 4Hakataekimae, Fukuoka, 812, Japan.
E-mail address: akhirata@gmail.com (A. Hirata).

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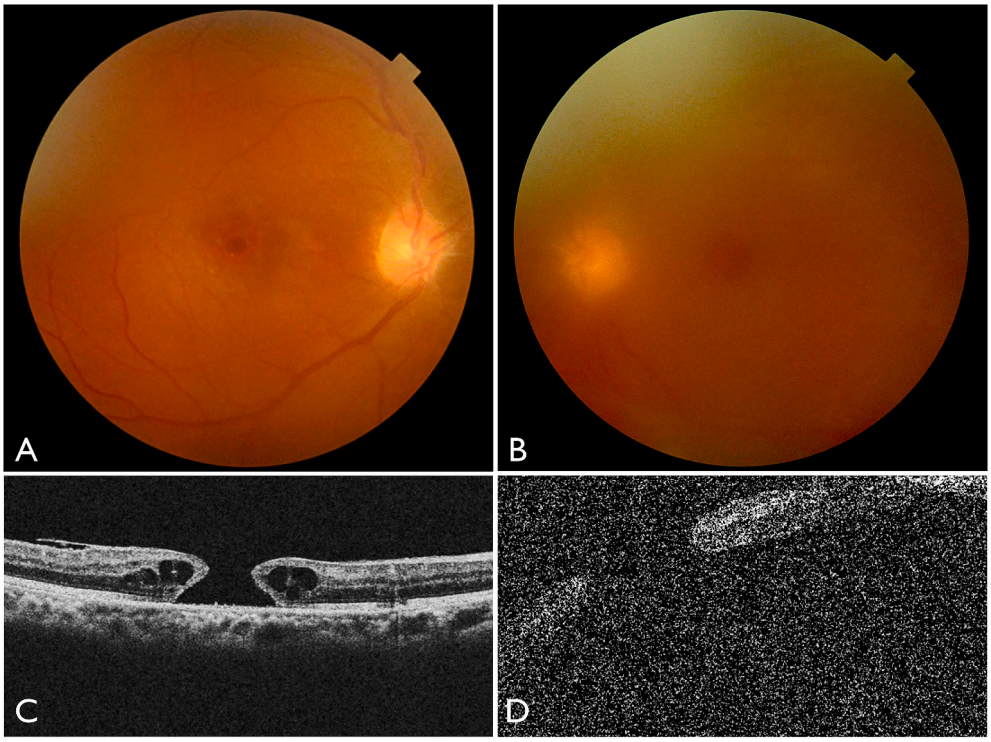

Fig. 1. Fundus and optical coherence tomography (OCT) findings obtained at the time of the first visit.
A, B: Fundus photographs at the first visit in the right (A) and left (B) eyes. A macular hole was noted in the right eye (A). Although difficult to observe due to cataract and vitreous opacity, retinal detachment was observed in the left eye (B). C, D: B-scan OCT findings for the first visit. A stage 3 macular hole was observed in the right eye (C). A macular hole was also found in the macula of the detached retina in the left eye (D).

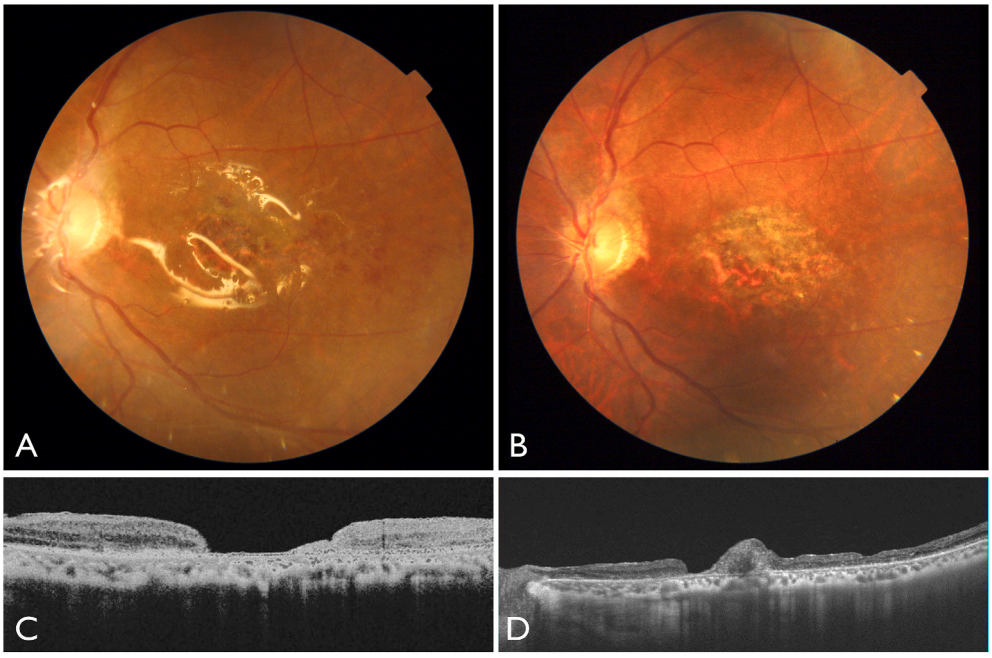

Fig. 2. Postoperative fundus and optical coherence tomography (OCT) findings in the left eye. A, B: Fundus findings after the second surgery with silicone oil (A). Fundus findings after silicone oil removal (B). The retina was reattached.
C, D: OCT findings after the second surgery with silicone oil (C). OCT findings after silicone oil removal (D). The macula was closed by gliosis.

with preretinal and subretinal proliferative membranes. Following core vitrectomy, peeling of the preretinal proliferative membranes, peeling of the ILM around the macular hole, fluid-air exchange, and air tamponade were performed. However, re-detachment of the retina occurred ten days after the surgery. Vitrectomy with additional removal of the proliferative membranes and injection of the silicone oil was performed on February 27, 2018. Silicone oil removal was conducted on October 23, 2018. The retina was reattached and appeared stable (Fig. 2). There was slight improvement of the visual acuity to 20/400.

For the right eye, we obtained informed consent from the patient to perform silicone oil injection at the time of the first surgery for the purpose of early social rehabilitation. On March 27, 2018, a vitrectomy combined with cataract surgery was performed in his right eye. Following the cataract surgery and core vitrectomy, peeling of the ILM was performed around the macular hole (Fig. 3A, C). A 1.5-disc diameter ILM flap was created in the temporal area and used to cover the macular

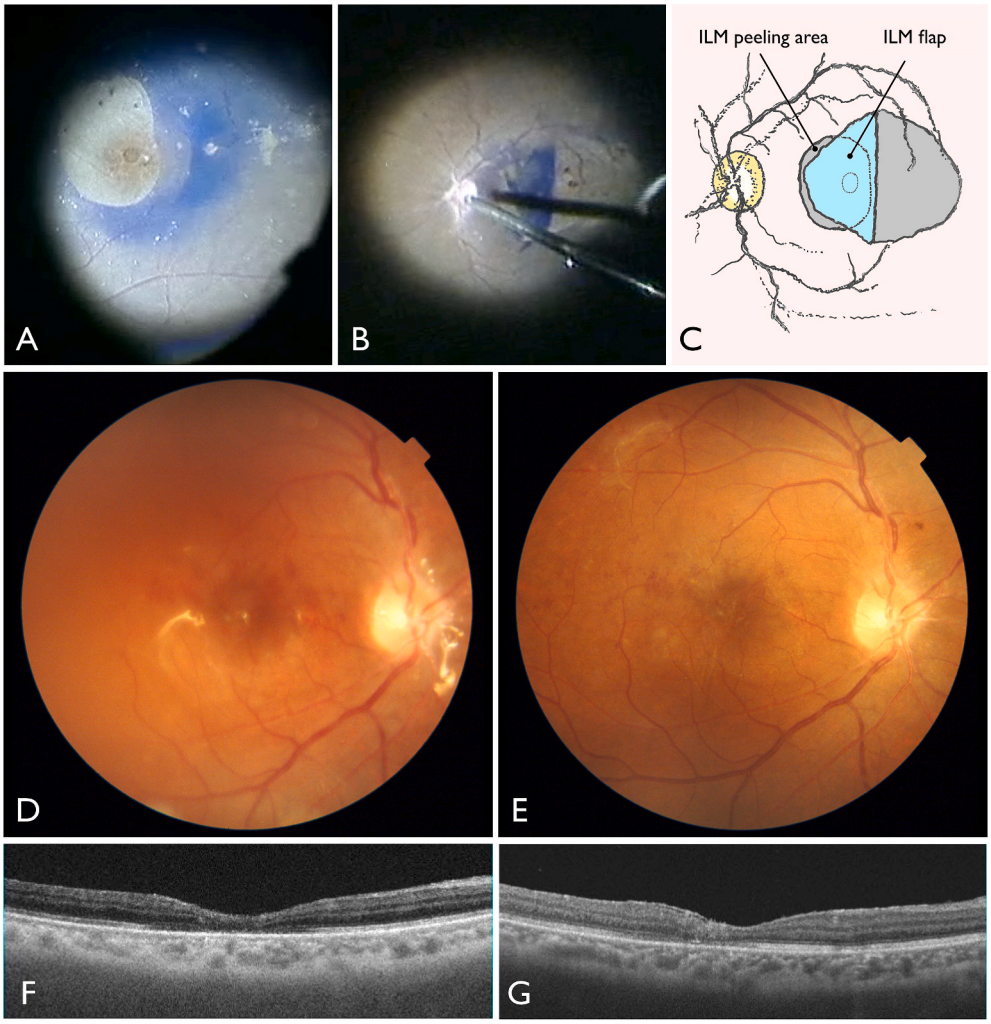

Fig. 3. Intraoperative and postoperative findings in the right eye. A, B: Intraoperative findings in the right eye. Internal limiting membrane (ILM) around the macular hole is peeled off (A) and the macular hole is covered with ILM flap of the temporal area (B). C: Schema of intraoperative findings. D, E: Fundus findings after the first surgery with silicone oil (D). Fundus findings after silicone oil removal (E). The macular hole was shown to be closed. F, G: OCT findings after the first surgery with silicone oil (F). OCT findings after silicone oil removal (G). The macular hole was closed and there was no abnormal foveal structure observed.

region as reported previously (Fig. 3B and C).[6] The surgery was completed by performing fluid-air exchange in the vitreous cavity and then replacing with silicone oil. The patient was instructed to maintain a prone position for three days. Two months after the surgery, the visual acuity in his right eye improved to 20/50 (Fig. 3D, F). Silicone oil removal was performed on May 22, 2018 (Fig. 3E, G). The visual acuity of his right eye on the day after the operation was 20/40, and there was also improvement in the macular morphology. However, one month after silicone oil removal, epiretinal membrane formation was observed in the macula, and OCT showed deformation over the entire retinal layer and an increase in the retinal thickness that subsequently resulted in a macular pucker (Fig. 4A, C). The epiretinal membrane of the macula coincided with the position of the ILM flap. One month later, the visual acuity gradually declined to 20/70. Therefore, the ILM flap was peeled on July 24, 2018. Although the ILM flap was firmly adhered to the retina, it was possible to detach it as a single piece. As there was no obvious reopening of the macular hole observed during the operation, the surgery was completed without having to use a tamponade substance. Postoperatively, the macular hole has remained closed, the foveal structure has improved, and there has been improvement in the visual acuity to 20/25 (Fig. 4B, D).

Light microscopy (LM) and focused ion beam/scanning electron microscopy (FIB/SEM) were used to further examine the extracted ILM as reported previously (Fig. 5).[7] Innumerable cells were found adhered to the surface of the ILM on the vitreous side after the ILM was inverted by surgery (Fig. 5A). Countless round vacuole-like structures were found in the cells (Fig. 5B). There was no cell attachment observed on the surface of the ILM that was newly facing the retina (Fig. 5A). Three-dimensional observation of the fine structure of a part of the extracted ILM by FIB/SEM revealed that cells with fine foot processes were attached to the ILM surface facing the vitreous body (Fig. 5C). These cells adhered to each other and formed puckers in the ILM (Fig. 5D). The round, vacuolar structure inside the cell was a sphere, which had been taken up by the cell (Fig. 5E). Pigment granules were also found in these cells.

3. Discussion

In the current case, the patient's left eye that had previously undergone surgery was shown to have an old macular hole retinal detachment, along with poor postoperative visual acuity. Therefore, silicone oil injection was performed at the time of the first surgery for the purpose of early social rehabilitation of his right eye.

Three months after the surgery in his right eye, macular pucker became apparent, and the ILM flap that had been used to close the macular hole was peeled off. In this case, peeling of the ILM flap restored the macular shape, it did not reopen the macular hole, and thus, the visual acuity improved. To the best of our knowledge, there have been no previous reports that have documented the contraction of the inverted ILM flap.

The ILM is a basement membrane-like membrane that is originally located in the innermost layer of the retina. This membrane is known to thicken and harden with age. It is believed that the preparation of the ILM flap stimulates the migration and proliferation of glial cells in the

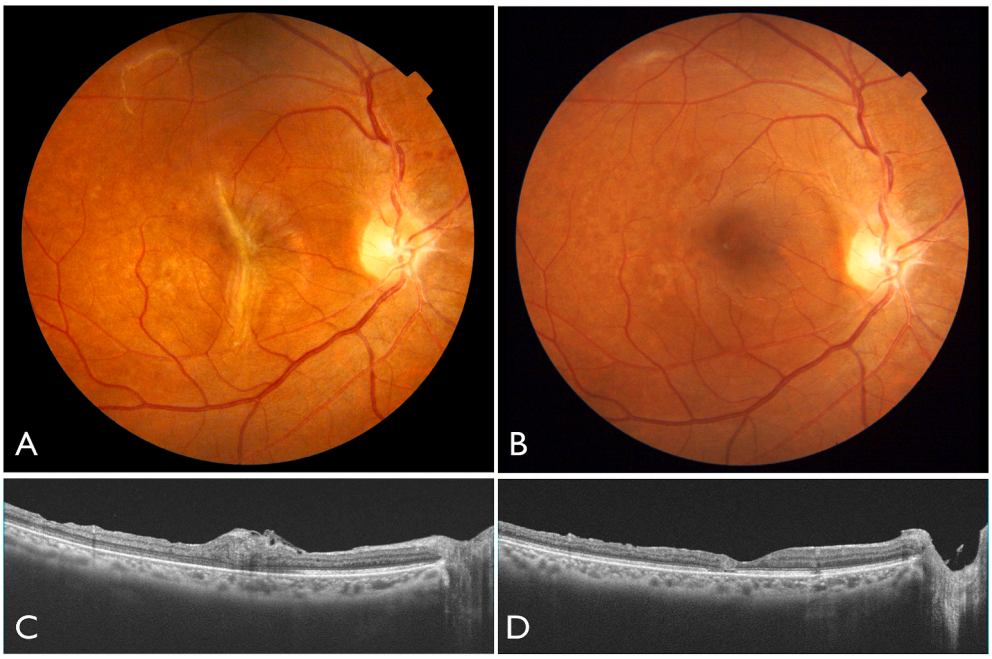

Fig. 4. Fundus and optical coherence tomography (OCT) findings in the right eye before and after the third surgery. A, C: Fundus and OCT findings in the right eye at one month after the second surgery. Formation of macular pucker was observed (A), and the macular region was thickened (C). B, D: Fundus and OCT findings in the right eye after the third surgery. Macular area was smooth (B) and the macular hole remained closed (D).

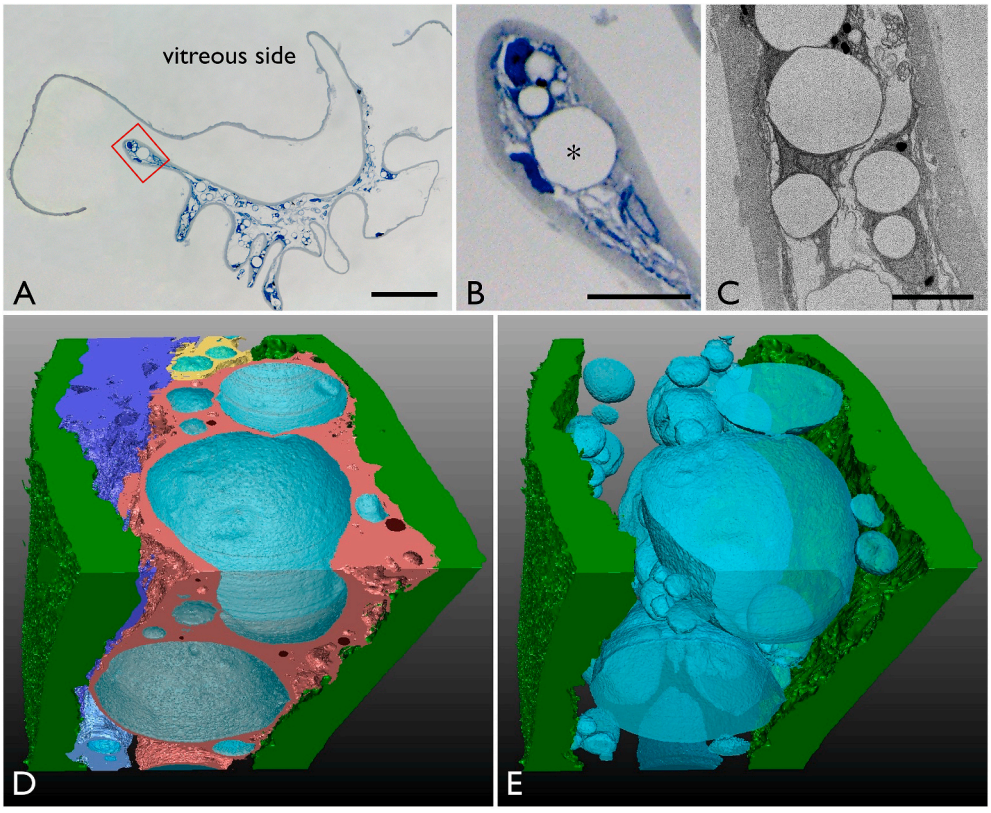

Fig. 5. Histological findings for the removed membrane. A, B: Light micrographs of the membrane. On the vitreous-side surface of the internal limiting membrane (ILM), contraction by numerous cells and proliferating tissues were observed (A). Magnification showed that there were round vacuoles (asterisk) found in the cells on the surface of the ILM (B). Bars = 50 μm for A and 20 μm for B. C–E: Focused ion beam/scanning electron microscopy findings. C: In the sectional image of the specimen, cells attached to the ILM showed numerous foot processes and intracellular vacuoles. Pigment granules were also seen. Bar = 10 μm. D, E: In the three-dimensional reconstructed membrane preparation, cells were in close contact with each other (D). When the cell components were made transparent, which allowed for the three-dimensional structure of the vacuoles to be examined, it was clear that they had a spherical shape (E). Green indicates the ILM, while pink, blue, white and yellow indicate cells attached to the ILM. Light blue, indicates the intracellular vacuoles. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

retina. Thus, this is expected to lead to changes in the morphology of the retina after the surgery. FIB/SEM analysis of the removed ILM specimen in this patient showed that the cells attached to the ILM were mainly on the surface that was newly facing the vitreous side. These cells were not found to be associated with the morphological changes observed on the retinal side.

The use of silicone oil as a tamponade substance for the treatment of macular holes is reported to be equivalent to gas tamponade in terms of anatomical and visual function improvements, and with regard to complication frequency.[8] The use of silicone oil in our current case was

performed in order to enable early social rehabilitation. However, injections of silicone oil can potentially cause pre-ILM cellular proliferation on the vitreous side. A previous study reported that macrophages incorporating emulsified silicone oil were seen in silicone oil tamponade cases in which epiretinal membrane formed after retinal detachment surgery.[9] Similarly, in the current case, we found that the cells attached to the excised ILM accumulated numerous spherical vacuoles, which were considered to be the presence of silicone oil inside of the cells. Moreover, multiple foot processes were observed for these cells. Although immunostaining was not performed in the current case, we assumed that these were macrophage-like cells. Wickham et al. reported that the use of silicone oil was accompanied by an inflammatory response mediated primarily by bloodborne macrophages.[10] This reaction was observed within a month of silicone oil injection and continued after removing the oil. In this case, it was considered that the preparation of ILM flaps, which could be a scaffold for cell proliferation, and postoperative inflammation with silicone oil, had a significant role in the formation of proliferative tissues on the flap.

4. Conclusions

In conclusion, the use of silicone oil tamponade during macular hole surgery can cause the ILM flap designed to cover the macular hole to contract and subsequently form macular puckers. However, it is unlikely that once the macular hole is closed it will reopen, even if the ILM flap is removed. Thus, silicone oil should be used with caution when using an ILM flap during macular hole surgery.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and the use of any accompanying images.

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Authorship

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

Declaration of competing interest

The following authors have no financial disclosures: AH, AT, TK, KN.

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References

1. Michalewska Z, Michalewski J, Adelman RA, Nawrocki J. Inverted internal limiting membrane flap technique for large macular holes. *Ophthalmology*. 2010;117(10):2018–2025.
2. Aurora A, Seth A, Sanduja N. Cabbage leaf inverted flap ILM peeling for macular hole: a novel technique. *Ophthalmic Surg Lasers Imag Retina*. 2017;48(10):830–832.
3. Chen SN, Hsieh YT, Yang CM. Multiple free internal limiting membrane flap insertion in the treatment of macular hole-associated retinal detachment in high myopia. *Ophthalmologica*. 2018;240(3):143–149.
4. Chen SN, Yang CM. Double internal limiting membrane insertion for macular hole-associated retinal detachment. *J Ophthalmol*. 2017;2017:3236516.
5. Michalewska Z, Michalewski J, Dulczewska-Cichecka K, Adelman RA, Nawrocki J. Temporal inverted internal limiting membrane flap technique versus classic inverted internal limiting membrane flap technique: a comparative study. *Retina*. 2015;35(9):1844–1850.
6. Hirata A, Hayashi K, Murata K, Nakamura KI. Removal of choroidal neovascular membrane in a case of macular hole after anti-VEGF therapy for age-related macular degeneration. *Am J Ophthalmol Case Rep*. 2018;9:14–17.
7. Hirata A, Murata K, Hayashi K, Nakamura KI. Three-dimensional analysis of peeled internal limiting membrane using focused ion beam/scanning electron microscopy. *Transl Vis Sci Technol*. 2018;7(1):15.
8. Ivanovska-Adjievska B, Boskurt S, Semiz F, Yuzer H, Dimovska-Jordanova V. Treatment of idiopathic macular hole with silicone oil tamponade. *Clin Ophthalmol*. 2012;6:1449–1454.
9. Tanaka Y, Toyoda F, Shimmura-Tomita M, et al. Clinicopathological features of epiretinal membranes in eyes filled with silicone oil. *Clinc Ophthalmol*. 2018;12:1949–1957.
10. Wickham LJ, Asaria RH, Alexander R, Luthert P, Charteris DG. Immunopathology of intraocular silicone oil: retina and epiretinal membranes. *Br J Ophthalmol*. 2007;91(2):258–262.