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## Case Report

# Autologous Transplantation of the Internal Limiting Membrane for Refractory Macular Hole following Ruptured Retinal Arterial Macroaneurysm: A Case Report

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## Keywords

Macular hole · Inverted internal limiting membrane flap technique · Retinal arterial macroaneurysm · Autologous transplantation of the internal limiting membrane · Refractory macular hole

## Abstract

**Purpose:** To report a case of macular hole (MH) secondary to a retinal arterial macroaneurysm (RAMA) which was successfully treated with an autologous transplantation of internal limiting membrane (ILM). **Case Report:** An 87-year-old female presented with a sudden decrease in central vision in the right eye. A fundus examination revealed a RAMA in the superonasal macular region, a subretinal hemorrhage (SRH), involving the macula, and a sub-ILM hemorrhage. A pars plana vitrectomy (PPV) was performed. Intraoperatively, an MH filled with coagulum was detected. We tried to blow off and drain the SRH with a current of BSS and a gentle suction with a 27-gauge vitreous cutter from the MH, but some amount of SRH at the bottom of the MH remained. The ILM was peeled off for 2 disc diameters around the MH. The vitreous cavity was filled with air at the end of the operation. Two weeks after the surgery, the MH was not closed. One month following the initial PPV, a second PPV was per-

formed to achieve closure of the MH. **Results:** An autologous transplantation of ILM was performed as second PPV. Six months after the final surgery, the MH was successfully closed and the best-corrected decimal visual acuity was 0.6. **Conclusions:** Autologous ILM transplantation can be an effective treatment option for MH closure following RAMA rupture.

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## Introduction

The onset of a macular hole (MH) after the rupture of a retinal arterial macroaneurysm (RAMA) is a sight-threatening complication of RAMA [1–3]. Previous reports have suggested that MH can develop in 5.9–12.5% of RAMA ruptures, and that the postoperative primary anatomic success rate of surgical intervention for MH secondary to RAMA is between 57.1 and 75.0% [4, 5], which is lower than that of surgical intervention for idiopathic MH [6, 7]. Moreover, there is limited information with respect to the most effective surgical procedure for MH after RAMA rupture. Several studies have reported the efficacy of the inverted internal limiting membrane (ILM) flap technique [8–11] and autologous transplantation of ILM [12–15] for treating refractory MHs. In this case report, we have described the application of autologous transplantation of ILM for treatment-resistant MH secondary to RAMA rupture.

## Case Report

An 87-year-old female presented with a 2-day history of a sudden decrease in central vision in the right eye (OD). Her best-corrected decimal visual acuity (BCVA) was 0.01 OD at the initial visit. The slit-lamp examination of the anterior segment revealed a normal OD. The lens status was pseudophakic OD. A fundus examination revealed a RAMA in the superonasal macular region, a subretinal hemorrhage (SRH) approximately 4 disc diameters in size, involving the macula, and a sub-ILM hemorrhage in the inferior macular region with a niveau formation (Fig. 1a). Preoperative optical coherence tomography (OCT) OD images depicted a swollen retina due to bleeding from the ruptured RAMA, but it was difficult to visualize the retina in detail because of the signal reduction due to SRH (Fig. 1b). The findings in the left eye were unremarkable. One week after the onset of symptoms, a 27-gauge pars plana vitrectomy (PPV) was performed with the written informed consent of the patient. Intraoperatively, an MH filled with coagulum was detected after the removal of a sub-ILM hemorrhage by gentle active suction using a 27-gauge vitreous cutter without touching the macular area. It is possible that the MH occurred preoperatively and was not derived from inadvertent aspiration of retinal tissue. We tried to blow the SRH off with a current of BSS, and to drain using a gentle suction with a 27-gauge vitreous cutter from the MH, but some amount of SRH remained at the bottom of the MH. The ILM was peeled off for 2 disc diameters around the MH. The inverted ILM flap technique was not performed because the remaining SRH at the bottom of the MH may provoke foveal photoreceptor damage when the MH was closed. The vitreous cavity was filled with air at the end of the operation. Patients are asked to remain face down for 1 week after the surgery, as much as possible. Two weeks after the surgery, a subfoveal hemorrhage remained (Fig. 2a), and the MH was not closed (Fig. 2b). One month following the initial PPV and after the SRH at the bottom of the MH was absorbed, a second PPV was performed to achieve closure of the MH. The detailed method of

the second PPV is described below. Six months after the final surgery, the MH remained closed (Fig. 3a, b) and the BCVA was 0.6 OD.

### PPV Method: Second Attempt

Local anesthesia was administered using sub-Tenon's injection of 2% lidocaine. A 3-port system for a 27-gauge PPV was placed 4 mm posterior to the limbus. First, residual ILM was stained with indocyanine green (ICG) (0.025%). Excessive ICG was immediately removed by active suction using a 27-gauge vitreous cutter. We created an appropriately sized, free ILM flap, which was larger than that of the MH, from the temporal-upper side of the macula, but the ILM was not removed completely from the retina. Then, we covered the MH with Viscoat (Alcon Laboratories, Fort Worth, TX, USA) as an adhesive, removed the ILM flap completely from the retina, delivered the free ILM flap on the fovea to cover the whole MH, and gently massaged it flat. Finally, supplementary Viscoat was applied on top of the free ILM flap as a ballast, as reported previously [16]. The vitreous cavity was filled with air at the end of the operation. As mentioned above, patients are asked to remain face down for the week following the surgery as much as possible.

### Discussion

As previously reported, MH can develop in 5.9–12.5% of RAMA ruptures [4, 5], and the primary anatomic success rate of surgical intervention for MH secondary to RAMA is lower than that of idiopathic MH [4–7]. The formation of MH following a ruptured RAMA is not entirely uncommon; thus, it is very important to discuss the most effective surgical methods for the treatment of MH secondary to RAMA.

Although the numbers of reports regarding the effectiveness of the inverted ILM flap technique and autologous ILM transplantation for refractory MHs are increasing [8–15], to the best of our knowledge, this is the first case report of MH secondary to RAMA rupture that was successfully closed by autologous ILM transplantation. In this case, during the first surgery, we recognized the existence of an MH intraoperatively, but we did not perform an inverted ILM flap technique simultaneously, because there was significant concern about the potential for macular toxicity from the remaining subfoveal hemorrhage. Imai and Azumi [17] previously reported that some quantity of ICG, which was left at the bottom of an MH and enclosed by the overlying ICG-stained ILM flap, might have provoked retinal pigment epithelium damage after the inverted ILM flap technique. Similarly, in our case, it was possible that the enclosed subfoveal hemorrhage might have induced damage of the foveal photoreceptors, which may have been in contact with the subfoveal hemorrhage when the MH was closed. These are the reasons why we performed ILM peeling and air tamponade first, then at a later date performed the second surgery after the absorption of the SRH at the bottom of the MH. We believe that these decisions contributed to both anatomical and functional successes. On the other hand, it might be one of the effective treatment options to try to remove SRH aggressively by using adjuvant therapy such as tissue plasminogen activator [18] and to perform the inverted ILM flap technique simultaneously during the initial operation. The results of large-scale, randomized, comparative, and prospective studies in the future may be warranted.

The most difficult part of the inverted ILM flap technique and autologous ILM transplantation is fixing the flap in position during fluid-air exchange [8, 9], and several methods have been reported to make that easier [16, 19]. Recently, Song et al. [16] reported that the Viscoat-assisted single-layered inverted ILM flap technique can stabilize the inverted ILM flap, which tends to turn over and move away from the opening of the MH. In this case, we applied this method to autologous ILM transplantation and achieved good results. This Viscoat-assisted method may be useful in increasing the success rate of the initial surgery even after autologous ILM transplantation for the treatment of refractory MHs including MH secondary to RAMA rupture.

In conclusion, our report may suggest that autologous ILM transplantation can be an effective treatment option for MH closure following RAMA rupture.

### Statement of Ethics

The authors have no ethical conflicts to disclose.

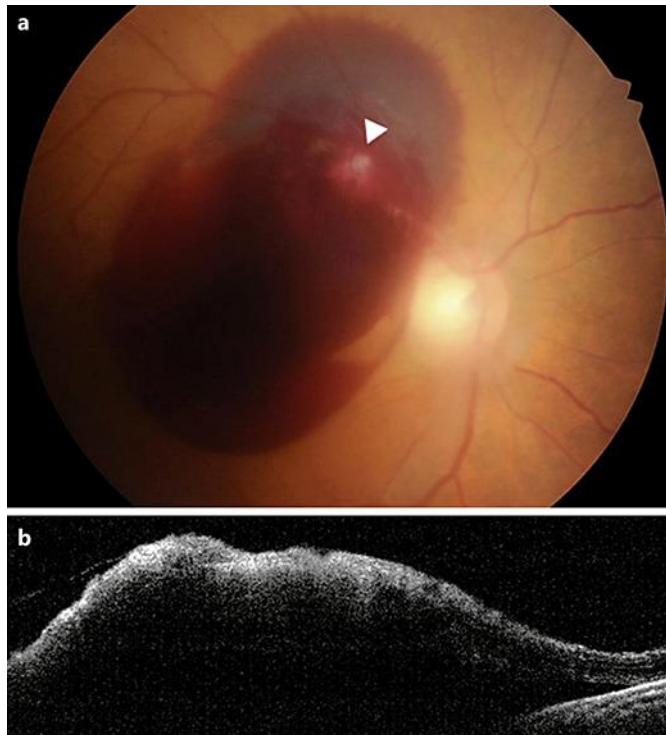
### Disclosure Statement

There are no conflicts of interest.

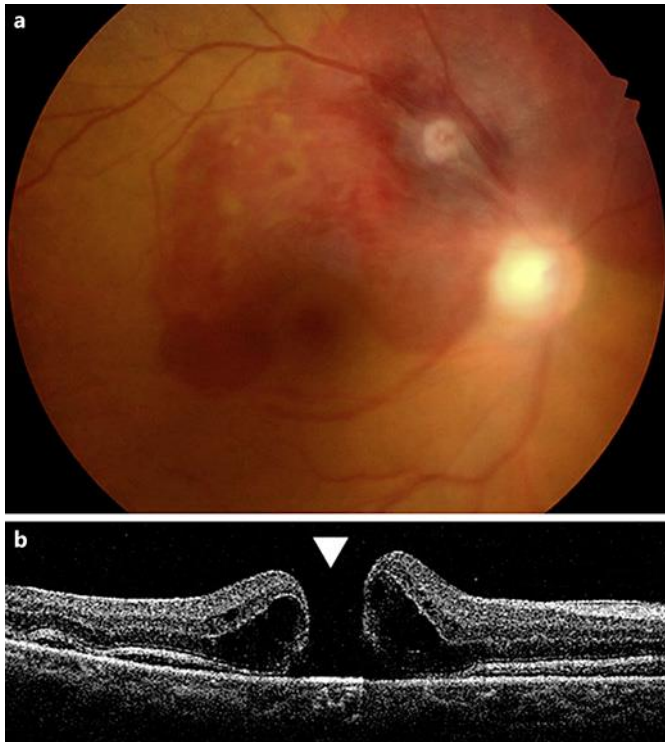
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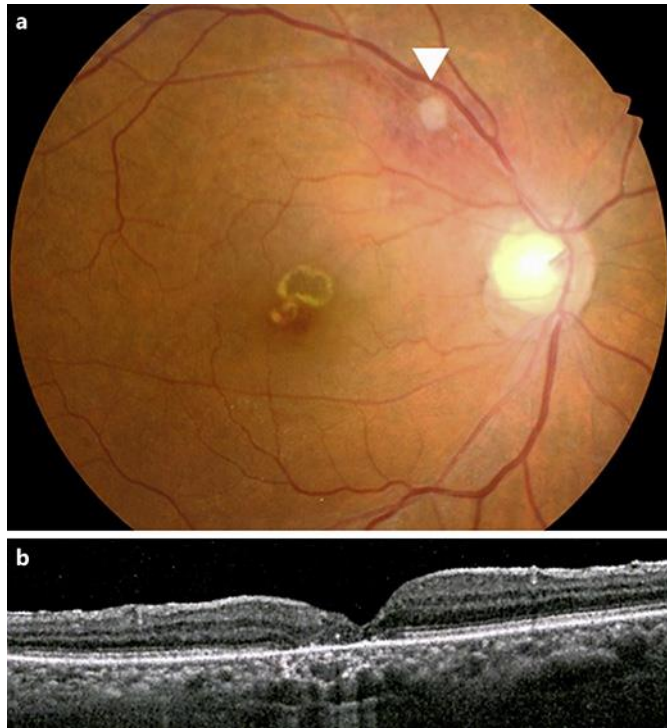
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**Fig. 1.** **a** Fundus photograph at the initial visit. Note a ruptured retinal arterial macroaneurysm (RAMA) in the superonasal macular region (white arrowhead), a subretinal hemorrhage involving the macula of approximately 4 disc diameters in size, and sub-internal limiting membrane hemorrhage in the inferior macular region with a niveau formation. **b** Optical coherence tomography finding at the initial visit. Note a swollen retina due to bleeding from the ruptured RAMA, but it was difficult to visualize the retina in detail because of the signal reduction due to SRH.



**Fig. 2.** **a** Fundus photograph 2 weeks after the first surgery. Subfoveal hemorrhage still remained. **b** OCT finding 2 weeks after the first surgery. MH was not closed (white arrowhead).



**Fig. 3.** **a** Fundus photograph 6 months after the last surgery. RAMA was organized (white arrowhead) and subfoveal hemorrhage was absorbed. **b** OCT finding 6 months after the last surgery. MH was successfully closed.