

# Severe phimosis-like epiretinal membrane proliferation following internal limiting membrane peeling for macular hole

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**Dear Editor,**

**I**nternal limiting membrane (ILM) peeling with gas tamponade has been the preferred surgical technique for the macular hole treatment<sup>[1-2]</sup>. Eckardt *et al*<sup>[1]</sup> achieved a complete closure of 92% of the macular holes. Generally, the procedure involves peeling of ILM from the macula using a microvitrectomy blade with an intraocular forceps 500 mm superior to the hole, removal of membranes and releasing the traction around the macular hole. If a macular epiretinal membrane (ERM) is present, it should be peeled during the surgery<sup>[1-2]</sup>.

The size of maculorrhesis has been still a debate; Eckardt *et al*<sup>[1]</sup> adopted a 3 to 4-disc diameter area, while Park *et al*<sup>[2]</sup> adopted a smaller 2-disc diameter maculorrhesis. Yao *et al*<sup>[3]</sup> showed better visual and anatomical results with the 4-disc diameter peeling group compared to the 2-disc diameter group in macular holes less than 500 microns. The size of ILM peeled area was found to be a predictor in anatomical success of macular hole surgery<sup>[4]</sup>.

ILM plays an important role as a scaffold for cellular proliferation and ERM formation<sup>[5]</sup>. Accordingly, removal of the ILM is associated with lower recurrence of ERM<sup>[2]</sup>. Also, silicone oil enhances cellular and membranes proliferation<sup>[6]</sup>.

We present a case of ERM proliferation following ILM peeling with a small maculorrhesis for a previous macular hole repair tamponade with silicone oil. ERM has a characteristic of phimosis-like pattern using the edge of previous maculorrhesis as a scaffold and sparing the central peeled area from attachment.

**Ethical Approval** Written informed consent was obtained from the patient for publication of this case report and any accompanying images. This case study was conducted in accordance with the Declaration of Helsinki.

## CASE REPORT

A 62-year-old male, not known to have any medical illness, underwent combined surgery of cataract extraction and pars plana vitrectomy with ILM peeling for macular hole in his left eye. Silicone oil was used as a tamponade due to the presence of multiple retinal breaks as stated by the medical records. The patient presented to our center one year later with significant loss of central vision in the same eye. On examination, the best corrected visual acuity was 0.3 in the right eye and counting fingers at 3 m in the left eye. Anterior segment examination revealed no abnormality apart from nuclear sclerosis cataract in his right eye and posterior chamber intraocular lens in the left eye. The intraocular pressure was within normal range in both eyes. Fundus examination of the left eye revealed central dense glial tissue covering the fovea with 360-degree laser marks in the far periphery in a silicone-filled eye. Both optic nerves were normal. Optical coherence tomography (OCT) was performed and showed dense epiretinal membrane with distorted macular morphology (Figure 1A).

The decision was to remove the silicone oil and to peel the ERM of his left eye. Using Lumera 700 microscope with Resight system for visualization (Zeiss), 23-gauge pars plana silicone oil removal was performed (Stellaris PC system by Bausch & Lomb) followed by serial fluid-air exchange to ensure maximum silicone oil removal. Under fluid filled eye, the phimosis-like dense glial tissue (Figure 2A) was removed using 23-gauge end grasping forceps.

During removal, a strong perifoveal attachment of the glial tissue sparing around one-disc diameter of the foveal center was noticed along with a closed macular hole (Figure 2B). Following removal, ILM staining was performed using

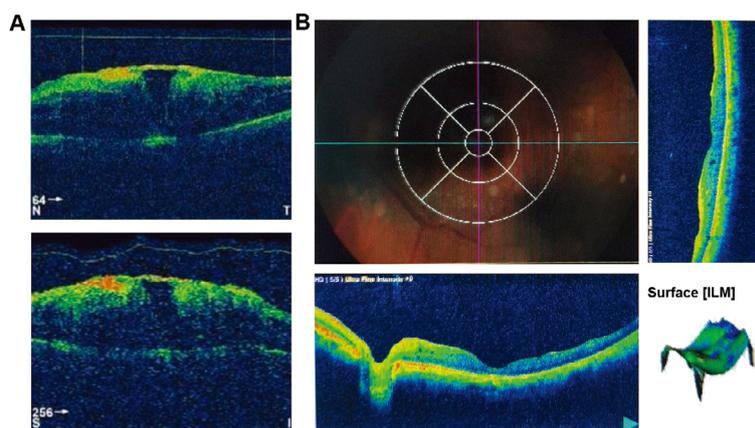


Figure 1 Optical coherence tomography of the macula indicating severe epiretinal membrane (A) and after the peeling of the epiretinal membrane (B).



Figure 2 Intraoperative images indicating the phimosi-like ERM proliferation (A, black arrow) with circumferential attachment for the previously ILM peeled area (B, black arrow). After the application of brilliant blue, the central foveal area had no attachment for the ERM proliferation (C, red arrows) ILM: Internal limiting membrane; ERM: Macular epiretinal membrane.

Brilliant Blue G stain in the air-filled eye. All the macular area took the stain except the central area (around one-disc diameter) that had no attachment with the peeled glial tissue, corresponding to the previously ILM peeled area in the primary surgery (Figure 2C; Video 1, online supplementary). Maculorrhexis was then enlarged, and the eye filled with 20% SF<sub>6</sub> gas.

The patient was given prednisone acetate 1%, and moxifloxacin 0.3% drops 6 times daily for one month and advised for face down positioning in the first 5d. Best corrected visual acuity continued to improve over the next year reaching 0.2. Postoperative OCT showed improving in macular morphology with no residual membrane (Figure 1B).

## DISCUSSION

To the best of our knowledge, we report the first case of dense phimosi-like ERM proliferation following ILM peeling for macular hole in silicone oil-tamponade eye. Uemoto *et al*<sup>[7]</sup> reported two cases of faint ERM proliferation following ILM removal for idiopathic macular hole. In the first case, a thin ERM developed nasally to the macula area where the ILM had been peeled. The patients complained only of metamorphopsia. In the second case, the macular hole was sealed but an ERM developed in the macular region; however, after two years of the operation, the visual acuity was still 1.0<sup>[7]</sup>. In this case, the patient complained of a decrease of vision one year following the primary ILM peeling with small maculorrhexis, and a dense phimosi-like ERM was disclosed which required surgical removal.

Removal of ILM is not risk free and can carry complications. Most of the complications related to vitrectomy, like cataract progression, retinal detachment, retinal tears, high intraocular pressure, hypotony, macular toxicity, dislocation of intraocular lens in pseudophakic eyes and endophthalmitis<sup>[8]</sup>. However, complication related to ILM peeling can be listed as focal retinal hemorrhages and edema, paracentral scotomas, and visual field defects, dissociated optic nerve fiber layer and reduction of retinal sensitivity associated with relative or absolute microscotomas<sup>[8]</sup>. Liu *et al*<sup>[9]</sup> studied risk factors for glial tissue proliferation following ILM flap technique for macular hole. They found that patients with larger minimum linear diameter of the macular hole have a greater chance to develop secondary glial proliferation. Also, the greater the defect of external limiting membrane and ellipsoid zone, the greater the chance of glial proliferation<sup>[9]</sup>. Another factor may contribute for the formation of ERM in our patient is the presence of silicone oil. Both fibroblast growth factor and interleukin 6 have the potential to contribute to the formation of ERM and the high concentration of these growth factors found in the silicone oil fluid points to the role of silicone oil in ERM formation<sup>[10]</sup>.

Recent *in vivo* studies suggest that glial tissues are essential for macular hole healing. However, severe gliosis may indicate a worse visual prognosis, and the disappearance of glial proliferation in the early postoperative period predicted better visual recovery<sup>[11]</sup>.

In this case, the primary maculorrhexis was relatively small according to the evidence from staining pattern with Brilliant

Blue G after removal of the glial tissue, along with the almost exact attachment of the ERM to the edge of the ILM and sparing the previously peeled area.

Tissue proliferation in our case is most likely a proliferative vitreoretinopathy process, given the fact that the patient had multiple retinal breaks in his primary surgery where the surgeon had to do 360-degree laser and use silicone oil as a tamponade. Regardless of the source of the glial tissue, we believe that ERM formed an attachment with ILM bridging the small maculorrhexis, contracted both tangentially and circumferentially causing this characteristic pattern.

This circumferential growth of the ERM around the peeled area gives it a phimosis-like picture, resembling the capsular phimosis complicating small capsulorrhexis in cataract surgery. To the best of our knowledge, macular phimosis as a term was not mentioned or described previously in literature. This case highlights the role of ILM in the proliferation of glial tissue, as well as the importance of large maculorrhexis not only to achieve a better success in macular hole closure, but also to avoid similar postoperative complication.

#### ACKNOWLEDGEMENTS

**Conflicts of Interest:** Mohidat H, None; El-Mulki O, None; Aleshawi A, None.

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