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# Serous retinal detachment accompanied by pachychoroid in hypotony maculopathy after trabeculectomy for diabetic neovascular glaucoma



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ARTICLE INFO	A B S T R A C T
<i>Keywords:</i> Vascular endothelial growth factor Bevacizumab OCT Blood-retinal barrier	Purpose: Two diabetic case reports of serous retinal detachment (SRD) accompanied by pachychoroid in hypotony maculopathy after trabeculectomy for neovascular glaucoma (NVG). Observations: Case 1: A 66-year-old female with stage 3 NVG and decreased vision acuity in the left eye. After trabeculectomy, postoperative laser suture lysis (LSL) resulted in development of hypotony maculopathy, followed by pachychoroid and SRD. Injection of C3F8 gas in the anterior chamber was unsuccessful and trans- conjunctival scleral re-suturing was performed. Intraocular pressure (IOP) consequently increased and SRD
	Trabeculectomy was uneventful, but postoperative LSL also resulted in development of hypotony maculopathy

re-suturing was performed. IOP consequently increased and SRD improved. *Conclusions:* SRD accompanied by pachychoroid was observed in hypotony maculopathy in diabetic cases. VEGF-independent exudative change in hypotony maculopathy may be due to hydrostatic pressure elevation in choroidal blood vessels based on Starling's hypothesis with the consequent breakdown of retinal pigment epithelium barrier in diabetic patients.

followed by pachychroid and SRD. Intravitreal bevacizumab injection had no effect and transconjunctival flap

## 1. Introduction

Hypotony maculopathy was first regarded as a visual acuity loss associated with choroid wrinkle folds.<sup>1</sup> While various causes such as trauma and uveitis exist, most reported cases occur secondarily after trabeculectomy and develops with a probability of 3-18%. This risk is increased with mitomycin C (MMC) combination.<sup>2</sup> The maculopathy is characterized by hypotony associated with fundus abnormalities including hypotony maculopathy, choroidal effusion, papilledema and vascular tortuosity, resulting in permanent visual acuity reduction.<sup>1,3</sup> A case report by Kokame et al. demonstrated that cystoid macular edema (CME) and serous retinal detachment (SRD) also occurred in hypotony maculopathy although this is uncommon.<sup>4</sup> These macular disorders are also reported to cause visual loss in various diseases including diabetic macular edema which is related to vascular hyper permeability.<sup>5</sup> However, the pathogenesis of hypotony-related SRD is still unknown. Here, we report two cases who developed SRD accompanied by pachychoroid from hypotony maculopathy after trabeculectomy for diabetic neovascular glaucoma (NVG).

#### 1.1. Case report

Case 1: A 66-year old woman with type 2 diabetes (HbA1c 10%) showed rubeosis iridis in her left eye. Her best-corrected visual acuity (BCVA) was 20/200 in the left eye. The intraocular pressure (IOP) of her left eye was 52 mmHg. Slit-lamp examination showed hyphema, rubeosis iridis and peripheral anterior synechiae (PAS; PAS index was 0.5). Fundus examination of the left eye revealed papilledema. Optical coherence tomography (OCT) showed normal choroidal thickness (263 µm) (Fig. 1A). She was diagnosed as NVG stage 3. As anti-VEGF injection was ineffective in reducing IOP, she underwent pars plana vitrectomy with endophotocoagulation and pars plana filtering. Although the IOP initially decreased, it started increasing again three months after surgery. Therefore, trabeculectomy with MMC was performed. Although there was no intraoperative complication, hypotony maculopathy (8 mmHg) developed after laser suture lysis (LSL). On postoperative day (POD) 9, OCT showed choroidal wrinkles and increased choroidal thickness (448 µm) (Fig. 1B). Furthermore, SRD and a persistent thicker choroid (489 µm) could be observed on POD 28, and

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Fig. 1. Optical coherence tomography (OCT) image before and after trabeculectomy in Case 1 (66-year old woman). (A) OCT image before surgery shows no apparent abnormal findings. (B) OCT image shows choroidal wrinkles and pachychoroid on postoperative day (POD) 9. (C) OCT image shows serous retinal detachment (SRD) on POD 28. (D) SRD resolved upon resolution of hypotony on POD 42. The choroidal thickness on POD 9, 28, and 42 was 448, 489 and 362  $\mu$ m, respectively. Vertical scale bar in C is 200  $\mu$ m.

exacerbated the condition (Fig. 1C). As the IOP remained under 8 mmHg for one month despite injection of gas and viscoelasticity into the anterior chamber, transconjunctival flap re-suturing was performed on POD 34. After treatment, the IOP increased to 30 mmHg and OCT showed the disappearance of SRD and decrease of choroidal thickness (362  $\mu$ m) in the left eye (Fig. 1D). Four months after surgery, her BCVA was 20/63 in the left eye with an IOP of 19 mmHg.

Case 2: A 60-year-old male with type 2 diabetes (HbA1c 13.6%) was diagnosed with diabetic retinopathy in both eyes. His BCVA was 20/25 in the right eye and 20/20 in the left eye. IOP was 18 mmHg in both eyes. Fundus examination of both eyes revealed hard exudates and intraretinal microvascular abnormalities. OCT showed macular edema in the right eyes (Fig. 2A). He underwent panretinal photocoagulation



**Fig. 2.** Optical coherence tomography (OCT) image before and after trabeculectomy in Case 2 (60-year-old male). (A) OCT image before surgery shows intraretinal cyst and discontinuous ELM line. (B) OCT image shows choroidal wrinkles and pachychoroid on postoperative day (POD) 6. (C) OCT image shows serous retinal detachment (SRD) on POD 22. (D) SRD resolved upon resolution of hypotony on POD 64. The choroidal thickness on POD 6, 22, and 64 was 333, 510 and 457  $\mu$ m, respectively. Vertical scale bar in C is 200  $\mu$ m.

and sub-Tenon's capsule triamcinolone acetonide injection. Under these treatments, an increase in IOP was observed and a large number of new blood vessels was observed on the iris surface of the right eye. The IOP increased to 36 mmHg despite anti-VEGF injection. His BCVA was 20/50 in the right eye. PAS was observed in a part of angle (PAS index < 0.1). He was diagnosed as NVG stage 2. Trabeculectomy with MMC was performed to control the IOP and the procedure was uneventful. After LSL, his BCVA was 20/20 in the right eye with an IOP of 3 mmHg. Fundus examination showed disc swelling and several fine retinal folds as hypotony maculopathy and OCT showed slightly increased choroidal thickness (333  $\mu$ m) (Fig. 2B). OCT revealed apparent SRD and thick choroid (510  $\mu$ m) on POD 22, and exacerbated the condition (Fig. 2C). Injection of gas and viscoelasticity into the anterior chamber and anti-VEGF injection were performed on POD 47 but no improvement was seen. Transconjunctival flap re-suturing was

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performed on POD 54. After treatment, IOP increased to 20 mmHg and OCT showed the disappearance of SRD but pachychoroid persisted (Fig. 2D). Four months after surgery, -his BCVA was 20/200 in the right eye with an IOP of 23 mmHg.

#### 2. Discussion

Kokame et al. reported a case with both SRD and CME in hypotony maculopathy.<sup>4</sup> However, we experienced two cases secondary to SRD accompanied by pachychoroid but not CME in hypotony maculopathy after trabeculectomy for NVG.

We consider three hypotheses for the causes of SRD accompanied by pachychoroid in hypotony maculopathy. First, vascular hyperpermeability in diabetic retinopathy occurs as a result of blood-retinal barrier breakdown due to many factors including vascular endothelial growth factor (VEGF) upregulation.<sup>6</sup> Kokame et al. reported diffuse microvascular leakage in the macula with fluorescein angiogram.<sup>4</sup> However, an administration of anti-VEGF drug was not effective in our case (case 2). Therefore, other factors aside from vascular hyperpermeability may be involved in the pathogenesis of our cases. Second, Kokame et al. reported that low intraocular pressure represents low tissue hydrostatic pressure, resulting in a higher hydrostatic pressure gradient across retinal capillaries, which promotes a net movement of fluid into the extracellular spaces, as presented in a modified concept of the Starling hypothesis.<sup>4</sup> Third, various reports have demonstrated breakdown of the retinal pigment epithelium (RPE) barrier in diabetic retinopathy.7 Lains et al. reported that swept-source OCT demonstrated a significant reduction of choroidal thickness in proliferative diabetic retinopathy.<sup>8</sup> However, a previous report by Saeedi et al. showed that lowering of IOP after trabeculectomy is associated with increased choroidal thickness.<sup>9</sup> In our cases, after surgery, lowering IOP induced choroid thickening with a modified concept of the hypothesis by Starling. Furthermore, in this case, diabetic retinopathy with poor glycemic control could have led to the breakdown of the RPE barrier, and the stored fluid in the choroid may have caused SRD.

Hypotony maculopathy after trabeculectomy for NVG in diabetic eyes may involve SRD accompanied by pachychoroid. IOP correction may be required for hypotony maculopathy after trabeculectomy in diabetic cases.

#### 3. Patient consent

Written informed consent for the research and publication of this study and any accompanying images was obtained from the patients.

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# Authorship

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

## Declaration of competing interest

All authors have no financial disclosures.

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