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Permanent ocular remodeling in the setting of chronic hypotony after trabeculectomy: A case report

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ABSTRACT

Purpose: Trabeculectomy surgery is a commonly performed procedure for treatment of glaucoma. While the goal is to lower intraocular pressure, over-filtration may cause hypotony with ocular structural changes and vision loss.

Observations: A 53-year-old woman with primary open-angle glaucoma was referred to our service for further evaluation. The patient previously underwent trabeculectomy 9 years prior and was found to have a cataract and hypotony maculopathy in the right eye. Treatment options included cataract surgery alone, bleb revision alone, or combined cataract extraction and bleb revision. Biometry revealed corneal astigmatism in the right eye, and significant disparity in axial length between the two eyes. Since the axial length and corneal astigmatic changes were presumed to be at least partially reversible, measurements from the non-operative left eye influenced the lens selection for the hypotonous right eye. The patient underwent combined phacoemulsification and bleb revision. While IOP increased and hypotony was partly reversed, there was hyperopic and astigmatic refractive surprise after surgery.

The patient subsequently underwent intraocular lens exchange using biometric values of the previously hypotonous eye and met the target post-operative refractive goal.

Conclusions and importance: This case demonstrates changes to the axial length and ocular structure following longstanding hypotony maculopathy may be permanent, even after restoration of normotensive intraocular pressure.

1. Introduction

Trabeculectomy surgery remains the gold standard intervention in many glaucoma practices for patients requiring low intraocular pressure[1] with over 20,000 procedures being performed annually among Medicare patients alone.[2] While the primary goal of surgery is to lower intra-ocular pressure (IOP) to slow or stop the progression of retinal ganglion cell loss, over-filtration may occur and cause hypotony, defined as structural changes to the eye resulting in vision loss. Vision loss in the setting of hypotony may result from corneal edema, corneal astigmatism, cystoid macular edema, subretinal fluid, optic nerve head edema, or chorioretinal folds.[3–6] The visual effects can be profound as the average visual acuity in the setting of hypotony maculopathy reaches that of legal blindness at 20/200.[7] Younger age, myopia, and antimetabolite exposure are all risk factors for the development of hypotony maculopathy after trabeculectomy surgery.[6,8]

The standard treatment for hypotony maculopathy is to increase the IOP. Conservative medical management may be attempted with aqueous suppressants to induce some degree of bleb failure or bandage contact lens; however, surgical intervention is typically required with either *trans*-conjunctival compression sutures or an open revision of the trabeculectomy bleb to re-suture the scleral flap.[7,9–12] Successful elevation of the IOP can lead to structural and visual recovery for many patients. However, long-standing hypotony has the potential to cause retinal, choroidal, or scleral fibrosis, which may impair resolution of chorioretinal folding.[3] In addition to vision loss, these architectural changes may carry refractive consequences. Subsequent cataract surgery may pose a unique challenge due to possible changes in axial length measurements.[13,14] Herein, we present a case of presumed permanent remodeling of the eye induced by chronic hypotony that resulted in a refractive surprise following cataract surgery, requiring a lens exchange for visual rehabilitation.

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2. Case report

A 53-year-old woman diagnosed with primary open angle glaucoma nearly two decades prior was referred to our glaucoma service for further evaluation. The patient previously underwent argon laser trabeculoplasty (ALT) in 2000 and a trabeculectomy with mitomycin C in 2010 of her right eye. On presentation in 2019, the patient was taking 4 glaucoma medications in each eye despite an IOP of 1 mmHg in the right eye post-trabeculectomy. The IOP was 19 mmHg in the fellow left eye. Best corrected visual acuity (BCVA) was 20/125 (pinhole 20/60-1) in the right eye and 20/15 in the left eye. Anterior segment examination of the right eye revealed evidence of anterior segment hypotony with

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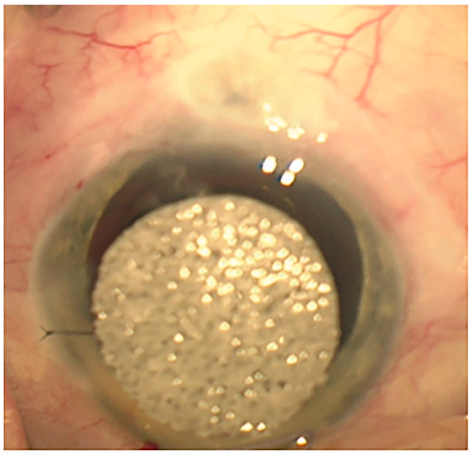

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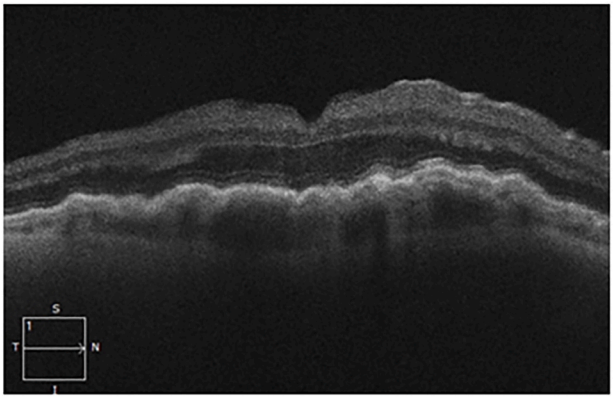

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		right eye		left eye	
Measuring mode	Mode	Phakic		Phakic	
Axial length	AL	21.70 mm	±0.019 mm	24.83 mm	±0.007 mm
Cornea thickness	CCT	543 µm	±2.2 µm	542 µm	±1.4 µm
Aqueous depth	AD	2.23 mm	±0.007 mm	2.64 mm	±0.006 mm
Anterior chamber depth inc..	ACD	2.78 mm	±0.005 mm	3.18 mm	±0.005 mm
Lens thickness	LT	4.19 mm	±0.008 mm	4.33 mm	±0.003 mm
Retina thickness	RT	200** µm	±0.0 µm	200** µm	±0.0 µm
Flat meridian	K1	42.07 D @ 174°	±0.108 D	43.69 D @ 74°	±0.031 D
Steep meridian	K2	47.33 D @ 84°	±0.186 D	44.19 D @ 164°	±0.155 D
Astigmatism	AST	5.26 D @ 84°	±1.4°	0.50 D @ 164°	±11.0°
Keratometric index	n	1.3375		1.3375	

Fig. 1. Pre-bleb revision and cataract extraction. Baseline testing prior to cataract extraction and trabeculectomy revision. A) Photograph taken at the start of surgery for bleb revision and cataract extraction showing avascular bleb. Corneal light shield in place B) Optical coherence tomography of the macula showed choroidal folds. C) Ocular biometry showed significant differences in axial length and corneal astigmatism between fellow eyes (right eye had chronic hypotony).

microstriae in the cornea under cobalt blue light, a low-lying cystic and avascular bleb (Fig. 1A), a relatively deep anterior chamber, and a likely visually significant nuclear sclerotic and cortical cataract. Fundoscopy showed glaucomatous cupping with choroidal folds that were also visible by optical coherence tomography of the macula (Fig. 1B). Given the IOP of 1 mmHg, all glaucoma medications were stopped in the right eye after which the IOP increased to 4 mmHg. Surgical options were discussed with the patient. The patient was subsequently lost to follow-up in the context of the COVID-19 pandemic.

Two years later, IOP and BCVA visual acuity remained relatively stable at 3 mmHg and 20/200 (pinhole 20/100), respectively, in the right eye. Given the visually significant cataract and hypotony maculopathy, several options were discussed with the patient including cataract surgery alone with the possibility of inducing scarring to the bleb and subsequent IOP elevation, bleb revision alone noting that future cataract surgery may induce bleb failure once the IOP was stable, or combined cataract extraction and bleb revision. The patient elected to proceed with combined cataract extraction and bleb revision. Preoperative refractive error was $-1.75 + 3.00 \times 084^{\circ}$ in the right eye and $-3.50 + 0.50 \times 001^{\circ}$ in the left eye (Table 1). Of note, pre-trabeculectomy refractive error was similar between the two eyes: $4.00 + 1.25 \times 029^{\circ}$ and $-3.50 + 0.50 \times 173^{\circ}$ for the right and left eyes, respectively. Ocular biometry indicated axial lengths of 21.70 mm and 24.83 mm, and corneal astigmatism of 5.3 diopters and 0.5 diopters, in the right and left eyes, respectively (Fig. 1C). The anterior chamber depth was 2.78 mm in the right eye and 3.18 mm in the left eye, with a lens thickness of 4.19 mm in the right eye and 4.33 mm in the left eye. Of note, no pre-trabeculectomy axial length or other biometric data was available.

Given evidence of anterior and posterior segment hypotony that was presumed to be at least in part reversible, ocular biometric calculations from the fellow left eye were used to influence selection of an intraocular lens (IOL) for the right eye. Using the Barrett formula, a +19.0 D hydrophobic acrylic IOL was selected to aim for a desired post-operative refraction of −2.0 D to balance the fellow eye. For the bleb revision, a peritomy was made superiorly through the prior ischemic, thin-walled bleb. After exposure, the prior scleral flap was found to have melted therefore requiring a pericardial patch graft to cover the sclera over the fistula. Ischemic conjunctiva was resected, and healthy vascular conjunctiva was advanced to the limbus and sutured with a running 10-0 nylon. The bleb revision was followed by uncomplicated phacoemulsification and intraocular lens insertion.

Three months after surgery, IOP improved to 10 mmHg with healthy bleb morphology (Fig. 2A). The choroidal folds were significantly reduced (Fig. 2B) and uncorrected visual acuity was 20/200. Despite targeting −2.0 D, postoperative manifest refraction was $+1.75 + 4.25 \times 085^{\circ}$ with BCVA 20/40. The hyperopic and astigmatic refractive surprise suggested that permanent scleral remodeling had occurred during the chronic period of hypotony. The patient was offered an IOL exchange. Repeat ocular biometry three months after bleb revision and cataract surgery showed axial lengths of 22.05 mm in the right eye and 24.85 mm in the left eye with corneal astigmatism of 4.8 D in the right eye indicating that the axial length and corneal astigmatism had minimally corrected after reversal of hypotony (Fig. 2C). An IOL exchange with a +29.0 D hydrophobic acrylic IOL with 6 D of astigmatism correction in the IOL plane at 85° was performed. Refractive error at 4 months after IOL exchange was −3.0 D sphere (no cylinder, Table 1) with best-corrected visual acuity of 20/25.

Table 1
Refractive error and acuity history. Best corrected visual acuity (BCVA).*Pinhole visual acuity.

Date & Surgery Status	Right eye			Left eye		
	Sphere	Cylinder	BCVA	Sphere	Cylinder	BCVA
2009, pre-trabeculectomy	−4.00	+1.25	20/20-2	−3.50	+0.50	20/20
2019, pre-bleb revision	−1.00	+2.75	20/60-1*	−3.75	+0.50	20/15
2021, pre-bleb revision	+0.25	+4.00	20/40-2	−3.75	+0.75	20/20
2022, 3 months post- bleb revision & CE/IOL	+1.75	+4.25	20/30-2	−3.75	+0.75	20/15
2022, 4 months post- lens exchange	−3.00	Sphere	20/25-1	−3.75	+1.00	20/20
2023, 14 months post-lens exchange	−3.00	Sphere	20/25-2	−3.75	+1.00	20/20

3. Discussion

We present a case of presumed permanent ocular remodeling after hypotony from trabeculectomy as evidenced by a hyperopic and astigmatic surprise after cataract surgery. The patient ultimately did well after an IOL exchange with 20/25 vision while meeting the refractive target of reading myopia without cylindrical correction.

Axial length shortening after trabeculectomy had been previously reported to be 0.14–0.42 mm.[13–15] As reviewed by Alvani and colleagues, which included patients from 25 different studies, showed an axial length reduction of up to 0.19 mm with contact biometry and up to 0.9 mm with noncontact biometry. Furthermore, up to 1.4 diopters of with the rule astigmatism was seen in this patient population.[16] In another study of 39 patients who underwent trabeculectomy, patients with IOP <4 mmHg had an axial length reduction of 0.39 mm while trabeculectomy eyes with IOP >4 mmHg had an axial length reduction of 0.14 mm.[13] Another report of 62 phakic patients found a decrease in axial length of 0.42 mm in patients who underwent trabeculectomy. Furthermore, hypotony maculopathy was associated with a decrease in axial length.[14] Based on this, various suggestions have been made to determine the correct IOL power in a phakic patient post-trabeculectomy needing cataract surgery from obtaining pre-trabeculectomy IOL measurements in phakic patients, though often this data is unavailable, to using an average of pre-trabeculectomy and post-trabeculectomy axial lengths.[14]

Since our patient had an axial length difference of 3.13 mm between the two eyes and 5.3 D of with-the-rule astigmatism prior to bleb revision, and we hypothesized that likely a significant a portion of these two metrics would be reversible with correction of hypotony. Repeated biometry four months after trabeculectomy revision (prior to IOL exchange) found that the axial length difference between eyes reduced to 2.80 mm and corneal astigmatism had reduced to 4.8 D suggesting that the vast majority of the axial length astigmatism changes actually did not correct with reversal of hypotony in our case. The 0.35 mm increase in axial length of the right eye after increasing the IOP to the normotensive range with bleb revision was more consistent with the studies above as far as axial changes after trabeculectomy. Therefore, we hypothesize that the residual 2.80 mm difference between the two eyes was related to scleral fibrosis contributing to the permanent shortening of axial length.

Improvement in vision in patients with hypotony after trabeculectomy following normalization of the IOP[11,15] is likely due to resolution of chorioretinal folds with recovery of retinal photoreceptors.[3] Consistent with this, since our patient ultimately achieved 20/25 BCVA after bleb revision and IOL exchange, the changes in the neurosensory retina were largely reversible while restoration of axial length was not. Though the patient presented with evidence of hypotony as evidenced by choroidal folds 9 years after initial trabeculectomy surgery, the duration of hypotony was not known. Noting an IOP of 4 at the end of the post operative period, no outside documentation of choroidal folds or other clinical findings of hypotony were present. This case highlights the degree and permanent nature to which these changes can occur even with

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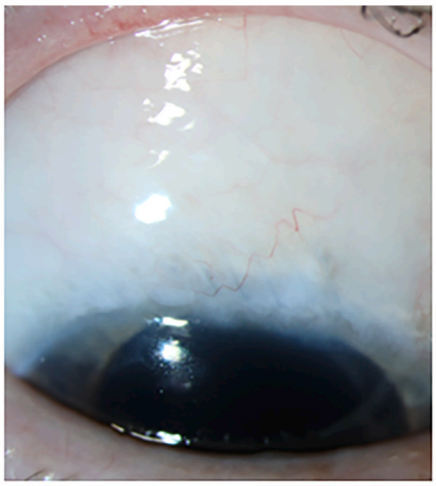

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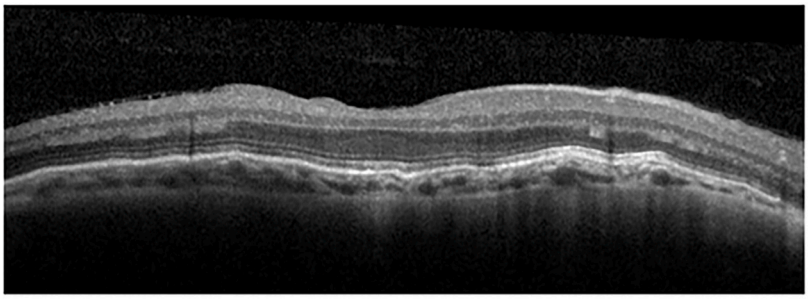

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		right eye		left eye	
Measuring mode	Mode	Pseudophakic default IOL		Phakic	
Axial length	AL	22.05 mm	±0.013 mm	24.85 mm	±0.019 mm
Cornea thickness	CCT	553 µm	±2.2 µm	538 µm	±3.0 µm
Aqueous depth	AD	-----	-----	2.65 mm	±0.003 mm
Anterior chamber depth inc..	ACD	-----	-----	3.19 mm	±0.004 mm
Lens thickness	LT	-----	-----	4.34 mm	±0.012 mm
Retina thickness	RT	200** µm	±0.0 µm	200** µm	±0.0 µm
Flat meridian	K1	42.23 D @ 175°	±0.165 D	43.46 D @ 89°	±0.147 D
Steep meridian	K2	47.08 D @ 85°	±0.236 D	44.39 D @ 179°	±0.248 D
Astigmatism	AST	4.84 D @ 85°	±0.6°	0.93 D @ 179°	±7.9°
Keratometric index	n	1.3375		1.3375	

Fig. 2. Post-bleb revision and cataract extraction. A) After revision, the bleb was low, diffuse, and without ischemia. B) Optical coherence tomography of the macula showed an incomplete improvement in choroidal folds. C) Ocular biometry measurements showed a slight increase in axial length and reduction in corneal astigmatism in the right eye.

reversal of hypotony and normalization of IOP.

It should be noted that our case may be somewhat atypical. A myopic surprise following cataract extraction post trabeculectomy is not uncommonly observed.[17] Furthermore, an IOP ≤9 following trabeculectomy has been shown to be a risk factor development of a myopic surprise when undergoing subsequent cataract extraction.[18] This study also suggested that an IOP spike after phacoemulsification was associated with a high risk of myopic surprise in the low IOP group.[18] Our patient had no documented IOP spike either after initial trabeculectomy, after combined cataract extraction and bleb revision or after IOL

exchange. Furthermore, astigmatic changes after trabeculectomy are common and tend to occur in the direction of the scleral flap.[19] A limitation to our study is the lack of pre-trabeculectomy biometric measurements including axial length measurements. However, given the symmetric pre-trabeculectomy refractive error of moderate myopia we hypothesize that the disparity of axial length measurements 9 years after trabeculectomy was secondary to chronic and permanent remodeling changes.

4. Conclusion

In summary, this case demonstrates permanent structural remodeling likely caused by chronic hypotony despite normalization of IOP. This can lead to refractive surprises when using the fellow eye as a basis for IOL calculations in anticipation of the hypotonous eye returning to its original structure. We recommend repeating biometry in the setting of prolonged choroidal folds when planning for eventual cataract extraction with IOL placement. With normalization of IOP and an appropriate IOL selection, visual potential may be excellent as in our patient's case.

Patient consent to publish

The patient described in this report granted their verbal consent to publish this case report and images contained within.

CRediT authorship contribution statement

Nathaniel Cameron: Writing – original draft. **Taylor Nayman:** Writing – review & editing. **Sanjay V. Patel:** Writing – review & editing. **Gavin Roddy:** Conceptualization, Data curation, Project administration, Supervision, Writing – review & editing.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Author SVP has the following disclosure: Consultant: Santen Inc., Design Therapeutics Inc.

Royalties: Developer of V-FUCHS, which Mayo Clinic licenses to Santen Inc., Trefoil Therapeutics, Inc., Kowa Research Institute, Aerie Pharmaceuticals, Iris Medicine.

References

1. Singh K, Sherwood MB, Pasquale LR. Trabeculectomy must survive. *Ophthalmol Glaucoma*. 2021;4(1):1–2. https://doi.org/10.1016/j.ogla.2020.08.009.
2. Ma AK, Lee JH, Warren JL, Teng CC. GlaucoMap – distribution of glaucoma surgical procedures in the United States. *Clin Ophthalmol Auckl NZ*. 2020;14:2551–2560. https://doi.org/10.2147/OPTH.S257361.
3. Costa VP, Arcieri ES. Hypotony maculopathy. *Acta Ophthalmol Scand*. 2007;85(6):586–597. https://doi.org/10.1111/j.1600-0420.2007.00910.x.
4. Camp AS, Weinreb RN. Hypotony keratopathy following trabeculectomy. *J Glaucoma*. 2020;29(2):77. https://doi.org/10.1097/IJG.0000000000001425.
5. Kokame GT, Leon MDL de, Tanji T. Serous retinal detachment and cystoid macular edema in hypotony maculopathy. *Am J Ophthalmol*. 2001;131(3):384–386. https://doi.org/10.1016/S0002-9394(00)00794-7.
6. Stamper RL, McMenemy MG, Lieberman MF. Hypotonous maculopathy after trabeculectomy with subconjunctival 5-fluorouracil. *Am J Ophthalmol*. 1992;114(5):544–553. https://doi.org/10.1016/S0002-9394(14)74481-2.
7. Cohen SM, Flynn HW, Palmberg PF, Gass JD, Grajewski AL, Parrish RK. Treatment of hypotony maculopathy after trabeculectomy. *Ophthalmic Surg Lasers*. 1995;26(5):435–441.
8. Nicolela MT, Carrillo MM, Yan DB, Rafuse PE. Relationship between central corneal thickness and hypotony maculopathy after trabeculectomy. *Ophthalmology*. 2007;114(7):1266–1271. https://doi.org/10.1016/j.ophtha.2006.10.024.
9. B Scoralick AL, Almeida I, Ushida M, et al. Hypotony management through transconjunctival scleral flap resuturing: analysis of surgical outcomes and success predictors. *J Curr Glaucoma Pract*. 2017;11(2):58–62. https://doi.org/10.5005/jp-journals-10028-1224.
10. Thomas M, Vajaranant TS, Aref AA. Hypotony maculopathy: clinical presentation and therapeutic methods. *Ophthalmol Ther*. 2015;4(2):79–88. https://doi.org/10.1007/s40123-015-0037-z.
11. Schwartz GF, Robin AL, Wilson RP, Suan EP, Pheasant TR, Prensky JG. Resuturing the scleral flap leads to resolution of hypotony maculopathy. *J Glaucoma*. 1996;5(4):246–251.
12. Quaranta L, Riva I, Floriani IC. Outcomes of conjunctival compression sutures for hypotony after glaucoma filtering surgery. *Eur J Ophthalmol*. 2013;23(4):593–596. https://doi.org/10.5301/ejo.500027.
13. Francis BA, Wang M, Lei H, et al. Changes in axial length following trabeculectomy and glaucoma drainage device surgery. *Br J Ophthalmol*. 2005;89(1):17–20. https://doi.org/10.1136/bjo.2004.043950.
14. Cashwell LF, Martin CA. Axial length decrease accompanying successful glaucoma filtration surgery. *Ophthalmology*. 1999;106(12):2307–2311. https://doi.org/10.1016/S0161-6420(99)90531-6.
15. Alagöz N, Taskoparan S, Altan AC, et al. Pressure restoration and visual recovery time in hypotony after trabeculectomy. *Int Ophthalmol*. 2021;41(9):3183–3190. https://doi.org/10.1007/s10792-021-01883-1.
16. Alvani A, Pakravan M, Esfandiari H, Safi S, Yaseri M, Pakravan P. Ocular biometric changes after trabeculectomy. *J Ophthalmic Vis Res*. 2016;11(3):296–303. https://doi.org/10.4103/2008-322X.188399.
17. Zhang N, Tsai PL, Catoira-Boyle YP, et al. The effect of prior trabeculectomy on refractive outcomes of cataract surgery. *Am J Ophthalmol*. 2013;155(5):858–863. https://doi.org/10.1016/j.ajo.2012.11.023.
18. Yeh OL, Bojikian KD, Slabaugh MA, Chen PP. Refractive outcome of cataract surgery in eyes with prior trabeculectomy: risk factors for postoperative myopia. *J Glaucoma*. 2017;26(1):65–70. https://doi.org/10.1097/IJG.0000000000000560.
19. Shiratori N, Nakamoto K, Nishio Y, et al. Statistical analysis of factors affecting surgically induced astigmatism following trabeculectomy. *Clin Ophthalmol Auckl NZ*. 2022;16:3833–3839. https://doi.org/10.2147/OPTH.S389480.