

Reversal of severe myopia by 24 years of hypotony with subsequent stable refraction after 2 years of normal intraocular pressure

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ABSTRACT

Purpose: To report sustained axial length shortening and hyperopic shift (refraction changed from -8 to -2 diopters) induced by 24 years of profound IOP (intraocular pressure) reduction with subsequent refractive stability 2 years after IOP rose to 11–17 mm Hg.

Observations: A 25-year-old woman with elevated episcleral venous pressure glaucoma underwent non-penetrating trabeculectomy and subsequent laser goniopuncture in her left eye. She had chronically low IOP (1–12 mm Hg) for the next 24 years. Hypotony maculopathy was present in postoperative years 13–14 and 18–24 but resolved at age 49 when emesis-induced iris prolapse obstructed the filtering passage and transiently raised IOP to 40. Medical management and iridectomy with flap suturing stabilized IOP between 11 and 17 mm Hg. Refraction before OS trabeculectomy was OD -7.50 /OS -9.00 . In postoperative year 24 spherical equivalent phakic refraction was OD -9.00 /OS -1.50 . Biometry 1 year after resolution of hypotony showed axial lengths OD 24.8, OS 22.6 mm. Cataract surgery was performed in postoperative years 24/26 (OD/OS) with Tecnis DCBOO intraocular lenses of powers 14.0/21.5 diopters; postoperative refractions, 2 years after OS IOP rose to 11–17 mm Hg, were OD $+0.25$ – 0.50 x 015/OS -0.75 – 1.25 x 160 with 20/25 corrected acuity in each eye. Cardiovascular symptoms 24 years after the onset of her glaucoma led to a diagnosis of severe pulmonary hypertension.

Conclusions and importance: This case demonstrates that 2 decades of chronic IOP reduction can reverse myopia (by > 2 mm reduction in axial length) with subsequent refractive stability 2 years after IOP normalization. In addition, the case shows that ocular signs of pulmonary hypertension may precede cardiovascular signs by 2 decades.

1. Introduction

Higher childhood IOP is known to increase axial length¹ and Mendelian randomization has shown that higher IOP makes myopia likelier.² Conversely, it is known that hypotony can transiently reduce axial length.^{3,4} (see Table 1) (Table 1)

We present here a case of sustained axial length reduction (>2 mm relative to the contralateral eye) induced by trabeculectomy and profound long-term IOP reduction which was stable 2 years after IOP rose to 11–17 mm Hg. Other notable features of this case are resolution of hypotony maculopathy that had been present for 6 years and a delay of 24 years between the onset of ocular and cardiovascular symptoms of pulmonary hypertension (which is associated with increased pressures in the right ventricle, superior vena cava, and episcleral veins⁵).

2. Case report/findings

This report gives 2 decades of follow-up on a case published in 2003. A previously healthy woman was a glaucoma suspect at age 23; 2 years later she presented with IOPs 16/50 (OD/OS) mm Hg and cup/disc ratios 0.4/0.9, along with episcleral venous congestion OS $>$ OD and blood in Schlemm's canal OS (external and gonio photographs may be seen in the original publication). Maximum medical therapy lowered IOP to 32, so filtering surgery was planned. To reduce the risk of choroidal effusions that might result from abrupt IOP reduction, non-penetrating trabeculectomy (deep sclerectomy) was performed with subsequent laser goniopuncture (*ab interno* YAG laser perforation of the trabeculo-Descemet membrane created by the surgical unroofing of Schlemm's canal and removal of corneal stroma external to anterior

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Table 1
Management, Refraction, Axial Length and IOP range over follow up period.

Age	Management	Refraction	Axial Length (mm)	IOP range (mmHg)
25	OS Trabeculectomy	OD -7.50 OS -9.00		OD 15-20 OS 1-12
39		OS -1.00–3.00 x 165		OD 15-20 OS 1-12
49	OD drainage tube and phacoemulsification OS trabeculectomy revision	OD -9.00 (before phacoemulsification) OS -1.50	OD 25.0 OS 22.6	Transiently OD 30 OS 40 Subsequently OD teens OS 11-17
51	OS phacoemulsification	OD +0.25–0.50 x 015 OS -0.75–1.25 x 160	OD 24.8 OS 22.6	OD teens OS 11-17

trabecular meshwork and posterior Descemet's).

She complained of having heard a pulsing water sound inside her head for several years, but a preoperative cerebral angiogram showed no arteriovenous fistula.

IOP in the contralateral eye was controlled topically from age 25 until age 49 when IOP reached 30; OD IOP was then lowered to teens with an Ahmed FP-7 drainage tube and phacoemulsification.

2.1. Hypotony maculopathy

The left eye was stable for the first 12 years after filtration surgery with IOPs 6–12 mm Hg and BCVA (best corrected visual acuity) 20/25. At age 38, hypotony maculopathy developed: BCVA was still 20/25 with IOP 6 mmHg, but she complained of blurred vision, and ocular coherence tomography (OCT) and fluorescein angiography showed retinal pigment epithelial (RPE) and choroidal folds. The maculopathy resolved a year later with IOP 6–8, but then recurred at age 43. Over the next 6 years, IOP gradually fell to 1 with an over-filtering but non-leaking bleb OS, and BCVA drifted down to 20/80; OCT at age 48 showed RPE folds (Fig. 1). Three months later, iris prolapse due to vomiting obstructed the filtering track and raised OS IOP to 40 mm Hg. Medications and trabeculectomy revision stabilized OS IOP in the teens and acuity initially improved to 20/25.

2.2. Delayed cardiovascular manifestation of pulmonary hypertension

A few months later, she developed serous macular detachment OS (Fig. 2), protrusion of the OD drainage device bleb between the closed lids (without implant extrusion), shortness of breath and marked jugular venous distention. She was diagnosed with pulmonary hypertension and transferred to a tertiary care hospital where her hospital stay was punctuated by resuscitation from a cardio-pulmonary arrest. She improved with sildenafil and treprostinil: 8 months after discharge, the maculopathy was resolved (Fig. 3) and she was able to walk 2 miles with supplemental oxygen.

2.3. Reversal of severe myopia by chronic hypotony

The patient's refraction was OD -7.50/OS -9.00 before filtering surgery at age 25. At age 39, refraction OS was -1.00–3.00 x 165 and the patient had evidence of hypotony maculopathy on OCT. At age 49, phakic refraction after resolution of hypotony OS was OD -9.00/OS -1.00–1.25 x 160.

Cataract surgery was performed OD (with Ahmed FP-7) at age 49 and OS at age 51. Biometry prior to each of those surgeries showed axial lengths 25.0/22.6 mm and 24.8/22.6 mm. Thus, the chronic hypotony OS was associated with a >2 mm reduction in axial length relative to OD. The intraocular lenses implanted were Tecnis DCBOO

(Johnson&Johnson, New Brunswick, NJ) OD 14.0/OS 21.5 diopters with subsequent 20/25 corrected acuity in each eye (refraction: OD +0.25–0.50 x 015/OS -0.75–1.25 x 160). The keratometry average dioptric powers were negligibly changed by surgery: OD 46.8/47.0 at age 49 and OS 46.4/47.0 at age 51.

3. Discussion

This 20-year follow up of a previously published case⁶ shows sustained hyperopic shift and axial length shortening consequent to chronic profound hypotony.

Axial length reduction is known to occur in children after IOP reduction.¹ In adults, several reports have documented axial length reductions <0.5 mm after IOP reduction for up to 1 year.^{7–10} IOP reduction in our case was of much longer duration (2 decades), which would seem to explain the sustained, large magnitude¹¹ axial length reduction.

Our literature search found that the longest previously reported periods of reversible hypotony maculopathy¹² were 7 years¹³ and 5 years.¹⁴ Our report of macular fold resolution after 6 years adds to the evidence that hypotony maculopathy may in some cases be reversible after many years if IOP is raised sufficiently.

There is some experimental evidence that IOP reduction can limit progression of myopia. Myopia-prone guinea pigs treated with latanoprost for 10 weeks had only 2 diopters of myopia compared to 8 diopters for controls¹⁵; interestingly, animal models have not shown beta-blockers to slow myopia, while prostaglandins and alpha-2 agonists have shown efficacy.¹⁶

A meta-analysis has found that timolol probably reduces myopic regression after laser refractive surgery¹⁷; however, we found no trials that have been reported of IOP reduction to prevent progression of myopia in humans.

A final point is the importance of considering the diagnostic possibility of pulmonary hypertension in the setting of increased episcleral venous pressure. Macular edema due to increased venous pressure has been reported to be present at the same time as pulmonary hypertension symptoms.¹⁸ This case, however, shows that ocular signs of pulmonary hypertension may precede cardiovascular signs and symptoms by 2 decades.

4. Conclusions

Chronic, substantial IOP reduction can cause significant and stable axial length reduction. The possibility that chronic IOP reduction on a population basis might reduce the incidence of myopia merits further study. Hypotony maculopathy may be reversed, even after 6 years, if IOP is increased substantially. Finally, episcleral venous congestion may present more than two decades before cardiovascular signs of pulmonary hypertension.

Patient consent

Oral consent to publish this case has been obtained.

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Authorship

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

CRedit authorship contribution statement

Dinukie-Chantal Perera: Data curation, Writing – original draft, Writing – review & editing, Investigation. **Peter E. Libre:**



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Technician:

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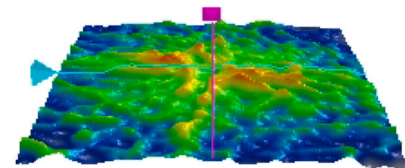
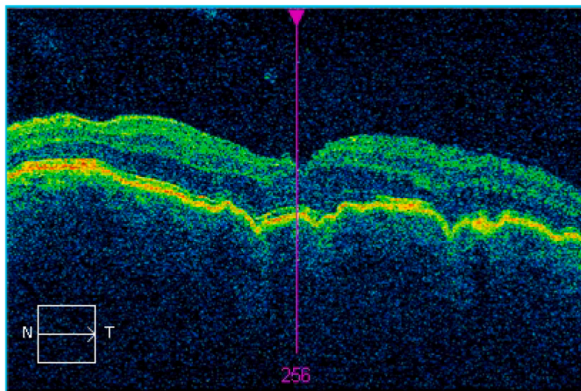
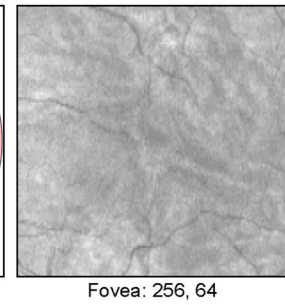
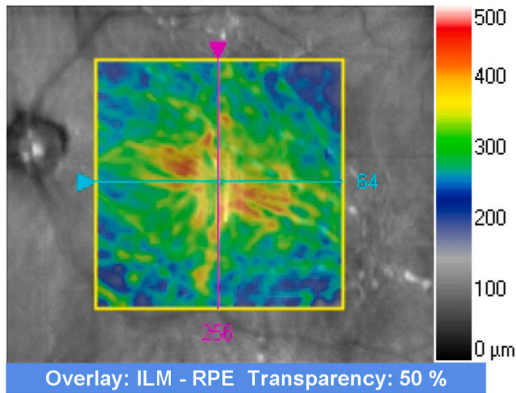
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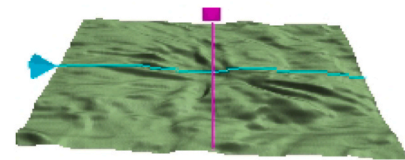
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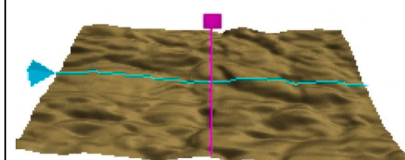
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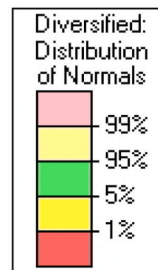
ILM - RPE



ILM



RPE



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	349	10.3	287

Comments

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Fig. 1. Optical Coherence Tomography OS hypotony maculopathy (retinal wrinkling) 23 years after trabeculectomy.



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Gender: Female

Technician:

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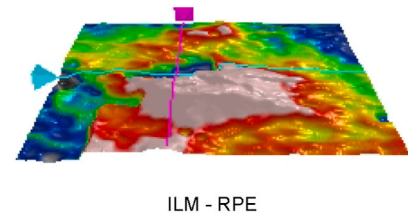
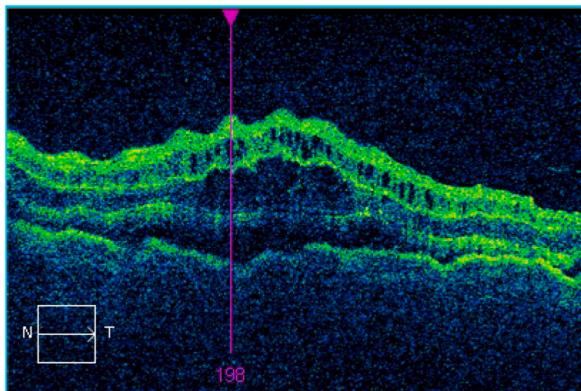
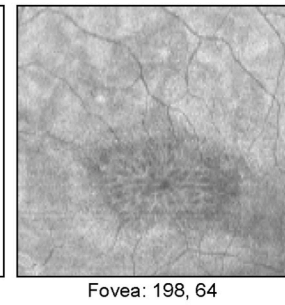
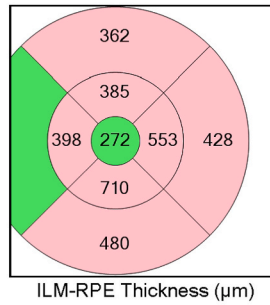
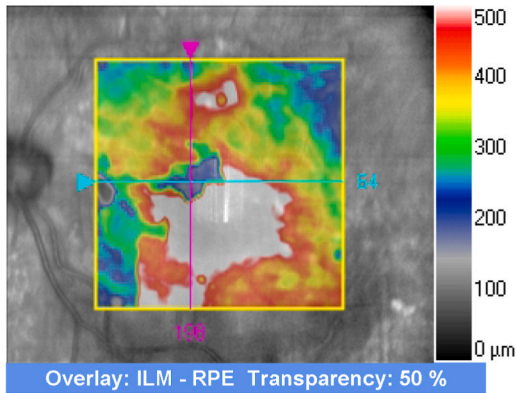
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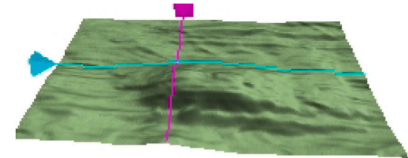
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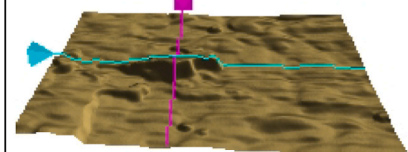
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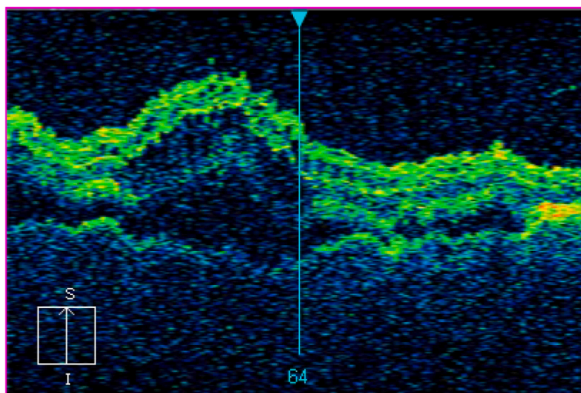
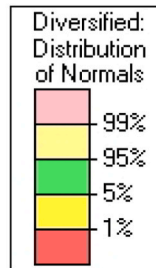
ILM - RPE



ILM



RPE



	Central Subfield Thickness (μm)	Cube Volume (mm³)	Cube Average Thickness (μm)
ILM - RPE	272	14.2	395

Comments

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Fig. 2. Optical Coherence Tomography OS macular edema and serous detachment due to worsened pulmonary hypertension.



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Gender: Female

Technician:

Exam Date:

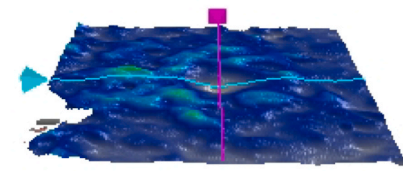
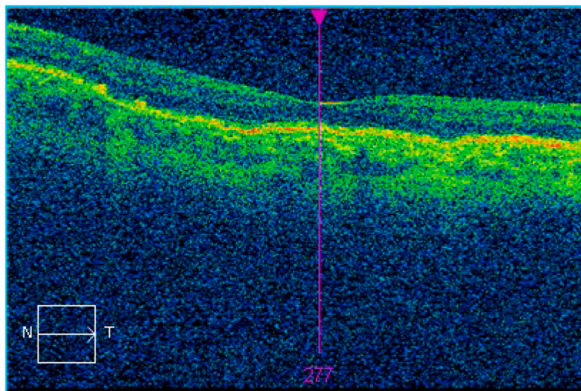
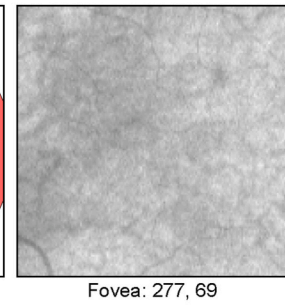
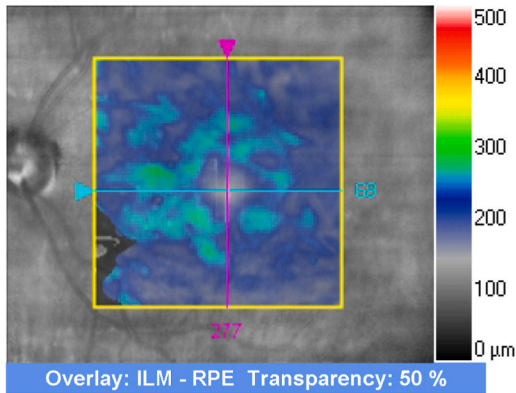
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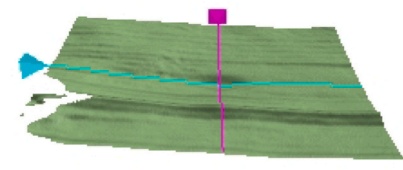
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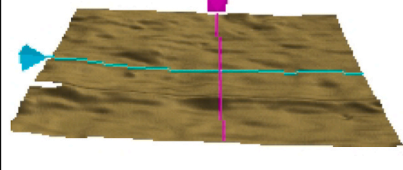
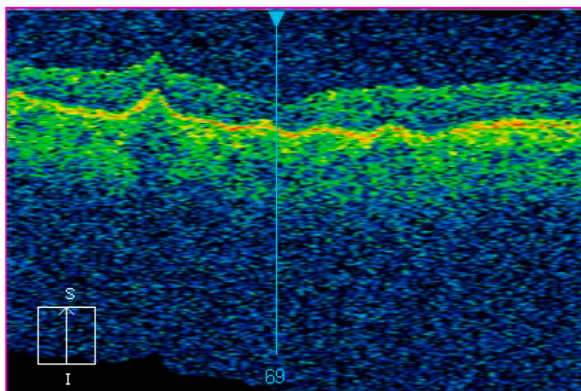
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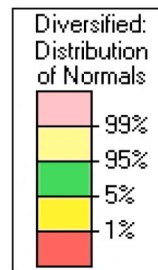
ILM - RPE



ILM



RPE



	Central Subfield Thickness (μm)	Cube Volume (mm³)	Cube Average Thickness (μm)
	179	7.7	214

Comments

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Fig. 3. Optical Coherence Tomography OS resolution of macular edema, detachment and wrinkles.

Conceptualization, Data curation, Investigation, 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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