American Journal of Ophthalmology Case Reports 7 (2017) 95–98

Contents lists available at ScienceDirect

American Journal of Ophthalmology Case Reports

journal homepage: http://www.ajocasereports.com/

Case report

Acute hypotony maculopathy following the initiation of a topical aqueous suppressant in a patient with a history of panuveitis without prior filtering surgery

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ARTICLE INFO

Article history:
Received 8 October 2016
Received in revised form
23 April 2017
Accepted 20 June 2017
Available online 22 June 2017

Keywords:
Cystoid macular edema
Glaucoma medication
Hypotony maculopathy

ABSTRACT

Purpose: To report a case of profound hypotony maculopathy as a complication of single-agent glaucoma therapy in a patient with a history of panuveitis without previous filtering surgery.

Observations: A 70-year old Hispanic male with a history of resolved bilateral panuveitis, chronic angle closure glaucoma, and pars plana vitrectomy was started on topical timolol 0.5% daily in the left eye for mildly elevated intraocular pressure (15 mmHg). The patient returned 1.5 weeks later with new onset hypotony (1 mmHg), chorioretinal folds, and cystoid macular edema in the same eye without associated signs of inflammation. The drop was discontinued. The patient returned 1 month later with normalized eye pressure and improved vision with near-resolution of chorioretinal changes on optical coherence tomography (OCT).

Conclusions and importance: Hypotony maculopathy is most commonly seen following glaucoma filtering surgery and ocular trauma. The development of hypotony maculopathy following the administration of topical glaucoma medication alone is rare. Our case is the first to our knowledge to describe the rapid onset of visually significant hypotony maculopathy characterized by profound OCT changes upon the administration of a single topical glaucoma agent in a patient without prior filtering surgery. Treatment with glaucoma medications in patients with complex ocular histories including uveitis and vitreoretinal surgery requires caution and close follow-up.

1. Introduction

Hypotony maculopathy, a term coined by Don Gass[1] in 1972, was first described by Dellaporta[2] in 1954 and represents the development of posterior fundus abnormalities including chorioretinal folds, vascular tortuosity, disc edema, and rarely cystoid macular edema in the setting of low intraocular eye pressure (IOP). Pederson[3] described “statistical” hypotony as an IOP less than 6.5 mmHg (3 standard deviations below the mean), whereas “clinically significant” hypotony is defined by an IOP below which results in visual loss. The pathogenesis of hypotony maculopathy, in particular, is deemed likely secondary to inward collapse of the scleral wall resulting in wrinkling and folding, especially of the thick perifoveal retina around the thin fovea. The subsequent photoreceptor distortion and decreased antero-posterior diameter of the eye leads to decreased visual acuity and relative hyperopia.[1] Hypotony maculopathy is most frequently seen following glaucoma filtering surgeries especially with the concurrent use of anti-fibrotic agents as well as in perforating eye injuries.[4,5] Other risk factors include young age, myopia, male gender, systemic illness, and elevated preoperative IOP.[6]

2. Case report

A 70- year old Hispanic male with a history of rheumatoid arthritis, resolved bilateral panuveitis, and chronic angle closure glaucoma presented for his first visit to our clinic for a routine exam. Examination of the right eye was unremarkable, but the left eye IOP was slightly above target at 15 mmHg. There were no signs of intraocular inflammation, and he was not taking any eye drops or systemic antihypertensive medications at the time. The patient was started on timolol 0.5% daily in the left eye and returned 1.5

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http://dx.doi.org/10.1016/j.ajoc.2017.06.016

weeks later with an IOP of 1 mmHg and decreased vision to 20/70 from 20/40 in the same eye. Fundus exam was notable for wrinkling of the macula as well as new onset cystoid macular edema on optical coherence tomography (OCT) (Fig. 1). There were no signs of choroidal detachment, retinal detachment, or optic nerve abnormalities. Timolol was discontinued and the patient returned one month later with subjectively improved vision (visual acuity 20/60) and an IOP of 18 mmHg. OCT demonstrated near resolution of chorioretinal folds and cystoid macular edema (Fig. 2).

A thorough review of his clinical record spanning over the past 12 years under the care of different providers revealed a history of similar repeated episodes of hypotony secondary to glaucoma medications. The patient initially presented in 2003 with visual acuity of 20/400 in the right eye and 20/200 in the left eye with elevated IOPs of 35 mmHg and 37 mmHg, respectively, secondary to bilateral panuveitis and acute on chronic angle closure glaucoma. Of note, both eyes were pseudophakic with a small degree of hyperopia (+0.50 spherical equivalent) and axial length around 22.5 diopters. His panuveitis was deemed mostly likely related to his underlying rheumatoid arthritis and was eventually controlled with bilateral pars plana vitrectomy, intravitreal fluocinolone acetonide implants, systemic immunosuppression with methotrexate, and chronic low-dose topical steroid treatment. IOPs improved to the mid-teens following bilateral laser peripheral iridotomies, and topical glaucoma medications were tapered and ultimately discontinued.

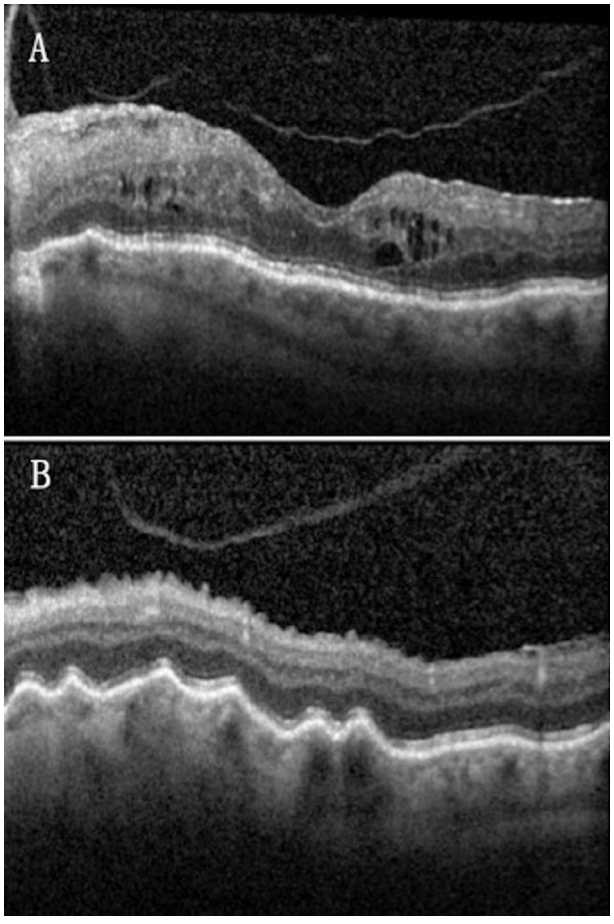

Fig. 1. Spectral domain-optical coherence tomography (SD-OCT) of the patient's central macula (A) and superior macula (B) demonstrating cystoid macular edema and extensive chorioretinal folds at 1.5 weeks following the initiation of timolol 0.5% daily in the left eye.

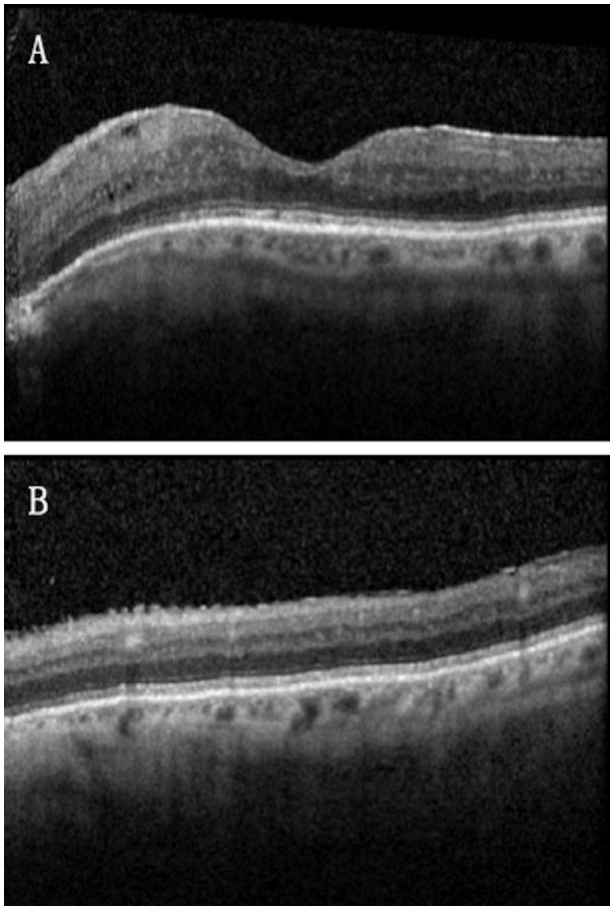

Fig. 2. Spectral domain-optical coherence tomography (SD-OCT) of the patient's central macula (A) and superior macula (B) demonstrating near resolution of cystoid macular edema and chorioretinal folds at 1 month following the cessation of timolol 0.5% daily in the left eye.

Nine years after his initial presentation, the patient was restarted on topical timolol 0.5% daily in the left eye for an elevated IOP of 22 mmHg at a routine visit. IOP of the right eye was acceptable at 11 mmHg. His next visit 3 months later revealed IOPs of 1 mmHg and 0 mmHg in the right and left eye, respectively. His vision remained unchanged at 20/60 and 20/50, and his fundus exam was normal with no intraocular inflammation noted. Gonioscopy was consistent with chronic angle closure. Timolol was discontinued, and the hypotony was treated with topical prednisolone acetate 1% three times daily in both eyes with improvement of his IOPs to 8 mmHg (right eye) and 16 mmHg (left eye) 1 month later. After a few years of relatively stable vision and IOPs on topical prednisolone acetate 1% twice daily, the patient presented with elevated IOPs of 18 mmHg and 31 mmHg in the right and left eye, respectively. He was treated with dorzolamide 2%/timolol 0.5% twice daily and latanoprost 0.005% nightly in both eyes. One month later, his IOPs had dropped to 2 mmHg and 1 mmHg, and the vision had decreased to 20/200 and 20/100 from his baseline of 20/60 and 20/40, respectively. Again no inflammation was seen, but macular striae were noted this time in both eyes. All glaucoma drops were again discontinued. He returned 3 months later with normalized IOP (9 mmHg and 15 mmHg), resolution of macular striae, and vision back to baseline.

3. Discussion

Our patient demonstrated three separate episodes of severe hypotony following topical glaucoma therapy, two of which resulted in visual impairment secondary to profound chorioretinal changes. Cystoid macular edema, a rare feature of hypotony maculopathy, was a prominent finding in this case. It has been hypothesized that low IOP translates to a higher pressure gradient across retinal capillaries that results in net movement of fluid into the interstitial space, especially in eyes with predisposing microvascular disease such as uveitis.[7] Interestingly, the bilateral hypotony seen after administrating timolol in only one eye is consistent with the known systemic effects of the drug, especially in patients with bilateral underlying ocular pathology. Similar to other cases of aqueous suppressant-induced hypotony, our patient demonstrated normalization of visual acuity with near resolution of chorioretinal findings following cessation of the inciting medication within one month, suggesting good reversibility with normalization of IOP.[8,9]

Since 1985, several cases of glaucoma drug-induced hypotony, mostly characterized by peripheral choroidal detachments, have been described in post-trabeculectomy patients.[8,9] Glaucoma filtering surgery, the most common cause of post-surgical hypotony, can result in bleb leakage, overfiltration, and even choroidal detachment, a hypotony-induced complication that can lower IOP further by decreasing aqueous humor production. Not surprisingly, hypotony can be exacerbated by topical glaucoma medications, several of which have been implicated including timolol.[8,9] Clinically significant maculopathy, however, is a less commonly seen outcome of hypotony. Our case is the first to our knowledge to describe acute, profound hypotony maculopathy following the administration of a single glaucoma medication in a patient without a history of filtering surgery.

The etiology of our patient's acute and drastic hypotensive response to topical glaucoma medications is unclear, but likely related to his complex ocular medical and surgical history. Cases of profound hypotony maculopathy in the absence of prior surgery and topical hypotensive therapy have been seen with acute idiopathic and HLA-B27 related anterior uveitis secondary to activation of prostaglandin-induced uveoscleral outflow or acute ciliary body dysfunction.[10,11] Each episode of acute hypotony in our patient, however, occurred in the absence of any intraocular inflammation. Patients with chronic uveitis can also develop long-term hypotony from tractional cyclitic membranes that damage ciliary processes and subsequently decrease aqueous production and increase outflow.[3] Our patient, however, did not demonstrate any signs of lingering inflammation in the years following his initial presentation. He did have a history of pars plana vitrectomy, a procedure which has been shown to predispose to hypotony due to anterior vitreous traction on ciliary processes, the surgical removal of which have been shown to resolve hypotony and improve vision.[12] However, these mechanical theories do not entirely explain our patient's clinical picture given that he had relatively normal to high IOP in between episodes off all glaucoma medications.

A more likely explanation is related to the alteration of dynamic forces required to maintain physiologic pressure in a post-surgical, uveitic eye. The balance of aqueous production and outflow resistance in such eyes is so precarious that the use of a single topical aqueous suppressant can result in large changes in IOP. The greater tendency for the eye to develop profound maculopathy in the setting of hypotony can also be explained by the patient's complex surgical history. It is known that pars plana vitrectomy, like trabeculectomy, can cause transient intra- and post-operative separation of the choroid.[13,14] Although there were no signs of choroidal detachment on exam, any egress of fluid into the widened suprachoroidal space from prior pars plana vitrectomy or even sclerotomy from fluocinolone acetonide implants could predispose to recurrent subclinical choroidal detachments and further hypotony, compromising the structural integrity of the eye wall at perhaps a higher IOP than would normally be expected.

4. Conclusions

In summary, we present a case of 'supersensitivity' to topical aqueous suppressants causing acute hypotony maculopathy and cystoid macular edema in a patient without a history of filtering surgery. Although we cannot explain the exact mechanisms by which this supersensitivity occurred, we postulate that our patient's complicated past ocular history of chronic uveitis and prior vitreoretinal surgery may have played a role. The initiation of glaucoma medications in similar patients, therefore, should be done so with caution and close follow-up.

Patient consent

Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the identification of the patient.

Funding

This work was supported by an unrestricted grant from Research to Prevent Blindness, New York, NY 10022.

Authorship

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

Conflict of interest

The following authors have no financial disclosures: DZ, HA, AR, NR.

Acknowledgements

None.

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