Case report

Hypotony maculopathy: a surgeon's nightmare

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Accepted 21 October 2019

SUMMARY

A 14-year-old myopic male patient diagnosed as steroid-induced glaucoma and cataract elsewhere presented to the eye opd with painless and progressive diminution of vision in the right eye (oculus dexter (OD)) for last 6 months. Phacotrabeculectomy was already done in the left eye (oculus sinister (OS)) 1 year ago and he was on topical latanoprost e/d-H.S in OD and brimonidine e/d-BD and dorzolamide e/d-TDS in both eyes (oculus uterque (OU)) postsurgery. Uneventful phacotrabeculectomy was performed in the right eye under 300 mL intravenous injection mannitol. He developed hypotony maculopathy postsurgery which was managed conservatively.

BACKGROUND

Trabeculectomy is the gold standard surgery in glaucoma. Complications associated with trabeculectomy can arise because of preoperative and postoperative factors. Apart from intraoperative events, age of the patient, use of mitomycin c, refractive status, biomechanical factors like scleral rigidity and thickness may affect the postoperative outcome. All these factors may lead to postoperative hypotony. Preoperative evaluation of all these factors is necessary to plan trabeculectomy surgery. Retrospectively, a number of preoperative factors were thought to predispose the hypotony rather than the intraoperative or postoperative events as seen in majority of cases.

CASE PRESENTATION

A 14-year-old male patient presented to the eye opd with chief complaints of painless and progressive diminution of vision in the right eye (OD) for last 6 months. He had recurrent episodes of redness and discharge for which he was locally treated with topical and systemic medications for last 2 years. Phacotrabeculectomy was done in the left eye (OS) 1 year ago and he was on topical latanoprost e/d-H.S in OD and brimonidine e/d-BD and dorzolamide e/d-TDS in both eyes (OU) postsurgery. On examination, the patient had a flat avascular bleb with pseudophakia in the left eye and posterior subcapsular cataract in the right eye. His best corrected visual acuity (BCVA) was 2/60 in OD and 6/9 in OS (Snellens chart). The intraocular pressure was 42 mm Hg in OD and 22 mm Hg in OS measured by applanation tonometer. Gonioscopy showed open angles in both eyes with a patent ostium in left eye. Fundus examination showed total cupping in both eyes ([figure 1](#)). Biometry showed axial length to be 25.44 mm with intraocular lens power

Figure 1 Fundus photograph of both eyes showing advanced cupping.

+13.0 dioptres as measured by SRK-T formulae (Biomedix). The patient was not compliant with the use of topical medications. So he was counselled properly and reviewed after 2 weeks. His intraocular pressure (IOP) came down to 20 and 14 mm Hg in OD and OS, respectively, at next follow-up. He was posted for phacotrabeculectomy with 2 mg% mitomycin c. Uneventful surgery was performed. On the third postoperative day, he developed hypotony maculopathy. Macular haemorrhage was seen in right eye ([figure 2](#)).

INVESTIGATIONS

Routine blood investigations prior to surgery were done which were normal. The third day postsurgery, optical coherence tomography (OCT) examination corroborated the finding of macular haemorrhage which gradually resolved ([figure 3](#)).

TREATMENT

Uneventful phacotrabeculectomy was performed in the right eye under 100 mL intravenous injection mannitol. He was put on topical steroids (1% prednisolone acetate) with 1% topical atropine eye drops. The eye was patched with pressure bandage and oral steroids (prednisolone 40 mg for 7 days and then tapered off) was started when hypotony developed

OUTCOME AND FOLLOW-UP

On the first postoperative day, right eye had a vision of 6/24 with an IOP of 28 mm Hg with a patent peripheral iridectomy. But his IOP dipped down to 12, 9 and 3 mm Hg on post operative day (POD) 2, 3 and 4, respectively. Fundus examination revealed total cupping with a macular haemorrhage. After 2 days, the patient was discharged with a vision 6/18 and IOP of 6 mm Hg in the right eye. After 1 week, the IOP was 10 mm Hg and visual acuity (VA) was 6/12 in OD. Oral and topical steroids were tapered and the patient was again followed up every 2 weeks

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To cite: Sen S, Parija S, Nayak B. *BMJ Case Rep* 2019;**12**:e231272. doi:10.1136/bcr-2019-231272

Figure 2 Fundus photograph showing macular haemorrhage.

for one and a half months. After 2 months following surgery, the IOP was 12 mm Hg and VA was 6/9 in OD. The macular haemorrhage had resolved and there were no signs of hypotony. Last follow-up was one and half years following surgery. The affected eye had an IOP of 12 mm Hg and vision 6/12 with a nice diffuse bleb (figure 4).

DISCUSSION

1. Hypotony maculopathy whether early or late is always sight threatening. Clinically in these cases, the IOP of the eye is not enough to maintain the shape of the eyeball. It is characterised by chorioretinal folds, vascular tortuosity and disc oedema. Statistically, it means the IOP is 3 SD below the mean or IOP is less than about 6 mm Hg.¹ Hypotony results from over drainage or underproduction of aqueous humour. Few causes are enumerated in the table 1.

Figure 3 OCT picture depicting macular haemorrhage and shadow beneath it.

Figure 4 Anterior segment photograph postoperatively showing diffuse bleb.

2. Significant wound alterations can occur postsurgery in human sclera. The biomechanical structure of the human sclera is influenced by a number of factors like young age, male gender, myopic eyes, eyes with primary filtering surgery done and use of antimetabolites like mitomycin c or 5-fluorouracil.² Post-trabeculectomy the IOP decreases and choroid thickens probably due to an increase in the intravascular volume. Studies have shown that eyes with hypotony maculopathy post-trabeculectomy with mitomycin c have shown to have greater reduction in axial length as compared with those without hypotony.³ Scleral shrinkage or low sclera rigidity especially in myopes is said to be a contributing factor. Myopic eyes have thinner sclera mostly at the posterior pole due to physical loss or alteration of extracellular matrix (ECM). The sclera of young myopes is more elastic, flexible and thinner than that of the older patients and is thought to shrink more in hypotony conditions. It facilitates the inward collapse of the scleral wall postsurgery resulting in chorioretinal folds. Human sclera consists of 90% collagen embedded in an ECM of proteoglycans. Proteoglycans have a core protein associated with glycosaminoglycan side chain (GAG). With ageing scleral rigidity increases due to loss of GAG, degeneration of collagen and scleral dehydration. Hence, the sclera is thinner and more vulnerable to collapse in event of hypotony. Usually, adult axial length is supposed to be reached by 13 years of age but in some cases of high myopia the axial length continues to increase thus causing thinning of sclera and the subsequent pathological changes.⁴ Scleral ECM remodelling is also thought to occur in ageing myopic eyes.⁵ These all increase the risk of hypotony maculopathy. Further, role of inflammation postsurgery cannot be undermined. Formation of a cyclitic membrane over the ciliary body postintraocular surgery or due to previous inflammation may cause hypotony. Intraoperative iridectomy

Table 1 Postoperative causes of hypotony

Overdrainage	Underproduction
Loose or breaking of flap sutures	Ciliary body shutdown due to inflammation
	Ciliochoroidal detachment
	Cyclodialysis cleft
	Antimetabolites (mitomycin c) toxicity

or improper visco wash leads to break in the blood–aqueous barrier causing subsequent ciliary body shut down and hypotony.

Use of antimetabolites in trabeculectomies cause reduced scar formation and increased filtration. This may prolong hypotony. But hypotony cannot be attributed to its antifibrotic property alone. Mitomycin-C (MMC) may have a direct toxic effect on

the ciliary body too.⁶ It induces conjunctival changes by making it more avascular. A study by Sihota *et al* in 2000 suggested that a dysfunctional conjunctival barrier and histopathological alterations in the epithelial barrier may lead to altered bleb morphology and subsequent hypotony.⁷

Contributors SS: preparation of manuscript, editing, literature search. SP: editing, review, analytical inputs, guarantor. BN: review, design.

Funding The authors have not declared a specific grant for this research from any funding agency in the public, commercial or not-for-profit sectors.

Competing interests None declared.

Patient consent for publication Parental/guardian consent obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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Patient's perspective

Parent's perspective: The thought of losing sight at such an early age for our son was very disturbing. After numerous visits to several hospitals, proper counselling and timely management helped salvage our child's vision. We intend to follow-up regularly and be compliant with the medications in future.

Learning points

- ▶ An uneventful trabeculectomy cannot ensure optimum pressure control each time.
- ▶ Patient's demography, biomechanical properties of the eyeball may modify the postoperative intraocular pressure and surgical outcome.
- ▶ Use of adjuvant antifibrotic agents should be made judiciously.

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