Patient creates filtering bleb: hypotony following traumatic rupture of an old scleral incision for intracapsular cataract surgery

Sophie Lemmens, Minh-Tri Hua, Ingeborg Stalmans

SUMMARY
An 86-year-old woman presented with symptomatic hypotony on the left eye since a few weeks, blurry vision and a very sensitive eye. She had a history of bilateral intracapsular cataract extraction (ICCE) in 1982 and secondary intraocular lens implantation in 1988. The patient mentioned a fall on the left side of the head 6 months earlier. The diagnosis of a superior scleral fistula was made, confirmed by gonioscopy and anterior segment optical coherence tomography. Direct surgical repair of the fistula led to a favourable outcome. This case demonstrates the occurrence of symptomatic hypotony due to the traumatic creation of a scleral fistula with an inadvertent filtering bleb many years after ICCE, and the resolution of signs and symptoms after surgical repair. Conventional as well as contemporary modalities can be valuable in the assessment of such fistulae. Management depends on the clinical course and the mechanism and extent of fistulation.

BACKGROUND
Cataract surgery has gone through a major evolution to become what it is today: the golden standard of refined, minimally invasive and safe phacoemulsification with placement of an artificial intraocular lens implant. The technique of phacoemulsification, introduced in the late 1960s, was preceded by conventional extracapsular and intracapsular cataract extraction (ICCE), both introduced in the second half of the 18th century. The first documented ICCE was performed in 1753 by Samuel Sharp.1 He created a large circumferential limbal incision and used his thumb to exert pressure on the globe to remove the lens and lens capsule from the eye in one piece. Obtaining zonulolysis is an indispensable part of the ICCE procedure and this step underwent several refinements. Application of alpha chymotrypsin in the 1950s and the development of a cryoprobe in the 1960s uplifted the elegance of this technique. The main difference between intracapsular and extracapsular techniques for cataract removal is the final destination of the capsular bag.2 As the capsular bag prevents the vitreous from prolapsing forward, sight-threatening complications such as retinal detachment, macular oedema and corneal decompensation are more likely to occur in ICCE. Moreover, the large incisions required for ICCE slow down healing and create significant surgically induced astigmatism. ICCE got outperformed by extracapsular techniques but despite its drawbacks, it remained the primary approach for cataract extraction in the western world, well into the 1970s, and modern ICCE is still in practice in developing countries.

We describe a case of a very late complication after ICCE, where a patient inadvertently created a scleral fistula with a filtering bleb and symptomatic hypotony.

CASE PRESENTATION
In February 2019, an 86-year-old Caucasian woman was referred to our centre by her ophthalmologist because of symptomatic hypotony on the left eye (OS) since a few weeks, with blurry vision and a very sensitive eye. She had a history of bilateral ICCE in 1982 because of quickly evolving subcapsular cataract, followed by secondary intraocular lens (IOL) implantation in 1988 OS and 1992 right eye (OD). No other ocular surgery had been reported. The patient mentioned a fall on the left side of the head with severe concussion in July 2018. At presentation, best corrected visual acuity (BCVA) was 20/22 OD and 20/40 OS. Intraocular pressure (IOP) was 15 mm Hg OD and 4 mm Hg OS on topical indomethacine two times per day and artificial tears since many years. Anterior segment examination showed bilateral anterior chamber IOL fixation with placement of an artificial intraocular lens (IOL) implantation in 1988 OS and 1992 OD. No other ocular surgery had been performed. The patient mentioned a fall on the left side of the head with severe concussion in July 2018. At presentation, best corrected visual acuity (BCVA) was 20/22 OD and 20/40 OS. Intraocular pressure (IOP) was 15 mm Hg OD and 4 mm Hg OS on topical indomethacine two times per day and artificial tears since many years. Anterior segment OCT showed bilateral anterior chamber IOL, as well as corneal guttata, endothelial pigment and a superior iridectomy. The right pupil had a pireform deformation and the left pupil showed a non-reactive semimydriasis. Some vitreum adhering to the IOL could be noted in the left eye. Additionally, the left eye unveiled a superonasal filtering bleb (figure 1A,B). The posterior pole of the right eye was without abnormalities, but the left fundus showed macular folds, confirmed by optical coherence tomography (OCT) (figure 1C), and a physiological optic nerve head and periphery on indirect ophthalmoscopy. The diagnosis of a superior scleral fistula was made. Because the patient mentioned a recent improvement of her reports, and because of the unpredictability of surgery, a wait-and-see attitude was first adopted. Two months later, the situation was re-evaluated. BCVA was 20/50 OS and IOP was 3 mm Hg OS. Biomicroscopic and funduscopic findings were unchanged.

INVESTIGATIONS
Gonioscopy showed a superonasal fistula in the anterior chamber angle (figure 1D), confirmed on anterior segment OCT despite poor image quality because of a significant degree of patient discomfort.
during image acquisition (figure 1E). B-scan ultrasound could not be performed successfully since deemed too uncomfortable by the patient. A causal relationship between the fall during the summer of 2018 and the presence of a scleral fistula seemed very likely, with the location of the fistula probably corresponding to a previously made surgical incision created while performing ICCE. The team of treating ophthalmologists decided to opt for surgical repair of the fistula, given the persisting symptoms and the risk of irreversible maculopathy in case of long-standing macular folds.

**TREATMENT**

Two months after initial presentation, closure of the scleral fistula in the left eye was performed under general anaesthesia to treat the patient’s symptomatic hypotony. After dissection of the superonasal conjunctiva, leakage of aqueous humour from a previously made scleral incision was clearly visible. The surgeon proceeded with the placement of three scleral sutures (Ethilon 10-0) to block the aqueous humour flow, followed by closure of the conjunctiva with four conjunctival sutures (one Ethilon 10-0 and three Vicryl 8-0). Postoperative treatment included acetazolamide 250mg two times per day for 3 days to decrease the production of aqueous humour and thus to release internal pressure on the fistula. Tobramycin and dexamethasone were administered four times per day for 1 month and the steroids tapered.

**OUTCOME AND FOLLOW-UP**

Two weeks after surgery, the patient achieved 20/33 BCVA and an IOP of 14 mm Hg in the left eye with resolution of the superonasal filtering bleb (figure 2A,B) and the macular folds (figure 2C), as shown on slit-lamp photography and OCT. On gonioscopy, the former scleral fistula appeared as a very discrete, narrow closing line (figure 2D). The patient was referred to her own ophthalmologist for further follow-up. Six months postoperatively, the clinical situation was stable.

**DISCUSSION**

Before the era of phacoemulsification, inadvertent filtering blebs due to scleral fistulation after cataract surgery were not uncommon. Yannuzzi and Theodore reported an incidence of 5% after ICCE, while others mentioned an incidence of inadvertent filtering blebs after cataract extraction between 1% and 5%. In 1979, Meredith and Theodore collected data on 1000 ICCE cases, and concluded that a younger age at the time of cataract surgery implied an increased risk of conjunctival bleb formation, while this risk was decreased by the use of monofilament nylon suture in a continuous closure. Inexact wound closure, abnormally thin limbal stroma and postoperative trauma were identified as predisposing factors for unplanned filtrations. Out of these, trauma to the eye is the only predisposing factor confined to the postoperative period. The introduction of phacoemulsification, small scleral tunnels and clear corneal incisions led to a rapid decline in the occurrence of post- cataract scleral fistulae with filtering blebs. However, these blebs still do appear every now and then, and are often due to intraoperative traumatic cyclodialysis, capsular bag shrinking in connection with ciliary body strain and leakage resulting from a persisting scleral tunnel. The case described here is the only one to our knowledge demonstrating an inadvertent filtering bleb after ICCE due to blunt trauma many years—37 to be precise—after the actual cataract surgery.

Regarding the management of such unintended scleral fistulae, several strategies have been adopted. One major strategy is a conservative attitude, which can be considered in the case of an asymptomatic bleb, or a symptomatic bleb persisting for no more than 3–6 months. A symptomatic fistula may cause irritation or visual disturbances as a result of bleb formation, astigmatism or hypotony possibly associated with several complications including chronic inflammation, anterior segment ischaemia, ciliochoroidal detachment, choroidal haemorrhage, maculopathy, epithelial ingrowth and phthisis. Additionally, the risk of delayed endophthalmitis should always be kept in mind.
Patient's perspective

I was referred by my treating ophthalmologist, where I presented with progressively worsening vision, and a very sensitive and uncomfortable left eye. From the start of the symptoms, I kept wondering whether this could have something to do with my fall with severe concussion 6 months earlier. I appreciated the joint decision-making in the treatment strategy and the comprehensive explanation my husband and I received from the medical staff, which could reassure me. During the weeks following the surgery, my vision gradually improved, and the discomfort decreased.

Learning points

- This case demonstrates the occurrence of symptomatic hypotony due to the traumatic creation of a scleral fistula with an inadvertent filtering bleb, many years after prior intracapsular cataract extraction (ICCE), and the resolution of clinical signs and symptoms after direct surgical repair of this fistula.
- Conventional techniques such as gonioscopy or B-scan ultrasound, as well as more contemporary modalities such as anterior segment optical coherence tomography, can be valuable in the diagnosis and structural analysis of such unintended scleral fistulae.
- Management depends on the patient’s reports, the duration and degree of overfiltration of aqueous humour, and the applied strategy needs to be individualised according to the mechanism and extent of fistulation.
- Because of the extensive time interval between ICCE and fistula formation, this case highlights the importance of thorough knowledge of obsolete surgical techniques and their potential long-term complications.

Approximately 80% of post-cataract blebs disappear spontaneously within 4 months after cataract surgery.1 This means that watchfully waiting may be the preferred approach in the majority of cases. However, a symptomatic bleb persisting for more than 4–6 months is generally regarded an indication for treatment.2,3 4 7 8 A wide variety of techniques for the repair of inadvertent filtering blebs have been described, such as the application of heat,9 trichloracetic acid,10 cryotherapy11 12 13 or continuous wave Nd:YAG laser14 to the conjunctiva, or administration of cycloplegics and/or topical or systemic corticosteroids.15

Surgical techniques include direct closure4 (as performed in the current case), closed external suture placement,16 corneal patch grafting,17 and direct excision of the fistula with or without a scleral donor patch.17

Acknowledgements The authors wish to express their gratitude for the additional clinical information they received from Dr Luc De Becker.

Contributors SL conceptualised the case description, examined the patient, acquired and interpreted the images, and drafted and finalised the manuscript. M-TH examined the patient, interpreted the images, performed the surgery and critically revised the manuscript. IS conceptualised the case description, examined the patient, interpreted the images and critically reviewed the manuscript. SL, M-TH and IS approved the submitted version, are accountable for the article, and ensure that all questions regarding the accuracy or integrity of the article are investigated and resolved.

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 Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

1 Hubbell AA, Sharp S. Samuel sharp, the first surgeon to make the corneal incision in cataract extraction with a single knife: a biographical and historical sketch. Med Library Hist 1904;2:242–68.
2 Davis G. The evolution of cataract surgery. Mo Med 1996;113:58.
3 Yannuzzi LA, Theodore FH. Cryotherapy of post-cataract blebs. Am J Ophthalmol 1973;76:217–22.
4 Swan KC, Campbell L. Unintentional filtration following cataract surgery. Arch Ophthalmol 1964;71:43–9.
5 Christensen RE, Rundle HL. Repair of filtering blebs following cataract surgery. Arch Ophthalmol 1970;84:8–11.
6 Meredith TA, Maumenee AE. A review of one thousand cases of intracapsular cataract extraction: II. visual results and astigmatic analysis. Ophthalmic Surg 1979;10:42–5.
7 Clinch TE, Kaufman HE. Repair of inadvertent conjunctival filtering blebs with a scleral flap. Arch Ophthalmol 1992;110:1652–3.
8 Jain SS. Inadvertent filtering bleb following sutureless cataract surgery. Indian J Ophthalmol 2005;53:196.
9 Machemer HF, Roters S. Ultrasound biomicroscopy of chronic hypotony after cataract extraction. J Cataract Refract Surg 2001;27:327–9.
10 Dunnington JH, Regan EF. Late fistulization of operative wounds. Arch Ophthalmol 1950;43:407–10.
11 Gehring JR, Ciccarelli EC. Trichloracetic acid treatment of filtering blebs following cataract extraction. Am J Ophthalmol 1972;74:622–4.
12 Douvas NG. Cystoid bleb cryotherapy. Am J Ophthalmol 1972;74:69–71.
13 Cleary GW, Fung WE, Webster RG. Cyoscopic closure of filtering blebs. Arch Ophthalmol 1972;87:319–23.
14 Geyer O, Large Mof. Management of large, leaking, and inadvertent filtering blebs with the neodymium:YAG laser. Ophthalmology 1998;105:983–7.
15 Shah VA, Majji AB. Ultrasound biomicroscopic documentation of traumatic cyclodialysis cleft closure with hypotony by medical therapy. Eye 2004;18:857–8.
16 Brunet VE, Edward Maumenee A, Stark WJ. A simple method of repairing inadvertent filtering blebs after cataract surgery. Am J Ophthalmol 1981;91:794–6.
17 Soong HK, Meyer RE, Wolter JR. Fistula excision and peripheral grafts in the treatment of persistent limbal wound leaks. Ophthalmology 1988;95:31–6.