Epithelial ingrowth of the bleb cavity, a true Tenon cyst, is a rare complication of a glaucoma drainage implant. Previous cases have been associated with persistent bleb leak, and most have occurred in eyes with prior extraocular surgery. We describe a case of a true Tenon cyst causing strabismus and an elevated intraocular pressure that was successfully treated by surgical revision.

Formation of an encapsulated bleb is a potential complication of glaucoma drainage implant devices (GDDs). Colloquially, an encapsulated bleb has been referred to by many ophthalmologists as a “Tenon’s cyst.” However, an encapsulated bleb is not a true cyst by histologic criteria (ie, an epithelial-lined fluid-filled cavity). An encapsulated bleb is a fluid-filled cavity with sclera and tense Tenon connective tissue comprising the inner wall of the cavity. We report the case of a true epithelial-lined cyst causing failure of a GDD and strabismus in a patient without a bleb leak.

REPORT OF A CASE

A 14-year-old girl sought care at the Bascom Palmer Eye Institute, Miami, Fla, for a large growth on her left eye. The growth had been present for approximately 6 months and caused an inward deviation of her eye and an unsightly appearance. She had a Baerveldt (350 mm²) GDD placed in the left eye 11 months prior to presentation at our institution. Her ocular history is significant for microcornea in the right eye, congenital rubella with congenital cataracts and glaucoma in both eyes, bilateral cataract extraction at age 6 months, esotropia and bilateral muscle surgery at age 2 and 7 years, macula sparing retinal detachment with subsequent pars plana vitrectomy, scleral buckling procedure, endolaser treatment, and silicone oil use in the right eye at age 12 years with removal of the oil 1 year later. She was using 0.5% timolol maleate, dorzolamide hydrochloride, 2% pilocarpine, and brimonidine tartrate in the right eye only.

The best-corrected visual acuity was 20/400 OD and 20/200 OS. Rotary nystagmus was present and motility is demonstrated in Figure 1. Slitlamp examination revealed a deep central chamber with iridocorneal touch medially and temporally in the right eye and mild corneal edema in the left eye. A large mass (Figure 2) that transilluminated easily was present over the previous GDD reservoir site. The drainage tube was not visualized. The intraocular pressure (IOP) was 54 mm Hg in the right eye and 40 mm Hg in the left by Goldmann applanation tonometry. Dilated fundus examination revealed a high scleral buckle with a flat retina and an old demarcation line in the right eye. Cup-disc ratio determination was difficult secondary to nystagmus and poor pupillary dilation, but was approximately 0.4 in the right eye and 0.8 in the left. The axial lengths were 22 mm in the right eye and 30 mm in the left. She was diagnosed with secondary chronic angle-closure glaucoma in both eyes and a giant encapsulated bleb in the left eye.

Because of the poor IOP control and motility abnormalities, surgical intervention was elected. Diode laser cyclophotocoagulation (13 spots over 270°) of the right
Intraoperatively, a fornix-based conjunctival flap was created and bluntly dissected off the cyst. Retroillumination and palpation of the cavity demonstrated the reservoir plate to be floating in the cyst. Rigid adherence to the sclera and an absence of a tissue plane prevented mobilization of the cyst. Clear fluid was present within the cyst. The wall of the cyst was incised and the entire cyst was removed and sent for pathologic examination. Retrograde irrigation of fluorescein-stained balanced salt solution through the tube using a 30-gauge cannula confirmed patency and location of the tube in the ciliary sulcus. The tube was tied off with a 7-0 Vicryl ligation suture as described by Trible and Brown, and the conjunctiva was closed with a running 8-0 Vicryl suture. The tube was left in place, since it is known that tubes in the posterior chamber perform better.2 Cryotherapy and cautery debridement were not done because epithelial downgrowth was not suspected. A forced duction test revealed marked restriction of the medial rectus muscle allowing only 10° of abduction.

Immediately after surgery, the patient’s visual acuity was 20/400 OD and 20/400 OS, and the IOP was 17 mm Hg in the right eye and 19 mm Hg in the left eye, while receiving 0.5% timolol, dorzolamide, brimonidine, prednisolone acetate, and atropine sulfate in both eyes. Six months after surgery, visual acuity was 20/200 OU, and the IOP was 16 mm Hg in the right eye and 5 mm Hg in the left eye while being treated with timolol and dorzolamide combination (Cosopt) in the right eye. External appearance and rotations are shown in Figure 3. Slitlamp, dilated fundus, and B-scan ultrasound examination findings were unchanged compared with the preoperative appearance.

PATHOLOGIC FINDINGS

Microscopic examination revealed a cystic structure lined by nonkeratinized stratified squamous epithelium consistent with conjunctival epithelium. No goblet cells were present. The wall of the cyst was also composed of dense fibrous connective tissue with apparent fibrosis compatible with bleb encapsulation (Figure 4).

COMMENT

In conjunction with postoperative antiglaucoma medications, GDDs have a 45% to 80% 2-year success rate for refractory pediatric glaucoma.3,4 In a retrospective review of patients with Molteno GDDs, Valimaki et al5 found that encapsulated blebs occurred in approximately 15% of their patients. When analyzed by diagnosis, 50% of their patients with congenital glaucoma developed bleb encapsulation. In adults, needling revision is effective at restoring IOP control in approximately 20% of patients with single-plate Molteno GDDs and 75% of patients with Baerveldt GDDs.6 Valimaki et al7 found that surgical revision with capsule excision restored IOP control in 75% of patients with Molteno GDDs. The cosmetically unacceptable appearance as well as the elevated IOP influenced our decision to elect surgical revision.

In our case, epithelial cells may have gained access to the capsule of the reseroir plate during the initial closure of the conjunctiva and Tenon layers. Epithelial proliferation resulted in the lining of the cyst wall with squamous epithelium causing a marked increase in the resistance of aqueous outflow. This may explain the elevated IOP despite the presence of a large bleb. Excision of the cyst wall allowed for reestablishment of adequate IOP control. This case is unique from the one previously reported case series of bleb cavity epithelial downgrowth for 2 reasons. Those cases were associated with an epithelialized conjunctival fistula causing a persistent wound leak and hypotony in eyes with concurrent or previous scleral buckling procedures. Our case had an elevated IOP and previous strabismus procedures, but no history of retinal surgery or fistulous tract in the affected eye. A recent randomized trial of Baerveldt GDDs in adults reported 2 of 107 eyes complicated by epithelial downgrowth.8 The authors did not specify if the downgrowth involved the bleb cavity or anterior chamber.

Orbital conjunctival cysts can be primary or secondary in origin. In a recent review,9 approximately two thirds of cases are secondary, with half of these occurring after enucleation. Of the remaining half, 90% were associ-
ated with previous strabismus or scleral buckling surgery typically performed within a few months before ocular surgery. However, patients may seek care decades later. Ninety percent of secondary conjunctival cysts are associated with ocular motility disturbances. Strabismus can occur following routine GDD surgery. With placement of the reservoir in the superior temporal quadrant, the patient may have some combination of exotropia and hypertropia. Our patient had a previously existing esotropia that was worsened by the mass effect of the cyst.

Epithelial ingrowth of the bleb capsule is a rare sequela of GDDs. It can be associated with hypotony or an elevated IOP. All of the reported cases have occurred in eyes with previous extraocular surgery.

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Corresponding author: Donald L. Budenz, MD, Bascom Palmer Eye Institute, University of Miami School of Medicine, 900 NW 17th St, Miami, FL 33136 (e-mail: dbudenz@med.miami.edu).

REFERENCES


