# Late leakage of filtering bleb in a patient with orbital pseudotumor

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PURPOSE. To report an unusual case of spontaneous late leakage of filtering bleb in a patient with orbital pseudotumor.

METHODS. Single case report.

RESULTS. A 53-year-old woman developed spontaneous leakage of bleb in her right eye 23 years after trabeculectomy with application of mitomycin-C (MMC). Two weeks later, her symptoms were blurring of vision, increasing redness, and dull ocular pain in the right eye. The inflammatory signs were suggestive of endophthalmitis, orbital cellulites, or pseudo-tumor. Absence of ophthalmoplegia, fever, and raised white cell count, together with the computed tomographic scan finding, confirmed the diagnosis of orbital pseudotumor. She responded well to oral steroids.

CONCLUSIONS. Orbital pseudotumor may initially present with spontaneous late leakage in a bleb augmented by MMC. Orbital pseudotumor should be added to the list of differential diagnoses when facing a patient with an inflamed, chemotic, proptotic eye in the presence of a late bleb leak. (Eur J Ophthalmol 2006; 16: 611-3)

KEY WORDS. Bleb leakage, Endophthalmitis, Pseudotumor

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#### INTRODUCTION

Late leakage of filtering bleb is a known complication of trabeculectomy, especially in those with augmentation by antimetabolites such as mitomycin-C (MMC) (1). Here we report an unusual case of spontaneous late leakage of filtering bleb in a patient with orbital pseudotumor. Without proptosis, the clinical picture of bleb leakage and inflammatory signs masqueraded as an endophthalmitis.

#### Case report

A 53-year-old Chinese woman had juvenile-onset open angle glaucoma and received unguarded Scheie-type full thickness sclerostomy in both eyes in 1976. A repeat trabeculectomy with MMC application was performed for failed filtration in the right eye in 1981, followed by a planned intracapsular cataract extraction (ICCE) in the right eye in 1987. In early 2004, best-corrected visual acuity (BCVA) of this aphakic eye was  $+8.25 -5.00 \times 145 = 0.1$ . Intraocular pressure (IOP) in the right eye was stable in the low teens without any medication. Vertical cup-to-disc ratio (VCDR) in the right eye was 0.8. The blebs in both eyes were reported to be cystic.

On November 1, 2004, the patient presented to our clinic with increasing discomfort over the right eye for a few days. The IOP of the right eye was elevated to 34 mmHg with mild injection and fibrosis around the cystic bleb. The internal ostium remained patent. IOP was lowered to the high teens with topical prostaglandin analogue (0.004% travoprost daily).

Two weeks later, she returned to us with a sudden-onset painless blurring of vision in her right eye. A spontaneous point leakage of bleb with a positive Seidel's sign



**Fig. 1** - *B*-scan ultrasonography of the right eye showing thickened posterior sclera.



**Fig. 2 -** Filtering bleb in the right eye remained cystic after cessation of leakage.



**Fig. 3** - Gonioscopy of the right eye revealed a patent internal ostium after cessation of leakage.

was detected. The IOP at that time was 0 mmHg, though the anterior chamber was formed and deep. An almost 360-degree shallow choroidal detachment was noted with no hypotonus maculopathy. The conjunctiva was mildly injected. The leakage was conservatively managed with eye padding and oral acetazolamide 250 mg qid. Over the next week the leakage sealed up itself with gradual resolution of the choroidal detachment.

At this point, the patient complained of increasing redness and dull ocular pain in the right eye. Within the same week, a 2-mm right proptosis developed, with prominent chemosis. At this juncture the differential diagnoses included right orbital pseudotumor, endophthalmitis related to bleb leakage, and orbital cellulitis. There were anterior chamber cells of grade 1 and no evidence of blebitis. The vitreous was relatively clear. The patient was afebrile with no ophthalmoplegia and normal white cell count. These made endophthalmitis and orbital cellulites less likely. One day later, B-scan ultrasonography of the right eye revealed scleral thickening and a clear vitreous (Fig. 1). Computed tomography of the orbit was performed and showed a right orbital soft tissue swelling and scleral thickening with no abscess or sinusitis. These clinical data were more suggestive of orbital pseudotumor. She was then treated with oral prednisolone 80 mg daily. Over a few days her proptosis, chemosis, and injection rapidly subsided with marked reduction in pain. In late December 2004, she needed three glaucoma medications over the right eye (0.5% timolol bd, 2% dorzolamide BID, and 0.005% latanoprost once in the evening to control her IOP at 10 mmHg. The bleb remained cystic (Fig. 2) with a patent internal ostium (Fig. 3). Her BCVA of the right eye remained at 0.1, and the VCDR 0.8. We offered the patient a conjunctival advancement with separation of peri-bleb fibrosis, to reduce the complication of a cystic bleb and to ensure adequate filtration. The patient declined the surgery.

## DISCUSSION

In the literature, spontaneous late bleb leakage was reported to occur after bouts of severe coughing in patients with chronic obstructive pulmonary disease (2) and a complete rupture of bleb was reported after eye rubbing during crying (3). To the authors' knowledge, this is the first reported case of spontaneous late leakage associated with orbital pseudotumor. Application of MMC was known to in-

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crease the chance of late bleb leakage (1). In our case as well as in most of the published case reports of spontaneous late leak or bleb rupture, the trabeculectomies were performed with antimetabolite application (2, 3).

One of the most serious consequences of a filtering bleb leak is a bleb-related infection (4, 5), which was reported to be over 25 times more likely to occur in a leaking than in a non-leaking filtering bleb (5). The concurrence of late bleb leak, proptosis, posterior scleritis, and vitreous haze posed some challenge in differentiating among orbital pseudotumor, orbital cellulitis, and bleb-related endophthalmitis. Careful consideration of all the clinical signs plus armamentarium such as B-scan ultrasound and computed tomography will help in the differentiation.

One may postulate that a possible increase in episcleral venous pressure may have occurred in the early phase of the inflammatory disease, which translates into an elevation of IOP. In our patient, the IOP was well maintained until weeks before the acute attack of inflammation. The inflammation might have also contributed to the peri-bleb fibrosis, which further decreased the drainage and elevat-

ed the IOP. There could be some component of trabecular inflammation which contributed to impeded aqueous outflow. The thin cystic wall finally would no longer withstand the grossly elevated pressure, which finally resulted in a late leakage.

In conclusion, orbital pseudotumor with rapidly progressive proptosis may present with spontaneous late leakage in a bleb augmented by MMC. One should include orbital pseudotumor on the list of differential diagnosis when facing a patient with an inflamed, chemotic, proptotic eye in the presence of a late bleb leak.

The authors have no financial or proprietary interest.

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