

Management of ocular surface squamous neoplasia extending up to a filtering trabeculectomy bleb

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A 73-year-old-gentleman was referred for ocular surface squamous neoplasia (OSSN) in his right eye (RE). He had history of combined cataract with trabeculectomy in RE and

was maintaining his intraocular pressure (IOP). He showed a corneoscleral lesion measuring 11 × 8 mm in nasal quadrant wherein, the superior edge of the lesion was extending up to the filtering bleb. After ruling out intraocular invasion or regional spread, he underwent complete tumor excision with “no touch” technique along with cryotherapy and surface reconstruction and a perilesional injection of Interferon α 2B. At 6-month visit, he shows no locoregional recurrence and has controlled IOP.

Key words: Bleb, ocular surface squamous neoplasia, perilesional interferon α 2B, surgical excision, trabeculectomy

Surgical excision has been considered the gold standard for management of ocular surface squamous neoplasia (OSSN).^[1-3] Topical chemotherapy with agents namely, Interferon α 2B (INF α 2B), Mitomycin- C (MMC), and 5-Fluorouracil (5FU) has been successfully used as primary or adjuvant therapy in treating OSSN in the last few years. Surgical excision is a good option in patients with lesions involving <4 limbal clock hours, rapidly growing lesions and those requiring rapid resolution of disease.^[4] Topical chemotherapy is preferred for

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large, multifocal lesions as it allows for complete resolution of disease with a decreased risk of limbal stem cell deficiency.^[5] The guidelines for management of OSSN in an eye with ocular co-morbidities are not extensively described in literature. Here, authors describe management of a novel case where the OSSN was extending up to a filtering trabeculectomy bleb. The patient consented to the use of his clinical photographs for academic and research purposes. This case report adheres to the ethical principles outlined in the Declaration of Helsinki 2013.

Case Report

A 73-year-old gentleman was referred for a suspected OSSN in RE noticed for 2 months. He was a known case of bilateral primary open angle glaucoma (POAG) without any significant systemic

medical illness. He had undergone cataract extraction (both eyes) with trabeculectomy procedure for his right eye about 17 years back. He was on topical antiglaucoma medication for his left eye and was maintaining IOP in right eye without medications.

Slit lamp examination of right eye revealed a 11 × 8 mm, partially pigmented corneo-conjunctival lesion in the nasal quadrant. The lesion was gelatinous with corneal extension, presence of surface keratin, intrinsic vascularity, prominent dilated feeder vessels as well as episcleral fixity [Fig. 1a]. The superior edge of the lesion was seen extending up to the filtering bleb at 11 o'clock from prior trabeculectomy [Fig. 2a]. Table 1 describes rest of the ocular and gonioscopy findings. There was no palpable lymphadenopathy. Anterior segment optical coherence tomography (AS-OCT) was performed [Fig. 1b].^[6] Though the tumor was found to extend till the edge of the bleb, the bleb itself was uninvolved by the tumor. The clinical diagnosis of OSSN in the right eye was hence made. The HIV serology along with other routine blood investigations were performed and found to be within normal limits.

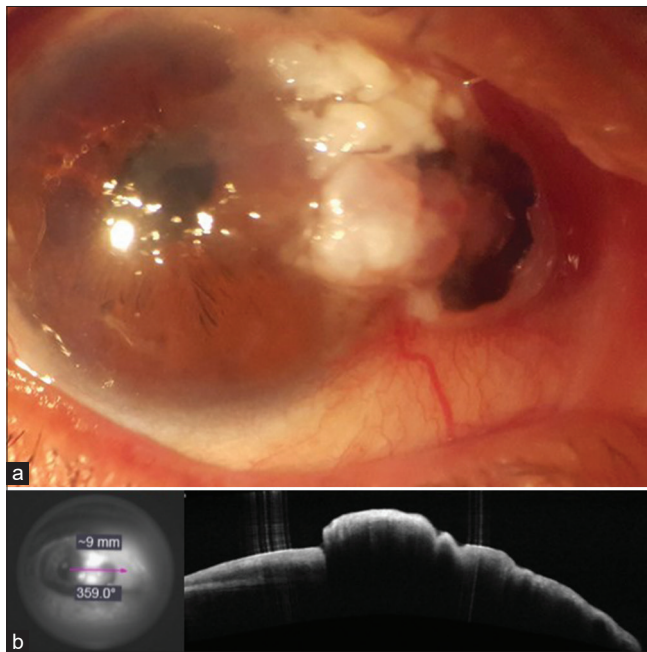


Figure 1: Clinical and AS-OCT findings; (a) Right eye, showing 11 × 8 mm partially pigmented lesion with surface keratin, corneal extension, intrinsic vascularity and dilated feeder vessels in nasal quadrant, trabeculectomy bleb is seen at 11 o'clock where the superior margin of lesion is seen, (b) AS-OCT showing hyper-reflective and thickened epithelium showing abrupt transition between normal and abnormal tissue

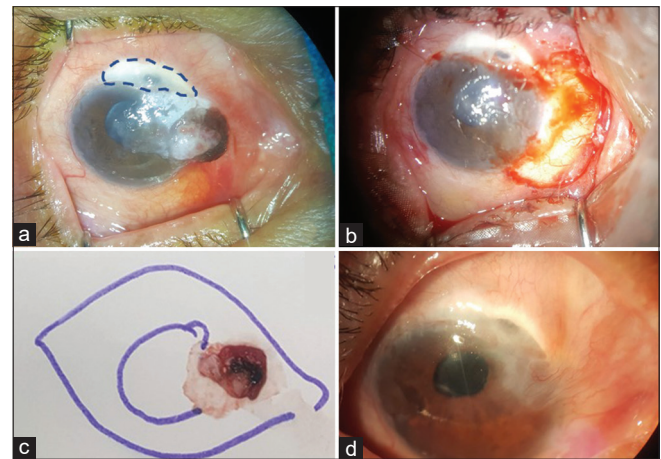


Figure 2: Intraoperative and post-operative images; (a) Pre-operative image with dotted blue line demarcating the area of conjunctival scarring and scleral thinning surrounding a filtering bleb, (b) After excision of the conjunctival lesion and alcohol assisted keratoepitheliectomy, (c) excised specimen mounted on a filter paper with rolled out margins which was sent to the pathologist in 10% formalin, (d) 8 weeks post-operative image of the eye showing complete tumor resolution and intact filtering bleb

Table 1: Ocular, intraocular pressure and gonioscopy findings

	Right Eye	Left eye
Best corrected visual acuity	20/200	20/40
Intraocular pressure (IOP) with applanation tonometry	14 mm Hg	14 mm Hg
Conjunctiva and Cornea	11 × 8 mm corneo-conjunctival lesion in nasal quadrant as described, trabeculectomy bleb at 11 o'clock.	Within normal limits
Iris	Normal	Normal
Pupil	Irregular, non-reacting	Irregular, partially reacting
Lens	Posterior chamber intraocular lens with posterior capsular opacity	Posterior chamber intraocular lens
Fundus:		
Posterior pole	C: D- 0.6	C: D- 0.7
Peripheral retina	Unremarkable	Unremarkable
Gonioscopy	360° open angles. No e/o angle involvement	360° open angles

The options for management

A. "No touch" surgical excision of the clinically visible tumor with a 4 mm clear conjunctival margin, 2 mm of clear corneal margin with alcohol assisted keratoepitheliectomy and lamellar sclerectomy and double freeze-thaw cryotherapy to the conjunctival margins and scleral base with ocular surface reconstruction using amniotic membrane graft.^[3]

B. Topical chemotherapy

The concerns for each option were, option A: the proximity of the filtering trabeculectomy bleb to the superior edge of the lesion would make it challenging to achieve 4 mm clear conjunctival margin without deroofting the functioning bleb. As for option B: the scleral thinning underlying the bleb was a risk factor for use of MMC. Moreover, the patient was not willing for frequent follow ups for long duration, required for of topical chemotherapy (INF α 2B).^[4]

After due considerations and explaining the patient about the pros and cons of available treatment modalities, he underwent a modified surgical excision under local anesthesia as explained above (option A) for all margins except for superior, where 1 mm margin was excised to avoid deroofting of the bleb. Cryotherapy was applied at the excised conjunctival edge without harming the bleb [Fig. 2b and c]. A perilesional injection of INF α 2B (3 million IU in 1 ml) was given in supero-nasal quadrant to take care of the possible residual tumor.^[7] Histopathology analysis confirmed presence of conjunctival intraepithelial neoplasia, grade 3- severe dysplasia (CIN) with negative margins of resection, closest (superior) being <1 mm. At follow up visit of 8 weeks, the wound was healed with intact filtering bleb and normal IOP [Fig. 2d]. He continues to do well at 6 months of follow up. There was no sign of local tumor recurrence, excessive scarring, symblepheron formation, significant rise in IOP or regional tumor spread on examination.

Discussion

When patient presents with OSSN, both surgical and medical approaches for tumor resolution should be considered. The authors describe a case of OSSN where the lesion was extending up to a functioning trabeculectomy bleb along with presence of conjunctival scarring and scleral thinning. Surgical excision alone had a risk of positive resection margins, whereas topical chemotherapy alone was not feasible in view of risk of further scleral thinning or melt (MMC) or longer treatment duration (INF α 2B). Thus, a combined approach of deliberate excision of lesser clinical margin in the area of the bleb with concomitant use of subconjunctival injection of INF α 2B in the same quadrant was utilized which resulted in complete resolution of tumor while preserving the filtering function of the bleb.

The importance of adequate surgical margins has been described in published studies with increased recurrence rates in those with incompletely excised tumors.^[8,9] Beneficial role of adjuvant interferon therapy (topical and/or perilesional/subconjunctival injection) in patients with high risk for recurrence after surgery as for those with positive margins, tarsal involvement, and recurrent disease has been discussed.^[10,11] After a detailed review of English literature with keywords "ocular surface squamous neoplasia,"

"OSSN," "trabeculectomy," or "bleb" the authors did not find a reported case of management of OSSN lesion involving a filtering trabeculectomy bleb. Hence, this case reports a novel presentation of OSSN with results of customized management protocol. The patient needs to be monitored for checking his IOP control and possibility of local recurrence in the future with a timely follow up schedule.

Conclusion

Surgical excision with adjuvant injectable interferon therapy can be applied in a case of OSSN for complete tumor resolution and to minimize the risk of recurrence in a challenging scenario where surgery alone would result in possible positive surgical margin. Judicious and customized use of available treatment options can aid in providing optimum tumor control with minimum side effects.

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Conflicts of interest

There are no conflicts of interest.

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