



Isolated primary sebaceous gland carcinoma of the bulbar conjunctiva

Kenneth Chang^{a,1}, Justin M. Wileman^{a,1}, Ryan D. Gabbard^{a,*}, Ryan J. Woodard^b,
Rakesh M. Patel^a, Shawn M. Iverson^a

^a University of South Carolina/Prisma Health, Columbia, SC, USA

^b University of South Carolina School of Medicine, Columbia, SC, USA

ARTICLE INFO

Keywords:

Sebaceous carcinoma
Bulbar conjunctiva
5-Fluorouracil
Cornea

ABSTRACT

Purpose: To report a rare case of primary sebaceous gland carcinoma of the bulbar conjunctiva without palpebral involvement.

Observation: A 76-year-old male was referred to the cornea service for a suspicious lesion on the limbal conjunctiva and cornea of his left eye. On exam, there was a large fibrovascular growth with conjunctivalization of the cornea from 2:00 to 10:00 extending 6mm onto the cornea. Patient underwent treatment with 5-fluorouracil, followed by excisional biopsy with cryotherapy. Histopathologic examination demonstrated poorly differentiated sebaceous gland carcinoma. Subsequent map biopsies of the palpebral and bulbar conjunctiva of the left eye were performed and ruled out pagetoid spread.

Conclusions and Importance: Primary sebaceous gland carcinoma of the bulbar conjunctiva without eyelid involvement is a rare phenomenon. Our patient joins a few case reports in the literature. Of these cases, we are the second case to utilize 5-fluorouracil preoperatively. We recommend clinicians consider sebaceous carcinoma on their differential when ocular surface neoplasms diagnosed as other conditions do not respond to conventional therapies.

1. Introduction

Sebaceous carcinoma is an aggressive malignancy that develops from sebaceous glands, most commonly originating from meibomian glands within the eyelid. Primary sebaceous carcinomas of the bulbar conjunctiva sparing the palpebral conjunctiva are extremely rare with only a few cases reported. We present a 76-year-old male who presented with a large fibrovascular growth with conjunctivalization of the cornea. Initial medical management for presumed ocular surface squamous neoplasia consisted of using multiple rounds of 5-fluorouracil preoperatively, followed by wide excision and cryotherapy to the conjunctival margins. Histopathological analysis demonstrated poorly differentiated sebaceous carcinoma. The collection and evaluation of protected patient health information of this patient were HIPAA compliant. This case report also adhered to the ethical principles outlined in the Declaration of Helsinki as amended in 2013.

2. Case presentation

A 76-year-old male was referred to our clinic for a pterygium in his left eye that was suspicious for squamous cell carcinoma. Patient reported symptoms including ocular redness and tearing for approximately one year. He denied pain, discomfort, or photophobia. His medical history was significant for diabetes mellitus, hypertension, atrial fibrillation, and coronary artery disease status post coronary artery bypass graft. He denied any significant ocular history or trauma. Additionally, he denied any known history of malignancy. On exam, visual acuity (VA) was 20/60 in the right eye (OD) and 20/100 in the left eye (OS). Systemic survey was unremarkable, and there was no evidence of enlarged preauricular or submandibular lymph nodes. Intraocular pressure was 23 OD and 22 OS. Slit lamp exam demonstrated diffuse injection OS with a large fibrovascular growth with conjunctivalization of the cornea from 2:00 to 10:00, thickest from 4:00 to 9:00, extending 6mm onto the cornea. The lesion also stained with fluorescein centrally (Fig. 1). The presumed diagnosis was ocular surface squamous

* Corresponding author. 4 Medical Park Rd, Columbia, SC, 29203, USA

E-mail addresses: kennethchang0@gmail.com (K. Chang), Justinwileman@yahoo.com (J.M. Wileman), gabbardsc42@gmail.com (R.D. Gabbard), Ryan.Woodard@uscmed.sc.edu (R.J. Woodard), Rakesh.Patel2@prismahealth.org (R.M. Patel), shawn.iverson2@prismahealth.org (S.M. Iverson).

¹ Contributed equally to this work.

<https://doi.org/10.1016/j.ajoc.2022.101675>

Received 11 April 2022; Received in revised form 24 June 2022; Accepted 22 July 2022

Available online 31 July 2022

2451-9936/© 2022 Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).



Fig. 1. Gross view of the sebaceous carcinoma involving the inferior limbus and inferonasal cornea.

neoplasia.

Treatment with topical 5-fluorouracil (5-FU) was started in attempt to treat or at least reduce the lesion size. Four cycles, each consisting of 1 week of 5-FU four times daily followed by a 3-week holiday, were completed. Despite an initial decrease in tumor size, the mass stopped responding to treatment and the conjunctiva remained persistently boggy. An excisional biopsy was performed in the operating room. The lesion dissected off the corneal surface en bloc. This was followed by cryotherapy to the conjunctival margins. Amniotic membrane was grafted to the conjunctival defect. The tumor was sent fresh for histopathologic analysis (Fig. 2). Immunohistochemical stains were positive for adipophilin (Fig. 3), AE1/AE3, CAM 5.2, and Ber-EPF and negative for CD3, CD20, CK20, S100, GATA3, CEA, GPAP, and mucicarmine; consistent with poorly differentiated sebaceous carcinoma. The patient was referred to an oculoplastic surgeon for conjunctival map biopsies to rule out pagetoid spread. Five biopsies each of the left upper and lower palpebral conjunctiva, along with four biopsies of the bulbar conjunctiva all returned negative for malignancy postoperatively.

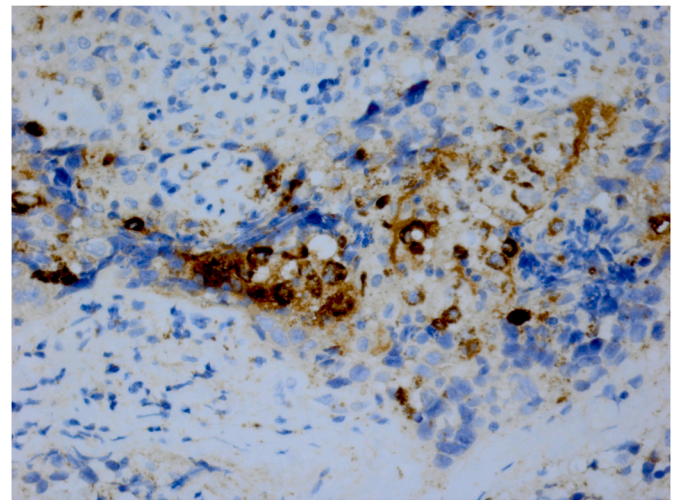


Fig. 3. Pathology slide demonstrating immunohistochemical staining positive for adipophilin (brown) at 100x magnification. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

No recurrence was seen at the 1-month follow-up. Extraocular movements remained full, and VA was unchanged from preoperative assessment. Slit lamp examination demonstrated trace residual boggy conjunctiva along the 6-8:00 limbus. There was no conjunctival staining or recurrent growth onto the cornea.

At the 9-month follow-up, the patient did not report any visual changes in either eye. The extraocular movements and VA remained unchanged from the preoperative assessment. IOP was 18 OU. Slit lamp examination was negative for symblepharon formation or concerning lesions on lids; however, a growth was visualized on the conjunctiva extending 1.7 mm inferiorly onto the cornea, concerning for a pseudopterygium or possible early recurrence (Fig. 4). The patient will continue to be followed in the clinic to monitor for potential recurrence.

3. Discussion

Sebaceous gland carcinoma accounts for 3.2% of eyelid malignancies and only 0.8% of all eyelid tumors, and rarely presents without lid involvement. Sebaceous carcinomas typically occur in the periocular region, head, and neck, but can appear anywhere in the body where

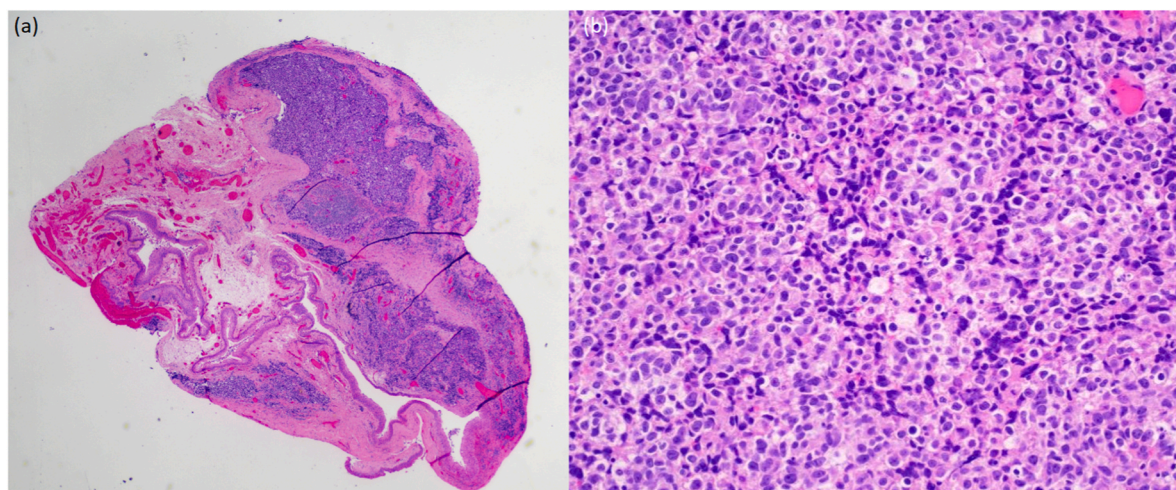


Fig. 2. (a) Histopathology slide demonstrating sebaceous carcinoma on the right with normal sebaceous glands present on the left portion of the slide at 2x magnification. (b) High power magnification (40x) view illustrating the mitotic figures (deep purple) of the tumor. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

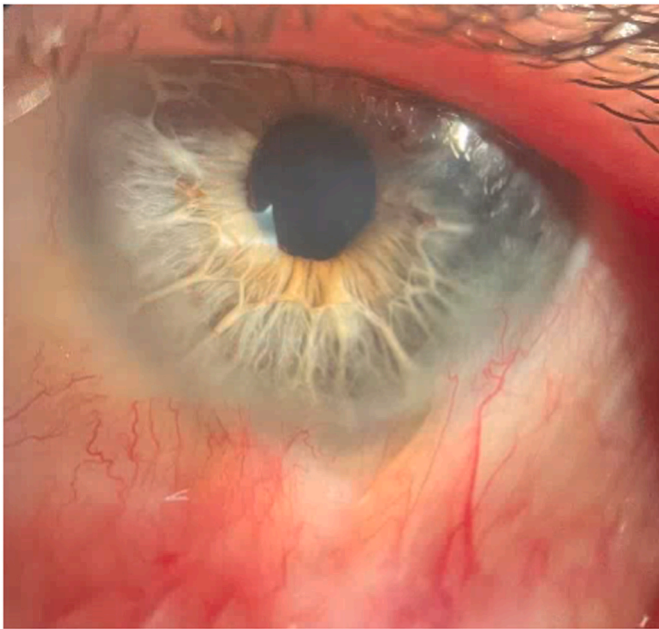


Fig. 4. Slit lamp photograph from the 9-month follow-up showing a growth extending inferiorly onto the cornea, concerning for a pseudopterygium or possible early recurrence.

sebaceous cells are present.¹ They typically originate in the upper eyelid due to the high number of meibomian glands present in this region with a prevalence of approximately 50%. Sebaceous carcinomas can be sporadic in nature or associated with Muir-Torre syndrome, a genetic condition characterized by sebaceous carcinomas and other internal cancers with colon cancer being the most common.² Sebaceous carcinomas can be difficult to diagnose as it often masquerades as a variety of conditions such as chalazion, blepharoconjunctivitis, sebaceous cyst, seborrheic keratosis, squamous papilloma, and other eyelid tumors such as basal cell and squamous carcinomas.³ Therefore, the diagnosis of sebaceous carcinomas should be suspected in elderly patients with a history of chalazion or unilateral blepharoconjunctivitis that does not respond to conventional therapies. One of the hallmark features of sebaceous carcinoma is that it tends to spread superficially in a pagetoid fashion, in contrast to basal cell and squamous cell carcinomas which typically spread radially.⁴

The mainstay of treatment is surgical excision and obtaining map biopsies of conjunctiva. Orbital exenteration may be indicated in invasive cases. Histopathology demonstrates sebaceous cells and undifferentiated cells that may mimic other carcinomas of the eyelid. Special stains can be utilized to distinguish sebaceous carcinomas that include oil-red O, Sudan IV, epithelial membrane antigen, and Leu-m1 immunostains.² Conjunctival epithelial involvement may be treated with cryotherapy, topical mitomycin C, 5-fluorouracil, or radiation therapy.⁵ Local recurrence rates can range from 11 to 23%; therefore, routine monitoring postoperatively is warranted.⁶

Primary sebaceous carcinomas of the bulbar conjunctiva are extremely rare with only a few cases noted in literature. Honavar et al. reported a 33-year-old female who had been previously treated for chronic blepharoconjunctivitis and was found to have a solitary conjunctival sebaceous carcinoma that underwent surgical excision and cryotherapy.⁷ Park et al. demonstrated that sebaceous carcinomas can present as multiple small corneoscleral masses.⁸ Margo and Grossniklaus reported two cases of intraepithelial sebaceous carcinoma involving the tarsal and bulbar conjunctiva.⁹ Only one case of primary bulbar conjunctival sebaceous carcinoma had been treated preoperatively with 5-fluorouracil.¹⁰

The purpose of this case report is to provide further evidence that

isolated sebaceous gland carcinomas should be included on the differential when ocular surface conditions do not respond to standard therapies. Additionally, routine postoperative monitoring is advised due to the high local recurrence rates of these carcinomas.

4. Conclusion

In summary, we present a rare case of primary sebaceous carcinomas of the bulbar conjunctiva sparing the palpebral conjunctiva.

Financial Support

None.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Disclosures

Kenneth Chang MD¹, kennethchang0@gmail.com – no disclosures.
Justin M. Wileman DO¹, Justinwileman@yahoo.com – no disclosures.

Ryan D. Gabbard MD¹, gabbardsc42@gmail.com – no disclosures.

Ryan J. Woodard BS², Ryan.Woodard@uscmed.sc.edu – no disclosures.

Rakesh M. Patel MD¹, Rakesh.Patel2@prismahealth.org – no disclosures.

Shawn M. Iverson DO¹, shawn.iverson2@prismahealth.org – no disclosures.

Acknowledgements and disclosures

No funding or grant support.

Patient consent

Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the identification of the patient.

Declaration of competing interest

The following authors have no financial disclosures.

Acknowledgements

None.

References

- Dasgupta T, Wilson LD, Yu JB. A retrospective review of 1349 cases of sebaceous carcinoma. *Cancer*. 2009;115(1):158–165.
- Buitrago W, Joseph AK. Sebaceous carcinoma: the great masquerader: emerging concepts in diagnosis and treatment. *Dermatol Ther*. 2008;21(6):459–466.
- Song A, Carter KD, Syed NA, Song J, Nerad JA. Sebaceous cell carcinoma of the ocular adnexa: clinical presentations, histopathology, and outcomes. *Ophthalmic Plast Reconstr Surg*. 2008;24(3):194–200.
- Shields JA, Demirci H, Marr BP, Eagle Jr RC, Shields CL. Sebaceous carcinoma of the eyelids: personal experience with 60 cases. *Ophthalmology*. 2004;111(12):2151–2157.
- Owen JL, Kibbi N, Worley B, et al. Sebaceous carcinoma: evidence-based clinical practice guidelines. *Lancet Oncol*. 2019;20(12):e699–e714.
- Orcurto A, Gay BE, Sozzi WJ, Gilliet M, Leyvraz S. Long-term remission of an aggressive sebaceous carcinoma following chemotherapy. *Case Rep Dermatol*. 2014;6(1):80–84.
- Honavar SG, Shields CL, Maus M, Shields JA, Demirci H, Eagle Jr RC. Primary intraepithelial sebaceous gland carcinoma of the palpebral conjunctiva. *Arch Ophthalmol*. 2001;119(5):764–767.

8. Park SJ, Wee WR, Lee JH, Kim MK. Primary sebaceous carcinoma of the corneoscleral limbus with pagetoid recurrence. *Kor J Ophthalmol*. 2009;23(2):104–107.
9. Margo CE, Grossniklaus HE. Intraepithelial sebaceous neoplasia without underlying invasive carcinoma. *Surv Ophthalmol*. 1995;39(4):293–301.
10. Pandey K, Singh P, Singh A, Pandey H. Primary sebaceous gland carcinoma of the bulbar conjunctiva without involvement of eyelid: a clinical dilemma. *Oman J Ophthalmol*. 2011;4(2):97–99.