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CASE REPORT

Sebaceous Cell Carcinoma of the Lower Eyelid in an Elderly Male: a Rare Case Report and Review of Literature

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Introduction

Sebaceous cell carcinoma (SCC) arises from the sebaceous gland and it is considered as one of the aggressive dermatological malignancy. It may have an ocular as well as extraocular origin however, the most common site is the ocular region with the accountability of 34–59% cases [1]. The most common site of ocular origin is the upper eyelid due to the dense concentration of meibomian glands and involvement of the lower eyelid is a very rare presentation [2]. However simultaneous involvement of both eyelids has been found in about 1–6% of cases [3]. It is considered the third most common malignancy in the USA after basal cell carcinoma (BCC) followed by squamous cell carcinoma (SCC) which is contrary to Asian people in whom it is more common than BCC with accountability of around 28–60% of all ocular malignancies [4]. SCC is most commonly found in elderly patients with age range from 70 to 72 with no gender differentiation [5].

The origin of SCC is found in the meibomian gland, caruncle, gland of Zeis, and skin of the eyebrow [6]. There are other ocular pathologies including both benign and malignant entities which may mimic SCC. These are chalazion, BCC, squamous cell carcinoma, ocular pemphigoid, conjunctivitis, blepharitis, and leukoplakia. Thus, during evaluation of SCC, all these pathologies should be addressed first and then definitive management of SCC can be planned. We are reporting a case of an elderly male who presented to us with post excision status with positive margins who underwent re-wide excision (Fig. 1).

Case Report

A 68-year-old gentleman with Eastern Cooperative Oncology Group (ECOG) Performance Status I with no comorbidity presented to our clinic with history of post excision status of right lower lid lesion. The histopathology report was suggestive of poorly differentiated carcinoma possibility of sebaceous cell carcinoma. We extracted all relevant history in relation with pre-surgery complaints, investigations and surgery details. Discharge summary was available where all relevant findings were already documented. There was no positive history of redness of eyes, loss of eyelashes, pain, trauma, discharge or history of radiation in the past. There was no significant medical and family history or hospitalization for any major illness in the past. The patient was evaluated by an ophthalmologist for chalazion and advised him to undergo excision. The patient underwent wide local excision and after getting a final histopathology report he was referred to our clinic.

On clinical examination, a scar mark was noted just near the midline of the lower eyelid with no palpable parotid, cervical lymph nodes. The rest of the systemic examination was unremarkable. The findings on the pathology report mentioned specimen size of 0.5 cm ×0.5cm. The size of the lesion was not mentioned but margins were involved by the tumor. We evaluated the patient again with review of the pathology report by our

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Fig. 1 Scar mark (post excision) over the right lower eyelid

expert team of oncopathologist. We did ultrasound of the neck for cervical lymph node status and contrast-enhanced computed tomography (CECT) of the neck, chest, abdomenand pelvis for metastatic staging work up. Our reviewed histopathology study reported presence of nodular or diffuse growth pattern surrounded by a dense fibrous stroma with infiltrative borders which was highly suggestive of poorly differentiated sebaceous carcinoma (Fig. Fig. 2). The radiology report was negative for enlarged parotid or neck nodes with no distant metastasis.

After discussing index case in our multidisciplinary tumor board, a decision was taken to plan for re-wide excision of the scar with adequate margins with reconstruction. Hence, after getting fitness from the anesthetist. We did re-wide excision of the scar with modified Hughes flap. The patient was discharged on the next postoperative day with uneventful postoperative course. Again for adjuvant treatment, there were conflicting evidences. Hence, our multidisciplinary tumor board decided adjuvant radiation therapy in view of poorly differentiated histology. Post radiation therapy, patient developed eyelid erythema, edema, conjunctival congestion and chemosis for the next 4 months which were resolved with symptomatic treatment. After 5th month of completion of treatment there was complete loss of eyelashes. The patient is still in periodic follow-up with us and after 18 months of completion of treatment, he is disease free.

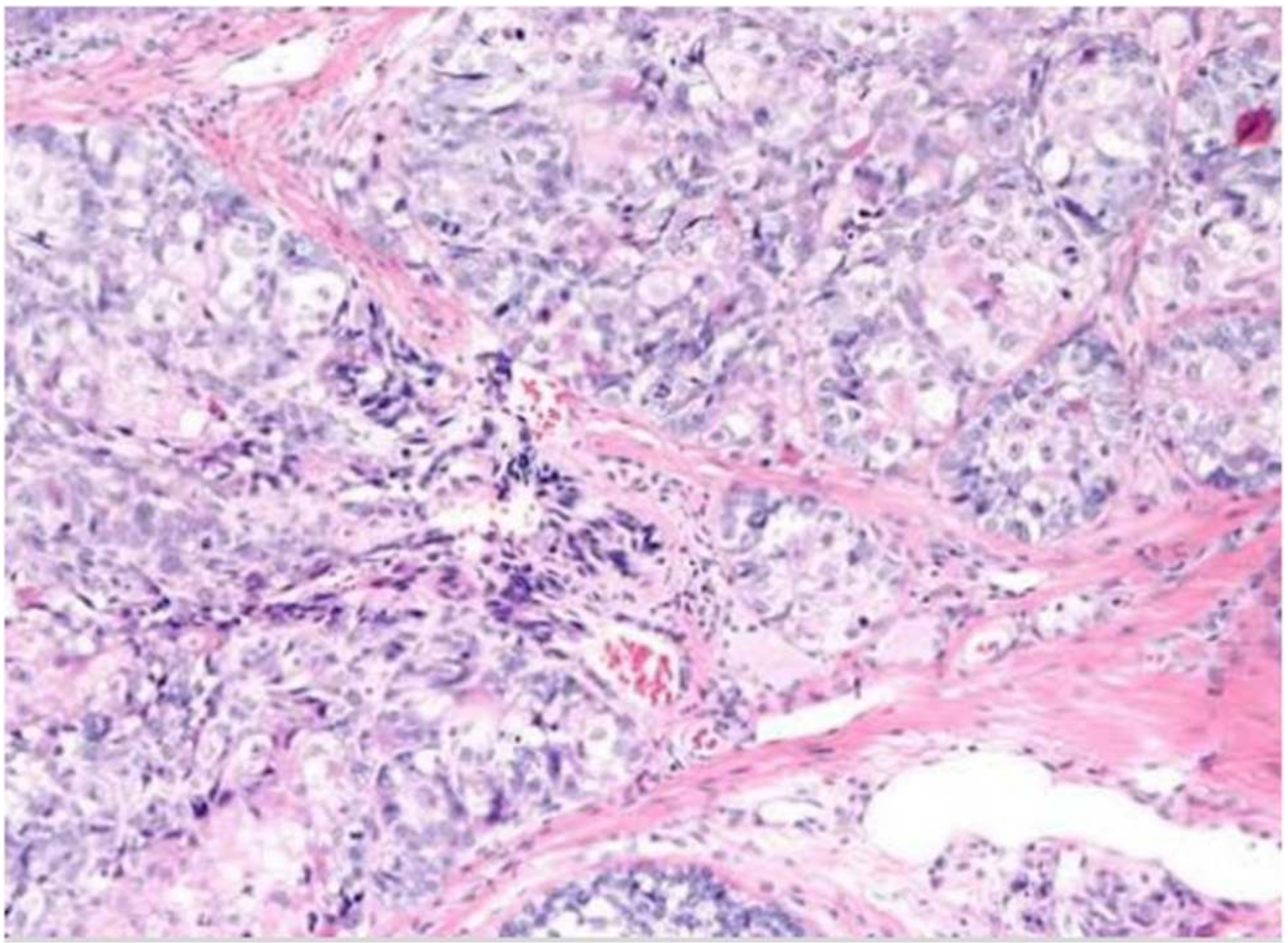

Fig. 2 Microscopic view of Sebaceous cell carcinoma

Discussion

There is scarcity of literature about the etiopathogenesis and the risk factors of SCC. The possible etiology includes immunosuppressive status, history of radiation treatment in the past, some syndromic association with Muir–Torre syndrome, production of carcinogen like nitrosamines and photosensitization because of use of diuretics in the past [7]. About 80% of SCC involves the skin of the head and neck with near about 40% involvement of the eyelids [5]. As we have already mentioned, several entities needs differentiation while ruling out SCC. The typical presentation of SCC of the eyelid is a solitary nodule which may be painless with some diffuse inflammation or sometimes with loss of cilia. The clinical picture of this finding mimics the presentation of chalazion. The second most common presentation of SCC is with diffuse unilateral thickening of the eyelid which simulates the common features of blepharoconjuctivitis.

The extra-ocular site of SCC is located over the head, neck, trunk, extremities and extracutaneous locations are the parotid gland, nasal cavity, breast, large bowel, ovary and prostate [8]. The local spread of SCC involves the palpebral conjunctiva, bulbar conjunctiva and cornea. In advanced cases, it may involve the orbital soft tissue, bone, and intracranial cavity. Metastatic spread involves the regional lymph nodes [9]. The upper eyelid tumor spreads to preauricular and parotid lymph nodes while the lower eyelid tumor spreads to submandibular and cervical lymph nodes. Distant metastasis may involve the parotid gland, liver, lung and bone [10].

The diagnosis of SCC is suspected when an elderly patient presents with history of unilateral blepharoconjunctivitis or recalcitrant chalazion which is resistant to standard ophthalmic treatment. The diagnosis is only confirmed with excision biopsy. Immunohistochemistry (IHC) is required to differentiate it from BCC and squamous cell carcinoma. Poorly differentiated SCC shows both, squamous and sebaceous differentiation. This finding makes it difficult to differentiate it from squamous cell carcinoma. There are some IHC markers which rules out SCC from other varieties. SCC is near about 100% positive for the following IHC markers—epithelial membrane antigen (EMA), adipose differentiation-related protein (ADP) and androgen receptor (AR) [11].

The differential diagnosis of SCC includes BCC, squamous cell carcinoma, benign sebaceous lesions, Merkel cell carcinoma and inflammatory lesions. The best way to differentiate these pathologies is by excision biopsy. BCC lesions are mostly ulcerative and SCC does not have ulceration. IHC markers EMA and ADP are negative in BCC and the same are positive in SCC. EMA may be positive in squamous cell carcinoma however, ADP and androgen receptor (AR) are consistently negative whereas these are positive in SCC. Benign lesions do not involve the periocular region. Inflammatory lesions mostly respond to standard ophthalmic treatment. Merkel cell carcinoma presents with red lesions over the eyelid and requires histopathological examination for diagnosis [12].

Patients with SCC should be examined carefully to define the clinical extent of the tumor and regional lymph nodes should be palpated properly. If an enlarged node is found, it should be posted for biopsy or fine-needle aspiration cytology. The diagnosed patient should be referred to an ophthalmologist for evaluation of conjunctival and corneal involvement. Radiological investigations to define the local extent and metastatic staging includes computerized tomography (CT) or magnetic resonance imaging (MRI) which are useful in evaluating invasion of orbital soft tissue, bone or intracranial cavity.

Surgical treatment is the first-line treatment for SCC and it includes wide local excision of the lesion with frozen section control or if available, Mohs micrographic surgery [13]. When conjunctiva gets involved, it may require surgical resection of the bulbar epithelium, cryotherapy or topical mitomycin. When there is extensive orbital involvement, it requires orbital exenteration. The role of radiation treatment for SCC is still not clear. Use of upfront radiation therapy has been reported by some studies [14]. Adjuvant treatment with all available modalities has been reported by few case series however, still there is no supportive strong evidence. When there is pagetoid invasion of conjunctiva, cryotherapy is frequently used as an adjuvant therapy but it has some side effects like symblepharon and corneal erosions [15]. Topical mitomycin has been used in few cases of pagetoid invasion of conjunctiva as an adjuvant treatment but it has side effects like persistent keratoconjunctivitis and epiphora [16]. When there is regional nodal involvement, it requires nodal dissection or radiation therapy [17]. There are isolated reported cases where cisplatin, capecitabine and fluorouracil were used successfully as a topical agents and radiation therapy for metastatic SCC [18]. Inadequate excision of the tumor is the most common factor responsible for local recurrence which in turn affects the prognosis. Local recurrence has been reported in 9–36% of patients and distant metastasis reported in 3–25%. Older age, poorly differentiated tumors and distant but not nodal metastasis are unfavorable prognostic factors [19]. Hence, all patients with SCC should be followed with extended long-term follow-up protocol as late relapses had been reported after 5–11 years [20].

Conclusion

Sebaceous cell carcinoma is considered a rare and highly aggressive tumor of the ocular origin. The diagnosis is confirmed with wide excision biopsy and requires

long-term periodic follow-up to prevent local recurrence and distant metastasis.

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Clinical Trial Transparency Not applicable

Declarations

Ethics Approval and Consent to Participate Not applicable

Informed Consent Informed consent was obtained from the patient for being included in the study.

Consent for Publication An informed consent to publish this case was obtained from the patient.

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Conflict of Interest The authors declare no competing interests.

References

1. Tryggvason G, Bayon R, Pagedar NA (2012) Epidemiology of sebaceous carcinoma of the head and neck: implications for lymph node management. Head Neck 34(12):1765–1768
2. Mahipathy SR, Durairaj AR, Kothandaraman K, Rajamanohar VC, Prabakaran A (2016) Sebaceous gland carcinoma of the lower eyelid reconstructed with a composite flap: a case report. J Clin Diagn Res 10(6):PD16–PD18
3. Bernardini FP (2006) Management of malignant and benign eyelid lesions. Curr Opin Ophthalmol 17(5):480–484
4. Shields JA, Demirci H, Marr BP, Eagle RC Jr, Shields CL (2005) Sebaceous carcinoma of the ocular region: a review. Surv Ophthalmol 50(2):103–122
5. Dasgupta T, Wilson LD, Yu JB (2009) A retrospective review of 1, 349 cases of sebaceous carcinoma. Cancer. 115(1):158–165
6. Pe'er J (2016) Pathology of eyelid tumors. Indian J Ophthalmol 64(3):177–190
7. Stockl FA, Dolmetsch AM, Codere F, Burnier MN Jr (1995) Sebaceous carcinoma of the eyelid in an immunocompromised patient with Muir-Torre syndrome. Can J Ophthalmol 30(6):324–326
8. Gnepp DR (2012) My journey into the world of salivary gland sebaceous neoplasms. Head Neck Pathol 6:101–110
9. Sawyer AR, McGoldrick RB, Mackey S et al (2009) Should extraocular sebaceous carcinoma be investigated using sentinel node biopsy? Dermatol Surg 35:704–708
10. Husain A, Blumenschein G, Esmaeli B (2008) Treatment and outcomes for metastatic sebaceous cell carcinoma of the eyelid. Int J Dermatol 47:276–279
11. Plaza JA, Mackinnon A, Carrillo L, Prieto VG, Sangueza M, Suster S (2015) Role of immunohistochemistry in the diagnosis of sebaceous carcinoma: a clinicopathologic and immunohistochemical study. Am J Dermatopathol 37:809–821
12. Mulay K, White VA, Shah SJ, Honavar SG (2014) Sebaceous carcinoma: clinicopathologic features and diagnostic role of immunohistochemistry (including androgen receptor). Can J Ophthalmol 49:326–332
13. Hou JL, Killian JM, Baum CL, Otley CC, Roenigk RK, Arpey CJ, Weaver AL, Brewer JD (2014) Characteristics of sebaceous carcinoma and early outcomes of treatment using Mohs micrographic surgery versus wide local excision: an update of the Mayo Clinic experience over the past 2 decades. Dermatol Surg 40:241–246
14. Hata M, Koike I, Maegawa J, Kaneko A, Odagiri K, Kasuya T, Minagawa Y, Kaizu H, Mukai Y, Inoue T (2012) Radiation therapy for primary carcinoma of the eyelid: tumor control and visual function. Strahlenther Onkol 188:1102–1107
15. Russell HC, Chadha V, Lockington D, Kemp EG (2010) Topical mitomycin C chemotherapy in the management of ocular surface neoplasia: a 10-year review of treatment outcomes and complications. Br J Ophthalmol 94:1316–1321
16. Khong JJ, Muecke J (2006) Complications of mitomycin C therapy in 100 eyes with ocular surface neoplasia. Br J Ophthalmol 90:819–822
17. Deo SV, Shukla NK, Singh M et al (2012) Locally advanced sebaceous cell carcinoma (T3) of eyelid: incidence and pattern of nodal metastases and combined modality management approach. Orbit 31:150–154
18. Jung YH, Woo IS, Kim MY, Han CW, Rha EY (2016) Palliative 5-fluorouracil and cisplatin chemotherapy in recurrent metastatic sebaceous carcinoma: case report and literature review. Asia Pac J Clin Oncol 12:e189
19. Thomas WW, Fritsch VA, Lentsch EJ (2013) Population-based analysis of prognostic indicators in sebaceous carcinoma of the head and neck. Laryngoscope 123:2165–2169
20. Park SJ, Wee WR, Lee JH, Kim MK (2009) Primary sebaceous carcinoma of the corneaoscleral limbus with pagetoid recurrence. Korean J Ophthalmol 23:104–107