Sebaceous gland carcinoma of eyelid in young boy: A rare case report and review of literature

Abstract: The sebaceous gland carcinoma of the eyelid is highly malignant with a high rate of local recurrence, regional and distant metastasis. It is a slow-growing tumor arising from Meibomian glands located in the tarsal plate, glands of Zeiss, sebaceous glands of the caruncle, and periocular skin. It usually occurs in elderly women and can be frequently misdiagnosed as chalazion or blepharitis or blepharoconjunctivitis. Herein, we present a case of sebaceous carcinoma of the upper eyelid in a young male previously incised and curetted for several times but without improvement and had to undergo wide local excision double freeze-thaw cryotherapy in all margins of skin and conjunctiva and reconstruction of the eyelid using cutler beard technique with a favorable outcome. There were no signs of local recurrence, regional or distant metastasis after surgery.

Keywords: Sebaceous carcinoma; Upper eyelid; Young male.

INTRODUCTION

The human eyelid is a complex structure that is frequently subject to pathological diseases such as inflammatory and benign and/or malignant tumors with similar clinical presentation. The sebaceous gland tumors originate in the Meibomian glands, glands of Zeiss, caruncle, and skin of the eyebrow (Pe’er J, 2016). The sebaceous gland carcinoma (SGC) being the third most common malignancy in the eyelid frequently occurs in elderly women (Snow SN et al., 2002). It can be misdiagnosed as blepharitis or chalazion or conjunctivitis and result in significant morbidity and mortality (Kass LG et al., 1989). We report a case of an eyelid sebaceous gland carcinoma of a young boy who had undergone incision and curettage (I and C) several times. The importance of reporting this case is based on a misdiagnosis of SGC in young boys and the need for full-thickness biopsy in case of recurrent chalazion.

CASE REPORT

A 21-year-old boy, presented in the oculoplastic outpatient department with a chief complaint of swelling in the left upper lid for 6 months. He gave history Incision and curettage done elsewhere for the same condition thrice. However, an incision biopsy done in one of the tertiary centers showed sebaceous cell carcinoma and was referred for wide excision and reconstruction in our oculoplastic and oncology department.

On external examination, an oval mass approximately 3 cm x 2.5 cm in diameter was visible in the upper lid with mass effect causing mechanical ptosis [Figure 1a]. The mass was firm to hard in consistency with adherent to deep tissues associated with distorted lid margin and loss of cilia. The skin overlying tumor revealed an ulcerative lesion. The eyelid eversion revealed scar of previous surgery, thickening of the tarsal plate, marked distortion of anatomy, and vascular engorgement around the lesion [Figure 1b]. On ophthalmic examination, Visual acuity, Hertel’s Exophthalmometry measurements, and extraocular muscle motility were normal. There was no regional lymphadenopathy and systemic workup revealed no metastasis.
The tumor was removed en-block with a 5 mm margin and sent for histopathological reconfirmation. Double freeze-thaw cryotherapy was applied to all the skin and conjunctival margins as well as cut end of the canaliculus. The large upper lid defect was reconstructed in the same sitting with 1st step cutler beard technique wherein post auricular ear cartilage was harvested for posterior lamella reconstruction [Figure 2a, 2b, 2c and 2d]. He was prescribed oral antibiotics and proteolytic enzymes for a week and topical antibiotics for a month. Histopathological examination showed tumor consisting of nests and irregular lobules of oval cells involving the full thickness of eyelid with foci of necrosis and coarse nuclear chromatin and prominent nuclei with foamy, multivacuolated cytoplasm [Figure 3a and 3b]. There was no lymphatic, vascular and perineural invasion.
At the 6 weeks, post-operative visit, a 2nd step cutler beard procedure was performed to separate the eyelids, and conjunctiva was sutured to reconstructed the upper eyelid. The lower eyelid wound margins were freshened, underlying tissues, and the skin sutured to the surrounding tissue. Postoperatively after one week after 2nd step of the cutler beard procedure, there was no lagophthalmos or exposure keratitis, and no residual periocular malpositioning.

**Figure 3.** Hematoxylin and eosin stained section from a full-thickness wedge resection. (a) The tumor consisted numerous of nests and irregular lobules of invasive disease throughout the eyelid. (b) High magnification shows poorly differentiated tumor with foamy, multivacuolated cytoplasm and abnormal mitotic figure.

After 2 months it remains with excellent eyelid closure and good aesthetic results [Figure 4a and 4b]. He is advised to follow up every 3 months for functional and aesthetic evaluation as well as regional or distant metastasis workup.

**Figure 4:** Post-operative photographs of patients after 2 weeks of 2nd step of cutler beard procedure. (a) Maintenance of eyelid contour (b) Absence of lagophthalmos

**DISCUSSION**

The sebaceous gland carcinoma can occur anywhere in the body where sebaceous glands are found. However, the upper eyelid is the most common site due to the presence of more Meibomian glands (Mahipathy SR et al., 2016 and Shields JA et al.2015). The incidence of sebaceous gland carcinoma on the eyelid varies from geographical areas accounting for approximately 5% in the USA (Shields JA et al.2015),7.9% in Taiwan (Lin HY et al.,2006, and 28% to 60 % in Asia (Shihota R et al., 1996). The sebaceous gland carcinoma of the eyelid is more commonly occurs in old aged over 55-years (Shields JA et al., 2005 and While B et al., 2014). The patient in our study is young which contradicts other studies in which most of patients were from older age group. There were no justifiable causes like Muir-Torre Syndrome, HIV, or retinoblastoma for the early onset of the disease (Hussain RM et al., 2014 and Burns SJ et al., 2005). The male patient in our
study also contradicts other studies in which most of the patients are women (Snow SN et al., 2002 and Veras D et al., 2020). However, the occurrence of eyelid sebaceous gland carcinoma was reported in 39 -years –old males in a Brazilian study (Veras D et al., 2020). In the context of our country, no case has been reported occurrence of sebaceous carcinoma of the eyelid in young males. Our case was a young boy without previous history of irradiation or genetic predisposition who presented with a tumor size of about 2cm x 2cm that resembled a giant chalazion. On eversion of the eyelid, we noticed a scar of previous surgery, thickening of the tarsal plate, marked distortion of anatomy, and vascular engorgement around the lesion. He had undergone incision and curettage thrice in different hospitals for misdiagnosis of chalazion. However, sebaceous carcinoma of the eyelid commonly masquerades as recurrent chalazion or blepharitis, or chronic blepharocconjunctivitis leading to clinical misdiagnosis (Shields JA et al., 2005 and Shields JA et al., 2004). We should examine the lid margin, conjunctiva, and tumor carefully to avoid clinical misdiagnosis. It is recommended that all types of malignant eyelid tumors should undergo wide excision biopsy under frozen section or Moh’s micrographic surgery control (Shields JA et al., 2005 and Jakobiec FA et al., 1993). In our study, we performed wide excision with a 5 mm margin. We couldn’t do a mapping biopsy at primary sitting due to lack of facility. However, double freeze-thaw cryotherapy was applied on all margins of skin and conjunctiva to prevent the local spread of the tumor (Kaliki S et al., 2016). The upper eyelid defect was reconstructed at the same sitting with post-aurlicular ear cartilage and cutler beard technique since the tumor was limited to the eyelid, and there was no regional lymphadenopathy. In our case, the histopathological report revealed poorly differentiated sebaceous carcinoma with the margins free of tumor invasion and no lymphatic, vascular, and perineural invasion TNM (AJCC) stage: pT2cNx. We advised our patients to consult with an oncologist for further systemic metastasis work up and needful. However, the patient didn’t require any postoperative adjuvant therapy and sentinel lymph node biopsy in accordance with the prognostic value of the staging system for eyelid tumors American Joint Committee on Cancer Staging Manual, 7th edition (Ford J et al., 2017).

CONCLUSION

This case report aims to highlight the need for a high degree of suspicion, sound clinical skills, and good pathology backup for successful diagnosis and treatment of sebaceous carcinoma of the eyelid in the young population.

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Conflicts of Interest: None

Consent: Informed written consent from Patient was obtained.

REFERENCES


