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Case Report

Kissing sebaceous carcinoma of the eyelids: a rare case report

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ABSTRACT

Sebaceous gland carcinomas are tumours of the skin adnexa which occur around the periocular region of elderly females. It is a rare malignancy usually involving the upper eyelid and has an indolent course. Treatment is surgical excision of the lesion with lid reconstruction. Here authors discuss a case of swellings involving both the eyelids of an elderly female diagnosed as sebaceous gland carcinoma on biopsy for which wide local excision and primary eyelid repair was done. This case has been presented as the lesion masquerades as a benign one but is malignant with multifocal spread.

Keywords: Carcinoma, Eyelids, Malignant, Rare, Sebaceous

INTRODUCTION

Sebaceous gland tumor of the eyelids may arise from the meibomian glands, glands of Zeis or glands associated with the caruncle.[1] They are included in the list of tumors of the epidermal appendages, so-called adnexal skin structures. Sebaceous gland carcinoma (SGC) might be the second most common lid malignancy after basal cell carcinoma (BCC). Its multifocal origin and pagetoid spread giving it a unique place among eyelid malignancies.

Sebaceous glands are located in the periocular skin, caruncle, and eyebrow skin follicles. The tumor is a very rare, slow growing, and commonly found in elderly population with female predisposition. Mean age at diagnosis is mid-sixties; however, the tumor has been reported in children as young as 3.5 years old.[2] It is rare in Caucasians and common in oriental Asiatics. The reported incidence of SGC varies from 0.5 to 5% of all lid carcinomas in USA and 28% in China.[3,4] SGC most commonly arises from the meibomian glands anterior to the gray line, occasionally from the glands of Zeis or Moll, and from sebaceous glands in caruncle; however, the cell of origin may not be certain in 50–60% of cases.[5]

In contrast to basal cell carcinoma (BCC) or squamous cell carcinoma (SCC), SGC is two to three times more common in upper eyelid due to more number of meibomian glands there.[3,6] Five percent cases may have simultaneous involvement of both eyelids due to intraepithelial spread and/or spontaneous development of multiple primaries.

On one hand it can mimic as benign lesion as blepharoconjunctivitis, whereas on the other extreme it can have widespread local and fatal distant metastases. Immunohistochemistry, molecular biology, and electron microscopy have greatly improved the diagnosis, management, and prognosis of SGCs overall. Surgery, chemotherapy, and radiotherapy all contribute to the treatment of SGC.

CASE REPORT

A 63-year-old female presented to the Department of Plastic and Reconstructive Surgery with a swelling each on the upper and lower eyelids since 9 months. The upper swelling started initially 9 months back following which the lower eyelid swelling started 8 months back. Both the swellings had a spontaneous onset and were not painful. The swellings were partially obstructing her vision. There was no ulceration or bleeding from the swellings. The lesions measured 8mm in the upper lid and 5mm in the lower lid. They were firm in consistency, non-tender and not friable (Figure 1). There was no regional lymphadenopathy.

Figure 1: Pre- operative photograph showing the lesion in the upper and lower eyelids.

Ophthalmologist opined that the swellings were suggestive of a chalazion. The patient had a history of cataract surgery in both eyes 2 years prior with a posterior chamber intra-ocular lens in situ. Surgical oncologist commented that the lesion was probably a nodular form of basal cell carcinoma. After obtaining fitness for surgery under general anaesthesia, the surgical oncologist performed a V-shaped full thickness wide local excision of both the tumours with a 5mm margin (Figure 2).

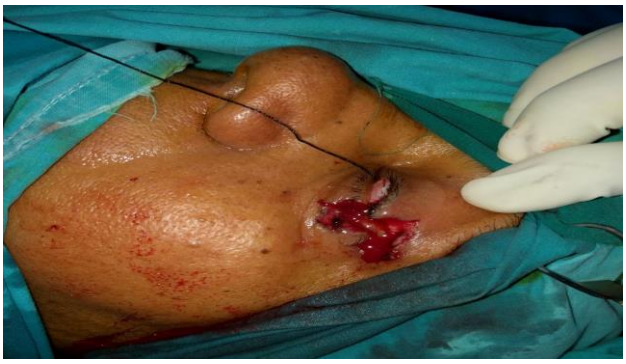

Figure 2: Picture following full-thickness wide local excision of the lesions.

The defects were closed in 2 layers, the conjunctiva was closed with 6-0 absorbable polyglactin sutures and the skin was approximated with 6-0 non-absorbable nylon sutures (Figure 3). Post-operative period was uneventful. The patient was discharged on the 5th post-operative day after removing sutures (Figure 4).

Figure 3: Immediate post-operative picture showing the suture line with mild edema of the eyelids.

Figure 4: Two weeks post-op picture after suture removal.

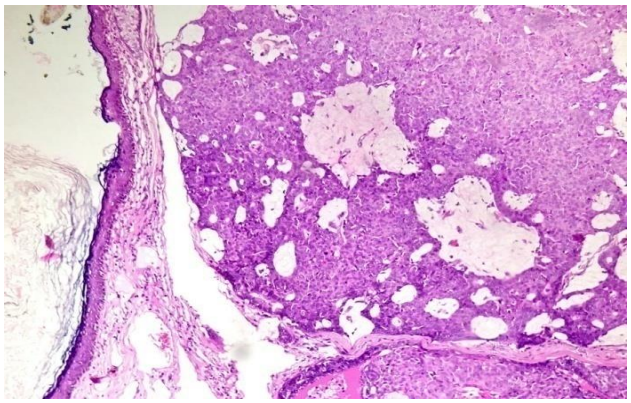

Figure 5: Section shows epidermis with the underlying dermis showing neoplasm composed of infiltrating sheets of malignant cells. H and E x 100.

Histopathology showed stratified squamous epithelium with skin adnexal structures with underlying dermis showing a malignant neoplasm composed of nests and

sheets of cells with moderate amphophilic to basophilic vacuolated cytoplasm and mild to moderately pleiomorphic vesicular nuclei with some showing prominent nucleoli.

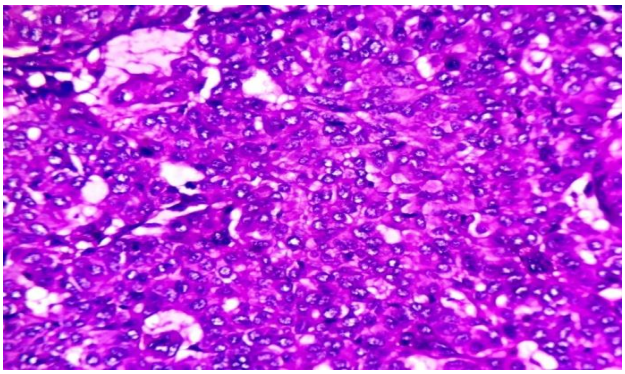

Figure 6: The malignant cells have scan to moderate eosinophilic cytoplasm and mild to moderately pleomorphic hyperchromatic nuclei with few cells showing vacuolated cytoplasm. H and E x 400.

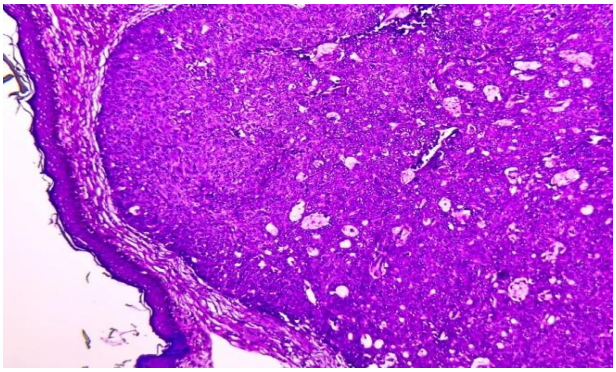

Figure 7: Infiltrating nests of cells showing cribriform pattern. H and E X 40.

Areas of necrosis were seen with margins free of tumour infiltration. The features were suggestive of sebaceous carcinoma (Figure 5,6,7).

DISCUSSION

Sebaceous gland carcinomas (SGC) are rare, slow growing skin adnexal tumours commonly arising in the periocular region. It constitutes about less than 5% of eyelid malignancies and has been considered to be the third most common eyelid malignancy following basal cell and squamous cell carcinomas. They arise from either the meibomian glands, glands of Zeis or glands in the region of the caruncle.[1] It usually occurs in elderly females with an increased incidence of local recurrence, regional, and distant metastases.[2] It is unique that it has a multifocal origin with pagetoid spread. Sebaceous glands are located in the dermis of periocular skin, tarsal plate, caruncle, and eyebrow skin follicles. Base of eyelashes have multiple modified sebaceous glands called Glands of Zeis. Upper eyelids are more commonly affected with sebaceous gland carcinoma due to the presence of plenty of Meibomian glands.[3,4]

There are two main pathological types of the SGC - nodular and spreading. In the nodular form, multicentric well circumscribed hard nodules spaced with adipose deposits are present around the upper tarsal plate. The spreading variety of SGC presents as a diffuse involvement of the eyelid margin, loss of eyelashes, and resembles other benign conditions like chronic blepharoconjunctivitis. It spreads in pagetoid form with diffuse intraepithelial infiltration of the eyelid skin of both the eyelids and conjunctival epithelium.[5-8] A rare form of SGC called Muir–Torre syndrome is characterized by squamous metaplasia of the tumor, multiple sebaceous adenomas or carcinomas in the skin, and visceral malignancies.

Histopathology shows large anaplastic cells with hyperchromatic nuclei and foamy cytoplasm due to lipid vacuoles, which is classical of sebaceous carcinoma. The cells stain positive for lipid such as Oil red O stain. Another characteristic feature is the spread of the tumour as infiltrating nests, cords or lobules as well as spread superficially within the epithelium. Sebaceous gland carcinomais invasive when there is hyperexpression of p53 on molecular biology. The tumor can metastasize by either direct, lymphatic, or haematogenous spread. The most common sites of metastasis are orbit, preauricular and submandibular lymph nodes, and parotid gland. It masquerades as a benign condition and therefore there is a delay in diagnosis, and this causes increased morbidity and mortality rates.[9] The literature reported a mortality of about 6%.[6]

Treatment depends on the stage of tumor. Wide local resection with or without neck node dissection forms the primary treatment. In advanced stages, orbital exentration may be needed. Radical surgical excision with a 4 mm tumor free margin is the treatment of choice. Moh's micrographic surgery is a more effective method of treatment. Approximately, 30% of SGCs recur after resection.[10,11] There is a high rate of metastasis (41%).[3,6] Composite full-thickness resections of eyelid will require reconstruction of all three layers, namely, anterior lamella (skin and subcutaneous tissue), muscle layer (orbicularis oculi) and posterior lamella (tarsal plate and conjunctiva).[12,13] The lower tarsus is a dense conjunctive structure which gives rigidity and stability to the eyelid, and containing the meibomian glands, which secrete a sebaceous material needed for lubricating the cornea.[12-14]

Tumors invading the tarsal plate cause full thickness defects of the eyelid. Defects of up to one-third of the total length of the lower eyelid are capable of being closed primarily. Larger tumors require surgical reconstruction through the preparation of flaps and with or without grafts.[15,16] There are many options to reconstruct the posterior lamella such as jugal mucosa grafts, mucosal grafts from the hard palate, chondro-

mucosal grafts from the nasal septum.[17,18] The presence of a free tissue graft in the posterior lamella requires a vascularised flap for the anterior lamella. Local nodal disease without distant metastasis is treated by radical neck dissection.[6] Topical mitomycin C, cryotherapy and proton electron irradiation has been tried for sebaceous gland carcinoma of the eyelid but with poor results.[11,20,21] Radiotherapy has been tried as a primary form of therapy with variable results.[6,21,22] Radiation is advised for poor surgical candidates due to advanced age or disease, for palliation, and in patients who refuse exenteration for advanced local disease.

The overall mortality rate is 6–11% because of delay in the diagnosis and treatment. The mortality from metastasis may go up to 30%. Involvement of upper or both eyelids and tumor size of more than 10 mm, duration of symptoms more than 6 months (mortality 35%), poorly differentiated and anaplastic tumors, haematogenous and lymphatic spread, orbital extension, multicentricity and pagetoid spread are poor prognostic factors. Tumors less than 6 mm and those arising from glands of Zeis are more favourable.

CONCLUSION

Sebaceous carcinomas are rare tumours which present as benign ocular lesions, and this requires proper diagnosis to start appropriate therapy due to the aggressive nature of the tumour. This case is presented due to the rarity of the simultaneous occurrence of sebaceous carcinoma of both the eyelids.

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