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Rare Eccrine Porocarcinoma of the Eyelid in A Non-Caucasian Patient

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Abstract

Eccrine porocarcinoma of the eyelid is a rare and locally aggressive tumor with a significant risk of metastasis and recurrence after surgical excision. There are only eight cases of eyelid porocarcinoma reported in the literature, most involve Caucasian patients. A 58-year-old non-Caucasian male had a recurrent mass of the left upper eyelid for a year. He underwent two prior mass removal procedures in the same location of the upper eyelid. At the time of this study, the patient underwent full-thickness excision, and a lateral canthal defect with superior palpebral involvement was sacrificed. The periosteal flap from the lateral orbital rim was attached to the edge of the intact tarsal plate of the upper eyelid as a part of the defect reconstruction. Good outcomes in terms of tumor recurrences, cosmesis, and upper eyelid functionality were observed one month following surgery. According to the histopathology, the tumor was identified as eccrine porocarcinoma, and the patient was recommended for chemotherapy. The 4-month follow-up showed no recurrence or metastasis. Eccrine porocarcinoma is a rare condition that should be taken into account when making a differential diagnosis for patient with malignant eyelid tumors. Wide excision is still a treatment of choice.

Keywords: Eyelid tumor, palpebral defects, periosteal flap, porocarcinoma, rare tumor

Introduction

Eccrine porocarcinoma (EPC), first described by Pinkus and Mehregan in 1963, is an extremely rare and aggressive malignant adnexal skin appendix tumor which arises from the intraepidermal ductal part of the eccrine sweat gland, also known as the acrosyringium.[1–4] Previous studies have reported that EPC accounts for approximately 0.005–0.01% of all cutaneous malignancies and most commonly affects people in the sixth to eighth decades of life, without sex predilection.[1,4–6] Owning to its rarity, the cause remains unknown and it could arise de novo or develop from pre-existing lesion (such as poroma with a long latency period).[1,2,5,7] History of radiotherapy, chronic ultraviolet exposure, trauma, and immunosuppression could increase the risk of EPC.[1,2,5,7] Approximately 18–50% of EPC cases are reported to originate from eccrine poroma.[1,4] White race seems to be more affected, which may be explained by an increased mutational charge in light skin phototypes due to UV radiation.[7]

Clinical differentation of EPC from other skin tumors remains challenging due to their lack of specificity in clinical and histopathological findings, frequently resulting in delayed diagnosis. in most cases, as its beningn counterpart (eccrine poroma) is called the "great imitator".[1,5] Previous studies reported that mean interval between tumor development and diagnosis has been reported to be five to nine years, although intervals several days to as long as 60 years have been documented.[5,7]

The most common predilection sites of EPC include the lower extremities (33.7–44%), followed by the head and neck region (18–30.6%), and the trunk (19.5–24%). Other reported location include the face, scalp, and ears (~20%), upper extremities (~11%), abdomen (~9%), and hands (3%).[4,5,8] In contrast, a Korean

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study involving 37 patients reported head and neck involvement in 29.7% of cases.[8] Eyelid involvement is exceptionally rare, accounting for only 0.7–0.8% of reported cases.[7,8] To date, only eight cases of eyelid EPC have been documented in the literature, seven of which occurred in Caucasian patients.[9–16]

The novelty of this report lies in the documentation of a rare case of EPC involving the upper eyelid in a non-Caucasian patient. This case represents the second reported instance of upper eyelid EPC in a non-Caucasian patient, while the first case was reported in Hong Kong.[16]

Case

A 58-year-old non-Caucasian male (Malay race, Sundanese ethnicity) presented with a reddish lump on his left upper eyelid that had been present for eight months. The mass grew larger without tenderness and did not bleed easily. The patient complained of experiencing similar incidents twice at the same location. The first lesion appeared 16 months earlier and was excised 11 months before presentation. Five months later, the same mass reappeared at the same place and was re-excised at a different hospital. However, the mass reappeared three months later. Unfortunately, he did not bring the past histopathologic results of previous mass. The patient had no history of eye redness and blurred vision, as well as of hypertension, diabetes, allergies, or other systemic diseases. Immunosupression condition, trauma, or chronic systemic disease were denied. However, he had been a heavy smoker since at young age and was a field worker.

Ophthalmologcal examination showed a firm well-defined, erythematous mass, measuring 8 x 6 x 4 mm on left lateral canthal upper eyelid,

Table 1 Reported Cases of Eyelid Eccrine Porocarcinoma

Author	Age/Sex	Location	Treatment	Follow-up	Outcome	Clinical Features
Boynton & Markowich[9]	68/F	Lower eyelid	Full-thickness excision	3 years	No recurrence	Erythematous, exophytic lesion with two distinct zones and central keratin plug
Orella et al.[10]	37/M	Lower eyelid	Wide excision	N/A	No recurrence	Not reported
D'Ambrosia et al.[11]	71/M	Lower eyelid	Mohs micrographic surgery	N/A	N/A	Asymptomatic lesion
Kim et al.[12]	75/M	Upper eyelid	Full-thickness excision	6 months	Orbital exenteration	Poorly defined, skin-colored, rubbery hard lesion
Greco et al.[13]	70/M	Lower eyelid	Full-thickness excision	2 years	No recurrence	Pagetoid spread involving the dorsum of the nose
Jain et al.[14]	70/M	Upper eyelid	Mohs micrographic surgery	6 months	No recurrence	Erythematous lesion with eyelash loss
Chua et al.[15]	86/M	Lower eyelid	Wide excision	18 months	No recurrence	Irregular nodule with distinct margins and eyelash loss
Mak & Li[16]	60/F	Medial canthus	Wide excision with frozen section	1 year	No recurrence	Firm, well-defined lesion with irregular crusting and telangiectasia
Present case (2019)	58/M	Lateral upper eyelid	Wide excision	4 months	No recurrence	Rounded, well-defined, reddish, fixed recurrent tumor

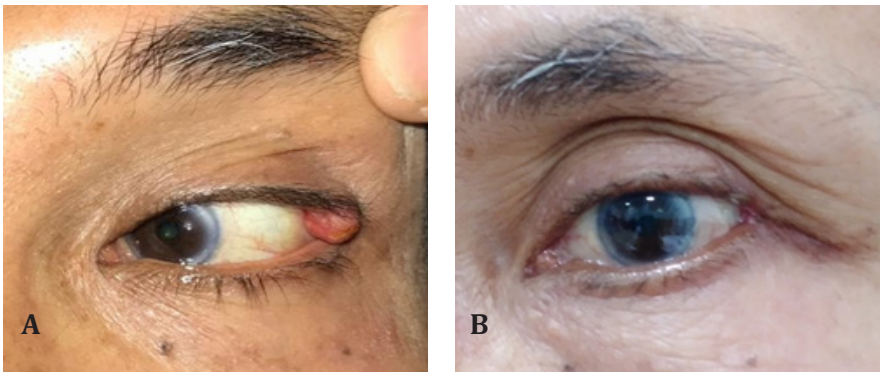

Figure 1 Clinical appearance: (A) Mass in the lateral upper eyelid; (B) 3 weeks follow-up

without other abnormalities of the ocular surface (Figure 1). The mass was spherical, chewy in consistency, fixed, reddish in color, non-tender, and associated with eyelash loss, raising suspicion of malignancy. Both eyes' visual acuity were 1.0 and intraocular pressure were within normal limits. Eye movement on both eyes showed no restriction. Other clinical examination revealed no enlarged lymph nodes around the neck or mandible, and everything was within normal limits. At the time, patient was diagnosed with suspected recurrent sebaceous gland carcinoma on left upper eyelid.

This patient underwent complete mass excision under general anesthesia. The mass was excised up to 3-mm from the margin, sacrificing about one-third full thickness of the lid. Periosteal flap from lateral orbital rim was attached to the edge of the rest of intact tarsal plate of the upper eyelid and then the skin was closed horizontally to reconstruct the eyelid and to achieve the good cosmetic aspect. The

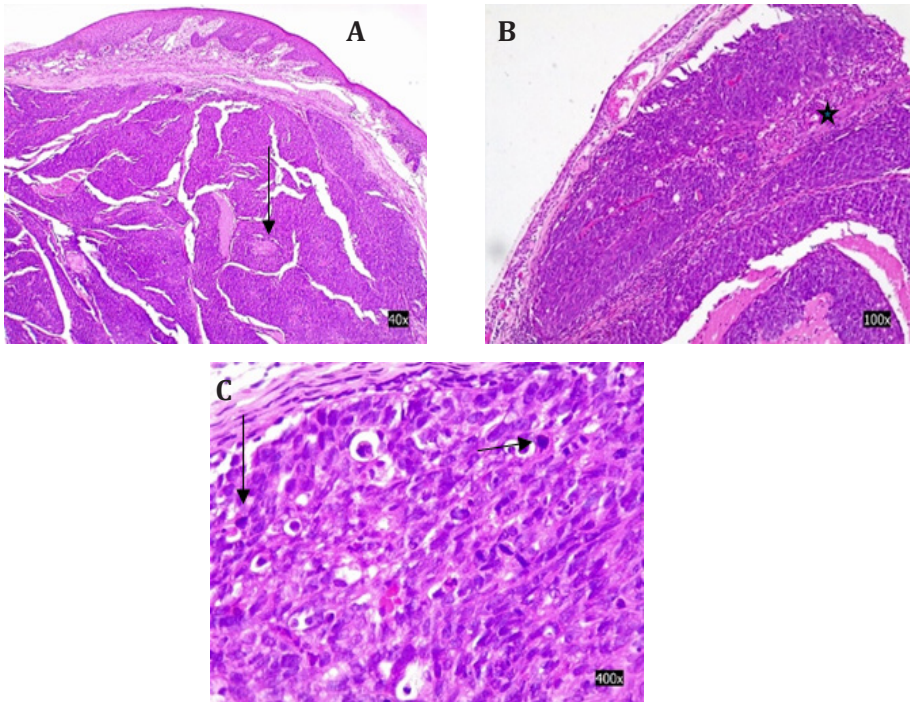

Figure 2 Histopathological Features Of Eccrine Porocarcinoma
(A) Comedonecrosis (arrow) (H&E, ×40); (B) Basaloid cells with clear cell change (asterisk) (H&E, ×100); (C) Frequent mitoses (>10 per 40 HPFs; arrow) (H&E, ×400).

excised specimen was submitted to Department of Anatomical Pathology.

According to macroscopic observations, the mass tissue was brownish-white and measured 1.0 x 0.8 x 0.6 cm. A white solid mass of 0.8 x 0.6 x 0.4 cm was visible in the lamellation, with a margin and base were clear from tumor cells. Furthermore, microscopic findings showed squamous epithelium covering the sample (Figure 2). The nucleus was within normal limits, and the subepithelial component was composed of a nest and islands of hyperplastic round, oval, and basaloid tumor cells. Pleomorphic and hyperchromatic nuclei and obvious mitotic nuclei, as well as clear cell areas and focal necrosis (comedonecrosis), were also found. Blood vessel dilation coincided with lymphocyte infiltration of the surrounding stroma, which was the infiltrated by tumor cells. Sebaceous glands, hair follicles, sudoriferous glands, mature fat cells, and muscle tissue were within normal limits.

The histopathological finding revealed an eccrine porocarcinoma and the patient was referred to the Medical Oncology Department at Hasan Sadikin Hospital for further management. The patient was advised to undergo six cycles of chemotherapy with paclitaxel and carboplatin, but he refused. At the four-month follow up, the patient experienced neither recurrence of the lump nor enlargement of lymph nodes after the surgical excision.

Discussion

The clinical presentation of EPC is highly variable. The lesion commonly appear as an asymptomatic erythematous or violaceous nodul or mass, more rarely, as a polypoid plaque of violet or erythematous color that enlarges over weeks to months. In some cases, EPC present with itching, ulceration, spontaneous bleeding, and pain.[5,6,17–19] In this case report, patient showed presentation of erythematous mass without tenderness and spontaneous bleeding, despite significant growth over an eight-month period.
Diagnosis of EPC is challenging because the disorder presents a wide variety of clinical, dermoscopic, and histopathological findings.[1,6] Moreover, EPC is often found in white race and it occured on eyelid, which is a rare location.[6,7] Patient already experienced similar tumor at the same location twice and underwent surgical treatment in different hospital. Unfortunately, the histopathological findings of previous tumor was not obtainable. The possibility of previously misdiagnose could happen in this patient since EPC is often misdiagnosed as othe skin lesions, particularly as squamous cell carcinoma, basal cell carcinoma, or other adnexal tumors.[1,5,6,16]

Histopathologically, EPC demonstrates features suggestive of its development from the acrosyringium, the intraepidermal spiral duct of the sweat glands. It is usually characterized by islands of atypical poromatous basaloid cells and the presence of intracellular lumina and/or true ducts and can be differentiated from squamous and sebaceous cell carcinoma by the presence of ductal differentiation (intracystoplasmic or actual duct formation).[5,15] The mitotic index, necrotic areas, and infiltrative growth pattern are frequently seen.[4,5] Moreover, nuclear hyperchromia is a notable characteristic of EPC cells. Consequently, employing histological staining techniques such as EMA, PAS, CK AE1/AE3, p63, S100 and Ki67, which have been reported as positive 100% of cases, is imperative for ensuring accuracy and minimising the risk of misdiagnosis.[1,4]

The therapeutic approach to porocarcinoma consists of surgery with adjuvant therapy, as it is an aggressive disease with a high potential for morbidity and mortality.[4,17] At time of diagnosis, 22.3% patients present with metastatic spread (17% with lymph node metastases, 3.9% with distant metastases, and 1.5% with a locoregional cutaneous spread).[4] Wide local excision (WLE) is the primary treatment, with a debate of optimal surgical margins ranging from 2 mm to 3 cm (resulting in cure rates of 70–80% when the margins are clear), while Mohs surgery (MMS) increasingly outperforms WLE in the reducing the likelihood of tumor recurrence.[1,4,5,17] In selected cases, sentinel lymph node biopsy (SLNB) should be performed, even in the absence of palpable lymph nodes, with success rate of 81.3% in identifying occult lymph node metastasis.[4] Further investigation with SLNB is highly indicated in specific scenarios, such as aggressive tumor types, lymphadenopathy, and histopathological criteria indicating a higher risk of metastasis (e.g. tumor depth >7mm, lymph node vascular invasion, and >14 mitoses in 10 high-power fields, poorly differentiated neoplasm, and tumor located in head/neck region).[1,4–6,18] Reconstruction of the eyelid after WLE can be challenging, depends on the size and depth of the tumor and the excision. Functionality aspect, as well as cosmetic, should be taken into account.

In conclusion, given its aggressive nature,

the primary treatment for EPC requires serious attention. Although EPC of the eyelid is a rare and may resemble other eyelid malignancies, its diagnostic and therapeutic principles are consistent with EPC arising at other anatomical sites. This case highlights the importance of accurate histopathological assessment and multidisciplinary collaboration, particularly involving dermatopathology and oncology specialists, to achieve optimal patient outcomes.

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