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Case Report

Membranous basal cell adenoma arising in the eyelid

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Abstract: Basal cell adenoma (BCA) is a specific entity that lacks the myxochondroid stromal component of pleomorphic adenoma. Membranous basal cell adenoma is a rare variant of BCA, which is characteristic by abundant eosinophilic extracellular hyaline material deposited either inside or at the periphery of the epithelial islands. Herin we describe the first case of membranous BCA arising in the upper eyelid in a 38-year-old woman. A well-demarcated nodule arising in the eyelid was composed of isomorphic basaloid cells organized with a prominent basal cell layer and distinct basement membrane-like material. Immunohistochemically, S100 protein and p63 highlighted the basal aspect of the peripheral epithelial cells, while CK7 expressed on the luminal cells. A diagnosis of membranous basal cell adenoma of the eyelid was made. At follow-up for 2 years and 3 months later, there was no evidence of recurrence. Further pathological characteristics of this disease are discussed.

Keywords: Basal cell adenoma, eyelid

Introduction

Basal cell adenoma (BCA) was a specific entity that lacks the myxochondroid stromal component of a pleomorphic adenoma, which accounts for 1% to 2% of all salivary gland epithelial tumors. Less frequently, these tumors may be found in minor gland sites, with the upper lip being the most common site. But BCA has not been described in the eyelid reviewing the English literature. The majority of the eyelid tumors, benign and malignant, are of cutaneous origin. Amongst the tumors arising from adnexal structures of the eyelid, pleomorphic adenoma is the most common [1]. We report a rare case and the first case of membranous basal cell adenoma arising in the eyelid.

Case report

A 38-year-old woman was referred to our hospital for evaluation of a lesion of the right medial upper eyelid. She had been aware of it for 2 years. The lesion grew slowly. CT image showed a well-defined, homogeneous mass in the upper eyelid and protruded into the orbit without bony resorption, measuring 12 mm × 5 mm (**Figure 1A**, **1B**). Physical examination revealed a well-defined, painless, and elastic-hard mass in the palpebra interna of the middle of upper eyelid. The mucosa of the eyelid was normal, and no ulceration or induration was found. Other ophthalmic examination results were normal. The tumor was soft and easily dissected without any demonstrable involvement of the orbicularis oculi muscle or the wall of the canaliculus. The mass was excised and was submitted for the pathologic examination.

Pathological findings

Microscopically, low power microscopy illustrates the lesion was well circumscribed, which was composed of basaloid cells with slightly eosinophilic cytoplasm, arranged as trabecular or tubular architecture (**Figure 2A**). The tumor islands comprise uniform basaloid cells with nuclear palisading at the periphery. Peripheral cells which showed a palisaded-like fashion were surrounded by abundant hyaline basement membrane material. The hyaline material was deposited at the epithelial islands. Some epithelial cells were entrapped in the basement membrane material (**Figure 2C**). Around the lesion, accessory lacrimal glands of Krause were presented (**Figure 2B**). These basaloid cells were small, with scant cytoplasm and dense basophilic nuclei. Central cells were

Figure 1. A, B. CT image showed a well-defined, homogeneous mass in the upper eyelid and protruded into the orbit without bony resorption, measuring 12 mm × 5 mm.

slightly large, cubial. Small ductal structures were observed. Immunohistochemically, S-100 protein, p63 highlight the basal aspect of the peripheral epithelial cells (**Figure 2D**), while CK7 expressed on the luminal cells (**Figure 2E**). Pan-Cytokeratin was positive both on luminal cells and peripheral basaloid cells (**Figure 2F**). Vimentin and glial fibrillary acidic protein (GFAP) were negative in tumor cells. The final diagnosis was membranous basal cell adenoma of the eyelid. 2 years and 3 months since the surgery, there has been no evidence of recurrence.

Discussion

The eyelids contain numerous histological elements that can be the origin of several types of benign or malignant tumors. The majority of the eyelid tumors are of cutaneous origin. Tumors arising in the eyelid might be divided into four groups: epidermal, adnexal, stromal and secondary tumors. Epidermal tumors might represent most of the eyelid tumors. In the adnexal tumors, sebaceous gland tumors most frequently occur. Some other tumors, such as sweat gland, hair follicle originated may arise in the eyelid. Salivary gland-type tumor virtually always develops in the deep orbital lobe of the gland. Rarely, the palpebral lobe might be involved [2]. The accessory lacrimal glands of Krause and Wolfring might present pleomorphic adenoma extremely rarely [3]. Our case of membranous BCA was found in the middle, and deep location of the right upper eyelid, which might be originated from glands of Krause of the accessory lacrimal glands. Lesions of the lacrimal gland account for approximately 5% to 14% of all biopsied orbital masses [9]. In lacrimal gland tumors, the incidence of pleomorphic adenoma ranges from 21% to 62% [10]. However, reviewed the literature of pleomorphic adenoma arising in the eyelid, Only 5 cases were reported to arise from either the accessory lacrimal gland of Wolfring or from the accessory lacrimal gland of Krause [3-7]. No BCA was described in such sites.

The term BCA was first described by Kleinsasser and Klein in 1967 [11]. On the basis of morphologic features BCA was divided into four subtypes: solid, trabecular, and membranous variants [8]. The most common type is the solid variant. According to previous studies of BCA, it accounts for 1-3% of all salivary gland tumors, but none occurred in the eyelid reviewing the English literature. Membranous BCA is a rare benign neoplasm characterized by the presence of abundant, thick, eosinophilic, and PAS-positive hyaline basal lamina material around the smooth-contoured tumor islands, and absence of the myxochondroid stromal component. The histogenesis of membranous BCA is uncertain, Yu et al [12] suggested that proliferating basal cell of the strated ducts may play a main role in the histogenesis of membranous BCA of the salivary glands.

The differential diagnosis at pathological aspects is other salivary-type benign or malignant entities, such as pleomorphic adenoma,

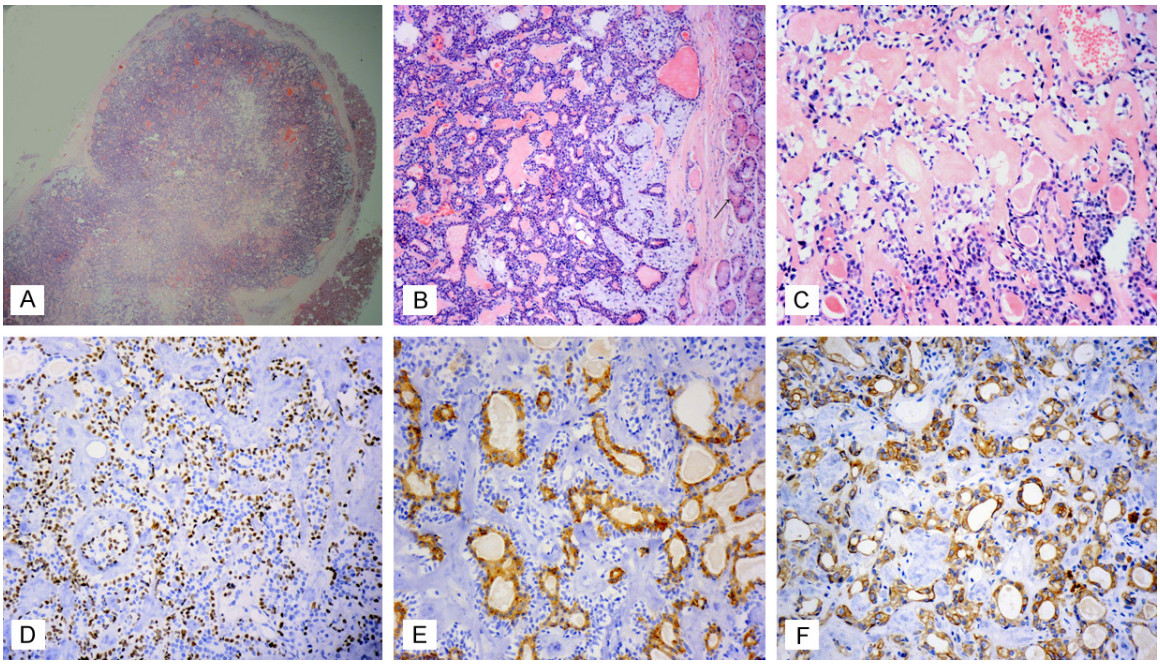

Figure 2. A. The tumor was well circumscribed, which was composed of basaloid cells with slightly eosinophilic cytoplasm, arranged as trabecular or tubular architecture. B. The tumor islands comprise uniform basaloid cells with nuclear palisading at the periphery. Around the lesion, accessory lacrimal glands of Krause were presented (arrow). C. Peripheral cells which showed a palisaded-like fashion were surrounded by abundant hyaline basement membrane material. Some epithelial cells were entrapped in the basement membrane material. D. p63 highlighted the basal aspect of the peripheral epithelial cells. E. CK7 expressed on the luminal cells. F. Pan-Cytokeratin was positive both on luminal cells and peripheral basaloid cells.

basal cell adenocarcinoma, epithelial myoepithelial carcinoma or adenoid cystic carcinoma. In salivary-type tumors, pleomorphic adenoma, basal cell adenocarcinoma, epithelial myoepithelial carcinoma and adenoid cystic carcinoma all have two-layer ductal structures. Benign entities such as pleomorphic adenoma may share some common architectures, but pleomorphic adenomas usually have myxoid stroma, and sometimes exhibit a chondroid (pseudocartilaginous) appearance. Basal cell adenocarcinoma may also share some simulated morphology with BCA. But basal cell adenocarcinoma has cellular atypia, the number of mitotic figures, and also presents invasive pattern. Histologic hallmark of epithelial myoepithelial carcinoma is the presence of two cell types identified in varying proportions in any given tumor, included luminal or inner epithelial cell layer, which is dark-staining cuboidal to columnar cells with central or basally placed nuclei, the outer myoepithelial cell layer was polyhedral cells with eccentrically placed nuclei, abundant clear cytoplasm, and distinct cell borders; the myoepithelial cell layer may predominate. Adenoid cystic carcinoma might also be confused with BCA, which also can present basal like cells. If grasped these features: invasive growth, no capsulated, myoepithelial cells constituted pseudo-glandular architectures, adenoid cystic carcinoma can differentiate with BCA. Because such rare case arising in the eyelid, some cutaneous origin tumors also should be ruled out. For example, basal cell carcinoma might have basal-like arrangements, and follicular originated tumors also have basal-like features. If we note that basal cell carcinoma attached to the epidermis and having the retraction artifact, follicular tumors having well-formed keratocysts or papillary mesenchymal bodies, and associated with dermis, we could easily rule out such cutaneous origin tumors.

Membranous BCA is usually multilobular and frequently unencapsulated; however, the case we reported was well-circumscribed. After 2 years and 3 months of follow-up, no recurrence has been detected in the case. Reviewing the literature, a case of malignant transformation

of a basal cell adenoma has been reported [13]. Therefore, careful follow-up observation is important.

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Disclosure of conflict of interest

The authors declare no conflicts of interest.

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