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CASE STUDY

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"Save my tears": Rare presentation of *Hidradenoma papilliferum*

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ABSTRACT

Hidradenoma papilliferum (HP) is a rare benign adnexal tumor, most commonly arising in the anogenital region of middle-aged women. *Ectopic hidradenoma papilliferum* (EHP) is an uncommon variant that occurs in apocrine-rich or modified apocrine gland-bearing sites, such as the eyelid, external auditory canal, and scalp. Due to its atypical presentation and non-specific clinical appearance, EHP can mimic a wide range of benign and malignant lesions, often leading to misdiagnosis. We present the case of a 57-year-old male with an unusual nodulocystic lesion on the right lower eyelid margin, which was histopathologically confirmed as HP. The clinical, dermoscopic, and histopathological features are discussed with emphasis on the diagnostic challenges and differential diagnoses. Complete surgical excision remains the treatment of choice, with excellent prognosis and minimal risk of recurrence. Awareness of this rare entity is crucial for dermatologists and pathologists to ensure accurate diagnosis and prevent unnecessary aggressive interventions.

KEY WORDS

Hidradenoma papilliferum; Apocrine Glands; Prognosis; Skin Neoplasms; Eyelid tumor;Surgical excision

ARTICLE HISTORY

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Introduction

HP is an uncommon benign adnexal tumor that arises predominantly in the anogenital region of adult women, believed to originate from apocrine sweat glands. First described by Werth in 1878, it is typically localized to vulva, particularly the labia majora. However, a subset of these tumors occurs in extragenital sites, described as EHP. These ectopic tumors are rare and often misdiagnosed due to their atypical location and non-specific clinical features [1].

EHP can present as solitary, slow-growing nodules in regions rich in apocrine or modified apocrine glands, such as the head and neck. Despite being benign, their unfamiliar location often leads to confusion with malignant tumors like basal cell carcinoma or syringocystadenoma papilliferum. Understanding its histopathological and immunohistochemical profile is essential for accurate diagnosis and appropriate management [2].

This paper presents a case of atypical presentation EHP on the eyelid of a middle-aged male and discusses the diagnostic challenges, histological features, and clinical significance of this rare entity.

Case Report

A 57 year old male presented with an asymptomatic growth over his right lower eyelid margin since 2 years gradually increasing in size. Patient also complains of restricted vision, irritation and excessive lacrimation. Excisional biopsy was performed under local anaesthesia and histopathology showed tubular pattern along the cystic spaces with papillary fronds projecting into cystic spaces suggesting HP. His past medical history was unremarkable, and there was no history of trauma, prior surgery, family history or similar lesions [3] (Figure 1).

Figure 1. (A) 57-year-old male with hidradenoma papilliferum over the right lower margin of the eyelid. (B)After Suturing. (C) Histopathology showed a . tubular pattern along the cystic spaces with papillary fronds projecting into the cystic spaces, suggesting Hidradenoma papilliferum (Black Arrow). (D) Intra-operative picture after excision.

Clinical Examination

A well-circumscribed, dome-shaped, tense, shiny, nodulocystic mass measuring approximately 2 cm x 2cm was observed near

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the right lower lid margin. The lesion was brownish black, non-tender, immobile, and without any signs of inflammation or ulceration. There was no regional lymphadenopathy. Given its appearance, differential diagnoses included junctional nevus, basal cell carcinoma, sebaceous adenoma, and syringoma.

Investigations and Histopathology

Dermoscopy evaluation was done with Dermlite DL5 and iPhone 14 Pro Max camera as secondary magnification, which showed features like tan brown- whitish to mild bluish homogenous area with mild scaling and a few terminal hair emerging out of the lesions. These features were inconspicuous and suggestive of junctional melanocytic nevi.

An excisional biopsy was performed under local anesthesia. Grossly, the excised lesion appeared as a flaccid after (exsanguination of cystic fluid following puncture), tan-brown nodule. Histopathological examination revealed a well-circumscribed dermal tumor with no connection to the overlying epidermis. The tumor was composed of papillary structures and cystic spaces lined by a double layer of epithelial cells: an inner layer of columnar cells exhibiting decapitation secretion and an outer layer of cuboidal to myoepithelial cells. No cellular atypia or mitotic figures were noted.

Treatment and Follow-up

Patient was prescribed a short course of oral & topical antibiotics for 5 days, and the suture was removed after 1 week. The patient recovered uneventfully, and no recurrence was noted during a 6-month follow-up.

Discussions

Previous studies have found that HP is most common in the anogenital region of middle-aged women, particularly the labia majora, likely due to the high density of apocrine glands. Ectopic HP, however, is a rare phenomenon and can occur in areas with modified or vestigial apocrine glands such as the eyelids (glands of Moll), external auditory canal (ceruminous glands), axillae, scalp, and nasal region [4, 5]. A systematic review by Michel et al. found that among all reported HP cases, fewer than 10% were ectopic. EHP affects both sexes but shows a slight female preponderance and tends to occur later in life (typically between 40–70 years). The eyelid is among the most frequently reported ectopic sites due to its abundant modified apocrine structures. [3] Etiopathogenesis of HP, including its ectopic variant, is believed to originate from apocrine glands or anogenital mammary-like glands. The presence of decapitation secretion, characteristic of apocrine differentiation, supports this theory. Some researchers propose that ectopic lesions may arise from pluripotent adnexal progenitor cells capable of apocrine differentiation under unknown triggers. [5] HP is typically non-hormone-responsive, although some reports suggest hormonal influence due to its resemblance to mammary ductal tissue and occasional expression of estrogen and progesterone receptors in vulvar lesions. However, ectopic lesions rarely show such receptor positivity [6,7]. Clinical Features EHP usually presents as a solitary, asymptomatic, slow-growing, dome-shaped or cystic nodule measuring 0.5–2 cm in diameter. It is typically skin-colored to pinkish, non-tender, and fixed to the skin but mobile over deeper tissues. Due to its non-specific appearance, it can be mistaken for epidermoid cysts, sebaceous tumors, or adnexal carcinomas [6]. Histopathology and Immunohistochemistry remains the gold standard for diagnosis. Characteristic findings include: A well-circumscribed dermal nodule without epidermal connection [1]. Cystic and papillary architecture with a bilayered epithelium. Inner layer: columnar cells showing decapitation secretion. Outer layer: cuboidal/myoepithelial cells. Immunohistochemical markers help confirm apocrine differentiation. Common findings include: Positive: CK7, EMA, GCDFP-15, CEA, S-100 (occasionally), GATA3 [2]. Negative: CK20, p63 (inconsistent), hormone receptors (usually). These features help distinguish EHP from syringocystadenoma papilliferum, papillary eccrine adenoma, and tubular apocrine adenoma [5]. Differential Diagnosis EHP can mimic various benign and malignant adnexal tumors. Key differentials include: Syringocystadenoma papilliferum: Typically connected to the epidermis and has plasma cell-rich stroma. Tubular apocrine adenoma: Lacks prominent papillary projections. Basal cell carcinoma: Shows peripheral palisading and clefting. Sebaceous adenoma: Contains lobules of sebaceous cells. Apocrine carcinoma: Shows cytological atypia and high mitotic index. Complete surgical excision is the treatment of choice. Local recurrence is rare but may occur with incomplete excision. There is no role for radiotherapy or systemic therapy in benign lesions. Malignant transformation is extremely rare, though a few cases of hidradenocarcinoma papilliferum have been documented in anogenital lesions [6]. The prognosis of EHP is excellent. Long-term follow-up is recommended only in cases where excision margins are uncertain or in lesions with atypical histology. Malignant transformation has not been reported in ectopic cases to date.

Conclusions

EHP is a rare but benign tumor that can occur in apocrine-rich sites outside the anogenital region, such as the eyelid. Due to its non-specific clinical presentation and rare occurrence, it is often misdiagnosed. Histopathological examination, supported by immunohistochemistry, is essential for accurate diagnosis. Complete surgical excision is curative, and prognosis is excellent. Awareness among dermatologists and pathologists is key to preventing misdiagnosis and overtreatment of this benign entity.

Conflicts of Interest

NIL

Patient Consent

The patient provided written informed consent for publication of clinical details and images.

Ethical Approval

Institutional ethical approval was obtained prior to publication.

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