

Rare and unusual occurrence of ectopic hidradenoma papilliferum in maxillofacial region

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Abstract

Hidradenoma Papilliferum (HP) is a rare, benign, cutaneous adnexal tumor of middle-aged females arising mainly in the apocrine sweat glands of perianal and genital region. Initially, HP was considered to be an apocrine sweat gland tumor however recent research showed that it descends from the anogenital mammary-like glands, HP can be regarded theoretically as the cutaneous complement of mammary intraductal papilloma. Nongenital HP is an infrequent manifestation, ectopic forms of this entity have been defined with bulk of them occurring on the head and neck region (60%) and 85% of cases being 1.5 cm in the greatest diameter or lesser. Average age of patients with ectopic HP is 1–2 decades older than the age range of onset in patients with anogenital HP. Here, we present a 15-year-old adolescent female with an unusually large, ectopic occurrence of nodular HP on right cheek since 2 years.

Keywords: Adnexal tumor, anogenital mammary-like glands, apocrine tumor, ectopic hidradenoma papilliferum, facial hidradenoma

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INTRODUCTION

Hidradenoma papilliferum (HP) is a slow growing benign, cystic, papillary adnexal tumor showing apocrine differentiation. The tumor initiates from the apocrine glands or anogenital mammary glands occurring almost solely in the female perianal region.^[1,2] First mentioned in 1878 by Werth. However, on some occasions, these tumors present themselves in other regions of the body apart from the anogenital area and are stated as “ectopic” HP.^[1,3,4]

Ectopic forms of (HP) usually tend to transpire in the head and neck or the maxillofacial region; on majority of circumstances ectopic lesions of HP ensue themselves in specific parts of the face like external ear/external auditory

canal or upper and lower eyelids, where the presence of modified apocrine glands are found in ample amounts.

Although some of the ectopic apocrine glands are indigenous to the skin of the nose, tumor arising in this region is rare and there has been only one case reported of ectopic HP arising in nasal skin.^[5]

Although HP is mostly seen in the vulvar/peri-genital region of adult females,^[6,7] some Ectopic forms can also occur in males and usually appear as a nodule, a subcutaneous tumor, or a cyst, most commonly in the head and neck regions. Apocrine type of glands is present in the skin, breast, eyelids and external ear. These glands in the breast conceal

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fat droplets into the mammary secretions (breast milk) and those present in the external auditory canal help form earwax. Apocrine glands in the skin and eyelid are usually sweat glands and their secretions typically have an odor. In this article, we present an unusual presentation of this tumor in an adolescent female occurring on the cheek where apocrine glands are not usually associated and of a fairly large size, showing a pedunculated appearance as compared to its usual presentation that of a subcutaneous nodule or cystic appearance. The lesions usually presents as an asymptomatic slow-growing, red, firm, mobile, well-delimited nodule that grows for a long time before resection.^[8]

CLINICAL HISTORY

A 15-year-old adolescent girl presented to us with an asymptomatic recurring growth on the right cheek. The patient's caregiver gave a history of being operated twice in the past 2 years for the same by means of a laser excision (paper work not present). The growth was approximately 30 mm × 20 mm × 30 mm in size, exophytic, pedunculated mass with skin showing areas of necrosis and ulceration. The underlying surface of the lesion appeared erythematous, mulberry like in texture [Figures 1 and 2]. The growth was firm in consistency and sessile in nature. No associated symptoms or discharge was noted.

The underlying skin showed mild erythema associated with a vertical scar due to previous surgeries. Owing to the initial presentation a differential diagnosis of cutaneous papilloma, pyogenic granuloma and sebaceous cyst were made and a decision was taken to perform an excisional biopsy under local anesthesia with 0.5 cm healthy skin margin keeping in mind the recurrence potential of the lesion.



Figure 1: Lesion showing parts of necrosed overlying skin and mulberry like surface texture

Histopathological examination

Revealed a solid-cystic, partly circumscribed, lobulated tumor in the dermis showing a sheet-like and papillary architecture. Individual cells are round to oval having vesicular nuclei, inconspicuous nucleoli and eosinophilic cytoplasm. Few cells show cytoplasmic clearing. Small and large lumina lined by cuboidal ductal cells are seen at places. Focal cystic degeneration noted. No significant cytological atypia or mitotic figures were seen [Figures 3 and 4].

DISCUSSION

The development of the adnexal apparatus and apocrine glands begins *in utero* where they continue to remain dormant until stimulated by the hormonal splurge at puberty. These glands void their secretions into the isthmus region of the pilosebaceous apparatus; hence, their presence is seen near the hair follicle regions. Therefore, making the occurrence of apocrine glands concentrated majorly in the anogenital region and axilla.^[9] Hence, majority of HP cases tend to originate in these regions. Literature search reveals that estrogen production which occurs during and after puberty, aids in the development of this tumor in females and so there is a tendency of this tumor to arise after the onset of puberty. The cases reported in the male population, however, needs more research toward this hypothesis. A search in Medline of Dermatology Research and Practice only about 20 cases of ectopic HP are reported. To the best of our knowledge, the case reported here is unique as it is only the second case of ectopic HP reported on the cheek region and it also presents a novel appearance of this tumor in this region of the face, as apocrine glands are normally not associated with this part of the head and neck region.



Figure 2: AP view showing a pedunculated appearance of the lesion

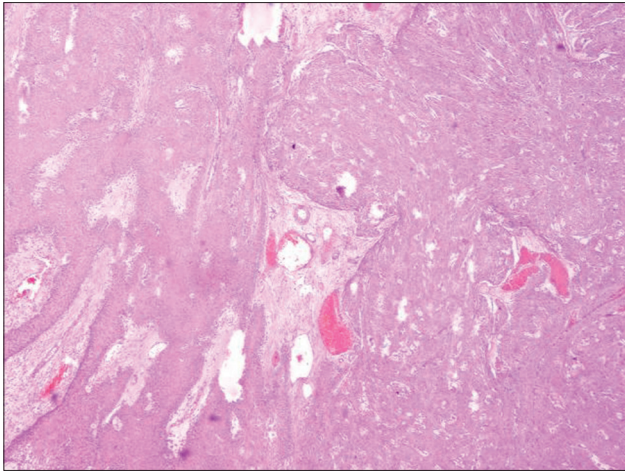


Figure 3: Low power view shows a solid-cystic dermal tumor arranged in lobules and sheets (H&E, x40)

This tumor is commonly seen in middle-aged white females (range 25–66 years), independent of its classic or ectopic form.^[10] Although differing from anogenital HP, nearly 50% of ectopic cases of HP are seen to occur in men. The ectopic form of the tumor is more consistently seen at the eyelid, nose, scalp and external ear, where modified apocrine glands (Moll and ceruminous glands) are found usually in the head and neck region, other regions include arm, thigh, back and nipple.^[11]

Clinically both the forms are alike in their presentation, most are asymptomatic, are likely to grow for a long period of time prior to being noticed. Some of them can be associated with pain, severe itching and may show ulceration at the time of presentation. Similar to other adnexal cutaneous neoplasms they may show a morphological resemblance to basal cell carcinoma and spinocellular carcinoma.

Immunohistochemistry

The HP expresses epithelial membrane antigen, carcino embryonic antigen, gross cystic disease fluid protein -15 and human milk fat globule membrane antigen and are usually positive for the markers of apocrine differentiation.^[5]

Their prognosis is usually good after excision and remains the most preferred treatment. Recurrence of these tumors is mostly due to incomplete removal of the primary lesion and presently there is no reported knowledge of recurrence in the ectopic forms.

The malignant transformation in anogenital HP is not very common but has been documented (intraductal carcinoma resembling apocrine carcinoma and invasive adenosquamous carcinoma) whereas no such documentation is present for the ectopic form. It has been hypothesized that Human

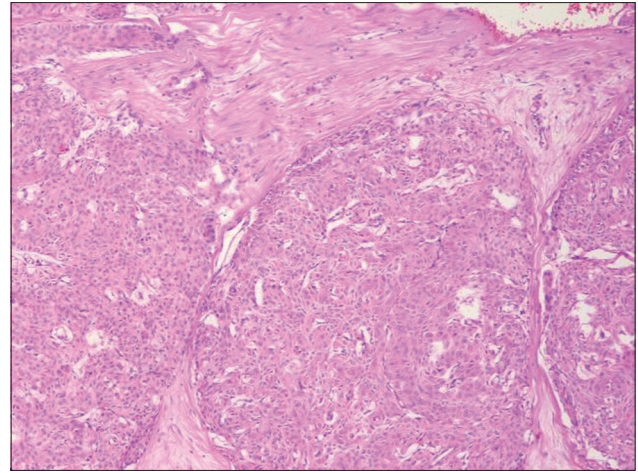


Figure 4: Higher magnification shows round to oval cells with vesicular nuclei and inconspicuous nucleoli. Few cells with clear cytoplasm also seen (H&E, x100)

Papilloma Virus may have a role in inducing malignancy, though the association still needs to be verified.^[12]

CONCLUSION

We suggest that HP occurring on the cheek region is a peculiar and interesting event, as it is not commonly associated with this region of the face.

Though ectopic forms of this tumor do occur in other parts of the body, apart from the typical perianal and genital areas of females. Their management remains the same more or less. Malignant transformation of these lesions is rarely reported and there is a need to investigate it further.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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