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ECTOPIC HIDRADENOMA PAPILLIFERUM ASSOCIATED WITH PILOMATRICOMA - A CASE REPORT

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ABSTRACT: Hidradenoma papilliferum is a rare, benign adnexal tumor considered to be of apocrine origin that occurs almost exclusively in females on the anogenital area. Ectopic or non- anogenital hidradenoma papilliferum is rare. But ectopic hidradenoma papilliferum associated with pilomatricoma has not been described in literature to our knowledge. We describe a case of hidradenoma papilliferum associated with pilomatricoma presenting with an intradermal nodule, 1cm in diameter on the right side of the back of the neck with history of 15 years duration. This case is reported because of its rarity.

KEYWORDS: Hidradenoma papilliferum, pilomatricomas, adnexal tumor.

INTRODUCTION: Hidradenoma papilliferum is a benign, cystic papillary tumor considered to be of apocrine origin that occurs almost exclusively on the skin of the anogenital region in middle aged females.¹ Even though it generally behaves in a benign fashion, rare malignant changes in the form of aggressive adenosquamous or squamous cell carcinoma can occur. Non-anogenital or ectopic hidradenoma papilliferum is rare and malignant change does not occur.² Pilomatricomas (calcifying epithelioma of Malherbe) are benign appendage tumors related to hair cells matrix.³ They are usually solitary, asymptomatic, intradermal nodules with predilection for head and neck region. A 52 year old male with ectopic hidradenoma papilliferum in the neck associated with pilomatricoma is presented here.

CASE REPORT: A 52 year old male with an intradermal nodule of 1cm diameter on the right side of the back of the neck reported for Fine needle aspiration cytology (FNAC) examination. The overlying skin exhibited a bluish discoloration. History of more than 15 years duration with gradual increase in size was given.

Aspiration smears showed a moderately cellular smear with a monomorphic population of benign looking epithelial cells in dispersed and loose clusters. A tentative diagnosis of benign adnexal lesion was made. Subsequent histopathological examination using hematoxylin and eosin (H&E) stain showed a well circumscribed dermal nodule surrounded by a fibrous capsule with no connection with overlying epidermis. The nodule consisted of a cyst with large arborizing papillae having fibrovascular cores. The papillary folds were lined by a double layer of cells consisting of an inner layer of secretory (columnar) cells showing evidence of decapitation secretion and an outer layer of small cuboidal cells having basophilic nuclei, which are myoepithelial cells (Figure 1). A few heterotrophic apocrine glands were also seen adjacent to the tumor. A diagnosis of hidradenoma papilliferum was made. Histochemically, the luminal cells contained many large PAS positive diastase resistant granules in the secretory cells (Figure 2). Myoepithelial nature of the outer cell layer was confirmed by expression of smooth muscle actin and calponin (Figure 3). In the dermis, there were

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many cystic spaces lined by basaloid cells with shadow cells on the luminal surface and central eosinophilic areas of keratinization. The basaloid cells were round to oval with basophilic nuclei and scanty cytoplasm. Shadow cells were eosinophilic with a central unstained area as a shadow of the lost nucleus (Figure 4). Focal multinucleated foreign body type giant cells and calcification were also present representing reaction to keratin. The findings were consistent with pilomatricoma.

DISCUSSION: Hidradenoma papilliferum occurs as an intradermal nodule of 1cm or less in diameter but may be as large as 1.5 cm.⁴ It occurs almost exclusively on the skin of the anogenital region in middle aged females. Lesions occurring in anogenital region in males have also been reported.⁵ It usually occurs in third to fifth decades of life.⁶

Ectopic hidradenoma papilliferum is rare, of which 60% occur in head and neck region.^{4, 6-8} Other sites are extremities, chest and back.^{2, 4, 9} It has been postulated that these ectopic lesions arise in modified apocrine glands.² It may be true in this case as a few ectopic apocrine glands were seen near the tumor. Association of hidradenoma papilliferum with nevus sebaceous of Jadassohn has also been described in literature. But such association is not observed in this case. In contrast to anogenital lesions, nearly one-half of ectopic cases are seen in males. The median age for ectopic cases is 1 to 2 decades older than the average age range of onset in anogenital lesions. Eighty five percent of cases are 1.5 cm in diameter or smaller. Clinical features, treatment and prognosis for lesions occurring in anogenital and ectopic region are similar.⁴

Pilomatricoma is the most common hair follicle tumor. It occurs predominantly in children and young adults but are now being increasingly recognized in adults and the elderly.^{10, 11} Slight male predilection has been reported in some studies.³ Though it generally occurs as a solitary lesion, multiple occurrences have been reported.¹² The most common site was head and neck.^{3, 13} Most of the cases were less than 2 cm in diameter. Malignant change is rare and tends to occur in middle-aged or elderly patients.¹³ Such changes are not observed in this case.

To conclude, ectopic hidradenoma papilliferum in the neck associated with pilomatricoma is a rare occurrence. These should be considered in the differential diagnosis of skin nodules on the head and neck, particularly in older patients for accurate diagnosis, appropriate treatment and to rule out malignancy.

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Fig. 1: Hidradenoma papilliferum (H&E, X10)

Fig. 2: Hidradenoma papilliferum (PAS, X100)

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Fig. 3: Hidradenoma papilliferum (Calponin, x40)

Fig. 4: Pilomatricoma (H&E, x40)

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