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Verrucous carcinoma and Syringocystadenoma papilliferum of thigh – An unusual collision tumor

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Abstract

Collision tumor is an entity used to describe the occurrence of two separate primary tumors at one anatomic location. Collision tumors occur at different locations in the body including skin. Syringocystadenoma papilliferum (SP) is an adnexal tumor of apo-eccrine differentiation and is juxtaposed to a nevus in one third of the cases and to a basal cell carcinoma in 10% of the cases. We report an unusual collision of SP and verrucous carcinoma occurring at an unusual location.

Keywords: Syringocystadenoma papilliferum, Verrucous carcinoma, Thigh, Skin

Introduction

Merging of two separate primary tumors at one anatomic location occurs rarely. There is no site or type specification for these tumors. Reports of collision tumors occurring at multiple locations including gastrointestinal tract, genitourinary tract, endocrine organs like thyroid and adrenals are on record.(1) The colliding tumors can be either both benign, both malignant or a combination of benign and malignant. Individual component of these collision tumors can be independent of each other, i.e., both having different cell of origin. If not so, the two lesions may arise from a single pleuripotent stem cell.

In cutaneous pathology, the commonest combination includes basal cell carcinoma occurring with nevus, seborrheic keratosis and neurofibroma. Adnexal tumors are also known to juxtapose with other neoplasm, but the association is rare.(2)

Syringocystadenoma papilliferum (SP) is an adnexal tumor of apo-eccrine duct differentiation and is juxtaposed to a nevus in one third of the cases and to a basal cell carcinoma in 10% of the cases.(3) However the occurrence of SP with verrucous carcinoma is very unusual with only two such cases published in the literature.(4,5) Identification of both the lesions in a biopsy is essential for adequate future management.

Herein we report a rare coexistence of SP with verrucous carcinoma detected serendipitously in a single biopsy. The report is presented because of its rarity and to put forth the fact that thorough examination of the biopsy provided is essential to increase the chance of identifying collision tumors.

Case Report

A 45 year old male patient presented with a mass on the posterior aspect of left thigh of one year duration. There was no history of associated fever, pain, rapid increase in the size of the swelling, oozing from the growth or trauma to the site. On local examination the swelling was 4 x 3 cm, covered with skin which was unremarkable. On palpation, the swelling was firm and non-tender. There was no evidence of regional lymphadenopathy. A clinical diagnosis of benign soft tissue tumor was made. The lesion was excised and sent for histopathological examination.

Gross: We recieved skin covered mass measuring 4 × 2 × 2 cm. Cut surface was solid grey white a small area of 1 × 1 cm which was brownish and different from the rest of the lesion. Entire tissue was embedded.

Microscopy: Sections revealed a composite tumor of exophytic and endophytic component. The lesion consisted of stratified squamous epithelium showing hyperkeratosis, parakeratosis, papillomatosis and acanthosis. The endophytic component showed squamous epithelium extending into deeper tissue as bulbous pushing margin. The second tumor showed an invaginating tumor composed of papillary and tubular pattern which was lined by bilayered epithelium. The epithelium was made up of outer tall columnar and inner layer of small cuboidal cells with plenty of plasma cells in the stroma. (Fig. 1, 2, 3 and 4)

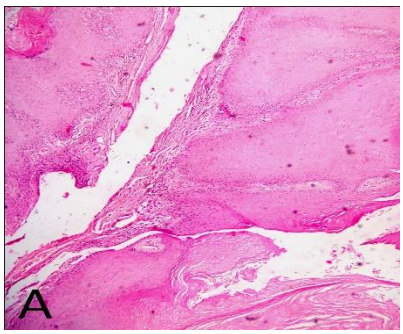

Fig. 1: Section shows verrucous carcinoma with an endophytic component- squamous epithelium extending into deeper tissue as bulbous pushing margin. (Hematoxylin & Eosin, × 40)

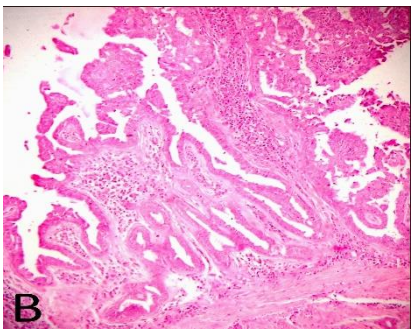

Fig. 2: Section shows an invaginating tumor composed of papillary and tubular pattern which was lined by bilayered epithelium - syringocystadenoma papilliferum. (Hematoxylin & Eosin, × 40)

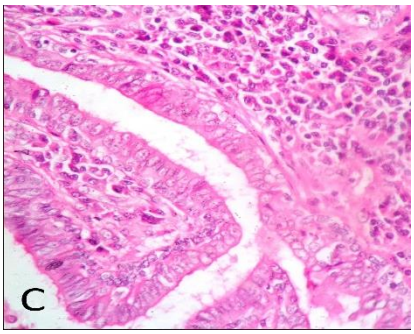

Fig. 3: Section shows the high power view of figure 3 with epithelium made up of outer tall columnar and inner layer of small cuboidal cells with plenty of plasma cells in the stroma. (Hematoxylin & Eosin, × 100)

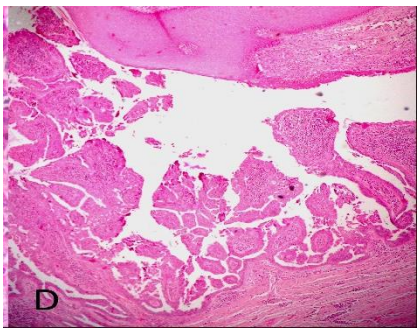

Fig. 4: Sections shows the collision of syringocystadenoma papilliferum and verrucous carcinoma in one focus. (Hematoxylin & Eosin, × 40)

Hence a final diagnosis of collision tumor - SP with verrucous carcinoma was made.

Discussion

SP is a rare adnexal tumor commonly located in the head and neck region. Its occurrence in the thigh is unusual and till date only nine cases have been reported.[6] It is a tumor of childhood and adolescence and rarely presents in elderly age group. In one third of the cases SP is associated with nevus sebaceous and in 10% of the cases with basal cell carcinoma.[3] Many benign tumors are known to coexist with SP. These include sebaceous epithelioma, apocrine hydrocystadenoma, trichoepithelioma, and eccrine spiroadenoma.[7] There are occasional cases reports of association of SP with squamous cell carcinoma, verrucous carcinoma and ductal carcinoma.[8,9] Verrucous carcinoma is a Human Papilloma Virus (HPV) associated tumor, commonly located in the mucosal sites like oral cavity and reproductive tract. Rarely, it can occur in the skin, commonly over the foot and occasionally on the nail.[10] Clinically and histologically oral lesions grow as infiltrative masses while perineal lesions demonstrate an exophytic growth. Microscopically the cytological features are well differentiated and this leads to a misdiagnosis of keratinous cyst. At this point, it is prudent to remember that recreation of a three dimensional image of a two dimensional picture is essential for a pathologist, especially while interpreting skin biopsies. Several biopsies and proper clinical correlation is desirable to reach at a correct diagnosis.

Histogenesis of SP is controversial. Apocrine mode of differentiation has been proposed based on IHC and EM studies. Origin of verrucous carcinoma has been attributed to HPV infection, irritation and inflammation.[3]

Coexistence of SP and verrucous carcinoma has been rarely reported. English literature reveals only two reported cases the details of which are tabulated below.

Table 1: Details of two reported cases of SP and verrucous carcinoma

Case Number	Age	Site	Duration of lesion in Years	Year of publication	Follow up
1	Elderly male	Thigh	15 years	1987	-
2	62 Years	Sacral area	Since birth (increase in size since 6 months)	2002	Unremarkable

In the previous and the present case the lesions were treated by wide surgical excision without any evidence of recurrence or metastasis.

Conclusion

Identification of the existence of collision tumor is essential. This can be achieved by complete embedding of the tissue which will help maximizing the yield. Similarly careful vigilance at the time of grossing to look for any different appearing area is a critical link to precise microscopic interpretation.

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