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Case Report

Trichoepithelioma of thigh. Third reported case

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ABSTRACT

Trichoepithelioma a small benign tumor derived from basal cells in the hair follicle. A trichoepithelioma can undergo malignant transformation into the basal cell carcinoma. The recognition of trichoepithelioma is important because of its close resemblance to basal cell carcinoma and other skin adnexal tumors, both clinically and histopathologically. Here we present case of 40 year old male presenting with painless swelling in right thigh. Wide excision of swelling was done and sent for histopathology which showed the swelling to be trichoepithelioma.

Keywords: Trichoepithelioma, Basal cell carcinoma, Skin adnexal tumor, Brooke's tumor

INTRODUCTION

A trichoepithelioma is a rare hair follicle tumor with mixed epithelial and mesenchymal proliferations, consisting of basaloid epithelial strands and cellular fibromyxoid stroma. It is a benign adnexal neoplasm and common sites comprise the face, head and neck, with rare occurrence at the thigh region. The gene involved in the familial form of trichoepithelioma is located on band 9p21. An abnormality in this gene may result in one of three syndromes Brooke-Spiegler syndrome, familial cylindromatosis, and multiple familial trichoepithelioma.[1] We can find about thirteen reported cases in literature, and there is two reported trichoepithelioma of the thigh. Our case is the 3rd cases reported trichoepithelioma of thigh to our knowledge.

CASE REPORT

A 40 year male patient came with complaint of pedunculated swelling in mid portion of right thigh since 3 month. The local examination revealed a pedunculated pigmented swelling measuring 3 × 2.5 cm (Figure 1). It was a firm, non-tender swelling which bleed on touch. The general physical and systemic examinations were normal. There were no palpable lymph nodes in the right inguinal region. The wide excision of the tumor was done and sent for histopathology (Figure 2). Histopathology and microscopy report show tumor arising from stratified squamous epithelial lining displaying segmentation of epithelium in the form of abortive pillar differentiation showing one or more layers of basaloid cells arranged in cords and strands. Individual cell are bland with very low mitotic activity that is feature suggestive trichoepithelioma.

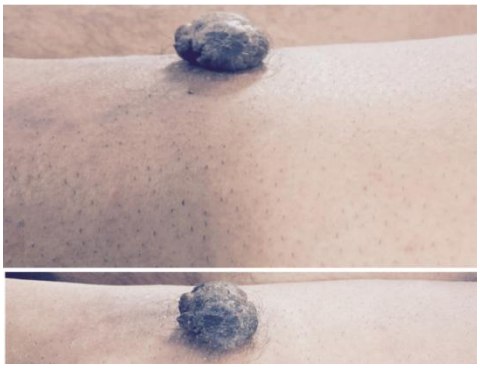

Figure 1: Local examination revealed a pedunculated pigmented swelling measuring 3 × 2.5 cm.

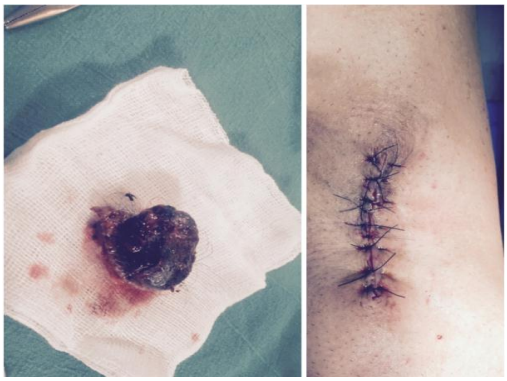

Figure 2: The wide excision of the tumor was done and sent for histopathology.

DISCUSSION

Trichoepithelioma also known as Brooke's tumor/ Epithelioma adenoids cysticum is a commonly encountered entity midway between trichofolliculoma and keratotic basal cell carcinoma in its degree of differentiation towards mature hair structure.[1] It probably arises from a pluripotential cell. Trichoepitheliomas occur as multiple skin colored papules of size 2-4 mm over the nasal cleft. Solitary lesions occur as subcutaneous nodules. Giant solitary trichoepithelioma of size >2 cm occurs rarely. Mostly giant solitary trichoepithelioma occur around the perianal region.[2] This case is presented for its rarity and location (Table 1).

Table 1: Previous case reports of giant solitary trichoepithelioma.

Age (years)	Sex	Site	Duration (years)	Tumor size (cm)	Recurrence/Follow up	Reported by
58	M	R Thigh	20	8	None/?	Czernobilsky et al.1972 [4]
70	M	Nose	?	2.5x1.5	None/1 year	Dvir E et al. 1981[7]
53	M	R Thigh	3.5	6.5x4.5x3	None/9 months	Filo GB et al. 1984[3]
77	F	Natal cleft	7	3.5x3.5x2.5	None/18 months.	Tatnall FM et al. 1986[2]
71	M	Buttock	Many	5x3.5x2.5	None/ 1 year	Tatnall FM et al. 1986[2]
70	F	Natal cleft	10	3.5x2.5x2	None/6 months.	Tatnall FM et al. 1986[2]
31	M	Scrotum	-	2	Recurrence/17 years	Beck S et al. 1988[8]
-	-	Scar	0.5	3	None/?	Beck S et al. 1988[8]
67	F	Abdomen	15-20	17x8	None/?	Oursin C et al. 1991[9]
48	M	L shoulder	?	4x2x1	None/3.5 years	Jemec C et al. 1999[10]
80	M	R side of nose	1	3x2	None/?	Krishnamurthy J et al. 2010[11]
45	F	R Forearm	25	9.5x4x2.5	None/6 months	Goyal et al. 2012[12]
80	M	R Elbow	50	2.8x2.5x2.5	?/ ?	Murugananthan et al. 2013[13]
40	M	R Thigh posterior aspect	0.25	3x2.5	None/1 year	Present case

Giant solitary trichoepithelioma needs to be differentiated from keratotic basal cell carcinoma. Trichoepitheliomas present as dermal tumor composed of basophilic cells that have the same appearance as the cells in basal cell carcinoma, except that they tend to lack high grade atypia and mitosis. Horn cyst shows abrupt keratinisation called "trichilemmal" keratinisation. The tumor islands composed of basophilic cells that are arranged in lace like or adenoid and in solid aggregates. These tumor islands show peripheral palisading of their cells surrounded by dense fibroblastic stroma without retraction artefact typical of basal cell carcinoma. Both adenoid and solid aggregates show invaginations, which contain numerous fibroblasts and resemble follicular papillae, also known as papillary bodies.[14] Basal cell carcinomas show predominant basal cell type, peripheral palisading of lesional cell nuclei, specialized stroma and clefting artefact between the epithelium and the stroma. In more than 90% of basal cell carcinomas, a connection between tumor cell formations and the surface epidermis can be shown to exist.[15] Basal cell carcinomas showing differentiation toward hair structures are called keratotic basal cell carcinoma. Keratotic basal cell carcinoma shows parakeratotic cells and horn cysts in addition to undifferentiated cells. The horn cysts, which are composed of fully keratinized cells, represent attempts at hair shaft formation.[16]

CONCLUSION

Trichoepitheliomas share s with basal cell carcinoma the presence of horn cysts, and it is sometimes difficult to decide whether a lesion represents keratotic basal cell carcinoma or a trichoepithelioma. In such a situation, clinical data may be necessary to reach a diagnosis. As trichoepitheliomas are benign lesions, surgical excision is curative and hence it needs to be differentiated from malignant keratotic basal cell carcinoma. Close follow up

of the cases of giant solitary trichoepithelioma is required as there is possibility of recurrence and rarely malignant transformation.

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