Proliferating trichilemmal tumor - Case report

Tumor triquilemal proliferante - Relato de caso

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Abstract: Proliferating trichilemmal tumor (PTT) is an uncommon neoplasm arising from the follicular isthmus. Its histological characteristic is the presence of trichilemmal keratinization. PTT usually presents as a solitary nodule on the scalp of elderly women. We describe a case of a PTT on the gluteal region (buttocks) of a 16-year-old female, presenting as a solitary nodule.
Keywords: Cysts; Histology; Buttocks

Resumo: Tumor triquilemal proliferante é uma neoplasia incomum que surge do istmo folicular, cuja característica histológica é a presença de ceratinização triquilemal. Apresenta-se usualmente como nódulo solitário no couro cabeludo de mulheres idosas. Descreve-se um caso de tumor triquilemal proliferante que se apresenta como lesão tumoral nodular na região glútea de uma jovem de 16 anos de idade.
Palavras-chave: Cistos; Histologia; Nádegas

INTRODUCTION

Proliferating trichilemmal tumor (PTT) is an uncommon neoplasm, first described in 1966 by Wilson and Jones [1] as a "proliferating epidermoid cyst." Most lesions occur on the scalp of elderly women as a solitary nodular lesion. [2-4] We report an unusual case, rarely reported in the international literature, of a TTP which presented as a nodular tumor located on the buttock of a young female patient.

CASE REPORT

16-year-old female patient reported the appearance five months ago of a pruritic papule on the gluteal region, which gradually evolved as a tumor, with episodes of bleeding at the site. Dermatological examination showed a pedunculated, cracked, bleeding and ulcerated tumoral lesion measuring about 2.5cm located on the patient´s left buttock (Figure 1). We performed complete excision of the lesion. Histological examination revealed an intradermal proliferation with lobular architecture formed by clusters of pleomorphic epithelioid cells with eosinophilic cytoplasm and nuclear atypia, surrounded by a fibrous pseudocapsule (Figure 2). The tumor presented foci of acantholysis and areas consisting of a set of clear cells with keratinized centers, representing trichilemmal differentiation (Figures 3 and 4). The stroma exhibited numerous vessels and granulation tissue on epidermal hyperplasia. The correlation of clinical findings with histopathology confirmed the diagnosis of PTT.

DISCUSSION

TTP is an uncommon adnexal neoplasm differentiating towards the follicular outer root sheath epithelium. It is believed that it is caused by a trichilemmal cyst (TC). TTP and CT possess trichilemmal keratinization as a histological marker (abrupt transition from epithelial nucleated cells to anucleate, keratinized cells, without the formation of granular layer). [2] The TTP are generally larger and more atypi-

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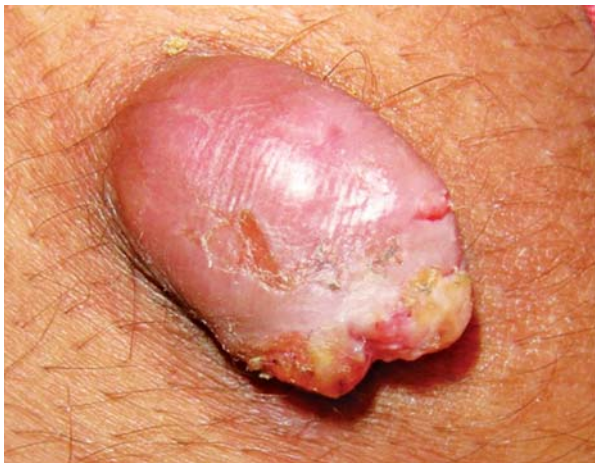

Figure 1: Pedunculated ulcerated tumor on the left buttock

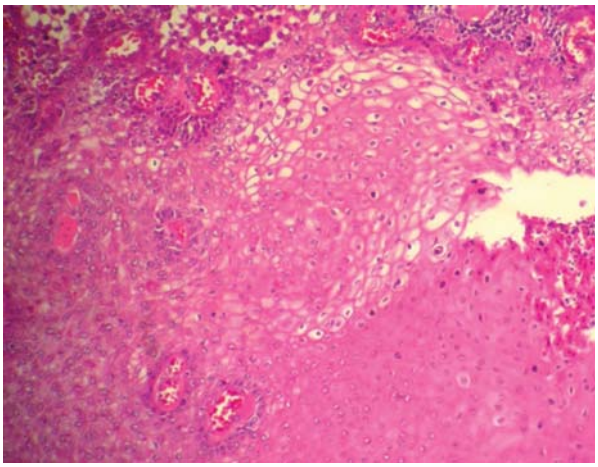

Figure 3: Cluster of clear cells with trichilemmal-type keratinization and ectatic blood vessels

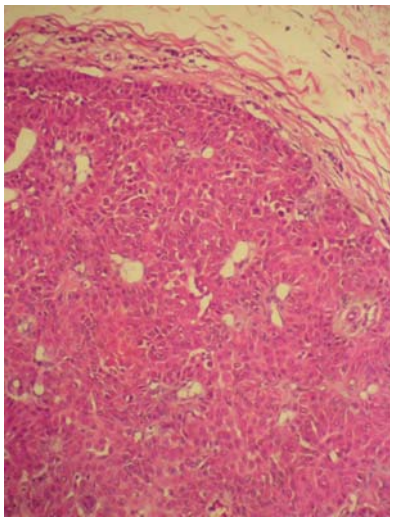

Figure 2: Fibrous encapsulated lobular cluster of neoplastic , epithelioid, eosinophilic, pleomorphic cells, with nuclear atypia

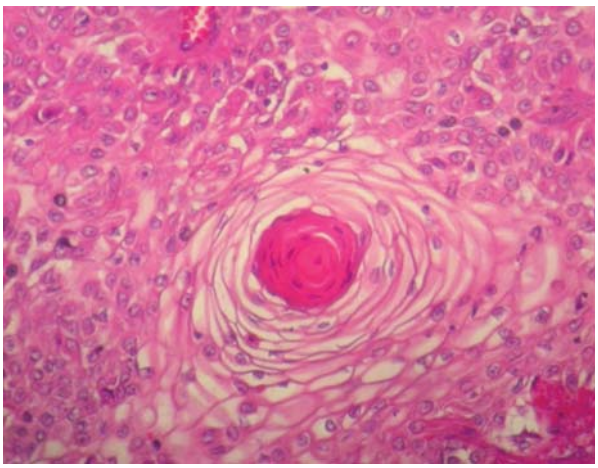

Figure 4: Cluster of clear cells with trichilemmal-type keratinization

cal when compared with CT and in some cases may simulate squamous cell carcinoma. [3, 4] About 90% of cases involve scalp lesions, described clinically as slow growth solitary nodular lesions. [3,4,5] Other less common locations include the neck, trunk, armpits, groin, vulva, lower and upper limbs, upper lip and buttocks. [2,6,8] Reports exist of multiple lesions, ulceration and bleeding [2,5]. Women are more affected than men, with age at onset of over 60, although cases have been reported in young people. [3,5,6,9] The tumor is characterized histologically by the proliferation of basaloid and squamous cells with abrupt trichilemmal keratinization and varying degrees of cytologic atypia. [5] Some cells may appear clear or vacuolated and the periphery of the epithelial cells displays PAS-positive eosinophilic membrane. [5] In the majority of reports it is characterized as a benign lesion. [4,10] Reports exist of clinical and histological malignant forms with local or lymph node spread but these are rarely hematogenous. [5,10 to 12] The higher risk of metastasis is present when the lesion occurs in areas other than the scalp, when it is fast-growing and infiltrative, is over five centimeters in diameter and/or presents cytological atypia and mitotic activity. [10] The treatment of choice is surgical resection with a safety margin of one centimeter. [4,9] Due to the high local recurrence rate, Mohs micrographic surgery would appear to be a therapeutic option. [13] ❑

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