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Clear Cell Hidradenoma of the Gluteal Region: A Case Report

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SUMMARY Clear cell hidradenoma is a rare skin appendage tumor. A 41-year-old female presented with right gluteal mass. Excisional biopsy of the mass was performed. Under the epidermis, an eosinophilic-cytoplasm, uniform-appearance, oval-round-nucleus, benign tumor with cystic and solid components was detected. These results were consistent with clear cell hidradenoma. The patient had not been given postoperative adjuvant treatment and has been under follow up free from disease for 2 years.

KEY WORDS: clear cell, hidradenoma, hip

INTRODUCTION

Clear cell hidradenoma is among the skin appendage tumors and gluteal localization is rare (1). Generally, it grows slowly and appears as a solitary dermal nodule under the skin for many years. It is mostly a benign, occasionally capsular (2,3), well-limited neoplasm containing solid and cystic components together.

CASE REPORT

A 41-year-old female presented to our clinic with the complaint of gradually increasing swelling that had been present in her right hip for 3 years. Her physical examination revealed a 4x4-cm mass in her right gluteal region. No regional lymphadenopathy was detected and other system examinations were normal. Excisional biopsy of the mass was performed with 1 cm clear margin. Macroscopically, tissue sample was of a cystic nature. Pathological examination revealed a well-limited, eosinophilic-cytoplasm, benign neoplasm with a thin fibrous capsule under the epidermis that constituted solid cell layers within the cystic space and was composed of oval-round-nucleus cells (Fig. 1). Immunochemistry showed positive staining with cytokeratin (Fig. 2), PAS (Fig. 3) and p63 (Fig. 4). Based on these results, the diagnosis of clear cell hidradenoma was made. Wide surgical excision was not required because the tumor

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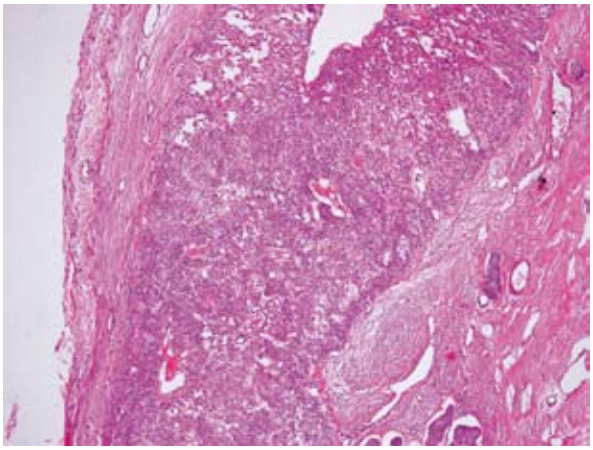

Figure 1. Benign tumor under the epidermis with a thin fibrous capsule, eosinophilic cytoplasm and oval-round nucleus (hematoxylin and eosin; X20).

was excised with a wide and safe margin on all sides. Adjuvant therapy was not administered. The patient has been under follow up at six-month intervals, free from recurrence on tomography taken at the last control visit. Considering the localization site, only one case has been reported in the literature besides this one.

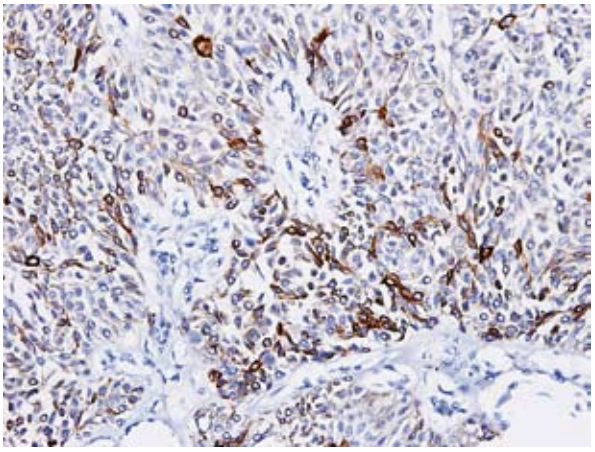

Figure 2. Positive cytoplasmic staining with cytokeratin (X40).

DISCUSSION

Although it can occur nearly at any age, clear cell hidradenoma is mostly frequently diagnosed from the fourth to eighth decade of life (4). It is more common in women than in men (5). The first case in the literature was described by Delacretaz *et al*. in 1958 (6). There are different opinions on its origin. Some authors suggest that it is of eccrine origin, based on the number of mitochondria, excess glycogen granules, electron microscopy images showing microvillus processes, and enzyme histochemistry (7). Many different terms are used for clear cell hidradenoma, i.e. clear cell myoepithelioma, nodular hidradenoma, eccrine sweat gland adenoma of clear cell type, solid cystic hidradenoma, and eccrine acrospiroma (7,8). However, more emphasis is put on the opinion that it is mainly of apocrine origin (9,10). In general, there are solitary lesions with a 1- to 2-cm diameter. However, there also are larger in size and multiple cases reported in the literature. Commonly, they tend to be located in the upper part of the body such as the head, face, upper extremity and abdomen (5,11,12).

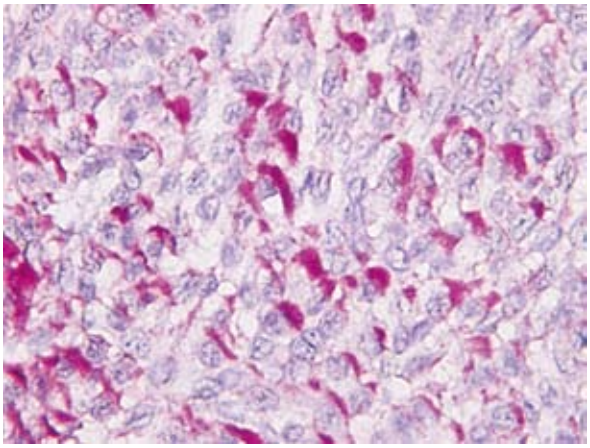

Figure 3. Positive cytoplasmic staining with PAS (X100).

Pathologically, clear cell hidradenoma consists of two cell types. The first includes epidermal polyhedral-structure cells with generally round nuclei and mildly basophilic cytoplasm. The second includes round and transparent-cytoplasm clear cells (8,11). Cystic areas probably result from degeneration of tumor cells.

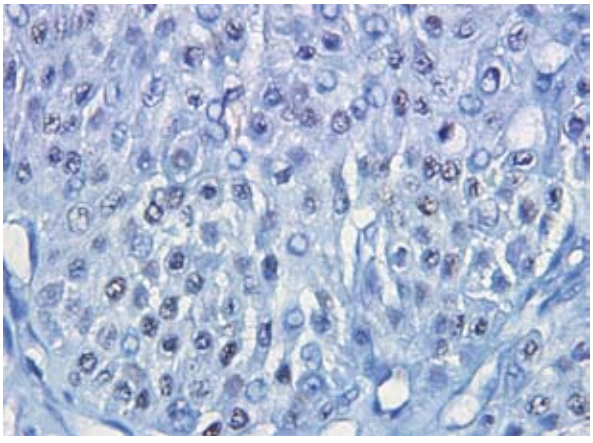

Figure 4. Positive nuclear staining with p63 (X100).

Malignant form is rare. It is characterized by nuclear atypia, mitosis, necrosis and invasion to the surrounding tissue (5,11). Malignant transformation may occur after inadequate surgical excision. Metastasis sites commonly include lymph nodes, bones and lungs (2,13).

Fine-needle aspiration biopsy is not enough to make the diagnosis. Excisional biopsy is required for both diagnosis and treatment. Immunohistochemistry staining is a supplementary technique in diagnosis. While clear cell hidradenoma exhibits a positive staining with cytokeratin, PAS and p63, it shows negative staining with S100, CD10, CD34 and vimentin (2,3,11,14). Our patient had the same immunohistochemistry staining results.

Although optimal treatment has not yet been definitely identified, the currently accepted approach is extensive surgical excision with negative surgical margins (4,11). There is no adjuvant treatment recommended postoperatively. Adjuvant treatment options including chemotherapy and/or radiotherapy have only been proposed for malignant clear cell hidradenoma. Therefore, we did not use any adjuvant treatment in our patient. We put our patient under follow up after extensive surgical excision with wide and safe margin to leave intact tissue space. Until now, no recurrence was detected on physical examination and tomographic images taken at six-month intervals.

Local recurrence rate is high and may be observed following inadequate surgical resection (2,11). In recurrent cases, surgical re-excision is usually performed (13).

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