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Case Series

Clear cell hidradenoma: a series of three cases

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ABSTRACT

Described by various terms as clear cell hidradenoma, eccrine acrospiroma and clear cell myoepithelioma, clear cell hidradenoma is an uncommon adnexal tumor of eccrine or apocrine differentiation and common anatomic sites encountered are the head and neck region, trunk and extremities. It usually presents as a single papule, nodule or mass and are covered by intact skin or have ulcerative look. Occasionally, the development of brown, blue or red discoloration with superficial ulceration and serous discharge may mimic malignancy. Peak incidence of the disorder is fourth to eight decades, although all ages can be affected. On histopathology, tumour shows periodic acid–Schiff-positive clear cells and polyhedral cells. Recurrence rate is low if the lesion is completely excised. Here, we report three cases of clear cell hidradenoma presenting as a solitary nodule on the scalp, lower back and eyelid. The aim of this study is to create awareness about the various clinical presentations and morphological spectrum of this tumour as it can be a potential mimicker for some malignant tumours/metastasis particularly when it occurs in uncommon sites.

Keywords: Clear cell hidradenoma, Eccrine, Adnexal tumour

INTRODUCTION

Clear cell hidradenoma is an uncommon benign tumor of eccrine glands and a variant of hidradenoma first described by Liu in 1949.[1] They are usually seen as solitary, well-defined, non-encapsulated, slow and self- limited growth pattern in the dermal layer of head, face and extremities, rarely show aggressive clinical behavior.[2] The lesions are most commonly red, but may also be brown or skin colored and has a preponderance in females.[3] The tumor may be solid or cystic in varying proportions, however clear cell hidradenoma is considered to be a solid tumor. Histologically, the tumor is composed of lobulated masses located in dermis and extending into subcutaneous fat.[4] It is composed of two types of cells, PAS-positive polygonal clear cells surrounded by myoepithelial cells.[5] Wide local excision is the treatment option. It has recurrence rate of approximately 12%, which may be attributed due to incomplete resection but rarely undergo malignant transformation.[6] The common differential diagnosis of a clear cell hidradenoma on clinical examination are epidermal inclusion cyst, ganglion cyst, vascular malformation, peripheral nerve sheath tumor, synovial sarcoma and basal cell sarcoma. This tumor can be a potential differential diagnosis for malignant tumors/ metastasis when occurs in uncommon sites. Hence aim of this study is bring to the attention about various clinical presentations and morphological spectrum, and the lesion should be differentiated where clear cells may be present.[7]

CASE SERIES

The first case was a 26-year-old female who presented with an asymptomatic, globular, slow-growing lesion over the scalp for 3 years duration. On clinical examination it was a 2.5 cm single, soft swelling over the temporal region

of scalp. A clinical impression of pilar cyst was given and complete excision was performed. We received the excised specimen in the histopathology department. On gross examination, the specimen received was single, skin covered, grayish nodule measuring 2.5x1.5x1 cm. On cut section, the lesion was gray -white, solid cystic.

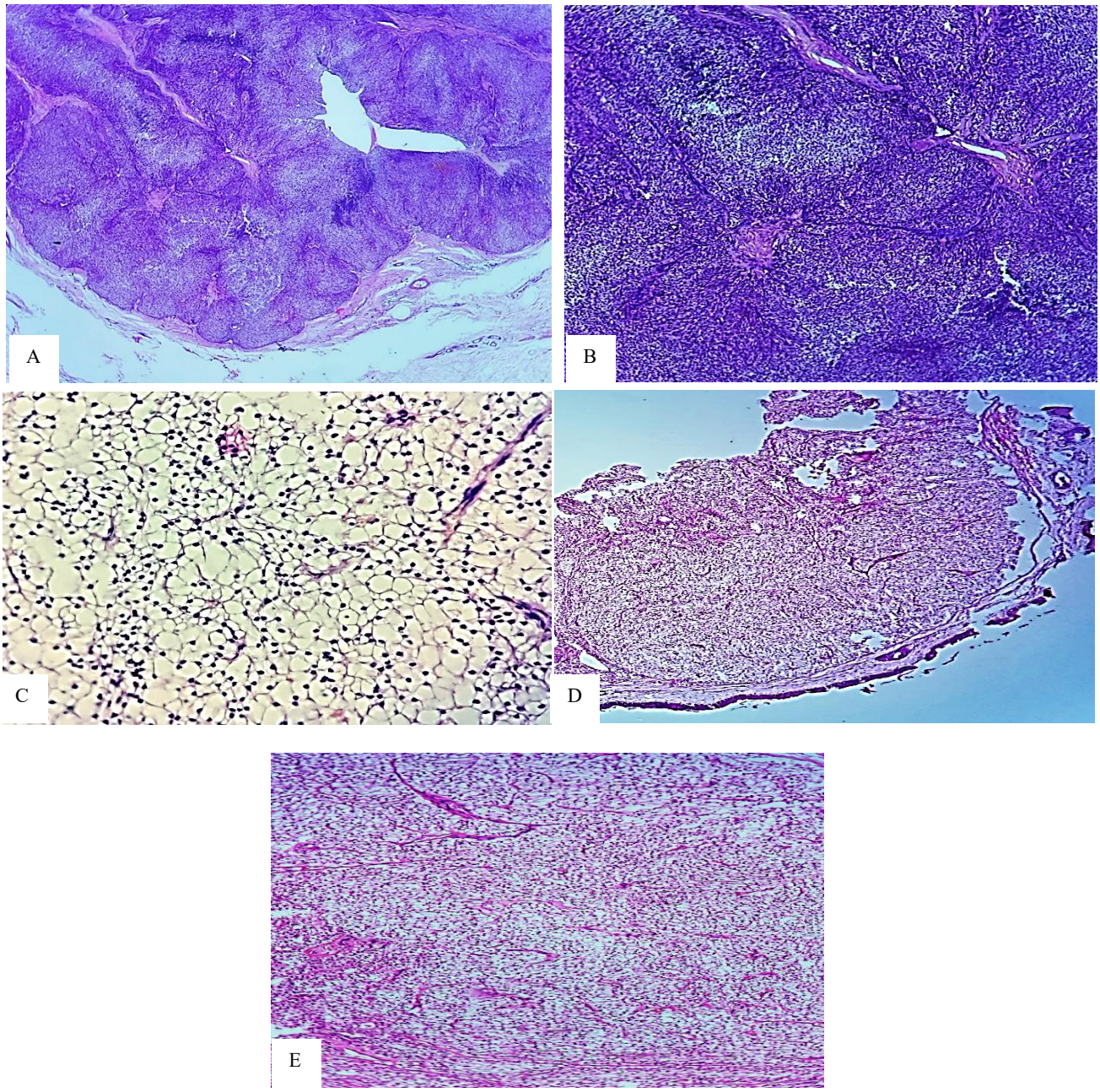

Figure 1: (A) low power (40X) view of H and E stained showing well circumscribed lobulated nodule, (B) histopathology showing fibrovascular core surrounded by eosinophilic cells, which show oval nucleus, bland nuclear chromatin. Clear cells show abundant clear cytoplasm and small eccentric nucleus (H and E x 100), (C) histopathology showing prominent clear cells with vacuolated cytoplasm (H and E x 100), (D) low power (40X) view of H and E stained showing epidermis with underlying well circumscribed lobulated dermal nodule, (E) histopathology showing pale and clear cells with eccentric nuclei and eosinophilic polyhedral cells (H and E x 100).

The second case was a 24-year-old male who presented with an asymptomatic solitary lesion over the lower back for 3 months duration. Clinically it was a solitary, non-tender, soft tissue swelling in the lower back with restricted mobility. It was diagnosed as epidermoid cyst and was excised. Grossly, the specimen received was a single, skin covered, nodular mass measuring 1.2x1.1x1 cm. The lesion was solid cystic and filled with whitish fluid with areas of hemorrhage on cut surface. The third

case was a 28-year- old male who presented with complaint of a single asymptomatic right lower lid mass for 1 month. There was no history of trauma or bleeding or pain. It was a 0.6 cm firm, painless nodule near the outer canthus of right lower eyelid. The left eyelid was normal with no other complaints. Clinically it was suspected to be dermoid cyst. The nodule was excised and sent to histopathology department. We received a single, partially skin covered, grey brown nodular tissue measuring 0.6 x0.5x0.5 cm. On cut section, the nodule appeared solid grey white.

In all the three cases there was no evidence of regional lymphadenopathy or history of trauma or pain. All of them underwent complete surgical excision of the nodules. All these cases showed similar histomorphological features. Histopathological examination with H and E stain was done in all three cases. Microscopy revealed well circumscribed, non-encapsulated dermal tumor showing lobules of tumor cells separated by collagenous stroma. The tumor showed dual population of cells nests of clear cells with abundant cytoplasm admixed with polygonal cells with granular cytoplasm. The clear cells showed abundant clear cytoplasm with a small, round eccentric nucleus. The elongated cells were round to polygonal, with round to ovoid nucleus, small distinct nucleoli and finely granular eosinophilic cytoplasm (Figure 1). There were no atypia, necrosis or mitoses seen. The clear cells were stained with periodic acid Schiff's (PAS) reagent due to presence of glycogen or a diastase resistant substance in cytoplasm and absence of lipid. All these above features were suggestive of a benign clear cell hidradenoma. Immunohistochemistry was not done as the diagnosis was definitive on histopathology.

DISCUSSION

Clear cell hidradenoma (CCH) is a rare sweat gland tumor. Liu first described in 1945 as clear cell papillary carcinoma of the skin.[8] Previously described as eccrine differentiation, it is now accepted that these can have apocrine origin as well.[9] Hernandez-Perez and Cestoni-Parducci revealed more common during fourth to eighth decade of life and twice as common in women as in men and can be seen in all age groups.[10] Although infrequent, malignant transformation is seen in 6.7% cases.[6] It is marked clinically by rapid growth with cutaneous ulceration and histologically by nuclear atypia, increased number of mitotic figures, infiltrative pattern with area of necrosis.

Clinically CCH presents as single, small, slow- growing, well- demarcated, intradermal tumor that varies in size from 0.5 to 3 cm.[11] Occasionally, there may be a cystic appearance. The lesions may be skin- colored, red, blue or brown. Some tumors ulcerate and bleeding surface, while few discharge serous material.[6] Hidradenomas have histomorphology variants such as nodular hidradenoma, eccrine acrospiroma, solid-cystic hidradenoma, clear cell hidradenoma and clear cell acrospiroma. Immunohistochemistry analysis is not required in routine cases as most cases are reliably diagnosed on histopathology with H and E-stained sections.[12]

Clear cell hidradenomas have biphasic cellular population of round, fusiform to polygonal cells admixed with clear cells having abundant cytoplasm. On immunohistochemistry, CCH shows reactivity for p63, p40, CK5/6 and AE1/AE3. Negativity for SMA and S100. Immunostaining for EMA and CEA highlight ductal differentiation when present.[13] Metastatic clear cell carcinomas including renal, lung or thyroid cell carcinomas are differential diagnosis of clear cell hidradenoma in adults. Nodules of hidradenoma lacks classic vasculature which is prominent in malignant deposits of renal cell carcinoma and the later are distinguished by their positivity to thyroid transcription factor-1 (TTF-1).[14] The other differential diagnosis of clear cell tumors in dermis includes metastatic and primary skin tumors such as salivary gland tumors. The malignant counterpart has a characteristic infiltrative tumor, cellular atypia and numerous atypical mitoses.[4] Our all the three patients were followed up for 6 months with no recurrence.

CONCLUSION

The diagnosis of clear cell hidradenoma solely based on the clinical appearance of the lesion is challenging. The sites of occurrence are non-specific and lack any distinctive clinical attributes. Thus, histopathology is important for diagnosis of these lesions. Complete surgical excision with wide margins is recommended for this tumor. As there is high rate of local recurrence, hence regular postoperative follow-up is important.

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