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Case Series

A study on clinicopathological spectrum of nodular hidradenoma

Krishnan Govindaraman Padmanaban*

Department of Pathology, Thanjavur medical college, Tamil Nadu Dr. MGR Medical University, Chennai, India

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***Correspondence:**
Dr. Krishnan Govindaraman Padmanaban,
E-mail: padhunkg@gmail.com

ABSTRACT

Nodular hidradenoma (NH) is an uncommon benign skin adnexal tumor that presents usually as solitary intradermal nodule. This study aims to discuss the various clinicopathological features of NH as it mimics other adnexal tumors, malignancies or metastasis particularly in uncommon sites. Twenty-four cases of histo-pathologically diagnosed NH during study period from 2016 to 2021were taken up for study. Their clinical, gross and microscopic findings were collected from records. Most tumors occurred in head in fifth decade. Cystic change was seen in majority of the cases. Tumor had dual population of cells with predominant clear cell component in five cases. Definitive cytological diagnosis was difficult requiring excision biopsy. Characteristic clinical and histological features help to distinguish NH from other potential mimickers NH need wide excision as they can recur and rarely transform in to malignancy.

Keywords: Hidradenoma, Clear cell, Adnexal tumor, Pathology

INTRODUCTION

Nodular hidradenoma (NH) is a benign skin adnexal neoplasm that exhibits eccrine differentiation.[1] Described by various terms as clear cell hidradenoma, eccrine acrospiroma and clear cell myoepithelioma. Apocrine origin has also been proposed for clear cell variations of tumor. It can arise from various anatomic sites, commonly seen in head, neck, extremities and trunk region. It frequently occurs in 4-8th decades of life though all ages can be affected.[2] NH usually presents as a solitary intradermal nodule that may or may not be ulcerated. [1]

The aim of this study is to create awareness about the various clinical presentations and morphological spectrum of this tumor as it can be a potential mimicker for some malignant tumors/ metastasis particularly when it occurs in uncommon sites.[2]

CASE SERIES

It's a descriptive cross sectional retrospective study carried out in the department of pathology, Thanjavur medical college and hospital from January 2016 to December 2021 after obtaining ethical committee approval. All cases diagnosed as NH by histopathological examination during the study period were included in the study. Clinical data such as age, sex, site, clinical presentation was obtained from case records. Gross findings including size of tumor, presence of cystic change, ulceration and microscopic picture were noted from pathology records and reviewed.

A total of 24 cases of NH were included in this study. Age at presentation varied from 9 years to 78 years with majority of them seen in 5th decade (37.5%). Gender distribution was equal. Site, age, gender distribution, size and pathological findings of the cases in our study are given in Table 1. Majority of the cases occurred in head (50%) followed by trunk (25%).

Most of tumors between 2-4 cm in size (16 cases-66.7%). Grossly, all cases presented with nodular swelling except 1 with warty growth over chest wall, 2 cases had surface ulceration in elbow and cheek, and cystic change was observed in more than half of the cases (13 cases-54.2%).

Microscopically, all tumors had dual population of cells in varying proportions-polygonal cells with clear cytoplasm and cells with pale eosinophilic cytoplasm (Figure 1). Clear cell component was predominant in five cases (20.8%), mostly in head region as well as were diagnosed as clear cell hidradenoma-clear cell hidradenoma (Figure 2). Similarly, five cases had squamous metaplasia (Figure 3). Melanin pigmentation, oncocytic change (Figure 4), calcification, myxoid changes were also noted.

Table 1: Clinicopathological profile of nodular hidradenoma.

Site	Age (years)	Gender (M-male, F-female)	Size (cm)	Other findings
Vulva	51	F	4×3	Oncocytic change
Thigh	55	F	2×1	Pigmentation
Inguinal region	78	M	5×3.5	CCH, cystic change
Eyebrow	55	M	2×2	Cystic change
Scalp	76	F	3×2	Squamous metaplasia
Breast	41	M	6×5	Cyst+
Scalp	24	F	3×2	Cyst+
Vulva	48	F	2×2	Squamous metaplasia
Chest wall	57	M	3×3	Warty, squamous metaplasia
Elbow	25	M	4×3	Ulceration, squamous metaplasia, cyst+
Anterior abdominal wall	26	M	2×2	Cyst+
Lip	60	F	1.5×1	Cyst+
Face	46	M	3×1	Squamous metaplasia
Back of trunk	12	M	2×2	Cyst +
Preauricular	56	F	2×2	Clear cells
Trunk	40	M	2×2	CCH
Thigh	25	F	3×3	Cyst +
Postauricular	40	M	2×2	CCH
Scalp	23	M	2.5x2	Cyst +
Scalp	60	M	3×2	CCH
Cheek	60	F	6×5	Ulceration, myxoid change, calcification
Face	55	F	2×2	CCH
Breast	35	F	4×4	Cyst +
Scalp	9	F	1×1	Cyst +

CCH- clear cell hidradenoma.

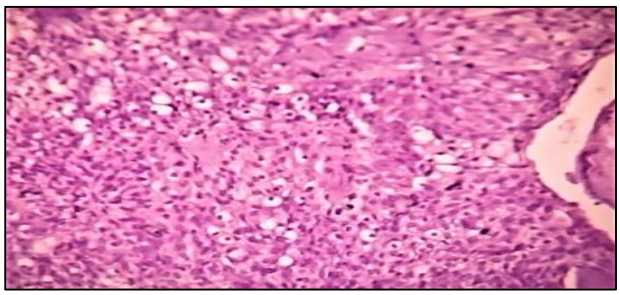

Figure 1: Dual population of cells, H and E stain, 100x.

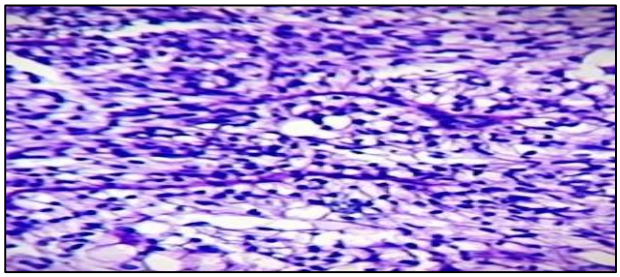

Figure 2: Prominent clear cells with vacuolated cytoplasm, H and E stain 100x.

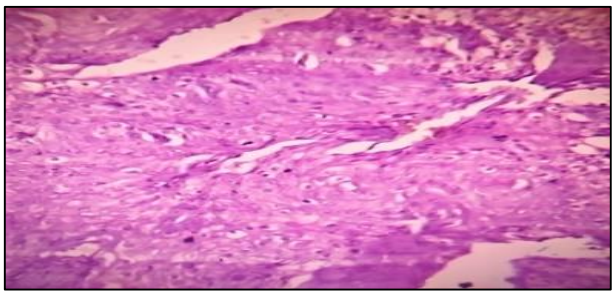

Figure 3: Prominent squamous metaplasia in NH, H and E stain 100x.

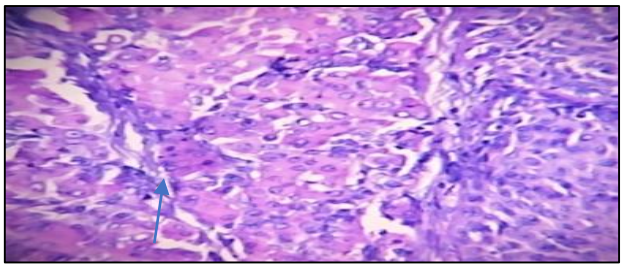

Figure 4: Oncocytic cells with abundant eosinophilic cytoplasm (arrow), H and E stain 100x.

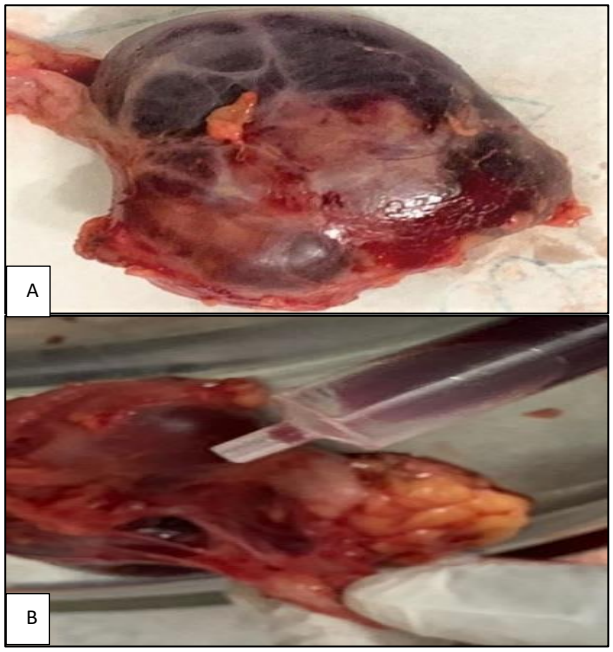

Figure 5 (A and B): Post operative picture of excised inguinal mass. Cut surface of inguinal mass showing cystic component.

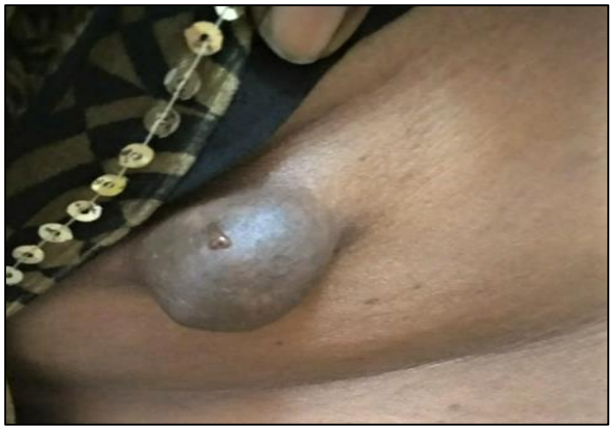

Figure 6: Nodular mass in inframammary region.

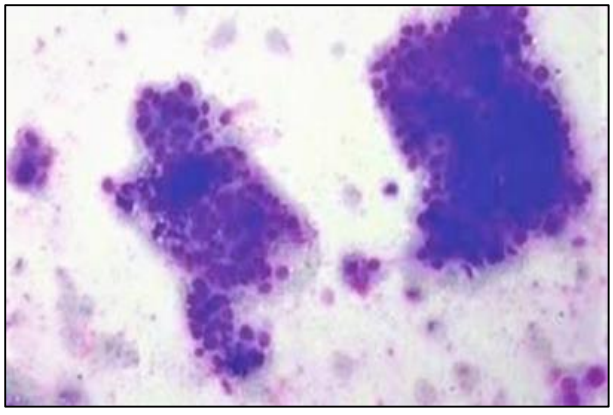

Figure 7: Smear shows clusters of epithelial cells with eosinophilic cytoplasm. May Grunwald Giemsa (MGG) stain 100x.

DISCUSSION

NH s were historically interpreted as a lesion of eccrine lineage, current information indicates that it may be of either apocrine or eccrine origin: some cases involve decapitation secretion that is classically observed with apocrine origin, whereas other cases exhibit ultrastructural features suggestive of eccrinedifferentiation.[3] Hidradenoma with sebaceous differentiation are probably best thought of as reflecting folliculosebaceous-apocrine lineage.[4] NH are commonly encountered on face, upper extremity, axilla, trunk, thigh, scalp and pubis.[5] It can occur in all ages, commonly females in fourth to eighth decades of life.[2] However, we encountered equal sex distribution of cases. Ozawa et al reported clear cell hidradenoma of the forearm in a 5-year male child.[6] Youngest case in our series was in a 9-year female child occurring in scalp. Tumor size varies from 0.8 cm to 8 cm.[3] NH usually presents as intradermal nodule with intact skin, although ulceration with leakage of serous fluid may be seen.[7]

Histologically, it is composed of solid and cystic spaces. Solid areas consist of two types of cells: Predominant cell type is the polyhedral cells with more basophilic cytoplasm and the round cells with a clear cytoplasm due to the presence of glycogen.[3,7]A preponderance of clear cells, as seen in CCH, is noted in less than one third of hidradenomas, as observed in this study.[7] The clear cells chiefly contain glycogen and in addition periodic acid-Schiff (PAS)-positive, diastase resistant material.[1,7,8] Epithelial lobules may have tubular lumina and cystic spaces, which are lined by cuboidal or columnar cells and may contain decapitation secretions. Cystic spaces are formed as a result of tumor degeneration and are filled by eosinophilic material. Squamoid and sebaceous differentiation may be seen. The stroma is more commonly fibrous or hyalinized but myxoid or chondroid stroma may rarely be seen.[3,7,8] A case of vulval NH had oncocytic differentiation (Figure 4). Roth et al reported a rare case of oncocytic hidradenoma and this variant does not carry any prognostic significance.[9]

We encountered a CCH presenting as inguinal mass in 78-year male. Clinically, it was mistaken for lymphnode swelling suspecting metastasis. Fine needle aspiration cytology yielded straw coloured fluid and had only cyst macrophages in smear. Excision biopsy revealed a solid - cystic mass (Figure 5 A and B). Histologically, it had predominantly clear cells (Figure 2), cyst macrophages and no lymphoid cells were noted. Possibility of renal primary was ruled out by radiological evaluation.

We had another case of NH in 35 years female, in inframammary region, clinically suspected as carcinoma of breast (Figure 6). FNAC obtained straw coloured fluid and cytology smear revealed clusters of epithelial cells with eosinophilic cytoplasm admixed with cyst macrophages (Figure 7), so excision biopsy suggested which turned out to be NH. Superficial location in dermal plane

and characteristic morphology in biopsy were helpful in arriving at a definitive diagnosis in both cases. However, in difficult cases, immune-ohistochemical panel including CK 5/6, p63, p40, ER, and PR may be helpful in distinguishing NH from breast carcinoma (negative for CK5/6); and carbonic anhydrase 9 (CAIX), renal cell carcinoma, CD10 negative immunoreactivity differentiates NH from metastatic RCC.[3]

In literature diagnosis of NH on cytology is rarely reported. Most of cases are inconclusive or misinterpreted on FNAC.[3,8,10] Cytologically, presence of dual population of cells-eosinophilic and clear cells, three-dimensional clusters with papillary-like fronds, tubules, rosette-like structures and extracellular hyaline material are the key features in NH.[11]

Malignancy in NH can be de novo or a transformation from benign tumor, is seen in up to 7% of cases. They are aggressive tumors with a high frequency of local recurrence and metastasis. Metastatic lesions have been reported in the regional lymph nodes and the lung.[12] They exhibit necrosis, marked cytologic atypia, increased mitosis including atypical forms, infiltrative borders, vascular invasion, perineural invasion.[3]We did not encounter any malignant NH in our study.

Immunohistochemistry (IHC) is not routinely required as most cases can be reliably diagnosed with h hematoxylin and eosin (H and E) stain.[2] Hidradenoma does not demonstrate a specific phenotype due to the combination of different types of cells. They express strongly and diffusely p40, p63, CK5/6, whileS100 and SMA are usually negative. Immunostaining for EMA and CEA highlights ductal differentiation when present. Most cases demonstrate MAML2 (master mind-like 2) gene fusion.[13]

As NH lesion is limited, surgical resection is usually used for the treatment. But incomplete removal may lead to recurrence, so surgical excision with adequate margins and close follow up is recommended.[2,14] House et al have suggested the use of the Mohs micrographic surgery for recurrent or large hidradenoma, with encouraging results, but it is not widely available in developing nations.[15]

CONCLUSION

NH is an uncommon skin adnexal tumor with potential of aggressive behaviour. Characteristic clinical and histological features help to distinguish NH from other potential mimickers. Emphasize benefit of wide surgical excision with appropriate margins to prevent local recurrence.

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