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MALIGNANT NODULAR HIDRADENOMA: A REPORT OF TWO CASES AND REVIEW OF THE LITERATURE

Pathology

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ABSTRACT

INTRODUCTION: Malignant nodular hidradenoma is a rare tumor of sweat gland.It was first reported as clear cell eccrine carcinoma by Keasbey and Hadley in 1954.The biological behaviour of the tumor is aggressive and are highly prone for recurrence.

CASE REPORT: We report two cases of malignant hidradenoma,one presented with a scalp swelling and other chest wall swelling.FNAC and biopsy was done in both the cases.The histomorphology was confirmed by running a panel of immunohistochemical marker and the diagnosis of malignant hidradenoma was ascertained.

DISCUSSION: Sweat gland tumors are mostly benign.Malignant nodular hidradenoma according to WHO classification comes under malignant tumor of skin appendages with apocrine and eccrine differentiation.MNH displays an infiltrative or invasive pattern,atypical mitosis,necrosis & angiolymphatic invasion which favours its malignant nature.It can take its origin from a pre-existing hidradenoma or can arise denovo.The tumor is extremely rare with less than 50 cases ever reported in literature.The prefered management is early wide excision with atleast 2cm clear margin for both primary disease and local recurrences.

KEYWORDS

Cock's peculiar tumor, malignant hidradenoma, scalp, sweat gland tumor

INTRODUCTION:

Malignant tumor of the sweat glands represents rare oncological entities,characterized by nonspecific clinical presentation and equivocal pathological features. Their precise diagnosis and histological classification can be very difficult[1]. Malignant hidradenoma is a rare tumor of sweat gland. It was first reported as clear cell eccrine carcinoma by Keasbey & Hadley in 1954.[2]Clear cell hidradenoma is an extremely rare tumor with lee than 50 cases reported[3,4].

The biological behaviour of the tumor is aggressive & are highly prone for recurrence. Hidradenoma arise as intradermal nodule from eccrine sweat glands. Ultrastructural and enzyme histochemical studies have shown nodular hidradenoma to be intermediate between eccrine poroma and accrine spiradenoma[5].

We report two case of malignant nodular hidradenoma, one presented with a scalp swelling and other as chest wall swelling.

CASE REPORT:

Patient 1: patient 1 was a 80 year male who presented with a swelling over scalp gradually increasing in size since 1year. The swelling was of size 15x15x10 cm , firm in consistency, with a ulceration over it. A clinical diagnosis of cock's peculiar tumor was given and sent our department for FNAC. Cytosmears were cellular with cells arranged in sheets, discohesive clusters and also dispersed singly(fig 1a & b). Individual cells were moderate to large sized with abundant eosinophilic cytoplasm,round to oval large nuclei, high N:C ratio and prominent nucleoli. The background containing plenty of inflammatory cell seen and an impression of malignant adnexal lesion with superadded inflammation was given. Biopsy fromthe ulcer margin was taken. Histopathology revealed a grenz zone between epidermis and dermis(fig1c). Tumor cells are arranged in nodular pattern in dermis. Cells are almost uniform looking with ill defined cell outline and eosinophilic cytoplasm. Good number of cells show clear cytoplasm(fig1d). Nucleus is round to oval, vesicular, having prominent nucleoli. Areas of pleomorphic cells with frequent mitosis seen. Prominent cystic spaces containing macrophages and few tubular structures also present. Immunohistochemistry was performed for EMA,P^{53} and Ki 67. EMA was found to be positive(fig1e) around tubular structures, P^{53} was negative and Ki 67 decorated >14% of neoplastic cells(fig1f). From histopathology report presence of a clear cell malignant nodular hidradenoma arising from the sweat gland was ascertained. On follow up the patient was found to be dead 4 month later.

Patient 2: The second patient was a 48 year male with a chest wall swelling rapidly increasing in size over last two month. The swelling was of size 6x6x4 cm, firm in consistency, without ulceration. The draining area showed no palpable lymphnodes. Abdominal ultrasonography was normal. Incisional biopsy was taken and histopathology revealed pleomorphic polygonal cells present in nodular pattern with infiltrating border. Mitotic figures are frequent(fig2b). Areas of necrosis seen. A diagnosis of malignant nodular hidradenoma was made from the findings.

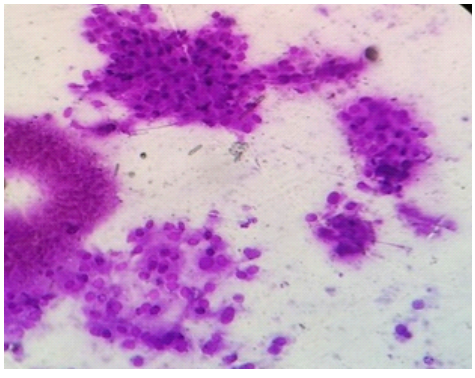

FIG 1 a:DIFFQUIK(X400)

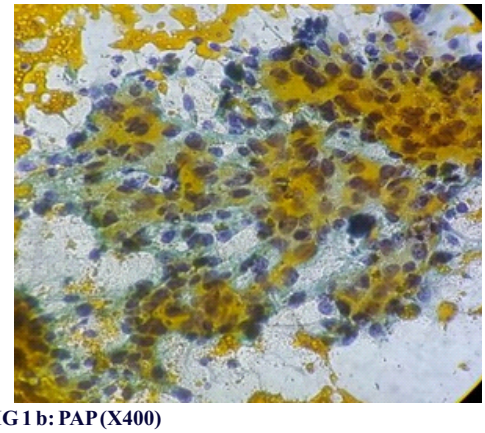

FIG 1 b: PAP (X400)

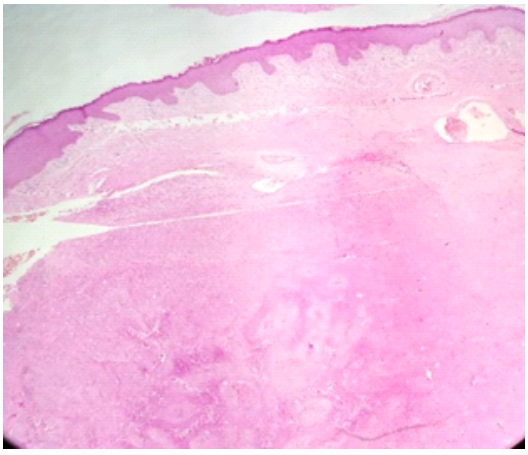

FIG 1c: H &E (X100)

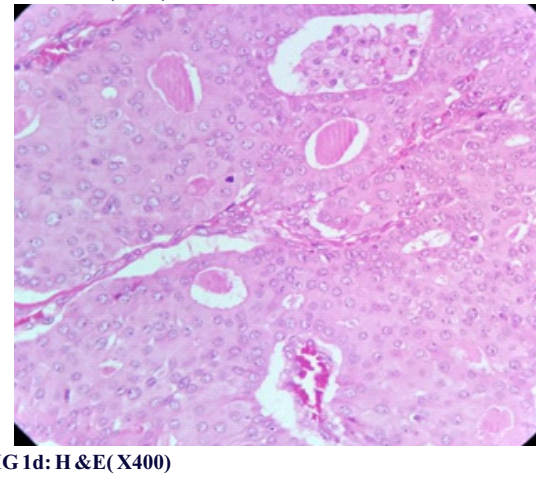

FIG 1d: H &E(X400)

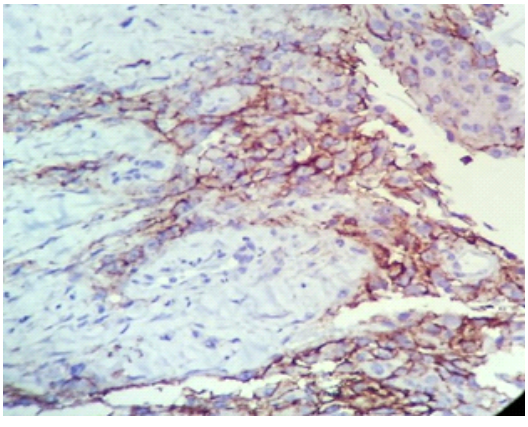

FIG 1e: EMA POSITIVE

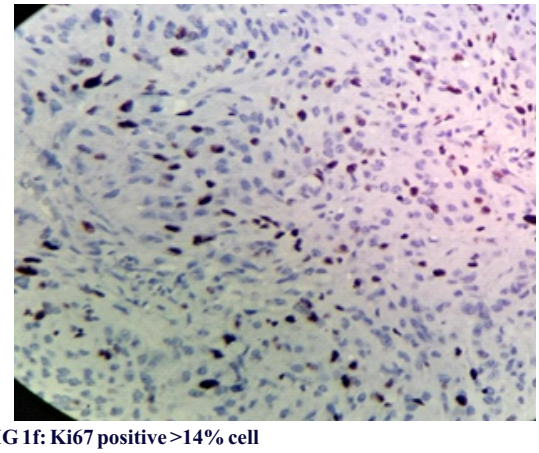

FIG 1f: Ki67 positive >14% cell

FIGURE 1.(a and b) showing FNA smear[scalp] of malignant adnexal lesion with superadded inflammation.(c) Grenz zone between epidermis and dermis. Tumor cells in nodular pattern in dermis.(d) Good number of cell showing clear cytoplasm. Prominent cystic spaces containing macrophages.(e) Immunostain showing EMA positivity around tubular structure. (f) Ki67 immunostain decorating >14% of neoplastic cell.

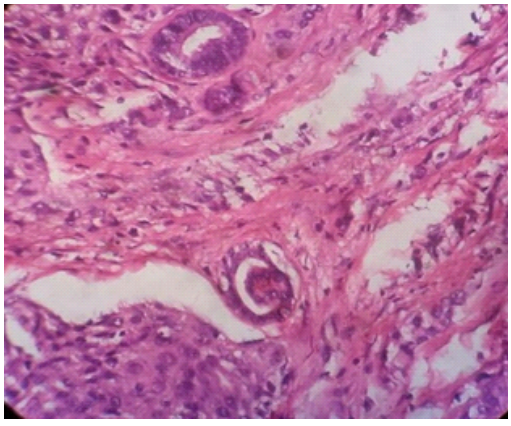

FIG 2a: H&E (X400)

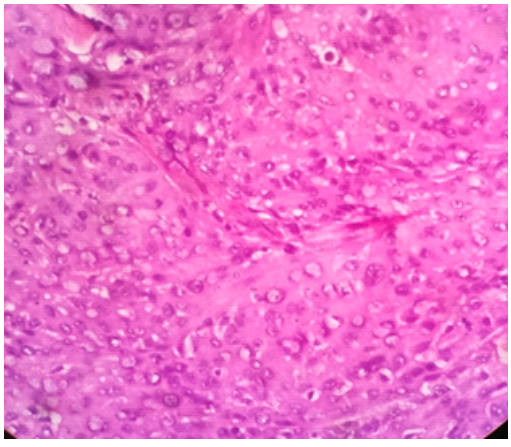

FIG2b: H&E(X400)

FIGURE 2[chest wall]: (a)Tubualr structure embedded in tumor cells.(b) Pleomorphic cell with mitotic figures.

DISCUSSION:

The recognition of hidradenoma as a distinct entity was first reported in 1941 by Mayer, whereas the term "clear cell hidradenoma" was proposed in 1954 by Keasbey and Hadley [2]. The malignant form of hidradenoma is extremely rare with less tha 50 cases ever reported in the literature. All these cases were characterized by a significant rate of locoregional recurrence. Some patient developed distant metastatic spread as well [4,6].

The disease is usually expressed as a small intradermal mass which remains inactive for a long period of time before increasing in size. Its aggressive behaviour is more apparent after each local relapse with faster growth and invasion of the surrounding tissue. In most cases no epidermal connection is encountered. Hidradenoma usually affects middle aged women, although its malignant form shows no age and gender predilection[7,8,9].

Malignant nodular hidradenoma according to WHO classification comes under malignant tumor of skin appendages with apocrine and eccrine differentiation. Sweat gland tumors are mostly benign. Histologically sweat gland may be either eccrine or apocrine in nature. Eccrine glands are present through out the skin but are most abundant in palm,sole &axilla. Apocrine glands are found mainly in axilla, around the nipples and anogenital region. The malignant nodular hidradenoma is an uncommon malignant cutaneous adnexal tumor that can show differentiation towards various components of eccrine sweat gland[7]. It is difficult to estimate the incidence and exact number of cases reported, as malignant nodular hidradenoma is described by varied names in the literature.

Histologically, clear cell hidradenoma seems to originate from the ductal epithelium of the sweat glands, whereas histogenetically it appears to represent a transitional tumor, sharing features of eccrine poroma and eccrine spiradenoma[7]. There is no clear distinction between eccrine and apocrine glands or between their malignant counterparts. The characterization of a sweat gland tumor, however, as of eccrine or apocrine origin, still remains useful[10,11]. Clear cell hidradenoma consists of two main cell subpopulations. The first one is composed of round or polygonal cells with round nuclei and clear cytoplasm, which is the result of abundant glycogen storage. The second subpopulation consists of multifaceted cells comprised of oval nuclei and basophilic cytoplasm evenly arranged at the periphery of the first cellular line. Clear cells may also be seen in other, more frequent malignancies of the head and neck, such as squamous and basaloid tumors, as well as in metastatic deposits of renal cell carcinoma[7]. Clear cell hidradenoma may contain keratinocytes forming keratin congregates. In addition, some cystic configurations noted on the specimen most probably represent empty spaces between degenerated cells rather than ductal formation of the tumor itself[7].

Although most cases of malignant nodular hidradenoma arise de novo, the tumor may also arise in pre-existing hidradenoma[12]. Malignant nodular hidradenoma also referred to as hidradenocarcinoma, malignant clear cell acrospiroma, clear cell eccrine carcinoma or primary mucoepidermoid cutaneous carcinoma. Malignant clear cell hidradenoma usually develope de novo and invades the dermis and subcutaneous tissue. The histology of malignant nodular hidradenoma is similar to that of its benign form. Malignant nodular hidradenoma displays a infiltrative or invasive pattern, atypical mitosis, necrosis and angiolymphatic invasion which favours its malignant nature[7].

The recognition of the eccrine origin of a malignant hidradenoma may be accomplished through specific immunohistochemical techniques, a positive PAS stain, as well as with the presence of lobules with epidermal differentiation[6]. The tumor cells stain positively for LMWK, and the ductal structure/luminal surfaces are highlighted by EMA & CEA. In our cases, the ductal structures are highlighted by EMA, p53 was negative and ki67 decorated >14 % of neoplastic cells. Interestingly, clear cell hidradenoma may occasionally mimic metastatic clear cell carcinomas including thyroid, lung or renal cell carcinoma. However, the first two are usually distinguished by their positivity to thyroid transcription factor-1 (TTF-1), and the latter by its prominent vascularity and the presence of haemorrhage and focalgranular necrosis within the lesion[13]. Renal cell carcinoma also express both EMA and Cd10.

Rosen etal [14], presented a case of a clear cell hidradenoma of the eyelid complicated by multiple recurrences and invasion of nearby structures. Histologically, no atypia or increased mitosis were found. Similar findings were noted in other case reports of clear cell hidradenoma[15].

Surgical excision remains the therapeutic modality of choice. Wong etal[6] supported wide surgical resection with atleast 2 cm of clear margin for both primary disease and local recurrences. Elective regional lymphadenectomy after lymphoscintigraphy should also be performed. Locoregional recurrence even after wide surgical excision has been reported inmore than 50% of cases.

CONCLUSION:

Malignant clear cell hidradenoma is a rare oncological entity, with no particular clinical or histopathological features. It should be included in the differential diagnosis of dermal lesions with an aggressive behaviour and multiple recurrences, despite aggressive surgical treatment.

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