Malignant Adnexal Tumour—Eccrine Hidradeno Carcinoma a Case Report

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Case Report

A 55 year old male patient presented with an ulceroproliferative lesion over the left thigh with left inguinal lymphadenopathy (Fig. 1a). Clinically it was suspected to be squamous cell carcinoma with inguinal lymphnode metastasis. Wedge biopsy of the lesion and fine needle aspiration cytology of the inguinal node (Fig. 3a) surprisingly revealed it to be Malignant adnexal tumour of skin with inguinal node metastasis. High frequency ultrasound scan and computed tomography revealed a malignant mass lesion in the subcutaneous plane of left thigh with left superficial inguinal lymphnode enlargement. Metastatic work up was done with base line chest X-ray and MRI abdomen and pelvis. Patient was treated with wide local excision (Fig. 1b) with left sided ilio-inguinal block dissection (Fig. 2a–d). Histopathology showed normal skin lined by stratified squamous epithelium (Fig. 3b) with neoplastic cells in the deeper levels arranged in nests which are tightly surrounded by fibrous septa (Fig. 3c) with infiltration into cutaneous vasculature. The neoplastic cells having scant to moderate amount of cytoplasm and the nuclei showing atypia and hyperchromatism, some showing prominent nucleoli. There is absence of decapitation reaction or comedone reaction (Fig. 3d). Based on the histologic features a final diagnosis of Malignant adnexal tumor of Eccrine differentiation (Hidradeno carcinoma) was made. All circumferential and deep resected margins free of tumor and 7/16 nodes showed metastatic deposits. Then the patient was subjected to post operative radiotherapy (70 Gy for primary field and 50 Gy for the inguinal region). The patient was followed up thrice a year with proper clinical examination. MRI of the

left thigh and pelvis and chest X-ray after a year showed no evidence of recurrence. The patient is still in follow up for more than 20 months and is in good control of the disease.

Discussion

Skin adnexa comprises hair follicles, sebaceous glands and sweat glands. The sweat glands are further classified into Eccrine and Apocrine glands. Sweat gland tumors are rare and among these eccrine carcinomas are commonest [1]. Wick et al. reported only 14 cases in a span of around 70 years [2]. Mehregan and associates reported 35 cases of eccrine carcinomas and only two had regional and distal metastatic features [3]. Hence Tumors of skin adnexa are of numerous types such as Hair follicle tumors, External root sheath tumors, Hair matrix tumors, Sebaceous gland tumors, Apocrine gland tumors, Eccrine gland tumour, etc. our discussion is confined to our case- Malignant hidradenoma which is a malignant Eccrine gland tumor.

Malignant hidradenoma is otherwise known as Hidradenocarcinoma or Malignant acrospiroma. It is defined as “a malignant tumor traditionally regarded as displaying eccrine differentiation and arising from a pre existing hidradenoma or de novo” [4]. These rare tumors seem to arise from ductal epithelium of the sweat glands and it represents a transitional tumor sharing features of eccrine poroma and eccrine spiradenoma [5]. This commonly occurs in elderly adults as slow growing ulcerated nodules in extremities with or without regional lymphnode involvement. Grossly they appear as connective tissue tumor, like a firm nodular dermal mass with or without skin changes. Rarely it may have an aggressive course with pulmonary metastasis. It represents less than 0.005 % of the skin tumors reported in literature. At times it may mimic metastatic carcinomas that may involve the skin. Around 50 % tumors recur after initial excision while

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Fig. 1 Wide local excision of the tumor (gloved finger in 1a pointing enlarged inguinal nodes)

**ADNEXAL TUMOR IN THIGH WITH
MALIGNANT LYMPH NODES**

a

WIDE LOCAL EXCISION

b

others spread to lymph nodes and distant sites in due course of time [6].

Pathologically eccrine tumors are characterized by infiltrative dermal proliferation of variable sized lobules and nests showing cyst formation. Composed of a mixture of eosinophilic polygonal cells, squamous cells, mucinous and clear

cells with no connection to the epidermis. Pleomorphism and necrosis with variable mitosis is common. Glycogen is demonstrated in the cytoplasm of some of the clear cells. It lacks Comedone reaction (which is a feature of sebaceous gland tumor) and Decapitation reaction (which is a feature of Apocrine gland tumor) [7]. Hence eccrine carcinoma is a

RADICAL ILIO INGUINAL BLOCK DISSECTION

a

b

COMPLETED BLOCK DISSECTION AND CLOSURE

c

d

RESECTED SPECIMEN

Fig. 2 Radical ilio inguinal block dissection and resected specimen

Fig. 3 Histo pathology **Table 1** Diagnostic algorithm of eccrine carcinoma

HENCE ECCRINE CARCINOMA IS A HISTO PATHOLOGIC DIAGNOSIS OF EXCLUSION

diagnosis of exclusion after ruling out sebaceous gland tumor and apocrine gland tumor. Ki-67 and p53 staining may be useful histologic parameters. Various markers such as GCDPF-15, CEA, EMA, S-100 protein, bcl-1, bcl-2, cytokeratin 5/7, cytokeratin 7 were studied with inconsistent results. It may be unnecessary to separate hidradenocarcinoma into eccrine and apocrine categories since closer examinations revealed existence of eccrine and apocrine counter parts as well (<http://www.ncbi.nlm.nih.gov/pubmed/23677677>). The histologic features of hidradenocarcinomas resemble the features of adenocarcinoma of breast and salivary gland. This led to investigations that used immunohistochemistry to compare HER2/*neu*, estrogen receptor, and gross cystic disease fluid protein 15 levels. The diagnosis of eccrine carcinomas should be considered with the staining of CK7, CK5/7, P63 and ER, PR receptors (<http://www.diagnosticpathology.org/content/8/1/15>). The relevance of HER2/*neu* positivity to hidradenocarcinomas and the use of trastuzumab in the treatment of hidradenocarcinomas remain to be further addressed.

Benign and malignant differentiation can be said by virtue of its poor circumscription, presence of nuclear atypia and high mitotic activity, infiltrative growth pattern and angiolymphatic permeation [8]. Hence diagnosis of malignancy through standard pathological examination may prove extremely difficult [9]. In our case we made a diagnosis of malignant hidradenoma with the presence of vascular invasion and high mitotic index (Table 1).

Eccrine gland tumors are treated with wide local excision with safe margins and Regional lymph node dissection which remains the main stay of treatment (<http://www.ncbi.nlm.nih.gov/pmc/articles/PMC3336933/>). Role of sentinel node biopsy and lymphoscintigraphy is controversial and is under trial. Small lesions near the vital structures are treated with

Mohs microsurgery [10]. Eccrine tumors have a predictable response to post operative radiotherapy and it avoids local recurrence (<http://www.ncbi.nlm.nih.gov/pmc/articles/PMC3336933/>). Role of chemotherapy is under trial. Strict follow up for recurrence is mandatory. The prognosis for survival with newly diagnosed hidradenocarcinoma is generally good and, notably the overall survival is around 74 % with the age adjusted survival was 94 %. Patients with differentiated tumors demonstrated good OS compared to its poorly differentiated/anaplastic counter parts (<http://www.ncbi.nlm.nih.gov/pubmed/23677677>).

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