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Unmasking the Uncommon: A Case Report on a Rare Malignant Adnexal Tumour

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ABSTRACT

Introduction: Malignant nodular hidradenoma, also called as hidradenocarcinoma is a rare neoplasm arising from the eccrine glands. The tumour has a slow onset with varied disease course and is generally associated with recurrence and metastasis. It usually affects head, trunk and extremities region and commonly seen in the fifth decade. Patient presents as a solitary nodule. These lesions can arise de novo as well as from a benign nodular hidradenoma.

Case summary: We present a case of a 49 years old lady who presented with a nodule on the right side of the upper abdomen since 10 years. The swelling was gradually increasing in size and was associated with watery discharge since 3 years and intermittent pain. Examination showed it to be a 3x4cm mass in the right hypochondrium, it was mildly tender and overlying skin was normal. Wide local excision was done with rhomboid flap. Histopathological examination along with Immunohistochemical studies concluded a diagnosis of Malignant hidradenocarcinoma.

Conclusion: Literature shows few cases of Malignant hidradenocarcinoma, since the incidence is low. As it is associated with poor prognosis, hence an accurate diagnosis is very essential for treatment of the patient.

Keywords: Hidradenocarcinoma, adnexal tumour, eccrine tumour.

INTRODUCTION

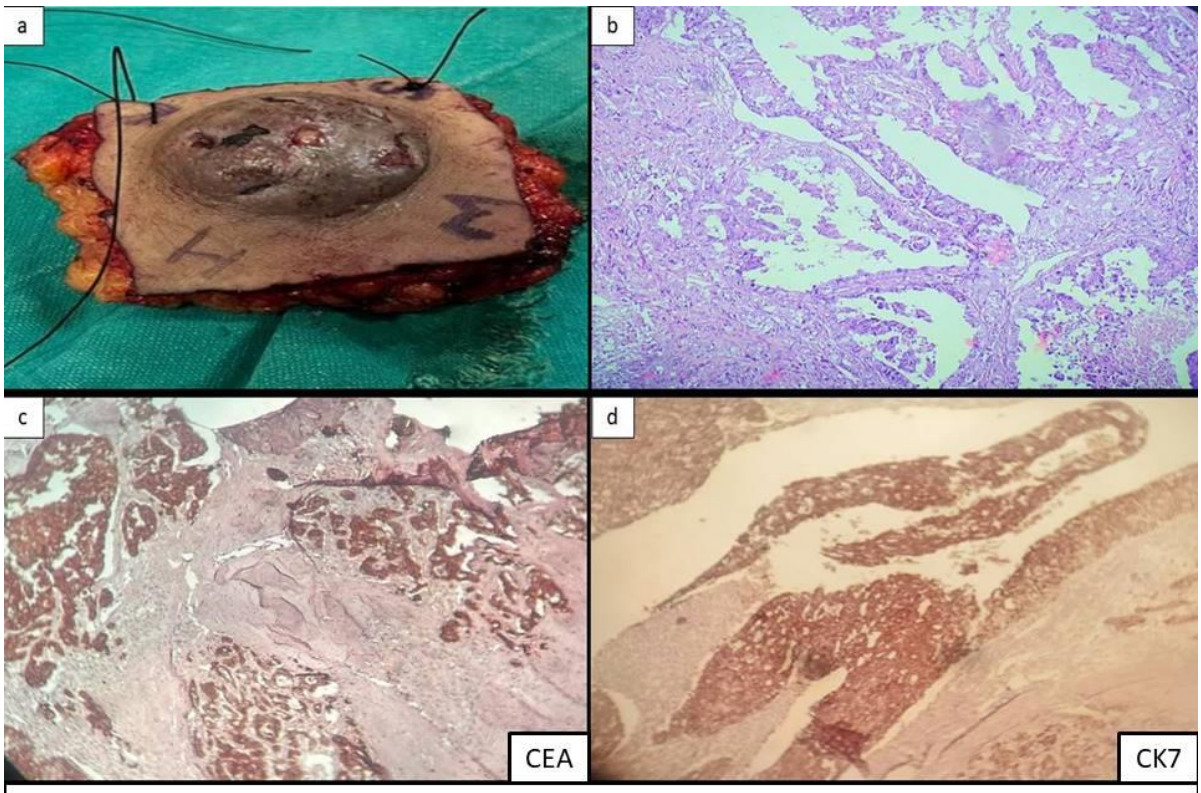
Malignant nodular hidradenoma, also called as hidradenocarcinoma or clear cell hidradenoma is a rare neoplasm arising from the eccrine glands. The tumour has a slow onset with varied disease course and is generally associated with recurrence and metastasis.[1] It usually affects head, trunk and extremities region and commonly seen in the fifth decade. Patient presents as a solitary nodule. These lesions can arise de novo as well as from a benign nodular hidradenoma. [2]

CASE REPORT

We present a case of a 49 years old lady who presented with a nodule on the right side of the upper abdomen since 10 years. The swelling was gradually increasing in size and was associated with watery discharge since 3 years and intermittent pain. No history of bleeding from this lesion. Examination showed it be a 3x4cm mass in the right hypochondrium, it was mildly tender, overlying skin was normal. CECT abdomen showed a well-defined heterogenously enhancing mass (3.5 x4.1cm) with areas of necrosis in the subcutaneous plane of anterior abdominal wall in the right hypochondrium with partially exophytic component. Overlying skin is irregular. No infiltration into the underlying muscle was seen. A possibility of malignant change in superficial

abdominal wall lesion was kept. Wide local excision was done with rhomboid flap.

Grossly, we received a well oriented skin covered soft tissue mass measuring 8x8x2 cms. The external surface showed a greyish white nodular growth measuring 3x3x2cms. Few ulcerations were also seen. Cut sections showed a well circumscribed greyish white to yellow tumor, soft to firm in consistency. The tumour was 1.5 cms from the superior margin, 2.8cm from the inferior margin, 2cm from the lateral, medial and deep margins each and 0.5cm from the anterior margin.



A. Gross image showing a nodular growth with few ulcerations. B. Photomicrograph showing trabeculae and nests of tumour cells.(100x) C. Photomicrograph showing diffuse positivity of tumour cells for CEA. D. Photomicrograph showing positivity for CK7. (100x)

Microscopically, the lesion was lined by keratinised stratified squamous epithelium. The dermis showed an invasive tumor comprising of solid and cystic areas. Tumour was arranged in the form of papillae, tubules, sheets and trabeculae. Tumour cells showed marked pleomorphism with vesicular to hyperchromatic nuclei, variably conspicuous nucleoli and moderate to abundant clear to eosinophilic cytoplasm. Atypical/ bizarre mitosis (6-7/10 HPF) were seen along with foci of necrosis. No heterologous elements were identified. PAS stain was done which highlighted granules in the cytoplasm of the tumour cells. All the surgical resection margins were free of tumour involvement. Immunohisto-chemistry was done which showed diffuse positivity in tumour cells for CEA and CK7 highlighted ductal differentiation. (figure 1) Keeping in view the above findings, diagnosis of Malignant hidradenocarcinoma was given.

DISCUSSION

Malignant nodular hidradenoma is a rare entity with few cases reported in the literature. Most commonly seen in fifth decade and lesion is seen on the head and neck region but few case reports also report rare sites like dorsum of hand, eyelid, scalp and lip.[3, 4] These tumours are associated with aggressive disease progression and with local recurrence and lymphatic invasion.[5,6]

Microscopic findings followed by immunohistochemistry are required for a correct diagnosis and to rule out other differential diagnosis. Microscopically it shows an invasive solid growth pattern with intracytoplasmic vacuoles, moderately to markedly pleomorphism, lymphovascular invasion and increased number of mitosis along with necrosis. [7] On Immunohistochemistry, it shows a positivity for p63, keratin AE1/AE3 and cytokeratin with high Ki67 proliferation rate while it is negative for CEA, S100, GCDFP-15 and EMA. [8,9]

Benign nodular hidradenoma, basal cell carcinoma with eccrine differentiation and metastatic renal or thyroid or lung carcinoma and squamous cell carcinoma are closed differential diagnosis which should be kept in mind. Nodular hidradenoma will be well circumscribed with less atypia and pleomorphism and mitosis. There will no necrosis. Basal cell carcinoma with eccrine differentiation will show peripheral palisading with no cystic areas, stromal epithelial retraction and with an epidermal connection. Metastatic renal cell carcinoma will show absence of ductal differentiation/sweat production and on immunohistochemistry will be positive for PAX8 and CD10 and negative for p63. TTF-1 will be positive for metastatic thyroid and lung carcinoma. Squamous cell carcinoma will show keratin pearls and have no secretory activity. [10] The choice of treatment is usually surgical resection followed by chemotherapy and radiation.

CONCLUSION

Malignant hidradenocarcinoma is a rare neoplasm with limited studies in the literature. Hence a correct diagnosis is very critical for the treatment and prognosis of the tumour

Declaration by Authors

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