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Solid – Cystic Hidradenoma: A Case Report

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Abstract: *Solid – Cystic Hidradenoma is a tumor of sweat gland origin with incidence rate of 5.1 cases per 1 million people annually [1] accounting it to be a rare case to be reported. We discuss here, a 24 year old female patient presenting with a cystic nodule in the thigh with clear cell changes in histomorphology. The differential diagnosis of clear cell renal cell carcinoma was ruled out based on morphological patterns. The case is reported here owing to the rarity of the tumor, its unique morphological features and importance of distinguishing it from metastatic renal cell carcinoma deposits.*

Keywords: *hidradenoma, morphology, renal cell carcinoma.*

I. Introduction

Tumors of sweat gland and of other adnexal structures of skin has increased between 1978 and 2005 by about 170 %, according to 16 cancer registries included in the Surveillance, Epidemiology and End results Program for the 1978 – 2005 period[1]. Solid – cystic hidradenoma is a unique tumor of sweat gland origin with distinct morphological features. It is one of the benign adnexal tumors with clear cells hence cutaneous renal cell carcinoma deposits has to be considered in the differential diagnosis.

II. Case Report

2.1A 24 year old female patient presented with 3 x 4 cm swelling in the thigh of one year duration. The lesion was about 3 x4 cm in size, nodular, soft and cystic. Overlying skin was normal. There were no other similar lesions elsewhere. Her complete hemogram and biochemical parameters including blood sugar, serum urea and creatinine were normal. Excision of the swelling under local anesthesia was performed.

2.2 Gross: The gross features of the lesion is 4 x 3 cms with overlying skin. The lesion was partly solid and cystic. Papillary excrescences were seen in the inner wall of the cyst. Cyst wall was otherwise smooth. Wall thickness ranged from 0.2 cms to 0.3 cms. The lesion was primarily located beneath the skin.

2.3 Microscopic Features: The lesion was circumscribed with solid and cystic areas. The cyst was located in the mid-dermis. It was neither connected to the overlying epidermis nor extended into the subcutaneous fat. The solid area of the tumor had a biphasic population of cells. One was composed ofporoid cells that were round to oval with abundant eosinophilic cytoplasm. Nucleus was round and vesicular. The other pattern was composed of cells with clear cytoplasm and eccentric nuclei. The poroid cellscomposed 75 % of the tumor admixed with the clear cell pattern which constituted the rest 25 %. The nuclei of the clear cells were smaller than those of eosinophilic cells. The clear cell cytoplasm appears to represents glycogen rich areas.

Squamous differentiation in the form of concentricpolygonal cells with slit like lumen representing squamous eddy formation was present. Duct like structures lined by a layer of cuboidal cells were present. Our slide is unique in that cystic area constituted the bulk of the lesion. Variable sized cystic cavities lined by flattened cells and composed of fibro collagenousstroma were seen. The cyst formation is believed to represent cystic degeneration.

Glandular structures with irregular lumen surrounded by cuticle resembling acrosyringium were seen. Mitotic activity was not increased. Atypical features indicating hidradenocarcinoma including nuclear pleomorphism and hyperchromatism, macronucleoli and giant cells were not seen.

2.4 Diagnosis: Owing to the presence of solid and cystic areas with biphasic pattern of tumor resembling hidradenoma, we named the tumor as solid – cystic hidradenoma.

2.5 Differential Diagnosis: The importance of differentiating clear cell hidradenoma from clear cell renal cell carcinoma is self- explanatory. Clear cell renal cell carcinoma metastatic to skin has relatively numerous delicate vascular channels as compared to clear cell hidradenoma in which there are fewer vessels. Also, in our patient the lesion is grossly cystic. Metastatic renal cell carcinoma is seldom cystic. The immunohistochemical panel for renal cell carcinoma is positivity for CD10, EMA and renal cell carcinoma antigen (RCA). Our

patient's ultrasound abdomen had features of fatty liver with normal echo texture of renal parenchyma. We relied on these clinical findings and histomorphology of the slide to rule out cutaneous renal cell carcinoma deposits.

Our slide has clear cells, squamoid cells and squamous eddies that brings clear cell squamous cell carcinoma into differential diagnosis. However, ductal and glandular structures are present in our slide and the lesion is cystic.These features usually do not occur in clear cell squamous cell carcinoma.Moreover, squamous cell carcinoma has continuity with the epidermis and has infiltrating borders. Our slide had dermal lesion with rounded contour and did not reveal features of malignancy.Hidradenocarcinoma, the malignant counterpart usually is cellular and shows nuclear atypia. It also has mitotic figures and has infiltrative features. In our slide these features are conspicuously absent.

III. Discussion

3.1 Epidemiology: Annual incidence rate of sweat gland tumors is about 5.1 cases per 1 million people. Men were more affected than women. As age increases, the incidence rate appears to increase with no site predilection for cutaneous appendageal carcinoma. Five year survival rate were 99% for localized tumors while metastatic diseases had 43% of 5 year survival rate [1].

3.2"Nosological Jungle" as named by Wilson Jones [2], Hidradenoma is also known as 'solid-cystic hidradenoma', 'eccrineacrospiroma'. 'eccrine sweat gland adenoma', 'clear cell hidradenoma' and 'clear cell myoepithelioma'.

3.3 Clinically, both eccrine and apocrine variants appear similar and are solitary. They are solid tumors or partially cystic nodule. It has a slight female preponderance. In our patient, the lesion was solid with cystic areas and papillary formations and moreover ours was a female patient.

3.4Age: Hidradenoma is a lesion of middle age although occurrence in a 13 year old boy had been reported in literature [3].Age of our patient was 24 years.

3.5Siteand Size: There is as yet no site predilection for Hidradenoma although occurrence in vulva has been reported [4].Involvement of scalp, face, anterior trunk and proximal limbs have been reported [5]. The average size of Hidradenoma is about 1 to 3 cm in diameter, although cases have been reported upto 6 cm in size [6]. In our patient, the size of the lesion is 4 cm in diameter and it was present in the proximal limb.

3.6Morphology: Furthermore,

Apocrine hidradenoma	Eccrinehidradenoma / PoroidHidradenoma
Typified by clear, polygonal and mucinous cells	Usually has poroid and cuticular cells
Has apocrine differentiation.	Has both apocrine differentiation and eccrine differentiation.
Well circumscribed, non-encapsulated, multilobulartumor centered in the dermis and also extending into the subcutis	Well circumscribed, non-encapsulated tumor centered in the dermis.

Although eccrinehidradenomahas poroid and cuticular cells, it usually lacks polygonal, clear cells and mucinous cells. Our patient, in addition to having poroid and cuticular cells, 25% of the tumor had clear cells. This feature contributed to the differential diagnosis of metastatic renal cell carcinoma deposits in skin.

3.7Genetic profile: t (11:19) translocation has been reported in cutaneous clear cell hidradenoma similar to the translocation in breast parenchyma. The fusion genes are the MucoEpidermoid carcinoma translocated 1 (MELT1) gene on chromosome 19p13 and the mastermind-like family 2 (MAML2) gene on chromosome 11q21[7].

3.8Immunohistochemistry: The immunohistochemistry panel includes positivity for Low Molecular Weight Cytokeratin, CarcinoEmbryonic Antigen and Epithelial Membrane Antigen. Variable expression of smooth muscle actin and S100 have been noted [8].

3.9 Electron microscopy: The tumor cells are connected by desmosomes. Clear cells have abundant glycogen and few tonofilaments, while the other cell type has abundant tonofilaments and small amount of glycogen [8].

3.10Prognosis: The lesion was excised in toto and follow-up for a period of one year revealed no tumor recurrence. However, local recurrence have been noted in incomplete excision of the tumor. The malignant variant, hidradenocarcinoma may arise ab initio or by transformation of a benign lesion [8].

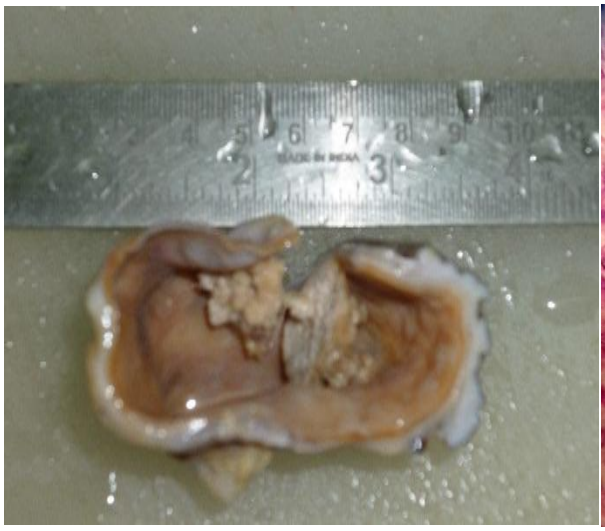
3.11 Review of Literature: To the best of our knowledge, we reviewed the literature and our findings are as follows:

S.No	Author	Year	No.of cases	Gender	Age	Site	Conclusion
1.	R.K.Winkelmann et al	1968	41	F>M	Any age	Any site	Solid-cystic hidradenoma is a distinct tumor. None of the 41 cases showed evidence of malignancy [5].
2.	B Wang, D Sarma et al	2007	1	Male	79yrs	Scalp	Carcinoma-in situ was a component, warranting complete excision of the tumor in order to prevent recurrence [9].
3.	P.Bagga, M Shahi et al	2008	1	Female	3 yrs	Supra clavicular region	Occurance of hidradenoma is rare in this age group [10].
4.	Keith E Volmaret al	2005	1	Male	59 yrs	Right axillary nodule	Patient was treated for conventional type of renal cell carcinoma [11].
5.	Gianotti R, Alessi E et al	1997	5	-	-	-	Hidradenomas were considered to be of uncertain cell of origin. Apocrine hidradenoma could possibly derive from pluripotentialgerminative cells in follicles [12].

IV. Conclusion

Solid – Cystic Hidradenoma, a tumor of sweat gland origin is a rare tumor. It displays biphasic pattern, including cells with eosinophilic cytoplasm and clear cells. Other morphological features like reduced vascular channels, presence of duct and gland formations, squamoid eddies differentiate this tumor from metastatic renal cell carcinoma deposits.

Our slide had clear cell changes for which clear cell renal cell carcinoma is a close differential diagnosis. The management differs for clear cell renal cell carcinoma metastatic deposits and clear cell changes in Hidradenoma being follow-up with chemotherapy for the former while excision for the latter. This case report highlights the morphological perspectives to be considered for differentiating these two entities and to choose appropriate management strategy, thereafter.Hence, in low resource setting, these morphological clues along with ancillary diagnostic tests can render accurate diagnosis and channelize better treatment option for the patient.



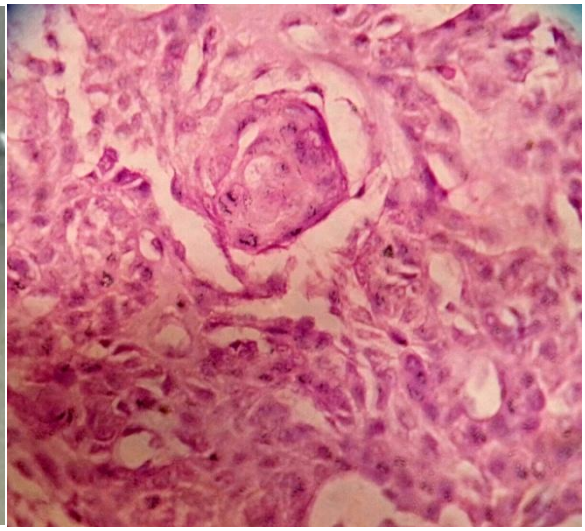


Fig.1 Gross: Cystic nodule with solid areas.**Fig 2:** Squamous eddies H & E stain (x 40 X)

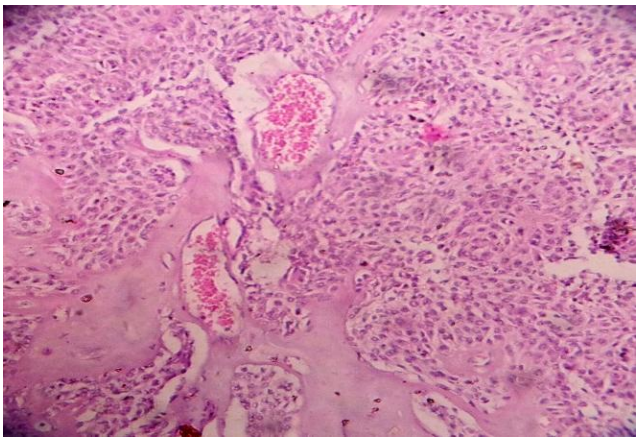


Fig 3: Vascular channels H & E stain (x 10X)

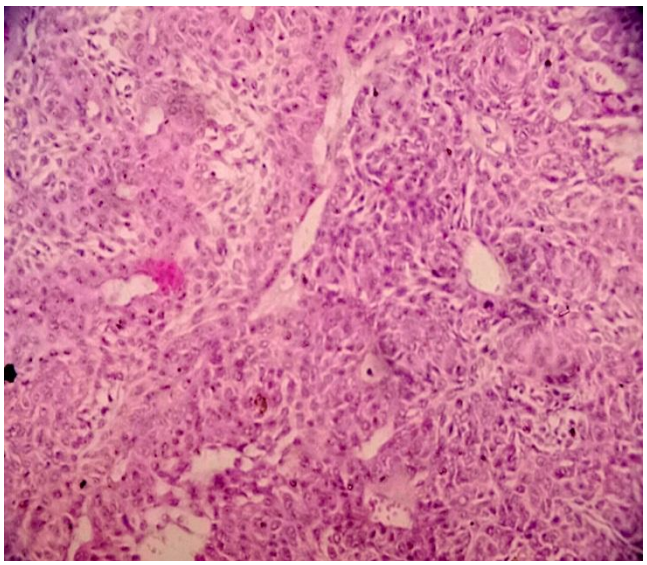


Fig 4: Clear cells and poroid cells H & E stain (x 40 X)

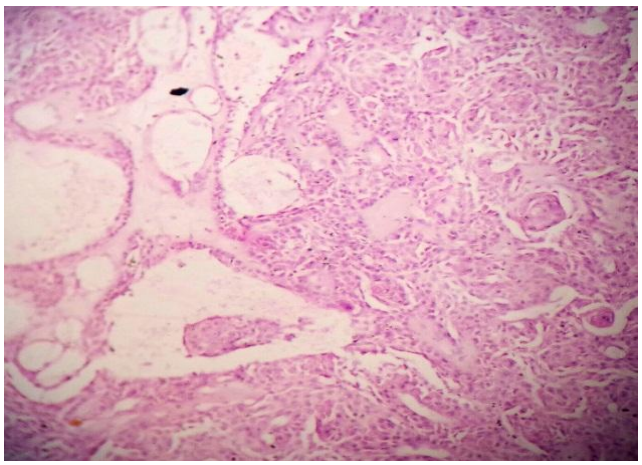


Fig 5: Cystic area and solid areas H & E stain (x 10 X)

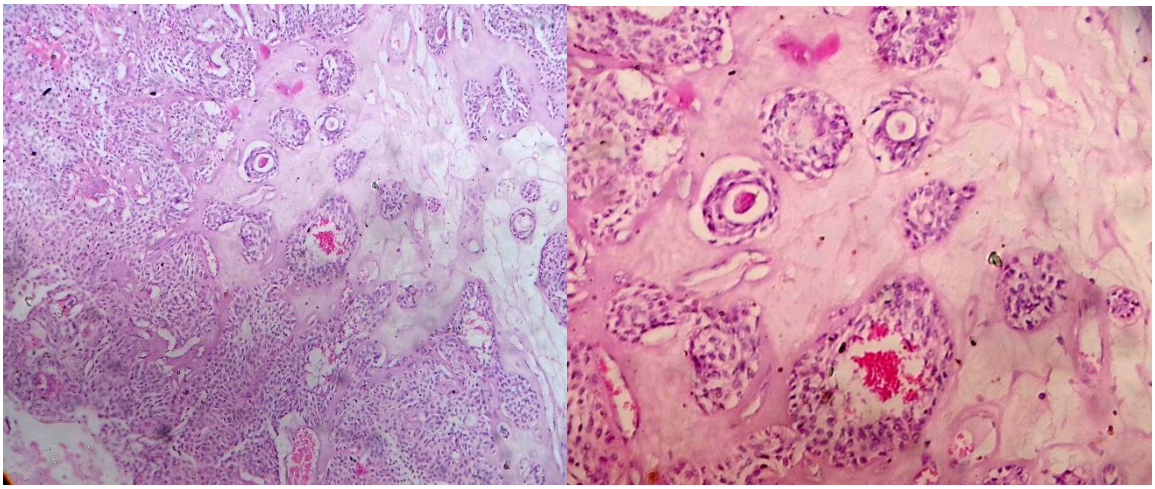


Fig:6, 7 Duct and gland like formations with clear cells (H&E)

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