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Case Report

Nodular hidradenoma of eyelid – A rare case with cyto-histo corelation

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ABSTRACT

Nodular hidradenomas is an uncommon benign adnexal tumor that arises as nodules from eccrine sweat glands. Although it can appear at various sites, its occurrence in eyelids is extremely rare. The diagnosis is usually confirmed on histopathology, and the lesion poses a diagnostic challenge for cytopathologist and is rarely diagnosed on fine aspiration needle cytology (FNAC), which prompted us to report. Here we report a rare case of Nodular Hidradenoma in a 50-year male, that was initially diagnosed on FNAC and later confirmed on histopathology and IHC. Thus, our study emphasizes that FNAC can be used as a first line diagnostic modality for nodular lesions of skin.

1. Introduction

Nodular Hidradenoma is a benign adnexal tumor of sweat gland origin. It is commonly being referred as clear cell hidradenoma, sweat gland adenoma of eccrine origin, a solid cystic hidradenoma, clear cell myoepithelioma, and eccrine acrospiroma. It usually affects any age group with a peak incidence in middle age group individuals. Most common sites of predilection being head and neck region with a predominance in female population. Clinically it presents as a slow growing solitary, solid to cystic intradermal nodule. A rare case of nodular hidradenoma in a 50 year old male is being discussed here who presented as a nodule in the lower eyelid which is an uncommon site, along with its cytological and histopathological correlation.

2. Case Report

A 50-year-old man presented with a nodular mass in the right lower eyelidfor 4 years.(Figure 1) The swelling was initially small, but gradually increased in size, that compelled him to seek for medical attention. There was no history of earlier trauma to the site complained by the patient. On physical examination a solitary painless, firm globular mass of 1x0.5 cm was seen in the lower eyelid margin that didn't extend to deeper structures.There was no ulceration or oozing of any kind of exudates (pus) from the mass. Routine hematological and biochemical parameters were done that was within normal limits. With a provisional diagnosis of Adenoid cystic carcinoma, the patient was sent to us for fine needle aspiration cytology (FNAC). FNAC smears were moderately cellular, showing few polygonal cells (epithelial)cells in clusters having round to ovoid nucleus with dense cytoplasm admixed with good number of spindle cells in clusters. Squamoid cells were also present and some cells showing an oncocytic change were also noted in a background of fibromyxoid stroma.(Figure 2 A-D) There were no mitosis or nuclear atypia henceexcluding the features of malignancy. The cytology report was signed out as Benign Adnexal tumor with a possibility of Chondroid syringoma and Nodular Hidradenoma. A complete wide local excision of the

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mass with a 3mm clear margin of healthy surrounding tissue was performed, together with primary closure and sample was send to us for histopathological analysis. Grossly we received two pieces of skin lined grayish brown tissue altogether measuring 1x1 cm. On microscopy microscopy the tumor was well circumscribedcomposed of epithelial lobules with intervening hyalised stroma. The tumor lobules composed of polyhedral cells with pale eosinophilic cytoplasm, some cells having clear cytoplasm. Tubular lumina of varying sizes were also present in the tumor. Some foci showed fusiform appearance of the cells. No nuclear atypia or mitosis were present.(Figure 3 A-C) Based on distinct histomorphological findings and IHC positivity to CK (diffuse membranous positivity) and EMA (focal/patchy pos) and negative to SMA and CD3, a final diagnosis of Nodular hidradenoma was rendered.

Figure 1: Nodular mass in the left lower eyelid

3. Discussion

Nodular hidradenoma is a rare cutaneous tumor that arises from distal excretory ducts of eccrine glands. Most common sites of predilection for nodular hidradenoma are face, upper extremities, axilla, trunk, thigh and scalp[1] Very few cases have been reported in eyelid as its rare. It can affect any age group.[2] First case of Nodular hidradenoma in the eyelid was reported in 1964.[3] Nodular hidradenoma is considered to be intermediate between eccrine poroma and eccrine spiradenoma proved in ultrastructural studies and in enzyme histochemical studies.[4] They are usually solitary and size ranges between 5mm to 20mm in diameter, however on rare occasions multiple lesions have been reported. They are usually covered by intact skin, but sometimes the skin may be flesh colored, erythematous or blue with surface ulceration and spontaneous oozing. In the present case the tumor appeared benign with no surface ulceration / spontaneous oozing.

Histologically sweat glands may be either of eccrine or apocrine nature. Eccrine glands are present throughout the skin but are most abundant in palms ,soles and axilla. In the eyelid eccrine glands are present at the lid margin and in the surface dermis. In our case the lesion was present in the margin of right lower eyelid. Apocrine glands are found in fewer regions of the body mostly axillae, nipple and anogenital region.

Cytological diagnosis of Nodular hidradenoma is challenging and most of the cases are misdiagnosed, inconclusive or misinterpreted on FNAC. Smears are usually cellular with predominance of two types of cell population –eosinophilic /polygonal cells and clear cells. Polygonal cells contain round to oval nuclei with faint eosinophilic cytoplasm. Sometimes these cells show squamoid /spindle cell like appearance. Clear cells have round eccentric nuclei, finely granular chromatin, small nucleoli and more abundant clear cytoplasm. Eosinophilic cells form large cohesive, three dimensional, papillary like or closely packed clusters. Clear cells form medium sized, flat clusters. The cytology of our case shows all features as mentioned above except a prominent clear cell component. Scanty or absent clear cells may be considered as one of the diagnostic pitfalls. Extracellular hyaline and amorphous material also present in the background. The cytological appearance of our case closely resembles that of chondroid syringoma (pleomorphic adenoma), adenoid cystic carcinoma and cutaneous cylindroma. Histopathology of Nodular hidradenoma shows well circumscribed dermal nodule with no connection with epidermis. Both solid and cystic components are seen. Cystic spaces are filled with homogenous eosinophilic material. In the solid area's tumor cells are arranged in lobules which have tubular lumina lined by cuboidal to columnar epithelium. The polyhedral and clear cells are mixed with varying proportions, which depends from tumor to tumor. Polyhedral cells are evident with a rounded nucleus and slightly basophilic cytoplasm. These cells at times may appear fusiform and show elongate nucleus. Clear cells are round and contain watery clear cytoplasm and small dark nucleus. These cells have considerable amount of glycogen, which are PAS positive and diastase resistant material at the periphery. In certain cases, squamoid differentiation and horn pearl formations are seen. The features indicative of malignancy includes poor circumscription, presence of nuclear atypia, mitotic activity, presence of predominantly solid cell islands, infiltrative growth pattern, necrosis and angiolymphatic permeation.[5–7] No features suggestive of malignancy were found in histosections. IHC positivity for CK, EMA and CEA has been reported.[8] In our case we did EMA that showed membranous positivity.

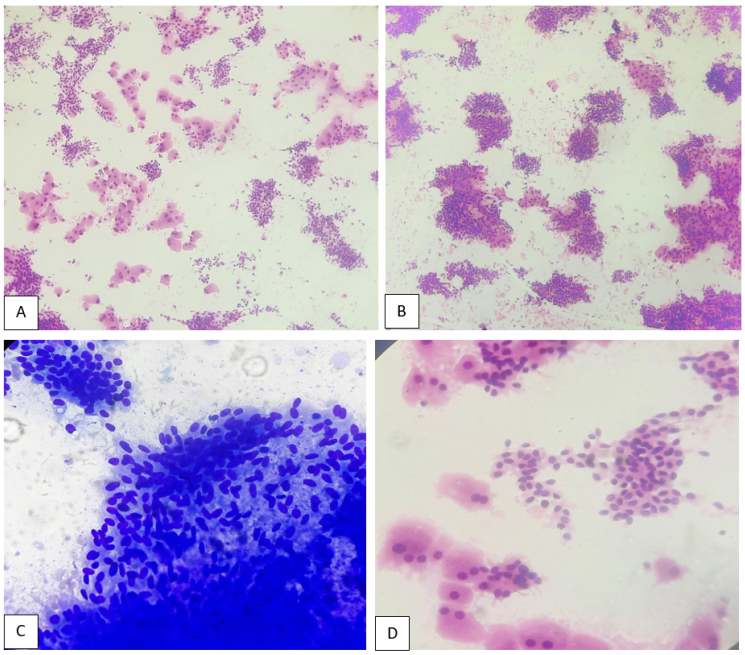

Figure 2: A): Scanner view 40x; **B**): LP 100x, **C**): LP 100x; **D):** LP 100x; **A-D**): FNAC - Few polygonal cells (epithelial) cells in clusters having round to ovoid nucleus with dense cytoplasm admixed with good number of spindle cells in clusters. Squamoid cells were also present and some cells showing an oncocytic change were also noted in a background of fibromyxoid stroma

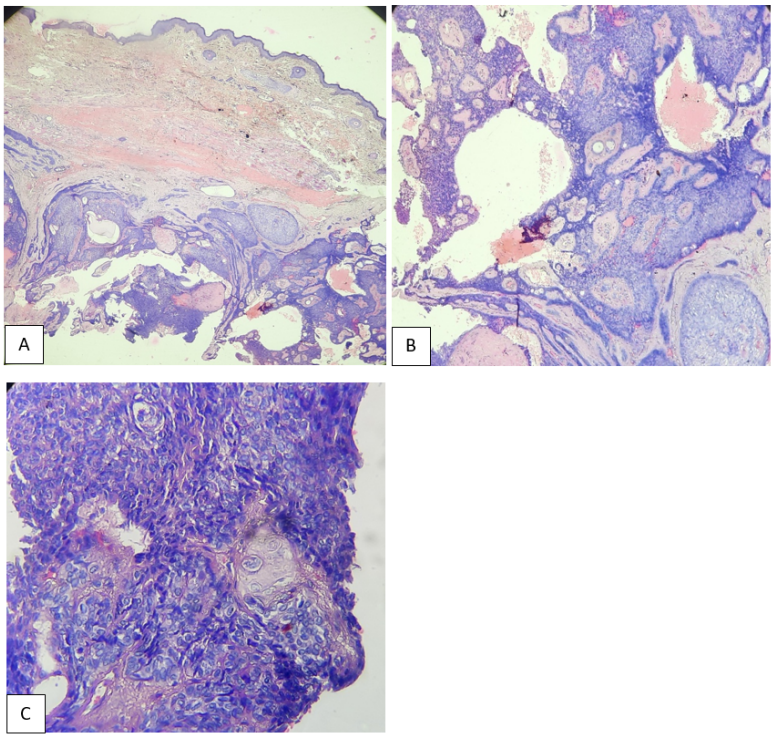

Figure 3: A): Scanner view 40x- Showing well circumscribed nodule in the dermis; **B**): LP100x; **C**): HP 400x; **A-C**): Both solid and cystic components are seen. Cystic spaces filled with homogenous eosinophilic material. In the solid areas-tumor cells are polygonal with dense eosinophilic cytoplasm and clear cells with clear cytoplasm

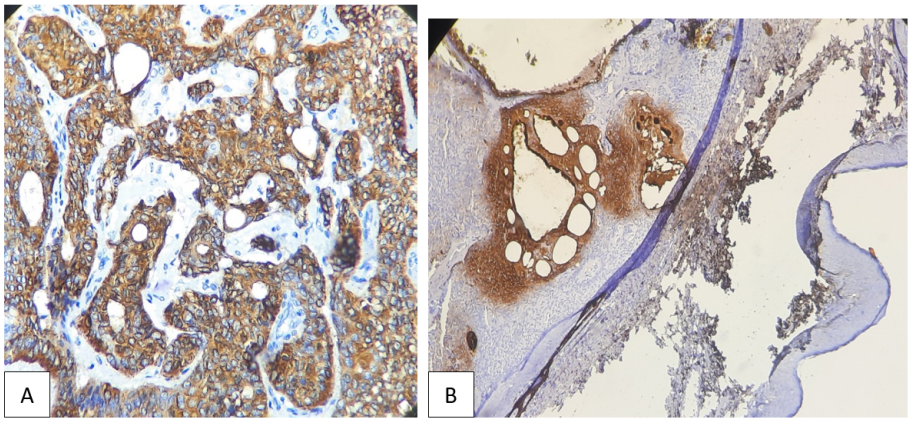

Figure 4: a): LP100x- CK - Diffuse membranous pos; **b)**: LP 100 x-EMA patchy positive

4. Conclusion

FNAC is not confirmatory for diagnosis of Nodular hidradenoma, however it plays an adjunct to histopathology and IHC. In order to sign out a cytology report of adnexal tumor, the cellularity must be adequate. One should always think, nodular hidranenoma as one of the possibilities in view of round to ovoid cells and few spindles to squamoid cells on cytology smears at an unusual site. Sweat gland tumor although rare, one should be kept in the list of differentials of eyelid tumor which can be correlated and confirmed by histopathology and IHC.

5. Source of Funding

None.

6. Conflict of Interest

None.

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