

Cyto-diagnosis of Papillary Hidradenoma of Vulva: A Case Report from A Tertiary Hospital of Rajasthan, India

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Abstract— *Papillary Hidradenoma is a rare benign tumor of apocrine glands. Very limited number of case reports are available in the literature detailing the cytological features of papillary hidradenoma. Such a rare case specimen came in notice here at Pathology Department of SMS Medical College, Jaipur (Raj.) India. So, one such a rare case of papillary hidradenoma was explored with its cytological findings. A 30 year old female presented with a vulval cyst. Fluid from this cyst was sent for cytology with a clinical diagnosis of Bartholin cyst. The cytology suggested it to be a benign adnexal tumour by the presence of Biphasic pattern of cell arrangement which was further confirmed histologically. So whenever an middle aged female presents with a nodular lesion in the anogenital area, hidradenoma papilliferum should be kept in mind along with other conditions.*

Key words – *Cystic Papillary Hidradenoma, Vulval Cyst, Bartholin Cyst, Skin Adnexal Tumors*

1. Background

Papillary Hidradenoma is also known as hidradenoma papilliferum, is an uncommon lesion which occurs in middle aged women usually of 30-49 years. It is a rare benign tumor of apocrine glands. Papillary hidradenoma of the vulva is a rare, benign neoplasm arising from apocrine sweat glands of the skin. Frequently, this lesion has been mistaken for carcinoma

It usually presents as an asymptomatic flesh-colored nodule in the anogenital area of women.¹ It may presents as a firm, flesh to red-colored nodule in the anogenital area. Rarely, it can be seen over the head and neck, and over modified apocrine gland areas such as breast, external ear canal, and eyelid. It is then referred to as ectopic hidradenoma papilliferum.² Histopathology shows a partly solid or solid-cystic dermal lesion with anastomosing papillary structures and tubules as well as some glandular areas. The epithelial lining consists of an inner layer of columnar cells with decapitation secretion in the lumen and frequently an outer myoepithelial layer. The tumor has got a good prognosis and malignant transformation is extremely rare.³

Very limited number of case reports⁴⁻⁶ are available in the literature detailing the cytological features of papillary hidradenoma. Such a rare case specimen came in notice here at Pathology Department of SMS Medical College, Jaipur (Raj.) India. So, one such a rare case of papillary hidradenoma was explored with its cytological findings.

2. Case Report

Clinical Manifestation: A 30 year old female presented with a cystic mass in the labia majora measuring 4x3 cm.

Investigation: Cyst fluid was aspirated and sent for cytology. Fluid was centrifuged and smears were prepared and stained with hemotoxylin and eosin (H&E) and papanicolou stain (PAP).

Microscopic Examinations: Microscopy revealed following salient observations:-

- A distinct epithelial component arranged in sheets and papillary clusters having moderate amount of eosinophilic cytoplasm, round to oval nucleus with fine evenly distributed chromatin. (Figure 1 & 2)
- Second type of cells appeared singly or in small cell balls having abundant bright foamy appearing cytoplasm which was basophilic on PAP staining, these cells were considered to be of apocrine origin and raised the suspicion of adnexal tumour. (Figure 1 & 2)
- Third cell type was small round or cuboidal cells with a dark pyknotic nucleus, probably myoepithelial cells and were relatively scarce in number. (Figure 1 & 2)
- Background showed presence of eosinophilic amorphous material on the H&E stain associated with foam cells, consistent with a cystic component in the tumour. (Figure 1 & 2)
- The cytologic diagnosis was that of a benign adnexal tumour and probably papillary hidradenoma of the vulva which was confirmed histologically. (Figure 3)

Figure 1

Figure 2

Figure 3

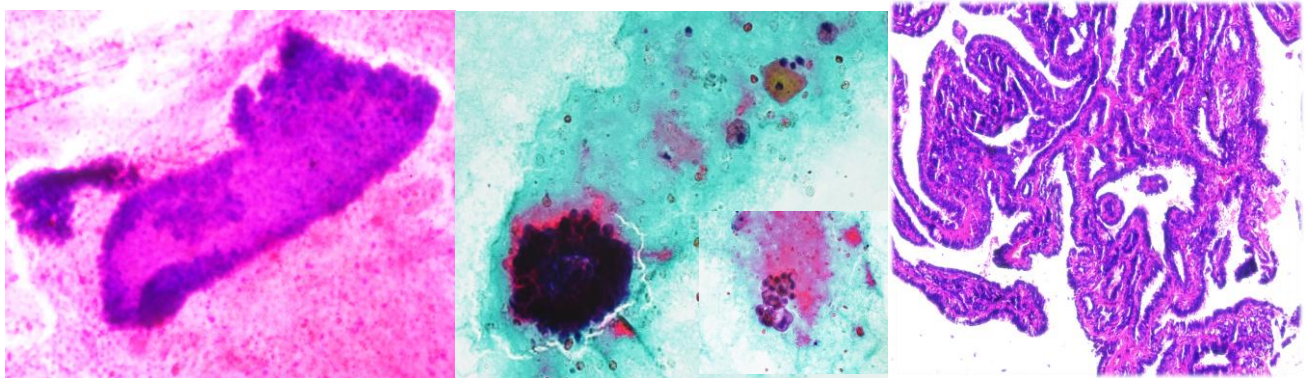


Figure 1: Sheet of ductal cells having eosinophilic cytoplasm round to oval bland nuclei (H&E, 400X).

Figure 2: Cuboidal cell having dark nuclei are myoepithelial in origin, amorphous material and foam cells forming cystic component. Inset showing biphasic pattern of cell having clear and eosinophilic cytoplasm (PAP Stain, 400X).

Figure 3: Histology shows ductal and tubular structures lined by two type of cells apocrine snout also present in lumen of these ducts (H&E, 400X).

3. Discussion:

In the present study it was observed that Skin adnexal tumours are rarely subjected to FNA and therefore there is lack of literature on cytology of adnexal tumors. It may pose diagnostic difficulties with Bartholin cysts/abscesses, lipoma and some premalignant/malignant lesion;⁶ However, recognition of lesion as skin adnexal tumour and if possible subtyping of tumour allows the patient to be managed for excision on an out patient basis and minimizing the risk of wide excision. The following features must raise an awareness for adnexal tumour including cystic degeneration represented by amorphous background material and foam cells with presence of epithelial and duct like structures and or clear cells and no nuclear features of malignancy⁷ The case was presented to us as Bartholin cyst fluid and a

suspicion of adnexal tumour was kept because of presence of biphasic pattern of cell arranged as duct like and tubule like structures and background of amorphous material with foam cells. Limited case reports of vulvar papillary hidradenoma are available till date and reports on exfoliated cytology is all the more rare. Among few of these rare cases, first case was described by RollinsSD⁸ and other one presented by TatsujiHoshno et al⁹ who described two type of cells on cytology. The possible pitfalls of cytological diagnosis of adenocarcinoma due to presence of extensive vacuolation and or clearing of cytoplasm should be removed by awareness of adnexal tumour. All skin adnexal tumour even if appears overtly benign should have histopathologic confirmation.^{10,11} Adenocarcinoma rarely occurs in vulvar papillary hidradenoma.¹² Pelosi G et al¹³ reported and immuno-histo-chemically documented the first case of malignancy in vulvar papillary hidradenoma resembling apocrine breast cancer. Immunoreactivities for low and high molecular weight cytokeratin, alpha smooth muscle specific actin, carcinoembryonic antigen, s-100 protien, and gross cystic disease fluid protein 15 show features that resemble those of an apocrine breast cancer.¹³ This case highlights the cytologic features of hidradenomapapilliferum on cytologic smears. The clinical presentation should not mislead the cytologic interpretation of the tumour cells. Subsequent histologic confirmation is essential for good management.¹⁴

This study case, a 30 year old female presented with a cystic mass in the labia majora measuring 4x3 cm. On cytological examination of aspirated cystic fluid a distinct epithelial component arranged in sheets and papillary clusters having moderate amount of eosinophilic cytoplasm, round to oval nucleus with fine evenly distributed chromatin. Other type of cells appeared singly or in small cell balls having abundant bright foamy appearing cytoplasm which was basophilic on PAP staining, these cells were considered to be of apocrine origin and raised the suspicion of adnexal tumour. Another type of cell were small round or cuboidal cells with a dark pyknotic nucleus, probably myoepithelial cells and were relatively scarce in number. Another salient feature was that background showed presence of eosinophilic amorphous material on the H&E stain associated with foam cells, consistent with a cystic component in the tumour. This cytological observations suggesting a benign adnexal tumour and probably papillary hidradenoma of the vulva which was confirmed histologically.

Other authors like Shrinath et al⁴ and Veerana et al⁵ also reported an asymptomatic a well-circumscribed red nodule of size 1 × 1 cm on inner aspect of right labium majus in a 32-year-old female. Excision biopsy of the nodule was done and sent for histopathological examination. Histopathological findings were suggestive of hidradenoma papilliferum.

CONCLUSIONS

Whenever an adult female presents with a nodular lesion in the anogenital area, hidradenoma papilliferum should be kept in mind along with other conditions such as sexually transmitted diseases and other benign and malignant tumors.

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