

Hidradenoma Papilliferum of the Vulva Presenting as a Hydrocele of the Nuck's Channel – Case Report

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Received: February 20, 2018; **Published:** April 30, 2018

Abstract

Background: Hidradenoma papilliferum is an uncommon benign neoplasm arising from apocrine glands which very rarely occurs at the skin of anogenital area, predominantly in middle-aged women. It usually presents as a slow-growing, solitary, asymptomatic skin colored or red nodule less than 1cm in diameter, although sometimes it ulcerates and bleeds. The tumor has got a good prognosis and malignant transformation is extremely rare.

Case Report: We describe a case of the hidradenoma papilliferum of the vulva in a 41-year-old women, which persisted as an asymptomatic nodule on the anterior part of the left major labia during last three months. On examination, a well-circumscribed skin colored nodule of 2 cm size on the anterior part of the left major labia was noted. The nodule was extirpated in toto and the histology revealed a hidradenoma papilliferum.

Discussion: Hidradenoma papilliferum is a benign neoplasm, cured only by simple excision. The differential diagnosis includes: Bartholin cyst, abscess, true tumors (lipoma, leiomyoma, sarcoma), or hydrocele of the Nuck's channel. Although the canal of Nuck normally disappears in the first year of life, it can cause an indirect inguinal hernia or hydrocele of the canal of Nuck, which present as painless edema in the inguinal area or labium.

Conclusion: When 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.

Keywords: *Hidradenoma Papilliferum; Vulva; Hydrocele Nuck's Channel*

Abbreviation

CK7: Cytokeratin 7

Introduction

Hidradenoma papilliferum is a slow-growing benign skin-adnexal tumor with apocrine differentiation. The skin of the vulva and perianal regions are the most frequently involved areas, with rare cases being reported in other skin localizations (ectopic hidradenoma papilliferum) [1]. The clinical presentation as well as the pathologic features, treatment, and prognosis are similar in both forms. It usually presents as a slow-growing, solitary, asymptomatic skin colored or red nodule less than 1cm in diameter [2], although sometimes it is accompanied with ulceration and bleeding. The most common site of ectopic hidradenoma papilliferum is the head and neck, although a giant ectopic hidradenoma papilliferum on the scalp has been reported recently [3]. The diagnosis can be made only by histopathological examination since they clinically mimic other cutaneous neoplasms. The differential diagnosis includes: Bartholin cyst, abscess, true

tumors (lipoma, leiomyoma, sarcoma), or hydrocele of the Nuck's channel. This channel represents a small evagination of the parietal peritoneum, which is formed when the genitoinguinal chord i.e. round ligament descends through the inguinal canal during the fetal period. Although the Nuck's channel normally disappears in the first year of life, it can cause an indirect inguinal hernia or hydrocele which present as a painless edema in inguinal area or major labia.

Case Presentation

A 41 year-old female patient presented with an asymptomatic nodule in the vulva with 3 months duration. On examination, the inspection and palpation revealed a well-circumscribed skin colored, tender nodule of 2 cm-size with intact surface located on the anterior part of left major labia. Our patient had regular menstrual cycles with normal intensity of menstrual bleeding. Her obstetric history included two pregnancies, which ended with normal vaginal labors. There was no family history for a malignancy of any kind.

All standard laboratory analyses were in normal ranges. The transvaginal sonography showed ordinary finding for the patient reproductive age. Uterus was in normal size with thin postmenstrual endometrium, both ovaries showed regular macromorphology and no free liquid in Douglas space was found. The perineal ultrasound revealed the cystic multilocular formation filled with dense liquid, with unclear margin from the surrounding tissue, located in the region of left major labia. Our differential diagnosis considered: cyst of the Nuck's channel, pyogenic granuloma or other skin-adnexal tumor because of similar localization, ultrasound picture and clinical presentation.

The surgical treatment included total excision of the tumor. During the preparation, the capsule rupture occurred and flesh-like content pulled out. After the complete tumor extirpation, the capsule and all inner content were sent for histopathology examination.

The microscopic examination revealed few pieces of collagen connective tissues associated with a well-circumscribed papillary neoplasm with papillary extensions, tubular structures and microcystic formations lined by basophilic cuboid to columnar cells with benign appearance with focuses of hyperplasia and apocrine metaplasia. Additional immunohistochemical staining showed WT1 positivity for the basal cells, CK7 positivity for the epithelial cells, and low proliferative index detected by Ki67. These findings confirmed the diagnosis of hidradenoma papillary.

The patient was discharged the next day after the surgery, with no complications and in stable clinical condition.

Results and Discussion

The first vulvar hidradenoma papilliferum was reported in 1878 by Werth [4]. He believed the lesion was derived from aberrant epithelium. According to Woodworth, et al. 38% of hidradenoma papilliferum originate in the labia majora and 26% originate in the labia minora [5]. An obstruction of the ductal drainage can result in cystic formation which can secondarily be inflamed [5]. On the other hand, a hidradenoma papillary should be taken into consideration in presence of recurrent cysts/abscesses in the anogenital region [2]. The differential diagnosis should include a cyst of Nuck's channel, which can show a focal reactive hyperplasia of the surrounding stromal tissue and focal inflammation, because of its nearby localization. So, the maintaining an awareness of differentiation between cyst of Nuck's channel and hidradenoma papilliferum, as well as inguinal hernia is of great importance. During the surgical preparation of the cyst of Nuck's channel or unrecognized hernia in the region of major labia, there is a possibility of intestinal damage with consecutive life-threatening diffuse peritonitis. Diagnostically, hidradenoma papilliferum tends to occur exclusively in post-pubescent white women between the ages of 30 and 70, occurring most commonly in the fourth decade.

Hidradenoma papilliferum is a benign neoplasm, cured only by simple excision and extirpation of the lesion. Malignant transformation is a very uncommon event. There is a reported case of adenosquamous carcinoma of the vulva from pre-existing hidradenoma papilliferum [7]. Malignant association of anogenital hidradenoma papilliferum with intraductal carcinoma resembling apocrine carcinoma or malignant transformation in invasive adenosquamous carcinoma has been documented [8,9]. It is speculated that infection with high-risk

HPV may play a role in inducing malignancy, but the association still needs to be proved [7]. On the other hand, the differential diagnosis between hidradenoma papilliferum and true benign or malignant tumor of Bartholin gland is of great importance. The main point is the localization of the vulvar tumor. The tumor of Bartholin gland has more posterior localization in comparison with hidradenoma papilliferum.

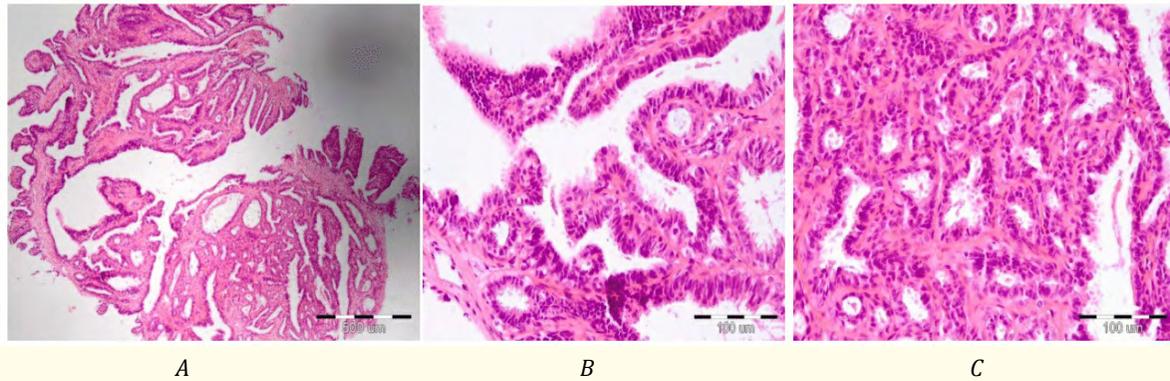


Figure 1: The microscopic structure of Hidradenoma papilliferum as well-circumscribed papillary neoplasm with papillary extensions, tubular structures and microcystic formations lined by basophilic cuboid to columnar cells with benign appearance: A: low magnification; B: high magnification; C: glandular tubules lined with apocrine cells.

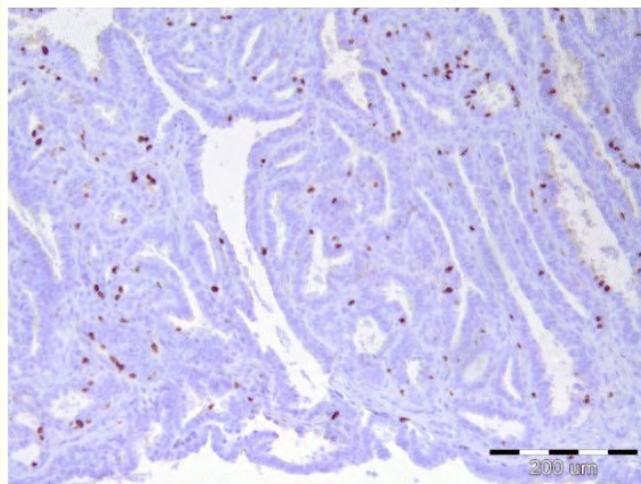


Figure 2: Immunohistochemical staining showed WT1 positivity for the basal cells, CK7 positivity for the epithelial cells, and low proliferative index detected by Ki67.

Conclusion

When 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 vulvar tumors. Patient history and clinical findings are nonspecific to distinguish these different entities and only surgical treatment and histopathology of the excised tissue can make the exact diagnosis. We present this case in order to point out the importance of histology in differential diagnosis between two vulvar lesion which have very similar presentation, hydrocele of the Nuck's channel and hidradenoma papilliferum, which is true adnexal-skin benign tumor.

Conflict of Interest

All authors have no conflicts of interest to declare.

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Volume 7 Issue 5 May 2018

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