

## Case Report

# Hidradenoma papilliferum on labia majora mimicking Bartholin's duct cyst: A case report

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**Abstract** Hidradenoma papilliferum is a relatively rare benign apocrine gland neoplasm that involve perianal, vulva, groin and with lower frequency axilla and periumbilical area. Here we describe a case of Hidradenoma papilliferum mimicking Bartholin's duct cyst.

### Key features

Hidradenoma papilliferum, Bartholin's duct cyst.

## Introduction

Hidradenoma papilliferum is a relatively rare benign apocrine gland neoplasm that involves perianal, vulva, groin and with lower frequency axilla and periumbilical area. It nearly exclusively is reported in middle-aged women. Size of the tumor usually is less than 1 cm, but there are rarely reports of giant lesions over 4 cm. It is four times frequent in vulvar area relative to perianal area and average age of involvement in perianal lesions is higher than vulvar lesions. Labia majora is involved most commonly and in descending order labia minora, interlabial vestibule, clitoris, posterior fourchette and mons pubis are involved.<sup>1,2</sup>

It can be seen as an ectopic tumor developing from heterotopic apocrine glands in head and neck, external ear (ceruminous gland), palpebral (Moll's gland), breast, cheek, eyebrow, trunk, nose and limb. Clinical and pathologic

presentation and prognosis are the same in both ectopic and nonectopic types. Rarely cases have been reported in men. We describe a case of vulvar hidradenoma papilliferum.<sup>3</sup>

## Case Report

A 30-year-old female with an anogenital cystic lesion referred to dermatology clinic in Afzalipour hospital, Kerman, Iran. On physical examination, an erythematous nodule with size of approximately 1 cm<sup>2</sup> was detected on inner surface of labia majora. The lesion was mobile with firm consistency in palpation. She had no history of pain, tenderness, pruritus, bleeding or ulceration (**Figure 1**). There was no other remarkable finding in history and physical examination.

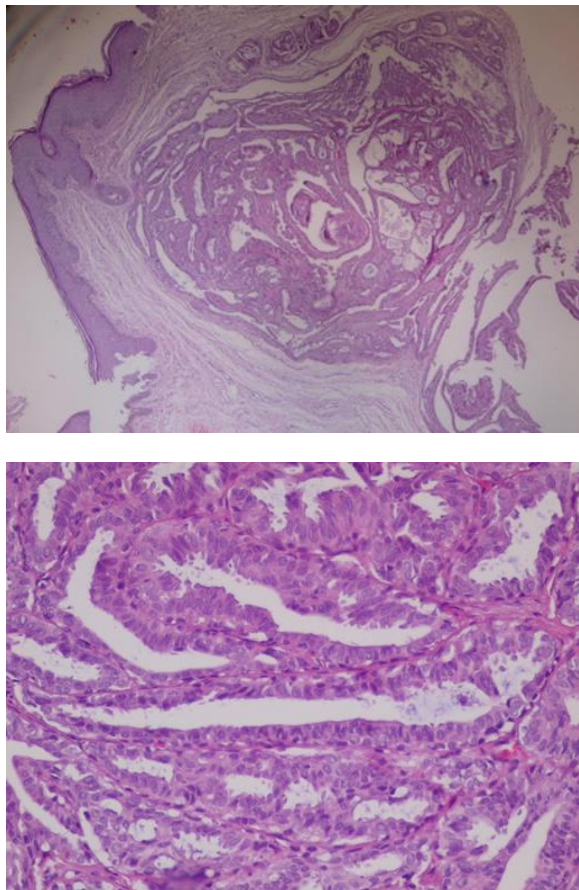
The lesion was excised completely and sent for histopathological evaluation. Clinical differential diagnosis were epidermal inclusion cyst, mucous cyst, fibroma and adnexal tumors. Pathological assessment revealed hyperkeratosis, epidermal acanthosis and a well-circumscribed neoplastic tumor composed of papillary projection lined by columnar epithelium with decapitating secretion that was

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**Figure 1** An erythematous nodule on inner surface of labia majora.



**Figure 2** Neoplastic growth composed of papillary projection lined by columnar epithelium with decapitating secretion, hematoxylin-eosin stain, X10 (A), (B).

diagnostic for Hidradenoma papilliferum (**Figure 2**).

### Discussion

Hidradenoma papilliferum appeared as a firm, red to flesh coloured mobile nodule with rapid or slowly growing that is usually asymptomatic. It can present as an umbilicated lesion with cystic or solid consistency. Ulceration, bleeding, discharge, burning sensation and pruritus have been reported less commonly.<sup>4</sup> Because of histological similarity of this tumor to intraductal papillary breast carcinoma, its origin is speculated to be from mammary like gland in anogenital area.<sup>5</sup>

Estrogen receptors and with less prevalence progesterone and androgen receptors have been recognized in this neoplasm. This can be explained for the absence of this tumor before puberty and also preferentially involvement in female.<sup>6</sup>

Pathologically, lesion is located in dermis and occasionally in subcutaneous tissue. It reveals as a cystic lesion with anastomosing papillary folds that projected into cyst wall and includes two different layers of cells. Inner layer is composed of larger columnar cells with decapitation and outer layer includes smaller cuboidal cells. Fibrosis tissue surrounding the tumor develops pseudocapsule in the periphery of the lesion. Epidermis can be normal, acantotic or ulcerated. Classically, lesion is not attached to overlying epidermis.<sup>7</sup>

There are very rare reports for malignant transformation in vulvar hidradenoma papilliferum. If there are cellular pleomorphism, mitotic activity, necrosis and atypical change; we should consider malignant transformation.<sup>8</sup> HPV types 16, 31, 33, 53, 56 have been detected in anogenital hidradenoma papilliferum lesions.

But their roles in the disease pathogenesis are still unclear.<sup>3,9</sup>

In immunohistochemistry (IHC) low molecular weight keratin (LMWK), epithelial membrane antigen (EMA), carcinoembryonic antigen (CEA), gross cystic disease fluid protein 15 (GCDFF-15), CD15 (Leu MI) are positive, also myoepithelial cells in S100 and smooth muscle actin (SMA) are positive.<sup>4,10</sup>

Clinical differential diagnosis includes Bartholin's duct cyst or abscess, epidermal inclusion cyst, mucous cyst, fibroma, lipoma, leiomyoma, endometriosis, amelanotic melanoma and squamous cell carcinoma.<sup>1,3,4,11</sup>

Bartholin's duct cyst is characterized as a unilateral cystic lesion on labia majora and vestibular area. Cystic formation results from obstruction of duct can lead to retention of secretion and dilation of duct. Absence of papillary projection and decapitating secretion can be differentiated this cystic lesion from hidradenoma papilliferum. Epidermal inclusion cyst usually appeared as a mobile, nontender cyst in labia majora. Mucous cyst presents as a soft asymptomatic lesion on labia minora, vestibule or periclitoral surface areas. Fibroma or leiomyoma develops as a firm nodule in labia majora, perineal or introitus areas. Skin biopsy can differentiate these lesions from hidradenoma papilliferum.<sup>11</sup>

Pathologically, it should be differentiated from tubular apocrine adenoma, syringocystadenoma papilliferum and clear cell hidradenoma.<sup>4</sup>

Absence of attachment to overlying epidermis and infiltration of lymphocyte and plasma cell in stromal can differentiate hidradenoma papilliferum from syringocystadenoma papilliferum. Tubular apocrine adenoma has different histology without papillary structures,

also it is more commonly seen on scalp area.<sup>1,4</sup> Clear cell hidradenoma can be differentiated by tubular structure and clear cells.

Treatment of hidradenoma papilliferum is by total excision of the lesion. Recurrence is uncommon, if the lesion excised completely.

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