



Hidradenoma papilliferum Presenting as a Painful Clitoral Mass: A Rare Case Report

Ağrılı Klitoral Kitle ile Prezente olan *Hidradenoma papilliferum*: Nadir Bir Olgu Sunumu

✉ Meral Koyuncuoğlu¹, ✉ Zercan Kalı², ✉ Şule Gençoğlu³

¹Private Sağlık Hospital, Clinic of Pathology, İzmir, Türkiye

²Gözde Hospital, Clinic of Obstetric and Gynecology, Malatya, Türkiye

³Malatya Turgut Özal University Faculty of Medicine, Department of Dermatology, Malatya, Türkiye

ABSTRACT

Hidradenoma papilliferum (HP) is a rare, benign, slow-growing tumor of apocrine origin, primarily located in the anogenital region of adult women. We report a rare case of HP in a 52-year-old postmenopausal woman presenting with a painful, nodular lesion on the clitoral area. Histopathologic examination confirmed the diagnosis after surgical excision. The lesion showed positive expression for estrogen and progesterone receptors, and p63 in myoepithelial cells. Although HP is typically asymptomatic and commonly seen on labial structures, its occurrence in the clitoris with painful presentation is rare. Awareness of such atypical localizations is crucial for proper diagnosis and management.

Keywords: *Hidradenoma papilliferum*, apocrine tumor, clitoris, vulvar mass

ÖZ

Hidradenoma papilliferum (HP), genellikle erişkin kadınlarda anogenital bölgede görülen nadir, benign ve yavaş büyüyen apokrin kökenli bir tümördür. Bu çalışmada, klitoral bölgede ağrılı nodüler bir lezyon ile başvuran 52 yaşında postmenopozal bir kadının nadir bir HP olgusu sunulmuştur. Cerrahi eksizyon sonrası yapılan histopatolojik inceleme ile tanı doğrulanmıştır. Lezyonda myoepitelyal hücrelerde p63 ve lüminal hücrelerde östrojen ve progesteron reseptörlerinin pozitif ekspresyonu saptanmıştır. HP genellikle labial yapılarda ve asemptomatik olarak izlenirken, klitoris yerleşimi ve ağrılı klinik bulgularla seyretmesi oldukça nadirdir. Bu tür atipik lokalizasyonlara yönelik farkındalık, doğru tanı ve yönetim açısından önem taşımaktadır.

Anahtar Kelimeler: *Hidradenoma papilliferum*, apokrin tümör, klitoris, vulvar kitle

INTRODUCTION

Hidradenoma papilliferum (HP) is a rare tumor with apocrine features, typically arising in the vulvar region. According to clinical observations, these lesions grow slowly and are generally benign. Although initially described in the anogenital area, cases have also been reported in less typical locations such as the head, neck, breast, and even extremities (1-3). Recent studies have identified specialized glandular structures in the anogenital region that embryologically resemble mammary tissue. It is thought

that these mammary-like glands may play a role in the development of lesions such as HP (4).

In clinical practice, HP lesions are usually 1-2 cm in size, well-circumscribed, and superficially nodular. Based on our observations, these lesions may appear skin-colored or reddish and are typically solitary. Although often asymptomatic, some patients may present with pain, itching, or bleeding (5,6). Accurate diagnosis, which is critical for distinguishing HP from other benign or malignant neoplasms, requires both clinical and pathological evaluation.

Address for Correspondence: Zercan Kalı, MD, Gözde Hospital, Clinic of Obstetric and Gynecology, Malatya, Türkiye
E-mail: zercankali@gmail.com **ORCID ID:** orcid.org/0000-0002-7128-7550

Cite as: Koyuncuoğlu M, Kalı Z, Gençoğlu Ş. *Hidradenoma papilliferum* presenting as a painful clitoral mass: a rare case report. Med J Bakirkoy. 2026;22(2):199-201

Received: 25.04.2025

Accepted: 02.09.2025

Publication Date: 08.06.2026



CASE REPORT

A 52-year-old postmenopausal woman presented with a three-month history of a small, painful mass in the clitoral region. Clinical examination revealed a nodular, tender lesion localized to the clitoris. Despite the typical asymptomatic nature of HP lesions in the anogenital area, this case exhibited pronounced symptoms, including sharp pain and discomfort. Surgical excision was performed for both symptom relief and definitive diagnosis.

Histopathological evaluation of the excised papillary lesion revealed features consistent with HP. The papillary lesion was lined by a bilayered epithelium forming gland-like structures (Figure 1A, 1B; x40, x200). Myoepithelial cells in the basal layer showed positive expression for p63 (Figure 1C; x200), and positive staining was observed for estrogen and progesterone receptors in the luminal cells (Figure 1D, 1E; x200).

Although recurrence rates are low in such cases, the patient was placed under clinical follow-up. Written informed consent was obtained from the patient for publication of the case and accompanying images.

DISCUSSION

HP is a rare apocrine tumor, most commonly observed in females aged between 20 and 90 years. The youngest reported case in the literature involved a 16-year-old patient (5). While the majority of cases are asymptomatic, symptoms such as pain, bleeding, itching, and ulceration can occasionally occur (6). For instance, a case of HP located in the external auditory canal was reported to cause an unusual symptom hearing loss (7).

HP most frequently affects the labia minora and labia majora. According to published data, 50% of cases are located on the labia minora, 40% on the labia majora, 7% on the fourchette, and only 3% are localized to the clitoris (8). Approximately 60% of ectopic HP cases have been identified in the head and neck region (9). Lesion sizes generally range from 3 to 25 mm, presenting as solitary, bluish, or reddish nodules (10).

In the case presented in this study, HP was localized to the clitoris, representing a rare location. The lesion measured approximately 12 mm in diameter, had a smooth surface with no ulceration, appeared yellow, was painful, and

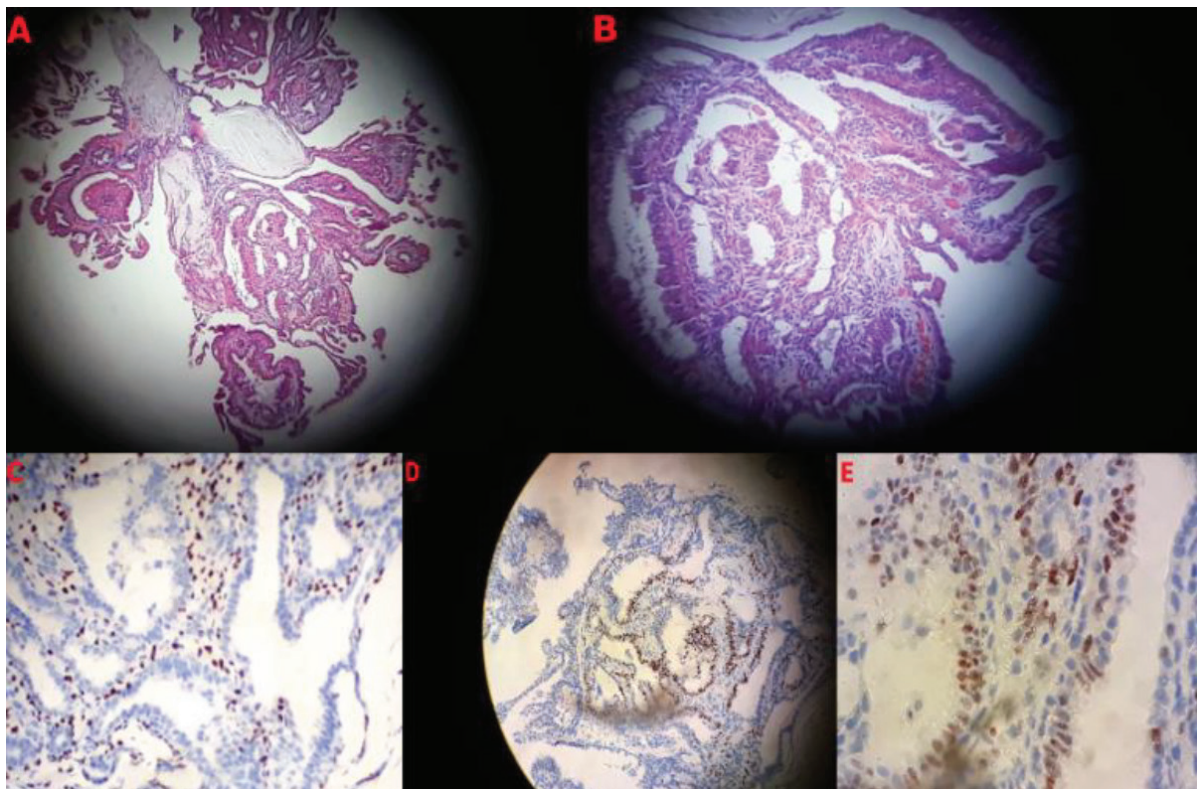


Figure 1. Histological and immunohistochemical features of hidradenoma papilliferum. **A)** The papillary lesion is lined by a bilayered epithelium resembling glandular structures (x40). **B)** The papillary lesion is lined by a bilayered epithelium resembling glandular structures (x200). **C)** Myoepithelial cells located at the basal layer exhibit positive expression for p63 (x200). **D)** Estrogen receptors show positive staining in the luminal epithelial cells (x200). **E)** Progesterone receptors show positive staining in the luminal epithelial cells (x200)

resembled a lipoma. Despite its benign appearance on clinical examination, histopathological evaluation confirmed the diagnosis of HP.

Current literature indicates that HP originates from mammary-like glands in the anogenital region (11). These glands, thought to arise from ectopic mammary tissue developed during embryogenesis, may provide the basis for HP development. They are considered normal anatomical components of the anogenital area. Due to histological similarities, such lesions can be mistaken for benign breast tumors (11).

Hormonal influences are also thought to contribute to the development of HP. Estrogen receptor expression is commonly observed in these tumors (12). An increase in tumor incidence has been reported following puberty, which corresponds with rising estrogen levels. However, in some cases, estrogen and progesterone receptors were negative, while prolactin receptors were positive. This suggests that other hormones may also play a role in the pathogenesis of HP (13).

Although the etiology of HP remains unclear, its higher prevalence among individuals of reproductive age and those who are sexually active has led to the hypothesis that sexually transmitted infections may play a role. While a few cases associated with HPV have been described, current findings are not sufficient to establish a direct causal relationship (14,15).

Study Limitations

This report describes a single case, which limits the generalizability of the findings. Immunohistochemical analysis was restricted to a few markers, and no molecular studies were performed to further explore possible etiological factors. Additionally, long-term follow-up data are currently lacking.

CONCLUSION

This case highlights the clinical and pathological variability encountered in the diagnosis and classification of lesions originating from anogenital mammary-like glands. The presence of the lesion in the clitoral region and its atypical clinical appearance distinguish it from similar cases in the literature. Surgical excision proved to be an effective method for both diagnosis and treatment. Further studies are warranted to investigate the molecular pathogenesis of such lesions in detail and to standardize diagnostic criteria, thereby improving clinicopathological comparisons.

ETHICS

Informed Consent: Written informed consent was obtained from the patient for publication of the case and accompanying images.

FOOTNOTES

Authorship Contributions

Surgical and Medical Practices: M.K., Concept: M.K., Design: M.K., Data Collection or Processing: Z.K., Analysis or Interpretation: Z.K., Literature Search: Ş.G. Writing: Z.K.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declare that this study received no financial support.

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