

Vulvar Hidradenoma Papilliferum: Clinical and Dermoscopic Features in a Case Series

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Introduction

Hidradenoma papilliferum (HP) is a rare benign tumor of the vulva, perineum, and perianal skin, in particular the vulvar interlabial sulcus and, much less commonly, “ectopic” sites. For a long time, it was thought to be a benign tumor of apocrine origin. However, several pathologists have suggested that HP originates from glandular tissue with eccrine and apocrine characteristics. It is clinically polymorphous. Dermoscopy may be a useful tool for diagnosis. This study aimed at describing the clinical and dermoscopic characteristics of HP in the vulva.

Case Presentation

This retrospective observational study included seven cases of histopathologically-proven HP consulted by our dermatology department.

The average age of the patients included in our study was 44.3 years. Six of the seven tumors arose in the interlabial sulcus. All lesions had a nodular aspect; one was polylobed, and another was ulcerated. The predominant color was red; a normal skin color was observed in the two remaining cases. Clinical onset ranged from 4 to 30 months (mean 12). Four patients were asymptomatic, one patient had mild pruritus, and another was bleeding on contact. Dermoscopy imaging was available in six cases revealing a triad of features made up of a reddish-pink background, shiny white structures (5/6 cases), and vessels: hairpin vessels in one patient, dots in one patient, telangiectasias in three patients, and irregular linear vessels in another. A bluish globule was also observed in one patient, which corresponds histologically to intratumoral hemorrhage. In our case series, the direct approach was sufficient for complete excision of six lesions; however, the patient who had the polylobed tumor benefited from surgical reconstruction. Histological examination showed

Table 1. Patient Characteristics and Clinical and Dermoscopic Aspects of Lesions.

Case	Age (Years)	Onset (Months)	Symptoms	Site	Size (mm)	Surface	Color	Morphology	Dermoscopic Features
1	24	4	None	Left labium majora	15	Non-ulcerated	Red	Well-circumscribed, light red nodule	Pink background, irregular dotted and linear vessels, bright white structures
2	36	3	None	Right interlabial sulcus	12	Non-ulcerated	Red	Well-circumscribed, light red nodule with a shiny bright pink surface	Pink background, shiny white areas, telangiectatic vessels
3	45	30	None	Right interlabial sulcus	12	Non-ulcerated	Red	Well-circumscribed, firm to touch with a shiny pink surface	Red background, shiny white areas, irregularly arranged serpentine vessels
4	39	7	Bleeding	Right interlabial sulcus	8	ulcerated	red	Small tumor with an ulcerated surface bleeding to the touch	Pink background, hemorrhagic suffusion
5	30	24	None	Right interlabial sulcus	15	Non-ulcerated	red	Pink nodule tender to touch	Pink background, shiny white structures of reticular layout, dotted vessels
6	80	12	None	Right interlabial sulcus	10	Non-ulcerated	Skin-colored	Small nodule with normal skin color	Pink background, shiny white structures hairpin vessels, bluish globule
7	56	4	Pruritus	Vulva and Perineal area	50 mm for the largest	Non-ulcerated	Skin-colored	Polylobate tumor of firm consistency taking the vulva and the perianal region	Not available

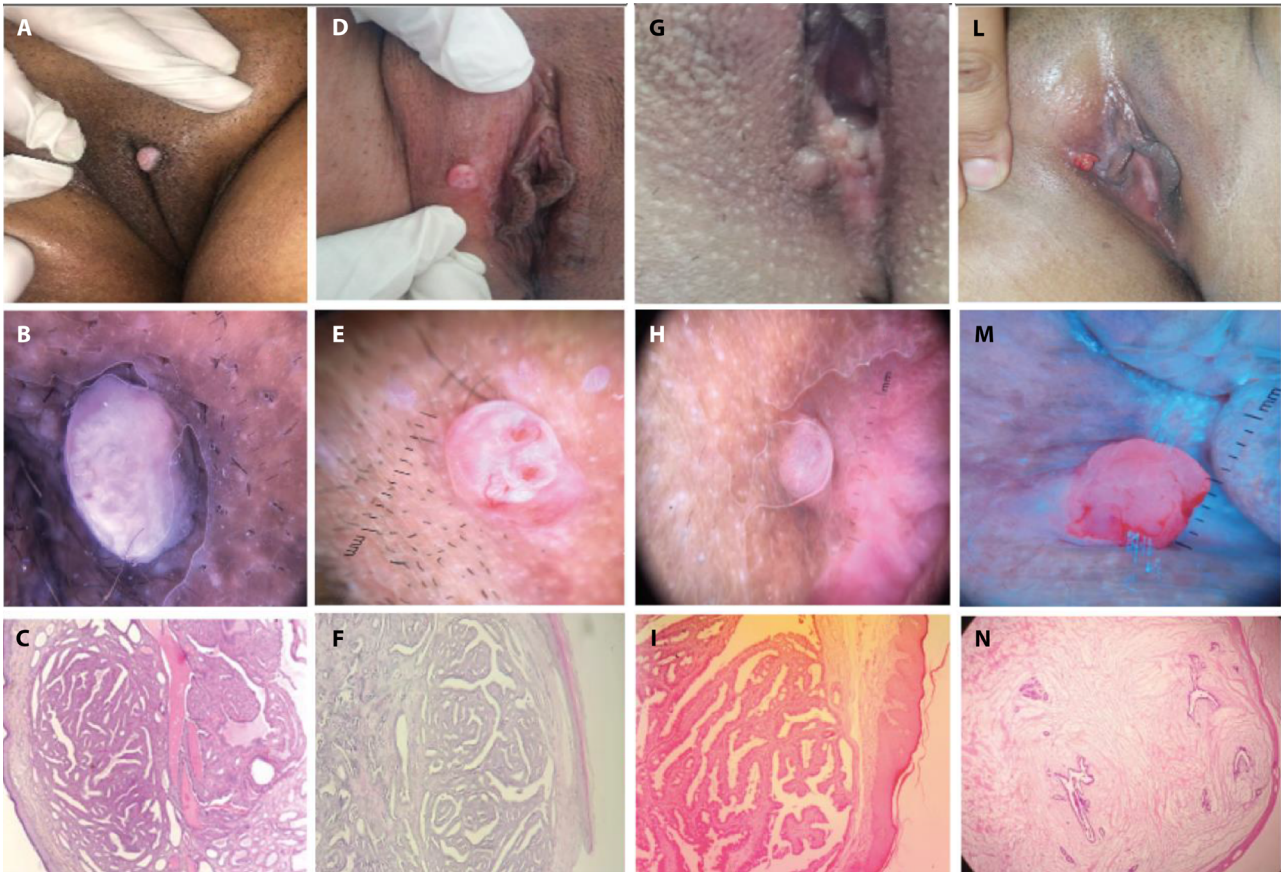


Figure 1. Case 1: (A) A well-circumscribed light red nodule; (B) pink background; irregular dotted and linear vessels; bright white structures; and (C) epithelial proliferation of tubular and acinar structures covered by a double layer of abundant ductal cells and myoepithelial cells on the outer layer.

Case 2: (D) Well-circumscribed light red nodule with a shiny bright pink surface, (E) pink background, shiny white area, telangiectatic vessels; and (F) tumor proliferation is made of tubulo-papillary structures within a cystic cavity (H&E, $\times 40$).

Case 3: (G) Well-circumscribed nodule, firm to touch, with a shiny pink surface; (H) red background, shiny white area, irregularly arranged serpentine vessels; and (I) well-limited tumor proliferation, papillary and glandular architecture, surrounded by sparse fibrous tissue.

Case 4: (L) Well-circumscribed nodule, firm to touch, with a shiny pink surface; (M) red background, shiny white area, irregularly arranged serpentine vessels; and (N) proliferation is made up of two cellular contingents: an epithelial component with ducts of variable size without atypical character and a connective component made of regular fibroblasts, which are not very cellular.

well-limited tumor proliferation and papillary and glandular architecture surrounded by sparse fibrous tissue. In the case where dermoscopy revealed the presence of a bluish globules, histology showed an intra-tumoral hemorrhage. Table 1 summarizes patient characteristics, clinical, and dermoscopic aspects of lesions.

HP is a benign tumor that occurs primarily in the vulva of Caucasian women [1]. It tends to occur most often in the fourth decade [2,3]. It is a rare, slow-growing, adnexal tumor mainly located in the vulvar interlabial sulcus, the most common location for mammary-like anogenital glands. HP have a good prognosis, and malignant transformation is extremely rare [4].

Dermoscopic findings of HP have been poorly reported in the literature. A case series of seven vulvar HPs

in a multiracial population has been reported, and it also showed similar dermoscopic features to our case series [1]. The majority of lesions had a triad made by a red background, shiny white structures which corresponds histologically to fibrous tissue, and polymorphous vessels which corresponds histologically to expanded vessels. Another dermoscopic feature was reported: a blue homogeneous pattern associated with peripheral, large vessels without pigmentation [1].

Considering the pitfall of clinical and dermoscopic polymorphism, vulvar HP may mimic other benign or malignant neoplasms. The differential diagnosis includes vulvar melanoma, syringocystadenoma papilliferum, eccrine porocarcinoma [5], squamous cell carcinoma, and nodular lesions in extramammary Paget diseases [3].



Figure 2. Case 5: (A) Pink nodule of the right interlabial sulcus; (B) pink background, shiny white structures of reticular layout, dotted vessels. **Case 6:** (C) Small nodule with normal skin color;(D) pink background, shiny white structures, hairpin vessels, bluish globules. **Case 7:** (E) Polylobate tumor of firm consistency taking the vulva and the perianal region; (F) clinical photo after reconstructive surgery; and (G, H) proliferation has 2 cellular components: an epithelial one with ducts of variable size without atypical character and another conjunctive one made of regular fibroblasts (magnification $\times 100x$, $\times 400$).

Conclusion

HP is clinically polymorphous, mimicking other benign or malignant neoformations, so histological examination is necessary to confirm the diagnosis and provide treatment.

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