

CASE REPORT

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Perianal hidradenoma papilliferum in a male: A case report and review of the literature

Lily Li, Hui Sze Wong, Danielle Taylor, Ruchira Fernando

ABSTRACT

Introduction: Hidradenoma papilliferum is a rare, benign, predominantly cystic tumor that predominantly affects women between 30 and 60 years of age. It is thought to originate from anogenital mammary-like glands, and therefore most commonly arises in the anogenital region. Clinically, it typically presents as a slow-growing, solitary, circumscribed nodule measuring between 0.5 cm and 2.0 cm in diameter.

Case Report: We report a rare case of a perianal hidradenoma papilliferum in a 70-year-old male who presented with a 2 cm highly pedunculated perianal lesion at the anal verge. This lesion had reportedly been present for several months and had recently increased in size. A literature review was completed and a total of 15 cases of hidradenoma papilliferum were identified in men, of which only three involved the perianal region. The majority of cases (9/15) were located in the head and neck region.

Conclusion: We therefore report a new case of this rare entity. To our knowledge, this is only the fourth documented case of perianal hidradenoma papilliferum in a male patient.

Keywords: Benign tumor, Hidradenoma papilliferum, Papillary hidradenoma, Perianal

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INTRODUCTION

Hidradenoma papilliferum (HP) is a rare, benign cystic tumor that occurs predominantly in women aged 30 to 60 [1]. It is thought to originate from anogenital mammary-like glands, and therefore most commonly arises in the anogenital region with vulvar lesions reported approximately four times more frequently than perianal lesions [2]. Rare ectopic cases have also been reported, typically in the head and neck. Interestingly, while anogenital HP occurs almost exclusively in women, ectopic forms demonstrate a more balanced sex distribution, with nearly half of reported cases arising in men [3]. Here, we report a rare case of perianal HP in a 70-year-old man and review the literature on male HP. To our knowledge, this is only the fourth documented case of perianal HP in a male patient.

CASE REPORT

A 70-year-old man presented to our General Surgery clinic for a surveillance colonoscopy following previous findings of multiple tubular adenomas with low-grade dysplasia. During the procedure, a 2 cm polyp at the anal verge was incidentally identified but not removed (Figure 1). On return to clinic for discussion of the colonoscopy results, further assessment of the anal lesion was undertaken.

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On examination, there was a highly pedunculated mass measuring approximately 2 cm in diameter at the 11 o'clock position. This lesion had reportedly been present for several months and had been recently increasing in size. There was no history of bleeding, pain, or pruritus. The patient's past medical history included obesity, hypertension, and hypercholesterolemia.

Pathology Findings with IHC Confirmation

Surgical excision under general anesthesia was performed for histological evaluation. Informed consent was obtained from the patient for publication of this case report and accompanying images.

Gross Findings

Soft tissue specimen measuring 18 × 12 × 4 mm. The specimen was sectioned into five pieces for histological examination.

Histopathological Findings

Microscopic examination demonstrated a polypoid lesion with anal squamous epithelial covering (Figure 2). There was a well demarcated cystic lesion in the dermis lined by double layered epithelium with myoepithelial cells and inner cuboidal to columnar cells with apocrine snouts. Focally, there was an intramural solid area with tubular and cystic structures with benign eccrine and apocrine epithelium. Low papillary-like structures were also seen. There were no features of atypia or malignancy.

Immunohistochemistry (IHC) Findings

Immunohistochemistry showed tumor cells positive for GATA3, estrogen receptor, and progesterone receptor (Figure 3). Taken together, the morphological and immunohistochemical findings supported a diagnosis of HP.

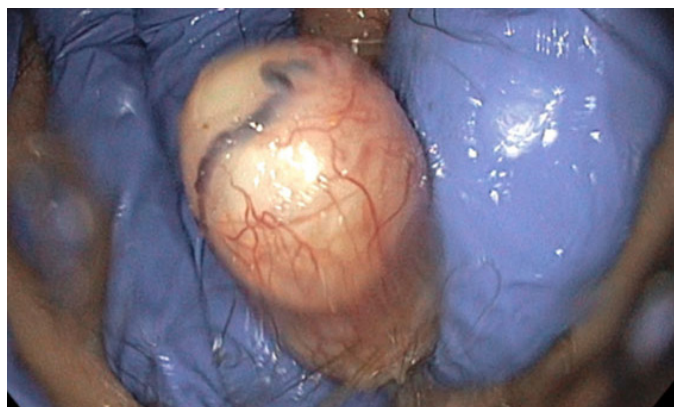


Figure 1: Endoscopic image of a perianal hidradenoma papilliferum obtained during colonoscopy. The lesion appears as a well-circumscribed, firm, nodular mass with a smooth surface and overlying telangiectasia.

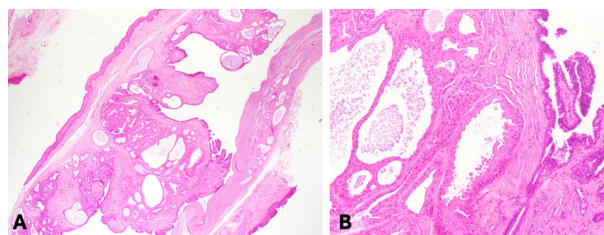


Figure 2: Histopathological images showing tumor composed of irregular glandular structures and papillary folds, characteristic of hidradenoma papilliferum. (A) (hematoxylin and eosin, ×2), (B) (hematoxylin and eosin, ×10)

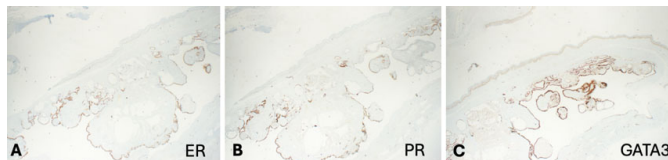


Figure 3: Immunohistochemical staining of hidradenoma papilliferum. Tumor cells demonstrating positivity for (A) estrogen receptor (ER) (B) progesterone receptor (PR) and (C) GATA 3.

DISCUSSION

Though first described in 1878, HP remains under-recognized, and its pathogenesis poorly understood. Typically presenting as an asymptomatic, slow-growing, raised nodule, some patients may report pain, pruritus, ulceration, and bleeding [2]. Lesions usually range from 0.5 cm to 2.5 cm in diameter, though sizes up to 10 cm have been reported [4].

Initially believed to arise from apocrine sweat glands, current evidence suggests that HP originates from anogenital mammary-like glands which are morphologically similar to breast tissue but are normally present in the interlabial sulcus [2, 5]. Like normal breast tissue, most lesions express estrogen and progesterone receptors and are influenced by female sex hormones, likely accounting for their predominance in post-pubertal women. Nevertheless, cases have also been documented in men and at ectopic sites. Ectopic HP is believed to arise from modified apocrine glands such as the glands of Moll at the eyelid margin or the ceruminous glands of the external auditory canal [6, 7]. While HP in women is strongly linked to the anogenital region, cases in men are more frequently ectopic. Additionally, the role of sex hormones in the pathogenesis of male HP is thought to be less significant than in women; however, our patient's tumor did demonstrate positivity for both estrogen and progesterone receptors. More research is required to clarify the contribution of hormonal pathways to HP development in both sexes.

To better characterise HP in the male population, we conducted a review of the literature and identified 15 case reports of HP in men (Table 1). Only three of these cases involved the perianal region and thus, our case represents the fourth reported instance, highlighting the rarity of the condition.

Consistent with the literature, the majority of ectopic HP in our review occurred in the head and neck region (9/15, 60%) though other sites such as the arm, chest wall, and areola were also described [3, 8, 9]. We found that in men, perianal HP seemed to present at a higher mean age compared with ectopic HP (61.5 years vs 52.5 years). This observation differs slightly from existing literature, which reports that ectopic HP presents on average 10–20 years later than anogenital lesions [3]. Considering our exclusive focus on male patients and the limited number of reported cases, these findings should be interpreted with caution. Nevertheless, clinicians should be aware of the possibility of HP in an older male presenting with a perianal lesion.

Table 1: Cases of male hidradenoma papilliferum in the English-language literature

Author, year	Age (year)	Location	Size (cm)
Binns, 1974 [10]	55	Post-auricular	2 × 0.3 × 0.3
Netland, 1990 [6]	78	Eyelid	0.6
Loane, 1998 [11]	68	Perianal	2.5 × 2 × 1
Vang, 1999 [3]	72	Arm, overlying triceps muscle	0.6
Tanaka, 2003 [8]	62	Chest wall	0.8
Smith, 2003 [12]	37	Nasal tip	
Minami, 2006 [13]	52	Eyebrow	1.6
Rosmaninho, 2010 [14]	26	Eyelid	1
Jain, 2012 [15]	80	Eyelid	1.2
Fernandez-Flores, 2012 [16]	38	Perianal	0.3
Panasiti, 2014 [17]	62	Scalp	0.5
Huddleston, 2016 [18]	70	Perianal	0.3
Kondo, 2018 [9]	36	Areola	1
Shukla, 2018 [19]	16	Upper and lower eyelid (2 lesions)	1 1
Laababsi, 2020 [7]	56	External auditory canal	1.5

The overall prognosis of HP is excellent, with surgical excision being curative in the vast majority of cases. Rare instances of local recurrence have been described [20], and there are occasional reports of malignant transformation in the forms of mammary-type hidradenocarcinoma, adenocarcinoma in situ and adenosquamous carcinoma [21, 22, 23]. Human papillomavirus (HPV), particularly HPV-16, has been detected in some of these lesions; however, a causal relationship has not yet been established [24].

CONCLUSION

In conclusion, we describe a fourth case of male perianal HP, a rare benign tumor most commonly found in women. It is often asymptomatic and may be overlooked or misdiagnosed due to its lack of distinctive clinical features. Surgical excision remains necessary for both definitive diagnosis and curative treatment.

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Author Contributions

Lily Li – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Hui Sze Wong – Conception of the work, Design of the work, Acquisition of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Danielle Taylor – Conception of the work, Design of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Ruchira Fernando – Acquisition of data, Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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