

## Case Report

# Imaging findings of big hidradenoma papilliferum in ischiorectal fossa: a case report

Xiang Chen, Jingtao Wu

Department of Medical Imaging, Subei People's Hospital, Clinical College of Yangzhou University, Yangzhou, Jiangsu, P. R. China

Received October 25, 2016; Accepted April 14, 2017; Epub August 15, 2017; Published August 30, 2017

**Abstract:** Hidradenoma papilliferum is a rare, benign skin tumor. However, when it locates uncommonly in ischiorectal fossa, it is very difficult to make preoperational diagnosis correctly. There haven't been any relative CT and MRI literature reported so far. Hence, we reported a case of big ischiorectal hidradenoma papilliferum in a 65-year-old woman with CT and MRI data. The lesion demonstrated a well circumscribed cystic-solid tumor with mild homogeneous enhancement of wall and intracystic nodule. Such imaging feature will improve our knowledge of hidradenoma papilliferum and expand the scope of thought in the differentiating diagnosis of perianal disorders.

**Keywords:** Hidradenoma papilliferum, ischiorectal fossa, magnetic resonance imaging, tomography, X-ray computed

### Introduction

Hidradenoma papilliferum (HP) is a rare adenoma of the mammary like anogenital glands [1]. It almost locates in the skin. Perianal area, ocular region, vulva are its predilection sites. The skin lesion usually manifests single solid or cystic nodule in or under the derma with clear margin. Its diameter is about 0.5 cm to 1.5 cm. Most cases are asymptomatic, but it may cause hemorrhage or serous effusion when the surface ulcers. The common disorders in ischio-rectal fossa are mesenchymoma and perianal abscess. HP that locates ischio-rectal fossa is particularly uncommon and unpredictable, so far there haven't been any relative imaging reports on such cases. A correct preoperational diagnosis may contribute to less invasive treatment strategies. We described the imaging findings of a case of HP arisen from perianal area.

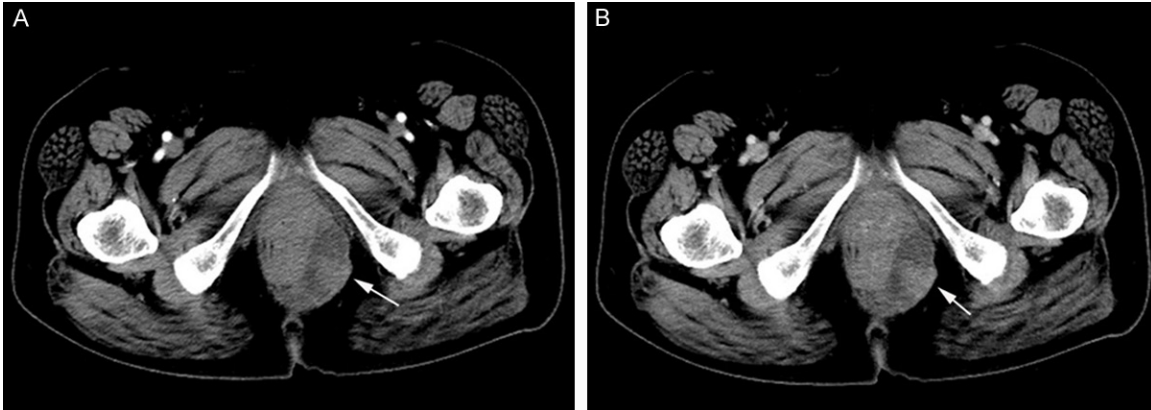
### Case report

A 65-year-old woman was admitted to our hospital with anal bulge for one month. She complained no fever and hematochezia. On physical examination, a palpable mass was felt under the perianal skin, but it was invisible on

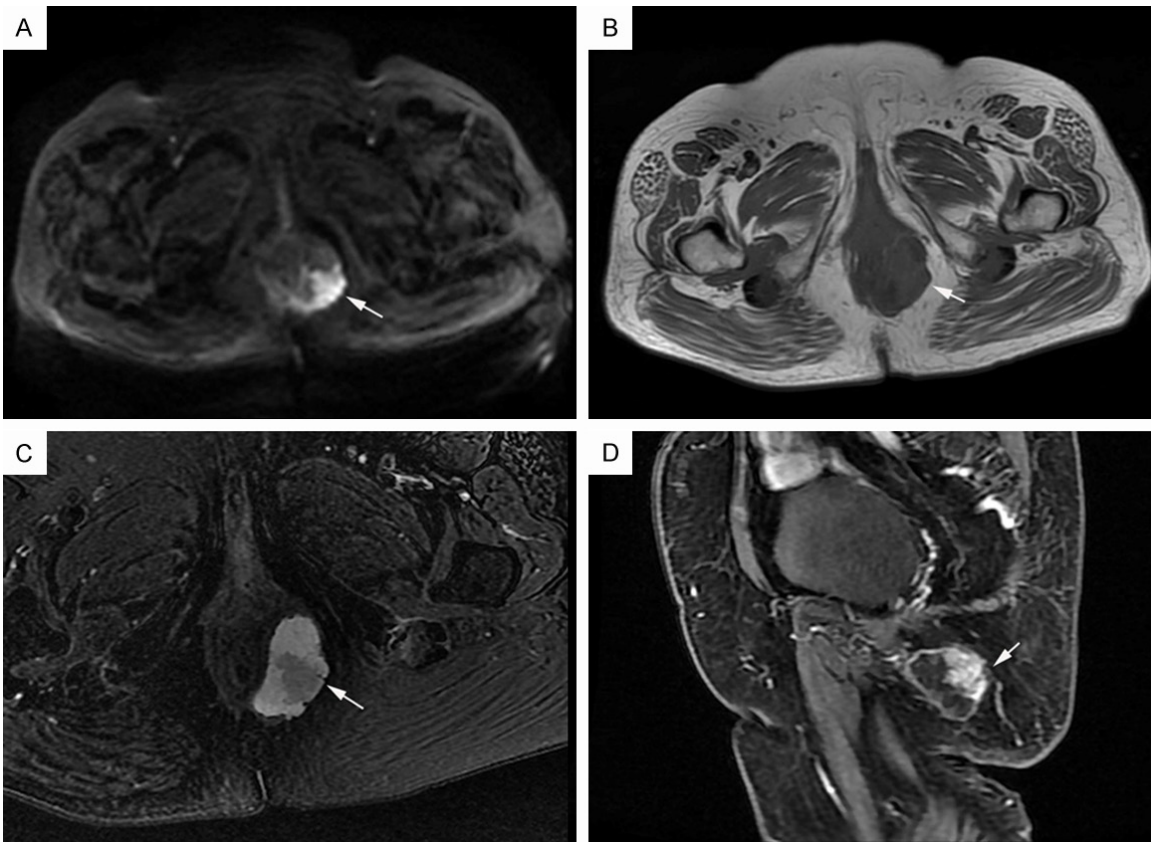
the surface of the skin. Digital rectal examination revealed a mass protruding from the left wall of the rectum. The white blood cell count was normal. Serum tumor markers including carbohydrate antigen (CA) 50, CA199, carcino-embryonic antigen were all negative.

Enhanced computed tomography (CT) revealed an oval, cystic-solid mass located in the ischio-rectalfossa. The size was 52 mm×27 mm×61 mm. The boundary of the mass was clear. The cystic wall and intracystic solid component exhibited mild and homogeneous enhancement in the arterial phase (**Figure 1A**), and presented more strong enhancement in the venous phase (**Figure 1B**), respectively. No enlarged lymph nodes were observed in the pelvic cavity and bilateral inguinal folds. Magnetic resonance imaging (MRI) confirmed the location of the lesion. The solid portion exhibited hypointensity on T1-weighted imaging (**Figure 2B**), slightly hyperintensity on T2-weighted imaging (**Figure 2C**) and hyperintensity on diffusion-weighted imaging (**Figure 2A**). Enhancement of the solid nodule and cystic wall were homogeneous (**Figure 2D**). As it was located outside of the rectum, the tumor was beyond the power of enteroscopy can reach. Preoperative biopsy was rejected by the patient.

## Radiology of perianal HP



**Figure 1.** A, B. Enhanced CT images of the HP in ischiorectal fossa. A. Enhanced CT showing a tumor in the left perirectal area with the cystic wall and intracystic solid component mild enhancement in the arterial phase. B. Further enhancement of the intracystic solid component in the venous phase.

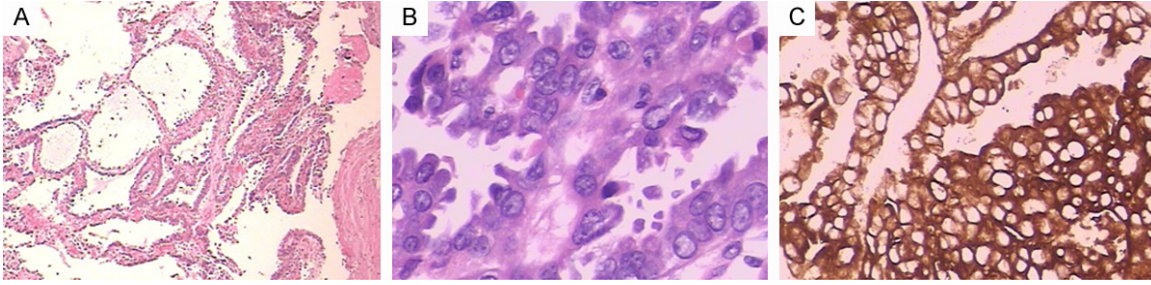


**Figure 2.** A-D. MRI images of the HP in ischiorectal fossa. A. The intracystic solid component exhibiting hyperintensity on diffusion weighted imaging. B. Axial T1-weighted imaging demonstrating well-defined hypointense cystic-solid mass. C. The solid portion exhibiting slightly hyperintensity on axial T2-weighted imaging. D. Sagittal enhanced imaging illustrating homogeneous enhancement of the solid component.

Therefore, the tumor was suspected as a gastrointestinal stromal tumor (GIST) and removed by perineal excision subsequently. During the operation a partly cystic degenerated tumor was found beside the rectum with a size about 5 cm×3 cm×6 cm.

Microscopic examination demonstrated papillary or tubular structures formed with epithelium cells (**Figure 3A**). The epithelium cells were rich of eosinophilic cytoplasm with apical secretion. Some cells were surface hobnailing (**Figure 3B**). Dilation of glandular tubes was found

## Radiology of perianal HP



**Figure 3.** A-C. Microscopic views of the HP. A. HE staining demonstrating papillary or glandular tube like structures formed by epithelium cells (4×10). B. The epithelium cells were rich of eosinophilic cytoplasm with apical secretion, some cells were surface hobnailing (×400). C. Immunohistochemistry indicating the neoplastic cells positive for CK7 (×200).

because of secretion accumulation. Immunohistochemistry indicated that the neoplastic cells were positive for Galectin-3, NapsinA, cytokeratin1 (CK) 7 (**Figure 3C**), and negative for CK20, Villin, gross cystic disease fluid protein (GCDFFP)-15, cluster of differentiation (CD) 10. Therefore, the lesion was final diagnosed as a HP. No postoperative complications were observed and the patient recovered well. There was no recurrence after 8 months follow-up.

### Discussion

HP is a rare, benign, cutaneous appendages tumor which occurs normally in the skin of the anogenital region. It mainly strikes women aged between 30 and 50, occasionally occurs in men [1-3]. Usually, the lesion is single, multiple focuses are relatively infrequent. It exclusively locates on the surface of labia minora. Such lesions in the breast, oter, eyelids, external auditory canal, auricular area, the limbs are uncommon [2], about 60% ectopic HPs locate in the head and neck regions [4].

The tumor is usually small with diameter about 10 mm [5] and asymptomatic, sometimes it may cause local itching. However, big lesion with diameter up to 40 mm had been reported [6]. On physical examination, the lesion is usually mobile, dense and often associated with tenderness. When the tumor is big and the surface ulcerates, it may be revealed by bleeding or discharge, which could be erroneously assessed malignancy [6]. There are some glands mimicking mammary gland around the vulva and anus, which do not express gross cystic disease fluid protein (GCDFFP)-15. Recently, most scholars accept that HP origins from these glands. The Histologic feature of HP

shows papillary or glandular structures with the epithelium cells rich in eosinophilic cytoplasm [7]. On immunohistochemistry most epithelial cells were positive for estrogen receptor (ER), progesterone receptor (PR), cytokeratin1 (CK) 7, but negative for CK20 and GCDFFP-15 [8]. In the present case, the perianal HP presented as a cystic mass with a wall-attached solid nodule. The epithelium cells formed glandular tube structures, and expressed CK7. These histopathologic features were in favor of HP, which was similar to that reported in the literature.

Generally, pathological features determine the imaging manifestations and the enhancement pattern. Subcutaneous HP manifests a subcutaneous hypoechoic cystic-solid nodule on ultrasonic examination [1]. In the present case, the HP was also a cystic-solid tumor. Daniel et al. [2] reported a perianal HP, which was a cystic lesion surrounded by a fibrous capsule with a wall-attached solid component presenting tubular and papillary structures on the microscopic view, the cystic cavity was filled with mucoid content. So the tumor manifested a cystic lesion with wall-attached solid nodule on the CT and MRI imaging. The wall and inner portion mild enhancement was possibly due to the fibrous cystic wall and parenchyma cellular area with slight blood supply. The mucus in the cystic area exhibits hypodensity in CT imaging, hypointensity in T1WI, hyperintensity in T2WI. Because of its benign nature, the boundary of the tumor is clear.

Perianal HP can be confused with other perianal conditions, including perianal abscess and mesenchymoma. The definite diagnosis relies on pathology. Mesenchymoma is a mixed mesenchymal tumor composed of two or more cel-

lular elements that are not commonly associated, exclusive of fibrous tissue. Mesenchymomas locate in the pelvic cavity rather than subcutaneous area. Mesenchymomas are not cystic. Most mesenchymomas present extraversion growth, heterogeneous enhancement, necrosis in the center with rare lymphatic metastasis, and malignant ones may have peripheral invasion [9]. An overwhelming majority of the stromal tumors express cluster of differentiation (CD) 117 on immunohistochemical examination [10, 11]. The typical characteristic of perianal abscess is perianal U shaped low density with obvious abscess wall enhancement on CT scans. MRI has more diagnostic value on showing this feature [12]. Abscesses don't contain solid nodule. Whereas, HPs locate in the surface of the skin or in the subcutaneous area. HPs are well-marginated, cystic-solid tumor with slight cystic wall and intracystic nodule homogenous enhancement. There are no regional invasion and no lymph node enlargement. Such features may alert the radiologist to a possible HP diagnosis. MRI is the best preoperative imaging for HP diagnosis.

Resection is the most effective treatment for HP, and the probability of late recurrence is rare for long-term follow-up [1]. HP can accompany with a ductal carcinoma in situ component [13] or extramammary Paget disease [14]. Human papillomavirus (HPV)-16 may contribute to the malignant transformation [13]. In this case, no recurrence was detected after 8 months follow-up.

### Conclusion

HP has some CT and MRI features, such as clear margin, cystic with solid components, homogenous enhancement of the cystic wall and intracystic nodule, no regional invasion and no lymph node enlargement. The present case suggests that an HP can occur in ischio-rectal fossa and should be taken into consideration in the differential diagnosis when a cystic-solid mass has been detected in the perirectal area.

### Disclosure of conflict of interest

None.

**Address correspondence to:** Jingtao Wu, Department of Medical Imaging, Subei People's Hospital, Clinical College of Yangzhou University, No 98 West

Nantong Road, Yangzhou 225001, Jiangsu, P. R. China. Tel: 13852551359; E-mail: witsby2014@163.com

### References

- [1] Scurry J, van der Putte SC, Pyman J, Chetty N and Szabo R. Mammary-like gland adenoma of the vulva: review of 46 cases. *Pathology* 2009; 41: 372-378.
- [2] Daniel F, Mahmoudi A, de Parades V, Flejou JF and Atienza P. An uncommon perianal nodule: hidradenoma papilliferum. *Gastroenterol Clin Biol* 2007; 31: 166-168.
- [3] Minami S, Sadanobu N, Ito T, Natsuaki M and Yamanishi K. Non-anogenital (ectopic) hidradenoma papilliferum with sebaceous differentiation: a case report and review of reported cases. *J Dermatol* 2006; 33: 256-259.
- [4] Vang R and Cohen PR. Ectopic hidradenoma papilliferum: a case report and review of the literature. *J Am Acad Dermatol* 1999; 41: 115-118.
- [5] Kazakov DV, Mikyskova I, Kutzner H, Simpson RH, Hes O, Mukensnabl P, Bouda J, Zamecnik M, Kinkor Z and Michal M. Hidradenoma papilliferum with oxyphilic metaplasia: a clinicopathological study of 18 cases, including detection of human papillomavirus. *Am J Dermatopathol* 2005; 27: 102-110.
- [6] Handa Y, Yamanaka N, Inagaki H and Tomita Y. Large ulcerated perianal hidradenoma papilliferum in a young female. *Dermatol Surg* 2003; 29: 790-792.
- [7] Zywica M, Hartschuh W and Hamsch C. Firm nodule in the perianal region of a 59-year-old woman. *J Dtsch Dermatol Ges* 2013; 11: 682-685.
- [8] Nishie W, Sawamura D, Mayuzumi M, Takahashi S and Shimizu H. Hidradenoma papilliferum with mixed histopathologic features of syringocystadenoma papilliferum and anogenital mammary-like glands. *J Cutan Pathol* 2004; 31: 561-564.
- [9] Schoellnast H, Preidler K and Szolar D. Malignant perirenal mesenchymoma with renal infiltration. *Imaging in CT and MRI. Rofo* 2004; 176: 263-264.
- [10] Yamamoto H, Oda Y, Kawaguchi K, Nakamura N, Takahira T, Tamiya S, Saito T, Oshiro Y, Ohta M, Yao T and Tsuneyoshi M. c-kit and PDGFRA mutations in extragastrointestinal stromal tumor (gastrointestinal stromal tumor of the soft tissue). *Am J Surg Pathol* 2004; 28: 479-488.
- [11] Fletcher CD, Berman JJ, Corless C, Gorstein F, Lasota J, Longley BJ, Miettinen M, O'Leary TJ, Remotti H, Rubin BP, Shmookler B, Sobin LH and Weiss SW. Diagnosis of gastrointestinal stromal tumors: a consensus approach. *Hum Pathol* 2002; 33: 459-465.

## Radiology of perianal HP

- [12] Halligan S and Stoker J. Imaging of fistula in ano. *Radiology* 2006; 239: 18-33.
- [13] Vazmitel M, Spagnolo DV, Nemcova J, Michal M and Kazakov DV. Hidradenoma papilliferum with a ductal carcinoma in situ component: case report and review of the literature. *Am J Dermatopathol* 2008; 30: 392-394.
- [14] Stefanato CM, Finn R and Bhawan J. Extramammary Paget disease with underlying hidradenoma papilliferum: guilt by association? *Am J Dermatopathol* 2000; 22: 439-442.