



Case Report

Villous adenoma of bladder with uncommon location in a super-aged patient without gross hematuria

Yoon Soo Hah  and Hyun Jin Jung 

Department of Urology, Catholic University of Daegu School of Medicine, Daegu, Korea

Abbreviations & Acronyms

CK = cytokeratin

CT = computed tomography

Correspondence: Hyun Jin Jung M.D., M.M.S., Department of Urology, Catholic University of Daegu School of Medicine, 33 Duryugongwon-ro 17-gil, Nam-gu, Daegu 42472, Republic of Korea.
Email: hnjini@cu.ac.kr

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Introduction: There have been few reported cases of villous adenoma of the bladder. It commonly occurs in the superior area of the bladder with hematuria as the most common symptom. Here, we have presented a case of villous adenoma of the bladder neck and reviewed the existing literature.

Case presentation: A 90-year-old man presented with voiding difficulty. Although urine analysis revealed microscopic hematuria and pyuria, the patient never complained about gross hematuria. Ultrasonography and cystoscopic examination revealed a bladder tumor located at the bladder neck. Transurethral resection was performed and villous adenoma was diagnosed by histopathologic examination.

Conclusion: Villous adenoma of the bladder is a rare disease, which is difficult to diagnose when the patient presents with uncommon clinical features. Although villous adenoma is known as a benign tumor, some reports suggest its association with malignancy. Therefore, careful management and follow-up are necessary.

Key words: aged, hematuria, rare disease, urinary bladder neoplasms, villous adenoma.

Keynote message

Villous adenoma of the bladder is a rare disease and usually has typical symptoms and location. Most commonly it is found in the superior area of the bladder and associated with gross hematuria. However, it is difficult to diagnose this lesion if the patient presents with uncommon clinical features. Although villous adenoma of the bladder is known as a benign tumor, some reports suggest that it may have malignant potency, and therefore, careful management and follow-up are necessary.

Introduction

Villous adenoma is presumed to be a premalignant tumor mainly found in the gastrointestinal system. Villous adenoma in the bladder known as benign glandular neoplasm has been reported in very few cases. However, its epidemiology has not been clearly identified. In an article that reported on 23 cases, the male to female sex ratio was reported as 1.1:1 and the average age was 62 years.^{1,2} Villous adenoma was mainly found in the bladder dome or urachus, and the main symptom was gross hematuria. It is not easy to diagnose this rare disease without predominant symptoms and features. Only one case of bladder villous adenoma has been reported in Korea in 2008, and to the best of our knowledge, there have been no reports of villous adenoma in the bladder neck without gross hematuria in patients >90 years old.³

Case presentation

A 90-year-old man with well-controlled hypertension and diabetes mellitus presented to our institution with voiding difficulty. The maximum flow rate of uroflowmetry was under 5 mL/s and transabdominal bladder and transrectal ultrasonography were performed. An ambiguous mass lesion of the bladder or intravesical protruding prostate was observed on ultrasound (Fig. 1a,b). Digital rectal examination showed no abnormal findings. Microscopic hematuria

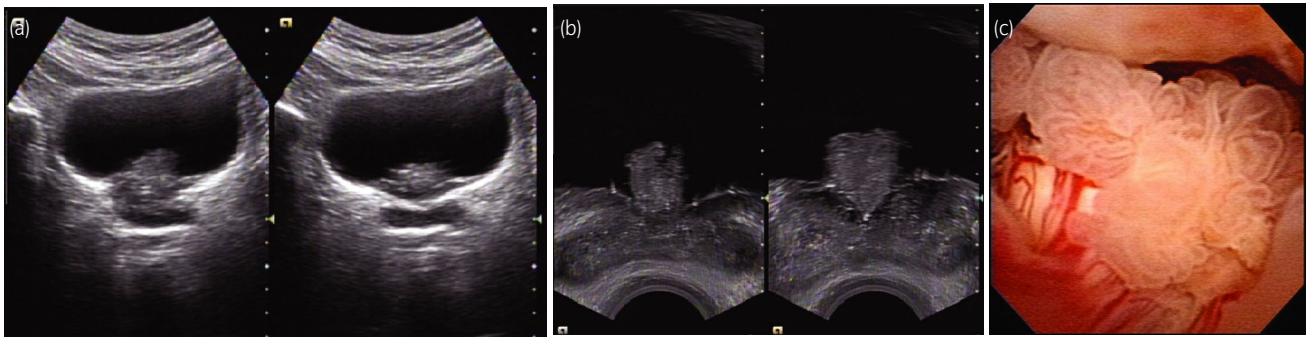


Fig. 1 Preoperative examination. (a) Transabdominal ultrasonography of the villous adenoma. The origin of mass is not clearly visible on the ultrasonogram. (b) Transrectal ultrasonography of the villous adenoma. The ultrasonogram shows the mass which seems to have risen from the bladder neck or intravesical protruding prostate. (c) Cystoscopic examination shows multiple papillary lesions sized approximately 2 cm surrounding the bladder neck.

and pyuria were observed on urinalysis and the prostate specific antigen level was 3.01 ng/mL. Cystoscopic evaluation was performed to confirm the ambiguous bladder lesion and multiple papillary lesions sized approximately 2-cm were observed surrounding the bladder neck (Fig. 1c). Bladder washing cytology was performed during cystoscopy and reported as negative for malignancy. An enhanced CT showed a 2-cm sized bladder neck mass with enhancement and further invasion to the prostate or bladder wall was not clear. The CT scan did not reveal any other remarkable findings such as enlarged lymph nodes or suspicious visceral metastatic lesions.

We made a diagnosis of malignant bladder tumor and performed transurethral resection. The pathological structure showed villous or tubulovillous architecture, finger-like projection with epithelial pseudostratification, and hyperchromasia (Fig. 2a). The mucin was found in the cytoplasm through alcian-blue stain (Fig. 2b). Histopathology confirmed the papillary and villous proliferation of columnar cells with nuclear stratification. The surrounding mucosa showed small gland proliferation with inflammation. Immunostaining for

CK20 and CK7 were positive (Fig. 2c,d) and expression of p53 was confirmed by molecular study. No local recurrence was found at follow-up 6 months after surgery.

Discussion

Symptoms of voiding difficulty are common in old age, and therefore, we sometimes start empirical treatments without detail examination. Aging is considered a reason for no clear improvement in clinical findings in super-aged patients, and thus, conservative treatment is maintained. Benign tumor in the bladder often exhibit symptoms of voiding difficulty rather than gross hematuria, depending on their location.⁴ Further study should be actively considered if the patient presents with voiding difficulty accompanied by obstructive symptoms that do not respond to medication.

The mean age at which villous adenoma usually appears is not clear yet. Several case series reported the average age to be 62 or 69.6 years.^{1,2,5} We reviewed 21 cases of pure villous adenoma without any associated malignancy (Table S1). The

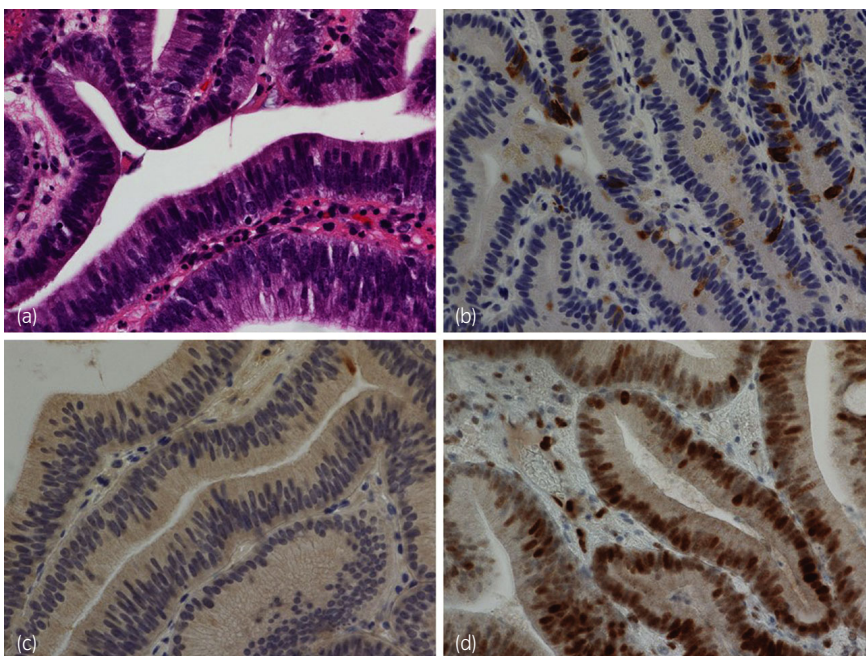


Fig. 2 Histopathology of the tumor specimen. (a) The villous adenoma resembles colonic tubulovillous adenoma having finger-like projection with epithelial pseudostratification and hyperchromasia ($\times 400$). The tumor cells are positive for (b) CK20 ($\times 400$), (c) CK7 ($\times 400$) and (d) p53 ($\times 400$) was expressed.

Table 1 Summary of clinical features of villous adenoma in previously reported cases

	n (%)
Age	59.5 ± 15.9
Sex	
Male	13 (65.0)
Female	7 (35.0)
Symptoms	
Hematuria	10 (47.6)
Obstructive voiding symptom	2 (9.5)
Other voiding symptoms	1 (4.8)
Urinary tract infection	4 (19.0)
Mucosuria	3 (14.3)
Other symptoms	1 (4.8)
Sites	
Dome or urachus	9 (45.0)
Anterior wall	1 (5.0)
Posterior wall	3 (15.0)
Lateral wall	1 (5.0)
Trigone	2 (10.0)
Bladder neck	1 (5.0)
Urethra	3 (15.0)

average age was 59.5 years (Table 1). However, the range was so wide that it was not useful for clinical reference. Furthermore, it was often confirmed in super-aged patients (>90 years old). Although the clinical manifestations of villous adenoma were various, gross hematuria was the most common chief complaint (47.6%) and cases with symptoms of urination disorders were relatively rare. The urinary bladder develops from the endodermal urogenital sinus, mesonephric duct, and allantois structure. During development, the bladder shares the cloacal origin with some part of colorectum and the rest of cloaca; this could be presumed as a cause of epithelial neoplasm. Embryologically, it is possible to find villous adenoma in the urachus, bladder, urethra, and prostate; however, it has been reported to frequently occur at the dome of the bladder or urachus (45.0%) rather than the bladder neck (5.0%), which can have a physical effect of bladder emptying. While the villous adenoma is rare and has a wide range of onset age, the common symptom and location is characteristic. Therefore, diagnosis of villous adenoma may be difficult or delayed if the patient presents with uncommon features. Villous adenoma located in the bladder neck not accompanied by gross hematuria is not easy to predict, although surgical treatment is necessary to improve symptoms.

It is difficult to distinguish villous adenoma from urothelial carcinoma by image study or cystoscopy. However, endoscopic resections must be performed if the bladder tumor is confirmed by evaluation. A cold biopsy might also be useful for patients who are inoperable due to old age or comorbidity. It is not challenging for a pathologist to distinguish between villous adenoma and urothelial carcinoma. All immunohistochemistry of villous adenoma was positive for CK20 with complete membranous and intense signal. However, 33–50% of the cases

were positive for CK7 with incomplete membranous and less intensity signal.⁶ Absence of GATA3 and p63 expression can distinguish villous adenoma from urothelial carcinoma.⁷ Villous adenoma appearing in urinary tract is known to be a benign tumor. However, some reports suggest that villous adenoma may have malignant potency or may coexist with other malignant tumors.^{5,8,9} Furthermore, cases of recurrent pure villous adenoma have been reported in the literature, and therefore, careful follow-up is necessary. Most of the cases showed favorable prognosis even without radical treatment.

In summary, we experienced a case of villous adenoma of bladder with uncommon clinical features. Villous adenoma of bladder is rare; however, with typical symptoms and location. Rare disease with uncommon clinical features is difficult to diagnose and leads to delayed treatment of patients. We believe that our study makes a significant contribution to the literature because to the best of knowledge, this is the first case to report villous adenoma in the bladder neck without gross hematuria in patients >90 years old.

Acknowledgment

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Conflict of interest

The authors declare no conflict of interest.

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Supporting information

Additional Supporting Information may be found in the online version of this article at the publisher's web-site:

Table S1. Detailed clinical features of villous adenoma in previously reported cases.