

Case Report

Clear cell adenocarcinoma of the prostatic urethra: A case report

Rumiko Sugimura,¹ Takashi Kawahara,^{1,2}  Go Noguchi,³ Daiji Takamoto,¹ Koji Izumi,¹ Yasuhide Miyoshi,¹ Takeshi Kishida,³ Masahiro Yao,² Mikiko Tanabe⁴ and Hiroji Uemura¹

¹Departments of Urology and Renal Transplantation, Yokohama City University Medical Center, ²Department of Urology, Yokohama City University Graduate School of Medicine, ³Department of Urology, Kanagawa Cancer Center, and ⁴Division of Diagnostic Pathology, Yokohama City University Medical Center, Yokohama, Kanagawa, Japan

Abbreviations & Acronyms

AC = adenocarcinoma

CCA = clear cell adenocarcinoma

MRI = magnetic resonance imaging

MVEC = methotrexate-vinblastine-epirubicin-cisplatin

PSA = prostate-specific antigen

RC = radical cystectomy

SCC = squamous cell carcinoma

UC = urothelial carcinoma

Introduction: Clear cell adenocarcinoma of the prostatic urethra in men is an extremely rare disease, with only eight case reports published.

Case presentation: A 56-year-old man visited our hospital for gross hematuria. Urinary cytology detected class V, cystoscopy showed no abnormal findings, and contrast-enhanced computed tomography also showed no abnormal findings in his upper urinary tract except for a low-enhancement lesion on his left prostate lobe. Magnetic resonance imaging revealed a cystic lesion surrounding the prostate that was suspected of being urethral or prostate cancer, so transurethral resection was performed. A papillary tumor was detected at the prostatic urethra, and after resecting this tumor, a cavity showing multiple tumors was observed. The final pathological diagnosis was clear cell adenocarcinoma. Laparoscopic radical cystectomy and urethrectomy were thus performed. The pathological diagnosis was the same as at the primary tumor site.

Conclusion: We herein report a case of clear cell adenocarcinoma of the prostatic urethra.

Key words: clear cell adenocarcinoma, prostatic urethra, urethra, urethral tumor.

Correspondence: Takashi Kawahara M.D., Ph.D., Departments of Urology and Renal Transplantation, Yokohama City University Medical Center, 4-57 Urafunecho, Minami-ku, Yokohama, Kanagawa 232-0024, Japan. Email: takashi_tk2001@yahoo.co.jp

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Received 4 October 2018; accepted 3 November 2018. Online publication 24 November 2018

Keynote message

- 1 Clear cell adenocarcinoma in men is an extremely rare entity, with only eight cases been reported in total.
- 2 Radiological findings showed diverticulum in the prostatic urethra in clear cell adenocarcinoma.
- 3 Radical cystectomy is effective in male cases of clear cell adenocarcinoma.

Introduction

CCA of the lower urinary tract, especially the urethra, is an extremely rare disease. Due to the higher incidence of urethral diverticulum in women than in men, most cases in CCA arise in women, and only eight cases have been reported in men.¹ Typical clinical complaints are gross hematuria, repeated urinary tract infections, and dysuria. Pathologically, cuboidal- or columnar-shaped clear cell and hobnail cells proliferate with tubulocystic, papillary, and diffuse pattern.^{1,2} We herein report a rare case of CCA of the prostatic urethra.

Case presentation

A 56-year-old man visited our department for his asymptomatic gross hematuria. Urinary cytology revealed class V, and cystoscopy showed no abnormal findings. A digital rectal examination revealed no remarkable findings. Contrast-enhanced computed tomography revealed a low-density lesion on his left prostate lobe. MRI showed a cystic lesion in his prostatic urethra, so transurethral resection and a prostate needle biopsy were planned (Fig. 1). PSA showed a normal value (0.599 ng/mL). We created three-dimensional images to understand the tumor location easily using the Osirix and MeshLab software programs (Fig. 2).

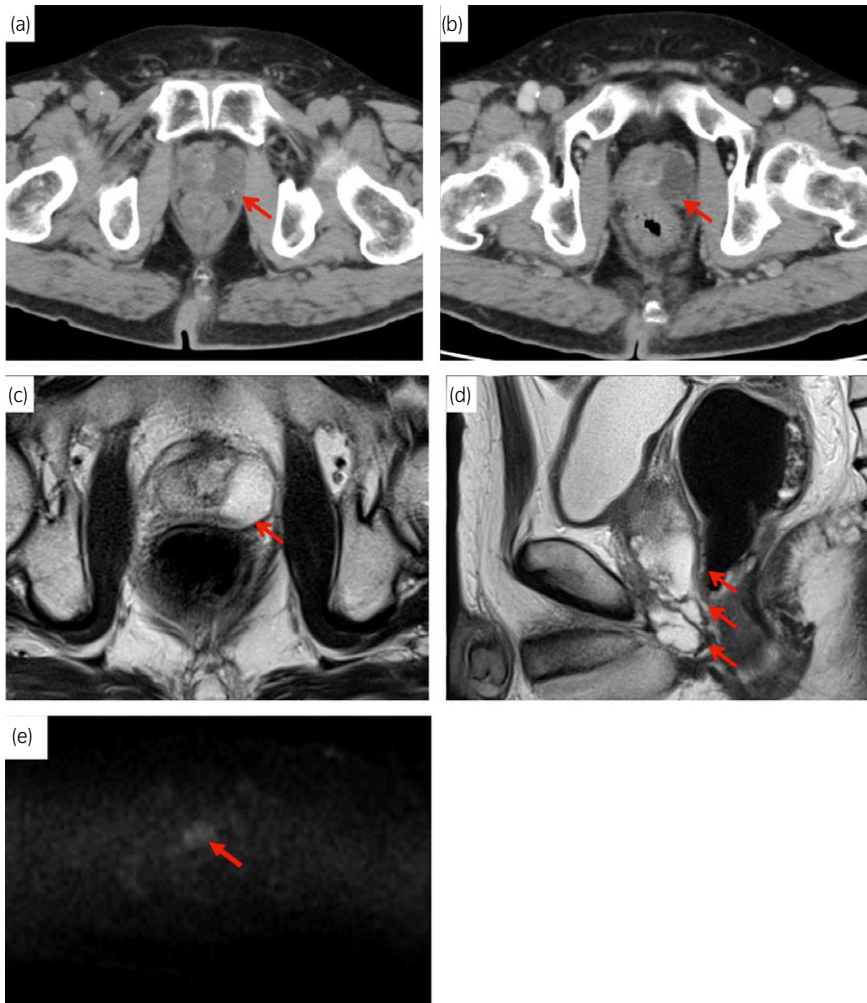


Fig. 1 (a) Non-contrast and (b) contrast-enhanced computed tomography. (c) Axial, (d) sagittal T2WI, (e) diffusion MRI findings.

At the time of transurethral resection, a papillary tumor nodule was observed in the cystic lesion (Fig. 3). A pathological examination showed epithelial tumor cells with clear or eosinophilic cytoplasm and irregularly enlarged nuclei. Immunohistochemical staining showed positivity for CK7, PAX-8; weak positivity for CK20, CK-HMW, and negativity for p63 and PSA. Based on these findings, CCA was diagnosed and a residual tumor was suspected based on transurethral resection findings (Fig. 3). RC with an ileal conduit and urethrectomy were therefore performed. The final pathological diagnosis was the same as the primary diagnosis, and lymph node metastasis was not observed. He was free from recurrence 5 months after RC.

Discussion

Urethral cancer is a relatively rare disease and is detected more often in women than in men, accounting for only 1% of all cancer among men. The most frequent sites of occurrence are the urethra bulb and membranous urethra followed by the pendulum part and prostatic urethra. Histologically, UC, AC, and SCC account for 24.9%, 46.7%, and 25.4% of urethral malignancies in female patients and 53.6%, 11.6%, and 34.8% of urethral malignancies in male patients,

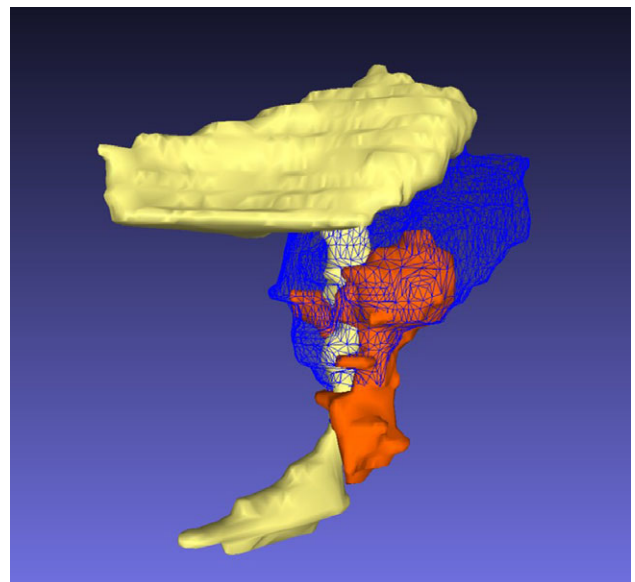


Fig. 2 Three-dimensional image of the anatomic location.

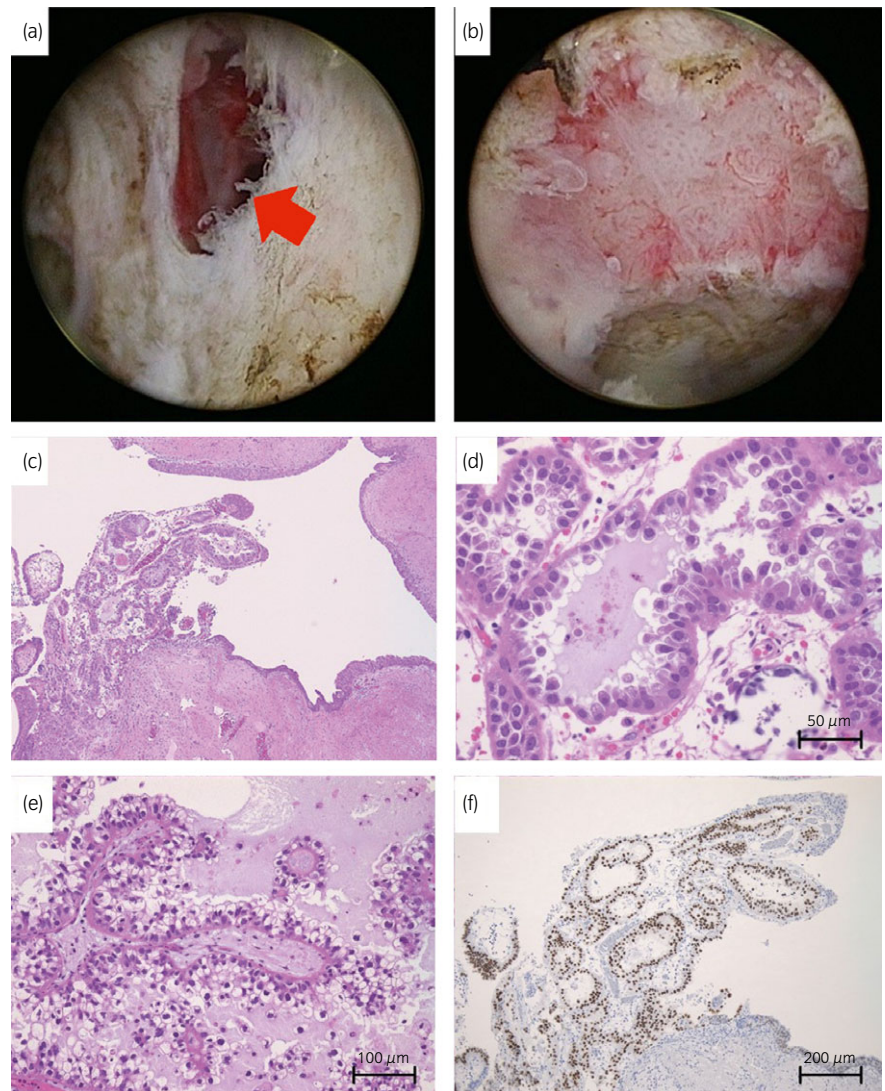


Fig. 3 (a,b) The tumor was located in the urethral diverticulum. HE findings: (c) the carcinomatous element consisted of CCA in the urethral diverticulum, (d) hobnail cells and (e) typical cytoplasm, and (f) immunohistochemical staining of PAX-8.

Table 1 Previous reports of CCA

Case	Report	Symptoms	Location	Treatment	Prognosis
1	Cantrell <i>et al.</i>	Gross hematuria, urinary retention	Verumontale	Radiation therapy	2 years 6 months, death by cancer
2	Oliva <i>et al.</i>	Urinary retention	Unknown	Unknown	Unknown
3	Seseke <i>et al.</i>	Urinary retention	Pendulum	Urethrectomy	2 years 6 months, death by cancer
4	Gogus <i>et al.</i>	Urinary retention	Bladder neck ~ urethra	RC + MVEC	10 years, death by cancer
5	Sun <i>et al.</i>	Gross hematuria, frequent urination	Unknown	Transurethral resection	Unknown
6	Varachhia <i>et al.</i>	Periurethral abscess	Bulbous	No treatment	2 months, death by cancer
7	Gandhi <i>et al.</i>	Urinary tract infection, perineal nodule, obstructive symptoms	Membranous, bulbous and penile	RC	Unknown
8	Lewis <i>et al.</i>	Micturition burning	Membranous, bulbous and penile	Brachytherapy	Alive, 11 months after diagnosis
9	Our case	Gross hematuria	Prostatic urethra	RC	Alive, 2 months after surgery

respectively.³ Clear cell carcinoma is categorized as an AC with columnar and mucinous cell carcinoma types.^{1,2}

CCA is frequently seen in women, and some cases have been reported as diverticulum tumor. Only eight cases in men have been reported (Table 1).^{2,4-9} Four of

these eight cases died of their cancer. No established treatment is available, although radiotherapy, cystectomy, and urethrectomy have been reported. In our case, due to the risk of recurrence and a poor outcome, RC was performed.

We performed RC because the diverticulum extended to the bladder neck and we could not rule out that the tumor extended to the bladder. However, the final pathological diagnosis revealed that the tumor had not extended to the bladder. Based on these findings, RC might not have been needed. No previous reports have described the follow-up and evaluation after bladder-preserving treatment. The present case was initially suspected of being non-UC based on urinary cytology, and repeated cystoscopy failed to detect any urethral tumor. MRI might therefore be useful for detecting non-cystoscopically finding urothelial tumors.

Hundreds of cases of CCA arising from urethral diverticulum in women have been reported.^{10,11} However, due to the extreme rarity of CCA in men, CCA with urethral diverticulum has not been reported in men. CCA of the urethra in men is extremely rare, with only eight cases reported to date. We herein report a rare case of CCA of the urethra with urethral diverticulum in a male patient. If positive urinary cytology and radiographic findings of urethra diverticulum are detected, CCA should be considered as a differential diagnosis.

Conflict of interest

The authors declare no conflict of interest.

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