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Ureteral ectopic prostate tissue: a case report and review of the literature

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Abstract

Background Ectopic prostate tissue is a rare clinical lesion that typically originates within the male urogenital tract. It is even more uncommon to find outside the urogenital tract or in female patients. The clinical symptoms of ectopic prostate tissue vary depending on the location of the disease.

Case presentation We present the case of a 56-year-old Chinese male of Asian descent who presented with a space-occupying left ureter during a physical examination but did not exhibit any specific clinical symptoms. Preoperative urinary tract color ultrasound and pelvic magnetic resonance imaging examinations revealed nodular, abnormal signal shadows in the lower segment of the left ureter, necessitating further investigation into their nature. The initial preoperative diagnosis leaned towards a ureteral tumor owing to the uncertain nature of the lesion. During laparoscopic left ureteral exploration, rapid pathological sections indicated the presence of mature prostate gland tissue within the affected area. Subsequent laparoscopic left ureteral bladder replantation was performed, and postoperative pathology, along with immunohistochemistry results, confirmed the presence of ureteral ectopic prostate tissue. A comprehensive review of relevant literature, both domestic and foreign, was conducted to analyze the location of the ectopic prostate tissue, clinical characteristics, and various diagnosis and treatment options for this condition.

Conclusion Ureteral ectopic prostate tissue is a rare disease that can be challenging to diagnose and treat accurately. Surgical removal of the affected tissue and subsequent histopathological examination are crucial steps to confirm the diagnosis. While the short-term prognosis for this condition is generally positive, long-term follow-up is necessary for optimal management.

Keywords Ectopic prostate, Ureteral mass, Case report, Literature review

Background

Ectopic prostate tissue (EPT) refers to prostate tissue that occurs outside the normal part of the prostate. It is clinically rare and was first reported by Jores in 1894 [1]. While EPT is most commonly found in the genitourinary system, it is uncommon in areas outside this system and in female patients. The clinical symptoms of ectopic prostate may vary depending on the location and size of the lesion, with some patients exhibiting no obvious symptoms. In this case study, we present a patient with a ureteral ectopic prostate who underwent surgical treatment to remove the affected tissue, with postoperative pathological results confirming the presence of ectopic prostate tissue. Through a review of relevant literature, both domestic and foreign, we have analyzed and summarized the clinical characteristics and experiences related to the diagnosis and treatment of ectopic prostate cases.

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Case presentation

The patient, a 56-year-old Chinese male, was found to have a space-occupying lesion in the left ureter during a physical examination conducted 7 months prior to admission. Following 3 months of monitoring, a color ultrasound continued to reveal the presence of the lesion, prompting the patient to request admission for further evaluation of its nature. Throughout the course of his condition, the patient reported no symptoms such as back pain, frequent urination, urgency, dysuria, gross hematuria, or any other uncomfortable symptoms. Upon inquiry about his medical history upon admission, the patient denied any prior history of hypertension, coronary heart disease, diabetes, or other malignancies, and there were no notable findings in his personal or family medical history. Upon digital rectal examination, a slightly enlarged prostate with a smooth and firm texture was noted, without any palpable nodules. The patient's serum total prostate-specific antigen (TPSA) level was 1.55 ng/mL, and other laboratory tests were within normal limits. Computed tomography urography (CTU) imaging revealed the thickening of soft tissue in the lower posterior part of the bladder, with uneven enhancement and bulging towards the bladder, but with clear separation from normal prostate tissue. The pelvic magnetic resonance imaging (MRI) examination revealed a nodular abnormal signal shadow in the lower segment of the left ureter, characterized by clear boundaries and complete separation from the prostate. Further investigation was warranted to determine the nature of this finding. (Fig. 1). Emission computed tomography (ECT) results showed slightly impaired left renal function, with smooth drainage in the upper urinary tract. Urine cytology examinations did not reveal any tumor cells. Cystoscopy showed no urethral lesions, mild prostate hyperplasia, normal bladder mucosa, and no abnormalities in the left ureteral orifice. Attempts to access the ureteroscope failed, preventing the biopsy of the mass.

The patient was initially diagnosed with a space-occupying lesion at the end of the left ureter. Preoperative imaging was inconclusive regarding the origin and nature of the tissue. Following a thorough discussion, it was decided to explore the ureter during surgery and conduct an intraoperative frozen pathology examination for further clarification. After meticulous preoperative preparation and addressing any surgical contraindications, a laparoscopic left ureteral exploration was performed under general anesthesia. The operation revealed a slightly firm tissue at the end of the ureter, clearly distinguishable from surrounding tissue, which was completely excised. Upon incision, the diseased tissue appeared gray-white with a narrow lumen, measuring approximately $2.5 \times 2.0 \times 1.0$ cm. Rapid frozen pathology indicated chronic inflammation of the ureteral mucosa, with visible mature prostate glands on the muscle wall. Subsequently, laparoscopic left ureterovesical reimplantation was successfully carried out. Postoperative immunohistochemistry results confirmed cytokeratin (CK)7 (+), CK20 (−), natural killer homeobox (NKX) 3.1 (+), tumor protein (P)63 (basal cell +), prostate-specific antigen (PSA) (+), GATA3 (weak +), and antigen Kiel (Ki)67+ < 1%, consistent with prostatic ectopia (Fig. 2). The patient was discharged following the removal of the pelvic drainage tube on the fifth postoperative day, with no surgical complications reported postoperation. We conducted intravenous urography and urinary tract computed tomography (CT) examinations at 3, 6, and 12 months after the surgery. The results indicated that the drainage of the left ureter was unobstructed, and no relevant recurrent tissue was observed at the ureteral orifice. The patient expressed high satisfaction with the postoperative recovery, and his condition remains stable after 14 months of follow-up.

Discussion

EPT refers to the presence of prostate tissue in atypical locations. This tissue can be found in various parts of the body, with a common occurrence in the urogenital system including the bladder, urethra, testicles, seminal vesicles, and ureteral orifices [2–5]. While rare outside the urogenital system, there have been documented cases of EPT in organs such as the spleen, small intestinal mucosa,

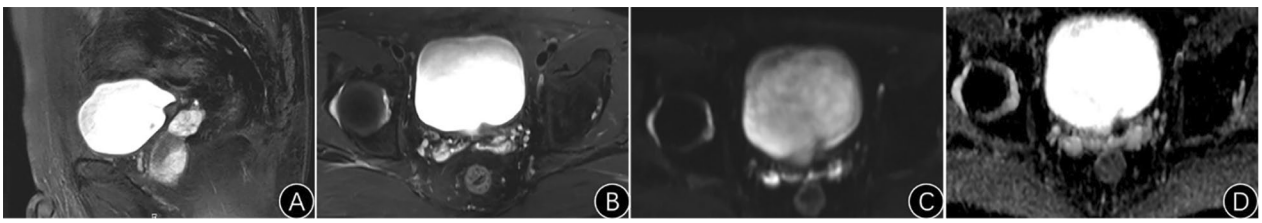

Fig. 1 **A**, T2-weighted imaging demonstrating complete separation of the mass at the end of the ureter from the prostate; **B**, T2-weighted imaging revealing a slightly low signal from the lesion; **C**, diffusion weighted imaging showing a slightly high signal from the lesion; **D**, apparent diffusion coefficient indicating a low signal from the lesion

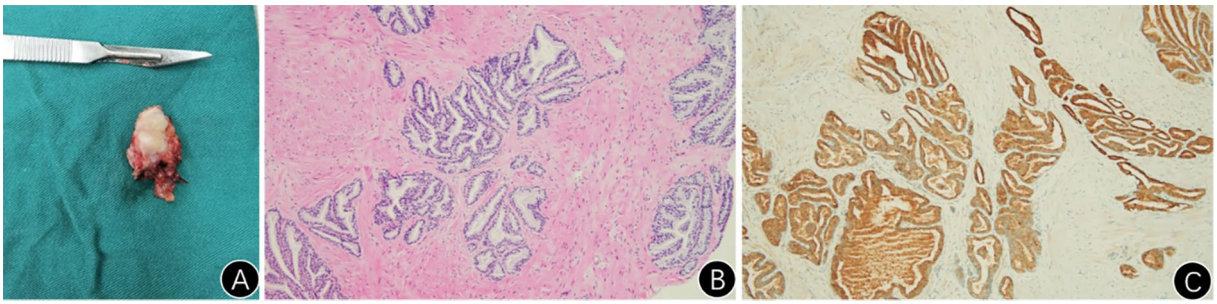

Fig. 2 **A**, Gross specimen postresection of the ureteral mass; **B**, microscopic view of pathological sections (hematoxylin and eosin, 40×); **C**, immunohistochemical staining results showing prostate-specific antigen positivity (hematoxylin and eosin, 100×)

retrovesical space, and sigmoid colon [6–9]. Additionally, EPT can also manifest in the female urogenital tract, affecting areas such as the cervix, vagina, and ovary [10–12]. A comprehensive search of literature databases such as PubMed and China National Knowledge Infrastructure revealed that most EPT cases are situated in the genitourinary tract, particularly in the bladder, presenting symptoms such as gross hematuria or dysuria. Cases of EPT outside the genitourinary tract exhibit varying clinical presentations depending on the specific location. Diagnosis of EPT is typically achieved through biopsy or surgical resection, with some cases incidentally discovered during autopsy. Pathologically, most instances of EPT display characteristics of ectopic prostate tissue, although rare cases may involve the development of prostate cancer within the ectopic tissue (Table 1).

The exact origin of EPT remains uncertain, with three prevailing theories explaining this phenomenon: the persistence of embryonic remnants, metaplastic changes in response to chronic inflammation, and migration and dislocation of normal tissues [2]. The potential etiology of EPT largely depends on its anatomical location, such as in the prostatic part of the urethra and the trigone of the bladder, where it may signify remnants of embryonic development. In other locations, EPT may result from metaplasia and abnormal embryogenesis due to "migration" or "misplacement" of prostate tissue. Gardner *et al.* suggest that prostate tissue could appear in the trigone area and the urethral prostate during embryonic development, indicating that these lesions are likely embryonic remnants rather than true ectopia [35]. Halat *et al.* also refer to lesions in the prostatic part of the urethra as prostatic urethral polyps rather than true ectopic tissue [3].

Ectopic prostate can lead to a variety of clinical symptoms depending on the location and size of the lesion, with some patients being asymptomatic. Lesions in the bladder and urethra commonly present with hematuria, while lower urinary tract obstruction can cause difficulties in urination or urinary retention. Irritation of the urinary tract may result in symptoms such as increased urinary frequency, urgency, and nocturia. Lesions outside the genitourinary tract may manifest as abdominal pain, distension, intestinal obstruction, masses, or bleeding [7, 25, 27, 29]. EPT is exceedingly rare in the female urogenital tract and is primarily found in the cervix, with occasional cases in the vagina or ovaries. These cases are often asymptomatic but can sometimes present as cervical masses or vaginal polyps, typically discovered incidentally during microscopic examination of cervical tissue removed for various reasons [36, 37]. In this case, the patient was found to have a mass in the left ureter during a health examination, with no significant clinical features present. However, upon dissection of the resected tissue, we observed that the lumen of the terminal ureter was narrowed. This finding suggests that the diseased tissue may have compromised the patency of the left ureter; however, it had not yet resulted in hydronephrosis or other complications. This situation may be attributed to the short duration between tumor detection and the initiation of our timely and active treatment.

Preoperative diagnosis of EPT is challenging owing to nonspecific clinical manifestations and imaging findings, necessitating histopathological examination for confirmation. MRI has been highlighted in literature as a potential tool for preoperative diagnosis of EPT. For instance, Hanna *et al.* presented a case of perineal ectopic prostate on MRI, demonstrating a distinct solid lesion with clear borders separate from the prostate, later confirmed as prostatic hyperplasia tissue via biopsy [30]. Another study by Sun *et al.* detailed the detection of EPT in the bladder trigone area using multiparametric MRI, showing signal characteristics consistent with all magnetic resonance (Magnetic Resonance, MR) sequences in a patient with a bladder mass and gross hematuria. When the signal characteristics resemble those of the normal prostate without diffusion restriction, EPT should be strongly suspected. Research has shown that multiparametric MRI can offer valuable imaging data

Table 1 Incidence sites and clinical characteristics of ectopic prostate tissue

Incidence site	Gender	Age	Source documents	Size (cm)	PSA (ng/ml)	Clinical symptoms	Discovery pathways	Pathological diagnosis
Genitourinary								
Anterior bladder wall [13]	M	59	Huang *et al*., 2023	1.0 × 1.0	N/A	Asymptomatic	TUR	EPT
Trigone of bladder [2]	M	37	Sun *et al*., 2023	1.6 × 1.8 1.4 × 1.5	Normal	Hematuria	TUR	EPT
Posterior bladder wall [14]	M	18	Gupta *et al*.,2022	1.0 × 0.9	Normal	Abdominalgia,dysuria	TUR	EPT
Bladder neck [15]	M	44	Zhou *et al*., 2020	2.0 × 2.0	N/A	Palpitation during urination	TUR	EPPC
Internal urethral orifice [16]	M	15	Zhu *et al*., 2019	1.4 × 1.3	N/A	Dysuria	TUR	EPT
Bladder dome [17]	M	68	Munde *et al*., 2017	3.0 × 2.0	N/A	Hematuria, abdominalgia	Autopsy	EPT
Ureteral [5]	M	60	Vincenzo *et al*., 2016	1.7	N/A	Abdominalgia, fever, nausea	Biopsy	EPT
Epididymis [18]	M	64	Lee *et al*., 2004	N/A	N/A	Undescended testicles	SS	EPT
Testis [19]	M	59	Milburn *et al*., 1994	0.5	Normal	Asymptomatic	SS	EPT
Seminal vesicle [4]	M	80	Somwaru *et al*., 2016	1.5 × 1.2	13.1	PSA elevation	Biopsy	EPT
Bulb of urethra [20]	M	60	Heyderman *et al*., 1987	N/A	N/A	LUTS	Biopsy	EPT
Penile urethra [21]	M	16	Congleton *et al*., 1989	N/A	N/A	Hematuria	Biopsy	EPT
Extragenitourinary								
Sigmoid colon [9]	M	33	Zhang *et al*., 2023	4.6 × 3.2	N/A	Abdominalgia	ESR	EPT
Pelvic cavity [22]	M	60	Jeong *et al*., 2022	14 × 10/3.3 × 2.5	12.18	Asymptomatic	PMRS	EPAC/EPT
Retrovesical pace [23]	M	73	Tan *et al*., 2012	8.5 × 8.0	24.66	Dysuria	Tumorectomy	EPT
Paracolic gutter [24]	M	73	Ahn *et al*., 2020	1.7	N/A	Asymptomatic	Tumorectomy	EPT
Small intestine [7]	M	79	Franchini *et al*., 2018	N/A	11.4	Abdominalgia, abdominal distension	SIR	EPT
Rectum [25]	M	81	Dai *et al*., 2013	1.0	2.38	Rectal bleeding	COR	EPT
Spine [26]	M	57	Kalsi *et al*., 2011	N/A	N/A	Nocturia, urinary incontinence	Tumorectomy	EPT
Processus vaginalis [27]	M	11	VanBeek *et al*., 2005	N/A	N/A	Scrotal mass	Tumorectomy	EPT
Anal canal [28]	M	59	Tekin *et al*., 2002	2.5	N/A	Rectal bleeding	Tumorectomy	EPT
Presacral [29]	M	78	Fulton *et al*., 2001	2.5 × 2.3	0.8	Intestinal obstruction	Tumorectomy	EPT
Perineum [30]	M	72	Hanna *et al*., 2020	N/A	7.0	Urinary retention, PSA elevation	biopsy	EPT
Ureterosigmoid anastomosis [31]	M	64	Martland *et al*., 2007	2.0	N/A	Subacute ureteral obstruction	SS	EPT
Spleen [6]	M	49	Vogel *et al*., 1996	N/A	N/A	Asymptomatic	Autopsy	EPT
Female								
Cervix [10]	F	34	Tschaidse *et al*., 2022	N/A	0.013	Hyperandrogenism	CC	EPT
Vaginal [11]	F	75	Roy *et al*., 2021	N/A	N/A	Polyps or nodules	Polypectomy	EPT
Ovary [12]	F	20	Sahin *et al*., 2016	4.0 × 3.5	N/A	Abdominalgia	Tumorectomy	EPT

Table 1 (continued)

Incidence site	Gender	Age	Source documents	Size (cm)	PSA (ng/ml)	Clinical symptoms	Discovery pathways	Pathological diagnosis
Adenocarcinoma								
Retrovesical space [32]	M	68	Tolkach *et al*., 2017	4.0	2.48	Asymptomatic	Tumorectomy	EPAC
Bladder dome [33]	M	70	Backhouse *et al*., 2014	N/A	5.67	Asymptomatic	biopsy	EPAC
Pelvic cavity [34]	M	71	Lu *et al*., 2019	12.0 × 10.7	44.48	Abdominal mass, urinary retention	Tumorectomy	EPAC

M male, *F* female, *TUR* transurethral resection, *LUTS* lower urinary tract symptoms, *PSA* prostate-specific antigen, *SS* surgical specimen, *ESR* endoscopic resection, *PMRS* pelvic mass resection surgery, *SIR* small intestine resection, *COR* colonoscopic resection, *CC* cervical conization, *EPT* ectopic prostate tissue, *EPPC* ectopic prostate pheochromocytoma, *EPAC* ectopic prostate adenocarcinoma

for enhancing the preoperative diagnosis of EPT [2]. Diagnosing ectopic prostate tissue located outside the genitourinary tract before surgery is notably challenging, often requiring postoperative pathological confirmation or incidental discovery during surgery. While imaging modalities such as CT, MRI, ultrasound, and endoscopy can detect larger lesions, they may not effectively identify smaller lesions. Histologically, the presence of prostate acini and stroma with a double-cell layer structure of columnar epithelial cells and basal cells are key features for distinguishing EPT. Additionally, the presence of amyloid bodies can aid in differentiation. Halat [3] *et al*. conducted a study on the histopathological characteristics of ectopic prostate tissue, finding that EPT closely resembles normal prostate tissue in both histology and immunohistochemistry. The authors recommend utilizing PSA and prostate-specific acid phosphatase (PSAP) as initial markers, given their specificity and sensitivity to prostate tissue. In cases where both markers are positive, it is advised to supplement with additional prostate tissue markers, with prostein (Prostate-Specific Protein, P501S) being the preferred option. Notably, P501S has only been identified in male prostate tissue to date. In instances where the initial staining results are inconclusive, the authors suggest using basal cell markers such as P63 and high molecular weight cytokeratin (HMWCK, 34βE12) or a PIN4 mixture (P504S, P63, and HMWCK) to confirm the presence of basal cells and secretory epithelial cells [25]. In our case description, the lesion occurred at the ureteral orifice and exhibited no particularly relevant clinical manifestations, resembling the incidental findings reported in most literature. The patient did not present any clinical symptoms, which may be attributed to the short medical history and our proactive management strategy. Prior to surgery, we conducted thorough examinations for highly suspected ureteral tumors and developed a comprehensive surgical plan. After engaging in active communication with the patient and obtaining informed consent for the procedure, we decided to perform intraoperative ureteral exploration and excise the affected tissue. During the operation, upon incising the diseased tissue, it was observed that the ureteral lumen had narrowed, with the potential for hydronephrosis if left unaddressed. Preoperative imaging indicated that the lesion was situated beneath the mucosa of the ureteral orifice, and no suspicious diseased tissue was identified during cystoscopy, making cystoscopic removal unlikely. The laparoscopic approach we adopted offers the benefit of the minimally invasive resection of the lesion, allowing for immediate ureterovesical reimplantation once the pathological nature is established during the procedure, thereby mitigating the need for a second operation. Although this approach may still result in a prolonged postoperative recovery time for the patient, this case provides valuable insights for addressing similar situations in the future.

EPT found in the bladder often presents symptoms that mimic those of bladder tumors, necessitating a clear differentiation between EPT and bladder cancer. Endoscopically, EPT typically appears as a firm, smooth, dome-shaped tumor with a wide base and intact mucosa, distinguishing it from urothelial tumors. Magnetic resonance imaging (MRI) is also a valuable tool for identification, as the signal intensity of EPT on MRI sequences mirrors that of normal prostate tissue. Unlike bladder cancer, EPT does not exhibit diffusion restriction on MRI and lacks extramural invasion, making it useful for ruling out malignant tumors and inflammatory conditions that typically show diffusion restriction. EPT occurring in the intestine may present with symptoms such as intestinal obstruction or rectal bleeding, necessitating differentiation from digestive tract tumors, inflammatory bowel disease, and metastatic tumors [1, 7, 25]. While most cases of EPT present as benign prostate tissue, there have been reported instances of malignant transformation. For instance, Gardner [35] *et al*. documented a case

of an elderly patient with persistent hematuria who was diagnosed with low-grade prostate adenocarcinoma after ectopic prostate tissue was discovered in the bladder dome during radical cystoprostatectomy for suspected bladder cancer. Tolkach [32] *et al*. discovered a 4-cm nodule located behind the bladder. The patient's preoperative PSA examination and digital rectal examination revealed no abnormalities. Following surgical resection, the pathology confirmed the presence of prostate cancer in the ectopic prostate. It is worth noting that benign ectopic prostate tissue found in the bladder, seminal vesicles, or nearby skeletal muscle tissue could be mistaken for tumor invasion in the orthotopic prostate of patients with prostate cancer. The use of alpha-methylacyl-CoA racemase and basal cell-specific immunostaining can aid in achieving an accurate diagnosis.

Conclusion

This case report describes a rare submucosal mass located in the distal ureter of the bladder. The patient's diseased tissue was identified during a routine health check-up, and the condition did not resolve with observation alone. To further investigate the nature of the lesion, the patient requested hospitalization for comprehensive examination. A thorough medical history revealed no significant past medical issues and no clinical symptoms. Imaging evaluations indicated a high likelihood of a ureteral tumor. Following the patient's informed consent, laparoscopic surgery was performed, and pathological analysis confirmed the presence of ectopic prostate tissue. Despite the clinical rarity of EPT, surgical resection of lesions through diverse approaches is advised once EPT is considered. Short-term postoperative follow-up has shown no recurrence, but the long-term efficacy remains uncertain [38].

Abbreviations

EPT	Ectopic prostate tissue
TPSA	Total prostate-specific antigen
CTU	Computed tomography urography
MRI	Magnetic resonance imaging
ECT	Emission computed tomography
CT	Computed tomography
PSAP	Prostate-specific acid phosphatase

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Author contributions

SC: conceptualization, data curation, investigation, visualization, and writing—original draft. ML: conceptualization, investigation, and writing—original draft. ZZ: funding acquisition, investigation, project administration, supervision, visualization, and writing—review and editing.

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Availability of data and materials

The original contributions presented in the study are included in the article/supplementary material. Further inquiries can be directed to the corresponding author.

Declarations

Ethics approval and consent to participate

The studies involving humans were approved by the ethics committee of the Huaihe Hospital of Henan University. The studies were conducted in accordance with local legislation and institutional requirements. The participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests

The authors declare that they have no competing interests.

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