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Epidermoid Cyst of the Ureter Mimicking Recurrent Urothelial Carcinoma: A Case Report and Brief Literature Review

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

ADEF 1 Omar M. Halalsheh
ABEF 2 Atef F. Hulliel
BCD 1 Abdallah Abualasal
BDE 3 Marwan I. Aldeqes
AEF 1 Ahmad Obeid
ABF 4 Mohammad Sharayah
ADE 3 Sohaib Al-Khatib

1 Department of General Surgery and Urology, Faculty of Medicine, Jordan University of Science and Technology, Irbid, Jordan
2 Faculty of Medicine, Jordan University of Science and Technology, Irbid, Jordan
3 Department of Pathology, Faculty of Medicine, Jordan University of Science and Technology, Irbid, Jordan
4 Division of Urology, Department of Special Surgery, School of Medicine, University of Jordan, Amman, Jordan

Corresponding Author: Omar M. Halalsheh, Department of General Surgery and Urology, Faculty of Medicine, Jordan University of Science and Technology, P. O. Box: 3030, Irbid-22110, Jordan, Fax: +962 2 7201064, e-mail: omarhalalsheh@yahoo.com

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Patient: Male, 79-year-old
Final Diagnosis: Epidermoid cyst
Symptoms: Routine follow-up
Clinical Procedure: —
Specialty: Urology

Objective: Rare disease

Background: Epidermoid cysts are benign, slow-growing lesions lined by keratinizing stratified squamous epithelium. Although they are among the most common cutaneous cysts, their occurrence within the urinary tract is exceptionally rare. In patients under oncologic surveillance, a new ureteral filling defect presents a significant diagnostic challenge, as benign lesions may closely mimic tumor recurrence and potentially lead to overtreatment or radical surgery.

Case Report: We report the case of a 79-year-old man with a history of high-grade papillary urothelial carcinoma treated with transurethral resection of bladder tumor (TURBT), intravesical Bacillus Calmette-Guérin (BCG), chemotherapy, and immunotherapy. He remained in remission until a CT scan in 2026 showed a proximal right ureteral filling defect suspicious for recurrence. Given the high-risk oncologic context, malignancy was strongly suspected. Flexible ureteroscopy with complete endoscopic excision was performed. Histopathology demonstrated a keratinizing squamous-lined cyst with laminated keratin and no atypia. Immunohistochemistry confirmed a benign epidermoid cyst of urothelial origin, with no evidence of malignancy.

Conclusions: Ureteral epidermoid cysts are exceedingly rare benign lesions that can closely mimic urothelial carcinoma on imaging, particularly in patients with prior malignancy. This case highlights that imaging findings alone are insufficient for diagnosis in this context, and that definitive histopathological and immunohistochemical evaluation is essential. Accurate tissue diagnosis enabled minimally invasive management and avoided unnecessary radical surgical intervention. This diagnostic dilemma underscores the importance of considering benign etiologies in ureteral filling defects during oncologic surveillance.

Keywords: Case Reports • Dermoid Cyst • Ureter

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Introduction

Epidermoid cysts, also known as epidermal inclusion cysts, are benign slow-growing lesions that are lined by keratinizing stratified squamous epithelium with a well-developed granular layer, producing laminated keratin within the cyst lumen [1]. They are one of the most common cutaneous cysts and are typically found on the head, neck, trunk, and extremities [2]. However, their occurrence in the genitourinary tract is remarkably rare, with the majority of reported cases involving the testis [3]. Epidermoid cysts arising in the upper urinary tract, specifically in the kidney and ureter, are exceedingly uncommon, and only a handful of cases have been documented in the literature [4,5].

The pathogenesis of epidermoid cysts within the urinary tract remains controversial. Several hypotheses have been proposed, including embryonic entrapment of ectodermal remnants derived from the Wolffian duct, squamous metaplasia of the urothelium secondary to chronic irritation, and metaplastic change induced by vitamin A deficiency [6]. Regardless of the mechanism, these lesions are clinically significant because they can produce obstructive symptoms, may be misdiagnosed as malignant neoplasms on imaging, and have occasionally led to unnecessary radical surgical procedures such as nephrectomy or nephroureterectomy [4,7].

We present a unique case of an epidermal inclusion cyst discovered in the proximal right ureter of a 79-year-old man who had a known prior diagnosis of high-grade urothelial cell carcinoma. This report shows that ureteral epidermoid cysts, although exceedingly rare, can radiologically mimic recurrent urothelial carcinoma in patients with a prior malignancy, and that definitive diagnosis can only be established through endoscopic excision with histopathological and immunohistochemical evaluation, thereby potentially preventing unnecessary radical surgery. The diagnostic workup, histopathological findings, and immunohistochemical profile are discussed.

Case Report

A 79-year-old married man, an ex-heavy smoker with a past medical history of hypothyroidism and dyslipidemia, presented to the oncology outpatient clinic for routine follow-up. His oncological history was significant for high-grade papillary urothelial carcinoma with extensive spindle cell morphology, initially diagnosed in 2015 (pathologic stage pT1). At that time, histopathology from a transurethral resection of the bladder tumor (TURBT) revealed extensive lamina propria invasion, tumor necrosis, and a spindle cell component in 20% to 30% of the tumor with myxoid changes. Given the high-grade nature of the tumor and the presence of sarcomatoid differentiation, a repeat TURBT was performed for restaging and assessment of residual disease. During subsequent surveillance, he underwent additional TURBT procedures for management of recurrent bladder lesions, with the last procedure performed in 2017. His treatment regimen included 6 cycles of intravesical Bacillus Calmette-Guérin (BCG) therapy, 4 cycles of systemic chemotherapy (gemcitabine and cisplatin), and immunotherapy with pembrolizumab, which he received until 2018. Following treatment with pembrolizumab, the patient achieved partial remission, which subsequently remained stable on serial surveillance imaging and clinical follow-up over the ensuing years. The patient remained in clinical remission until 2026, when he reported 2 episodes of gross hematuria approximately 2 months before the February 2026 follow-up visit. He also had urinary frequency and nocturia, but denied current hematuria, fever, chills, nausea, or vomiting. On urine cytology performed on August 4, 2025, results were negative for high-grade urothelial carcinoma. A subsequent cytology on January 29, 2026 revealed atypical cells with a high nuclear-to-cytoplasmic ratio and hyperchromatic nuclei, raising concern for possible urothelial pathology.

As part of the preoperative assessment, laboratory investigations were performed. Complete blood count showed a white blood cell count of $5.60 \times 10^3/\mu L$, red blood cell count of $4.86 \times 10^6/mm^3$, hemoglobin of 13.9 g/dL, hematocrit of 41.9%, mean corpuscular volume of 86.2 fL, and platelet count of $170 \times 10^3/\mu L$. Renal function testing demonstrated preserved kidney function with a serum creatinine level of 107 µmol/L, urea of 7.6 mmol/L, sodium of 138 mmol/L, and potassium of 4.46 mmol/L. Coagulation studies revealed a partial thromboplastin time of 22.8 seconds. Urinalysis showed red-colored, turbid urine with a specific gravity of 1.003 and proteinuria (2+). Urine glucose was negative, white blood cells were 2 to 4 per high-power field, and numerous red blood cells were detected. Urine culture demonstrated no bacterial growth. Overall, the laboratory findings were consistent with hematuria without evidence of active urinary tract infection or significant hematologic, renal, or coagulation abnormalities. A follow-up computed tomography (CT) scan of the chest, abdomen, and pelvis with intravenous and oral contrast was performed in February 2026. CT imaging demonstrated a 0.6 × 0.5 cm endoluminal soft tissue density lesion along the posterior wall of the proximal right ureter. The lesion demonstrated mild enhancement on the portal venous phase and was visualized as a persistent filling defect on delayed excretory phase images (**Figure 1A-1C**). The lesion was well-defined without associated aggressive periureteric invasion. Notably, a tiny endophytic soft tissue density nodule was identified as a filling defect on delayed excretory phase images. This thickening appeared more prominent compared to a previous scan from April 2025, which had shown a stable eccentric lesion with a focus of calcification in the same region. The scan also demonstrated mild bilateral hydroureteronephrosis, with the

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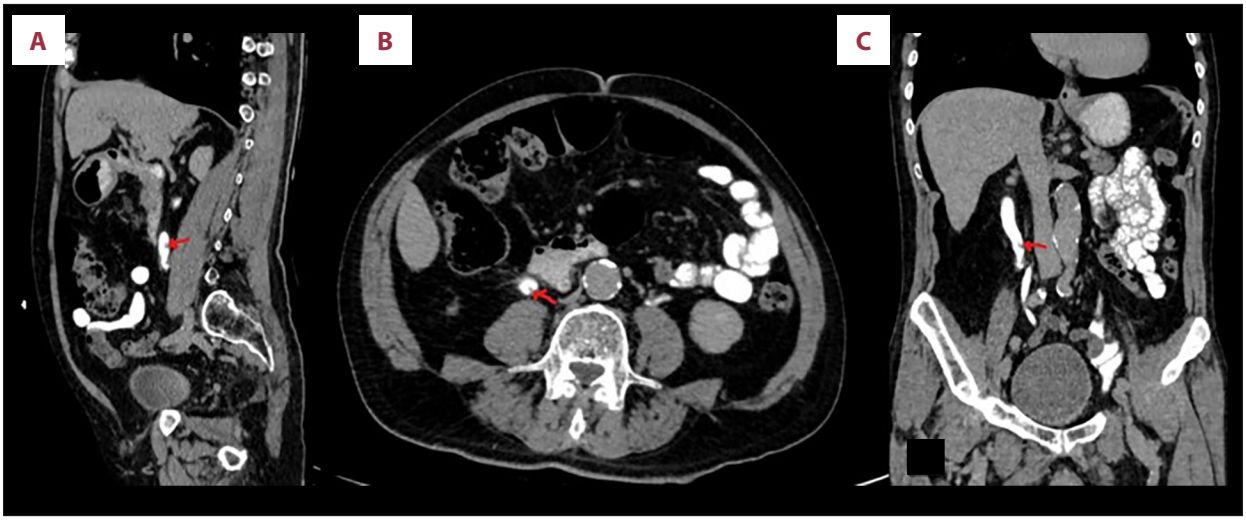

Figure 1. Contrast-enhanced computed tomography (CT) images demonstrating an endophytic filling-defect lesion (red arrows). (**A**) Sagittal view showing the lesion along the proximal ureter. (**B**) Axial view showing the intraluminal filling defect. (**C**) Coronal reconstruction confirming the location and extent of the endophytic lesion.

anteroposterior diameter of the right renal pelvis measuring 1.3 cm and the left measuring 1.0 cm.

On diffusion-weighted and delayed imaging sequences, no definite diffusion restriction or progressive delayed enhancement was identified. The radiological differential diagnosis included a slipped ureteral calculus given the patient's history of nephrolithiasis, a blood clot, or upper tract urothelial carcinoma recurrence (noting that upper tract involvement occurs in approximately 2% to 7% of patients with prior bladder urothelial carcinoma). Less likely considerations included a fibroepithelial polyp or keratinizing desquamative squamous metaplasia secondary to chronic irritation.

Given the patient's extensive history of high-grade urothelial carcinoma and the concerning new filling defect in the right ureter, a diagnostic and therapeutic surgical intervention was planned. In March, 2026, the patient underwent cystoscopy and flexible ureteroscopy under general anesthesia. Initial cystoscopy using a 22 Fr scope revealed an enlarged "kissing" prostate with a prominent median lobe, but the bladder mucosa was normal with no evidence of recurrent lesions. Ureteroscopic examination demonstrated a small, pedunculated, cauliflower-like lesion resembling papillary urothelial carcinoma; however, it lacked the typical friable appearance, white keratinized debris, or sessile invasive morphology. A 6 Fr flexible ureteroscope was then advanced through the right ureteric orifice under fluoroscopic guidance. The procedure successfully identified the lesion in the right proximal ureter. The lesion was biopsied and completely removed using a Dormia basket. Following the removal, a ureteral stent was placed over a guidewire to ensure ureteral patency. The surgical specimen, labeled as a right ureter lesion, was received in formalin and consisted of multiple pieces of white, soft tissue measuring 0.5 × 0.2 × 0.2 cm in aggregate. The fragmented nature of the specimen precluded reliable histologic assessment of surgical margins; therefore, complete excision could not be confirmed microscopically.

Microscopic examination of the tissue revealed a cystic structure predominantly lined by stratified squamous epithelium. This epithelium exhibited a well-developed granular layer and contained abundant laminated keratin within the cyst lumen (**Figure 2A, 2B**). Focally, areas of adjacent, normal appearing urothelial lining were also identified. There was no evidence of cytologic atypia, dysplasia, or invasive malignancy observed in the examined sections. The lesion showed the characteristic features of an epidermal inclusion cyst, consisting of a cyst lined by keratinizing stratified squamous epithelium with a granular layer and containing laminated keratin debris. The cyst wall was fibrous and did not contain adnexal structures. No significant chronic inflammatory infiltrate, calcification, foreign-body giant cell reaction, or keratin granulomatous response was identified.

To definitively rule out a recurrence of the patient's known sarcomatoid urothelial carcinoma and to confirm the nature of the epithelial lining, a panel of immunohistochemical (IHC) stains was performed. Immunohistochemistry was undertaken due to the patient's prior history of both high-grade urothelial carcinoma and suspected prostatic disease, necessitating exclusion of recurrent or metastatic malignancy. The epithelial lining of the cyst was negative for CK20 (**Figure 2C**) and NKX3.1 (**Figure 2D**), the latter effectively ruling out a prostatic origin for the tissue. Staining for p53 demonstrated a wild-type pattern, characterized by scattered, weak positivity, which differs from the diffuse overexpression or complete absence typically seen in high-grade malignancies (**Figure 2E**). Notably,

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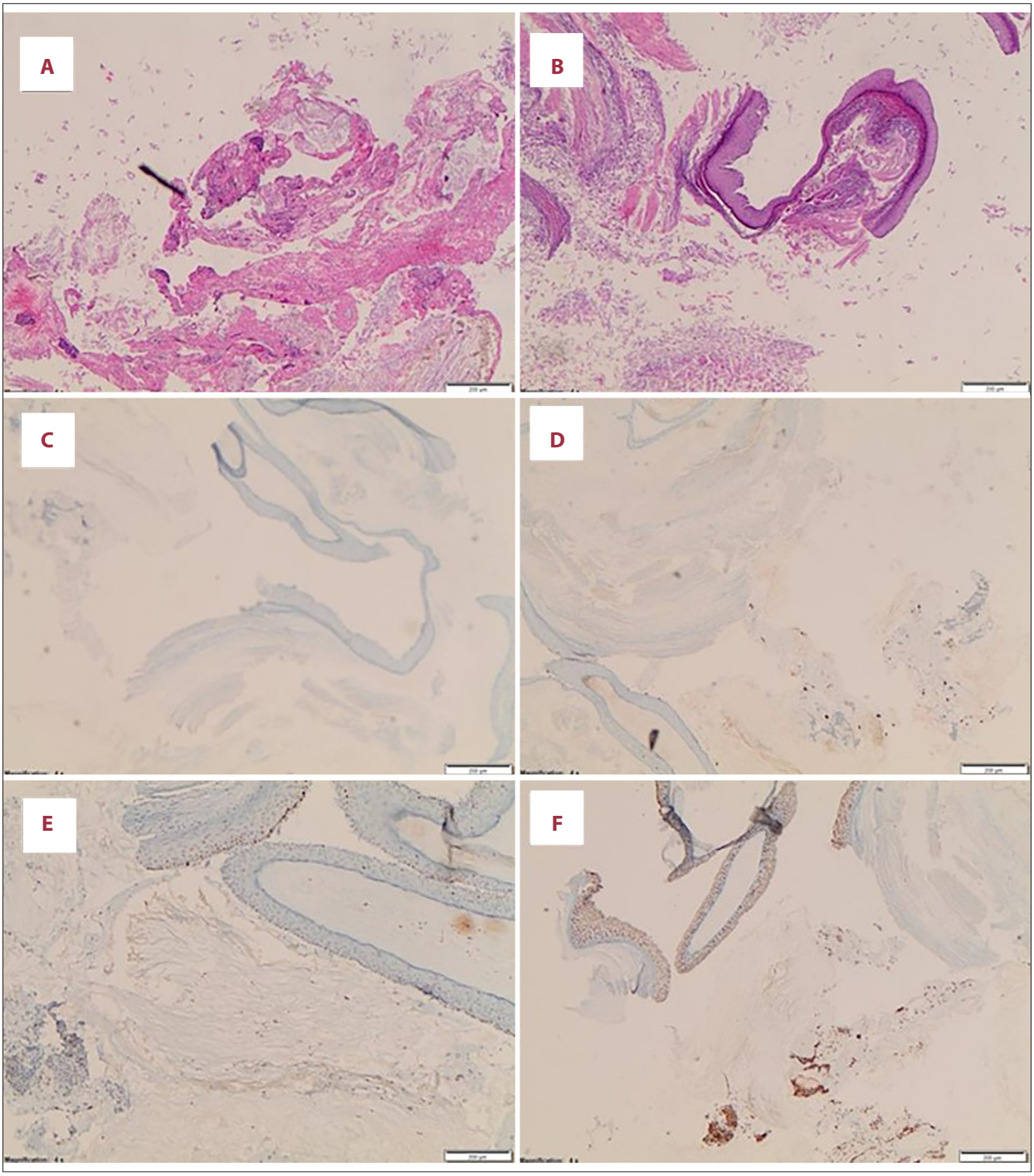

Figure 2. Histopathological and immunohistochemical evaluation of the ureteral cyst. (**A, B**) H&E 4 × sections showing a cystic lesion lined by stratified squamous epithelium with a well-formed granular layer and abundant laminated keratin within the lumen, with focal adjacent normal urothelial lining and no evidence of dysplasia or malignancy. (**C**) CK20 immunostain negative in the cyst epithelium 4 ×. (**D**) NKX3.1 immunostain negative 4 × (**E**) p53 immunostaining demonstrating a wild-type (scattered weak) pattern 4 ×. (**F**) GATA3 immunostain highlighting native urothelium and showing strong positivity in the squamous epithelial lining 4 ×.

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GATA3 immunostaining highlighted both the native urothelial epithelium and showed strong positivity within the squamous epithelial lining of the cyst (**Figure 2F**). However, GATA3 expression was interpreted with caution, as it is known to be expressed in both urothelial and squamous epithelium and therefore lacks specificity in this setting; its positivity alone could not be used to infer a urothelial origin. Based on these morphological and immunohistochemical findings, the final pathological diagnosis was an epidermoid cyst of the right ureter, with no evidence of malignancy.

The postoperative course was uneventful. The patient reported resolution of his lower urinary tract symptoms, and there was no recurrence of hematuria during early follow-up. The ureteral stent was removed approximately 2 weeks after the procedure without complications. He was subsequently placed on routine urologic and oncologic surveillance with planned imaging follow-up.

Discussion

Epidermoid cysts of the urinary tract are remarkably rare, and their occurrence in the ureter is an exceptionally uncommon clinical entity. The first reported case of an epidermoid cyst involving the ureter was described by Ishizaki et al in 2007, in a 72-year-old man [4]. Since then, only a small number of additional cases have been documented, including the report by Jing et al in 2022, describing a 48-year-old woman with an epidermoid cyst in the middle ureter that had been misdiagnosed as a ureteral calculus for 8 years [5].

The histogenesis of epidermoid cysts within the urinary tract remains a subject of debate. Several theories have been proposed to explain their development in a location normally lined by urothelium [8].

Squamous metaplasia of the urothelium is the most widely accepted explanation for epidermoid cysts of the upper urinary tract. Prolonged irritation from factors such as calculi, chronic infection, or other inflammatory stimuli may induce metaplastic transformation of transitional epithelium into keratinizing stratified squamous epithelium [6].

In the present case, the identification of focal urothelial lining adjacent to the squamous epithelium, together with GATA3 positivity in both epithelial components, supports a urothelial origin of the lesion. GATA3 is a transcription factor strongly expressed in normal urothelium and is considered a sensitive marker of urothelial differentiation [9]. Its expression in both components is compatible with a metaplastic process, in which the keratinizing squamous epithelium may have arisen from native urothelium rather than from embryonically entrapped ectodermal remnants.

In this patient, the long and complex urologic history likely provided a background for a metaplastic process to develop. Over more than a decade, he underwent repeated transurethral resections and multiple endoscopic procedures involving both the upper and lower urinary tract. In addition, he received 6 cycles of intravesical BCG, systemic gemcitabine–cisplatin chemotherapy, and pembrolizumab immunotherapy. He also had a history of nephrolithiasis, with a previously noted calcified focus in the same ureteral segment. Taken together, these factors represent repeated and sustained sources of local irritation and inflammation.

Such chronic stimuli are well recognized in the development of keratinizing squamous metaplasia of the urothelium, which can arise in response to long-standing mechanical, infectious, or inflammatory stress, including urinary tract stones, recurrent infections, indwelling instrumentation, and repeated surgical manipulation. In particular, intravesical BCG is known to induce a persistent granulomatous inflammatory reaction within the urothelium, further supporting this potential pathway [10].

The differential diagnosis of a keratin-filled cystic lesion in the ureter includes several entities. The primary concern in this patient was urothelial carcinoma with squamous differentiation, given the clinical history. However, the absence of cytologic atypia, the well-organized granular layer, and the benign immunohistochemical profile effectively excluded this possibility. Dermoid cysts may also contain keratinizing squamous epithelium but would be expected to show additional skin adnexal structures such as hair follicles and sebaceous glands, which were absent in this case [11]. Epidermoid cysts located in the upper urinary tract can produce an obstructive effect, leading to hydronephrosis and symptoms such as renal colic and hematuria [5]. Importantly, because current medical imaging modalities lack adequate specificity and sensitivity to accurately diagnose an epidermoid cyst in the ureter, these lesions are frequently misdiagnosed as urothelial tumors or ureteral calculi [5].

On CT scans, epidermoid cysts typically appear as hypodense, non-enhancing lesions, sometimes with peripheral calcification [12]. MRI may show inhomogeneous hypointense T1 and hyperintense T2 signal, with absent or minimal peripheral contrast enhancement [12]. However, given the narrow ureteral cavity and the typically small size of these lesions, CT diagnostic accuracy may be inferior to MRI.

Management options range from conservative observation to endoscopic excision and radical resection, depending on the clinical circumstances. Endoscopic approaches, including holmium laser excision via ureteroscopy, have been successfully employed with excellent outcomes and minimal morbidity [5]. In the present case, the biopsy-confirmed benign nature of the lesion spared the patient from more aggressive surgical intervention.

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Although epidermoid cysts are benign, long-term follow-up is recommended because published reports, primarily based on epidermoid cysts in sites other than the ureter, state that there is a small risk of malignant transformation to squamous cell carcinoma (approximately 2%) and a 3% chance of recurrence after surgery [13].

This case adds several incremental contributions to the limited literature on ureteral epidermoid cysts. First, to the best of our knowledge, this is among the very few reported cases occurring in a patient with a prior history of high-grade urothelial cell carcinoma under oncologic surveillance, a context in which radiologic findings strongly favor tumor recurrence and may bias clinical decision-making. Second, it demonstrates that such lesions can present as a ureteral filling defect mimicking malignant recurrence on imaging, highlighting a specific diagnostic pitfall in a high-risk oncologic population. Third, the case provides histopathological and immunohistochemical evidence (including GATA3 expression in both epithelial components) supporting a metaplastic urothelial origin, rather than embryonic inclusion, which adds diagnostic insight into the pathogenesis of these rare lesions. Finally, complete endoscopic excision allowed definitive diagnosis and organ preservation, reinforcing the role of minimally invasive management in avoiding unnecessary radical surgery.

From a practical standpoint, this case highlights that not all ureteral filling defects in patients with prior urothelial carcinoma represent recurrence, and that tissue diagnosis through endoscopic evaluation should be considered when feasible to avoid overtreatment.

Conclusions

Ureteral epidermoid cysts are rare benign lesions that can mimic urothelial malignancy on imaging, especially in patients with prior urothelial cancer. This case shows that imaging alone is insufficient for diagnosis, and histopathological and immunohistochemical evaluation is essential to confirm benignity. Here, accurate tissue diagnosis allowed minimally invasive endoscopic management, avoiding unnecessary radical surgery. However, this finding is based on a single case and applies to carefully selected patients where endoscopic biopsy is feasible. Given the rarity of ureteral epidermoid cysts and the patient's prior history of high-grade urothelial carcinoma, long-term surveillance is recommended to monitor for potential recurrence or delayed complications.

Department and Institution Where Work Was Done

King Abdullah University Hospital, a tertiary hospital fully affiliated with Jordan University of Science and Technology, Jordan.

Consent for Publication

Written informed consent was obtained from the patient for the publication of this case report and accompanying images. A copy of the written consent form is available for review by a journal. We confirmed that the privacy of the participants was preserved and that the data were anonymized and kept confidential.

Availability Data and Materials

Data supporting the findings of this study are available from the corresponding author upon reasonable request.

Declaration of Figures' Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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