

## Squamous cell carcinoma of the upper urinary tract revealed by severe infection in a duplicated ureter with Lithiasic ureterocele: A case report and literature review

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### Abstract

**Introduction:** Primary squamous cell carcinoma (SCC) of the upper urinary tract is a rare tumor representing less than 1% of renal neoplasms. It is strongly associated with chronic irritation, particularly related to urinary calculi and recurrent infections. Diagnosis is often delayed, due to a non-specific clinical presentation that may mimic severe infectious conditions. The association of ureteral duplication with a complicated ureterocele is extremely rare.

**Case Presentation:** We report the case of a 60-year-old patient with no significant medical history who presented with febrile acute left-sided pyelonephritis. Imaging revealed a duplicated left kidney with marked dilation of the upper pole collecting system secondary to a tissue thickening at the ureterovesical junction, suggestive of a complicated lithiasic ureterocele. A multiloculated retroperitoneal abscess extending along the psoas muscle was also identified. Initial management included percutaneous drainage and internal urinary diversion with placement of two double-J stents. Subsequently, a nephroureterectomy with bladder cuff excision was performed. Histopathological examination revealed a well-differentiated squamous cell carcinoma infiltrating the renal parenchyma (pT3NxMx), with vascular emboli and a positive ureteral margin. The clinical course was marked by the rapid development of necrotic mediastinal and retroperitoneal lymphadenopathy suggestive of metastatic progression.

**Conclusion:** This case highlights the aggressive nature of upper urinary tract squamous cell carcinoma and underscores the importance of considering a malignant etiology in any chronic obstruction complicated by severe infection, particularly in the presence of underlying anatomical abnormalities.

**Keywords:** Upper urinary tract squamous cell carcinoma; Ureteral duplication; Ureterocele; Urinary lithiasis; Pyelonephritis; Nephroureterectomy; Upper urinary tract tumor; Case report

### 1. Introduction

Primary squamous cell carcinoma of the upper urinary tract is a rare entity, representing approximately 0.5% to 1% of renal malignancies[1,2]. In contrast to urothelial carcinoma, it is almost invariably associated with chronic inflammation secondary to urinary lithiasis, prolonged obstruction, or recurrent infections[3,4].

Carcinogenesis is thought to result from progressive squamous metaplasia of the urothelium, which may evolve into dysplasia and eventually invasive carcinoma under persistent inflammatory stimulation.

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The clinical presentation is typically non-specific and often dominated by infectious or obstructive symptoms, leading to delayed diagnosis[5]. As a result, most patients are diagnosed at a locally advanced stage ( $\geq$  pT3), which contributes to the poor overall prognosis [5].

The association of ureteral duplication with a lithiasic ureterocele complicated by squamous cell carcinoma is exceptionally rare and has only been reported in isolated cases.

## 2. Observation

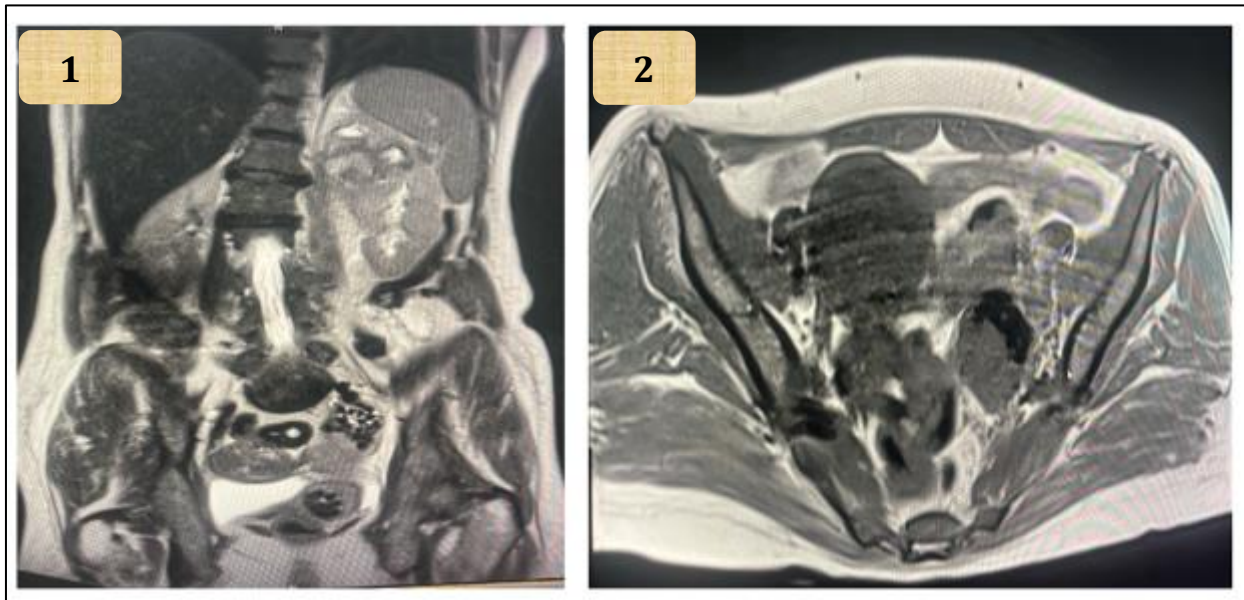
A 60-year-old female patient with no significant medical or surgical history presented to the emergency department with left-sided lower back pain that had been present for 5 days, associated with fever, chills, and a decline in general health.

The biological workup showed:

- Hb: 7.9 g/dL
- WBC: 14,000/mm<sup>3</sup>
- CRP: 257 mg/L
- Normal kidney function
- Normal blood ionogram

An emergency abdominal and pelvic MRI was performed and revealed:

- Left kidney composed of two renal segments, the upper one of which shows significant ureteropelvic-caliceal dilatation upstream of a probable degenerated ureterocele with tissue thickening extending back to the level of the pelvic ureter and multiple upstream stasis stones.
- Mild dilation of the lower nephron due to compression
- Left retroperitoneal and posterior perirenal collection in close contact with the upper nephron
- Renal hilar and external iliac lymphadenopathy measuring 24 × 20 mm



**Figures 1 and 2** Coronal (1) and axial (2) sections showing a degenerated ureterocele with calculi

Initial treatment combined double internal drainage using two double-J catheters, one in each renal pelvis, with percutaneous drainage of the collection via a nephrostomy tube, with clinical improvement.

A chest CT scan was performed as part of the staging evaluation and revealed:

- Right hilar pulmonary lymphadenopathy
- Presence of peripheral calcifications

- A few bilateral parenchymal micronodules (upper and lower lobes)

Given these findings, lymph node tuberculosis was suspected. BK tests of sputum samples were performed and came back negative. After the infection stabilized and her biological condition improved, a radical nephro-ureterectomy with bladder cuff was performed via open surgery, revealing a well-differentiated squamous cell carcinoma infiltrating the renal parenchyma with vascular emboli and invaded ureteral margins, stage pT3NxMx



**Figure 3** Surgical specimen from a nephroureterectomy showing the kidney, ureter, and degenerated ureterocele

The immediate postoperative course was simple.

A follow-up thoraco-abdominal-pelvic CT scan at 3 months revealed:

- Bilateral proximal pulmonary embolism
- The development of multiple bilateral mediastinal and hilar lymphadenopathies with extensive necrosis, the largest measuring 23 × 22 mm
- The appearance of necrotic celiomesenteric and lumbosacral lymph nodes, the largest measuring 15 mm in short axis.

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### 3. Discussion

#### 3.1. Epidemiological data and risk factors

Primary squamous cell carcinoma of the upper urinary tract is a rare tumor, accounting for less than 1% of malignant kidney tumors. It is distinguished from urothelial carcinoma by its strong association with chronic irritation of the urothelium.

Several series have shown that urinary stones are the primary risk factor for this tumor. In the classic series reported by Michael K Li [1], an association with urinary stones was found in more than 80% of cases. This observation has also been confirmed in more recent series.

Ramon Holmäng's [2] study of patients with squamous cell carcinoma of the upper urinary tract showed that the majority of patients had a history of urinary stones or chronic urinary tract infection. The authors also reported that most tumors were diagnosed at a locally advanced stage.

Similarly, the analysis conducted by Seung Hwan Lee [3] found a strong correlation between the presence of urinary stones and the development of squamous cell carcinomas of the upper urinary tract, suggesting a major role for chronic inflammation in carcinogenesis.

In our study, the presence of multiple stasis stones associated with obstruction due to a ureterocele likely constitutes the primary factor contributing to chronic irritation of the urothelium.

**Table 1** Main series published on squamous cell carcinoma of the upper urinary tract

Author	Year	Number of cases	Lithiasic association	Stage $\geq$ T3	5-year survival
Michael K Li	1987	22	82 %	68 %	0 %
Ramon Holmång	2007	65	76 %	71 %	<10%
Seung Hwan Lee	2014	29	79 %	65 %	11 %
David Berz[6]	2012 (SEER)	179	NR	majority advanced	median survival 10 months
Our observation	2025	1	yes	pT3	metastatic evolution

### 3.2. Pathophysiology

The carcinogenesis of squamous cell carcinoma of the upper urinary tract is based on a progressive process of adaptation of the urothelium to chronic irritation.

Under the influence of persistent inflammation associated with obstruction, urolithiasis, or recurrent urinary tract infections, the urothelium may undergo squamous metaplasia. This histological transformation constitutes an adaptive response that is initially benign but may progress to dysplasia and then to invasive squamous cell carcinoma[7,8].

This physiological mechanism explains the strong association between urinary stones and this type of tumor, an association widely described in published series.

### 3.3. Anatomical feature of our case

The combination of a squamous cell carcinoma of the upper urinary tract with ureteral duplication and a lithiasic ureterocele is extremely rare.

Congenital anomalies of the urinary tract can contribute to urinary stasis and recurrent infections, thereby creating an environment conducive to chronic inflammation of the urothelium.

In our case, the dilation of the upper renal pelvis secondary to the ureterocele likely led to prolonged urinary stasis and the formation of stones, creating a favorable environment for the development of a squamous cell carcinoma.

### 3.4. Diagnostic challenges

Preoperative diagnosis of squamous cell carcinoma of the upper urinary tract is often challenging. Clinical manifestations are generally nonspecific [9] and may be dominated by signs of infection or obstruction.

Radiologically, the images may show a combination of hydronephrosis, an infiltrating renal mass, and urinary stones. However, these signs are nonspecific and may be observed in other inflammatory conditions such as xanthogranulomatous pyelonephritis.

Therefore, in many cases, a definitive diagnosis is not established until after histopathological analysis of the nephrectomy specimen. [10]

### 3.5. Prognosis

The prognosis for squamous cell carcinoma of the upper urinary tract is generally poor. Most patients are diagnosed at stage  $\geq$  pT3, with a 5-year survival rate of less than 10% in the most recent studies. [5]

Studies by Ramon Holmång and Seung Hwan Lee have shown that overall survival remains limited, particularly in locally advanced or metastatic stages.

Our case also illustrates the aggressiveness of this tumor, with the rapid development of necrotic mediastinal and retroperitoneal lymphadenopathy suggestive of early metastatic progression despite surgical management.

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### 4. Conclusion

Squamous cell carcinoma of the upper urinary tract is a rare but particularly aggressive tumor, most often associated with chronic irritation of the urothelium secondary to urolithiasis or recurrent urinary tract infections. Diagnosis is frequently delayed due to nonspecific clinical presentation that may mimic a severe infectious disease.

Our case report highlights several features rarely described in the literature, notably the association of squamous cell carcinoma with ureteral duplication complicated by a lithiasic ureterocele, responsible for chronic urinary stasis.

This case highlights the importance of considering a neoplastic etiology in any chronic upper urinary tract obstruction associated with stones and recurrent infections, particularly in patients with anatomical abnormalities.

Early surgical intervention remains the standard of care, but the prognosis remains poor due to the often delayed diagnosis and the aggressive nature of this tumor.

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### Compliance with ethical standards

#### *Disclosure of conflict of interest*

No conflict of interest to be disclosed.

#### *Statement of informed consent*

Informed consent was obtained from all individual participants included in the study.

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