

Case report

## High Grade Urothelial Carcinoma in Neobladder Replacement: An Unusual Form of Recurrence 9 Years After Radical Cystoprostatectomy

Richepin Tidahy<sup>1,2</sup>, Ouima Justin Dieudonné Ziba<sup>2</sup>, Adrian Roman<sup>1</sup>, Malek Bargoud<sup>1</sup>, Moulay Hassan Farih<sup>2</sup>

<sup>1</sup>*Urology Department, Roanne Hospital, Auvergne-Rhône-Alpes, France*

<sup>2</sup>*Department of Urology, Andrology, Renal Transplantation, Hassan II University Hospital, Sidi Mohammed Ben Abdellah University, Fez, Morocco*

### HIGHLIGHTS

- Urothelial carcinoma in orthotopic neobladder is unusual.
- There are no specific guidelines for managing recurrent urothelial carcinoma.

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#### \*Corresponding Author:

Richepin Tidahy

Email: [tidahy15@gmail.com](mailto:tidahy15@gmail.com)

Address: Urology Department, Roanne Hospital, Auvergne-Rhône-Alpes, France.

### ABSTRACT

#### Introduction

The recurrence of urothelial carcinoma on a neobladder made from ileal is an infrequent entity. Radical cystectomy is the standard treatment for muscle-invasive bladder cancer.

#### Case presentation

We report here the case of a 68-year-old patient with a history of radical cystoprostatectomy with Hautman-technique of enterocystoplasty replacement 9 years ago, followed by adjuvant chemotherapy, for urothelial carcinoma pT2G3N1M0+CIS. The patient was followed up regularly and did not return for his control visit. He was admitted to the emergency room with acute urinary retention on clotting hematuria. A cystoscopic examination with a tumor biopsy was performed, and the anatomic-pathological result favoured a recurrence of a high-grade urothelial carcinoma infiltrating the intestinal mucosa. The extension workup by abdominopelvic CT showed a secondary location in the liver with a suspicious left latero-aortic lymph node mass. In agreement with a multidisciplinary consultation meeting, the patient was referred to oncology for chemotherapy.

#### Conclusions

Tumor recurrence should be evoked in patients with hematuria after replacement enterocystoplasty. Although urothelial carcinoma in orthotopic neobladder is unusual. However, in these rare situations, it is essential to quickly manage the patient in a reference center with multidisciplinary consultations to optimize adequate care.

**Keywords:** Urothelial Carcinoma; Neobladder; Abdominopelvic; Radical Cystoprostatectomy

#### Introduction

Radical cystectomy is the standard treatment for muscle-invasive bladder cancer (1), and orthotopic ileal neobladder replacement is the technique for continuous

urinary diversion after radical cystectomy (2). This shunt appears to be the most accepted because it is the most physiological while maintaining an intact body image without needing an appliance and maintaining micturition

by natural means (3). Recurrence of urothelial Carcinoma in the urethra has been widely reported (4), as well as in the upper urinary tract (5), or in the uretero-neobladder anastomosis (6, 7).

However, isolated recurrences at the level of ileal neobladder replacement remain exceptional, with only 4 other case reports in the literature (8). We report a recurrence of high-grade urothelial Carcinoma infiltrating the intestinal mucosa 9 years after ileal neobladder in a patient diagnosed with pT2G3N1M0 urothelial Carcinoma associated with Carcinoma in situ (CIS) who was treated by radical cystoprostatectomy with Hautman-enteroplasty replacement technique.

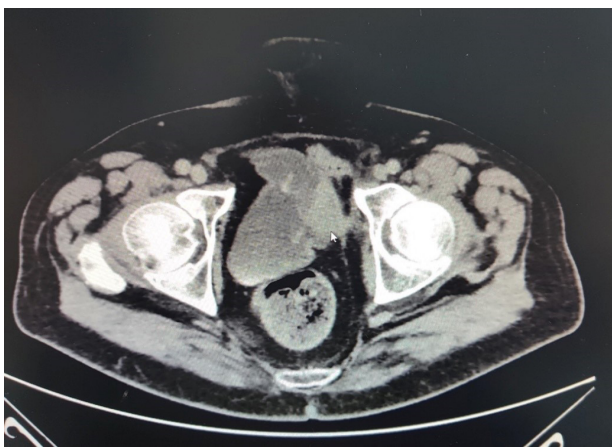
### Case presentation

This is a 68 years old patient admitted to the emergency room with acute urine retention due to clotting hematuria.

In this history, we note arterial hypertension, non-insulin-dependent diabetes, chronic smoking weaned for two months, radical cystoprostatectomy with enteroplasty replacement by Hautman technique in September 2009, followed by adjuvant chemotherapy for urothelial carcinoma pT2G3N1M0 associated with a CIS. The patient had regular follow-ups (abdominopelvic CT and urine cytology) until 2016 and then lost to follow-up. The last imaging in 2016 was within normal limits, except for a slight trend of atrophy of the left kidney. Two years later, nine years after radical cystoprostatectomy and neobladder replacement, the patient presented with macroscopic hematuria complicated by acute urinary retention motivating his emergency room visit.

Urgent bladder catheterization, declotting, and continuous irrigation were performed. The biological workup was routine except for anemia. Urine cytology was positive for high-grade urothelial carcinoma (according to the Paris 2016 system).

An abdominopelvic CT scan showed tissue thickening



**Figure 1.** Axial section CT scan showing tissue thickening on the left lateral part of the neovessel

on the left lateral part of the neobladder (Figure 1). A cystoscopic examination (Figure 2) objectifies a tumor localized at the left lateral side of the neobladder. No apparent abnormalities were observed in the urethra and urethro-neobladder anastomosis.

A transurethral resection (TUR) of the tumor was performed, and the anatomic-pathological result with the complementary immunohistochemical examination was in favor of a recurrence of a high-grade urothelial carcinoma infiltrating the intestinal mucosa (Figure 3).

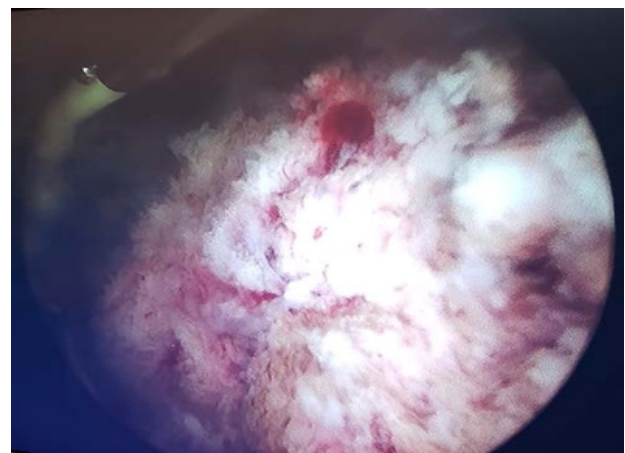
The extension workup by thoracic-abdominal-pelvic CT scan showed a secondary location in the liver with a suspicious left latero-aortic lymph node mass.

A biopsy of the liver lesion showed tumor infiltration of the liver parenchyma by a poorly differentiated urothelial carcinoma, whereas the biopsy of the latero-aortic lymph nodes showed no neoplastic lesion. In agreement with a multidisciplinary consultation meeting, the patient was referred to oncology for chemotherapy.

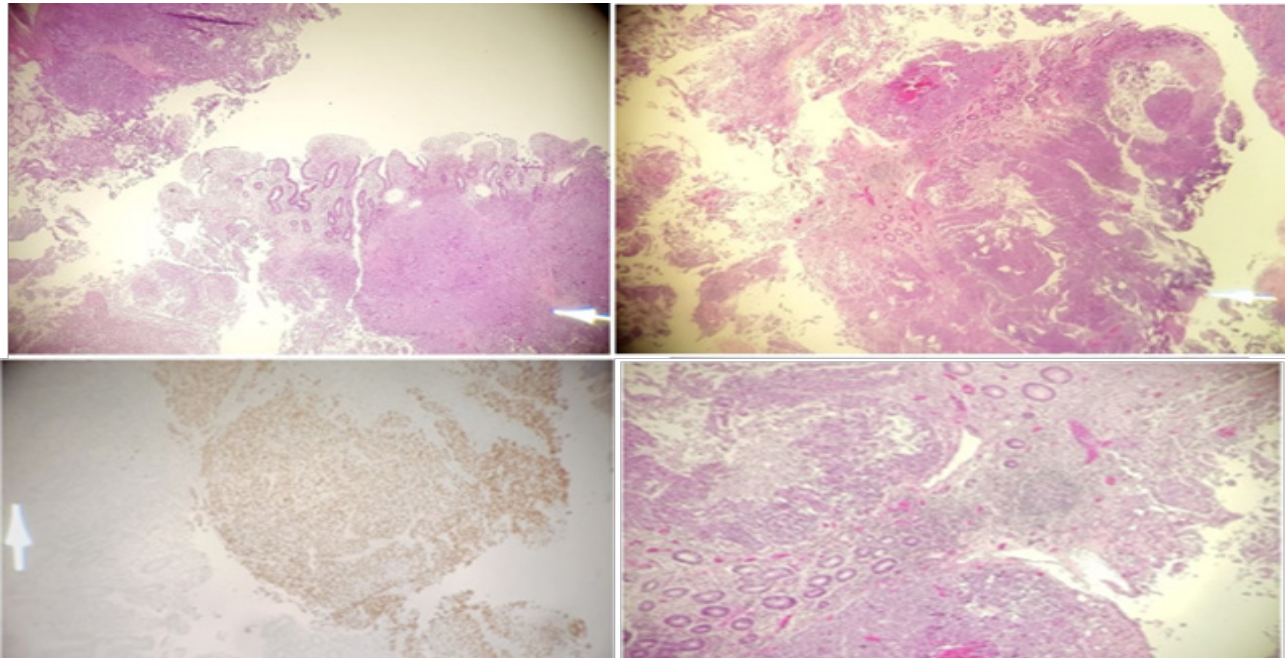
### Discussion

The risk of urethral recurrence of the urothelial tumor in patients who have undergone orthotopic neobladder has been a deterrent to performing neobladder in many patients (9). Several studies have reviewed the risk factors for urethral recurrence after orthotopic ileal neobladder following radical cystectomy (10, 11). These risk factors included tumor location, multifocality of carcinoma in situ, prostatic involvement, and positive urethral margin (4). By applying these risk factors to patient selection, the number of patients who could benefit from orthotopic neobladder after radical cystectomy was significantly reduced (12).

Compared to urethral recurrence, the recurrence of urothelial carcinoma on a neobladder made from the ileal is a rare situation (13). To our knowledge, only 4 case reports have been published in the literature, and the



**Figure 2.** Cystoscopic view of the tumor lesion in the left lateral wall of the neovessel



**Figure 3.** Pathological image of a recurrence of a high-grade urothelial carcinoma infiltrating the intestinal mucosa.

mean age of diagnosis was 60 years (8). In the previously reported cases, the circumstances of the discovery of the recurrence were the occurrence of macroscopic hematuria in the majority of cases and positive urine cytology in some cases (8). Our patient was admitted to the emergency room with acute urine retention clotting hematuria.

The recurrence time after neobladder varies from one to 11 years (13). The occurrence of carcinoma in situ in a colonic neobladder 8 years after radical cystectomy has also been reported (13). We report the occurrence of a recurrence of a high-grade urothelial carcinoma infiltrating the intestinal mucosa 9 years after ileal neobladder.

There have also been case reports of transitional cell recurrences in the ileal duct (7). Herawi et al., reported low-grade, non-invasive papillary carcinomas in the colonic mucosa and peritoneum after neobladder (14).

Some studies have concluded that urothelial carcinomas can disseminate and implant, even in non-urothelial mucosa (8). Hara et al., considered that recurrence of urothelial carcinoma in a segment of the intestine could occur by two mechanisms: direct invasion or implantation (15). In our case, the exact mechanism remains unknown, we can think that a direct invasion of the primary tumor is at the origin.

There are no specific guidelines for managing recurrent urothelial carcinoma (1). In most cases, the management of previously reported cases included transurethral resection (8). For some authors, surgical removal of the recurrence with curative intent is an option, neobladder may be replaced by another urinary diversion type continent or

non-continent (16). Some recurrences in carcinoma in situ (CIS) have been successfully treated with BCG instillations; chemotherapy, when administered, is done with palliative intent (17). In the case of our patient, because of the metastatic evolution of the recurrence and in agreement with a multidisciplinary consultation meeting, he was referred to oncology for chemotherapy.

### Conclusions

Recurrence of high-grade urothelial carcinoma infiltrating the intestinal mucosa on the neobladder is still a rare situation. The excellent selection of patients for this type of referral surgery and the increasing expertise of urologists in the technique is a considerable asset that limits the number of cases. Although urothelial carcinoma in orthotopic neobladder is unusual, tumor recurrence should be considered in patients with hematuria after enterocystoplasty replacement. However, in these rare situations, it is essential to quickly manage the patient in a reference center with multidisciplinary consultations to optimize adequate care.

### Authors' contributions

All authors contributed equally.

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**Conflict of interest**

The author declares that there is no conflict of interest.

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**Ethical statement**

Our case reports are based on CARE guidelines, and the patient's parent agreed to report his case after signing the written informed consent.

**Data availability**

Data will be provided on request.

**Abbreviations**

CIS Carcinoma in situ

TUR Transurethral resection

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**Author (s) biosketches**

**Tidahy R**, MD, Urology Department, Roanne Hospital, Auvergne-Rhône-Alpes, France and Department of Urology, Andrology, Renal Transplantation, Hassan II University Hospital, Sidi Mohammed Ben Abdellah University, Fez, Morocco.  
Email: [tidahy15@gmail.com](mailto:tidahy15@gmail.com)

**Justin Dieudonné Ziba O**, MD, Department of Urology, Andrology, Renal Transplantation, Hassan II University Hospital, Sidi Mohammed Ben Abdellah University, Fez, Morocco.  
Email: [ouimzib@gmail.com](mailto:ouimzib@gmail.com)

**Roman A**, MD, Urology Department, Roanne Hospital, Auvergne-Rhône-Alpes, France.  
Email: [adrianroman@gmail.com](mailto:adrianroman@gmail.com)

**Bargoud M**, MD, Urology Department, Roanne Hospital, Auvergne-Rhône-Alpes, France.  
Email: [malek.bargoud@ch-roanne.fr](mailto:malek.bargoud@ch-roanne.fr)

**Hassan Farih M**, Professor, Department of Urology, Andrology, Renal Transplantation, Hassan II University Hospital, Sidi Mohammed Ben Abdellah University, Fez, Morocco.  
Email: [pr.farih@yahoo.fr](mailto:pr.farih@yahoo.fr)

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