

**Case Report**

A Rare Case of Rectal Metastasis from Sarcomatoid Variant of Urothelial Carcinoma: A Case Report

Jacklyn Yek Jia Lin¹, Po Yin Tang², Puay Hoon Tan², Chee Yung Ng³,
Lee Lui Shiong^{1*}

¹ Department of Urology, Singapore General Hospital, Outram Road, Singapore 169608

² Department of Pathology, Singapore General Hospital, 20 College Road, The Academia,
Singapore 169856

³ Department of Coloectal Surgery, Singapore General Hospital, 20 College Road, The Academia,
Singapore 169856

Abstract

Introduction: Urothelial carcinoma of the bladder with sarcomatoid differentiation is known to display aggressive biological behaviour. To our knowledge, this is the first reported case of isolated rectal metastasis from sarcomatoid urothelial carcinoma of the bladder following curative surgery.

Presentation of Case: A 72-year-old male presented with tenesmus 6 months after radical cystoprostatectomy, lymph node dissection and ileal conduit formation for pathological T1N0M0 bladder carcinoma.

Digital rectal examination revealed thickening of the distal rectum with no bleeding. Computerised tomography demonstrated thickening in the rectal mucosa and submucosa with intact perirectal fat. Rectal biopsy performed via colonoscopy confirmed metastases of urothelial carcinoma origin. The patient was treated with palliative radiation.

Conclusion: This case report illustrates an unusual location of urothelial carcinoma metastasis. A high clinical suspicion in patients with this aggressive variant of cancer is required.

Keywords: bladder cancer; metastasis; rectum

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Consent: We confirm that family members of the patients have given their informed consents for the case report to be published.

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***Correspondence to:** Lee Lui Shiong, Department of Urology, Singapore General Hospital, 20 College Road, The Academia, Singapore 169856; Email: lee.lui.shiong@sgh.com.sg

Introduction

Sarcomatoid variant of urothelial carcinoma (SVUC) is defined by WHO as a biphasic malignant neoplasm which exhibits histological and/or immunohistochemical evidence of both epithelial tumors and mesenchymal differentiation with the presence or absence of heterologous elements [1].

SVUC is a rare and aggressive variant of urothelial carcinoma, accounting for 0.2% to 4.3% of urothelial malignancies. There is a relatively low incidence of this variant and hence, no randomised controlled trials have been conducted by far to dictate optimal management for such tumours. The existing literature is reliant on case series, and there are limited systemic options available following local treatment with no consensus regarding the best treatment option.

SVUC has been associated with inferior outcomes in comparison with conventional urothelial carcinoma (CUC). SVUC is associated with advanced stage and exhibits distant metastases at presentation [2,3]. Comparing stage for stage, patients with SVUC are at greater risk of mortality than conventional urothelial carcinoma [2-6].

We present an unusual case of solitary SVUC metastasis in the rectum following radical surgery for curative intent. To our knowledge, this is the first report to document an unusual site of systemic metastasis in urothelial carcinoma of the bladder [6,7].

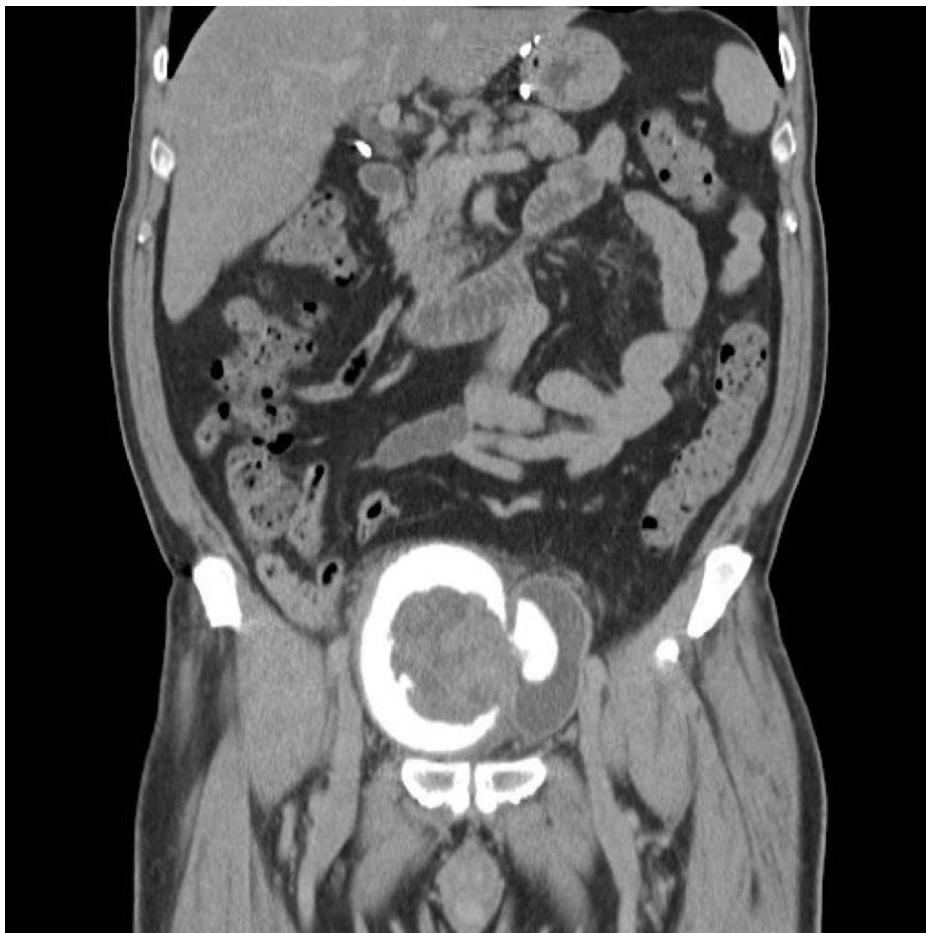


Figure 1 Pre-operative computed tomography (CT) scan showing bladder tumour of 5.2 x 5.1 cm with diverticulum measuring 8cm x 3cm in the left lateral wall. There was no lymphadenopathy or distant metastases.

Case Report

A 71-year-old Chinese gentleman presented 12 months ago with hematuria. He had no significant past medical or surgical history of note. He was a smoker of 20 pack-years. A computerised tomography (CT) urography showed a 5.1 cm x 5.2 cm irregular mass in the bladder with no evidence of distant metastases (Figure 1). Transurethral resection of the tumor revealed a necrotic malignant spindle cell tumour with immunohistochemical evidence of epithelial differentiation, consistent with high grade urothelial carcinoma with sarcomatoid features. The muscularis propria fibers identified in the specimen were negative for tumour, thereby classifying the tumour as pathological T1. Immunohistochemical stains of broad spectrum keratins AE1/3 and MNF116 that indicate epithelial differentiation and p63-HMWCK that identify cells of urothelial origin showed patchy but distinct positivity in tumor cells (Figure 2). He subsequently underwent radical cystoprostatectomy, lymph node dissection and ileal conduit formation. Post-operative recovery was uneventful.

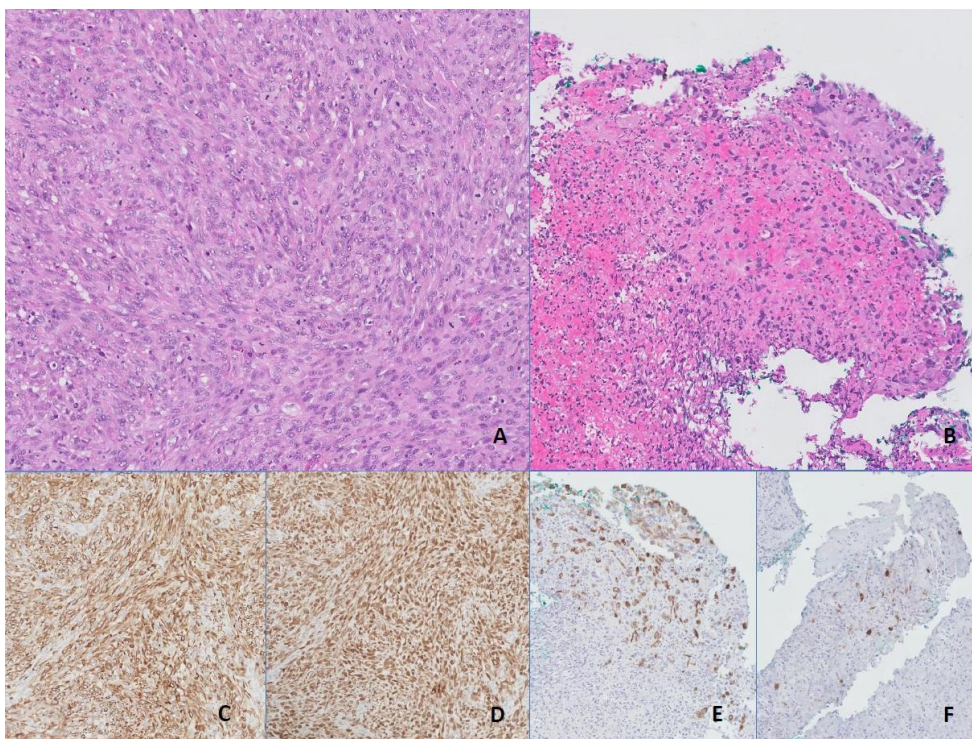


Figure 2 Histological examination of the bladder transurethral resection specimen showed high grade urothelial carcinoma with sarcomatoid features (A; H&E, x10).

Similar morphological appearance demonstrated by the abnormal cells were found in the rectal biopsy (B; H&E, x10). Immunohistochemical staining of the transurethral resection specimen showed strong positivity for MNF116 (C; MNF116, x10) and p63-HMWCK (D; p63-HMWCK, x10). The abnormal cells in the rectal biopsy were suggestive of urothelial origin as demonstrated by the positive immunohistochemical staining of MNF116 (E; MNF116, x10) and CK7 (F; CK7, x10).

Surgical margins of the resected specimen were clear of tumor. All 24 lymph nodes excised were examined and negative for tumour involvement. Therefore, the bladder tumour was pathologically staged as pT1N0M0.

Six months later, he presented with tenesmus. Digital rectal examination revealed a thickening of the distal rectum with no contact bleeding seen. CT abdomen and pelvis revealed thickening in the mucosa

and submucosa of the rectum with intact perirectal fat. There was no recurrent disease detected elsewhere. (Figure 3)

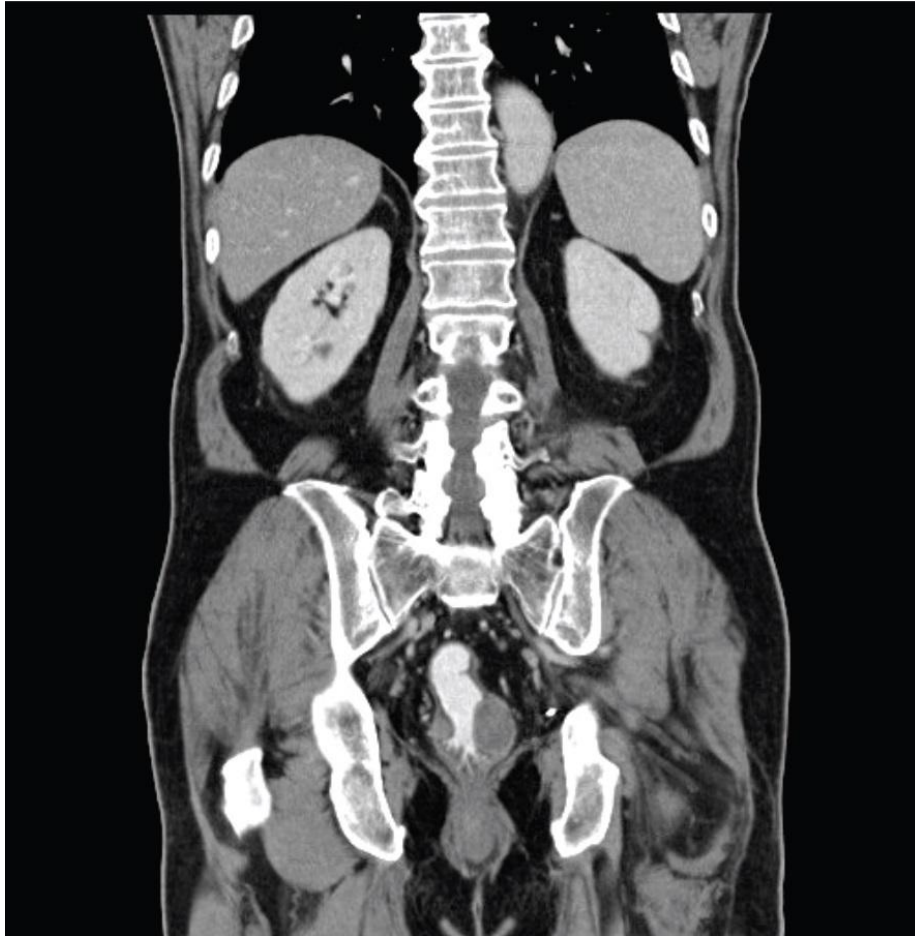


Figure 3 Post-operative CT scan showing rectal thickening of 2.5cm. There is no lymphadenopathy or distant metastases.

During colonoscopy, a circumferential submucosal mass with normal overlying mucosa was seen at the distal rectum. Biopsy confirmed the presence of urothelial carcinoma with microscopic features similar to the bladder tumour and immunohistochemical stains showing positivity for AE1/3, MNF116 and CK7, indicating an epithelial origin consistent with urothelial derivation (Figure 2).

The patient was started on palliative radiotherapy immediately but developed systemic metastases in the lungs and liver and passed away a few months later.

Discussion

This clinical case is unusual in the following aspects.

Metastatic spread from bladder cancer to the gastrointestinal tract is uncommon and tends to be a local extension or surgical implantation involving the rectum. To our knowledge, this is the first case report of isolated rectal metastasis, wherein lung and liver are spared in urothelial carcinoma. The most frequent sites of metastases were regional lymph nodes, liver, lung and bone [5].

The diagnosis of rectal metastases via hematogeneous spread can be supported by radiological, histological and immunohistochemistry assessment.

Radiological investigations demonstrated the preserved integrity of the peri-rectal fat, serosal and mucosal plane, consistent with haematogenous spread rather than direct implantation. Histopathological assessment confirmed radiological findings with evidence of anatomically intact rectal serosal and mucosal layers.

Immunohistochemistry is able to assist in determining cell lineage, with epithelial elements reacting with cytokeratins, whereas mesenchymal components react with vimentin and other markers corresponding to the specific mesenchymal differentiation [4].

The clinical significance lies in its rapid clinical course and short interval between organ involvements. This could possibly suggest that a presentation of rectal metastases could be a harbinger of diffuse systematic metastases with a poor clinical prognosis.

To conclude, rectal metastases from bladder urothelial carcinoma are rare, and predicts for subsequent organ involvement. A high index of clinical suspicion is essential for expedited and accurate diagnosis of this rare clinical entity.

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