

## Case Report

# Clear cell adenocarcinoma of the urinary bladder arising from endosalpingiosis: a case report and review of the literature

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**Abstract:** We present a case of a rare urinary bladder cancer. The patient was a 78-year-old lady who had a cystoscopy following an episode of macroscopic haematuria. Her endoscopic biopsy revealed a high-grade invasive carcinoma with clear cell differentiation. She underwent a radical cystectomy, which showed a clear cell adenocarcinoma (CCA) in an area of endosalpingiosis (Mullerianosis). Most bladder CCAs are not associated with Mullerianosis, have been diagnosed in patients with a previous history of urothelial carcinoma, and have an immunohistochemical profile overlapping that of usual urothelial carcinoma. They are thought to originate from glandular differentiation in urothelial neoplasms. Endosalpingiosis, fallopian tube epithelium outside the fallopian tube, is associated more with serous tumours than clear cell tumours. Although CCA of bladder has previously been called mesonephric carcinoma, there is no convincing evidence for a mesonephric origin. This case supports the postulated Mullerian origin for CCA in bladder cancer.

**Keywords:** Endosalpingiosis, mullerianosis, adenocarcinoma

### Clinical presentation

A seventy eight year old woman with a past history of type 2 diabetes mellitus, hypertension presented with one episode of macroscopic haematuria. She was a non-smoker with no significant family history or occupational exposure.

Ultrasound and CT scan demonstrated a 3.6 cm tumour in the bladder. Initial endoscopic evaluation showed a solid appearing bladder mass on the right posterior wall. The histological assessment revealed a high-grade invasive carcinoma with glandular and clear cell differentiation, invading submucosa but not into detrusor muscle. After discussion in a multidisciplinary forum she underwent evaluation for metastatic disease, which was negative. Tumour markers Ca125, 19.9, AFP and CEA were all within normal limits.

A radical cystectomy, anterior exenteration and diversion with an ileal conduit were performed.

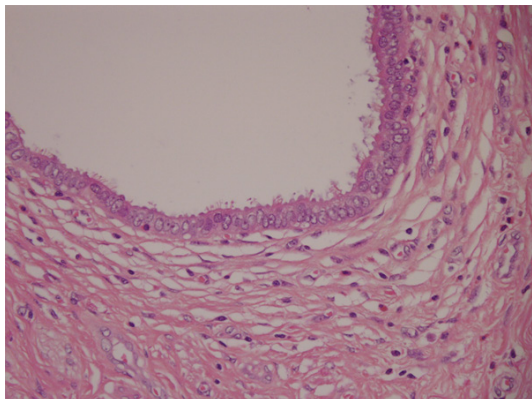
Her post-operative course was complicated by acute renal injury secondary to hypotension from rapid atrial fibrillation. She recovered from her surgery and was discharged home 18 days afterwards.

### Histopathology

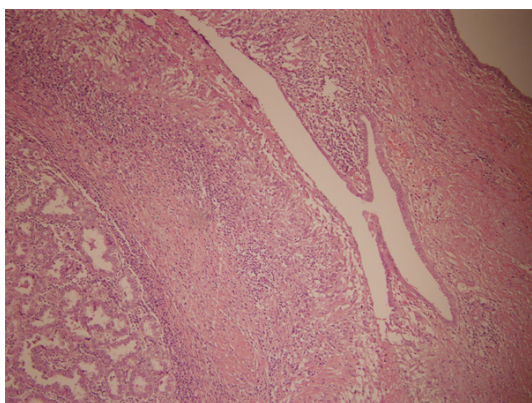
The specimen was a radial cystectomy and hysterosalpingo-oophorectomy, which showed an ulcer involving bladder mucosa.

Microscopically, there was a tumour arising in bladder wall. This measured 6.5 mm in maximum dimension and was composed of irregular tubules, sheets and acini of epithelial cells with large pleomorphic nuclei and prominent nucleoli with frequent mitoses and apoptosis. In some areas tumour cells had clear or vacuolated cytoplasm while other areas had a hobnail appearance. No lymphovascular or perineural invasion was seen. Tumour was seen to invade into the detrusor muscle. The urothelium at the edge of the defect showed reactive changes.

## Endosalpingiosis and clear cell adenocarcinoma



**Figure 1.** H&E section of endosalpingiosis. ( $\times 200$  objective).



**Figure 2.** H&E section showing clear cell adenocarcinoma and endosalpingiosis. ( $\times 40$  objective).

Tumour arose in a background of endosalpingiosis with focal mild atypia. Several foci of tubal-type, with ciliated columnar epithelium were present in the bladder stroma (**Figures 1** and **2**). No metastatic tumour was seen in any of twelve lymph nodes. Tumour cells showed positive immune staining for CK7, P53 and P504S. The Ki67 labelling index was 25%. Tumour cells were focally positive for CK20. No immune staining for CA125, CEA, ER, PR or Pax5 seen.

The diagnosis was clear cell adenocarcinoma (CCA) of the urinary bladder arising from an area of endosalpingiosis (mullerianosis).

### Discussion

Though CCA of bladder has previously been called mesonephric carcinoma there is no convincing evidence for a mesonephric origin. Most bladder CCAs are not associated with Mull-

erianosis, have been diagnosed in patients with a previous history of urothelial carcinoma, and have an immunohistochemical profile overlapping that of usual urothelial carcinoma. They are thought to originate from glandular differentiation in urothelial neoplasms.

A Mullerian origin for CCA has been postulated [1-3] due to its female preponderance and morphological similarity to CCA of the female genital tract, which is often associated with benign Mullerian epithelium.

Endosalpingiosis, fallopian tube epithelium outside the fallopian tube, is associated more with serous tumours than clear cell tumours. This case supports the postulated Mullerian origin for CCA in bladder cancer.

### Acknowledgements

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### Disclosure of conflict of interest

None.

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