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## Oncology

## Tubulovillous Adenoma in a Urethral Neobladder Anastomosis



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#### ABSTRACT

We present a case of a tubulovillous adenoma arising in a neobladder that was managed by cystoscopic resection. A 64 year-old male underwent a cystectomy with creation of an ileocolic neobladder urinary diversion for T2 urothelial carcinoma of the bladder. Nine years following his surgery, the patient noted several episodes of gross hematuria. Cystoscopic evaluation revealed the rare occurrence of a 3 cm tubulovillous adenoma with high-grade dysplasia at the neck of the neobladder.

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#### Introduction

We report a case of a tubulovillous adenoma with high-grade dysplasia presenting at the neck of an ileocolic orthotropic neo-bladder (ON). Individuals with invasive bladder cancer have the option of one of three urinary diversions after cystectomy: an ileal conduit (IC), ON or Indiana pouch. The rare potential complication of secondary tumor development is of important consideration for patients with intestinal diversions.

Tumor development has been well described following ureterosigmoidostomy, augmented bladders, and other conduits. The spectrum of tumors found in isolated intestinal diversions include, but are not limited to, urothelial carcinoma (UC), squamous cell carcinoma, signet ring cell carcinoma, and adenomas. Kälble et al analyzed 17,758 patients with urinary diversion and found that the tumor risk of ureterosigmoidostomies (2.58%) and cystoplasties (1.58%) were significantly higher than other types of continent diversions. In addition, they note ileocolic ON (1.25%) had a significantly higher rate when compared to ileal ON (0.14%). Raman et al reported the first case of tubulovillous adenoma in an Indiana pouch. We report the first case of tubulovillous adenoma in an ileocolic ON at the bladder neck.

#### Case presentation

In 2004, a 64 year-old patient presented with gross hematuria. Hematuria workup revealed a large papillary tumor on the right lateral wall of the bladder. The patient underwent a transurethral resection of bladder tumor that demonstrated T1 UC and carcinoma in situ. The patient then received induction intravesicle therapy with bacille calmette guerin (BCG) and interferon. Subsequently, he recurred with a T2a high grade UC. In 2005, he underwent a radical cystoprostatectomy with the creation of an orthotopic ileocolic ON. The patient was followed semiannually for 5 years. At his 5 year follow-up, the patient experienced difficulty with self-catheterizing. Cystoscopic examination revealed only scarring at the urethral neobladder anastomosis.

In 2014, 9 years postoperatively, the patient noticed blood in his urine. Workup for gross hematuria was performed including cystoscopy and an MRI, both confirming the presence of a large polyploid mass at the anastomosis of the neobladder and urethra (Fig. 1). His urine cytology was negative. He underwent a resection of this polypoid mass.

Pathological examination demonstrated tubulovillous adenoma with predominantly low-grade dysplasia (Fig. 2). The adenomatous epithelial cells showed mild hyperchromasia with enlarged, oval nuclei, occupying basal half of the cell cytoplasm. There was retention of basal polarization with no significant nuclear pleomorphism. Focally, high grade dysplastic features were present, showing crypt budding with marked loss of polarity and pseudostratification of neoplastic nuclei that extend toward the luminal half of the cells (Fig. 3). Lamina propria invasion was not identified.

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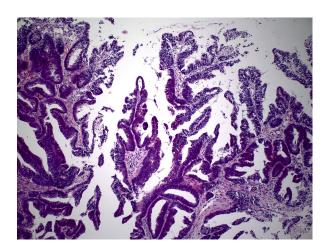
**Figure 1.** MRI of abdomen and pelvis demonstrating presence of a large polyploid mass at the interface of the neobladder and urethral structure.

#### Discussion

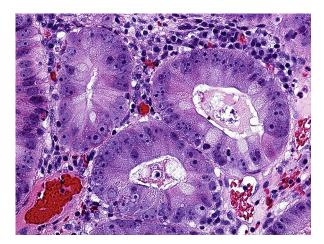
The presence of a malignant growth in urinary diversions using bowel segments for bladder substitution is rare. The majority of growths are adenomatous lesions such as tubular, tubulovillous, or villous adenomas. However, invasive lesions such as adenocarcinomas are seen. Important differences have been noted with different types of diversions. In ureterosigmoidostomies, nearly 100% of these lesions are either adenomas or adenocarcinomas. However, isolated intestinal segments have more variable histology with 27.4% of tumors being UC, squamous cell, signet ring cell, oat cell carcinomas or leimyosarcoma.

The mechanism for carcinogenesis is still unclear, however, increases in nitrosamines, ornithine decarboxylase, sialomucins, and chronic infection with inflammation are all thought to be contributing factors. <sup>1,3</sup> In isolated urinary reservoirs, one explanation might be the more intensive contact between urine and the urointestinal mucosa. The hydrolytic urine enzymes activate carcinogens throughout the diversion and hence, only 58% of the tumors are found at the urointestinal anastomosis with the remaining being found more distally. However, in ureterosigmoidostomies, urine is most concentrated at the anastomosis compared to being diluted throughout the rest of the colon. Hence, it is thought to be the reason why almost 100% of the lesions are seen in this region. <sup>1</sup>

Few reports of tumor development in neobladders have been presented in the literature. Austin et al note one case of an adenoma



**Figure 2.** Tubulovillous adenoma with low-grade dysplasia (H&E, original magnification  $\times$  10).



**Figure 3.** Tubulovillous adenoma showing high-grade dysplasia component (H&E, original magnification ×40).

in a colonic neobladder, a nephrogenic adenoma in a sigmoid neobladder, and a carcinoid tumor in an ileal neobladder. <sup>1,5</sup> The tumor sites for secondary tumor growth in the neobladder cases were all found at the neobladder base (urethral anastomosis). Kälble et al report two tumors, one adenocarcinoma and one desmoid, in ileal ONs, and one benign adenoma in a colonic ON, which were located at the ureterointestinal anastomosis. In addition, they report three masses in the ileocolic ONs (one adenoma, one adenocarcinoma, and one carcinoid) with all three occurring in the intestinal segments.<sup>2</sup>

Our case is the first report of a tubulovillous adenoma presenting at the neck of an ileocolic neobladder. It is evident that there are differences in location and type of tumor found depending on the type of diversion used. This case will aid in better understanding secondary tumor growth following bladder substitution and the need to follow and recognize these occurrences in this population of patients.

### Conclusion

Secondary tumor parthenogenesis is still unclear. Follow-up care with patients is imperative. Any abnormalities, such as hematuria require investigation, as it may be an indication of disease. Early recognition and detection is essential to preserve the integrity of the urinary diversion.

## Consent

Informed consent was obtained from the patient for publication of this case report and any accompanying images.

## Conflict of interest

The authors declare they have no conflict of interest.

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