



Inverted urothelial papilloma: A case report

Raj A. Kumar^{a,*}, Nikhil Batra^b, Michael Madaa^b, Kevin Campbell^b, Jon Frankel^c,
Udaya Kumar^c

^a Catherine & Joseph Aresty Department of Urology, Keck Medicine of USC, University of Southern California, Los Angeles, CA, USA

^b Department of Urology, University of Florida College of Medicine, University of Florida, Gainesville, FL, USA

^c Citrus Memorial Hospital, Inverness, FL, USA

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ABSTRACT

Inverted urothelial papilloma (IUP) is an unusual tumor that typically presents as a non-invasive, endophytic urothelial neoplasm. Though fairly well described in the bladder, IUP of the upper urinary tract is fairly rare, with only 68 cases documented in English language literature. Our patient presented with an IUP of the distal left ureter and was treated with a distal ureterectomy and reimplant of the ureter with a psoas hitch.

1. Introduction

Inverted urothelial papilloma (IUP) is a rare tumor typically occurring in the bladder neck, trigone, or the prostatic urethra.¹ Histologically, IUPs are similar to inverted papillary tumors of the nasal cavity and paranasal sinuses. IUPs have rarely been described in the upper urinary tract. A recent systematic review of the literature by Santi et al. described 68 cases of IUP in the renal pelvis and ureter in English literature.¹ Patients typically present with symptoms similar to those of other upper urinary tract neoplasms including hematuria, flank pain, and renal colic.^{1,2} However, such tumors can rarely be asymptomatic, diagnosed as an incidental finding. Historically, treatment of IUP of the upper urinary tract involved broad surgical resection such as nephrectomy or nephroureterectomy. More recently, localized resection and ureterectomy have been used to good effect with minimal recurrence.

Here we present a case of IUP of the left distal ureter treated with distal ureterectomy and reimplant of the ureter with a psoas hitch.

2. Case presentation

A 54-year-old male patient presented to his primary care physician with complaints of painless intermittent gross hematuria for approximately the last two years. Patient reported a smoking history of 1 pack per day for thirty years. Computerized tomography (CT) imaging with delayed phase contrast was obtained which revealed a 2.3 × 3.0cm filling defect of the left ureter at the level of the iliac crest. A representative coronal image in the excretory phase is seen in Fig. 1. There

was an intraluminal soft tissue mass within the ureter with no evidence of urinary obstruction.

The patient was referred to urology. Voided urine cytology showed atypical urothelial cells. Patient subsequently underwent diagnostic cystoscopy with bilateral retrograde pyelograms and left ureteroscopy. A large, papillary mass was noted at the junction of the distal and mid ureter highly suspicious for urothelial carcinoma. Ureteral washings with cytology and tumor biopsy were performed. Selective cytology from the left ureter revealed only benign urothelial and squamous cells. Left ureteral biopsies showed only benign squamous cells.

Considering the long patient history of smoking and voided urine cytology revealing atypical cells, the patient was counseled and underwent an open partial distal ureterectomy. Intraoperative ureteroscopy of the left upper urinary tract showed no abnormal findings proximal to the mass following excision. Reimplant of the left ureter was performed with a psoas hitch and bladder elongation using the Turner-Warwick modification.³

Pathology revealed a polypoid mass measuring 3 cm in diameter (Fig. 2). Tumor histology was described as areas of papillary and sessile proliferation of urothelial cells showing minimal atypia (Fig. 3). Proliferating nests of cells were observed to invaginate downward into the underlying fibrovascular stalk. Foci of cystic change were present and focal squamous metaplasia of the urothelial cells was noted. These histologic findings were consistent with a diagnosis of IUP. Margins were negative for malignancy.

The patient had an uneventful postoperative course, and Foley catheter was removed after 10 days following a negative cystogram.

* Corresponding author. Research Assistant Urology Institute, University of Southern, California.

E-mail address: rkumar28@uic.edu (R.A. Kumar).

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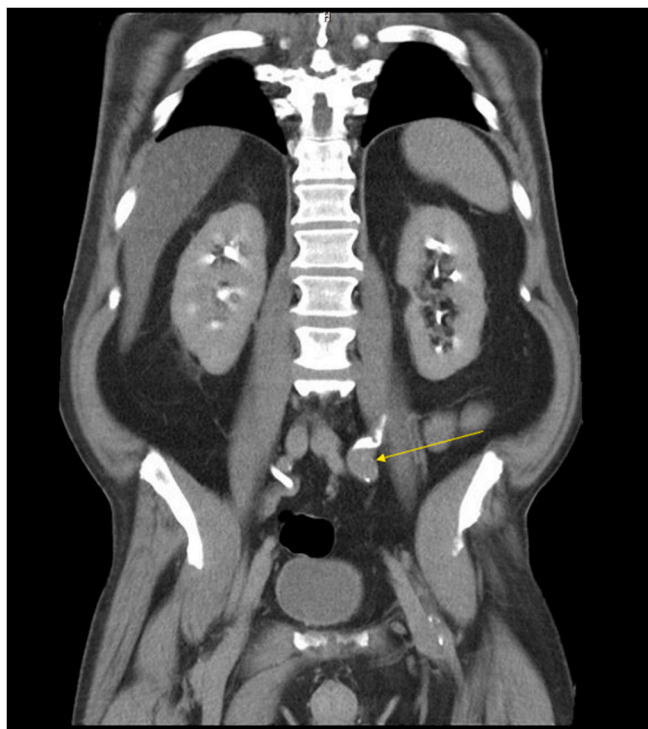


Fig. 1. Coronal image in the excretory phase.

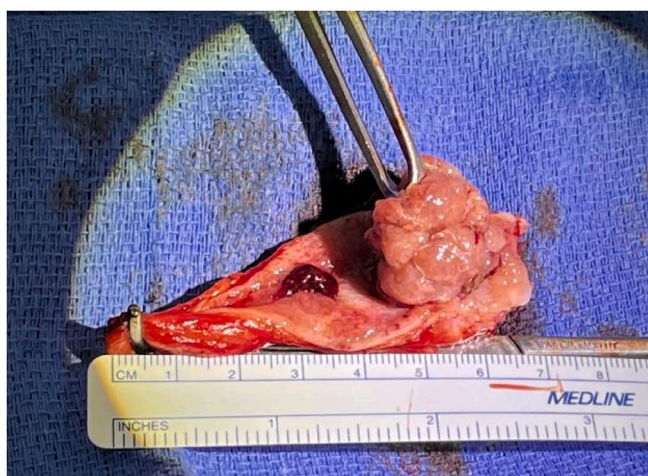


Fig. 2. Excised specimen measuring 3 cm in diameter.

Ureteral stent was removed after 1 month, and the patient currently reports no complications.

3. Discussion

While more than 1000 cases of IUP have been reported, occurrence in the upper urinary tract is fairly rare.⁴ The tumor occurs overwhelmingly in males, typically in the 6th and 7th decade of life. Though the etiology of the tumor remains unclear, it has been suggested that the human papillomavirus may play a role similar to the occurrence of inverted papilloma tumors of the nasal cavity.⁵ Differential diagnosis may pose a challenge, and often includes urothelial carcinoma and nephrogenic adenoma.

In their clinical series, Asano et al. reported that 3 out of 48 cases developed recurrence, all of which were in the bladder.⁴ IUP has historically been considered a benign tumor. However, in the 1980s,

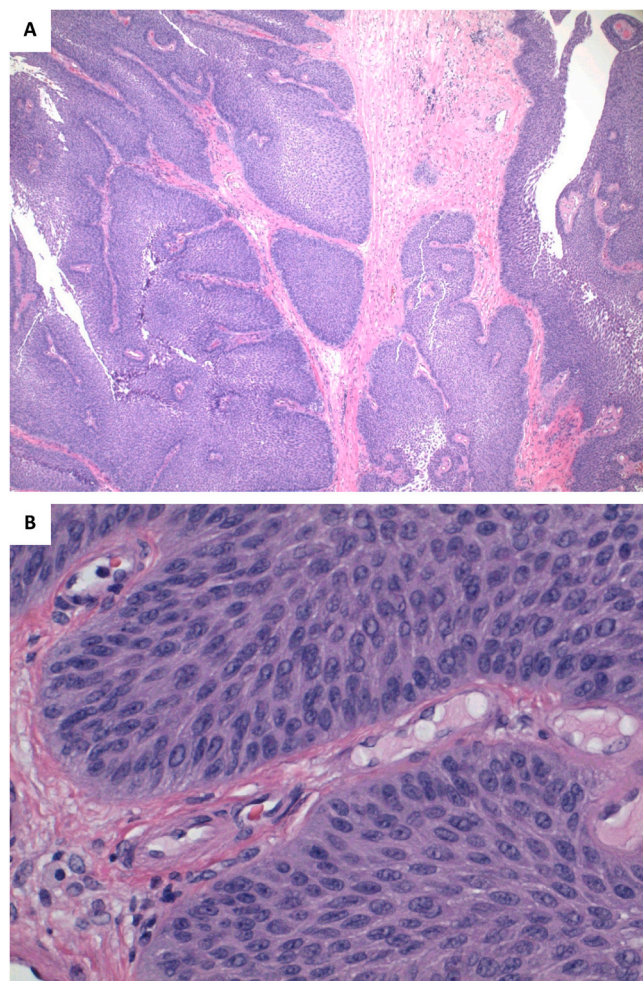


Fig. 3. Tumor histology showing areas of papillary and sessile proliferation of urothelial cells showing minimal atypia. (A) Displays low power histology and the architecture of an inverted urothelial papilloma involving downward growth lined by urothelium. (B) Medium power histology showing cytologic characteristics.

reports of recurrence complicated with concurrent malignant urothelial cancers drew attention to the malignant potential of IUPs.⁴ While there have been efforts to predict malignant potential using ploidy patterns and proliferative activity, the true malignant potential remains unclear.

Treatment of IUPs is similar to other tumors obstructing the upper urinary tract. Though this has historically involved broad resection of the tumor with nephrectomy or nephroureterectomy, more recent therapy involves local resection and ureteral reimplant as in our patient. When a firm diagnosis of IUP of the upper urinary tract can be made with endoscopic biopsy, transurethral resection may be considered as a viable therapy.^{1,4} However, most cases would still require a local segmental resection or extirpative surgery.

Given that there are well documented cases of tumor recurrence, follow up must include periodic surveillance with appropriate imaging and endoscopic evaluation for at least 2 years.⁴ Tumor recurrence typically occurs within the bladder, but has been demonstrated to occur in the upper urinary tract as well.^{1,4}

4. Conclusion

Here we have presented an unusual case of IUP of the upper urinary tract. IUPs are characterized by an inverted pattern of growth without atypical features or other characteristics of malignancy and are usually managed with surgical resection. A follow up of at least two years is

advised to surveil for tumor recurrence.

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