

## Giant Inverted Papilloma in the Ureter: An Extremely Rare Urological Condition

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### Abstract

We report a rare case of inverted papilloma in the ureter, in a 75-year-old female who presented with haematuria. During workup, CT and MRI showed filling defect – a soft tissue mass, masquerading as transitional cell carcinoma (TCC) in the right lower ureter. Ureteroscopy showed papillary lesions along the ureter, definitive biopsy could not be obtained due to difficult access. Subsequently on a suspicion of transitional cell carcinoma, nephroureterectomy was performed. Surprisingly, histology confirmed inverted papilloma of the distal ureter, which is an extremely rare to occur in the ureter.

**Keywords:** Biopsy, Inverted Papilloma; Nasal Cavity; Paranasal Sinuses; Soft Tissue; Ureter

### Background

Inverted papilloma in urology, is most common in the bladder. As per a systematic literature review in 2020, there has been report of only 68 cases of inverted papilloma in the upper tracts [1]. This is in light of a 2020 review article. Inverted papilloma in the urinary tracts is histologically similar to inverted papilloma of the nasal cavity and paranasal sinuses. Out of the 68 reported cases, 36 cases occurred in the ureter, and 32 in the renal pelvis [1].

Initial description was by Paschkis in 1927 [2], and since then more than 1,000 cases reported in the literature regarding urothelial inverted papilloma [3]. Most inverted papilloma cases occur in the bladder neck, trigone, or prostatic urethra [1]. At presentation patients are usually in their fifth or sixth decade of life, with age range of 44-81 years [4]. This clinical condition is observed significantly more commonly in men, with nearly six men affected for every woman [4].

Inverted papilloma comprises 1.4–2.2% of total urothelial neoplasms [5]. Approximately <1% of them occur in the upper tracts [3]. Of those, around half are in the ureter [5]. Rest of the lesions are observed in the urinary bladder.

Haematuria (blood in the urine) and dysuria (painful micturition) are the most common presentation of inverted papilloma in the urinary tract [6]. In some cases, it can be an incidental finding on CT/MRI scans, performed due to other clinical reasons. Although definitive diagnosis requires histopathological examination. There is no clear recommendation from recognized urological organizations; rather there are multiple schools of thoughts regarding follow up regime.

### Case Files

A 75-year-old female was referred to our outpatient clinic on 2-week-wait

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urgent pathway due to visible hematuria. Flexible cystoscopy showed a papillary lesion <2 cm in vicinity of right ureteric orifice and another papillary lesion bulging through the right ureteric orifice. Bimanual examination revealed no masses.

CT Urogram as part of haematuria workup, showed filling defect in right lower ureter, mass near the ureteric orifice as well as moderate proximal hydroureteronephrosis of right side. CT Urogram and MRI confirmed soft tissue mass in the right distal ureter which was protruding into the bladder (Figure 1-4).

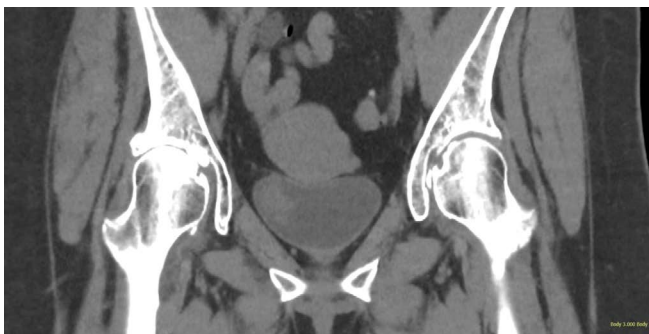


Figure 1: CT image showing lesion along right side of the bladder.

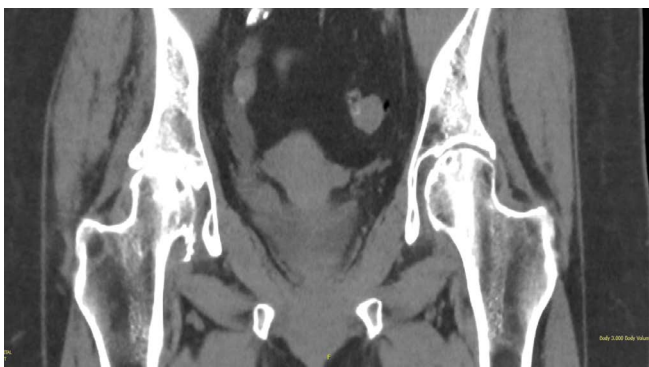


Figure 2: CT image showing lesion along right of distal ureter.

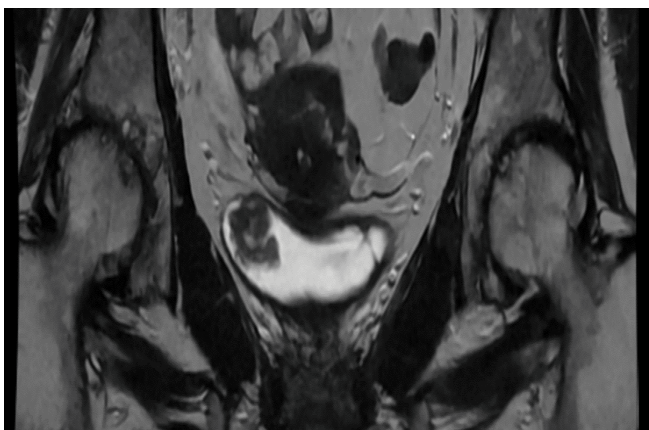


Figure 3: MRI image showing lesion along right side of bladder.

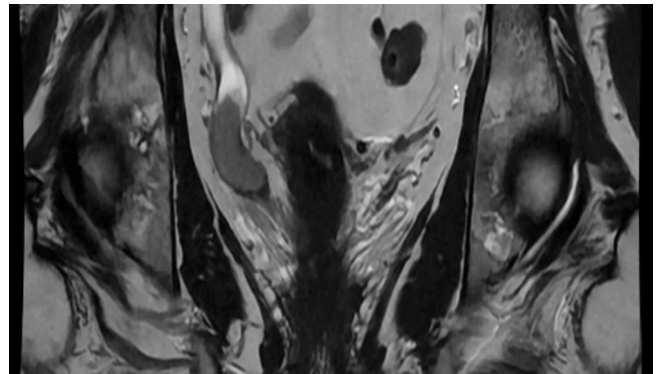


Figure 4: MRI image showing lesion along right distal ureter.

The patient's case was discussed in the Multi-Disciplinary Team (MDT) and the consensus was to proceed with a Trans-Urethral Resection of Bladder Tumor (TURBT), ureteroscopy and biopsy in the first instance. The reasoning behind this was, to obtain histological diagnosis first (Figure 5).

Cystoscopy revealed right distal ureteric tumor hanging from the ureteric orifice into the bladder, on a stalk. Ureteroscopy revealed right distal ureter full of papillary tumor. Mid and proximal ureter revealed no lesions (Figure 6).

Loop diathermy was applied to stalk of the right ureteric tumor, removed en-bloc and sent for histology. Histology was sent to tertiary center for further analysis, as differential diagnosis of this lesion was between inverted papilloma and inverted urothelial carcinoma.

Subsequent right nephroureterectomy was performed on a suspicion of transitional cell carcinoma (TCC). Surprisingly, histology confirmed inverted urothelial papilloma (IUP). No evidence of malignancy.

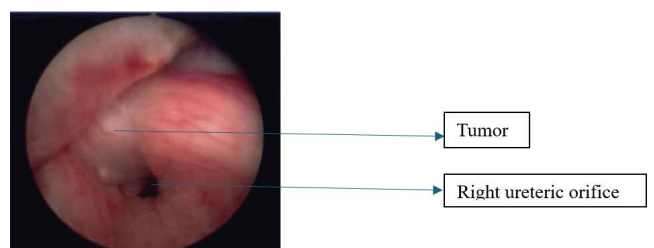


Figure 5: Cystoscopic appearance of tumor bulging through right ureteric orifice

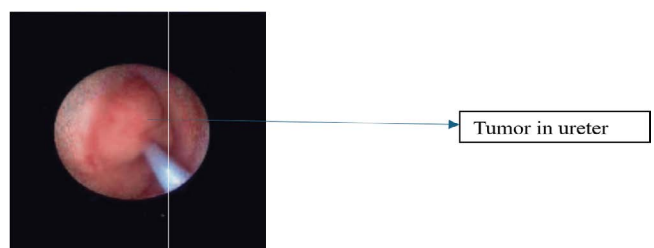


Figure 6: Ureteroscopy showing distal ureteric tumor.

## Discussion

The literature appears to be limited to smaller case series and reports describing the clinical presentation, pathology, treatment approaches and follow-up based on institutional experiences. There are no consolidated, widely accepted guidelines for managing inverted papilloma of the ureter from societies like the- European Association of Urology (EAU), American Urological Association (AUA), British Association of Urological Surgeons (BAUS) or British Journal of Urology International (BJUI).

Mainstay of treatment of inverted papilloma in the urinary tract is surgical resection – because of its appearance being very similar to a concerning urothelial lesion [7]. As per local policy, such cases will go through standard process of being discussed in the multidisciplinary meeting; and in presence of difficulty in distinguishing from a case of transitional cell carcinoma (TCC), nephroureterectomy is considered a definitive management in upper tract lesions. If found in the lower tracts, this lesion will undergo a surgical resection at first setting as well.

The option of nephroureterectomy is strongly considered, chiefly when dealing with large or indistinct ureteral masses, owing to the risk of either simultaneous transitional cell carcinoma (TCC) or subsequent malignant transformation of inverted papilloma.

Among a cohort of 348 reported cases with inverted urothelial papilloma (IUP), there were 5 (1.43%) and 4 (1.15%) patients with simultaneous and subsequent transitional cell carcinoma of the urinary tract, respectively [8].

Regarding inverted urothelial papilloma in the bladder, there is no concrete recommendation regarding follow up. A school has recommended that a trans-urethral resection is adequate treatment and surveillance protocols similar to carcinoma surveillance may be unnecessary [6]. Another school has commented that patients should be followed up with endoscopy and radiographic studies similar to those with low grade transitional cell carcinoma (TCC), particularly in upper tract cases [4].

For upper tract inverted papilloma, follow up ideally should include upper tract surveillance along with cystoscopy surveillance. Recommendation is cystoscopy and ultrasonic scans every 6 months for the first 2 years, and then annually. This is in light of a study encompassing 10 patients, the time to recurrence in the majority of cases is no more than 2 years following surgery [7].

## Conclusion

This case encompasses the importance of considering inverted papilloma as a differential diagnosis for ureteric

tumors, despite its rarity. Accurate diagnosis relies on histopathological examination, and due to its close resemblance to TCC, surgical excision is often required for definitive diagnosis and treatment. Long-term surveillance is recommended to monitor for recurrence or malignant transformation, particularly in cases involving the upper urinary tract.

## Ethical Implications

No experiments were conducted in this case study; only the clinical manifestations of the disease were noted. Therefore, ethical approval from any authority was not required.

## Consent of the patient

Only the disease course was observed in this case report. There were no unconventional treatment options mentioned in this article. Therefore, consent of the patient was not an ethical issue.

## Conflicts of Interest

The authors stated that they have no conflict of interest.

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