A rare case of successful endoscopic management of a fibroepithelial polyp with intussusception of the ureter and periodic prolapse into bladder

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ABSTRACT

Fibroepithelial polyps of the ureter are rare non-epithelial benign tumours with a slight female predominance. They are primarily located in the upper left ureter, incidentally diagnosed in patients who are evaluated for gross haematuria or renal colic. Fibroepithelial polyps are traditionally managed by open surgery with resection of the polyp and its stalk or partial resection of the ureter. We describe the case of a fibroepithelial polyp located in the distal right ureter, which occurred in a 42-year-old man with lower urinary tract symptoms, haematuria and flank pain. To our knowledge, this is the first reported case of ureteral fibroepithelial polyp acting as a lead point for ureteral intussusception and to protruding periodically into the bladder cavity, which was successfully resected by ureteroscopic electrocauterisation with good outcome.

KEYWORDS

Fibroepithelial polyp – Ureteral intussusception – Ureteroscopy

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Case history

A 42-year-old man presented with urgency, frequency and colicky abdominal pain in the right flank of 18 months duration with intermittent gross haematuria. He was a non-smoker and had not been exposed to any solvents or chemicals. His medical history was free of urinary tract infections and urolithiasis. Abdominal and digital rectal examinations were unremarkable. His urine culture and routine blood tests were normal.

Intravenous urography revealed a right distal ureteral enlargement containing a pedunculated polypoid filling defect with what was thought to be a ‘bell-shaped’ configuration in the upstream ureter, together with a filling defect in the ipsilateral bladder wall (Fig 1). There was no hydro-ureteronephrosis and the left urinary tract was normal. Contrast-enhanced computed tomography (CT) demonstrated a solid mass in the right distal ureter protruding into bladder (Fig 2), with no evidence of infiltration into surrounding tissues or distant organs.

Subsequent diagnostic cystoscopy demonstrated a solid papillary lesion which periodically prolapsed into the bladder from the right ureteral orifice, with no evidence of concomitant bladder tumors. <http://www.filedropper.com/ufpprolapse>. Samples of voided urine and ureteral washing were taken for cytology. In the same session, a right retrograde pyelogram revealed what was thought to be a ‘V-shaped ureter’ upstream from the mass, suggestive

of antegrade ureteral intussusception (Fig 3). A biopsy was taken from the prolapsed portion of the polyp; the specimen was negative for malignancy and cytology also proved negative. We decide at this point that endoscopic management should be attempted.

A ureteroscopy was performed four weeks later and confirmed ureteral intussusception at the attachment point of the pedunculated polyp with no evidence of ischaemic necrosis (Fig 4). The specimen was then resected en bloc with its stalk by ureteroscopic electrocauterisation (Fig 5) and extracted in its entirety. The intussusception was immediately resolved and no additional lesions were noticed throughout the entire ureter. A double J stent was inserted to prevent stricture, with a plan for it to be removed four weeks later. The excised tumour measured 4.5 × 1.0 cm and had an irregular cystic surface (Fig 6). Pathological examination confirmed a fibroepithelial polyp with cystically dilated von Brunn’s epithelial nests in the lamina propria (Fig 7). No dysplasia or malignancy was noticed.

The patient was symptomless during the 12 months following tumour excision and subsequent CT with intravenous urography revealed a mild residual distal hydroureter with no recurrence or ureteral stricture (Fig 8).

Discussion

Ureteral fibroepithelial polyp (UFP) is a rare non-epithelial benign tumour of mesodermal origin.^{1,2} Polyps usually

originate in the upper ureter, most commonly on the left side. They occur more often between the second and fourth decades of life, with a slight female predominance.^{1,2} How this entity arises remains unclear; obstruction, urolithiasis, trauma, irritation, infection, development anomaly and hormonal imbalance have all been proposed as causative agents. Necrosis of the polyp and ureteral obstruction are the most common complications.^{1,2} The current case, with no known comorbidities, showed a ureteral fibroepithelial polyp arising in the lower right ureter and leading to ureteral intussusception in a male patient.

Intussusception of the ureter is an uncommon complication of a ureteral mass (benign or malignant tumour),

urolithiasis or ureteric instrumentation; only 27 cases have been listed in literature.³⁻⁶ This entity may occur when the proximal ureteral wall (intussusceptum) telescopes into the distal ureter (intussusciens).

Since the first UFP was described in 1932, fewer than 13 cases of polyp-induced intussusception of the ureter have been reported.^{3,4} A pedunculated polyp may be pulled downstream by the ureteral peristalsis, urine flow and gravity, resulting in intussusception of the upstream

Figure 1 Excretory urogram showing a pedunculated polypoid filling defect in the right dilated distal ureter with a filling defect in the ipsilateral bladder wall. A 'bell-shaped' ureter was seen in the upstream ureter suggesting ureteral intussusception (arrow).

Figure 3 Right retrograde pyelogram showing a contrast in the upstream ureter terminating in a V-shaped configuration (arrow).

Figure 2 Coronal computed tomography of the abdomen showing a solid mass in the distal ureter protruding into bladder.

Figure 4 Intraoperative ureteroscopic view showing a telescoping of the proximal segment of the ureter (intussusceptum) within the lumen of the adjacent segment (intussusceptions). A wire was passed through the lumen of the invaginated segment.

Figure 5 Intraoperative ureteroscopic view showing electrocauterisation of the polyp

ureter.³⁻⁵ However, intussusception with periodical protrusion of the polyp into the bladder is even a rarer event; to

Figure 6 Gross specimen of the polyp.

our knowledge, only one such case has been documented, which was managed by laparoscopic ureterectomy.⁵ In the current case, the upstream ureter was thought to telescope only during the periodic prolapse.

Pathologically, a UFP consists of fibrous and vascular tissue with normal transitional epithelium.³ In this case, however, cystically dilated von Brunn's epithelial nests seen into the underlying mucosa may have resulted from chronic urothelial inflammation due to periodical prolapse of the polyp with repeated mechanical trauma to its urothelium.

UFPs are usually incidental findings on urinary tract imaging in patients who are evaluated for gross haematuria or renal colic.¹⁻⁵ In our patient, urgency and frequency were probably associated with the periodic prolapse into the bladder cavity, whereas intussusception might explain haematuria, flank pain and ureteral dilatation.^{5,6}

A UFP can usually be diagnosed by intravenous urography, ultrasound, CT or retrograde pyelogram. Radiological features of UFPs, however, are non-specific. Consequently, the differential diagnosis may include transitional cell carcinoma (TCC), blood clots or radiolucent calculi.¹⁻⁵ Moreover, although only two cases of UFP with concurrent malignancy have been reported,^{1,2} polyp-induced intussusception is frequently associated with TCC.^{3,4,6} In addition, blood supply to the intussuscepted segment may be compromised with the risk of ureteral ischaemia. Under these circumstances, ureteroscopy with biopsy is often helpful to rule out TCC, to screen intussusception with signs of ischaemia and to determine the optimal treatment approach. This case, with prolapse into bladder, cystoscopic examination also was diagnostic, ruling out malignancy after biopsy.

Telescoping of the ureter, however, shows characteristic radiological findings and can easily be diagnosed by intravenous urography or retrograde pyelogram. Other specific CT features,^{5,4,6} such as the 'line sign', 'concentric' or 'target sign' and 'stalk-of-corn appearance' were not seen in

Figure 7 Microscopic analysis showing a fibroepithelial polyp with cystically dilated focal islands of benign-appearing urothelium located in the lamina propria, most consistent with von Brunn's epithelial nests.

Figure 8 Follow-up computed tomography (left) and excretory urogram (right) showing residual right distal hydroureter (arrows), with no recurrence or stricture.

our case, as the patient presented with a short segment of transient ureteral intussusception, which might also explain the absence ischaemic necrosis.

There are currently no formal guidelines on the surgical management of UFPs. However, treatment could change according to the size and the site of the polyp, ipsilateral

renal function, status of the upper urinary tract (hydronephrosis) and the associated conditions (calculus, TCC or intussusception). It can include ureteroscopic resection or fulguration using electrocautery or holmium:YAG laser, polypectomy via open ureterotomy, partial ureterectomy, dismembered pyeloplasty or nephro-ureterectomy.¹⁻⁶

Conservative management, percutaneous resection or laparoscopic excision have been previously used in selected cases.^{2,5} The present case, with polyp-induced intussusception, could have been managed with radical surgery. However, no concurrent TCC or ischaemic necrosis of the intussusceptum was noticed. Consequently, endoscopic management is a good treatment option for such isolated small distal UFPs, especially in a young patient with a normal ipsilateral renal function. Retrograde ureteroscopic resection may be feasible, safe and effective, but potential risk factors include recurrence and ureteral stricture.^{2,5} It is therefore necessary to achieve complete resection and to suggest long-term follow-up as part of UFP management.

Conclusion

The case presented herein is unique and interesting. To our knowledge, this is the first reported case of endoscopic management of a UFP with concurrent ureteral intussusception and periodic prolapse into bladder. Patients

diagnosed with this condition should be assessed for concomitant TCC and/or ureteral ischemia prior to treatment. Radical en bloc excision of the affected ureter can be used in selected cases. Nevertheless, ureteroscopic resection of the UFP is the standard treatment option in this case as it seemed to be easier, less invasive and successful.

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