The rare case of an extraperitoneal ureteral herniation into the inguinal canal of a 12-year-old boy

K Cao¹, M Turner², P Mishrah³, P Cuckow¹

¹Urology Department, Great Ormond Street Hospital For Children NHS Foundation Trust, London, UK

²University of Cambridge Department of Public Health and Primary Care, Primary Care Unit, Cambridge, UK

³Urology Department, Evelina London Children's Healthcare, London, UK

ABSTRACT

Ureteric herniation into the inguinal canal is a rare condition in adults and even more so in the paediatric population. These hernia types can be categorised as paraperitoneal or extraperitoneal. The former are the only varieties encountered in the paediatric literature, where patients are generally infants, while the latter phenomenon is seen in 20% of ureteric herniations in adults. We present a case of an extraperitoneal ureteric herniation into the inguinal canal in a 12-year-old boy with previous surgery for cloacal exstrophy, analyse the existing literature on the condition and make the speculation that the extraperitoneal variety is an adult variant seen in this child because of weakness in the surrounding structures as a result of previous surgery.

KEYWORDS

Hernia – Ureter – Extraperitoneal – Paediatric urology

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CORRESPONDENCE TO

Kevin Cao, E: kevincaowork@gmail.com

Case history

A 12-year-old boy presented with fever and a painful left groin. He had a past medical history of cloacal exstrophy, untethering of the spinal cord and cross-fused pelvic kidney with lower moiety renal calculus. His past surgical history to his urinary tract included cloacal exstrophy closure as a neonate, injection of DefluxTM to the bladder neck followed by bladder neck reconstruction and ileocystoplasty.

Examination revealed a reducible mass in the left inguinal region. The remainder of the abdominal examination was unremarkable, with both testes found in the scrotum. Ultrasonography of the groin revealed the appearance of a left inguinal hernia and a cross-fused right ectopic kidney. No hydroureter or hydronephrosis was present. The acute episode was managed conservatively and the patient booked for elective open inguinal herniotomy.

In surgery, the groin was explored through the left half of the existing Pfannenstiel incision and the external oblique aponeurosis was defined. The external oblique was opened and the inguinal canal and its contents were then delineated. A mobile nodular structure was found in the inguinal canal, measuring 2.5 cm by 2.5 cm in size and was found to be reducible proximally (Fig 1). On further dissection, a tubular structure resembling a hernia sac was

identified terminating in the nodule with a thickened tube resembling the vas deferens found intimately attached to the underside. Both structures appeared to descend together in a 'U-shape' formation, terminating in the nodule. On further dissection, the sac-like structure became narrower and more tubular proximally (Fig 2). At this point, suspicion turned to this being a ureter, so it was incised and a feeding tube passed into it. The structure drained clear fluid continuously, confirming it to be a ureter. The nodular structure was excised and the vas-like structure was opened up and catheterised with a feeding tube, whereby it also drained clear urine, showing that it was the distal limb of the same loop of ureter. No hernia sac was seen in the inguinal canal. The ureter was joined back together by spatulating the incised ends and end-to-end anastomosis was performed over a JJ stent, which was removed via cystoscopy six weeks later.

To summarise, the nodule was adherent to the ureter making it long and redundant. The ureter emanated from the retroperitoneum and a loop lay inside the inguinal canal. Kinking and partial obstruction had occurred at the level of the nodule, leading to proximal dilation and appearance similar to a hernia sac. As the whole unit was exposed, without a hernia sac and reducible into the

Figure 1 Left inguinal canal, opened with cord contents displayed.

Figure 2 Left inguinal canal, demonstrating route of two limbs of the ipsilateral ureter originating from the retroperitoneum.

retroperitoneal space, it is classified as an extraperitoneal ureteroinguinal hernia.

Histology of the nodule showed circumferentially aligned smooth muscle fibres with alveolar-like arrangement of glands with double layered epithelial lining. There was central urothelial-lined channel and focal chronic inflammatory cells infiltrate – these findings are suggestive of a genital duct remnant.

Follow-up ultrasound of the renal tract three months after the JJ stent removal showed no dilatation of the upper renal tract and the renal function tests were normal.

Discussion

Ureteral herniation into the inguinal canal is relatively rare amongst adults and even more so in children. There

have been over 140 reported cases, but only five concerning paediatric patients. These were aged 4 weeks, 6 weeks, 4 months, 10 months and 18 months.¹⁻⁵

As this case demonstrates, ureteral hernias are difficult to identify preoperatively. Unless a high index of suspicion exists as to ureteral involvement in the inguinal canal, it is unlikely that any imaging would be employed. It is suggested that urinary symptoms of dysuria, frequency and urgency could suggest a ureteral herniation; however, for this boy and many others, no such symptoms were noted.

Ureteroinguinal hernias are classified into two types: paraperitoneal (80%) and extraperitoneal (20%). Paraperitoneal hernias have a true sac that drags the ureter into the inguinal canal via traction. All five reported paediatric cases were of this type. Extraperitoneal herniations do not have an associated hernia sac and are believed to be related to incomplete differentiation of the ureter from the Wolffian duct or adhesion between the primitive ureter and the genitofemoral ligaments. Extraperitoneal herniation may also be an acquired condition due to prolapse of retroperitoneal fat. Both types of ureteroinguinal hernias are most common in males in the fifth or sixth decade of life, with 46% of patients having associated anatomical urinary tract abnormalities, the most frequent being crossed-fused ectopia and nephroptosis. It is possible that in the paediatric population, paraperitoneal hernias are the only extant variety of ureteric herniation and the extraperitoneal ureteric hernia is an adult phenomenon. So how did our patient present with an extraperitoneal hernia?

We speculate that the patient's multiple pelvic operations weakened the support structures in the vicinity, both abdominal and pelvic muscles as well as retroperitoneal adipose, leading to a weakness that allowed herniation. In this model, the genital duct remnants are connected to the ureter and may function as a lead point, dragging the ureter into the inguinal canal via traction. The gradual nature of the pathogenesis may explain the long and redundant

ureteric length and lack of urinary symptoms or ureteric dilatation on imaging.

The management of ureteroinguinal herniation depends on the individual appearance of the herniated loop of ureter. Most advocate relocation of the ureter back into the retroperitoneum.¹ But in the case of redundancy of ureter or injury, resection and reanastomosis is recommended. End-ureterostomy has been reported for extremely dilated ureters,^{3,4} and ureteric repair can be performed using a psoas hitch, Boari flap, transureteroureterostomy, auto-transplantation and ileal or appendiceal interposition.

Conclusion

This case appears to be the first reported extraperitoneal ureteroinguinal hernia in a child with a history of abdominal operations. Ureteric herniation should be borne in mind when patients with urinary tract malformations and a history of major lower urinary tract surgery present with innocuous hernias. The consequences of ureteral damage are of course extremely detrimental to kidney function if not rectified urgently. An anuric child, post-inguinal repair, should be a warning sign for ureteric or bladder injury.

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