CASE REPORT

Symptomatic ureteric triplex in a child

William Calvert,¹ Dhanya Mullassery,¹ Simon E Kenny²¹Department of Paediatric Surgery, Alder Hey Children's Hospital NHS Trust, Liverpool, UK²Department of Paediatric Urology, Alder Hey Children's Hospital NHS Trust, Liverpool, UK**Correspondence to**Dhanya Mullassery,
drdhanyauk@doctors.org.uk**SUMMARY**

We describe a case of a triplex system in a 4-year-old girl that was associated with a ureterocele and vesicoureteric reflux and causing symptomatic culture-proven urinary tract infections. Undetected on preoperative ultrasound imaging and micturating cystourethrogram, this was discovered at the time of a planned left heminephrectomy. The redundant system having been removed the patient is now asymptomatic.

BACKGROUND

Urethral triplication is a rare anomaly. This case highlights the ease of missing the diagnosis despite imaging. A missed ureteropelvic system left in situ could become the source of recurrent urinary infections.

CASE PRESENTATION

A 4-year-old girl with a history of dysuria and fever and culture proven *Escherichia coli* urinary tract infections (UTIs) was referred to the tertiary paediatric urology clinic. The patient was otherwise fit and well and had no relevant medical history or family history of note. No anomaly had been detected on antenatal ultrasound scans.

INVESTIGATIONS

Preoperative ultrasound reported a left-sided duplex system (figure 1). Cystoscopy revealed a low ureterocele causing bladder outlet obstruction, and that was incised. There was evidence of left-sided vesicoureteric reflux (VUR) on cystoscopy and a micturating cystourethrogram (MCUG). MCUG did not show the duplex system. Dimercaptosuccinic

Figure 1 Ultrasound showing a duplex kidney.

Figure 2 DMSA showing no function in the upper pole of the left duplex.

CrossMark

To cite: Calvert W, Mullassery D, Kenny SE. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2013-202101

Figure 3 Resected bifid upper duplex pole (from a Smith type II triplex).

acid (DMSA) revealed 36% function on the left side with no function in the left upper pole (figure 2).

TREATMENT

Following initial incision of the ureterocele, the patient was asymptomatic on prophylactic nitrofurantoin. However, she developed UTIs again within a month of stopping the antibiotic. Therefore, the decision was made to undertake a laparoscopic heminephrectomy to excise the non-functioning upper pole of her left kidney. Surgical exploration, however, revealed a Smith type II triplex ureter with a bifid upper duplex pole. This was successfully excised (figure 3).

OUTCOME AND FOLLOW-UP

The prophylactic antibiotics were stopped 5 months following the heminephrectomy and she has had no further UTIs.

DISCUSSION

Urethral duplication has an incidence of 0.8%. The triplex system is much rarer. It has previously been reported to be associated with other urological anomalies.

Ureteral buds develop as a diverticulum from the distal part of the mesonephric duct in the fourth and fifth weeks of embryological life. The distal end grows dorsally and then cranially, makes contact with the metanephros and differentiates into the renal pelvis and major and minor calyces.¹ In triplication of ureter, the three ureteric buds could arise independently from the mesonephric duct or from early fission of one or more ureteric buds to join the metanephros. We would presume that the Weigert-Meyer law would be applicable to a triplex system with regard to the positional opening of the ureteric orifices in the bladder relative to the pole of origin.²

The landmark article by Smith³ in 1946 proposed the following classification system (figure 4).

1. Type I: Completely separate ureters where all three ureters open into the bladder or one opening may be ectopic.
2. Type II: Double ureters with one bifid.
3. Type III: Trifid ureters with one ureteric orifice.
4. Type IV: Inverted Y formation of one ureter with a separate ureter.

Since the description, there have been few case reports of triplex systems detailing associations with VUR and ureterocele, both of which were present in our case. Looking at the mechanical basis for these coexistent conditions in duplex systems and the positional finding of the ureterocele on cystoscopy, we can hypothesise that the lower pole ureter with its shorter intramural segment is predisposed to VUR and the upper pole insertion more likely to suffer obstructing pathologies such as ureterocele.² They are uncommon but important developmental anatomical variations to be aware of.

Presenting features include urinary infections, renal colic or incontinence. Ureteteric triplex systems are frequently associated with other urological anomalies including contralateral duplication, ureteral ectopia, renal dysplasia, reflux and ureterocele.⁴⁻⁶

Figure 4 Smith classification of the ureteric triplex.

Surgical intervention is indicated if there are symptoms including pain or recurrent infections due to potential obstruction by ureterocele or reflux in one of the systems.^{5 6} Treatment includes transurethral incision of the ureterocele and partial nephrectomy of the redundant parts of the pelviureteric systems.

Ureteral triplication is a rare congenital anomaly that requires a high index of suspicion for detection at surgery, as the diagnosis may not be evident on clinical features or even radiological investigations.

Learning points

- ▶ Consider the possibility of a third redundant ureter complex during heminephrectomy, especially if concurrent vesicoureteric reflux or ureterocele.
- ▶ A missed ureteric/ureteropelvic system could be left in situ as a source of ongoing urological pathology and morbidity including recurrent urinary infections.

Contributors WC and DM prepared the initial draft of the manuscript. SEK helped complete the manuscript. All the authors have approved the final version of the manuscript.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Benz-Bohm G. Urinary tract embryology, anatomy and anatomical variants. In: Fotter R, edn. *Pediatric uro radiology*. 2nd rev edn. Springer 2008:55–66.
- 2 Schulman CC. The ureters. In: O'Donnell B, Koff SA, eds. *Paediatric urology*. 3rd edn. Oxford: Butterworth-Heinemann, 1997:397–419.
- 3 Smith I. Triplicate ureter. *Br J Surg* 1946;34:182–5.
- 4 Perkins PJ, Kroovand RL, Evans AT. Ureteral triplication. *Radiology* 1973;108:533–8.
- 5 Park BS. Type IV ureteral triplication associated with ipsilateral ureterocele with ureterolithiasis and contralateral duplication with an ectopic ureteral orifice. *Int J Urol* 2008;15:1022.
- 6 Ander H, Ziyilano O, Cayan S, et al. A case of ureteral triplication (type 1) associated with vesicoureteral reflux in a solitary kidney. *Int J Urol Nephrol* 1997;29:537–40.

Copyright 2013 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow