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CASE REPORT

Syringocoele of the bulbourethral duct with additional lower genito-urinary anomalies

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Abstract The ultrasonographic (US) appearance of a syringocoele of the bulbourethral (Cowper) duct, with correlative urethrocystoscopic images, is demonstrated. An infant boy, 5 weeks of age, who presented with *E. coli* infection of the urinary tract also had bilateral hydroureteronephrosis, small bilateral simple ureterocoeles, and posterior urethral valve leaflets.

Keywords Bulbourethral duct · Syringocoele · Ureterocoele · Posterior urethral valve

Introduction

An infant boy presented with *E. coli* infection of the urinary tract. He had bilateral hydroureteronephrosis and several unusual anomalies of the lower urinary tract.

These included syringocoele of the bulbourethral (Cowper) duct that was diagnosed at transperineal US and confirmed at urethrocystoscopy.

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Case report

An infant boy, 5 weeks of age, was diagnosed with a symptomatic febrile infection of the urinary tract. Urine analysis demonstrated bacteria, and there was growth of *E. coli* in blood cultures. Renal US demonstrated dilatation of the renal collecting systems with debris in the left renal pelvis and calyces (Fig. 1). Neither system was duplex. Peristalsis could be observed in both distal ureters, each of which bulged into the bladder as a small ureterocoele (Fig. 2). Jets of urine from both distal ureters were documented; the left ureteric jet was narrow and of high velocity. The wall of the bladder measured 2-3 mm and subjectively seemed prominent on post-void images in spite of a small residual volume of urine. Transperineal US in the sagittal plane revealed a non-compressible fluid-filled mass in the bulbous urethra that tapered proximally to a channel that terminated in the region of the bulbourethral glands (Figs. 3). The posterior urethra measured 3 mm in diameter, an intermediate value compared to normal and obstructed posterior urethras [1].

After discussion, further imaging, specifically voiding cystourethrography, was considered unnecessary; direct visualisation and treatment was planned to follow clearance of the infection. Cystourethroscopy confirmed the presence of a syringocoele of bulbourethral duct, and this was marsupialised (Fig. 3). Posterior urethral valve leaflets were present at 5 and 7 o'clock and these were incised. A tall prostatic urethra consistent with outlet obstruction was noted. The small bilateral ureterocoeles were incised.

Micturating cystourethrography, 6 days following the surgical procedure, showed bilateral vesico-ureteric reflux without evidence of urethral obstruction. There was brief filling and emptying of the syringocoele (Fig. 4).

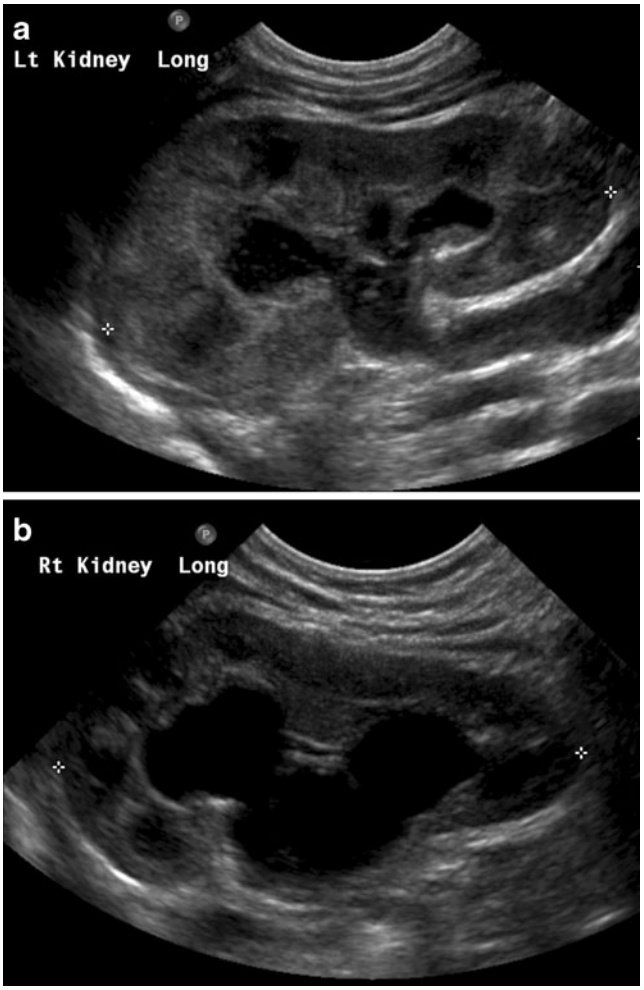

Fig. 1 **a** US image in the long axis of the left kidney demonstrates dilatation of the renal pelvis and calyces with echogenic debris, likely due to infection. **b** US image in the long axis of the right kidney demonstrates hydronephrosis without overt debris within the collecting system

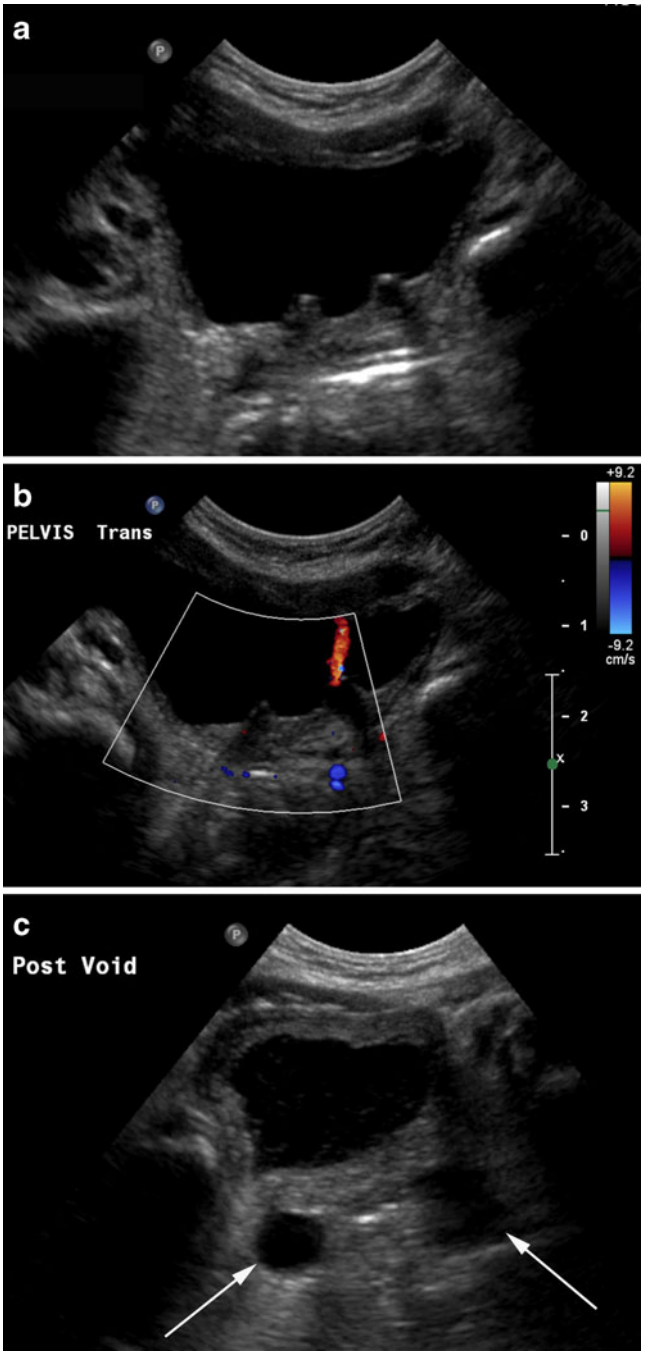

Fig. 2 **a** Transverse scan of the urinary bladder demonstrates small bilateral ureterocoeles. **b** A narrow jet of urine into the bladder is seen from the left ureterocoele. **c** Transverse view of the urinary bladder following micturition. The bladder wall is subjectively thickened, although the pre-micturition thickness was only 2-3 mm. Arrows point to the dilated distal ureters.

Discussion

In males, two bulbourethral glands, are located just inferior to the prostate, and a duct from each enters the anterior urethra. The two main ducts may join and enter the bulbous urethra via a single opening [2]. The glands secrete mucoid material that assists in lubrication and immune defense of the distal genitourinary tract [3]. "Syringocoele" in reference to dilatation of bulbourethral duct is a neologism, but it is the word most commonly used in recent literature. "Retention cyst" of the bulbourethral duct has also been used to describe dilatation [2]. The incidence was reported at 1.5% on cystography performed in 195 boys however, the true incidence is unknown [4, 5]. These lesions have been described as being more common in patients with ductal anomalies; for example, when paired bulbourethral ducts have joined to form a single distal duct [2]. Protrusion of the syringocoele into the urethra can be asymptomatic but may also cause dysuria, recurrent urinary tract infections, haematuria and symptoms referable to obstructed voiding [4, 6].

The etiology of a syringocoele is unclear and may be multifactorial. It has been considered congenital when

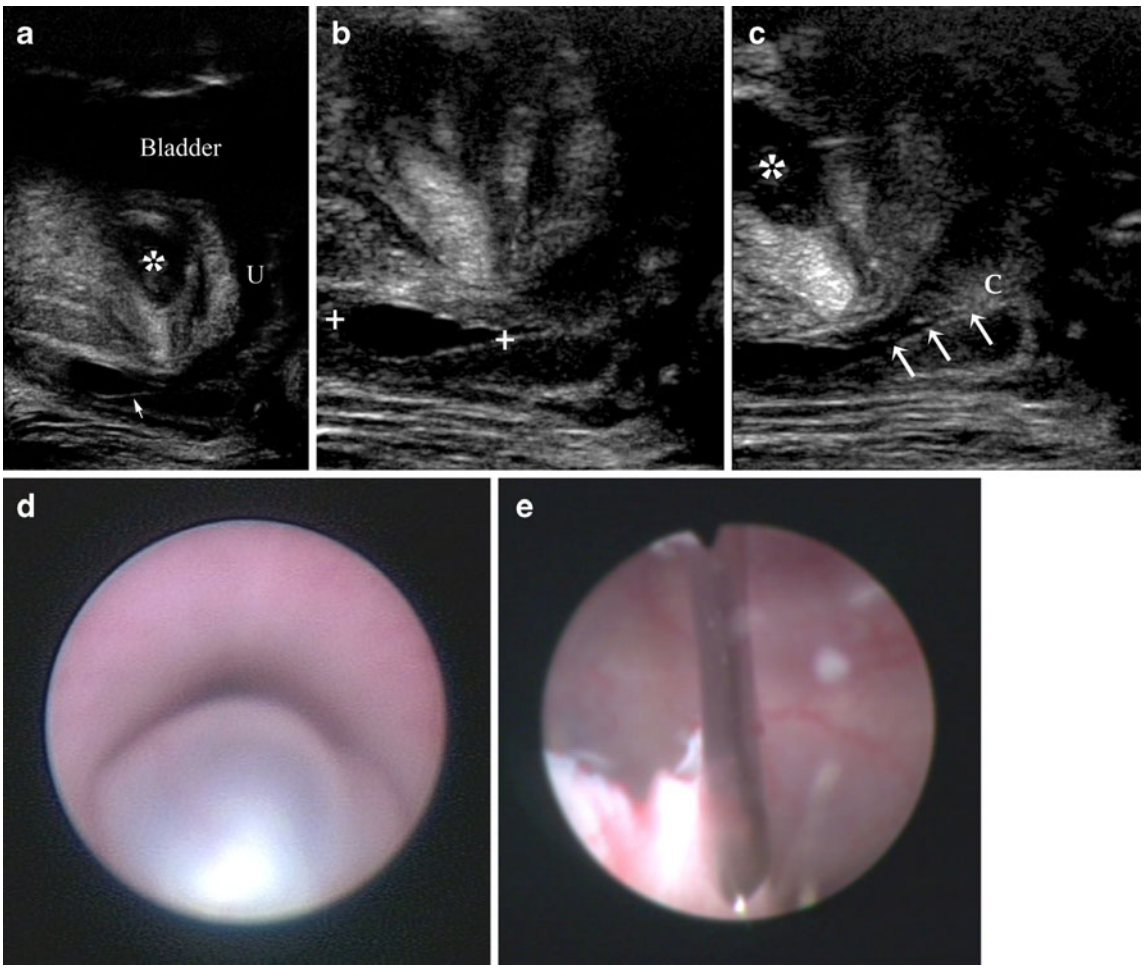

Fig. 3 **a** Midline sagittal transperineal US view (resembling a lateral view at voiding cystourethrography). The patient faces left; the asterisk marks the cartilaginous pubic symphysis. The bladder, posterior urethra (*U*), and the syringocoele (*arrow*) in the anterior urethra are demonstrated. **b** Transperineal sagittal sonogram slightly off the midline shows the syringocoele (between crosses), 1 cm in length. **c** Transperineal sagittal sonogram slightly off the midline is angled to show the proximal bulbourethral (Cowper) duct (*arrows*) and gland (*C*). The asterisk marks the cartilaginous pubic symphysis. **d** Photograph, via cystoscope, of the syringocoele in the anterior urethra. **e** Intra operative urethroscopic view of the incision of the syringocoele

discovered in a neonate. Severe outlet obstruction of the bladder has been reported in newborn infants with the condition. Children with no history of infection or trauma have also been diagnosed as having "retention cyst" [2]. In our patient, we believe that US revealed a congenital lesion. The distal dilatation of the duct was significantly greater than the calibre of the duct as it exited the gland, thereby suggesting malformation rather than acquired obstruction. Syringocoele has been ascribed by some to prior infection or trauma [5].

In the case presented, the mild prominence of the bladder wall identified at US prompted perineal scans of the urethra; this showed a cystic mass with extension to a bulbourethral gland. Reports in the literature discuss the diagnosis of syringocoele with voiding cystourethrography (VCUG), magnetic resonance imaging (MRI), retrograde urethrography, and/or urethrocystoscopy [2–4, 6]. The diagnosis is suggested on VCUG when a smoothly defined mass is identified in the bulbous urethra [6]. Reflux may outline a duct that runs parallel to the floor of the bulbous urethra [2]. Confirmation with MRI has been described in one adult, where a homogeneous non-enhancing cystic lesion with high signal intensity on T2-weighted and short-tau inversion recovery sequences and low signal on T1-weighted sequences, extended from the bulbourethral glands [6].

There have been reports of diagnosis of syringocoele via transperineal US in adults [7]. In children, sagittal transperineal ultrasonograms are generally more helpful than coronal views in establishing the anatomy of the urethra [1, 8]. With the patient lying supine, a high-frequency transducer placed along the mid sagittal plane allows identification of the urethra. In infants, the almond-shaped anechoic cartilaginous pubic symphysis is a good anterior

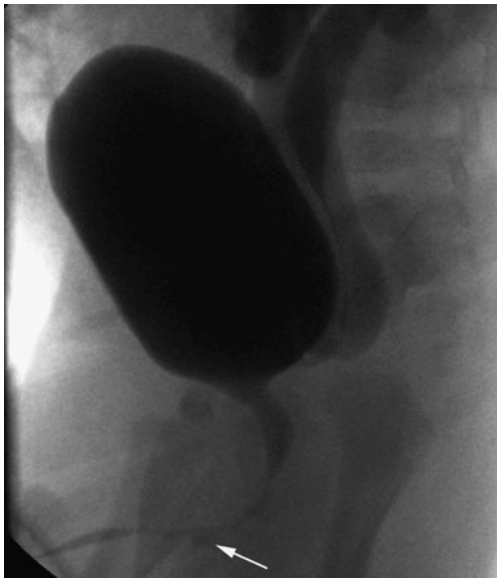

Fig. 4 Last image-hold at postoperative voiding cysto-urethrography demonstrates reflux of contrast (*arrow*) into the residual ectatic duct that parallels the urethra

midline landmark (Fig. 3a) [1]. The orientation of US images similar to images from VCUG and sagittal MRI aids the identification of anatomical structures. US during voiding assists in assessing urethral obstruction [1, 8]; however, timing is challenging. The presence of a catheter prevents visualisation of the anatomy.

The differential diagnoses of a cystic mass in the urethral region include urethral diverticulum, utricular cysts, partial urethral duplication or ectopic ureter [5–7]. Posterior urethral valves may be seen on transperineal ultrasonography as mobile, linear, hyperechoic structures [1]. The posterior urethral valve leaflets were not obvious on US in our patient; the syringocoele captured all the attention at the time.

This case is unusual because there were multiple abnormalities of the distal genito-urinary tract. As far as we are aware, no other such case of a bulbourethral syringocoele with posterior urethral valvelets, and bilateral small simple single system ureterocoeles has been reported. It is tempting to consider the presence of a systemic problem in genito-urinary tubular connections in this infant.

We encourage the use of transperineal US when transabdominal US shows bilateral hydroureteronephrosis and possible bladder outlet obstruction. Similarly, if VCUG demonstrates a urethral mass, perineal US is likely to be helpful in characterising the mass as cystic or solid, and identifying possible sites of origin.

Bulbourethral duct syringocoele is rare, but can be easily treated with transurethral marsupialisation. Follow-up urethrography may demonstrate reflux into the dilated ducts as seen in our case.

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