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Multiple urethral anomalies: Anterior urethral diverticulum, posterior urethral valves, and distal hypospadias

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Abstract We report an infant with antenatally detected bilateral hydroureteronephrosis and a penile cyst who was eventually diagnosed with an anterior urethral diverticulum (AUD), subcoronal hypospadias, and posterior urethral valves (PUV) after birth. To our knowledge, there are no reports where all three anomalies have been found to coexist. As per urethral catheterisation was difficult, cystoscopy-guided catheterisation was performed. On retracting the complete foreskin, a subcoronal hypospadias was identified. The AUD was resected partially, and during a cystoscopy for completion of AUD resection, a dilated anterior urethra was found collapsed, and thin PUV were identified and divided. He remains well at nine months with an improving renal cortical thickness. The vesicoureteric reflux had resolved.
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There are a number of congenital anomalies of the male urethra. An anterior urethral diverticulum (AUD) is a rare congenital anomaly associated with deficiency of the ventral part of the proximal corpus spongiosum, which creates an out-pouching of urethra that fills during micturition and obstructs the urethral flow [1]. The pathology differs from hypospadias which is also associated with defective formation of the distal part of corpus spongiosum leading to a triangular shaped defect of the glans penis and distal urethra, the apex of which is formed by the proximally situated urinary meatus and the base formed by urethral plate [2]. There is also a further deficiency of spongiosum proximal to the meatus. Finally, posterior urethral values (PUV) result from abnormal embryogenesis at the confluence of mesonephric duct and the urogenital sinus membrane around the seventh week of gestation [3].

There have been few reports on AUD in the literature [1,4,5,7–10] with the largest being a series from Turkey [11,12] of about 100 cases.

We now describe a male infant who had a large obstructive AUD, PUV and a coronal hypospadias. This unusual combination does not appear to have been previously reported.

1. Case report

A healthy primigravida woman was referred at 36 weeks of gestation for antenatal counselling due to antenatal ultrasound scan (USS) findings at 20 weeks of gestation of bilateral hydroureteronephrosis, bilateral dysplastic kidneys and a "penile cyst" (suggestive of AUD) (Fig. 1). She had an uncomplicated vaginal delivery at term.

The male infant had poor urinary stream and was referred on the second day of life. The AUD was seen to enlarge

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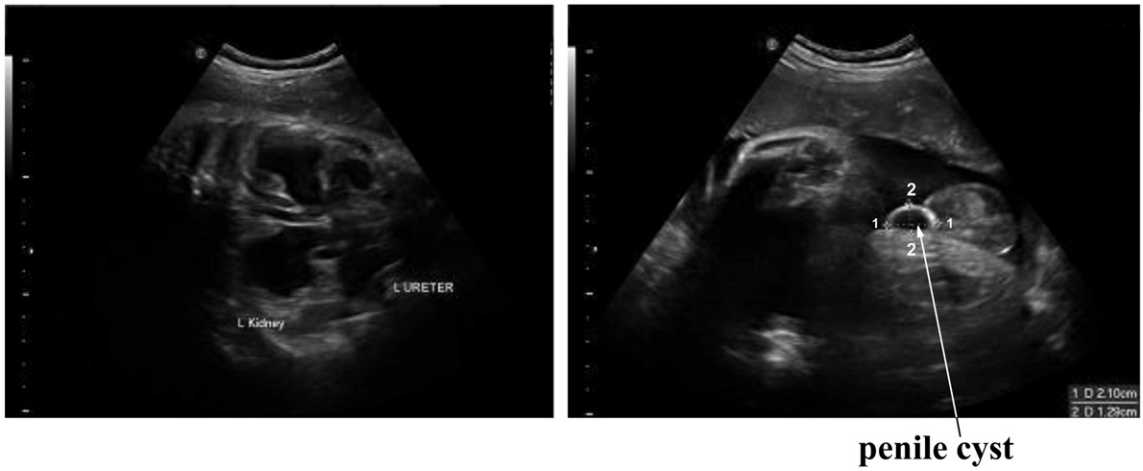

Fig. 1 Antenatal USS of the baby: Renal system and penile lump.

during voiding and appeared to obstruct urine flow. The initial serum creatinine level was 146 µmol/l (normal range µmol 10–50) suggesting significant renal impairment. A postnatal USS confirmed severely dysplastic kidneys with bilateral hydroureteronephrosis and thick walled small bladder (Fig. 2).

The diverticulum and overlying skin were found to be extremely thin and almost transparent. Significant digital pressure was required to compress the diverticulum and all attempts at urethral catheterisation failed.

Suprapubic catheter insertion was not performed because the bladder was felt to be small and therefore, cystoscopy-guided per urethral catheterisation was performed. During this insertion a subcoronal hypospadias was identified within a complete foreskin. Posterior urethral dilatation proximal to the diverticulum was confirmed and an endoscopic resection of the dorsal wall of the diverticulum was commenced, but not completed due to bleeding. A 10 Fr catheter was inserted into the bladder over a guide wire and left to drain. Post operative renal biochemistry normalised.

During the next cystoscopy performed on 17th day of life, the orifice of the AUD was clearly defined. Dilatation of the urethra proximal to the AUD but distal to the posterior urethra had collapsed and PUV were now obvious. The bladder was trabeculated and had a single ureteric orifice on each side. The PUV were resected. The dorsal wall of the AUD was completely incised from proximal to distal end of

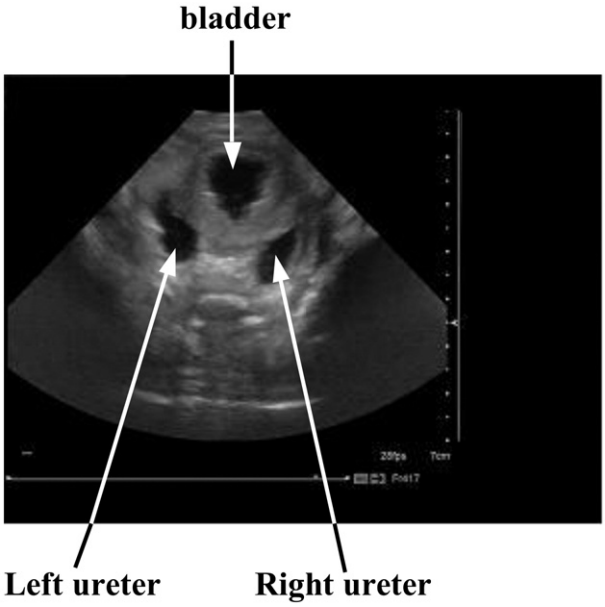

Fig. 2 Postnatal USS abdomen: level of bladder.

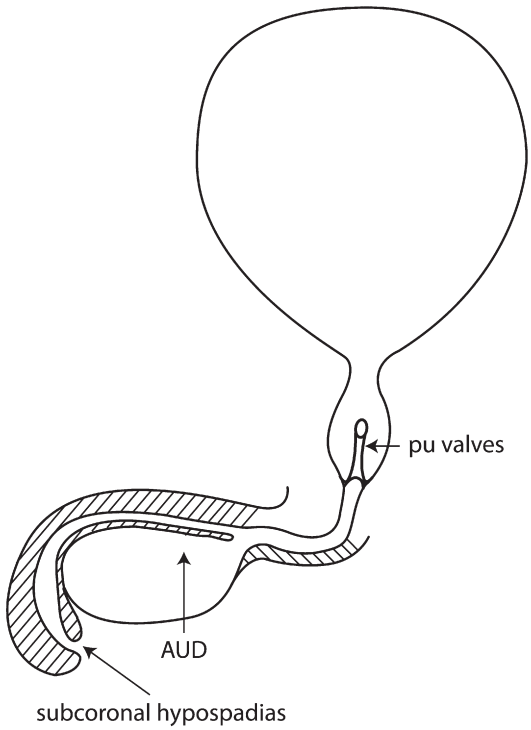

Fig. 3 Patient's anatomy: line diagram (Illustration by Harry Hayes, Department of Medical Illustrations, Central Manchester Foundation Trust).

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