CASE REPORTS

AN ANTERIOR URETHRAL VALVE WITHOUT URETHRAL DILATATION DIAGNOSED BY CYSTOSCOPY IN A NEONATE

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ABSTRACT
Anterior urethral valve (AUV) is a rare congenital anomaly that can lead to severe obstructive uropathy. Early diagnosis and management of AUV is very important to prevent further damage due to vesicoureteral reflux and infection. We present a neonate with febrile urinary tract infection and ultrasonography revealed hydronephrotic left kidney with a distended urinary bladder. Voiding cystourethrogram (VCUG) initially missed an AUV which was picked up on cystourethroscopy. When VCUG was examined again, an AUV without dilatation in the urethra was seen. Transurethral valve ablation was done.

Introduction
Anterior urethral valve (AUV) is a rare cause of congenital obstruction of the male urethra and it is 15-30 times less common than the posterior urethral valve (PUV).¹ AUV may cause a serious obstruction in the distal urinary tract. If left untreated, AUV can also result in end-stage renal disease.² Therefore, it should be promptly diagnosed and managed. We present a neonate with AUV that was missed initially on voiding cystourethrogram (VCUG) but diagnosed on cystourethroscopy.

Case Report
A 26 day-old boy was referred to us due to febrile urosepsis. He was born at 39 weeks of gestation by spontaneous vaginal delivery and had a birth-weight of 3100 gm. On presentation, his weight was 3750 gm, (50% percentile) and the temperature was 38.5°C. He had poor general condition with hypotonia, tachypnea (70 breaths/minute), tachycardia (180 heartbeats/minute), hypertension (systolic-diastolic arterial tension, 60/35 mmHg) and peripheral circulatory collapse (capillary refill time, 5 seconds). Other examination findings were normal. Laboratory investigations showed leukocytosis (white blood cells, 14.3x10³/µL), thrombocytopenia (platelets, 53x10³/µL) and high C-reactive protein (CRP 156 mg/L). Urinalysis showed bacteriuria and pyuria with a positive nitrite result. Biochemical parameters were as follows: serum urea 25 mg/dL, creatinine 0.5 mg/dL, sodium 134 mEq/L, and potassium 5.0 mEq/L. Enterobacter cloaca was isolated from urine (>105 CFU/ml) and blood cultures. Although prenatal ultrasound (USG) screen was reported as normal, current USG showed a distended urinary bladder with grade III hydronephroterephonosis in the left kidney. The craniocaudal length of the left kidney was 50 mm, transverse length 25 mm and the distal ureter diameter of 6 mm and the craniocaudal length of the right kidney was 45 mm, transverse length 30 mm. An indwelling urethral catheter to allow urine drainage was placed. Following intravenous meropenem treatment for 14 days, VCUG scan revealed left-sided grade V vesicoureteral reflux (VUR) without a posterior urethral valve (PUV). Cystourethroscopy showed a valve-like leaflet in the penile urethra arising from the ventral aspect of urethra. When VCUG was examined again, an AUV without dilatation in the urethra was seen (Figure 1). It was treated by the ablation of the valve with electrocautery (Figure 2). Two months after the operation, cystourethroscopy showed no urethral abnormalities.

Discussion
AUV is a congenital mucosal fold located distally to the membranous urethra. It can occur as an isolated entity or in association with a proximal diverticulum. Firlit et al classified AUVs as type 1 which consists of a demonstrable AUV with minimal proximal urethral distention and types 2, 3 and 4 that are associated with urethral diverticulum.³ On the other hand, some authors distinguish between AUV and anterior urethral diverticulum. They especially emphasized the incomplete spongious tissue formation in the anterior urethral diverticulum and normal corpus spongiosum development in patients with AUV.⁴,⁵ AUV have been effectively treated with transurethral valve ablation, whereas the anterior urethral diverticulum usually needs diverticulectomy with urethroplasty. VCUG is the diagnostic modality of choice in the diagnosis of AUV. The urethra generally appears dilated proximal to the valve and narrowed distal to it. A valve may be demonstrated as a linear filling defect along the ventral wall, or it may show a dilated urethra ending in a smooth bulge or an abrupt change in the caliber of the dilated urethra. It can also reveal...
Figure 1. Voiding cystourethrography showed; (a) left-sided grade V vesicoureteral reflux (arrow), and (b) linear filling defect in the penile urethra (arrow)

Figure 2. (a) Cystourethroscopy revealed a valve-like leaflet (arrow) in the bulbar urethra consistent with anterior urethral valve; (b) The endoscopic view of leaflet after transurethral ablation of the anterior urethral valve (arrow)

a thickened trabeculated bladder, a hypertrophied bladder neck, VUR, and urethral diverticula. However, it should be noted that the valves are likely to be missed on VCUG when urethral dilatation is not obvious. In addition, cystourethroscopy usually helps in confirming the diagnosis. In our patient too, the initial VCUG missed the presence of AUV and was picked up on cystourethroscopy. If there are no posterior urethral valves in cystourethroscopy, it would be appropriate to look carefully for the presence of AUV and to re-examine the VCUG.3-5

In conclusion, AUV without urethral dilatation is a rare congenital anomaly of the male urethra which can easily be overlooked during VCUG or cystourethroscopy and may require a re-look.

Compliance with Ethical Standards
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References: