Concomitant anterior urethral valve, distal urethral diverticulum and posterior urethral valve with five-year follow up; A case report and literature review

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ABSTRACT

Concomitant anterior urethral valve and diverticulum (AUVD) and posterior urethral valve (PUV) is an extremely uncommon congenital anomaly that causes infra-vesical obstruction. We present our experience with one case of concomitant AUVD and PUV as well as the related literature review. Early diagnosis and successful management of these anomalies can improve renal function and prevents recurrent urinary tract infections and subsequent renal failure.

1. Introduction

The anterior urethral valve and diverticulum (AVUD) and posterior urethral valve (PUV) are extremely rare congenital anomalies that present with various urinary symptoms. The presentation of urethral anomalies is dependent on the age of the patient and severity of the obstruction.1,2 High-quality oblique view voiding cystourethrography (VCUG) is the mainstay of diagnostic imaging for obstructive urethral pathologies in children.3 Here we present a case of concomitant AUVD and PUV with degenerative changes of the right kidney along with a five-year follow-up and a review of the literature.

2. Case presentation

A 3-year-old male patient with uncomplicated birth history and record of recurrent urinary tract infections (UTIs) was referred to our center by a pediatric nephrologist for evaluation of anatomical urinary tract abnormalities. The patient was admitted to the hospital on his seventh day of life due to urinary retention. Per-urethral catheterization was failed so vesicostomy was placed. The ante-grade VCUG demonstrated dilated urethra, suggestive of AUV. Endoscopic fulguration of AUVD was done at 6-month-old and vesicostomy was closed one month after the AUV repairing surgery. Kidney-ureter-bladder ultra-sonographies were recommended as post-operative changes of the right kidney along with a five-year follow-up and a review of the literature.

The patient was discharged with no complications the day after the operation.

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The urethral catheter was removed one week later, after voiding per urethra was noted with good caliber. Follow-up was every 3 months in outpatient’s clinic for one year and then annually on regular basis except during the COVID pandemic. VCUG was included in the first and fifth year follow-up sessions as demonstrated Fig. 1 B and C.

On the last follow-up visit, a magnetic resonance urography (MRU) and 3-dimensional virtual cystoscopy were performed for further evaluation of right kidney function and bladder. The MRU depicted atrophied right kidney with severe cortical loss, in accordance with previous results, normal-sized left kidney with normal cortical thickness; bladder diverticulum in the anterior portion with 24*15mm diameter; and angulation at the distal part of the right ureter at the UVJ (Fig. 2A, B & C). The above-mentioned findings required no further intervention.

### 3. Discussion

Among several theories explaining the embryologic basis of developing concurrent anterior and posterior urethral anomalies, the theory of delay in mesenchymal tissue migration and abnormal absorption of the Wolffian duct could clearly explain the mentioned concurrency. The incidence of AUVD is 10- to 30-times lower than PUV. The concomitant occurrence of anterior and posterior urethral valves is extremely rare. To the best of our knowledge, this is the sixth case of concomitant AUD and PUV and the second case of concomitant AUV, AUD, and PUV as reported in Table 1. This case is reported as another experience in the successful management of a rare congenital urethral anomaly in the literature in the hope of consensus on the management of concomitant AUVD and PUV.

Precise cystourethroscopy would be a valuable tool for diagnosing and managing urethral anomalies. Abbreviations

**Abbreviations**

| Abbreviation | Description |
|--------------|-------------|
| AUVD         | Anterior urethral valve and diverticulum |
| PUV          | Posterior urethral valve |
| AUV          | Anterior urethral valve |
| AUD          | Anterior urethral diverticulum |
| VCUG         | Voiding cystourethrography |
| UTIs         | Urinary tract infections |
| AP           | Anterior-posterior |
| DMSA         | Dimercaptosuccinic acid |
| DTPA         | Diethylenetriamine pentaacetic acid |
| UVJO         | Ureterovesical junction obstruction |
| MRU          | Magnetic resonance urography |

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**Fig. 1.** Voiding phase VCUG: A. Pre-operative voiding phase oblique-view VCUG which was performed at 3-year-old revealing AUVD and PUV, B. voiding phase of VCUG in the first year of follow-up revealing response to the endoscopic fulguration with no signs of AUVD and PUV, C. voiding phase AP view VCUG in the fifth-year follow-up revealed no signs of AUVD and PUV. The asterisk shows PUV, the short arrow shows AUV and the long arrow shows AUD.

**Fig. 2.** MRU and virtual cystoscopy: A. MRU view of the urinary tract in the fifth year follow-up demonstrating atrophied right kidney, normal-sized left kidney and angulation at the distal part of the right ureter at the UVJ. B. MRU view of the urinary tract in the fifth year follow-up demonstrating atrophied right kidney with severe cortical loss, normal-sized left kidney with normal cortical thickness and angulation at the distal part of the right ureter at the UVJ. C. Right lateral view of virtual cystoscopy of the bladder in the fifth year follow-up revealing bladder diverticulum in the anterior portion with 24*15mm diameter. The arrow shows the bladder diverticulum in the right lateral aspect of the bladder; the asterisk shows the right ureter orifice. D. Cystourethroscopic view of type II PUVs. E. Cystourethroscopic view of hypertrophied bladder neck indicating modified BNI intervention.
| Source | DOI | age presentation | Ultrasonography | VUR | Renal scan management | comment |
|-------|-----|------------------|-----------------|-----|-----------------------|---------|
| 1 | 10.1016/S0022-5347(00)00024-3 | 0-4 d | Urinary retention, posterior urethral valve, posterior urethral diverticulum, thickened bladder wall | Doppler of the left kidney, left hydronephrosis, thickened bladder wall | High creatinine | Resolved by surgery |
| 2 | 10.1016/j.purol.2020.11.002 | 1 y | Antenatal hydronephrosis, urinary tract infection, obstructive uropathy | - | - | - |
| 3 | 10.1095/aud.2012.059455 | 2 d | Straining during voiding, dribbling, thickened bladder wall | Bladder wall thickness | Left high-grade reflux | - |
| 4 | 10.1095/aud.2012.032914 | 18 mo | Weak stream, straining during urination | Bilateral severe hydrourephrosis, thickened bladder wall | Left high-grade reflux | - |

**Table 1**

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**Table 2**

Concluding remarks: The early surgical repair can prevent recurrent febrile UTIs and subsequent renal insufficiency. A precise cystourethroscopy and high-quality oblique view VCUG are the cornerstones of diagnosing urethral anomalies. In the matter of urethral anomaly, urologists must bear in mind the complete evaluation of the urinary tract.

**Informed consent**

Written consent was obtained by the authors from the legal guardian of the patient prior to submission.

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**Declaration of competing interest**

The authors declare no conflict of interest.

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