

Delayed Presentation of Posterior Urethral Valve with Enuresis

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ABSTRACT

Posterior urethral valve (PUV) is the commonest cause of lower urinary tract obstruction in male infants. Persistent obstruction can lead to obstructive uropathy and renal impairment. PUVs can be diagnosed in-utero using antenatal ultrasound and postnatally. There is a varied mode of presentations among infants with PUV. This 12-year old boy presented with enuresis due to overflow incontinence from chronic urinary retention. The diagnosis was confirmed with micturating cystourethrogram (MCUG) and he subsequently had valvotomy using Mohan's valvotome. This unusual presentation underscores the spectrum of clinical findings associated with PUVs. It adds to the small, but growing, body of literature on delayed PUV presentation and emphasizes the importance of a high degree of clinical suspicion when evaluating boys of any age with LUTS and unusual urologic complaints.

Keywords: Urethral valve, Uropathy, Incontinence, cystourethrogram.

INTRODUCTION

Posterior urethral valves (PUV) are the commonest cause of lower urinary tract obstruction in male infants with an incidence of 1:5,000 male infants and 1:25,000 live birth. They cause obstruction to the antegrade flow of urine from urinary bladder to the exterior. When the obstruction persists for a long time, the back pressure on the kidneys can lead to obstructive uropathy and renal failure.

PUV can be detected by antenatal ultrasound or postnatally. The presentation in the post-natal period

varies greatly in severity. Occasionally, the diagnosis is missed until adolescence or adulthood when the patient presents with lower urinary tract symptoms.

This congenital abnormality of proximal urethral development is thought to be due to Wolffian duct persistence or congenital obstructive posterior urethral membrane, and is usually associated with a high morbidity including urosepsis, urinary incontinence (over-flow), chronic renal insufficiency, and even death.

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This case is being reported because of the atypical presentation of enuresis that was not thought to be due to posterior urethral valve by parents, resulting in late presentation.

RESULTS

A 12-year-old boy, who was referred to our facility with a history of enuresis of five years duration. He has a history of recurrent intermittent fever and dysuria which was treated with antibiotics. On examination, he was a healthy-looking boy with stable vital signs. Abdominal examination revealed a non-tender suprapubic mass of about 5cm from the pubic symphysis, which was dull to percussion. He has normal external genitalia. A diagnosis of chronic urinary retention secondary to possible posterior urethral valve was made. Initial management included continuous per urethral catheter drainage of the bladder with size 8 Foley's catheter under strict aseptic condition. Empirical antibiotic therapy was commenced using levofloxacin. Investigations done included urinalysis, Full blood count, serial blood electrolytes, urea and creatinine, urine microscopy, culture, and sensitivity (M/C/S). Initial radiological investigations consisted of abdomino-pelvic ultrasound, which revealed pre-voidal volume of 511mls and post-voidal urine volume of 249mls with thickened bladder wall (1.16-1.26cm.) and features suggestive of PUV. It also showed that both kidneys were enlarged in size for patient's age. They measured 11.3 x 6.9 cm and 12.2 x 7.1 cm for the right and left, respectively. Corticomedullary differentiation of both kidneys were fair with slight increase in parenchyma echogenicity was observed. There was moderate dilatation of the renal pelvis and calyces, blunting of the fornices and flattening of the papillae with mild cortical thinning observed on both kidneys, suggestive of grade III hydronephrosis as shown in figures A and B.

The diagnosis was confirmed with Micturating cystourethrogram (MCUG), which showed dilated and elongated posterior urethral as shown in figure C. Ultrasound scan revealed "keyhole sign" as shown in figure D.



Figure A: Left kidney showing grade III hydronephrosis.

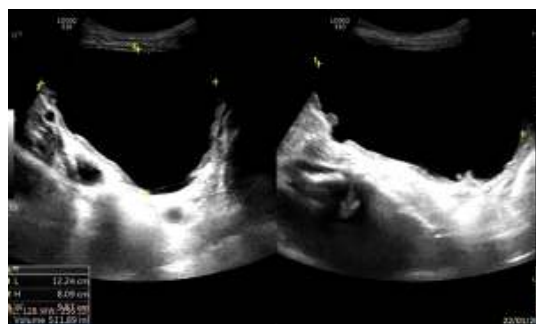


Figure B: Post-voidal residual urine volume before valve ablation.

Urinalysis results is as shown in table 1. Urine M/C/S revealed no bacteria growth. The full blood count and renal function tests were normal. The initial bladder drainage was 1000mls of turbid urine. He subsequently developed post



Figure C: Post Micturition film showing dilated PUV (straight arrow) with markedly dilated posterior urethra (elbow arrow).



Figure D: The keyhole sign in the patient.

obstructive diuresis with initial urine output at 5-6ml/kg/hour and urine specific gravity (SG) showing

Table 1: The urinalysis test results of the patient.

Variable	Result
Leucocyte	++
Blood	+
Pus cells	++++
Erythrocyte	+
Epithelial cells	+
Protein	++

hyposthenuric urine with a specific gravity of 1.005 but became normal after 72-hours with urine SG at 1.015. He was placed on continuous bladder drainage for one week after which he had avulsion of the valves using Mohan's valvotome. A silicon urethral catheter was left in-situ for three days to allow the edema to subside and to enable measurement of urinary output. He voided with good flow and repeat ultrasound showed a PVR of 34mls as shown in figure E.

DISCUSSION

In the past, before the advent of antenatal ultrasound, late presentation of PUV was considered a good prognostic sign, suggestive of lesser degrees of obstruction. Nowadays, late presentation has been reported to be uncommon. This is not the same in our

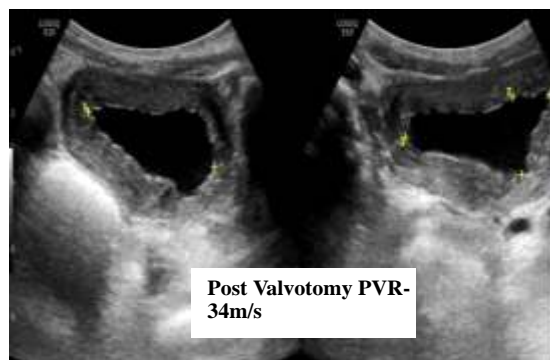


Figure E: Insignificant post-voidal residual urine after valve ablation.

environment where late presentation is still the norm. Odetunde et al. in Enugu Nigeria, reported that the bulk of patients seen with PUV in their practice presented late with urosepsis, anemia, uremia, and ESRD and he noted that 71% of patients presented with renal failure.(10)

The median age at presentation in our environment as reported by Odetunde and colleagues' in a study of 21 patients with late presentation of PUVs, was 3 years. The late presentation has been attributed to the fact that most developing countries including Nigeria compared to developed nations has low level of antenatal care. (10) A study carried out in Nigeria revealed that heavy financial cost of ultrasound scanning, negative perception of patients towards ultrasound scanning, unsatisfactory previous scan experience, long distance to service providers, and long waiting periods have been identified as major obstacles to prenatal diagnosis. The cause of delayed presentation in this index case could be attributed to ignorance on the part of the care givers and poor health seeking behaviour. Symptoms of late presentation of PUVs include: recurrent urinary tract infections, poor urinary stream, urinary frequency, nocturnal enuresis, urinary incontinence, anemia, and voiding pain or dysfunction incontinence, and sexual dysfunction. . The main symptom at presentation of this patient

was enuresis. This is in tandem with the report by Odutola and colleagues who reported enuresis in 47.6% of patients presenting late with PUV. This can be linked to overflow incontinence due to chronic retention in a decompensated urinary bladder resulting in nocturnal enuresis. Schober et al in evaluating the outcome of late-diagnosed PUV ablation, showed that 67% of patients with PUV presenting late had nocturnal enuresis. (15) The patient developed persisted nocturnal enuresis up to 12-years of age which became a cause of concern to both parents whose other siblings achieved continence at an earlier age. Pathological conditions are superimposed on late PUV, such as decompensated bladder with chronic urinary retention and overflow incontinence, sphincter-bladder dyssynergy and other differential diagnosis of PUVs like neurogenic bladder make the diagnosis of PUV a diagnostic dilemma in late presentation. The history of features consistent with recurrent UTI and findings of a palpable bladder mass that was not tender (CUR) raised the index of suspicion of PUV.

PUVs are now commonly diagnosed by the postnatal evaluation of infants who had prenatal hydronephrosis. The initial diagnostic tool in patients with suspected PUVs is renal and bladder ultrasound scan and often reveals a thickened bladder wall, bilateral hydronephrosis, elevated PVR volume, as well as a dilated posterior urethra. An MCU confirms these findings and may also demonstrate bladder trabeculation, a circumferential filling defect at the level of the pelvic floor, prominence of the bladder neck, diverticula, and VUR. This patient's PUV was confirmed with a voiding cystourethrogram as shown in figure B. This can also show bladder diverticulum and reflux if present. In a study by Ozen and colleagues' of 17 boys diagnosed with late-presenting PUVs (mean age 7.35 years), PUVs were observed by MCUG in 10 (58.8%) patients, consisting of 9 with a

dilated posterior urethra. Reflux was detected in 9 (52.9%) patients. Urethrocystoscopy was not done to confirm PUV in this boy because of non-availability of pediatric endourologic facilities in our centre. This would have also given first-hand information on post-obstructive bladder changes, confirming the presence of pop-off pressure mechanisms, such as bladder diverticulum, which helps in the preservation of renal function in this patient.

In the study, *Escherichia coli* accounted for 66.7% of the urinary tract infection (UTI) and 14.3% did not grow any organism. *Pseudomonas aeruginosa* was found in 4.8% of the urine culture of patients with UTI while 14.3% also grew *Klebsiella*. There was no organism cultured in this boy. This may be due to prolonged use of antibiotics in treating recurrent UTI. Elevated serum creatinine level has been shown to be associated with poor prognosis in PUV patients, and this may not be reversed even with the relief of the obstruction. Interestingly, this 12-year old boy had normal serum creatinine despite obstructive uropathy. The reason for this is not immediately apparent in this patient. However, it may be because of the large volume capacity bladder (decompensated), which helped to take off pressure from the kidneys. Other factors which may show poor prognostic are: include bilateral reflux, echogenic kidneys, loss of corticomedullary differentiation, sub cortical cysts (indicative of renal dysplasia). Though, this 12-year-old boy does not have some of the stated poor prognostic indices, a follow-up by the paediatric nephrologist was arranged due to the obstructive nephropathy at presentation as evidence by proteinuria. In addition, he reported improvement in enuresis after the valve ablation. This was however not surprising as the PVR after valve ablation as shown in figure E was insignificant and eliminates overflow incontinence.

This is supported by a study by Schober et al who showed that 68% of patients with PUV establish good bladder emptying after valve ablation despite late presentation. (15)

This unusual presentation underscores the spectrum of clinical findings associated with PUVs. It adds to the small, but growing, body of literature on delayed PUV presentation and emphasizes the importance of a high degree of clinical suspicion when evaluating boys of any age with LUTS and unusual urologic complaints.

CONCLUSION

Enuresis can be a pointer to the diagnosis of PUV in patients who present late especially in our environment. Therefore, a high degree of clinical suspicion is required when evaluating boys at any age with LUTS and uncommon urologic complaints.

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