Posterior Urethral Valve with Anterior Urethral Diverticulum, A Rare Cause Of Lower Urinary Tract Obstruction - A Case Report

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ABSTRACT

INTRODUCTION: Congenital anterior urethral diverticulum (CAUD) is a rare cause of urinary obstruction in children. Its association with posterior urethral valve (PUV) is an exceedingly unusual occurrence. We hereby report a case of 3 years male with straining on micturition as stated by the mother underwent micturating cystourethrogram (MCUG) revealing a posterior urethral valve (PUV) plus a diverticulum in the region of anterior urethra. Valve ablation was done for PUV. Open diverticulectomy and urethroplasty was done for the diverticulum. This case report represents a rare event in which two congenital causes of bladder outlet obstruction are combined.

KEYWORDS: Congenital Anterior Urethral Diverticulum, Posterior urethral valve, Urethral Diverticulum

INTRODUCTION:

Congenital anterior urethral diverticulum (CAUD) is an extremely rare entity in children and is defined as an outpouching of the anterior urethra through the corpus spongiosum Clinical presentation varies depending on the age and severity of urinary obstruction. It includes a poor urinary stream, post-void dribbling, recurrent urinary tract infections (UTI), and ventral penile swelling[1]. Urethral diverticulum is defined as epithelialized, saccular dilatation that is separate from the urethra but communicates with the lumen through a discrete orifice.

Radiologically two varieties have been described: saccular and globular[2]. A detailed clinical examination and imaging studies, such as MCUG, help in diagnosing the condition. Treatment options include either watchful waiting in minor cases, or endoscopic, or open surgical excision depending on the symptoms, type or size of the diverticulum, and upper urinary tract changes [3, 4].

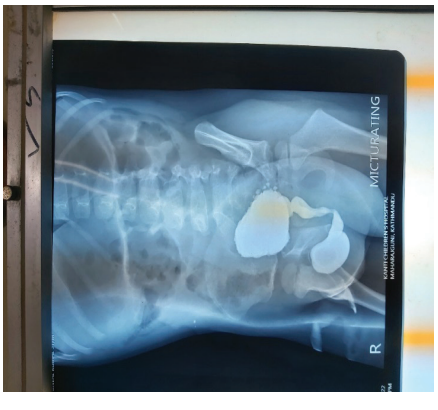
We report a rare association of congenital anterior urethral diverticula with posterior urethral valve in a 3 years boy with review of literature. We report a rare combination of the two anomalies in one.

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CASE SUMMARY:

A 3 years male was bought by her mother in the pediatric surgery OPD of Kanti Children's Hospital with complaints of thin and weak urinary stream, straining on micturition and post micturition dribbling since last 1 year. There was one episode of fever in between. Physical examination revealed a globular bulge over the penoscrotal area. Systemic examination was within normal limits.



(Fig.1). There was no reflux seen in MCUG.

Urine routine microscopic and culture was positive for UTI for which antibiotic was given. USG abdomen was sent which revealed bladder wall thickening suggestive of cystitis. MCUG was sent which revealed a dilated posterior urethral valve and a diverticulum as an outpouching in the junction of anterior and some part of bulbar urethra (Fig.1). There was no reflux seen in MCUG.

He was planned for cystoscopy, fulguration of PUV and excision of the urethral diverticulum. In the operation theatre cystoscopy was done under GA where there was a brilliant transillumination over the area of the diverticulum (Fig 2). The anterior urethra revealed a large distal urethral diverticulum approx.6 cm length with its proximal shelve till the external meatus as measured with the cystoscope.

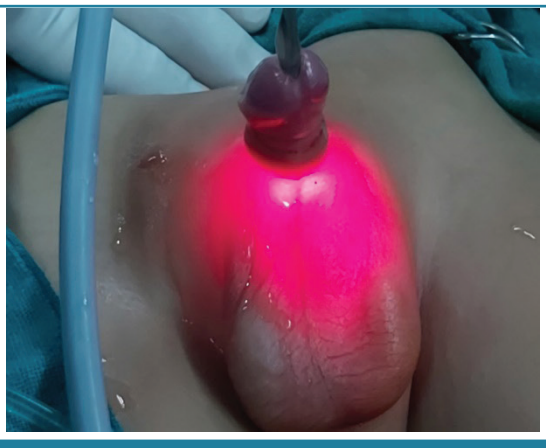


Fig.2 Examination of diverticula under

Next step was to do a fulguration of the PUV. After the fulguration of the PUV the patient was positioned for urethroplasty. The incision was given over the scrotal median raphae. The diverticulum was identified and after its isolation it was opened longitudinally and excised (Fig 3). The urethra was repaired (urethroplasty) with 6-0 size absorbable suture using the dartos flap imposition in two layers over an 8Fr. Foleys catheter. Postoperative period was absolutely uneventful and on the 10th postoperative day catheter was removed and the patient was discharged. Follow up MCUG was done which showed a good result

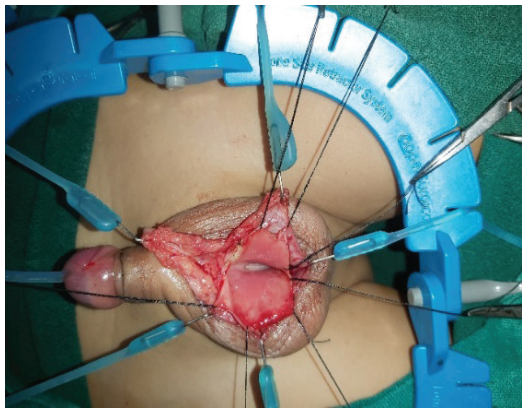


Fig.3 Diverticulum isolated and opened

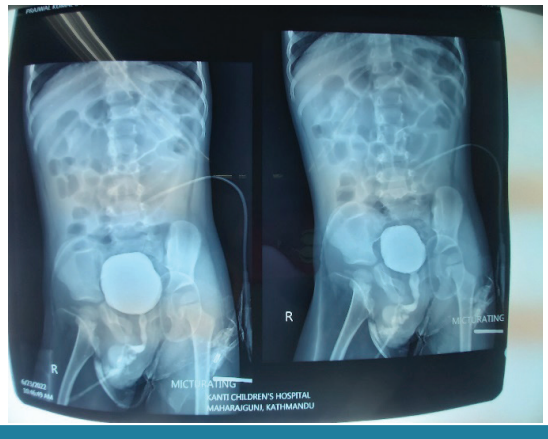


Fig.4 Post Surgery MCUG

DISCUSSION:

Lower urinary tract obstruction in children can occur due to diverticulum or PUV. It can be presented an isolated entity or together with a proximal diverticulum; illustrating the spectrum of the disease [5]. Its association with PUV is a rare event with only 2 cases previously reported [6]. The etiology of this condition remains unclear. It has been suggested that the lack of a corpus spongiosum results in urethral dilatation leading to a diverticulum [7]. The age of presentation for CAUD is variable based on the degree of obstruction to the urinary flow. The reported age of presentation in literature ranges from antenatal diagnosis to adulthood.

In the two case reports of children with PUV and CAUD, both patients presented at neonatal age which was a bit earlier than our patient [6]. In the majority of the cases, it is located at the penoscrotal junction while in one-third of the cases it occured distally [7]. Various proposed hypotheses have been described which can range from "an abortive attempt at urethral duplication, failure of alignment between the proximal

and distal urethra, imbalanced tissue growth in the developing urethra leading to a remnant of excess tissue acting as a valve and congenital cystic dilatation of periurethral glands leading to a flap-like valve [1]. AUVs can occur at the level of the bulbar urethra (40%), pendulous urethra (30%) and penoscrotal junction (30%) [5]. In our case the diverticulum was at the junction of bulbar and penile urethral area. Some children may present antenatally with an obstructive picture on antenatal ultrasound but the majority present postnatally with lower urinary tract symptoms including difficulty in micturition, dribbling of urine, poor urinary stream, and/or UTI. In severe cases, they may present with renal failure and gross upper tract changes [7, 8,].The symptoms were similar in our case with weak urinary stream, straining on micturition and post micturition dribbling.

MCUG can also show a dilated urethra with abrupt caliber change or ending in a smooth bulge [9]. MCUG serves as the diagnostic modality in the diagnosis of AUV. It can reveal a urethral caliber change such as the dilated or elongated posterior urethra, dilatation of the anterior urethra, and urethral diverticulum. Apart from these it shows thickened trabeculated bladder, a hypertrophied bladder neck and vesicoureteric reflux [9]. In our case the patient had bladder outlet obstruction with thickening of bladder suggestive of cystitis due to long standing obstruction. However there was no evidence of VUR in our case.

Cystourethroscopy is beneficial in confirming the diagnosis which would precede the treatment that comprises of the destruction of the AUV and PUV by electro cautery or by a resecting hook[10], like we did in our case. Treatment of CAUD depends on the size of the diverticulum and the degree of obstruction. Small and asymptomatic lesions may be followed by surveillance alone. Various treatment options for symptomatic diverticula include open diverticulectomy and urethroplasty and endoscopic resection [11]. Our patient was treated with open diverticula excision and urethroplasty which was also used in the report by Kumar and et al[12]. In our case repair was done using two layers of dartos flap over an 8 Fr catheter.

CONCLUSION

PUV with urethral diverticulum is a rare combination occurring as cause of bladder outlet obstruction in a child. Imaging studies, such as MCUG, can help in the diagnosis of the condition. In case of large diverticulum open diverticulectomy and primary repair are recommended.

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