Urology Case Reports 34 (2021) 101447

Contents lists available at ScienceDirect

Urology Case Reports

journal homepage: http://www.elsevier.com/locate/eucr

Pediatrics

Congenital anterior urethral diverticula with posterior urethral valve: A rare combination, case report☆

Hana Abebe *, Hiwote Girma, Fisseha Temesgen

Addis Ababa University, Ethiopia

ARTICLE INFO

Keywords:
Congenital anterior urethral diverticula
Posterior urethral valve
Diverticulectomy
Case report

ABSTRACT

Background: Congenital anterior urethral diverticulum is a rare cause of urinary obstruction in children. Its association with posterior urethral valve is an exceedingly unusual occurrence.
Case presentation: 18 month old male child for whom cystoscopic valve ablation was done for posterior urethral valve continued to have obstructive symptoms for which VCUG was done and revealed congenital anterior urethral diverticula. Open diverticulectomy and urethroplasty was done and he was discharged improved.
Conclusion: This case report represents a rare event in which two congenital causes of bladder outlet obstruction are combined and the presence of one masquaders the diagnosis of the other.

Background

Congenital anterior urethral diverticula represent a cystic dilatation of the anterior urethra and is one of the rare causes of bladder outlet obstruction in children. Its embryologic basis is debated.[1] It arises from the penile urethra in third of cases and radiologically two varieties have been described: saccular and globular.[2]

Clinical presentation varies depending on age and the severity of urinary obstruction. It includes poor urinary stream, post void dribbling, recurrent UTI and ventral penile swelling which is decompressed upon manual compression. The imaging studies used for diagnosis include ultrasound, VCUG, Computed Tomography and Magnetic Resonance Imaging. There are reports of antenatal diagnosis of CAUD.

The treatment options for CAUD are variable and guided by the presence of symptoms, upper urinary tract changes and the size of diverticula. Watchful waiting may be justified for patients without symptoms and upper urinary tract obstructive changes. Transurethral resection is the treatment of choice for small, well-supported diverticula while open diverticulectomy and urethroplasty with or without flap imposition or marsuplization is recommended for large diverticula.

In this report we describe a rare association of congenital anterior urethral diverticula with posterior urethral valve with review of literature. We report a rare combination of the two anomalies in which the proximal, more severe anomaly prevented the initial expression of the other.

Case summary

18 month old male child presented with poor urinary stream and straining during urination which was noticed since 2 month of age. He was also repeatedly treated for febrile UTI at a rural health facility. At presentation to our facility, he was febrile and tachycardic with a palpable bladder for which urethral catheter was inserted with difficulty and 100 ml urine was drained.

On further workup he had leucocytosis with left shift, serum creatinine of 0.8 mg/dl and many WBC on urine analysis. Abdominal ultrasound revealed bilateral severe hydroureteronephrosis with thickened bladder. After stabilization VCUG was done which revealed a trabeculated bladder with dilated posterior urethra and left high grade reflux. Cystoscopy was done with a finding of trabeculated bladder and Type I PUV which was ablated at 5,7 and 12 o'clock and he was discharged on the 3rd postoperative day (Fig. 1).

Two weeks later he presented to the emergency department with difficulty of urination and febrile UTI for which he was catheterized with difficulty and treated with IV antibiotics. Check cystoscope revealed anterior urethral outpouching otherwise there was no remnant valve. At the end of the procedure urethral catheterization was difficult and deferred. The patient developed acute urinary retention in the immediate

Abbreviations: CAUD, Congenital anterior urethral diverticula; PUV, Posterior urethral valve; UTI, Urinary tract infection; VCUG, Voiding cystourethrography.

☆ Study Area: Addis Ababa University, College of Health Sciences, School of Medicine, Tikur Anbessa Specialized Hospita.

* Corresponding author.
E-mail addresses: hanaabebe23@gmail.com (H. Abebe), hiwotesc@gmail.com (H. Girma), bemafish@gmail.com (F. Temesgen).

https://doi.org/10.1016/j.eucr.2020.101447
Received 9 September 2020; Received in revised form 5 October 2020; Accepted 8 October 2020
Available online 12 October 2020

post-operative day in the wards for which catheterization was tried but unsuccessful. Thus, he was taken to the operating theatre and vesicostomy was done. He was discharged improved subsequently.

On further follow up at outpatient department the mother noticed swelling at the ventral penis which was evacuated upon manual compression. VCUG was repeated and revealed dilated anterior urethra with diverticula of the bulbar urethra, collapsed posterior urethra and resolution of the vesicoureteral reflux (Fig. 2). Abdominal ultrasound at this time also showed bilateral mild hydronephrosis (Fig. 3).

Cystourethroscopy showed anterior urethral diverticula and through median raphe scrotal incision, the anterior urethra was approached and there was a diverticulum of about 4 cm length involving the bulbar and membranous parts. The diverticulum was opened longitudinally and excised. Urethroplasty was done in two layers over 8 Fr tube with a dartos flap imposition. Post operatively, catheter was kept for 10 days and he was discharged improved and currently waiting for vesicostomy closure.

Discussion

CAUD has been described to occur in association with various urologic and non-urologic anomalies like vesico-ureteral reflux, anterior urethral valve, penile torsion, patent ductus arteriosus and polydactyly. Its association with posterior urethral valve is a rare event with only 2 cases previously reported.[3,4]

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 reports of children with PUV and CAUD, both patients presented at neonatal age which was a bit earlier than our patient.[3,4]

The diagnosis of CAUD was initially missed in our patient as the ventral penile swelling and anterior urethral dilatation were not revealed prior to posterior valve ablation due to poor urinary flow distally. This was also the case in a report by Kumar and et al.[4]

Although the presence of persistent urinary obstruction after posterior valve ablation usually suggests incomplete valve ablation, it is also good to bear in mind the rare obstructive defects in the anterior urethra as a differential diagnosis. Check cystoscopy and voiding

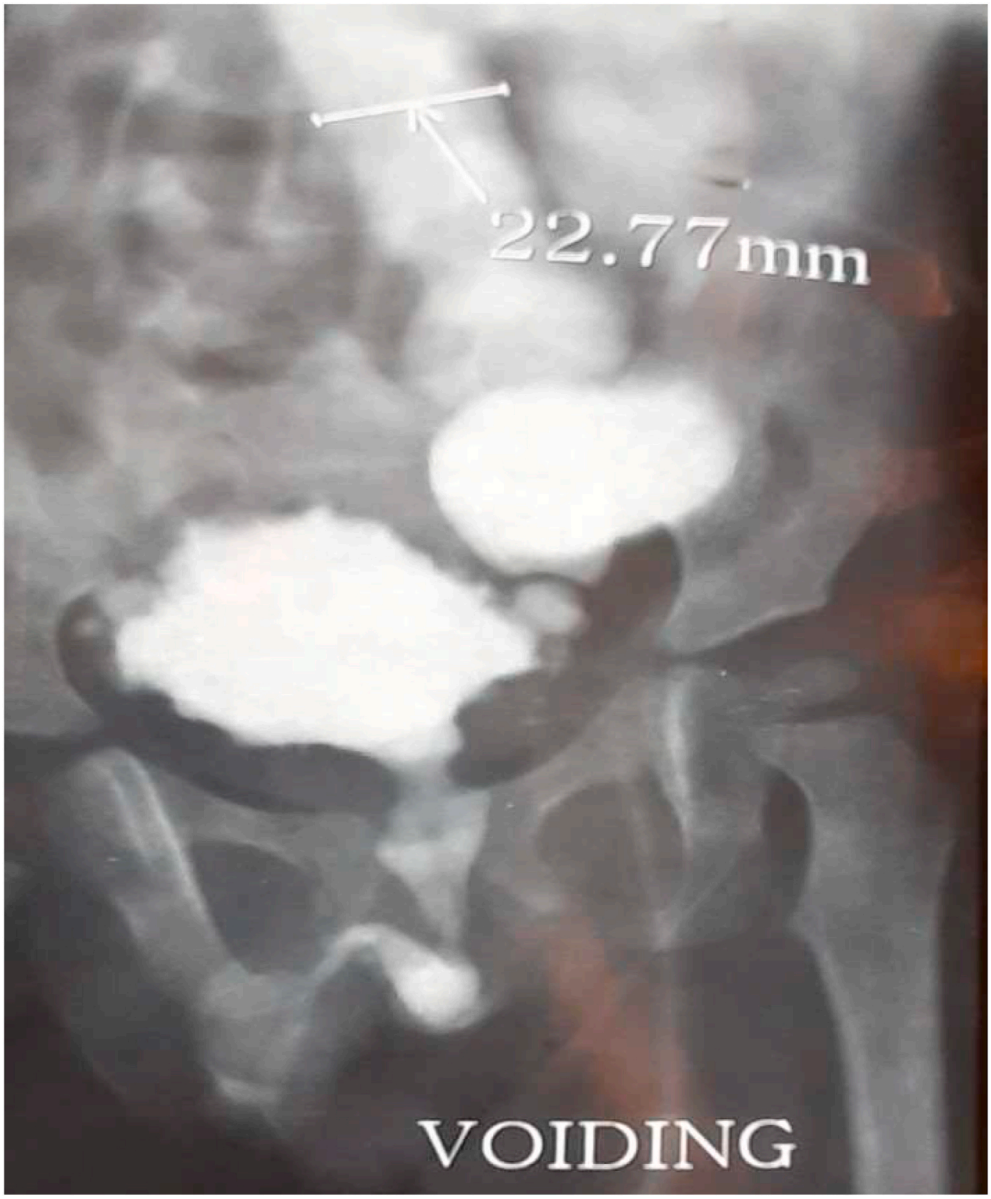


Fig. 1. Preoperative voiding cystourethrogram.

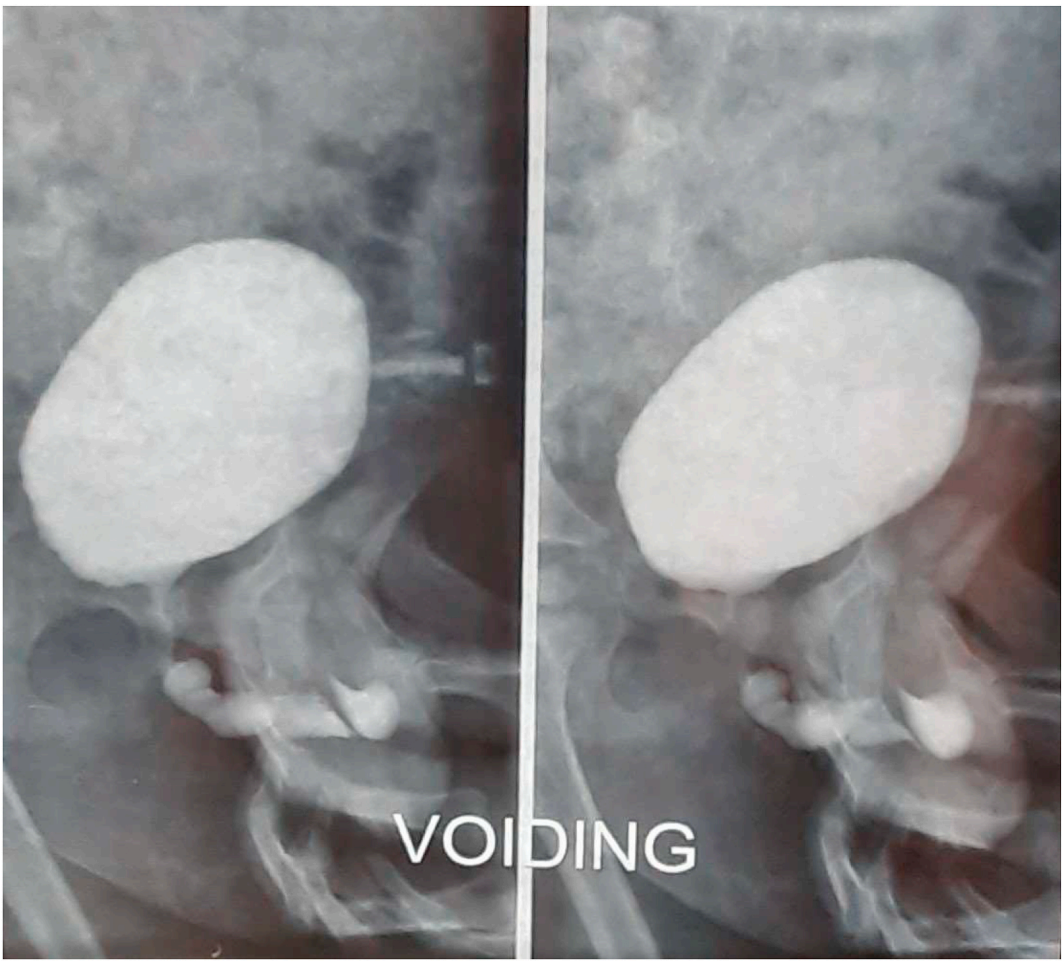


Fig. 2. Post posterior urethral valve ablation voiding cystourethrogram.

cystourethrography are helpful in differentiating among these causes.

The use of vesicostomy in stabilizing the patient with persistent urinary obstruction post PUV ablation and associated congenital anterior urethral diverticula was effective in our patient as evidenced by improvement of the serum creatinine (from 0.8mg/dl to 0.2 mg/dl), decrement in the degree of hydronephrosis on follow up abdominal ultrasound, reversal of obstructive bladder changes and resolution of vesicoureteral reflux on voiding cystourethrography. Temporary urinary diversion by vesicostomy was also used in a report by Kumar and et al.[4]

Treatment of congenital anterior urethral diverticula 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.[5] Our patient was treated with open diverticula excision and urethroplasty which was also used in the report by Kumar and et al.[4]

Ethics approval and consent to participate

Written informed consent was obtained from the parent for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Availability of data and materials

The datasets with more images and patient data are available from the corresponding author on reasonable request.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Authors' contributions

FT was primarily involved in the management of the patient. HAG and HG were also involved in the management of the patient and follow up. HAG wrote the case summary and the case report with review of the literature. All authors have read and approved the final case report.

Registration of research studies

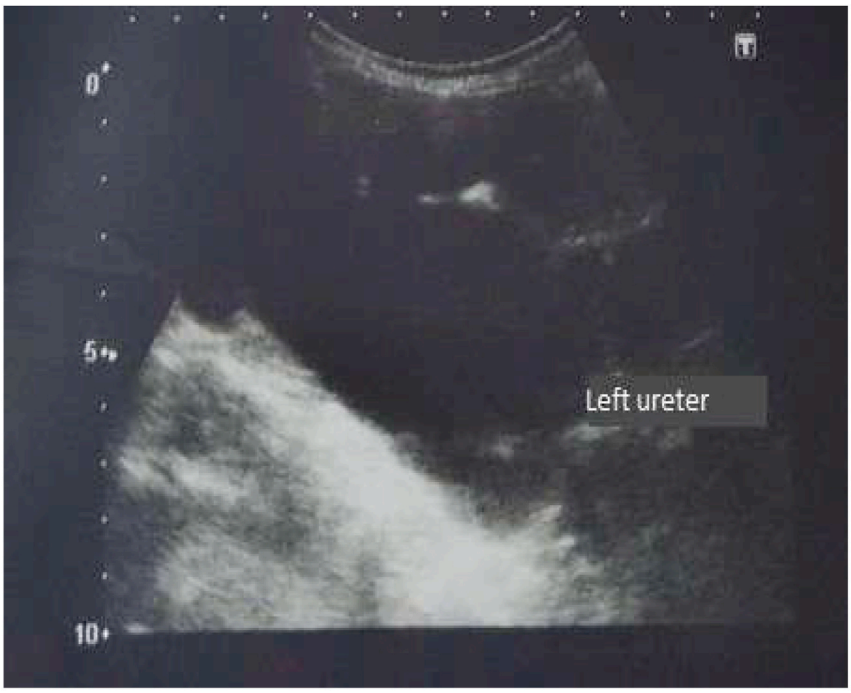
Registry not required as this is not a first-in-man case report.

Guarantor

Dr. Hana Abebe.
Department of Surgery, Paediatrics surgery unit.
Addis Ababa University, Ethiopia.

Provenance and peer review

Not commissioned, externally peer-reviewed.



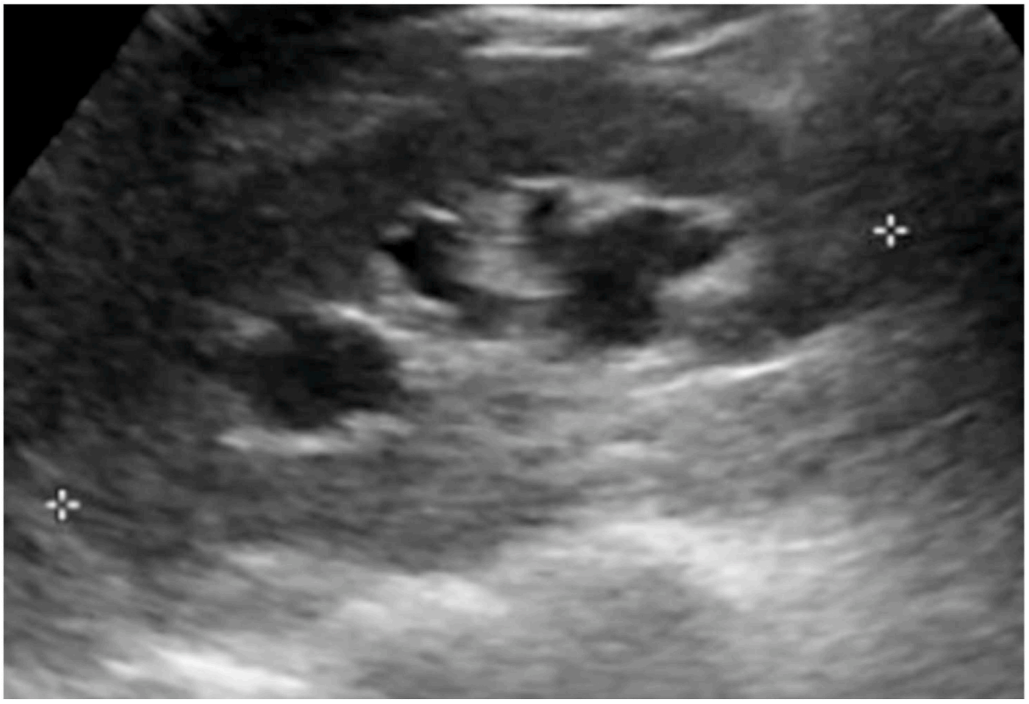


Fig. 3. Renal ultrasound images pre and post posterior valve ablation.

Declaration of competing interest

The authors declare that they have no competing interests.

References

1. Kadian YS, Rattan KN, Singh Mahavir, Kajal Pradeep. *Congenital Anterior Urethral Diverticulum in Children: A Case Report and Review ISRN Surgery*. 2011, 120307.
2. Gupta DK, Srinivas M. Congenital anterior urethral diverticulum. *Pediatr Surg Int.* 2000;16:565–568.
3. Graham jr Sam D, Krueger Ronald P, Glenn James F. Anterior urethral diverticulum associated with posterior urethral valves. *J Urol.* August 1982;128.
4. Kumar A, Bajpai M, Gupta AK. Double urethral obstruction in a neonate –A case report. *Eur J Pediatr Surg.* 2005;15:449–451.
5. Zia-ul-Miraj M. Congenital anterior urethral diverticula in children. *Pediatr Surg Int.* 1999;15:567–569.