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Duplication of Urethra with Primary Vesico-Ureteric Reflux in Solitary Kidney: A Rare Case Report

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Abstract

Urethral duplication is very rare congenital anomaly with ill defined etiology. Very few cases have been reported in literature till date. Patients may present with urinary incontinence, abnormal or dual urinary stream, recurrent urinary tract infections and sometimes associated penile deformity. Aim of presenting this case report is to limelight one of such rare presentations. Herewith reporting a case of 16 months old baby boy presented with history of recurrent urinary tract infection, solitary kidney with grade 4 primary vesico-ureteric reflux and duplication of urethra. Diagnosis and treatment plan in such cases require a multistage approach.

Keywords

Urethral Anomalies, Urethral Duplication, Congenital Anomalies

1. Introduction

Many theories have been put forward explaining the etiology for Urethral duplication like abnormal embryological closure of the Mullerian canal, incomplete mesodermal closure, ischemic injury during embryogenesis and abnormalities during development of urogenital sinus [1]. Patient presents with different presentations including recurrent urinary tract infections to penile deformities [2]. Urethral duplication is a rare urinary anomaly in children. So far, only 300 cases have been reported in literature [3]. Efmann classified urethral duplication in 3 types. There are many surgical reconstructions which have been explained depending upon the type of urethral duplication. Herewith reporting a case of 16 months old baby boy presented with history of recurrent urinary tract infection, solitary kidney with grade 4 primary vesico-ureteric reflux and duplication of urethra.

2. Case Report

16 months old baby boy was referred with complaints of recurrent urinary tract infections treated symptomatically with antibiotics. On examination there were two urethral orifices passing urine from both urethral meatus. On sonography abdomen there was evidence of solitary right side kidney with dilated lower ureter 9 mm and bladder was showing early changes of cystitis. On Micturating cystourethrogram (MCU), there was evidence of duplication of distal urethra.

(Type 1 b as per Efmann classification) with grade 4 vesico-ureteric reflux in solitary kidney (Figure 1). Renal scan showed mild scarring with maintained renal function on right side, Left side kidney showed no uptake of dye considering absent kidney. Patient was planned for surgical correction of duplication of urethra with ureteric reimplantation on right side. Laboratory examinations revealed a normal complete blood count (CBC) and creatinin level.

Patient was catheterised with infant feeding tube no 7 through normal urethral meatus, which was clearly visible through accessory urethral meatus (Figure 2). Accessory urethra was cored out along the tract after complete degloving of penis and excised completely (Figure 3). Primary wound closure was done along with glanspalsty same as done for glandular hypospadias repair over infant feeding tube no 7 (Figure 4). Skin flaps rotated. In the same sitting Cohens ureteric re-implantatios done for solitary right side kidney with grade 4 vesico-ureteric reflux with open approach through pfannenstein incision. 3 french DJ stent was kept in situ. Postoperative intravenous antibiotcs were given for 5 days and was discharged on per urethral catheter in situ for 10 days. After 10 days on follow up per urethral catheter was removed. There was no evidence of wound dehiscence and no urinary leak from urethral wound. After 4 weeks cystoscopic DJ stent removal was done. Patient was kept on caliberation with infant feeding tube no 7 at home for next one month after removal of catheter. On follow up of one year baby is passing urine in good stream from primary urethral meatus

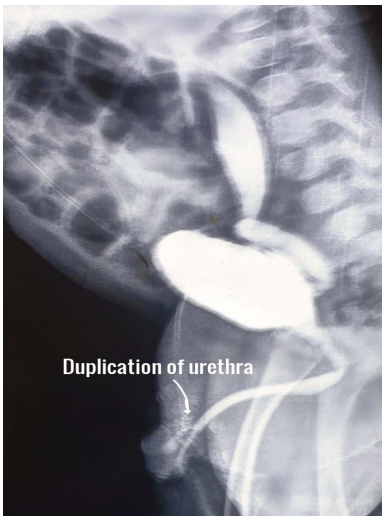

Figure 1. MCU with duplex urethra.

Figure 2. Post catheterisation.

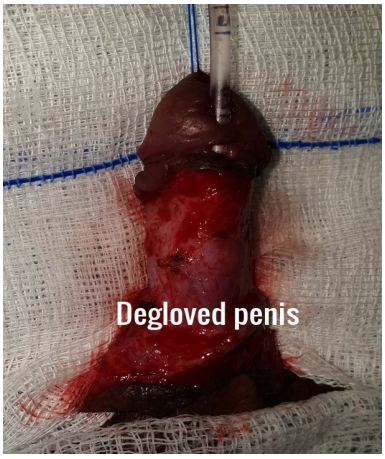

Figure 3. Degloved penis.

Figure 4. After surgical repair.

with good force and without straining. Further follow up of micturating cysto-urethrogram and renal scan for ureteric re-implantation is awaiting. But till date there is no breakthrough of urinary tract infection.

3. Discussion

Various classification methods have been proposed for a better understanding of urethral duplication. The most commonly used classification system is the Effman classification [4]. According to Effman classification, urethral duplications are divided into three main groups, as Type I, II, and III (**Figure 5**). Type IA is the most common type. This anomaly may be associated with penile deformities, other urinary anomalies but association with vesico-ureteric reflux is very rare to encounter with [5]. Complete evaluation of patient is important part of the diagnostic process. Despite this incidentally urethral duplication diagnosis can be made during circumcision and/or hypospadias surgery. Radiological Imaging tests should definitely be used for confirming the diagnosis. The main diagnostic imaging procedures include voiding cysto urethrography, intravenous pyelography, ultrasonography (USG), retrograde urethrography, and MRI [6]. Doppler ultrasonography is useful for prenatal diagnosis of urethral duplication by evaluating fetal micturation [7]. Micturating cystourethrogram helps in knowing more about anatomical course, shape, diameter and relationship with urinary bladder of both urethra. MRI and CT scan help in diagnosing the accompanying genitourinary and gastrointestinal abnormalities (including solitary kidney, Mullerian anomalies, duplicated colons and double anuses etc.) [8]. MRI provides excellent information on urethral width and length, and structure of periurethral soft tissues [9]. Few authors recommend no therapy for asymptomatic cases [10]. But in view of incontinence, obstructive symptoms, double micturation, recurrent infection and cosmetic concerns are among indications

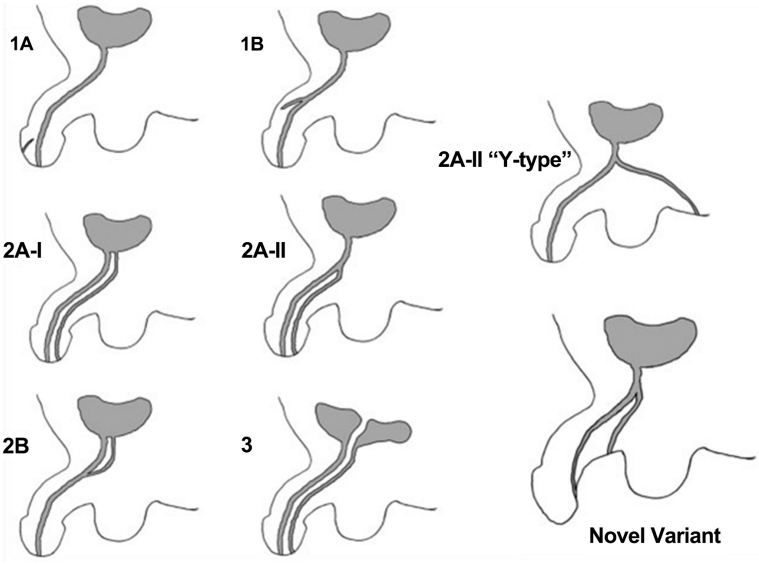

Figure 5. Effman classification for urethral duplication.

for surgery [11]. Treatment options for urethral duplication include follow-up without a specific therapy, serial urethral dilatation for well draining urethra among the primary and accessory urethra, perineal urethrostomy and surgical operations such as urethroplasty surgery [12]. A treatment plan is based on the anatomical type of the duplication. Further follow up is needed to watch for urinary tract infections, urinary stream and if associated with reflux then renal scans to assess renal functions.

4. Conclusion

Urethral duplication is a rare cause of recurrent urinary tract infections in children and associated with other urological anomalies, too. Exclusive plan of treatment should be explained to the parents exhaustively. For complete evaluation sometimes anesthesia is also required though diagnostic cystoscopy may miss the findings, other radiological imaging as mentioned above may help to delineate the course completely.

Disclosure

An informed consent has been taken from parents to publish this case report for academic purpose. I declare no potential conflict of interests, real or perceived.

Conflicts of Interest

The author declares no conflicts of interest regarding the publication of this paper.

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