Case Report

A case study of duplication of urethra

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Abstract Urethral duplication is a rare congenital anomaly; either isolated or associated with other anomalies, it presents with two different urine outlets, frequent episodes of Urinary tract infection. These conditions should be promptly managed by appropriate surgical intervention. We have reported here one case of such rare anomaly detected by micturating urethrogram.
Keywords: urethra.

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INTRODUCTION

Urethral duplication is a rare congenital anomaly with approximately188 cases described in literature[1]. These anomalies most commonly are seen in males however few cases are also reported in females. Embryogenesis of this condition is not clear. Several embryological theories have been postulated. Cassel man and Williams[2]stated that a partial failure or the growth of the lateral mesoderm between the ecto-dermal and endo-dermal layers of the cloacal membrane in the midline, accounts for the formation of dorsalepispadiac channel. Das andBrosman[3] reported that abnormal termination of the mullarian duct was responsible for urethral duplication. Rica *et al.*[4] suggested that asymmetry in the closure of theuro-rectal septum results in urethralperineal fistula. Inspite of the numerous theories proposed to explain this anomaly, no single theory explains allthevarious types of anomalies. Urethral duplication (UD) is a rare anomaly with varied clinical manifestations such as deformed penis, twin streams, urinary tract infection (UTI), urinary incontinence, serous discharge from sinus, out flow obstruction and associated anomalies.[5–8] It may be complete or incomplete. The accessory urethra often presents as dorsal or ventral midline openings and rarely opens eccentrically (Coronal/collateral). Dorsally opening accessory urethra is the most common type.

CASE REPORT

We report here a case of urethral duplication in 5 yrs. old male Patient who had a history of frequent Urinary Tract Infection, urine outflow through two separate openings, and difficulty in micturation. The X-ray :Cysto-urethrogram (Fig.1 to Fig. 4) showed- that there was duplication of urethra starting from mid penile urethra Opening separately – one at normal site- situated at tip of penis and second at base of penis above scrotum . Patient was treated for the UTI and was advised surgical Correction.

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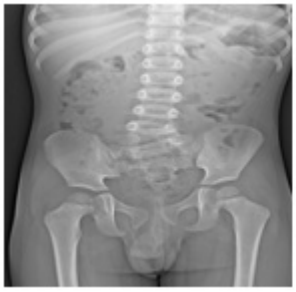

Figure1:

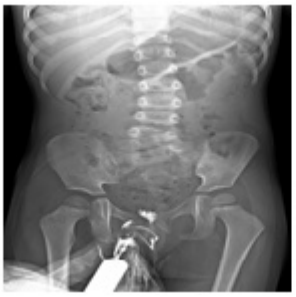

Figure 2:

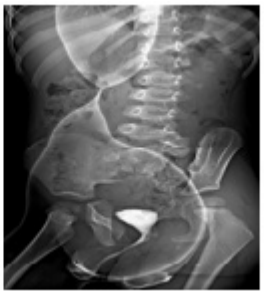

Figure 3:

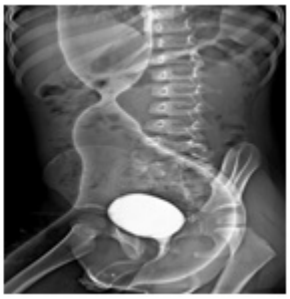

Figure 4:

DISCUSSION

Urethral duplication is a rare anomaly with about 188 cases reported till date, usually seen in males and often associated with genitourinary and gastro-intestinal anomalies.[5,6,8] Embryogenesis of urethral duplication is not well understood and various hypothesis exist.[5,8–9] The components of the male external genitalia are developed from the cloacal folds, labioscrotal swelling, urogenital sinus and preputial swelling and is composed of all 3 germ layers with timely interaction of Sonic hedgehog signals, fibroblast growth factors, Hox genes, bone morphogenetic proteins signals and androgens.[10,12] According to most studies, which are histology based, the male urethra develops mostly or completely from the endoderm[11] except the distal most glanular part which is of ectodermal origin.[12] According to Hynes *et al*., the entire urethra develops from migration and fusion of the paramedian folds of mesoderm sub-epithelially. This starts proximally and moves distally and involves 4 pairs of folds which are urethral, frenular, preputial and lacuna. This is preceded by cleavage of cloacal membrane leading to the formation of the urethral groove[13]. The commonly quoted and accepted hypothesis for complete urethral duplication is that of Patten and Barry and is supposed to result from an abnormal relationship between the lateral folds of the genital tubercle and the ventral end of the cloacal membrane.[8,12] But this does not explain all subtypes of urethral duplication. Failure of fusion of mesodermal bands or growth of mesoderm around 2 urethral anlages, duplication of cloacal membrane and defective mesenchymal proliferation around the cloacal membrane could explain the clinical presentations of our case of diphallus child.[14,15] Sometimes urethral duplication can also be associated with bladder duplication.[16] Complete duplication of bladder and urethra is more commonly associated with anomalies of other organ systems including gastrointestinal anomalies and vertebral anomalies. Campbell proposed splitting of the vesico-urethral anlage as the cause, suggesting that whether the split occurs before or after division of the cloaca by the urorectal septum determines if rectal anomalies will be present.[16] While there may be overlapping features among genital duplication and the urorectal malformation sequence, no genetic basis has been identified.[17] Patients with urethral duplication are usually asymptomatic, except for a urinary double stream and cosmetic problems. However, it has also been reported to be detected in the prenatal ultrasound findings of a male foetus,[18] and it can also present as recurrent urinary tract infection in an adolescent boy [19] and as bladder outlet obstruction in an adult male.[20] While some patients may opt for conservative management for the complete duplication of urethra; as it can sometimes only give rise to relatively mild symptoms such as double urinary stream, our patient opted for early urethral reconstruction as the pre-op voiding curve showed mild obstructive pattern, which may potentially be due to the narrowed urethral opening at the dorsal side. Furthermore, the associated anterior megalo-urethra may also have posed as an infection risk due to urine stasis and it was cosmetically unappealing.

CONCLUSION

Urethral duplication is a rare congenital anomaly; either isolated or associated with other anomalies, it presents with two different urine outlets, frequent episodes of Urinary tract infection. These conditions should be promptly managed by appropriate surgical intervention.

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