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High grade vesicoureteral reflux in hypospadias: Case report of a rare association and review of literature

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ABSTRACT

The Hypospadias represents a common urogenital anomaly amenable to surgical correction. Vesicoureteral reflux (VUR) are though may be found associated with hypospadias but it is unusual to have a severe VUR along with several episodes of high grade fever and culture positive urinary tract infections. We are presenting a rare association of high grade vesicoureteral reflux with febrile urinary tract infections (UTI's) with subcoronal hypospadias. We also reviewed the available literature on this rare association and present its work-up and management.

Key Words: Hypospadias, vesicoureteral reflux, meatal stenosis, urinary tract infection.

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Introduction

Hypospadias is one of the commonest urological abnormalities with an incidence of one in every 300 male births [1]. Surgery is the mandatory treatment of hypospadias which is done mainly at early age for various psycho-social reasons. Hypospadias may be associated with other urological anomalies like inguinal hernia or undescended testes [2]. Urethrocutaneous fistulas, urethral stricture and meatal stenosis are the common problems faced in patients with hypospadias in the postoperative period. Routine imaging of the upper urinary tract in not recommended [3,4]. Despite some controversy, anomalies are rarely detected in renal ultrasonography in hypospadias patients [3,4]. It is common to have vesicoureteral reflux (VUR) in pre / post operated cases of hypospadias that too asymptomatic and are of low grade (ie. Grade I and II). The VUR has been reported in approx. 17% of hypospadias patients [5,6]. It is extremely uncommon to have febrile urinary tract infection (UTI) associated with high grade VUR (grade V) in hypospadias cases before surgery. The causes of VUR in hypospadias patient are not clear but due to high risk of post-operative meatal stenosis / urethral stricture with impaired flow of urine may be the possible causes for aggravated VUR in these patients.

We present a case of such an extremely rare combination of associated anomaly (i.e. high grade VUR with febrile UTI's associated with

subcoronal hypospadias) in a 5 year old boy. As far as we know, this is the first example of such a relationship.

Case report

We report a case of 5 years old boy, referred to our hospital (Dr. Ram Manohar Lohia Institute of Medical Sciences, Lucknow, U.P. a tertiary teaching referral center) with complaint of urinary opening on the under-surface of the penis since birth and gradual narrowing of stream with frequent episodes of high grade fever and difficulty in micturition. On examination – he was having subcoronal hypospadias with downward bending of penis and glandular tilt with dorsal preputial hood and good urethral plate with bilateral testes descended (Fig. 1). The meatus was narrow but can be stented with 6 Fr infant feeding tube. At the time of admission he was having 2-episodes of high grade unrecorded fever, pain in supra-pubic region with difficulty in micturition and intolerance of food.

Fig. 1. Sub-coronal hypospadias with 8 Fr catheter in the meatus.

He was admitted and urinary catheterization with 6 Fr infant feeding tube was done and he was started on IV antibiotics. Urine for culture and sensitivity was sent which grew Klebsiella pneumonia with significant colonies. He was started on culture sensitive IV antibiotics (Piperacillin and Tazobactum) with which he stabilized. He was evaluated with ultrasonography of a kidney, ureter, and bladder (KUB) that showed bilateral hydroureteronephrosis with thinned out renal parenchyma and irregular trabeculated urinary bladder. After the repeat urine c/s showed no growth, he was evaluated with micturating cystourethrogram (MCU). MCU showed bilateral grade V- VUR with irregular shaped bladder mucosa (Fig. 2).

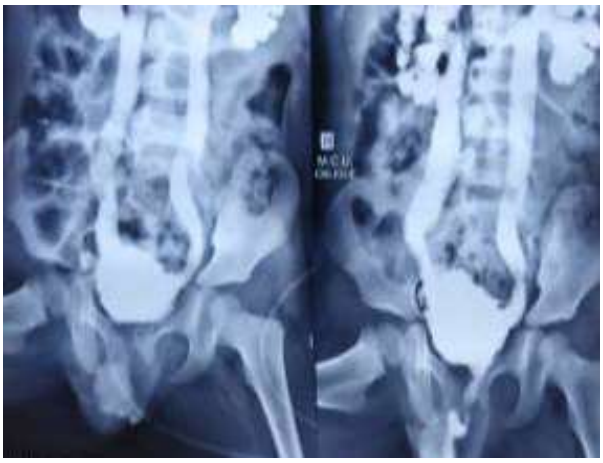

Fig. 2. Micturating cystourethrogram (MCU) of the patient showing high grade reflux.

He underwent check cystoscopy which showed bladder severely trabeculated with both ureteric orifices were dilated. Bladder was filled with dense muck due to which nothing was clearly visualized, so he was given twice bladder wash with normal saline. Bladder neck was normal. Posterior urethra was normal. No posterior urethral valve and rest of the anterior urethra was also normal. He is under follow-up and planned for dimercaptosuccinic acid (DMSA) scintigraphy and subsequent ureteric reimplantation and hypospadias repair.

Discussion

The prevalence of VUR in children with UTI is found to be around 30% compared with 0.4-1.8% in healthy children with 2.2% in girls and 0.6% in boys [7,8]. In hypospadias, it has been reported in approximately 17% patients [5,6]. There could be several contributory factors associated with hypospadias surgery which may result in decreased urinary flow due to edema or post-operative stricture or meatal stenosis and also due to non-compliant neo-urethra [9-12]. Wolffenbuttel et al. [12] found that patients less than 2 years of age at the time of surgery already had decreased flow and interrupted micturition. In patients with hypospadias, the urethra might already have a stricture or be less distensible, due to structural abnormality in or around the spongiosum body. The bladder condition might present another risk factor for VUR in these patients. It has been found that in hypospadias that VUR is of low grade and asymptomatic, without fever which resolves within 2years of age without any intervention [13]. Most cases of VUR are temporary and are secondary to instability of the urethra and bladder at younger age groups. They resolve in 90% of patients, 1-2 years following surgery [13]. In a study of 214 patients with primary reflux, VUR resolves only by 13% yearly during the initial 5 yeas follow-up [14]. Many studies and case reports have been done in this section. The upper urinary tract anomalies are most frequently found in patients with hypospadias than in general population [15]. Undescended testis is the most frequent associated anomaly seen in patients with hypospadias. There are controversies on whether USG/ micturating cystourethrogram (MCU) should be done routinely in all cases of hypospadias. In a study by Davenport et al. [4] showed that routine ultrasonography is not justified unless hypospadias is severe or there is an associated anomaly. In another study by Moore et al. showed that the overall incidence of surgical interference (not including hypospadias repair) was 11.7% and routine radiological screening in boys with hypospadias is recommended [16]. However there is always a dilemma in all physicians related to radiological evaluation. In our case report, MCU was followed after USG report. We recommend MCU to be done in specific cases were child is symptomatic or USG shows evidence of hydroureteronephrosis.

Conclusion

We can say that bilateral high grade VUR with febrile UTIs in patients with hypospadias before surgery is a very rare association which has never been reported that needs a high index of suspicion for diagnosis. Proper history, early diagnosis, evaluation and surgical intervention can lead to a successful outcome in these patients. We believe that increased awareness of these associations will improve the ability of surgeons to diagnose similar cases in a timely fashion leading to preservation of upper urinary tracts in cases with hypospadias.

Compliance with ethical statements

Conflicts of Interest: None.

Financial disclosure: None.

Consent: All photos were taken with parental consent.

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