Post-urethroplasty symptoms: Complications or pre-existing pathology?

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Abstract. Hypospadias is not usually thought to be associated with upper tract anomalies. However, in some cases there may be associated undiscovered upper tract anomalies which can complicate the post-operative period and cause anxiety to both the patient and surgeon. A 2-year-old boy presented with mid penile hypospadias and no other associated urinary complaints such as poor urinary stream, retention, dribbling or dysuria. He underwent an uncomplicated Snodgrass urethroplasty. After 6m of asymptomatic period, the child started suffering from repeated urinary tract infections (UTIs), episodes of retention and poor stream. Post-urethroplasty stricture was suspected but a retrograde urethrogram, cystoscopy and urethral calibration ruled out the possibility. Upper tract evaluation was then undertaken. Ultrasonography and micturating cystourethrogram were finally able to show a thick walled trabeculated bladder with bilateral dilated and tortuous ureters and bilateral grade 5 vesicoureteric reflux (VUR). The child underwent bilateral cross-trigonal ureteric reimplantation for the high grade reflux. Post reimplantation the patient remains well at 4 months follow-up with resolution of recurrent and obstructive uropathy. Pre-existing undiscovered anomalies may complicate the course of a simple case of hypospadias. Thorough investigations, high index of suspicion and preparedness for dealing with unexpected complications are required. If facilities permit, a screening ultrasound before undertaking even a simple hypospadias surgery can detect associated upper tract anomalies and thus circumvent unnecessary anxiety and investigations post-urethroplasty.

Key words: Urethroplasty, Symptoms, Stricture, Obstructive Uropathy

Introduction

Hypospadias is one of the most common congenital anomalies with an incidence of 1 in 300 live births. Though earlier thought to be an isolated anomaly, various studies over the years have revealed that it may be associated with significant genito-urinary, genetic, hormonal and extr-urological anomalies. Hypospadias can be a feature among many other abnormalities in about 33 syndromes that have been indexed by Smith. The incidence of associated urogenital anomalies has been found to be 35-40% whereas extra-urogenital anomalies have been found in 2-16% in various studies. Upper tract anomalies have been observed in 4-11% of patients with hypospadias.

Considering such a wide variation in incidence and severity of associated anomalies, the need for routine pre-operative investigations has been debated, with some authors recommending a complete workup including even an intravenous urogram, while others deeming any investigation as unnecessary. While the choice of performing preoperative screening investigations is currently considered a matter of individual preference, it is important to remember that the innocuous appearing hypospadias may also be associated with anomalies that can become symptomatic and complicate the recovery of a patient post-urethroplasty. Failure to do so may cause anxiety to the surgeon as well as the patient about the urethroplasty and lead to avoidable investigations. The following case report highlights the necessity of keeping in mind the possibility of associated extra-urological anomalies which may have a bearing on recovery of child post-surgery for hypospadias.

Case Report

A 2-year-old boy presented in our outpatient department with complaints of abnormal ventral position of urinary meatus and deviant urinary stream. He had no history of any other urinary complaints like urinary dribbling, straining, retention or poor stream. No other systemic complaints including abnormal bowel movements were reported. On examination,
the child was diagnosed to have a mid-penile hypospadias with moderate chordee. The glanular fossa was deep and urethral plate of acceptable width and quality. The child underwent a single stage Snodgrass urethroplasty over an 8Fr stent after complete degloving of phallus to remove the chordee. The catheter was kept for 10 days. The child had an uneventful recovery and was asymptomatic till 6 months after surgery when he presented to another hospital with acute onset of urinary retention in the emergency. A post-urethroplasty stricture complicated by acute urinary tract infection (UTI) was suspected by the attending surgeon. However an 8Fr catheter could be passed easily through the neo-urethra and the child was thus relieved of the retention. The child was treated for urinary tract infection according to the culture sensitivity. However, the child continued to have recurrent UTI and episodes of acute urinary retention with occasional poor stream. A retrograde urethrogram (RGU) ruled out a post-urethroplasty stricture and showed a good caliber urethra with no narrowing or stenosis (Fig.1a). Cystoscopy again revealed a normal caliber neo-urethra with no narrowing. Following this the child was referred back to our hospital. Since the neo-urethra was proven normal, a separate pathology was suspected and an ultrasonography (USG) done, which revealed a thick walled trabeculated bladder with bilateral hydroureretonephrosis. A micturating cystourethrogram (MCU) revealed bilateral grade 5 vesicoureteric reflux (VUR) with multiple bladder trabeculations and diverticulae (Fig.1b). Tc-99 Dimercaptosuccinic acid (DMSA) showed bilateral upper polar scars. Urodynamic studies could not be done due to lack of facilities. The child underwent bilateral Cohen's cross-trigonal ureteric reimplantation (Fig.2) along with behavioral changes such as frequent micturition. At 4 month follow-up the child is asymptomatic with no episodes of UTI or urinary retention and a good urinary stream.

Discussion

Hypospadias had been earlier considered an isolated anomaly. However recent studies now emphasize the presence of associated uro-genital and extra-urological anomalies with hypospadias. Though most of them do not require immediate surgical correction, the knowledge of their presence may be important in planning the long term management of the patient. The incidence of associated urogenital anomalies has been found to be 35-40%, most common being undescended testis, inguinal hernia, penoscrotal transposition and prominent uticulus. Congenital heart defects, musculo-skeletal anomalies, anorectal malformations and neurological defects are the most common associated extra-urological anomalies, present in 2-16% of cases. These anomalies are found to increase in incidence in proportion to the severity of the penile deformity, being highest in penoscrotal or scrotal hypospadias.

Upper tract anomalies are seen in 4- 12% of children with hypospadias. Kulkarni et al reported a 11% incidence of upper tract anomalies with approximately 5.5% incidence of vesicoureteric reflux; at least half of whom were symptomatic and presented with recurrent urinary tract infection. However, there was no direct relationship between the degree of hypospadias and upper urinary tract anomaly. This is corroborated in our case also as the child had a mid-penile hypospadias but bilateral severe Grade 5 VUR.

The upper urinary tract anomalies were more frequently found in patients with hypospadias than in general population. It is thought that the higher than normal incidence of upper tract anomalies may be due to some overlap of maturation of kidney, ureters and urethra. It is possible that the development of kidneys and the urethra is
influenced by a common factor. For this reason some authors even recommend a routine intravenous urogram before undertaking urethroplasty. However, this is debatable with others finding it unnecessary in all cases. Ultrasonography is a quick, non-invasive and convenient investigation for screening any associated upper tract anomaly. It can influence surgical planning such as prioritizing ureteric reimplantation before urethroplasty. It can also guide further investigations such as need for MCU in presence of back pressure changes. Most importantly a screening ultrasound prepares both the patient and surgeon for any complications during recovery. Thus it can prevent anxiety and suspicion about the urethroplasty and help in avoiding unnecessary investigations. While Kulkarni et al recommend a screening USG in all cases of hypospadias, Duckett recommends an USG to be done in cases of hypospadias associated with anomalies of other systems such as anorectal malformations and meningomyelocele.

Had our patient undergone a screening USG, the high grade reflux would have been discovered and managed appropriately before urethroplasty. RU and cystoscopy could have been avoided. The parents could have been mentally prepared and prognosticated accordingly.

Conclusion

Pre-existing undiscovered anomalies may complicate the course of a simple case of hypospadias. Unexpected post-operative symptoms may not always be due to surgical complications but may reflect an associated pathology. Thorough investigations, high index of suspicion and preparedness for dealing with unexpected complications are required. If facilities permit, a screening ultrasound before undertaking even a simple hypospadias surgery can detect associated upper tract anomalies and thus circumvent unnecessary anxiety and investigations post-urethroplasty.

References