Anterior bladder flap neo urethra as treatment for stress urinary incontinence due to developmental urogenital anomaly

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CASE REPORT

A 15 year old girl was referred to urogynecology department (April’08) with severe urinary incontinence since childhood.

Cloacal anomaly was detected soon after birth (March’93) which was managed successfully by the paediatric surgeon with colostomy, anal transposition and colostomy closure. In view of non visualisation of vaginal opening, vaginoplasty at a later date was suggested by the paediatric surgeon.

A complete workup was done in the urogynecology department. Local examination under anaesthesia showed a very wide and patulous urethra with urinary leakage. A narrow vaginal opening was lying lateral to the urethral orifice (instead being inferior) with a bridge of tissue separating the two [Figure 1]. The vaginal opening could be dilated to expose a normal cervix and adequate vaginal length that did not require vaginoplasty. Radiological imaging (Ultrasonography, Computed Tomography imaging, Micturating Cystourethrogram, intravenous urogram)

Key Words: Urethral reconstruction, urinary incontinence, urogenital sinus

Abstract

Congenital anomalies that involve the distal segment of urogenital sinus (giving rise to female urethra and vagina) may lead to abnormal urethral development ranging from absent to markedly deficient urethra. The abnormal division may also cause a short and patulous urethra. Sphincteric defects are likely to be associated and when combined with the short urethral length is a cause for severe urinary incontinence. Urinary incontinence due to a congenital cause requiring repeated urethral reconstruction to relieve symptoms is presented. A 15 year old girl was referred for bothersome urinary incontinence due to a short, wide, patulous urethra with defective sphincteric mechanism as part of urogenital sinus developmental anomaly. She was initially managed by reconstruction of bladder neck and proximal urethra with sphincter augmentation using autologous pubovaginal sling. Persistent urinary incontinence demanded a second urethral reconstruction using tubularised anterior bladder flap (modified Tanagho). Surgical reconstruction of the urethra achieved socially acceptable continence.

INTRODUCTION

The close embryological development of anorectal, genital and urinary system increases the occurrence of coexistent anomalies and may need complex multiple surgical corrections. Cloacal anomalies compel detection and management at infancy whereas urogenital anomalies become identifiable at puberty with onset of menstruation or later due to bothersome urinary symptoms. A rare case of urogenital sinus anomaly with short urethra causing severe urinary incontinence is reported as it posed difficulty in management requiring repeated surgeries.
confirmed normal upper urinary tract. Bladder appeared normal in capacity as well as contour. Features of stress urinary incontinence with markedly decreased valsalva leak point pressure were depicted in the urodynamic assessment. There were no features of bladder outlet obstruction or detrusor overactivity. Cystourethroscopy confirmed patulous short urethra with significant sphincter deficiency.

Developmental urethral anomaly as part of urogenital sinus maldevelopment was the cause of her incontinence so reconstruction of urethra and augmentation of urethral sphincter with autologous fascia lata as pubovaginal sling was done on 11.1.10. During the procedure the dense fibrous scar tissue between urethra and vagina was dissected. After mobilizing the remaining urethra vaginal wall flap was used to lengthen the urethra and reconstruct the external urethral meatus. A harvested strip of fascia lata was laid under the proximal urethra as pubovaginal sling.

Persistence of significant urinary incontinence was observed in the post operative period. Re-evaluation was done after six months and confirmed persistent short (approximately one cm) urethra with sphincter deficiency. A second reconstruction was planned to form a neourethra. Continent diversion was also an available option but was deferred considering her age and child bearing prospective. Restoring continence with a near normal functional anatomy remained the prime objective.

On 21.01.11 examination revealed a nonexistent urethra and bladder neck and the only opening discharging urine was identified as bladder. It was decided to reconstruct the urethra from tubularised anterior bladder wall (modified Tanagho flap). Parallel incisions were made in the anterior wall of the bladder to form a 5 cm × 2 cm rectangular bladder flap preserving its proximal attachment. Scarring around the bladder opening due to previous surgery demanded abdominal mobilization of the bladder. Further extraperitoneal mobilization of the bladder made the anterior wall flap tension free which was then tubularised to create the neourethra. The distal end was brought down at the expected site of meatus to form the external urethral meatus.

Laparotomy also revealed a lateral fusion defect that included a relatively well developed communicating left horn, rudimentary right horn and a right endometriotic cyst of 4 cm × 5 cm which was drained.

Urinary drainage was ensured by suprapubic and urethral catheter post operatively. Continuous periurethral leak (leak around the catheter) was observed in second post operative week which was treated with antimuscarinics. Menstrual suppression was given for six months in view of endometriotic cyst.

Initially frequency, urgency and urge incontinence was significant which improved with antimuscarinics and bladder training. Four months following the surgery, patient has achieved a continent period of 2 hours which is socially acceptable to her and is likely to improve further with bladder training. A normal urinary flow rate and no residual urinary volume excluded bladder outlet obstruction. However, regular and longer follow up with symptomatic and functional assessment is required.

**DISCUSSION**

Female urethral development occurs from pelvic and phallic part of urogenital sinus. The abnormal division of urogenital sinus with possible malrotation occurring during the development is the probable hypothesis for the short, wide urethra, a narrow and lateral placement of the vaginal opening in this patient. Also, differential growth with ageing due to the non addressed urogenital anomaly at birth may have contributed to the defect.

However, the exact embryological defect remains unknown and closely resembles congenital short urethra where the urethra seldom exceeds two cm in length and its orifice is typically wide and patulous. Although the bladder is normal in such cases, the sphincteric mechanism is wholly deficient. There is a symptomatic resemblance with female epispadias but absence of characteristic bifid clitoris, depressed mons, ill developed labia, occasional symphyseal separation and deficient dorsal urethral wall restrained us from making a clear cut diagnosis of female epispadias.

In patients with severe stress urinary incontinence attributed to short urethral length and defective sphincter mechanism, the goals of urethral reconstructive surgery should be directed to
achieve adequate urethral length and restoration of continence. Pubovaginal sling using autologous tissue is considered gold standard procedure for treatment of urinary incontinence in cases of defective sphincteric mechanism.\(^1\)

With an objective to restore adequate urethral length and continence, initially reconstruction and sphincter augmentation was attempted. Fascia lata was used as Pubovaginal sling and synthetic sling was intentionally avoided. The use of fibrotic anterior vaginal wall (due to developmental defect) during reconstruction had probably failed which resulted in persistent leakage and demanded a more elaborate urethral reconstruction by way of forming a neourethra. Non availability of adequate vaginal tissue made bladder flap a suitable choice. Decrease in bladder capacity was anticipated which improved in due course.

Anterior bladder flap neourethra (Tanagho’s procedure) originally described for developmental urethral defects\(^2\) is currently a widely accepted procedure for cases of extensive urethral loss\(^3\) due to any reason. The application of this procedure in the current case is based on the similar concept.

Anterior bladder flap neourethra was described by Tanagho in 1969\(^4\) for patients with urethral and sphincteric loss during transurethral resection of prostate. A square flap 2.5 cm × 2.5 cm was raised from the anterior bladder wall and turned into a tube around a 12 F catheter by interrupted sutures. After the tube is closed the apex of the trigone at the midline posteriorly is secured with a mattress suture to the base of the tube which will now be the level of internal meatus this is followed by closure of bladder on either side. The final closure suture line will be Y-shaped with the straight stem in the posterior midline of the tube and the upper V shape in the posterior aspect of the bladder.

The major modification in our surgical technique was the length of the flap. A rectangular 5 cm × 2 cm flap from anterior bladder wall was reconstructed into urethra over a catheter. The extra length of neourethra thus gained by a longer flap was helpful in achieving continence, other than the circular muscle fibres at the distal end acting as a sphincter.

Extensive urethral reconstruction may not restore complete continence due to the associated neuromuscular damage. The intricacies of anatomy and dynamics of urethral sphincter mechanism were difficult to reproduce requiring multiple attempts for functional restoration and continence.

The associated mullerian duct anomaly in this case belongs to class III (lateral fusion defect with asymmetric obstructed disorder of uterus or vagina) of American Fertility Society. Endometriotic cyst presently has regressed with menstrual suppression; however, intervention may be required for non communicating rudimentary right horn if it becomes symptomatic.

CONCLUSION
Management of short urethra and severe urinary incontinence due to impaired sphincteric mechanism as part of developmental anomaly warrants multiple surgical reconstructions. Socially acceptable continent periods can be achieved with anterior bladder flap neourethra.

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