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Case Report

Acquired hypospadias in females: an unusual presentation of obstetric injury

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ABSTRACT

Obstetric injuries are frequently encountered in caesarean and vaginal deliveries. Vaginal deliveries have been seen to be associated with vaginal lacerations, cervical tears, perineal body injury, anal mucosa tear, anal sphincter tear, forniceal tear, bladder injuries and urethral or periurethral injuries. Urethral injuries account for the rarest variety in obstetrics. More commonly occurring in direct trauma or pelvic fracture, urethral injuries can range from minor contusion to major transection injury. Here we report a unique case of urethral injury post-delivery, which presented to us after 37 years of infliction, with splaying of urine, poor stream, post void dribbling and nocturnal enuresis. It was diagnosed by clinical examination and cystovaginoscopy. The defect was repaired in one step surgery, with three layered closures.

Keywords: Hypospadias, Obstetric injury, Urethral injury, Urethral reconstruction

INTRODUCTION

Urethral injuries occur typically by trauma, or iatrogenic injury during pelvic surgery.[1,2] They are one of the rarest among the gamut of obstetric injuries. Difficult vaginal delivery can lead to maternal and fetal complications. Use of vaccum or forceps can add to the complication rate. The maternal complications like perineal tears, vaginal tears, cervical tears, and postpartum hemorrhage can occur.[1] The vaginal lacerations are mostly in middle and distal third of vagina. Urethral injuries are rarely reported in literature. Mostly the urethral injuries are associated with vaginal lacerations.[1]

Diagnosing them requires a proper history and a detailed pelvic examination. It should not be confused with stress urinary incontinence. Imaging can be used to delineate the diagnosis in case of confusion. The key of successful repair includes proper visualization, tension-free closure, and repair in multiple layers with avoidance of overlapping suture lines.[3]

CASE REPORT

A lady in her 50's presented to female pelvic medicine clinic with complaints of involuntary leakage of urine since her first vaginal delivery 37 years ago. She had an uncomplicated unsupervised pregnancy following which she had spontaneous onset of labor at term gestation. She had prolonged second stage of labor, and had a difficult vaginal delivery of a baby girl weighing 3.5 kg conducted by an untrained person at her home. Immediately postpartum, she had both day and night time incontinence. Thereafter she had five more vaginal deliveries in the next seven years, which did not change her urinary symptoms.

She presented to the outpatient department of female pelvic medicine clinic for the first time for an evaluation and treatment of a long-standing complaint.

She was a moderately built and poorly nourished female with complaints of increased day time urinary frequency (14-16 times), night time frequency (3-4 times), on and off

burning micturition. She had splaying of urine, poor stream, post void dribbling along with nocturnal enuresis. Minimal amount of self-void of urine was present. She did not get herself operated anywhere since all these years. Imaging done at a local village did not reveal any abnormality. She achieved her menopause at the age of 50 years. Her menstrual cycles were regular, flow lasting for 4 days and cycle length of 28-30 days with normal flow. She had blood in urine (menouria) each time she menstruated after her first vaginal birth.

Pelvic examination showed a bivalve dorsal urethra with loss of anterior vaginal wall tissue with urine seen leaking through an opening at the level of bladder neck (Figure 1).



Figure 1 : Local examination findings.

Investigations

Voiding diary showed a day time frequency of 16 times, nocturnal enuresis soaking 40ml/hr on an average. She had episodes of scanty dribbling (2-4 ml) of urine multiple times in a day irrespective of stress, cough, heavy weight lifting, and laughter. Uroflowmetry performed on outpatient basis showed a flow rate of 68.9 ml/sec, voided volume of 463 ml and post void residual urine of 90 ml. Computed tomography reported a vesicovaginal fistula measuring 7×13 mm at the base of urinary bladder communicating with the anterior vaginal wall. Cystovaginoscopy was performed on day care basis. It showed a bivalve dorsal urethra with ventral urethral and anterior vaginal wall tissue loss. A communication measuring 1cm was seen from the proximal urethra just below the bladder neck to the vagina. The edges were healed and fused. Bladder neck was normally visualised. Bladder with its both ureteric orifices were seen. Rest of the vagina and cervix was normal looking apart from the above-described defect.

Her hemoglobin was 12.6 g/dl, total leucocyte count was 5410/mm^3, S. creatinine 0.76 mg/dl. Her blood sugar, liver function test, viral markers are normal. Urine microscopy showed a ph 6.0, 8-10 pus cells and urine culture had *Escherichia coli* growth. After an antibiotic course, sterile urine culture was sought before posting her for urethral reconstruction.

Treatment

Patient was laid in prone split leg position. After adequate exposure, stay sutures were applied all around the edges of the bivalved urethra. A plane was created between the vaginal mucosa and the underlying pubocervical fascia all around the margins, extending up to the bladder neck. (Figure 2) A Silicon foley's no.18 was inserted into the bladder. After adequate mobilisation of edges, the edges of urethral mucosa were approximated over the foleys with vicryl 3-0 in a continuous fashion. Thereafter, the pubocervical fascia approximation was done with the help of buttressing sutures. Third layer of closure (vaginal mucosa) was done with vicryl 3-0 suture.

Outcome and follow-up

Postoperatively the patient was started on anticholinergic (oxybutynin 5 mg t.i.d) and injectable antibiotics for 24 hours followed by oral antibiotic. Patient was discharged on postoperative day 4 after obtaining a sterile urine culture sensitivity. Patient was followed up weekly till 3 weeks when her foleys catheter was removed. Patient is under regular follow up and is continent since last 1 year.

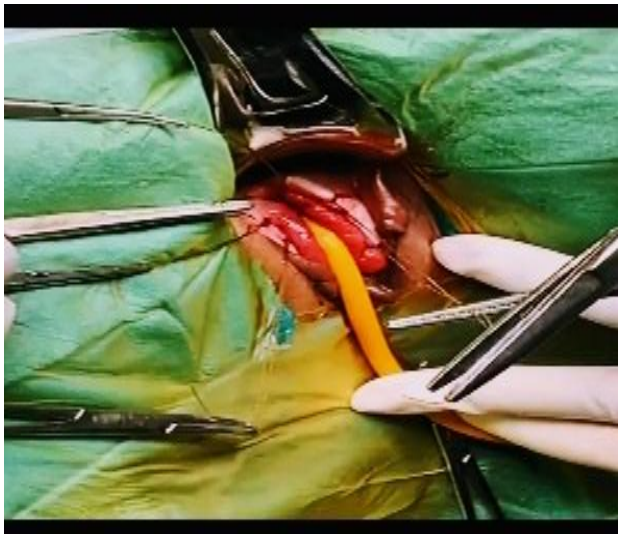


Figure 2: Dissection of plane between urethral mucosa and anterior vaginal wall circumferentially.

DISCUSSION

Injuries to pelvic organs to surrounding structures can occur in both, lower segment caesarean section, and a difficult vaginal delivery or instrumental delivery. Laceration in vaginal walls, torn anal sphincter, extending upto anal mucosa, vesicovaginal fistulas and rarely urethral injury can occur following a difficult prolonged vaginal delivery. Caesarean delivery with difficult dissection in case of adhesions in previous caesarean delivery or previous surgery for ectopic pregnancy or

abdominopelvic surgery, inflammatory condition can be a risk factor for bladder and ureteric injury, former being more common.

Literature has case reports of patients having bladder and ureter injuries after caesarean section. Urethra injury has been reported rarely among the vaginal deliveries. In a study done by Shantel et al incidence of obstetric associated bladder injuries was 0.14% and 0.94%, with majority occurring in caesarean delivery.[6] Incidence of ureteric injuries in caesarean delivery was reported to be between 0.013-0.09%. In their study, 10.5% (2/19 patients) had vaginal delivery induced lower urinary tract injury, both being urethral injuries. Another case of simultaneous urethral, bladder and uterine rupture has been reported in a 22-year patient following vaginal delivery. On vaginal examination, the bladder neck and proximal one third of the urethra were ruptured. An extensive rupture of the lower uterine segment to the left sidewall of the uterus was seen.[7]

Urethral injuries are infrequent in females oughing to its small length. Urethral trauma has been reported in 10.3/1000 vaginal deliveries.[8] Urethral injuries occur after direct trauma near vagina, cuts or pelvic fracture. Anterior urethral injuries tend to occur after a crushing injury while posterior one involving sheer forces. Improper catheterisation and instrumentation are the most common cause of urethral trauma worldwide.[9] The trauma can range from a mild contusion to complete open transection. It needs to be treated timely as scar formation in the injured tract can lead to fibrosis, stricture further causing upper tract changes in bladder, bilateral ureters and kidneys. Obstetric injuries have not been shown to be very extensive unless a complete transection as in our case.

Urethra can be evaluated by retrograde urethrography as the contrast extravasation delineates the broken tract. It is best for acute injury cases. Another imaging modality, computed tomography (CT) scan is particularly useful to evaluate kindness, bladder and ureter. Ultrasonography and magnetic resonance imaging remain challenging to assess urethral injury cases.[10]

In males, urethral injuries have been classified into five types based on location of injured segment: (Unified Anatomical Mechanical Classification).[11]

Type 1: Posterior urethra is stretched but intact. Type 2: Pure posterior urethral injury with a tear of the membranous urethra above the urogenital diaphragm (UGD); partial or complete. Type 3: Combined anterior and posterior urethral injury with the involvement of UGD; partial or complete. Type 4: Bladder neck injury with extension into the urethra Type 4a: Injury of the base of the bladder with periurethral extravasation and Type 5: Pure anterior urethral injury; partial or complete.

Management of urethral injury depends on the condition of patient and extent of injury. It can be divided into immediate (0-10 days), delayed (10-14 days) and late (>3 months). Bladder decompression is important in a urethral trauma, and placing a suprapubic cystotomy (SPC) is recommended. Early realignment should be tried transurethrally, following which the SPC is kept *in situ* for 1-3 months.

In our case, the patient had sustained the urethral injury following a very difficult vaginal delivery conducted by a untrained person at home 37 years ago, following which the urine kept on leaking from just below the bladder neck. Examination showed leakage of urine through ventral aspect with loss of anterior vaginal and ventral urethral wall, a picture similar to hypospadias in males. It was a healed urethral bivalve injury. Patient was operated after obtaining a sterile culture by a tranvaginal urethral reconstruction. Postoperatively, the patient remained continent and is under regular follow up since 1 year.

CONCLUSION

Urethral injuries are rare of its kind among the gamut of obstetric postpartum complications. It can present immediately postpartum or later in life with nocturnal enuresis and poor stream. The approach remains in two steps: One to reconstruct the urethra, and second to provide continence. Sometimes, the patients do not require anti-incontinence surgery, and remain continent by one step approach as in our case.

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