DOI: 10.7860/JCDR/2016/20106.8256

Case Report

Obstetrics and Gynaecology Section

Urethral Coitus in a Case of Vaginal Agenesis - Is Only Vaginoplasty Enough to Treat the Urinary Problems?

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ABSTRACT

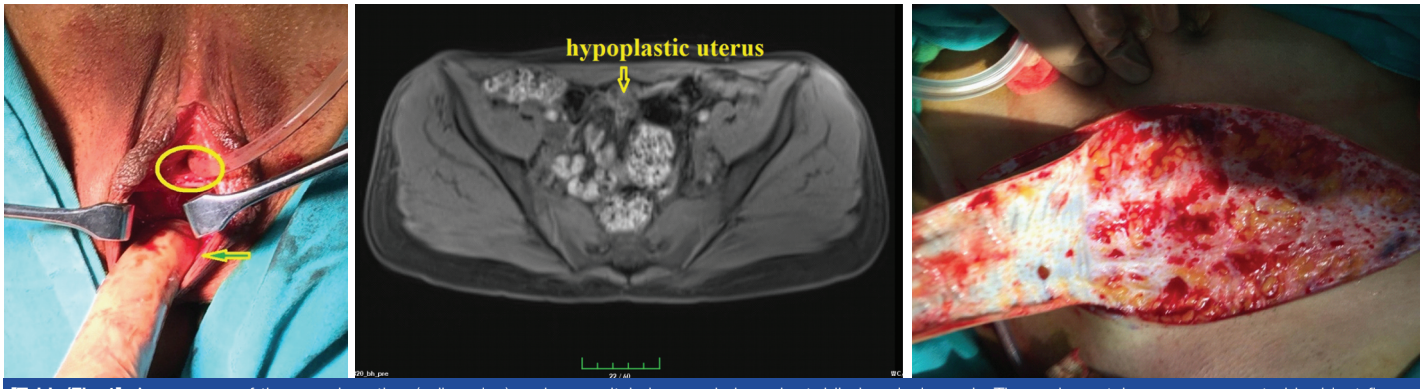
Urethral coitus is an extremely rare condition. Megalourethra and urinary incontinence due to urethral coitus in vaginal agenesis are unusual manifestations because these patients usually present with primary amenorrhea before becoming sexually active and receive treatment. A 24-year-old woman came to our clinic because of primary amenorrhea, sexual dysfunction, dyspareunia, megalourethra and urinary incontinence five months after her marriage due to urethral coitus. Based on these clinical and radiological findings a diagnosis of Mayer-Rokitansky-Kuster-Hauser Syndrome was made and patient underwent modified McIndoe Vaginoplasty. The elasticity of female urethra permits repeated coitus, together with the physical damage can probably lead to incontinence. In this case, after eliminating the underlying cause of disease with vaginoplasty, no other treatment was required. The integrity of sphincteric function and structural support of urethra might be regained without subjecting the patient to aggressive reconstructive surgical procedures.

Keywords: Full-thickness skin graft vaginoplasty, Mayer-rokitansky-kuster-hauser syndrome, Megalourethra, Urethral coitus

CASE REPORT

A 24-year-old woman came to our clinic because of primary amenorrhea, sexual dysfunction, dyspareunia and urinary incontinence five months after her marriage, but she did not seek medical attention for her persistent symptoms. On targeted questioning, she stated that she was too ashamed and afraid to tell anyone about her problems. The external gynaecological examination revealed completed puberty with normal female phenotype and secondary sexual characteristics including normal external genitalia, breasts, pubic and axillary hairs. External urethral meatus seemed dilated approximately 20mm in diameter, easily admitting one finger and the urogenital sinus ends in a short, blind vaginal pouch [Table/Fig-1]. The external urethral orifice and its supporting structures including the urethral sphincter were observed to be damaged. There was visible leakage of urine from the urethral meatus. Patient was wearing sanitary pads and changing them many times a day. Urethral Coitus (UC) was highly probable. A detailed anamnesis and a clinical examination revealed that they were using this "damaged area" for sexual intercourse. She had a history of recurrent microscopic haematuria and urinary tract infections. Additionally she was diagnosed as having honeymoon cystitis since some times. She has been treated with antibiotics and symptomatic medication but these managements provided only transient relief. Ultrasound and magnetic resonance imaging showed hypoplastic uterus measuring 40×28mm size [Table/Fig-2], associated with agenesis of the upper-middle third of the vagina. Both ovaries were normal. Hormonal analyses were within normal ranges and her karyotype was 46, XX. Based on these clinical and radiological findings a diagnosis of Mayer-Rokitansky-Kuster-Hauser Syndrome was made. Reconstruction was planned by modified McIndoe's vaginoplasty.

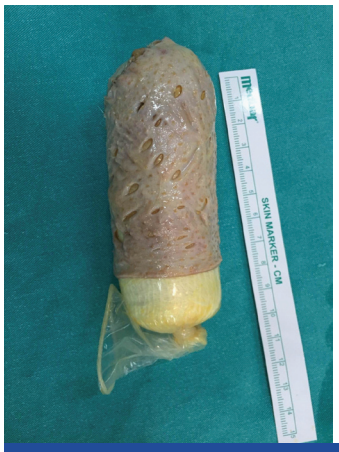
Patient was operated on under general anesthesia in the lithotomy position with urinary catheterization. An "X" shaped incision was made on the blind vaginal pouch and then a neovaginal space was achieved in vesicorectal space with blunt finger dissection [Table/Fig-1]. A suitable cavity size with vaginal dimensions (~ 12 cm

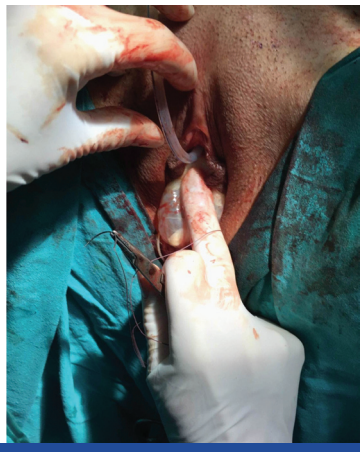


[Table/Fig-1]: Appearance of the megalourethra (yellow ring) and urogenital sinus ends in a short, blind vaginal pouch. The vesicorectal space was created by blunt finger dissection (arrow). **[Table/Fig-2]:** Magnetic resonance imaging of hypoplastic uterus. **[Table/Fig-3]:** A full-thickness skin graft is harvested from lower abdomen with a scalpel and remaining fat residuals of the subcutaneous tissue is carefully removed.

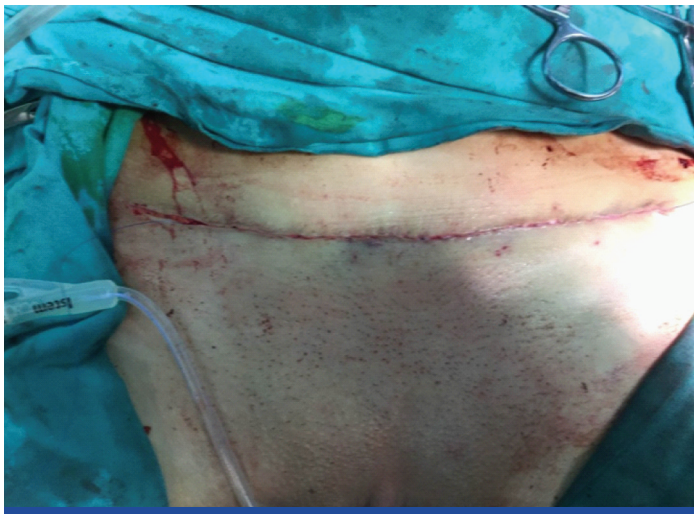
length and ~ 5cm width) was created. The full-thickness skin grafts were taken from lower abdomen [Table/Fig-3] and dressed over the mould with absorbable sutures [Table/Fig-4]. The graft was perforated with multiple slits and then inserted into the neovaginal cavity. Finally, the external edge of the skin graft was sutured to the perineal margins of the incisions [Table/Fig-5] and the abdominal incision was closed [Table/Fig-6]. The labia minora were sutured to keep the mould in position. The vesical catheter and the mould were kept in place for 7 postoperative days.

The diameter of the urethra decreased over time and had returned to almost normal appearance. At 3 months of follow-up the patient claims normal sexual function and the couple were more satisfied with their sex lives. Six months after surgery the patient voided normally and completes resolution of urinary incontinence was achieved. Additionally, a significant improvement of pelvic pain and dyspareunia was noted.





[Table/Fig-4]: The graft was perforated at multiple places with a scalpel to prevent separation of the graft from its wound bed by haematoma or seroma.
[Table/Fig-5]: A mould made by sponge and covered with latex condom which is simple, inexpensive and safe has been used for long period to maintain the space and to allow epithelialization.



[Table/Fig-6]: Primary closure of donor area.

DISCUSSION

Urethral Coitus (UC) is an extremely rare condition and only about 28 cases have been reported thus far [1]. Although UC in women with a normal vagina and introitus has been reported [1], in the majority of these cases, the main underlying cause of this problem is vaginal agenesis or hymenal anomalies [1-3]. Urethral incontinence and urethral dilatation are unusual presentations in vaginal agenesis which poses an important surgical challenge of restoring continence.

Herein, we describe a case of urethral dilatation and urethral incontinence caused by urethral intercourse in a woman with vaginal agenesis that we treated with only modified McIndoe's vaginoplasty. After eliminating the underlying cause of disease with vaginoplasty, no other treatment was required. There are only a few reports of the subject in the literature that the integrity of sphincteric function and structural support has been regained without subjecting the patient to urethral reconstructive procedures [3-5].

Congenital vaginal agenesis is a rare malformation with an incidence of one in 4000 to 5000 female newborns [6]. Megalourethra and urinary incontinence due to UC in vaginal agenesis are unusual manifestation because these patients usually present with primary amenorrhea before becoming sexually active and receive treatment. Urinary incontinence and dyspareunia are frequently expected symptoms of UC. However, this condition may occur in a heterogeneous group of patients and the clinical manifestations may differ from patients to patients [4]. Therefore, clinicians must have a low threshold of suspicion under many circumstances. Pelvic fractures, traumatic childbirth and iatrogenic causes are the most relevant underlying causes of urethral laceration and loss of sphincteric mechanism [7]. Stenotic vaginal introitus is an apparent risk factor for the UC.

The elasticity of female urethra permits repeated coitus, together with the physical damage can probably lead to incontinence. This

Author date	N	Age	Presentation	Surgical intervention
Present case	1	24	Urinary incontinence, dyspareunia, primary amenorrhea	Modified McIndoe's vaginoplasty
Aksakal et al., 2015 [1]	1	48	Urinary incontinence	Urethroplasty, urethral plication
Brown et al., 2012 [2]	1	27	Bowel evisceration through perforated bladder.	Repair of cystotomy, suturing bladder neck and planned for reconstructive surgery
Di Donato et al., 2008 [3]	1	-	Dyspareunia, infertility, incontinence during coitus, urethral dilatation, microperforate hymen	Surgical dilatation of hymen with dilators
Ryckman J et al., 2014 [4]	2	13 16	Case1: Small vaginal dimple and vaginal agenesis Case2: Dilated urethral orifice, primary amenorrhea and cyclic abdominal pain	Case 1: Declined vaginal dilatation or reconstruction Case 2: Stent/dilator therapy.
Khattar N et al., 2008 [5]	1	16	Incontinence during coitus	McIndoe's vaginoplasty and urethral plication
ZarghamM et al., 2014 [7]	2	21 19	Case 1: Urinary incontinence, dyspareunia, infertility, recurrent urinary tract infection Case:2: Enuresis, urinary tract infection, dyspareunia	Case1:Urethroplasty with fascial graft and Martius flap Case2: Urethroplasty with paravaginal flap
Sert Ü et al., 2012 [8]	1	24	Urinary incontinence	Modified Martius technique
Sakinci et al., 2012 [9]	1	23	Primary infertility with congenital adrenal hyperplasia, megalourethra	Creation of a functional neovagina and urethral plication
Borski & Mittemeyer 1971 [10]	1	42	Dyspareunia, urinary incontinence	None
Ayan et al., 2000 [11]	2	22 28	Case 1:Suprapubic pain, urgency Case 2: Incontinence, infertility	Urethroplasty Urethroplasty
Verma et al., 2012 [12]	1	27	Urinary incontinence, dribbling of urine	Surgical repair of the fistula and McIndoe's vaginoplasty
Deniz et al., 2002 [13]	1	32	Dyspareunia, Urinary incontinence	Bladder flap
Okeke et al., 2007 [14]	1	21	Urinary incontinence of 1-year duration after being raped	Repair of urethral laceration and plication of urethra and bladder neck

[Table/Fig-7]: Clinical features and management of urethral coitus with brief review of literature.
N indicates the number of patients.

elasticity may allow the urethra to return to its physiologic size without subjecting the patient to aggressive reconstructive surgical procedures [3]. Reinforcing this idea, in the present case, after eliminating the underlying cause of disease with vaginoplasty, no other treatment (urethroplasty, urethrosphincteric reconstruction, ureteral plication, etc.) was required. As previously reported by Khattar et al., present study has also shown that once the urethral intercourse stops and a correct passage for coitus is provided, the muscular tone of urethral rhabdosphincter will slowly improve, thereby further contributing to continence [5]. According to Ünal et al., when urethral dilatation is <120 F, conservative management should be considered but if it is greater than 120 F urethra loses its tone [8].

Various procedures may be required to regain anatomical and functional integrity, particularly in cases of urethral sphincter injury and massive urethral dilatation. Surgery may also be indicated for cosmetic reasons and to prevent future urinary problems. For correction of patulous urethra continence, procedures in the form of bladder neck plication, urethrosphincteric reconstruction using bladder flap, or urethral plication with excision of urethral mucosal strip have been described [9]. Reported treatment options have focused on corrections of the squeal of UC and creation of a proper vaginal orifice to accommodate intercourse. Types of management is briefly reviewed in [Table/Fig-7].

CONCLUSION

Modified McIndoe's vaginoplasty may be used successfully in the treatment of urinary incontinence and megalourethra caused by UC. The integrity of sphincteric function and structural support might be regained without subjecting the patient to aggressive reconstructive surgical procedures.

Informed Consent: Written informed consent was obtained from the patient who participated in this study.

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FINANCIAL OR OTHER COMPETING INTERESTS: None.

Date of Submission: **Mar 12, 2016**
Date of Peer Review: **May 10, 2016**
Date of Acceptance: **May 16, 2016**
Date of Publishing: **Aug 01, 2016**