Case Report

# Adolescent Urethral Coitus: 2 Cases and Review of the Literature

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A B S T R A C T

*Background:* Urethral coitus is a very rare finding in adolescent women. There are 26 reported cases in the literature, but only two were in adolescents. Urethral coitus has been most commonly described in women with müllerian anomalies and less commonly in other unusual clinical circumstances.
*Cases:* We report 2 cases of adolescent women with known müllerian anomalies who were unknowingly engaging in urethral coitus; 1 adolescent with known vaginal agenesis and VACTERL association and one 16-year-old with an oblique vaginal septum and non-communicating functioning uterine horn. Both young women had significant urethral dilation at the time of examination under anesthesia.
*Conclusions:* Urethral intercourse is rare but may have significant consequences. Ongoing follow-up, careful history, and physical examination in adolescent women with müllerian anomalies are important in order to avoid possible complications, particularly when they are contemplating sexual activity. A history of urinary incontinence, dyspareunia, and urinary tract infections in young women with müllerian anomalies should raise suspicion of urethral coitus. If possible, the müllerian anomaly should be corrected to allow vaginal intercourse and discontinuation of urethral coitus.
*Key Words:* Adolescent, Urethral coitus, Müllerian anomalies, Urinary incontinence

## Introduction

Müllerian anomalies may present a significant challenge for both patients and health care providers, particularly around the time of initiation of sexual activity. Urethral coitus is very rare, particularly in cases of vaginal agenesis (Mayer-Rokitansky-Hauser-syndrome) as these women usually present prior to coitarche with primary amenorrhea and receive treatment. To date, most cases of urethral coitus in the literature involve women in their early to mid-twenties; only two of the reported cases occurred in adolescents[1,2] In the reported cases of urethral coitus, most presented with urinary incontinence during intercourse. Dyspareunia and recurrent urinary tract infections are also possible symptoms. We present 2 cases of urethral coitus in adolescents. The first is an unusual case of an adolescent female with known vaginal agenesis and VACTERL association (vertebral anomalies, anal atresia, cardiac malformations, tracheo-esophageal fistula, renal anomalies, and limb defects). The second is a 16-year-old female with an oblique vaginal septum and non-communicating functioning uterine horn. Both had unknowingly engaged in regular urethral intercourse. The urethral coitus literature is also reviewed.

### *Case 1*

A 13-year old Asian female with known VACTERL association (vertebral anomalies, anal atresia, cardiac defects, tracheo-esophageal fistula, renal anomalies, limb defects) and müllerian anomalies was initially assessed by pediatric gynecology to document the precise nature of her müllerian anomalies. On examination she had a small vaginal dimple and vaginal agenesis. She was subsequently followed regularly throughout her adolescence. She developed to Tanner Stage V breast and pubic hair and had normal vulvar estrogenization. Multiple radiological investigations could not delineate the precise nature of her müllerian anomalies; ultrasound and MR imaging were suggestive of 2 uterine horns and possible cervical atresia/agenesis. Despite being pubertal, the patient did not develop cyclic abdominal pain and serial imaging throughout adolescence did not demonstrate a hematometra. Her endocrine workup, including FSH, LH, estradiol, and TSH, was normal. She became sexually active at age 18. After 1 year of regular satisfactory intercourse, she had no complaints of dyspareunia or coitally-related incontinence. On examination in the clinic at age 19, she had what appeared to be a blind vagina of 4 cm in length but no visible cervix. Examination under anesthesia and vaginoscopy were performed to determine the status of the cervix and the feasibility of reconstructive surgery; the examination revealed a vaginal dimple, normal vulva, and a dilated urethral orifice approximately 2 cm in diameter. Urology was consulted intraoperatively. Cystoscopy revealed acute cystitis, bilateral ureteral openings, and an otherwise structurally normal bladder. Urethral length was normal with good sphincteric closure. Urine culture grew pansensitive *Escherichia coli.* The patient had unknowingly been engaging in urethral coitus. Post-operatively, vaginal dilator therapy was initiated but she was not compliant. She declined further investigations to delineate her müllerian

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anomaly and declined vaginal dilatation or reconstruction. Despite counseling, she elected to continue with urethral coitus.

*Case 2*

A 16-year old sexually active female presented in consultation for primary amenorrhea and cyclic abdominal pain. Previous ultrasound and MR imaging had documented a left rudimentary non-communicating functioning uterine horn, left renal agenesis, a high oblique vaginal septum, and a significant hematocolpos. On history, she did complain of dyspareunia but no history of urinary incontinence or recurrent urinary infections. Diagnostic laparoscopy, hysteroscopy, and surgical resection of the oblique vaginal septum were performed. Significant urethral dilatation was noted at the time of surgery suggesting that she had been engaging in urethral intercourse. Although she was not aware of this, urethral coitus was confirmed during careful post-operative history taking and examination with the patient. Her post-operative care included the use a vaginal stent and subsequent vaginal dilator therapy in order to prevent stenosis at the site of the septum resection as well as to create/maintain an adequate vaginal patency. Although she initially experienced difficulty with the stent/dilator therapy, she persisted and began to have successful vaginal intercourse. After 2 years of successful menstrual suppression, a laparoscopic excision of the non-communicating functioning rudimentary horn was performed. Urethral dilatation was no longer seen.

## Discussion

The first case of urethral coitus was reported in 1965 by Zeigerman and Gillenwater.[3] Since then, approximately 26 cases have been documented worldwide (Table 1), including 9 in the non- English literature.[4–12] The majority of them were diagnosed in women in their twenties or early thirties.[2,13–22] The presentation frequently consists of dyspareunia and coital incontinence often, though not exclusively, following marriage.[15,16,18–21,23] Several authors have reported urethral intercourse presenting as primary infertility.[2,14–16] Unfortunately, there are cases of traumatic urethral intercourse including one in a woman with normal female anatomy,[17] as well as one in a young woman with an imperforate hymen[1] during sexual assault. In many cases, urethral intercourse is associated with müllerian anomalies.[1,2,15,16,19,21,22] More unusual presentations include post-urethral coitus bladder rupture and small bowel evisceration through the urethra in a paraplegic patient who was chronically catheterized.[13] Another case described urethral coitus in a woman with a stenotic vaginal introitus that developed due to surgically corrected ambiguous genitalia secondary to congenital adrenal hyperplasia.[14]

Evidently urethral coitus may occur in a variety of patients and the clinical presentation may vary, hence

**Table 1**
Case Reports of Urethral Coitus in the English Literature

| Author/Date | Age of patient | Presentation | Management |
|---|---|---|---|
| Zeigerman et al (1965) | 32 | Urinary incontinence, dyspareunia, atypical uterine bleeding, annular fibrotic hymen | Urethral plication, hymenectomy |
| Borski & Mittemeyer (1971) | 42 | Urinary incontinence, dyspareunia, absence of the vagina | None |
| Taneja et al (1973) | 22 | Urinary incontinence, absence of vagina | Urethral plication and enteric vaginoplasty |
| Khanam et al (1978) | 22 | Dyspareunia, congenital absence of vagina, primary amenorrhea | McIndoe vaginoplasty and stenting |
| Shukla & Tripathi (1982) | 16 | Urinary incontinence, amenorrhea, absence of the vagina | Enteric vaginoplasty, urethral plication and sphinteric excercises |
| | 28 | Unfounded male infertility (in partner), stress urinary incontinence, recurrent urinary tract infection | Urethral taping and suspension |
| Tandon et al (1983) | 22 | Urinary incontinence, urinary tract infections | Urethroplasty |
| Pierce (1999) | 14 | Total urinary incontinence, bleeding, sexual assault, imperforate hymen | Urethroplasty |
| Ayan et al (2000) | 22 | Urinary urgency, stress urinary incontinence, suprapubic pain | Urethroplasty |
| | 28 | Stress urinary incontinence, urinary tract infection | Urethroplasty |
| Deniz et al (2002) | 32 | Dyspareunia, urinary loss, Mayer Rokitansky syndrome | Bladder flap |
| Okeke et al (2007) | 21 | Total urinary incontinence, sexual assault | Urethral & bladder neck plication |
| Di Donato et al (2008) | 32 | Infertility & dyspareunia, incontinence during coitus, microperforate hymen and urethral dilatation | Surgical dilatation of hymen with dilators |
| Brown et al (2012) | 27 | Bowel evisceration through perforated bladder secondary to rupture following urethral coitus, paraplegia | Repair of cystotomy via low midline incision<br>Suprapubic catheter & temporary closure of bladder neck<br>Plan for definitive reconstruction |
| Sakinci et al (2012) | 23 | Urethral intercourse in a patient with congenital adrenal hyperplasia, megalourethra, primary infertility, previous clitorovaginoplasty | Correction of megalourethra via plication of urethral wall<br>Creation of a functional neovagina |
| Verma et al (2012) | 27 | Urinary incontinence with coitarche | Diagnosis via indirect magnetic resonance fistulography |
| Ryckman et al (2013) | 13 | VACTREL, asymptomatic; incidental finding of urethral dilatation during EUA | Vaginal dilatation |
| | 16 | Primary amenorrhea & cyclic abdominal pain, oblique vaginal septum, dilated urethra | Resection of oblique septum & vaginal dilatation |