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### Case report

# Asymptomatic urethral intercourse associated with Mayer Rokitansky Küster Hauser syndrome type II: A case with crossed fused renal ectopia and scoliosis



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#### KEYWORDS

Urethral coitus;  
Mullerian anomalies;  
CT;  
Crossed renal ectopia;  
Scoliosis

#### Abstract

**Introduction:** Urethral coitus is very rare with significant consequences and less than 30 cases were reported in the literature. It is most commonly associated with Mullerian anomalies.

**Observation:** We report a case of 28 years married woman with Mullerian agenesis and associated anomalies who was engaged in urethral coitus and urethral dilation was detected on examination under anesthesia. The patient did not accept vaginoplasty after she learned that it would not be a cure for her infertility problem.

**Conclusions:** Urethral coitus is very rare with few cases reported. Woman diagnosed with MRKHS II should be assessed for associated abnormalities especially skeletal and renal ones. Urinary incontinence associated with coitus and dyspareunia with Mullerian anomalies should raise the suspicion of urethral coitus. Follow-up is important to avoid urinary tract infections if the female rejects surgery and is satisfied with intra-urethral coitus. Mullerian anomaly should be corrected to allow vaginal intercourse if possible.

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#### Introduction

Urethral coitus is very rare, commonly associated with Mullerian agenesis (Mayer-Rokitansky-Hauser-syndrome I and II types). These anomalies usually present prior to coitarche with amenorrhea. Most cases reported in the literature occur in the mid-twenties [1,2]. Most cases presented with urinary incontinence during coitus and recurrent urinary tract infections.

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Urethral coitus is a serious health condition because it can lead to urinary system problems. This may be attributed to sexual abuse or ignorance of the female [2].

Mayer-Rokitansky-Hauser-syndrome type I (MRKH syndrome type I) is an isolated Mullerian agenesis while Mayer-Rokitansky-Hauser-syndrome type II (MRKH syndrome type II) is Mullerian duct aplasia with anomalies especially renal anomalies and skeletal malformations, mainly vertebral and less commonly cardiac and otologic anomalies [3].

We present a case of asymptomatic urethral coitus associated with MRKHS type II.

### Case report

A 28 years old, sexually active female, presented to our department on 8/2015 complained of infertility after one year of marriage with regular coitus and primary amenorrhea. She complained of dyspareunia with no urinary incontinence or recurrent urinary tract infections which might be related to ignorance of the patient.

Clinical examination revealed normal looking female about 170 cm height, normal breast development and pubic, axillary hair well developed. Her external genitalia looked normal for female. Karyotyping was done revealed 46, XX. Hormonal profile was normal for female complete urine analysis was normal.

Transabdominal Ultrasound revealed absent uterus, cervix, vagina but the ovaries were identified. CT was done by the patient for associated anomalies revealed same picture of ultrasound with crossed fused renal ectopia shifted to right side with lower left one and left fifth lumbar hemi vertebra with right sided lumbar scoliosis, while otherwise was normal (Fig. 1).

Diagnostic laparoscopy and examination were performed under general anesthesia. Significant urethral dilatation with inflamed and bleeding on manipulation external urethral meatus were detected suggesting urethral intercourse and she was not aware of this issue. Urethral diameter was measured with Hegar dilator revealing 2.5 cm diameter (Fig. 2). Rectal examination did not reveal the uterus, cervix or vagina.

Laparoscopy revealed absent uterus with large Douglas pouch and Mullerian ducts remnants. Both her ovaries were seen with fimbria ends of the fallopian tube (Fig. 3).

The patient did not accept corrective operation because infertility cannot be corrected afterwards and she was not complaining of this issue. She preferred nonsurgical management; so, she was advised to use dilators on the vaginal dimple for half an hour to 2 h per day if she desired that, with prophylaxis against urinary tract infections.

### Discussion

The first case of intraurethral intercourse was reported in 1965 [3]. Approximately, less than thirty cases were documented worldwide till now [4–12]. Most cases were diagnosed in early age of twenties or early thirties [2,13–22]. The complains are usually dyspareunia and urinary incontinence during coitus [18–21,23]. Also, primary infertility as in our case was reported by many authors [2,14–16]. Traumatic urethral intercourse including woman with normal female anatomy [17], and imperforate hymen [1] was reported. It is mostly associated with Mullerian anomalies as in our case [21,22].

Although our case was presented with infertility and dyspareunia in the absence of incontinence despite repeated urethral intercourse probably due to less urethral dilatation.

As urethral dilation increases, continence could not be maintained. Urinary incontinence is observed with urethra dilated to 120 F (4 cm) in diameter. Surgical approach is recommended as conservative management fails. The resolution of incontinence has been described [2,17,19,21] indicating that urethral anatomy can restore its normal structure and function with time. The long-term integrity was not documented, and needs further investigation. Urethral plication was described in these cases [1,2,13,17–20,22].

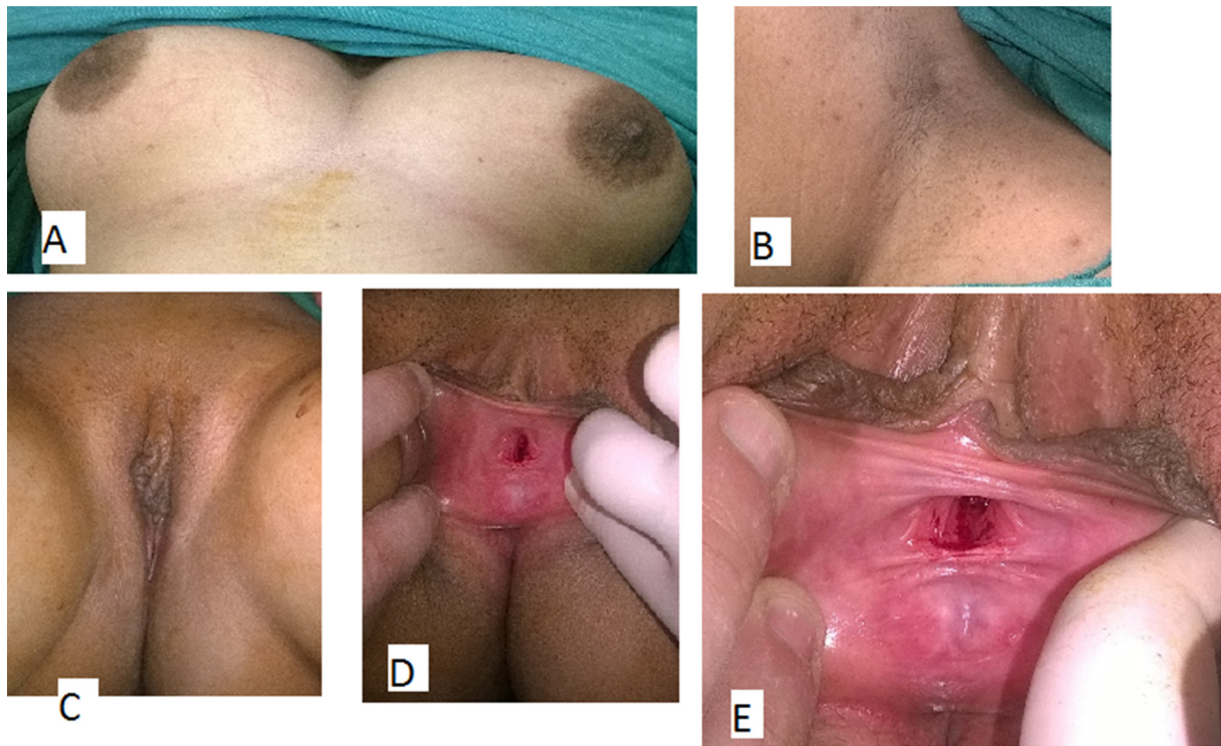
Other reported treatment is the creation of a vaginal orifice for intercourse [16], allowing the urethra to improve with rest or sphincter exercises [2]. This patient desired to continue urethral intercourse. As reported with other studies, some patients find urethral intercourse satisfactory with no problems resulting from it [14].

The American Academy of Pediatrics include external gynecologic examination in the examination of children and adolescents with complete pelvic examination in adolescent ones with amenorrhea [24].

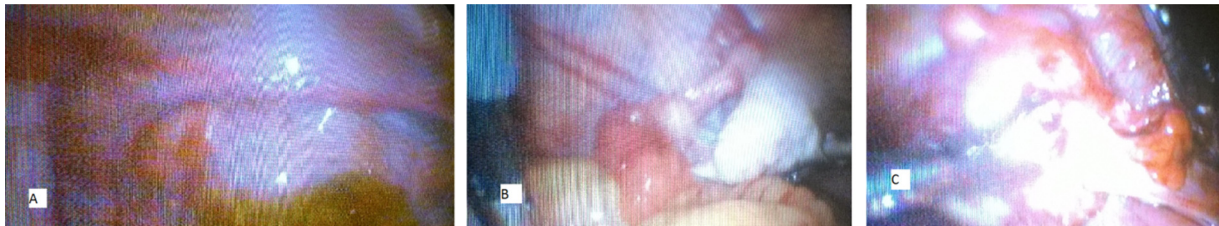
MRKH syndrome occurs 1 in 5000 women and it is the second most common cause of primary amenorrhea [25]. Treatment of Mullerian agenesis includes management of the physical findings and



**Fig. 1** CT revealed right kidney higher than the left (a), crossed fused renal ectopia (b) and right side scoliosis and left fifth lumbar hemi vertebra (c).



**Fig. 2** Examination of the patient revealed normal breast (a), normal axillary hair (b), normal external genitalia and pubic hair (shaved) (c), single opening in the vulva which is mega urethra with inflamed edges and bleeding on manipulation (d, e).



**Fig. 3** Laparoscopy showed Mullerian duct remnant (a), right ovary and fimbria (b) and left ovary with fimbria (c).

psychological support. Treatment consists of creating a neovagina for intercourse when they are ready for coitus. Treatment may be nonsurgical or surgical. Nonsurgical techniques are the first-line approach by plastic specially designed vaginal dilators or by fingers (Franck's dilator method) where a physician (and then woman herself) applies the dilator, which progressively widens the vagina. This procedure should be continued for six weeks to several months [26].

There is no consensus about which plastic surgical technique is the best one. Females will use vaginal dilators after surgery to enhance success. These females do not have a functional uterus and so they will have no children (infertile) except using in vitro fertilization of own eggs and surrogate pregnancy. The risk of passing the disease to off spring exists [27].

Patients with MRKH syndrome II who had renal anomalies as in our case have an increased risk to urinary tract infections and renal stones. Doctors should follow them for infection and prescribe antibiotics when needed. This is very important especially in cases with single functioning kidney to avoid losing it. Skeletal

abnormalities may need surgery, physical therapy and medical management according to the type and complains [27].

### Conclusion

Intercourse through the urethra is so rare with few reported cases. Risks associated as urinary tract infection can occurs. Coital urinary incontinence and dyspareunia with Mullerian anomalies should raise the suspicion of urethral coitus. They should be checked regularly for urinary tract infections on urine analysis and prescribe antibiotics when needed or using long term suppression if the patient refused corrective vaginoplasty. Many are asymptomatic and have satisfying intercourse while continent. Counseling these patients is important, especially when vaginal coitus is difficult. Where possible, the Mullerian anomaly should be corrected to allow vaginal intercourse.

### Conflict of interest

Nothing to declare.

## Funding

This study was not funded.

## Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

## Informed consent

Informed consent was obtained from the patient included in the study.

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