African Journal of Urology (2017) **23**, 48–51

African Journal of Urology

Official journal of the Pan African Urological Surgeon's Association
web page of the journal

www.ees.elsevier.com/afju
www.sciencedirect.com

Case report

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

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Received 6 January 2016; received in revised form 28 February 2016; accepted 20 March 2016
Available online 21 July 2016

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.

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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Peer review under responsibility of Pan African Urological Surgeons' Association.

http://dx.doi.org/10.1016/j.afju.2016.03.005

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

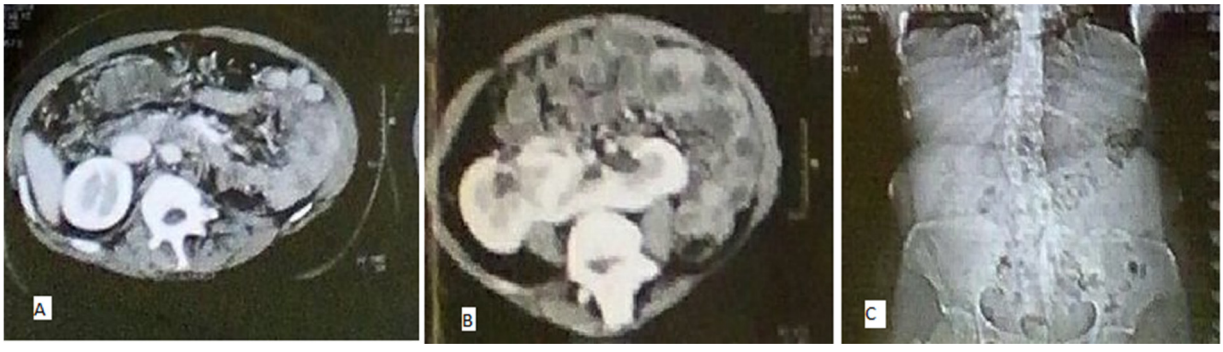

Fig. 1 CT revealed right kidney higher than the left (a), crossed fused renal ectopia (b) and right side scoliosis and left fifth lumbar hemi vertebra (c).