

Case Report

Urethral coitus associated with vaginal agenesis

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Abstract

Urethral coitus is a rare condition seen especially in cases with vaginal agenesis or hymen imperforatus. A female patient with sexual dysfunction and primary amenorrhea diagnosed with urethral coitus is presented in this report. These cases present recurrent urinary tract infections, dysuria, and dyspareunia besides primary amenorrhea. There was no megalourethra or vaginal introitus in our case. The patient did not accept corrective operation because infertility cannot be corrected afterwards. In this report, we present a case of urethral coitus associated with vaginal agenesis.

Key words:

urethral coitus, vaginal agenesis, clitoral hypertrophy

Introduction

Urethral coitus is a rare condition seen in cases with vaginal agenesis or hymen imperforatus. Urethral coitus is a serious health condition because it leads to especially urinary system problems [1,2]. This may be attributed to sexual abuse or ignorance of the partner. Vaginal agenesis is frequently seen with Mayer-Rokitansky-Küster-Hauser Syndrome [3]. Early diagnosis is important in order to have healthy sexual life and to avoid urinary system problems. Septate vagina and other anomalies of hymeneal orifice are predisposing factors at the time of initiation of sexual activity. Urethral intercourse in the female is very rare. When urethral coitus occurs, it is usually in association with rape or with vaginal atresia or other hymeneal anomalies.

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Case presentation

A 23 years old female married for eight months was admitted to the hospital with the complaint of primary amenorrhea and sexual dysfunction. The patient had premature secondary sexual characteristics such as clitoral hypertrophia, enlargement of urethral meatus and vaginal agenesis (Figure 1). Distal urethra was measured with hegar dilator no:16, proximal urethra was dilated with hegar dilator no:8. Uterus was measured 19x31x49 mm in diameter with ultrasonography. Endometrial line and ovaries could not be seen accurately with ultrasonography. Hormone profile was FSH: 102 mIU/ml, LH: 54.9 mIU/ml, PRL: 24.2 ng/ml, TSH: 0,78 uIU/ml fT3: 1.52 pg/ml, DHEA-S: 328 ug/dl. Intra venos pyelography was normal. Karyotype was 46 XX. Routine laboratory tests were in normal range except 10-15 leucocytes in urine sediment. Uterus and bilateral tubas were hypoplastic, and ovaries seemed sclerotic during diagnostic laparoscopy (Figure 2). Because of clitoral hypertrophy, partial clitorectomy was performed.

The patient did not accept vaginoplasty after she learned that it would not be a cure for her infertility problem. We prescribed hormone replacement therapy.

Figure 1.



Clitoral hypertrophy, urethral meatus enlargement and vaginal agenesis.

Discussion

Urethral coitus with vaginal agenesis or hymen imperforatus is rarely seen [1, 2]. This may be attributed to sexual abuse or ignorance of the partner. Vaginal agenesis is frequently seen with Mayer-Rokitansky-Küster-Hauser Syndrome [3]. There are cases having urethral coitus besides normal vaginal anatomy. Tandon and Saksena reported a case having urethral coitus after one year vaginal intercourse [4]. Although urethral coitus is frequently seen in cases with vaginal agenesis or hymen imperforatus, cases that underwent ambiguous genitalia correction operations having urethral coitus due to vaginal tightness have also been reported [5]. There were 26 cases in the literature and only two of them were adolescents [6]. To the best of our knowledge, our case, 23 years old adult, is the 27th Mayer-Rokitansky-Küster-Hauser syndrome in the literature. In our case, karyotype was normal. However, this is not sufficient in these cases because there may be partial Y chromosome insertion to the X chromosome [SRY translocation]. If this is the case, mullerian inhibiting factor will inhibit the development of the structures which are originated from the mullerian canal. Moreover excess androgen causes cliteromegaly. SRY translocation can be diagnosed by FISH tech-

nique. Due to malignancy potential of streak gonads in SRY translocation, gonads should be removed. If there is no SRY translocation the case could be congenital adrenal hyperplasia. The rarest etiology for this case is single gene disorders. We could not perform a FISH analysis in this case because of technological limitations in our institution. We wanted to refer the patient to a genetic laboratory for FISH analysis, and because nothing could be done to treat infertility, the patient did not accept FISH analysis. Laparoscopy is the best method in diagnosis and treatment of the mullerian canal abnormalities, and we performed laparoscopy for this patient. Urethral coitus should be suspected if there is recurrent urinary tract infections, dysuria, dyspareunia with vaginal agenesis. Urinary infections and incontinence in urethral coitus are inevitable complications in the long term[7]. Urethral sphincter, 3-4 cm proximal to the urethra, plays a role in urinary continence. As dilation increases in the sphincter, continence could not be sustained. It is reported that urinary incontinence is observed when urethra is dilated to 120 F in diameter [4]. At this level of dilation, urethra stays in atonic condition and cannot return to original caliper. Urinary incontinence is not observed when urethral smooth muscles and elastic tissue gain their tonicity [8].

Figure 2.



Laparoscopic view of hypoplastic uterus, tuba and streak gonad.

In our case, distal urethra was dilated but as proximal urethra had normal caliper and sphincter structure, there was no urinary incontinence. Distal urethra was measured with

hegar dilator no:16, which is 16 mm in diameter. From the anamnesis of the patient we learned that they had successful sexual intercourse despite dyspareunia. However, patient's husband was not aware of this unusual condition. When urethral caliber is more than 120 F, surgical approach is recommended as conservative treatment fails in that situation. In the surgical approach, urethra is mobilized and cut, then tightened and sutured. Paraurethral and ureteropelvic ligaments can be brought together with sutures in order to support midurethral complex [4]. Another surgical approach is making neourethra from proximal urethra and leaving distal urethra as vagina by means of a bladder flap [1]. In our case, because the surgery does not improve fertility, patient did not accept vaginoplasty. We only corrected clitoral hypertrophy. The patient was discharged from hospital with prescription of urinary infection and hormone replacement therapy.

In conclusion, rarely observed urethral coitus may have serious consequences. Mullerian anomalies may lead to many complications especially for sexual activity in women. Mullerian anomaly with symptoms of urinary incontinence, dyspareunia, frequent urinary tract infection, urethral coitus should come to mind first. Karyotype analysis is not sufficient in these patients and translocation can be diagnosed by FISH technique. Because of possible malignancy, streak gonads should be removed in case of SRY translocation. If possible, mullerian anomaly must be corrected in a way that enables vaginal intercourse.

Conflict of Interest

Authors declare no conflict of interest

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