Case Report

Iatrogenic urethrovaginal fistula with transverse vaginal septum presenting as cyclical hematuria

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ABSTRACT

We present a case of iatrogenic urethrovaginal fistula with transverse vaginal septum. The patient presented with cyclical hematuria and infertility. The vagina was blind-ending. The magnetic resonance imaging (MRI) showed normal uterus with transverse vaginal septum. The cystoscopic examination during cyclical hematuria revealed bloody efflux through a small fistula below the internal urethral sphincter. Vaginoplasty and repair of the urethrovaginal fistula was done. The vagina was reconstructed using an amniotic mould. The report emphasizes the importance of MRI and cystoscopy in diagnosing such rare and complex anomalies.

Key words: Cyclical hematuria, Vaginal septum, Urethrovaginal fistula

INTRODUCTION

Müllerian anomalies are congenital defects of the female reproductive tract resulting from failure in the development of the Müllerian ducts and their associated structures. Their cause is not known, but is currently believed to be multifactorial. Symptoms appear principally during adolescence or early adulthood, and affect the reproductive capacity of these women. When clinically suspected, investigations leading to diagnosis include imaging methods such as hysterosalpingography, ultrasonography, magnetic resonance imaging, and cystoscopy.

CASE REPORT

A 25-year-old nulliparous lady, presented to the gynecology OPD with complaints of inability to conceive. She was married for 7 years, cohabiting since 5 years. At 16 years of age, she consulted a local doctor in her village for primary amenorrhea and cyclical pain in abdomen, where some operative procedure was done. Since then the patient has passage of blood mixed urine periodically every month for 2 to 3 days with suprapubic abdominal pain. She had thelarche at 13 years and pubarche at 15 years of age. There was no urinary incontinence or dysparunia.

On general physical examination, secondary sexual characters were present. On local examination, external urethral meatus appeared dilated, and there was blindly ending vagina approximately 3 cm in length. On per rectum examination, uterus appeared normal in size and there was no bulge felt below it to suggest any collection. On investigations, the ultrasound showed normal uterus with endometrial thickness 9.5 mm, ovaries were normal, and bilateral kidneys were also normal. MRI of abdomen showed normal uterine body and cervix with small amount of fluid in endometrial cavity, endocervical cavity, and in upper part of vagina; below that there was a transverse vaginal septum of about 3.5 cm in thickness. No fistulous tract or communication was visualized between the genital and urinary tract. [Figure 1] On cystoscopy, a fistulous opening of approximately 2 mm was seen below the internal sphincter of urethra through which menstrual blood was seen coming out; the bladder and ureteric orifices were normal. Micturating cystourethrography also failed to provide any idea about the fistulous tract. Vaginoplasty and repair of fistulous communication between the genital tract and the urethra was planned for the patient.

The woman was subjected to operation using

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abdominoperineal approach. On laparotomy, uterus was normal in size, bilateral tubes were present, and ovaries were normal. The methylene blue dye was injected into the fundus of the uterus; dye was visualized coming out of urethra by the side of the indwelling catheter. From the perineal approach, a transverse incision was made through the vault of the short vagina, the dissection was done upward in the connective tissue between the bladder above and rectum below and vaginal space was created. After sharp and blunt dissection, cervix was visualized as dye was coming through it. The cervical os was dilated with Hegar's dilator. The urethrovaginal fistulous tract was identified and repair was done in two layers. Over the raw area of vagina, the amnion graft was employed over a sponge mould with condom. An indwelling catheter was applied. The mould change was done after a week; the amnion graft had successfully taken up. Dentine moulds were made and applied. Patient was taught how to change the mould daily. The patient was found to be fine at follow-up after a month, with no hematuria and normal menstruation. She was advised to have regular intercourse. She was explained about the fertility period and asked to come at 3 monthly follow-up.

DISCUSSION

The incidence of mullarian anomaly is estimated to be between 0.1% and 3%.[1] It ranges from imperforate hymen to uterine agenesis. In females, when a mullerian duct becomes obstructed, the patient may present with an abdominal mass and dysmenorrhea. If the patient is not treated in a timely fashion, the consequences can be severe, extending even to infertility.

In this case, there was transverse vaginal septum in the upper part of vagina leading to hematocolpus and amenorrhea. An iatrogenic fistulous tract was probably created which led to cyclical hematuria. Magnetic resonance imaging (MRI) is the mainstay in imaging for the evaluation of mullerian agenesis and is considered a very useful diagnostic tool. [2] In our case, MRI determined the precise mullarian anomaly giving us the details regarding the presence of normal cervix with precise length of vaginal septum to help us judge the prognosis and success of the operative procedure, but we got no information about the fistulous communication as it was quite fine and narrow. The site of the opening of fistulous tract in the urethra was apparent on cystoscopy and led to the assumption that the other end tract opened in the vagina. Therefore, apart from MRI, cystoscopy played a major role in determining the anomaly in this case.

Upon literature review of cases with genitourinary fistula, we found one previous report of urethrovaginal fistula with imperforate hymen.[3] The fistula in that case, however, was congenital, unlike our case in which it was iatrogenic in all probability. It also presented with cyclical hematuria

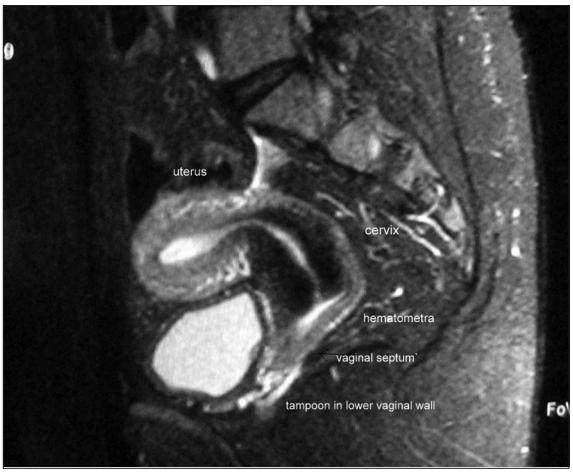

Figure 1: The MRI shows uterus with cervix and upper 2 cm of vagina containing some blood products. There is a septum between the upper and lower part of vagina (the lower part identified by tampon in vaginal canal)

with no incontinence of urine due to imperforate hymen. Congenital vesicouterine, cervicovesical, and vesicovaginal fistula associated with other mullarian anomalies has also been reported in literature.[4-6] Iatrogenic fistulous communication between the genital tract and the urinary tract, known to occur as a complication of cesarean section, has been described in literature as "Youssef's syndrome." The uterine side of the fistulous tract is above the uterine isthmus. It is characterized by cyclical hematuria, absence of vaginal bleeding, and complete urinary continence.[7,8] In all these reports in which there was a fistulous communication between the urinary and the genital tract, cystoscopy, hysterosalpingography, and dye test were required for confirmatory diagnosis and only computerized tomography (CT) or MRI did not give the full clinical picture.

This report highlights the diagnostic and therapeutic challenge faced by the clinician in dealing with a fistulous genital and urinary tract communication with obstructing transverse vaginal septum. Increasing awareness of this rare entity calls for more meticulous evaluation before any surgical intervention in patients with complex genitourinary anomalies.

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