Youssef’s Syndrome following Cesarean Section

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Youssef’s syndrome is characterized by cyclic hematuria (menouria), absence of vaginal bleeding (amenorrhea), and urinary incontinence due to vesicouterine fistula (VUF), the least common of the urogynecological fistulas. Youssef’s syndrome has a variable clinical presentation. A vesicouterine fistula is an abnormal pathway between the bladder and the uterus. The most common cause is lower segment Cesarean section. Conservative treatment may be appropriate in some cases, but surgery is the definitive treatment. Vesicouterine fistula should be suspected in cases presenting with urinary incontinence even years after Cesarean section. Diagnostic tests as well as necessary appropriate surgery should be performed on cases with suspected vesicouterine fistula. We present a 40-year-old multiparous woman with vesicouterine fistula after primary Cesarean section; she presented with urinary incontinence, hematuria, and amenorrhea 1 year after the birth. Here, we discuss our case with the help of previously published studies found in the literature.

1. Introduction

A vesicouterine fistula is an abnormal pathway between the bladder and the uterus. The first case was reported by Knipe and colleagues in 1908. Vesicouterine fistula is the least common of all the urogenital fistulas, representing 1–4% of all cases [1]. The vast majority of vesicouterine fistulae are secondary to iatrogenic causes, the most common being lower segment Cesarean section [2]. The less frequent causes include induced abortion, dilatation and curettage, vaginal birth after previous Cesarean section, obstructed labor, forceps delivery, placenta percreta, migrated intrauterine contraceptive device, and brachytherapy [2]. The main symptoms of VUF (vesicouterine fistula) are urinary incontinence, cyclic hematuria (menouria), amenorrhea, and urinary tract infection. Most of the cases present in a delayed fashion, from weeks to years after the inciting event [3]. In such cases, the diagnosis is mainly established by clinical detection of urine or dye passing through the external cervical os or by means of a hysterosalpingogram or micturating cystourethrogram, which will demonstrate the fistulous communication [3]. Conservative treatment may be appropriate in some cases, but surgery is the definitive treatment. Transabdominal, laparoscopic, or robotic methods can be used. We present a case with vesicouterine fistula after primary Cesarean section; the patient presented with urinary incontinence, hematuria, and amenorrhea 1 year after giving birth.

2. Case Presentation

A 40-year-old multiparous female patient presented to our clinic with urinary incontinence, hematuria, and amenorrhea a year after giving birth by Cesarean section due to fetal distress in our clinic. Patient history had no highlights except a lower segment Cesarean section by Pfannenstiel incision due to fetal distress in previous year. Transvaginal USG (ultrasound) showed normal uterine and ovary structures. Noticeable urine smell showed that the case had moist vulvar area and speculum examination showed a leakage consistent with hematuria that flowed from cervical os (Figure 1). After administering methylene blue through insertion of a urethral Foley catheter, there was an active outflow of methylene blue from cervical os. IV (intravenous) pyelography showed
no anatomical defects or significant leakage (Figure 2). Cys-
toscopy found the fistula focus on posterior bladder wall
(Figure 3). During cystoscopy, catheter pushed through the
fistula on posterior wall of bladder came out of cervical
external os (Figure 4). After informing the patient and her
relatives about the clinical situation thoroughly, laparotomy
was decided. Since the patient wanted to keep her fertility
intact, an extraperitoneal approach by Pfannenstiel incision
was used to reach the bladder. The bladder was removed
using O’Connor method and following the detection of fistula
focus between uterus and bladder, necrotic fistula tract was
removed and bladder and uterine mucosa were sutured shut
by 2 layers of 2-0 polyglycolic sutures. Leakage control was
done by administering sterile saline solution to the bladder
and no leakage was seen in suture lines. An 18-Fr Foley
catheter was placed into urethra and a drain was placed
on extraperitoneal area. The drain placed on abdomen was
removed 5 days after and urethral Foley catheter was removed
14 days after the spontaneous urination was normal and no
leaks were detected.

3. Discussion

In 1957, Youssef described a syndrome comprising of cyclic
hematuria, amenorrhea, menouria, and complete urinary
incontinence in a patient who had lower segment Cesarean
section (LSCS) [4]. The VUFs are among the least common
urogynecological fistulas. The VUF also occurs following
high vaginal forceps-aided delivery, external cephalic version,
curettage or manual removal of the placenta, placenta per-
creta, myomectomy, uterine rupture due to obstructed labor,
uterine artery embolization, perforation of an intrauterine
device, and brachytherapy for carcinoma of cervix. The
LSCS is the single most common cause of VUF [3]. Amen-
orrhea, cyclic hematuria without urinary incontinence in
combination with a history of LSCS, has been described as
pathognomonic of VUF [5]. The clinical presentation is often
nonspecific and findings on examination classically used to
depict the fistula may be negative, leading to considerable
delay in diagnosis [6]. The VUF may not manifest with con-
stant urinary incontinence because of a functional sphincter
at the internal uterine os. Urinary incontinence occurs if the
level of the VUF is at or below the internal os or if the os is
incompetent [5]. In our case scanty urine leak occurred even
in the presence of competent os with fistula communicating
with uterus above isthmus.

The diagnosis of VUF is often confirmed by imaging
studies and cystoscopy. Cystoscopy, even when repeated,
can fail to confirm the fistula [4]. Methylene blue instilled into the uterine cavity or through the urethra or through catheterization of a visible lesion in the bladder wall can confirm the fistula. This test, however, does not show directly the fistulous tract and its specific location. Moreover, this test can be negative in patients with a long and tortuous tract [4]. In radiological studies, both cystography and hysteroscopy have been used in the diagnosis of VUFs. In Tancer's review of published reports, he found that hysteroscopy was the most reliable diagnostic technique [5]. Intravenous urography can show the fistula when contrast medium enters the vagina, but distinguishing vesicovaginal and vesicouterine fistulae is difficult.

Although VUFs are difficult to diagnose using USG, Park et al. reported that sonography can demonstrate the fistulous tract as double echogenic lines between the endometrium of the anterior wall of the uterine body and the mucosal layer of the posterior wall of the bladder [3]. However, sonography has inherent difficulty in differentiating the VUF tract from different patterns of a noncomplicated Cesarean scar [6]. Also transvaginal sonography is recently being used for diagnosis [7]. Magnetic resonance imaging has now become the first reliable diagnostic technique [5]. In intravenous urography, he found that hysteroscopy was the most prevalent due to changes in modern obstetrical care.

Vesicouterine fistulas are uncommon, yet they are becoming more prevalent due to changes in modern obstetrical care. They should always be kept in mind in patients with a history of Cesarean section or who have experienced a gynecological procedure associated with signs of hematuria and/or urinary leakage.

Disclosure
Ozer Birge is the guarantor of submission.

Conflict of Interests
The authors declare no conflict of interests.

Authors’ Contribution
Ozer Birge made substantial contributions to conception and design, acquisition of data, analysis and interpretation of data, drafting the paper, revising it critically for important intellectual content, and final approval of the version to be published. Ertugrul Gazi Oz bey and Mustafa Melih Erkan, and Ilk en Kayar made substantial contributions to conception and design, acquisition of data, analysis and interpretation of data, drafting the paper, revising it critically for important intellectual content, and final approval of the version to be published. Ertugrul Gazi Oz bey and Deniz Arslan made substantial contributions to conception and design, acquisition of data, analysis and interpretation of data, drafting the paper, revising it critically for important intellectual content, and final approval of the version to be published.

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