

## Vesicouterine Fistula (Youssef's syndrome): Imaging Findings Case Report

## Mesane-Uterus Arasında Fistül (Youssef's sendromu): Görüntüleme Bulguları Olgu Sunumu

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### ABSTRACT

We report a case of vesicouterine fistula (VUF) secondary to cesarean section. This is a rare complication of cesarean section. Diagnosis was made clinically, radiologically, and endoscopically during the postoperative period. Conservative management with cystoscopic fulguration for 4 weeks failed. Hence the fistula was repaired surgically by an abdominal approach.

**Key words:** Urogenital tract, Imaging, Fistula

### ÖZET

Biz sezaryan operasyonuna bağlı mesane ile uterus arasındaki fistül olgusunu sunduk. Bu sezaryan operasyonunun nadir bir komplikasyonudur. Tanı operasyon sonrasında klinik, radyolojik ve endoskopik olarak konuldu. 4 haftalık sistoskopik fulgurasyon ile konservatif tedavi yetersiz oldu. Bu nedenle fistül abdominal yaklaşımla cerrahi olarak tamir edildi.

**Anahtar Kelimeler:** Ürogenital sistem, Görüntüleme, Fistül

## INTRODUCTION

Vesicouterine fistulas (VUF) is a rare condition, representing only 1 to 4% of all cases of urogenital fistulas (1). The majority of cases of VUF are iatrogenic, occurring mainly after cesarean section, usually a low segment incision (2). In Youseff's syndrome, which was first identified in 1957, VUF develops over the internal cervical os level, following lower segment cesarean section. The symptoms are amenorrhoea, infertility and menouria with the absence of urinary incontinence, while the cervix is intact (3). But in some cases the clinical presentation is nonspecific and findings on examination clasically used to depict the fistula are negative, leading to considerable delay in diagnosis (4). We describe the imaging findings in a 28 year old woman presented with the symptoms of the urinary incontinence and diagnosed as the vesicouterine fistula.

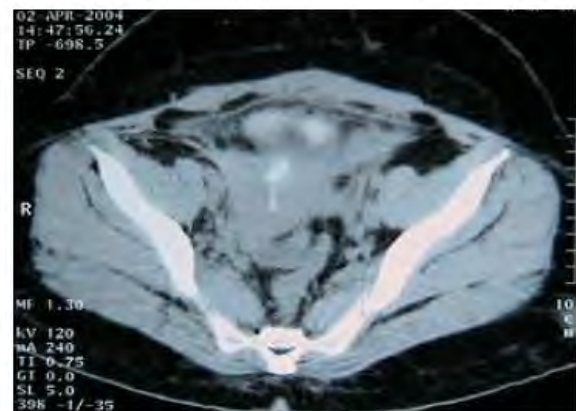
## CASE REPORT

A 28-year-old woman was referred to our urology department with symptoms of the urinary leak. In her personal history, the patient had undergone cesarean, using epidural anesthesia, two months ago. Because of the methylene blue test of the patient was positive, a vesicouterine fistula was suspected and the excretory urography was performed. In fifteenth minute of the excretory urography, the endometrial cavity was opacified (Figure 1). Then, pelvic computed tomography with intravenous contrast injection showed the uterine and the vaginal cavity opacification. But, no definite fistula was visualized (Figure 2). After one day, the hystero-graphy depicted the leakage of contrast material between the uterus and the bladder via 5 mm sized-fistula tract (Figure 3).

Cystoscopy demonstrated the open fistula and the fistula tract between the posterior wall of bladder and the anterior part of the isthmus uteri, so radiologic diagnosis was confirmed (Figure 4).



**Figure 1:** In 15th minute of excretory urography, endometrial cavity is opacify.



**Figure 2:** A,B, Pelvic CT with intravenous contrast injection shows uterine and vaginal cavity opacification. But, no definite fistula was visualized.



**Figure 3:** Hystero-contrast sonography depicts leakage of contrast material between the uterus and bladder via 5 mm sized fistula tract.



**Figure 4:** Cystoscopy demonstrates fistula open and fistula tract between the posterior wall of bladder and the anterior part of the isthmus uteri.

Cystoscopic fulguration of the fistula was attempted in our urology department and the patient used a transurethral Foley catheter for four weeks. Subsequently, at follow-up of the patient, there was no incontinence. But unfortunately, because of the cyclic hematuria (menouria) was appeared, the open surgical repair was performed.

## DISCUSSION

Vesico-uterine fistula is a rare but recognized complication of the lower segment cesarean delivery (2). Other causes include obstetrical-gynecological interventions such as uterine biopsy, radiation therapy, malign tumors, contraceptive device, pelvic adhesions, long labor, forceps delivery, abdominal pregnancy for perforation of the anterior wall of uterus and tuberculosis of genital tract (1, 2, 5). Delayed vesicouterine fistula formation may result from infection, devascularisation, clamping or hematoma of the urinary bladder (6). Tancer suggested that the most likely mechanism for vesicouterine fistula after lower segment cesarean section is occult injury to bladder during cesarean. The main symptom of a VUF is the urinary incontinence and it is usually immediate or occurs within a few days post low segment cesarean section. When the presentation is delayed, the combination of amenorrhea and cyclic hematuria in the absence of urinary incontinence has been described as pathognomonic of vesico-uterine fistula (2).

Youssef's syndrome is a common manifestation of VUF accounting for about 30% of presentations, and is characterized by apparent amenorrhoea, cyclic hematuria and urinary incontinence (3). After 1947, with more low segment cesarean being done, the occurrence of Youssef's syndrome has been increased (2). Symptoms depend on the level of the fistula, can be explained by the sphincteric mechanism of the uterine isthmus and the different pressure gradients. The shape and the diameter of the isthmus lumen change during the menstrual cycle. In normal menstruation, the blood to some extent distends the uterine cavity and this is followed by relaxation of the sphincter with rhythmic discharge of the blood (3,4). When a fistula is present above the isthmus, the blood passes through the fistulous opening into the bladder, no distension of the uterus takes place and the sphincter fails to relax. The result is

Youssef's syndrome that is amenorrhea with a patent cervical canal, periodic hematuria termed menouria by Youssef and the absence of urinary leakage through the vagina. Thus, if the fistula is above the uterine isthmus, cyclic hematuria ensues. If the communication is below the isthmus, the menstrual blood accumulates normally in the uterine cavity and when the sphincter of isthmus relaxes, the menstrual blood passes as it should through the cervix into the vagina. Conversely, when submitted to high pressure in the bladder, urine leaks through the fistula from the bladder into the uterine cervix and the vagina. However, occasionally both may occur and menouria can be associated with urinary leak through the vagina (3).

Youssef's syndrome is diagnosed by hystero-graphy, cystoscopy, vaginoscopy, cystography, excretory urography computed tomography and magnetic resonance imaging (2,3,7,8,9). Methylene blue test can confirm the fistula. This test is performed via catheterization of a visible lesion in bladder wall or via the urethra. In other technique, the methylene blue is instilled into the uterine cavity directly. Sometimes, this test can be negative related to the length and the tortuosity of fistulous tract. If the test is positive, the clinician suspect for fistula. In our patient, the test was also positive. For this reason, we turned towards the advanced investigations for fistula. Usually, the result of excretory urography are normal (3, 7). But, in our patient it was abnormal. The uterine cavity was opacified, however fistula tract was not seen. In spontaneous healed fistula the bladder contour may show 2 small radiological images of indentations of the the posterior vesical wall which are called 'parrot's beaks'. Hystero-graphy shows the fistulous tract as an abnormal communication between the uterus and the bladder (8). Transvaginal ultrasonography may show the disrupted bladder wall and bladder invaginated into uterine isthmus (4). Doppler ultrasound examination may assess the flow of fluids

between the 2 hollow cavities (10). Computerized tomography and MRI may show the patency of fistula; they may be both necessary sometimes in excluding the involvement of other organs (8,9,10). In addition MRI has a role confirming or ruling out the presence of bladder endometriosis which can be associated with VUF. Heavily T2-weighted single-shot RARE sequence which utilise long echo times (150-1000 ms) have been used to delineate fluid-filled structures (9).

The disease treatment options include conservative treatment as well as surgical repair. Rarely, patients refuse any kind of treatment because of the benignity of symptoms and prognosis of the disease. Conservative management by bladder catheterization for at least 4-8 weeks is indicated when the fistula is discovered just after delivery since there is good chance for spontaneous closure of the fistulous track. Hormonal management should be tried in women presenting with Youssef's syndrome. Surgery is the mainstay and definitive treatment of vesicouterine fistulas after cesarean section. Patients scheduled for surgery should undergo pretreatment of urinary tract infections. Surgical repair of vesicouterine fistulas are performed by different approaches which include the vaginal, transvesical-retroperitoneal and transperitoneal access which is considered the most effective with the lowest relapse rate. Recently, laparoscopy has been proposed as a valid option for repairing vesicouterine fistulas. The endoscopic treatment may be effective in treating small vesicouterine fistulas. The disease may be prevented by emptying the bladder as well as by carefully dissecting the lower uterine segment (10).

Vesicouterine fistulae, despite being infrequent, are no longer a rare diagnosis and are most commonly secondary to lower segment cesarean section. With patient history and selected investigations diagnosis is relatively easy. The surgical repair of these fistula is standard treatment, especially with delayed fistulae

with achievement of total continence, and complete resolution of cyclic haematuria.

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