

Spontaneous Vesicouterine Fistula: A Case Report

Spontan Vezikoüterin Fistül: Olgu Sunumu

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Abstract

Vesicouterine fistulae (VUFs) are the least common type of urogenital fistulae and they are usually associated with lower segment caesarean section. The classic triad of VUF has been defined as urinary incontinence, amenorrhea, and cyclic hematuria. In the literature, spontaneous VUF without a history of previous surgery or radiotherapy has not been reported before. In this report, we present a female patient who presented with continuous urinary incontinence and was diagnosed with VUF. The patient was managed with cystectomy and hysterectomy and ileal conduit diversion. To our knowledge, this is the only reported case of spontaneous VUF.

Keywords: Cystectomy, hysterectomy, urinary incontinence, urogenital fistula, vesicouterine fistula

Öz

Vezikoüterin fistül (VUF) ürogenital fistüllerin en az karşılaşılanıdır ve genellikle alt segment sezaryen ile ilişkilidir. VUF klasik triadı üriner inkontinans, amenore ve siklik hematüri olarak tanımlanmıştır. Literatürde geçmişinde cerrahi ya da radyoterapi öyküsü olmayan spontan VUF daha önce rapor edilmemiştir. Bu yazıda, tarafımıza sürekli idrar kaçırma ile başvuran ve VUF saptanan bir kadın hasta sunuldu. Hasta sistektomi, histerektomi ve ileal konduit ile tedavi edildi. Bildiğimiz kadarıyla, bu spontan VUF bildirilmiş tek olgudur.

Anahtar Kelimeler: Sistektomi, histerektomi, üriner inkontinans, ürogenital fistül, vezikoüterin fistül

Introduction

Vesicouterine fistula (VUF) is an abnormal connection between the bladder and the uterus and it is the least common fistula among urogenital fistulae with an incidence of 2-4% (1). It has been reported that VUFs were generally seen after obstetric operations and 83% of the VUFs were observed after caesarean section (2).

The classic triad of VUF is amenorrhea, cyclic hematuria (menouria) and discrete leakage of urine from the vagina (1). Observation of methylene blue at os cervix which is given during cystoscopy or with a urethral catheter or detecting opaque passage to the bladder or uterus during either hystero-graphy or cystography are the main diagnostic methods used for detecting VUF. Although conservative treatment methods such as observation, fulguration or hormonal therapy can be used

in the management of VUF, definitive treatment is appropriate surgery (3,4).

Spontaneous VUF without a history of previous surgery or radiotherapy has not been reported before. We present a female patient who was admitted with the complaints of continuous urinary incontinence and was diagnosed with VUF. The patient was managed with cystectomy and hysterectomy and ileal conduit diversion. To our knowledge, this is the only reported case of spontaneous VUF.

Case Presentation

A 49-year-old female patient admitted to our department with the complaint of continuous urinary incontinence that started one month ago. During vaginal examination, continuous

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leakage of urine from the vagina was detected and there was no cystocele or rectocele. Ultrasonography showed bilateral renal dilatation; the bladder was empty and the bladder wall thickness was increased. Her medical history revealed that she had diabetes mellitus for 5 years and she went through menopause 4 years ago. Her obstetric history included 3 normal spontaneous vaginal deliveries without any prolonged or difficult labour. Her physical examination showed a diabetic ulcer of the left foot. Laboratory examinations were as follows: creatinine: 2.01 mg/dL, K: 6.1 mmol/L, and Hb: 7.7 g/dL. The patient was hospitalized after placement of a urethral catheter.

Gynecological examination and transvaginal ultrasonography were performed and the adnexa and uterus were found to be normal without any pathology. Endocrinology consultation was requested for her HbA1c level being 9.5% and insulin treatment was started. Then, the patient underwent cystoscopy which showed that right (Figure 1a) and left (Figure 1b) ureteral orifices were lateralized and widened that the cystoscope was negotiated easily into the orifice. Under general anesthesia, the bladder capacity was found to be 50 cc. A 0.5 cm fistula with edematous and hyperemic mucosal borders was observed between the posterior wall of the bladder and dome of the bladder (Figure 1c). Simultaneous cystography showed the passage of iodine solution to uterine cavity (Figure 1d). Biopsies with transurethral resection were taken from hyperemic areas around the fistula. Bilateral hydronephrosis was detected during retrograde urography (Figure 1e), therefore, bilateral nephrostomies were inserted. Pathological examination of the biopsy specimens showed chronic inflammation and necrosis. Purified protein derivative skin test, acidoresistant bacilli microscopy and urine culture for tuberculosis were negative.

Treatment options were explained to the patient. Since she had a low bladder capacity, diabetes mellitus and renal failure, cystectomy, total abdominal hysterectomy and bilateral salpingo-oophorectomy and ileal conduit diversion were performed. Final pathological examination of the cystectomy and fistula track specimen showed chronic inflammation. Final pathological examination of the total abdominal hysterectomy and bilateral salpingo-oophorectomy showed chronic cervicitis with squamous metaplasia. During the operation and postoperative period, no complications were observed. After the procedure, the patient's creatinine level decreased to 1.27 mg/dL on the postoperative 1st week.

Discussion

Vesicouterine fistulae are the least common type of urogynecological fistulae. Caesarean section is the most common cause of these fistulae. Foreign bodies such as intrauterine devices, uterine rupture, placental anomalies,

uterine artery embolization, traumatic bladder cauterization, pelvic surgery, pelvic cancer, brachytherapy, pelvic external irradiation, and infectious and inflammatory diseases are the etiological factors that can cause VUF (3). In the present case, none of the above mentioned etiological factors were present. The only concomitant disease during her admission was diabetes mellitus, nephropathy, and contracted bladder probably related to diabetes. To our knowledge, this is the only reported case of VUF without any etiological factors and occurring spontaneously.

Cystography or hysterosalpingography can be considered as the gold standard in the diagnosis of VUF. Computed tomography can provide important information about the topography of the fistula tract. Magnetic resonance imaging (MRI) is a non-invasive method that produces detailed anatomical images without the use of contrast media causing allergic reactions or nephrotoxicity. MRI is able to identify the exact location and anatomy of the fistula and the presence of endometriosis within the bladder. In addition to these, transvaginal ultrasonography can also be used in the diagnosis of VUF (4). In our case, we performed cystography during cystoscopic examination under general anesthesia. This examination also enabled us to evaluate the bladder capacity and upper urinary tract dilatation.

Various kinds of approaches have been proposed for the treatment of VUF. Spontaneous closure of VUF with conservative approach has been reported (5). Surgical approach depends on the patient's desire for fertility as well as other surgical factors. If the patient does not desire to have further children, closure

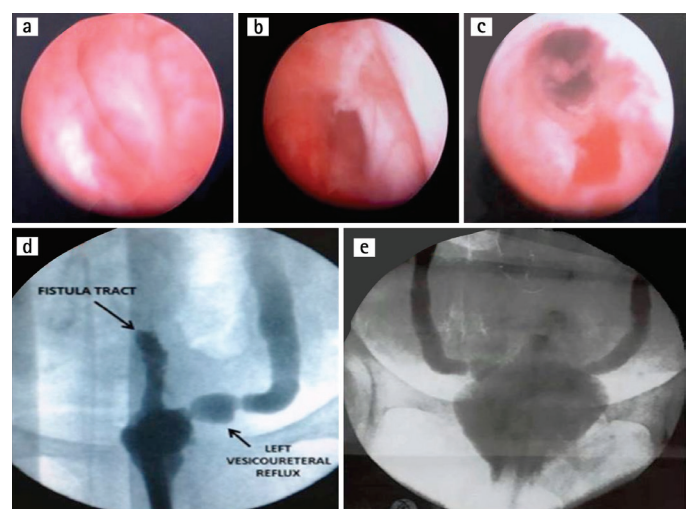


Figure 1. a, b) Right and left ureteral orifices lateralized and widened that a cystoscope was negotiated easily into the orifice, c) Fistula tract with edematous and hyperemic mucosal borders between posterior wall and bladder dome, d) Simultaneous cystography showed the passage of iodine solution to uterine cavity, e) Decreased bladder capacity and bilateral hydronephrosis was detected during cystography and retrograde urography

of the bladder site of the fistula tract can be performed with hysterectomy. If fertility is a major concern, uterine-sparing approaches are implemented. Laparoscopic and robotic surgery can also be used in VUF repair (4). Uterus-preserving approaches were not considered in our case because our patient was in menopausal age.

Cystectomy is widely used for invasive bladder cancer, however, patients with debilitating non-malignant bladder diseases, in whom conservative or minimally invasive treatment options have failed, may also undergo various forms of cystectomy and urinary diversion (6). There are mainly three alternatives for urinary diversion: abdominal and urethral diversions with either continent or incontinent forms and rectosigmoid diversions. Although augmentation cystoplasty and continent urethral diversions are preferred for the quality of life in these patients, there are contraindications for these complex forms of diversions such as debilitating neurologic and psychiatric illnesses, limited life expectancy and impaired liver or renal function (7). Augmentation surgery or continent diversion was not considered because our patient had nephropathy. Therefore, total abdominal hysterectomy and bilateral salpingo-oophorectomy and ileal conduit diversion were performed after taking the patient's informed consent. After the procedure, renal function recovered and hydronephrosis was resolved in both kidneys. Pathological examination revealed chronic inflammation in the bladder and vesicouterine fistula track which ruled out any malignancy or an etiology which was not diagnosed preoperatively.

Although an etiological factor exists in most of the cases, VUF can be found in women with urinary incontinence and continuous urinary leakage per vagina, as well as other urogenital fistulae. Treatment options are based on the closure of the fistula tract, however, the optimal surgical method should be decided individually for each patient.

Ethics

Informed Consent: Consent form was filled out by all participants.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: Aykut Başer, Zafer Aybek, Concept: Aykut Başer, Hülya Yılmaz Başer, Design: Aykut Başer, Hülya Yılmaz Başer, Data Collection or Processing: Ali Ersin Zümrütbaş, Cihan Toktaş, Analysis or Interpretation: Aykut Başer, Hülya Yılmaz Başer, Literature Search: Aykut Başer, Yusuf Özlülerden, Writing: Aykut Başer, Hülya Yılmaz Başer.

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