Intravesical contrast-enhanced ultrasound (CEUS) for the diagnosis of vesicouterine fistula (VUF)

A case report

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Abstract

Background: Vesicouterine fistula (VUF) is a very rare type of urogenital fistula, the incidence of which has increased in recent years due to increased cesarean section deliveries and other pelvic surgeries. The clinical diagnosis of VUF is typically challenging.

Case: A 31-year-old woman who presented with fever, increase urine frequency, and urinary incontinence at night, along with occasional vaginal discharge after cesarean section. The VUF was misdiagnosed on conventional ultrasound, but was successfully diagnosed by using intravesical contrast-enhanced ultrasound (CEUS) using SonoVue, which was confirmed by the subsequent cystoscopy.

Conclusion: Intravesical CEUS provides a new effective method for diagnosing VUF, which may become the first choice for diagnosing urogenital fistulas.

Abbreviations: CEUS = contrast-enhanced ultrasound, CT = computed tomography, MRI = magnetic resonance imaging, US = ultrasound, VUF = vesicouterine fistula, WBC = white blood cell.

Keywords: contrast-enhanced ultrasound (CEUS), urogenital fistula, vesicouterine fistula (VUF)

1. Introduction

Vesicouterine fistula (VUF) refers to an abnormal communication channel between the bladder and the uterus. It is a rare type of urogenital fistula, with an incidence of only 4% to 10% of all genitourinary fistulas.[1] In a majority of cases, VUF is caused by iatrogenic damage caused during pelvic surgery, such as cesarean section.[1] The incidence of VUF has increased in recent years owing to the rise in number of cesarean deliveries and other pelvic surgeries.[1]

The diagnosis of VUF is mainly based on the methylene blue staining test, cystoscopy, cystography, ultrasound (US), computed tomography (CT), and magnetic resonance imaging (MRI).[2] Treatment options include conservative treatment, laparoscopic repair or laparotomy depending on the location and size of fistula.[3] Therefore, accurate delineation of the fistula tract plays a vital role in the treatment of VUF.[2] We herein report a patient with VUF in whom the diagnosis was established by intravesical contrast-enhanced ultrasound (CEUS) using SonoVue. Our experience provides a new, simple, and effective method for the diagnosis of VUF.

2. Case

2.1. Ethical approval

The study was approved by the Institutional Review Board and Ethics Committee of The First Hospital of Jilin University. Written informed consent was obtained from the individual participant included in the study.

2.2. Case report

A 31-year-old woman presented with a history of fever since 16 days, which started from the third day post-cesarean section, and poor healing of surgical wound noticed since three days. She complained of increase urine frequency and urinary incontinence at night, along with occasional vaginal discharge. The peak body temperature was 39.5°C, which did not respond to treatment with anti-inflammatory drugs. The abdominal incision reopened without apparent cause three days prior to admission, and exuded purulent fluid. She had no pregnancy or abortion history, and no history of diabetes or pelvic inflammatory disease. Physical examination revealed a 10cm longitudinal surgical scar at the ventral midline, which was split into a rip of 2cm long and 6cm deep from the upper half of incision, along with purulent fluid exudation. Gynecological examination revealed that the vagina was smooth with soft and moist mucosa; the surface of cervix was rough; the uterus was slightly enlarged and exhibited poor mobility and tenderness; there was a moderate quantity of purulent uterine discharge. The fast C-reactive protein level was > 200.0mg/L. White blood cell (WBC) count at admission was 13.95×10⁹/L; lymphocyte percentage was 11.34% and neutrophil percentage was 83.64%.

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The conventional two-dimensional (2D) ultrasound (US) showed that the uterus was antepositioned (size: 58 × 40 mm) with an intrauterine anechoic area of about 11 × 4 mm. There was no definite discontinuity of the anterior uterine wall on 2D US. There was a small irregular anechoic dark space with poor acoustic signal in front of the uterus located between the right rear wall of bladder and right posterior abdominal wall. An anechoic dark area sized 30 × 15 mm was seen in the vagina, with poor acoustic signal. After real-time observation for 2 minutes, the intravaginal anechoic dark region increased gradually, with a maximum size of about 50 × 33 mm. A provisional diagnosis of intraperitoneal bladder fistula and VUF was considered.

After placement of an indwelling urinary catheter, intravesical contrast-enhanced ultrasound (CEUS) examination was performed. The contrast-agent SonoVue (Bracco, Italy) was diluted with 0.9% sodium chloride (1:100) and used for the first CEUS exam of bladder. The acoustic attenuation was obvious after injection of contrast agent; however, the deep organs were not clearly observed. Therefore, a repeat examination was performed after voiding of urinary bladder. SonoVue was diluted with 0.9% sodium chloride (1:400) for the second CEUS exam (Fig. 1), and 150 mL suspension was injected into the bladder through the urinary catheter. The bladder was filled with uniform contrast agent, and the deep organs were displayed very well. After observation for 3 minutes and 9 seconds, the contrast agent flowed out in front of the uterus; however, no contrast medium was discovered in the uterus and vagina (Fig. 1A). However, after the vagina was drained, there was contrast agent filling within uterine cavity at 3 minutes 28 seconds. The fistula was located between the anterior wall of uterus and the posterior wall of bladder (width: 4 mm; length: 5 mm). Subsequently, the contrast agent flowed into the vagina (Fig. 1B and C). The CEUS images confirmed the diagnoses of intraperitoneal bladder fistula and VUF. The methylene blue dye test showed that blue liquid flowed from the cervix into the vagina. Subsequent cystoscopy displayed a 2 mm split above the posterior wall of the bladder trigone surrounded by hyperemic and edematous tissues (Fig. 1D).

The patient was administered antibiotic therapy (intravenous drip 2.0 g ceftezole sodium and 0.5 g metronidazole, BID). With use of indwelling urinary catheter for four weeks, vaginal leakage of urine significantly subsided. After real-time observation for 2 minutes, the intravaginal anechoic dark region increased gradually, with a maximum size of about 50 × 33 mm. A provisional diagnosis of intraperitoneal bladder fistula and VUF was considered.

3. Discussion

VUF is a very rare urogenital fistula. The clinical manifestations of VUF depend upon the location and size of fistula. If the uterus fistula is located above the isthmus, patient may present with amenorrhoea, urinary incontinence, and menstrual hematuria. When the uterus fistula is below the isthmus, it is characterized by noncontinuous vaginal leakage of urine, which seriously impairs the quality of life. If the VUF is not diagnosed timely, patient may develop infection, which in severe cases can progress to bacteremia, shock, or even death. Methylene blue staining test, cystoscopy, cystography, conventional US, CT, and MRI are not dynamic modalities and cannot display the location, size, and detailed information of the fistula, which renders the diagnosis of covert or small and circuitous fistula difficult or may even cause misdiagnosis. In addition, cystography and CT involves x-ray
exposure besides exhibiting poor resolution of soft tissues; moreover, patients with iodine hypersensitivity are unable to tolerate cystography. \[2\] High cost of MRI is a major limitation to its wider use.\[3,4\]

CEUS, also referred to as acoustic contrast imaging, is a new technology that can significantly improve the resolution, sensitivity, and specificity of ultrasonic diagnosis through strengthening the scattering echo by use of a contrast agent.\[6\] The most commonly used contrast agent is SonoVue (Bracco, Italy); its key component is sulfur hexafluoride (SF\(_6\)) gas microvesicle, which is metabolized and excreted by lungs, is hypoallergenic, and does not damage the liver and kidneys. The clinical value of CEUS for diagnosis of liver diseases is well acknowledged.\[7\] Mao et al\[6\] obtained good results with injection of SonoVue in the biliary drainage tube to determine the patency of the bile duct. Chen et al\[6\] reported successful diagnosis of enterovesical fistula with injection of SonoVue into bladder through the urinary catheter, which clearly displayed the fistulous tract between the bladder and the rectum. Sparczech et al\[6\] reported their successful experience with use of SonoVue injection into a variety of surface catheters and drainage tubes for diagnosis of biliary fistula, enterovesical fistula, and archosyrinx, aided by dynamic observation of the flow and distribution of contrast agent. In this way, the location, size, shape, and course of the fistula could be directly visualized, especially for the complex fistula, with a diagnosis coincidence rate of up to 90%.\[7\] We presume that some of the misdiagnoses may be attributable to biliary fistula and intestinal fistulas, in which presence of intestinal gas is liable to affect ultrasonic imaging. However, in the case of VUF, the uterus is a substantive organ, and the bladder is a liquid containing organ; moreover, there are no intervening organs containing gas. In addition, there are no intestines in front of the bladder, as the bladder is adjacent to the abdominal wall. Therefore, ultrasound is undoubtedly the best way to observe the pathological changes in the bladder and uterus.

The VUF in the present case occurred secondary to cesarean section. The patient presented with postoperative fever, increased urinary frequency, nocturnal urinary incontinence, associated with abdominal infection and surgical wound infection. With use of contrast agent SonoVue, intravesical CEUS dynamically displayed the perfusion process of configurative complex fistulas of VUF and intraperitoneal bladder fistula, and accurately showed the location, size, and shape and the anatomical relationship of the fistula with the surrounding tissues. Our diagnosis was confirmed by cystoscopy. We used 2 different SonoVue concentrations (1:100 and 1:400 dilution with 0.9% sodium chloride) in this patient, and found that the ultrasonic imaging quality with 1:400 dilution was much better than that with 1:100 dilution; this can provide an empirical reference for intravesical CEUS. False negative results of intravesical CEUS were avoided by real-time observation of the perfusion process and vaginal drainage to reduce the intrauterine pressure. This provided a distinct advantage over use of cystography, CT, and MRI. With conservative treatment, the patient’s symptoms successfully resolved. Unfortunately, the patient compliance was poor, and we were unable to obtain further follow-up data.

In conclusion, as a rare urogenital fistula, imaging examination is crucial for the diagnosis of VUF. As compared with traditional cystography, CT, and MRI, intravesical CEUS allows for dynamic observation of the perfusion process of fistula, and offers the advantages of clear imaging, absence of radiation exposure, strong repeatability and cost-effectiveness. Intravesical CEUS provides a new, simple, effective, and safe method for the diagnosis of VUF, which may become the first choice for the diagnosis of VUF, even in cases of urogenital fistula.

**Author contributions**

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