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Case report

Trans-intrauterine contrast-enhanced ultrasound (CEUS) can be an effective approach for the diagnosis of vesicouterine fistula (VUF), especially for patients with fistulas flowing unidirectionally from the uterine cavity

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ABSTRACT

Background: Vesicouterine fistula (VUF) is a rare complication after cesarean section. It is challenging to diagnose VUF correctly.

Case presentation: A 34-year-old woman complained of recurrent hematuria and urinary tract infection for more than 4 years after cesarean delivery, mostly during menstruation, without vaginal leakage and with a normal menstrual cycle. Conventional transabdominal ultrasound showed no abnormal findings in bilateral kidneys and ureters, bladder and uterus, and transvaginal ultrasound showed that the scar at the lower part of the anterior uterine wall was closely adhered to the posterior wall of the bladder. Considering that the patient had hematuria but no vaginal leakage, we assumed that the fistula was flowing unidirectionally from the uterine cavity to the bladder cavity. Therefore, we chose to inject SonoVue (ultrasound contrast agent) into the uterine cavity rather than into the bladder. After intrauterine injection of SonoVue, the ultrasound contrast agent was seen flowing from the uterine cavity into the bladder cavity through the fistula, showing a hyperechoic fistula between the posterior wall of the bladder and the uterine wall, confirming the diagnosis of VUF. The accuracy of this diagnosis was then further confirmed by both MRI and cystoscopy.

Conclusions: Trans-intrauterine CEUS provides a new effective imaging method for the diagnosis of VUF, especially for patients with fistulas that flow unidirectionally from the uterine cavity.

1. Introduction

Vesicouterine fistula (VUF) is a very rare complication occurring after caesarean section, accounting for about 1–4% of all urogenital fistulas [1]. The main symptom is early postoperative urinary incontinence [2], and patients may develop periodic hematuria, amenorrhea or urinary tract infection months or years after surgery. Atypical clinical symptoms of VUFs often delay early and correct

Abbreviations: CEUS, contrast-enhanced ultrasound; VUF, vesicouterine fistula; MRI, magnetic resonance imaging.

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diagnosis and treatment [3]. Therefore, making a timely and accurate diagnosis is essential to solve a series of symptoms caused by VUF. Our report documents a case of VUF with recurrent hematuria and urinary tract infections and a normal menstrual cycle, which was successfully diagnosed using innovative *trans*-intrauterine CEUS.

2. Case report

A 34-year-old woman was admitted to hospital 4 years ago for emergency cesarean section due to premature rupture of membranes and two weeks of umbilical cord around neck at 36 weeks and 6 days of pregnancy. She presented with repeated hematuria and urinary tract infection 2 years after surgery, without vaginal leakage and amenorrhea, and had a normal menstrual cycle. Routine urine tests showed repeated increases in white blood cell count and red blood cell count. Conventional transabdominal ultrasound showed no abnormal findings in bilateral kidneys and ureters, bladder and uterus. Cystoscopy performed in another hospital one year ago showed a small bulging mass on the posterior wall of the bladder, with no obvious fistulae found. Pathological examination showed that this bulging mass was cystitis glandularis with lymphatic follicular formation.

After anti-inflammatory and hemostatic treatment, the above symptoms are still repeated. In order to seek further treatment, the patient came to our tertiary hospital for examination. Conventional transabdominal ultrasound showed no abnormal findings of both kidneys and ureters, but this time a hyperechoic mass was observed in the bladder cavity, which could move with position change (Fig. 1). Transvaginal ultrasound showed a scar at the lower part of the anterior uterine wall was closely adhered to the posterior wall of the bladder, and no sign of fluid accumulation in the uterine cavity and vagina. We asked her to have another ultrasound examination in a week. One week later, the patient states that the hematuria has disappeared. Conventional transabdominal ultrasound did not find the hyperechoic mass in the bladder cavity. A double-lumen contrast catheter was placed vaginally into the uterine cavity for *trans*-intrauterine CEUS examination. The ultrasound contrast agent SonoVue (Bracco, Italy) was diluted with 0.9% sodium chloride (1:20). On the first attempt to injection, the contrast agent was reflux to the vagina because the balloon was too small. Then enlarged the balloon for a second try, it was real-time displayed that intrauterine SonoVue was sprayed into the bladder cavity, clearly showing the hyperechoic fistula tract delineated by SonoVue between the lower anterior uterine wall and the posterior bladder wall (Fig. 2, Supplementary Video), so VUF was confirmed for diagnosis. Subsequent magnetic resonance imaging (MRI) showed a fissure in the posterior wall of the bladder communicating with the uterine cavity (Fig. 3). Under cystoscopy, a fistula of about 1.0cm*1.0cm in size was observed in the posterior wall of the bladder, and no stones or diverticulum were found. The patient underwent transabdominal fistula repair in the department of urology, and no recurrence of hematuria was found at follow-up.

3. Consent for publication

Written informed consent was obtained from the individual for the publication of any potentially identifiable images or data included in this article.

4. Discussion

VUF is a rare type of urogenital tract fistulas, which are often caused by iatrogenic surgery, with cesarean section accounting for the

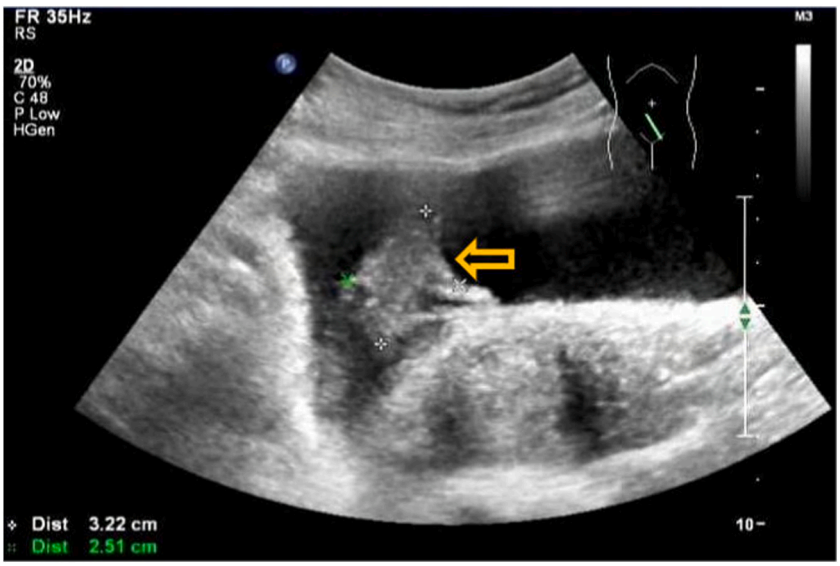

Fig. 1. Conventional transabdominal ultrasound. A hyperechoic blood clot in the bladder (arrow).

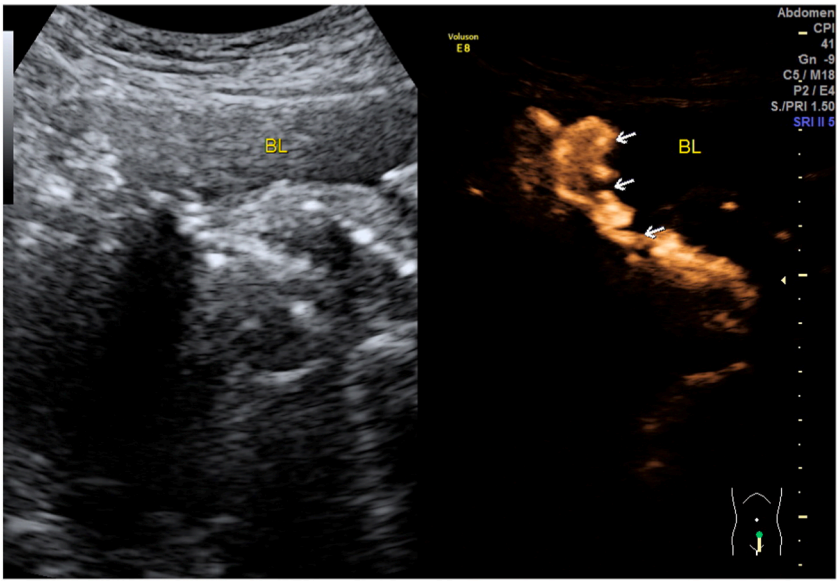

Fig. 2. *Trans*-intrauterine contrast-enhanced ultrasound (CEUS). SonoVue in the uterine cavity was sprayed into the bladder cavity, clearly showing the hyperechoic fistula tract delineated by SonoVue between the lower anterior uterine wall and the posterior bladder wall (arrow). A video of this process can be viewed in Supplementary Video. BL, bladder.

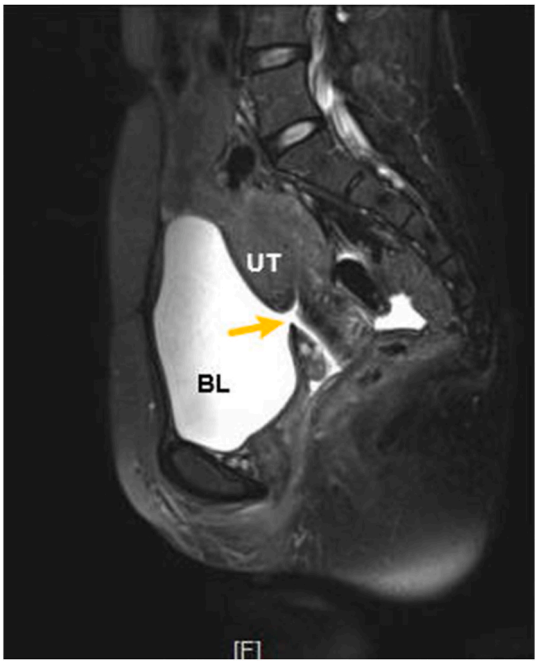

Fig. 3. MRI. A fissure in the posterior wall of the bladder and the uterine cavity (arrow).BL, bladder. UT, uterus.

highest proportion [4]. Clinical manifestations of VUF are diversified according to the location and size of the fistula. Jozwik et al. proposed to divide the clinical presentation of VUF into three types based on the routes of menstrual flow: type I—with amenorrhea and periodic hematuria, and urine can be completely controlled (without vaginal urine leakage), i.e. Youssef's syndrome; type II —urine in the bladder and menstrual blood in the uterus flowing in both directions through the fistula, with vaginal leakage and periodic hematuria; type III —with normal menstruation with or without vaginal urine leakage [5]. Type I accounts for the highest percentage and Type III is the least common. The patient we report belongs to type III, which presents with normal menstruation but not vaginal urine leakage.

Urogenital fistulas require a high level of suspicion by the clinician and the selection of the right tests for early diagnosis and

treatment. The main methods used to diagnose VUF are ultrasound (US), methylene blue staining, cystoscopy, cystography, computed tomography (CT), and MRI. However, using these regularly used techniques to appropriately diagnose UVF still presents considerable difficulties. Contrast-enhanced ultrasound (CEUS) enhances the acoustic impedance difference from the surrounding tissue through the scattering echo effect of the ultrasound contrast agent, resulting in high-resolution contrast-enhanced ultrasound images. Currently, the commonly used contrast agent is Sonovue (Bracco, Italy), its main component is sulfur hexafluoride (SF_6) gas microbubble, which is exhaled from the lungs, hypoallergenic and does not damage the liver or kidneys. Sun et al. reported a successful diagnosis of VUF by intravesical injection of SonoVue through a catheter into the bladder [6]. In the present case, VUF occurred after cesarean section. Because the fistula in the bladder was too small, cystoscopy performed at another hospital a year ago showed no fistula but only a small bulge at this site. Since the patient never experienced vaginal leakage, we hypothesized that the fistula might have a one-way valve function that allowed only menstrual blood to enter the bladder and not urine to enter the uterine cavity, which may be related to the one-way valve generated by the uterine isthmus sphincter [7]. In addition, a hyperechoic blood clot image was seen in the bladder cavity at the time of this patient's visit, suggesting that this may be the source of the blood in the urine. Considering these two points, we believe that a positive result cannot be obtained by intravesical injection of SonoVue into the bladder as in sun et al., so we innovatively proposed a contrast agent injection route for such patients by intrauterine injection instead of intravesical injection. With this new method, we can see the whole process of SonoVue entering the bladder from the uterus through the fistula in real-time and make a correct diagnosis. The accuracy of this method was supported by MRI and cystoscopy findings in our case, as well as subsequent treatment follow-up.

In conclusion, compared with previously reported methods, *trans*-intrauterine CEUS can dynamically visualize the unidirectional fistula in patients with type III VUF in real-time and has the advantages of no radiation, reproducible, and inexpensive. *Trans*-intrauterine CEUS provides a new simple, effective, and safe imaging method for the diagnosis of VUF, which can be used in the future for the diagnosis and differential diagnosis of patients with VUF-related symptoms.

Author contribution statement

All authors listed have significantly contributed to the investigation, development and writing of this article.

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Data availability statement

Data included in article/supp. material/referenced in article.

Declaration of competing interest

The authors declare no conflict of interest.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.heliyon.2023.e13268.

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