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Vesicouterine fistula presenting with cyclical haematuria mimicking bladder endometriosis: A case report

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ABSTRACT

Vesicouterine fistula (VUF) is an abnormal communication between the bladder and uterus, occurring 1–4% of all urogenital fistulas. Diagnosis is still a challenge because symptoms may appear late or fistula may be missed even after repeated examination. A 37-year old woman who has two children born through caesarean section complained of the absence of menstruation for the past three years. At the same time point, she experienced cyclic haematuria and amenorrhea. The diagnosis was made through ultrasonography, cystoscopy and hysteroscopy. She was then managed with laparoscopic bladder fistula repair continued with total laparoscopic hysterectomy. The VUF can present as an undesirable consequence of caesarean section.

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1. Introduction

VUF refers to a pathological communication between the bladder and uterus or cervix. This is a rare case and is estimated to account for l–4% cases of all urogenital fistulas [4]. It is usually a consequence of iatrogenic injury instigated through pelvic surgery. The lower segment caesarean section has been acknowledged as the most common cause of VUF [6]. Thus, the amount of cases has risen in the past few years, owing to the growing number of caesarean section [11]. The impact of genitourinary tract fistula on the family can be devastating since the patients may encounter social consequences and psychosocial distress that may become the sources of contention in marriage [5,15]. Therefore, an accurate and early diagnosis, as well as proper management are of paramount importance [9]. The diagnosis of VUF remains challenging due to the delayed onset of symptoms [7]. Likewise, a subtle fistula may be missed during the examination [19]. We herein report a case of VUF presented to our outpatient clinic with a chief complain of secondary amenorrhea, with a review and update of the literature.

2. Case illustration

A 37-year-old Javanese-Indonesian female patient with a history of two previous caesarean sections was referred by a urologist to our outpatient clinic with bladder endometriosis. Her chief complaint was the absence of menstruation for the last three years. Intriguingly, she was also experiencing cyclical haematuria, without cyclical vaginal bleeding, accompanied with typical premenstrual symptoms (such as breast tenderness), for the same time duration. There is no history of watery vaginal discharge or vaginal bleeding during menstruation. She had a history of bladder injury during the second cesarean section three years before admission, and a bladder restoration was performed concurrently. No specific drug history or family history of other diseases. The physical examination at presentation was unremarkable.

Gynaecologic ultrasound evaluation of the cervix and uterus displayed homogeneous myometrium with an endometrial thickness of 11 mm (Fig. 1A–E). There was no fluid appeared in the uterine cavity. Both ovaries were normal. An interconnection between the posterior bladder wall and the lower uterine segment was evident, measuring 8 mm in diameter, was related to vesicocervical fistula.

Hysterosalpingogram (HSG) revealed uniform contrast material filling the bladder, whereas no contrast media was discovered in the uterus or fallopian tubes. This result might designate the presence of vesicouterine fistula (Fig. 1F–I). She was then evaluated by cystoscopy (Fig. 1J) and hysteroscopy (Fig. 1K). Cystoscopy found a small opening in the posterior bladder wall, sized approximately 5 mm in diameter, located 1,5 cm from the bladder neck. Hysteroscopy examination showed a defect in endocervix at the 12-o'clock position measuring 10×5 mm^2, connecting the endocervix and the posterior bladder wall. There was no access to the uterine cavity as the cervical orifice was merged with the posterior bladder wall, signifying a vesicocervical fistula. Since all findings implied a vesicocervical fistula, the patient then underwent a laparoscopic vesicocervical fistula repair.

Cystoscopy was performed prior to the surgery, which aimed to insert a bilateral ureter catheter and a double J-stent (DJ stent). The fistula was identified at the 12-o'clock direction of the vesical trigone, measuring 5×5 mm. It was found that most of the cervix part fused with the bladder. The bladder was dissected from

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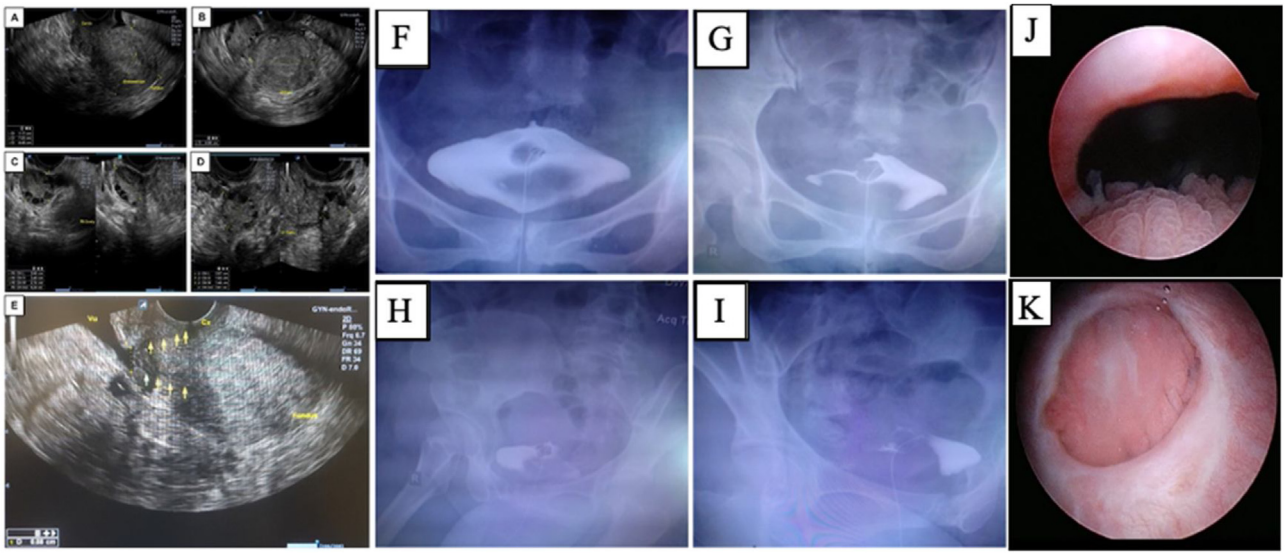

Fig. 1. A-E (**Ultrasonography examination of the uterus and vesicouterine fistula).** Transvaginal ultrasonography demonstrates (A) normal-sized retroflexed uterus with homogenous myometrium. (B) The endometrium thickness is 11 mm. There is no sign of fluid in the uterine cavity. Both right (C) and left (D) ovaries are normal, sized 26 × 24 × 27 mm and 20 × 16 × 14 mm respectively. (E) The appearance of an interconnecting tract between the posterior bladder wall and the lower uterine segment measuring 8 mm in diameter indicating vesicocervical fistula. Fig. 1F-I (HSG result displays a vesicouterine fistula). (F) An anteroposterior view of the pelvis depicting contrast media in the bladder. (G-I) The figures showing the contrast is slowly diminished in the bladder when the balloon insufflation was off and is no longer observed in the bladder after the patient urinated. Fig. 1J–K (Hysteroscopy and cystoscopy results). (J) The hysteroscopic view reveals that there is no access to the uterine cavity from the cervical canal as the orifice merged with the bladder, indicating vesicocervical fistula (K). Cystoscopy examination shows a fistula with endocervical tissue on the posterior part of the bladder measuring.

the uterus and we identify the fistulous tract. Initially, the plan was conserving the uterus by undertaking the bladder and uterine reconstructive surgery which performed by female pelvic and reconstructive gynecologist. Nonetheless, the presence of substantial loss of the isthmus and cervical tissue during the dissection caused the least probability that the uterus could be reconstructed. The procedure is then followed by a supracervical laparoscopic hysterectomy. The bladder was then sutured two layers interruptedly.

Postoperative evaluation was uneventful. The patient was discharged after five days of hospitalisation. Cystostomy was maintained for two weeks. While the Foley catheter was kept for one month and the DJ stent was successfully removed after two months. Eventually, three months following the surgery, the patient was subjected to CT-urography. It was observed that the contrast media filled the bladder and there was no visualisation of an abnormal connection between the cervix and the posterior bladder wall. The patient is currently in good condition without any complaints of urinary leakage or incontinence, and felt relieved with no further complains since being treated.

3. Discussion

Diagnosis of VUF is generally based on accompanying patient symptoms and physical examination [16]. The patient came with primary complaints of secondary amenorrhea and infertility for three years. Cyclic haematuria was the accompanying symptoms that occurred in parallel to amenorrhea. The onset of these symptoms was at the same time frame as her previous caesarean section with complication which is history of bladder injury. This accumulation of symptoms, consisting of amenorrhea, cyclic haematuria and a history of previous caesarean section, has been recognised as Youssef's syndrome. The symptom of VUF itself tends to be varied. If the presence of VUF is above the internal uterine ostium, it may be linked to amenorrhea, decreased urinary leakage, and cyclic haematuria. In contrast, VUF encompassing the cervix is generally depicted by urinary leakage and uninterrupted menstrual cycle [6,17]. The absence of urinary incontinence might be due to the functional valve made by the isthmic sphincter that also causes the menstrual blood flows into the bladder. Likewise, the pressure created by the sphincter is sufficient to restrain the leakage of urine into the vagina [13]. In some instances, the internal uterine orifice can be sealed by granulations situated on the posterior uterine wall, and the fibrotic tissue surrounded the cervix, which may also prevent the reverse flow of urine into the vagina. However, patients with VUF could also experience urinary incontinence that mainly occurs in cervix involvement or when the fistula located below the isthmus [12]. This is not the case in our patient highlighted that the fistula might be above or at the level of isthmus. This condition also one of the reason for the patient to delay seeking medical assistance due to no urinary leakage from vagina.

Endometriosis was also suspected initially due to the presence of cyclic haematuria. The absence of normal vaginal bleeding and the presence of typical Youssef's triad might exclude the probability of endometriosis; hence, the bladder wall biopsy to confirm the diagnosis is not compulsory. HSG and cystoscopy remain the "gold standard" evaluation to diagnose VUF. Despite its inherent difficulty in distinguishing the VUF tract from diverse patterns of noncomplicated caesarean scars, ultrasound is emerging as a feasible alternative imaging modality to diagnose VUF. Ultrasound evaluation for detecting VUF is intricate yet challenging. Fistulous tract in ultrasound examination visualised as double echogenic lines between the anterior wall of the uterus and posterior wall of the bladder. The presence of fistula in our patient was first suspected through transvaginal sonography that was then discerned by HSG and cystoscopy [8]. However, in some instances, when 2D ultrasonography and cystoscopy are not able to differentiate the presence of fistulous tract, intravesical contrast-enhanced ultrasound (CEUS) can be an alternative option [18]. The best imaging that can be used to evaluate VUF are computed tomography (CT) or pelvic MRI, but due to budgeting and resources limitation the examination was not performed to this patient [19].

A surgical approach is contemplated the mainstay and definitive management of the VUF following caesarean section [17]. Notably, a subset of women was able to resume a normal period after fistula

repair [14], though the data on how frequently this occurs is very limited [2,10,14]. Due to the low prevalence of VUF, although hysterectomy is not obligatory, it is not easy to find the most assuring approach to manage VUF. Surgeon's experience, patient's preferences, the type and location of fistula are of paramount importance in determining the choice of surgical repair [14]. Vaginal stenosis has been recognised as a common sequela of VUF that is least likely to recuperate and possibly worsen, impacting sexual function and the ability to conceive [1,14]. The patient was concerned to preserve the uterus owing to secondary infertility; thus, fistula repair was the initial plan proposed in this patient. However, this procedure could not be carried out due to the considerable loss of isthmus and cervical tissue during the dissection. Supracervical hysterectomy was done due to severe adhesion of fibrotic tissue between bladder and cervix.

4. Conclusion

The VUF can present as an undesirable consequence of caesarean section. The prevalence is considered low but tends to increase due to the growing number of caesarean sections. The diagnosis can be made by ultrasonography, cystoscopy and HSG. Although patients with cyclic haematuria and secondary infertility can be highly suspected as bladder endometriosis, we should be more cautious of the possibility of having VUF in constructing a differential diagnosis and choosing the diagnostic tests. Patients who are still concerned about fertility should be well informed that the probability of preserving the uterus can be high, but it depends on the nature of the case including the extent of tissue involved and the type of the fistula. In this context, hysterectomy may be unavoidable.

Declaration of Competing Interest

The authors declared have no conflicts of interest to disclose, and this study had been reported in line with SCARE 2020 criteria [3].

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Ethical approval

This study is exempt from ethnical approval.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Achmad Kemal Harzif: study concept or design.
Mila Maidarti: data analysis of interpretation.
Ivan Ginanjar: data collection.
Amalia Shadrina: writing paper.
Alfa Putri Meutia: study concept or design.

Registration of research studies

Not applicable.

Guarantor

The Guarantor was also the corresponding author.

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