

Bioscientia Medicina: Journal of Biomedicine & Translational Research

Journal Homepage: www.bioscmed.com

Diagnosis and Management of Vesicouterine Fistula (Youssef's Syndrome): A Case Report

Amir Fauzi^{1*}, Ratih Krisna¹, Hadrians Kesuma Putra¹, Sonia Prima Arisa Putri², Febi Stevi Aryani², Hana Andrina²

¹ Division of Urogynecology and Reconstructive Surgery, Department of Obstetrics and Gynecology, Faculty of Medicine, Universitas Sriwijaya/Dr. Mohammad Hoesin General Hospital, Palembang, Indonesia

² Department of Obstetrics and Gynecology, Faculty of Medicine, Universitas Sriwijaya/Dr. Mohammad Hoesin General Hospital, Palembang, Indonesia

ARTICLE INFO

Keywords:

Cystoscopy
Fistula repair
Hysteroscopy
Vesicouterine fistula
Youssef's syndrome

*Corresponding author:

Amir Fauzi

E-mail address:

amirfauzi61@yahoo.co.id

All authors have reviewed and approved the final version of the manuscript.

<https://doi.org/10.37275/bsm.v6i5.501>

ABSTRACT

Background. Vesicouterine fistula (VUF) is a rare pathological communication between the uterus or cervix and the bladder. Youssef's syndrome is an atypical presentation of a patient with a vesicouterine fistula, characterized by a triad of cyclic hematuria, amenorrhea, and absence of urinary incontinence. Because of this atypical presentation, the patient may go undiagnosed and patient management will be delayed. **Case presentation.** A 39-year-old woman complained of hematuria. The patient underwent a second caesarean section in 2008 and a few weeks after the procedure the patient complained of leakage of urine from her vagina. Six months later the patient complained of hematuria on her menstrual cycle and amenorrhea, with an absence of urinary incontinence. Ultrasound examination found adhesions between the uterus and vagina, and irregular bladder mucosa. The patient underwent cystoscopy with endometriosis and was suspected of vesicovaginal repair scars and a bladder biopsy was also performed with the results of no endometrial stroma and glands then followed by hysteroscopy. The Hysteroscopy result showed a 20 mm diameter hole with irregular margins. Subsequently, the patient was treated with hysterorrhaphy laparotomy and fistula repair. **Conclusion.** One type of vesicouterine fistula is Youssef's syndrome, which is a complication following a lower segment caesarean section with undiagnosed bladder injury. The diagnosis can be confirmed by cystoscopy, intravenous pyelography, hysterosalpingogram, sonography, and other types of imaging tests. Management of patients with VUF can be conservative, medical, or surgical.

1. Introduction

Vesicouterine fistula (VUF) is an uncommon pathological communication developing between the uterus or cervix and the urinary bladder most commonly after an iatrogenic injury during Caesarean section with a prevalence of approximately 1-4% of all urogenital fistulas.¹ The prevalence is increasing due to the increased rate of caesarean delivery. Youssef's syndrome is an atypical presentation of a patient with VUF, characterized by a triad of cyclical hematuria,

amenorrhea, and absence of urinary incontinence. Due to these atypical presentations, the patient might be undiagnosed and the management of the patient will be delayed. Management of the patient with VUF could be conservative, medical, or surgical. The usual surgical management is resection of the bladder part of the fistula with or without hysterectomy. Manifestation of VUF could affect patients' quality of life. We describe the manifestation of VUF, diagnostic approach, and

management of VUF patients in our facility.

2. Case Presentation

A 39-year-old woman came to the urogynaecology outpatient clinic with a complaint of haematuria. The patient underwent her second caesarean section in 2008 and weeks after the procedure patient complained of urine leakage from her vagina. The patient was referred to Cipto Mangunkusumo Hospital and

managed with silicon catheter insertion. After 1 month, the catheter was removed and spontaneous micturition resumed without further leakage of urine. Six months later the patient complained of cyclical hematuria on her menstrual dating and amenorrhea, with complete absence of urinary incontinence. On ultrasound examination uterus is within normal shape and size, with a finding of adhesion between uterus and vagina and irregular bladder mucosa.

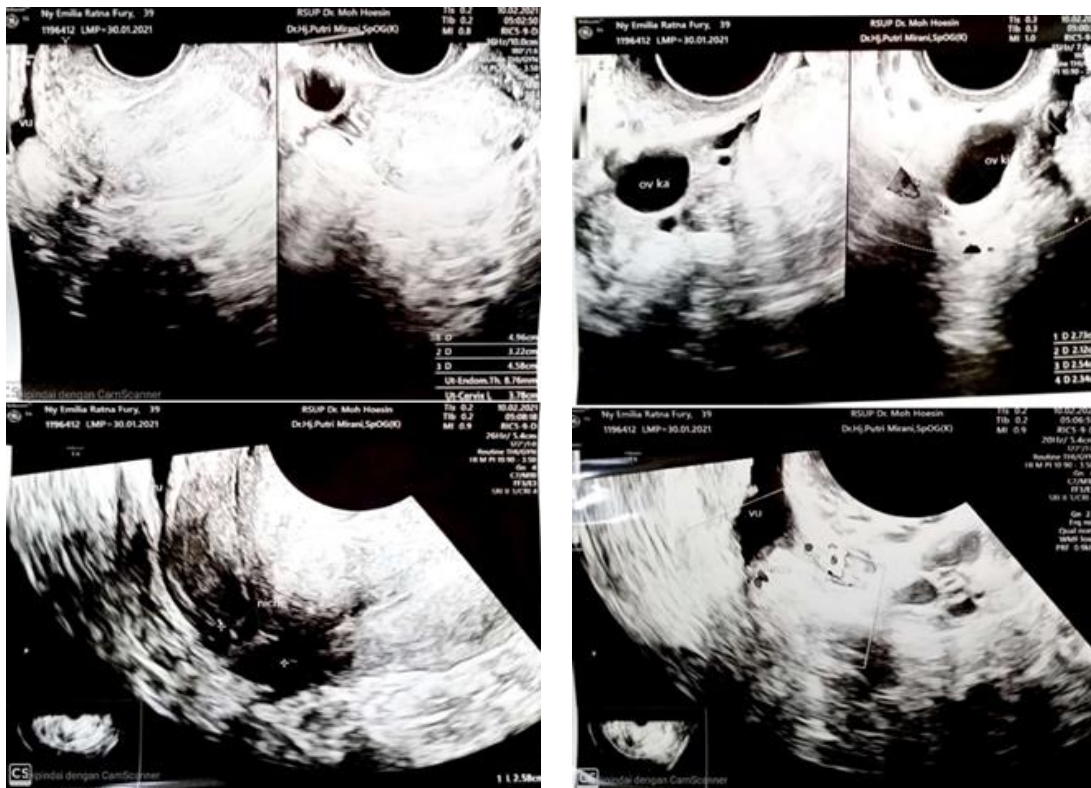


Figure 1. Ultrasound finding of irregular bladder wall susceptibility of bladder mass and adhesion between uterus and vagina

The patient underwent cystoscopy with suspected of endometriosis at vesicovaginal repair scar and bladder biopsy was performed with the result no stromal and endometrial gland from bladder biopsy then continued with hysteroscopy. Before the hysteroscopy procedure, a urinary catheter was inserted with the finding of

blood in the urine bag, and on speculum examination cervix was pushed to the anterior. The hysteroscopy revealed an opening approximately 20 mm in diameter with an irregular margin and a urinary catheter was found inside the uterine cavity.

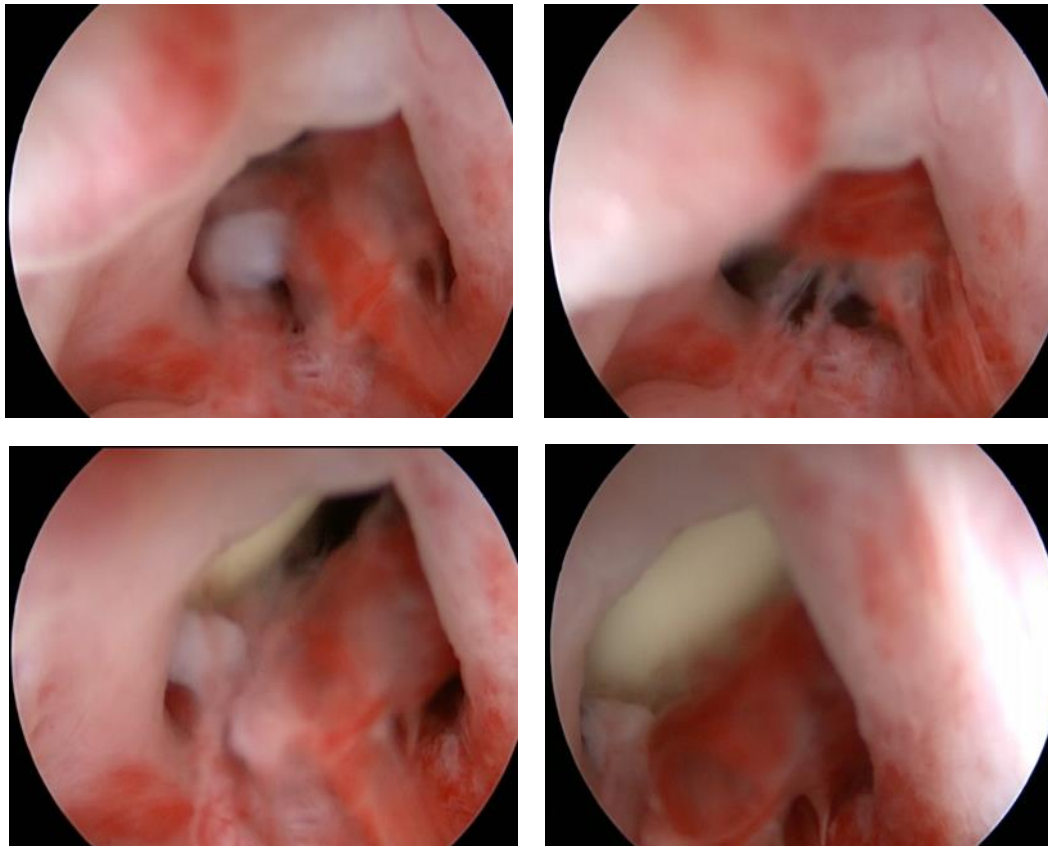


Figure 2. Hysteroscopy finding with defect size approximately 20 mm and urinary catheter was found inside the uterine cavity

The patient was managed with laparotomy hysterorrhaphy and fistula repair in join with the urology surgery department without hysterectomy. There was a defect of the bladder with size 10 cm after adhesion between uterus and vagina was released. The defect was sutured two layers with interrupted suture. The patient was discharged after 4 days of hospitalization and a urinary catheter was retained for 2 weeks. Postoperatively the patient had no complications and resumed her menstrual cycle 1 month after surgery.

3. Discussion

Vesicouterine fistula (VUF) is an uncommon pathological communication developing between the uterus or cervix and the urinary bladder.¹ Cesarean section is the single most common cause of VUF, approximately 83-93% of all cases. Although vesicouterine fistula represents only 1-4% of all urogenital fistulas its prevalence is increasing all over

the world due to the increased rate of caesarean section.^{2,3} The diagnosis of VUF is still challenging because of the atypical presentation of the patient. Youssef's syndrome is a rare presentation of a vesicouterine fistula. It was first described by Youssef in 1957. The fistula classically presents with a triad of cyclic hematuria, amenorrhea, and the absence of urinary incontinence.⁴

Youssef's syndrome is an iatrogenic complication following lower segment Cesarean section with an undiagnosed bladder injury.³ several mechanisms might be proposed incidence of VUF after caesarean section including the presence of a non-detected bladder rupture during emergency Cesarean section, inadvertent application of a suture in the base of the bladder while suturing the uterus, and abnormal blood supply to the base of the bladder, secondary to an abnormal vascular bed due to multiple dissections, usually after repeating Cesarean sections.⁵ Diagnosis of endometriosis on repair scar was suspected initially

due to the presence of cyclic haematuria but can be excluded because of the presence of amenorrhoea and absence of urinary incontinence, supported with bladder wall biopsy result. Once VUF is suspected, the diagnosis can be confirmed by cystoscopy, intravenous pyelography, hysterosalpingogram, sonography, and other types of imaging exams. Cystoscopy was the mainstay of diagnosis of VUF in the current study. In this case diagnosis of VUF was established with hysteroscopy. Imaging modalities sometimes were not able to show very small fistulae.

Management of the patient with VUF could be conservative, medical, or surgical.⁶ When conservative management fails, surgical treatment has been the mainstay of therapy.⁷ The usual surgical management is resection of the bladder part of the fistula with or without hysterectomy. Unless otherwise indicated, there is no need to remove the uterus even if the fistula is large.^{3,6} Although the repair is challenging, it was successful in all cases and pregnancy is possible after repair.^{8,9} 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.¹⁰

4. Conclusion

The diagnosis of Youssef syndrome can be confirmed by cystoscopy, intravenous pyelography, hysterosalpingogram, sonography, and other types of imaging tests.

5. References

1. Daniyan B, Sunday-Adeoye I, Ekwedigwe K, Dantani D, Eliboh M. Review of vesicouterine fistula at the National Obstetric Fistula Centre, Abakaliki, Nigeria. *Gynecol Obstet (Sunnyvale)*. 2016; 6: 380.
2. Bhattacharjee S, Kohli UA, Sood A, Tripathy S, Gupta M. Vesicouterine fistula: Youssef's syndrome. *Med J Armed Forces India*. 2015; 71(Suppl 1): S175-S177
3. Kettache R, Merad A, Buchholz NNP. Surgical

Management of Youssef's Syndrome: A Case Report. *Urologia Internationalis*. 2020;104(9-10): 833-6.

4. Youssef AF. Menouria following lower segment cesarian section: A syndrome. *Am J Obstet Gynecol*. 1957; 73: 759-64.
5. Machado RA Jr, Machado LC Jr, Lourenço LL. Vesicouterine fistula (Youssef syndrome): Case report and literature review. *Rev Bras Ginecol Obstet*. 2018; 40(9): 563-9.
6. Ali-El-Dein B, El-Tabey N, El-Hefnawy A, Osman Y, Soliman S, Shaaban AA. Diagnosis, treatment and need for hysterectomy in management of post caesarean section vesicouterine fistula. *Scand J Urol*. 2014; 48(5): 460-5.
7. Alamoudi OJ, AlTheneyan MA, Aldhaam N, Moazin M. Early robotic repair of vesicouterine fistula: a case report and literature review. *Urol Case Rep*. 2017; 11: 76-78.
8. Symeonidis EN, Sdralis E, Symeonidis A, et al. Vesicouterine fistula (VUF) as a rare urogenital complication managed with delayed surgical repair: A case report and review of the literature. *Case Rep Obstet Gynecol*. 2018; 2018: 2394896.
9. Kopp DM, Wilkinson J, Bengtson A, Chipungu E, Pope RJ, et al. Fertility outcomes following obstetric fistula repair: a prospective cohort study. *Reprod Health*. 2017; 14(1): 159.
10. Harzif AK, Maidarti M, Ginanjar I, Shadrina A, Meutia AP. Vesicouterine fistula presenting with cyclical haematuria mimicking bladder endometriosis: A case report. *Int J Surg Case Rep*. 2021; 80: 105709.