

## Youssef's syndrome - a need to review our ways

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### Abstract:

Vesicouterine fistulas are rare gynaecological fistulas. Multiple causative factors are known. A trending cause now a days is the increase in rate of lower segment caesarean section. We present a case of 25 year old primipara who presented with cyclical hematuria, amenorrhea and urinary continence, 7 months after her cesarean section. She was diagnosed with vesicouterine fistula by hysterosalpingography. After cystoscopic confirmation of size and relation to the trigone of bladder, the fistula was excised by transvesical approach via laparotomy.

**Keywords:** Cesarean section, fistula, hematuria, hysterosalpingography, cystoscopy.

Vesicouterine fistulae (VUF) are one of the rare gynaecological fistulae. Multiple factors are known to play a causative role in its formation. One of the less common but, a trending cause now a days is the increase in rate of lower segment caesarean section (LSCS) and repeat caesarean sections<sup>1</sup>. A VUF formed post LSCS was first explained by Youssef in this topic's maiden article "Menouria' following lower segment caesarean section: A syndrome"<sup>2</sup>. As this is mainly an iatrogenically formed VUF, it is of utmost importance for the gynaecologists to be aware of such an occurrence and hence to take necessary precautions while handling a case of prolonged or obstructed labour or a repeat LSCS. Surgery is the mainstay of treatment<sup>3</sup>, although non-surgical approach has been successful in a small number of early detected cases<sup>4</sup>.

### Case

A 25 years old lady, primipara, presented to our out-patient department (OPD), with a history of passing blood with urine every month for 4-5 days, during the time of expected menses from last 1 year and 3 months. This started 7 months after an emergency LSCS with a concurrent postpartum intra uterine contraceptive device (PPIUCD) insertion done in view of prolonged labor post induction. Patient doesn't give history of haematuria prior to LSCS. No history of urinary incontinence or any lower abdominal discomfort.

On examination, abdomen showed a healed pfannensteil incision, with no palpable mass or other scars. On per speculum and per vaginal examinations, no abnormality was detected. On suspicion of a VUF, she was advised transvaginal sonography (TVS), which showed a VUF from the LSCS scar with IUCD in-situ. A hysterosalpingography (HSG) was performed after removal of IUCD to confirm the location of the tract, which showed a VUF with right cornual block and left fallopian tube block (figure 1). Magnetic resonance imaging (MRI) pelvis showed a linear T2 hyperintense tract connecting the endometrial cavity through lower segment of anterior uterine wall into the posterior wall of urinary bladder suggestive of VUF (figure 2). A cystoscopy was done to locate the fistula opening with respect to the trigone and ureteric openings, which showed a 1.5 cm fistula on the posterior

wall of the bladder along the midline just above the trigone of bladder. On diagnosis of Youssef syndrome, patient and her partner were explained about the condition and the treatment options available. After obtaining consent, she was taken up for laparotomy.

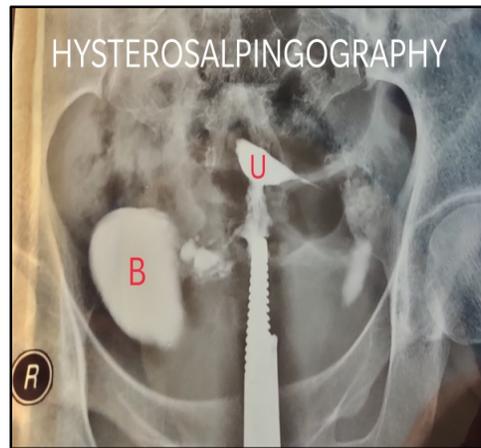


Figure 1: HSG showing filling of bladder with radiopaque dye along with a communicating tract.

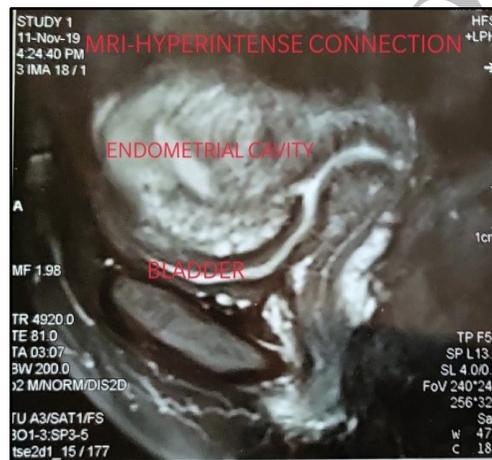


Figure 2: MRI showing the hyperintense communication tract between endometrial cavity and bladder cavity.

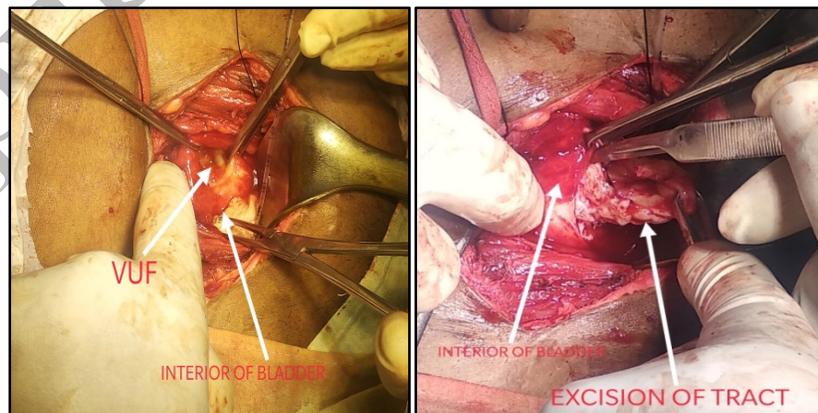


Figure 3, 4: Intraoperative visualization and excision of fistulous tract.

Upon opening of the abdomen, the bladder dome was identified and a transvesical approach was adopted to identify the fistulous tract, and the ureteric openings were identified and catheterized with infant feeding tubes. Fistulous tract was then excised along the margins (figure 3 & 4); the opening in the uterus was closed with polyglactin 1-0 continuous sutures and bladder was then repaired in two layers with 2-0 polyglactin sutures so as to avoid exposure of the inner layer of sutures. The fistulous tract was sent for histopathological examination, which showed fibrosed tissue. Postoperatively, the patient had bilateral ureteric catheters in-situ for 7 days, suprapubic catheter in-situ for 14 days and urethral catheter in-situ for 21 days. She was kept in hospital for 21 days with antibiotic coverage for the first 7 days. Recovery was uneventful and she was discharged on day 21 after removal of urethral catheter on being able to pass urine comfortably. She was advised regular check-ups and kept amenorrhic for 3 months with depot medroxy progesterone acetate (DMPA) injections monthly. Following which, the patient presented to the OPD with regular menses on follow-up.

## Discussion

Vesicouterine fistula is an abnormal connection between the bladder and the uterus, which may or may not be associated with menouria and/or urinary incontinence. It has been classified into 3 types; type 1 - menouria, amenorrhoea & urinary continence, type 2 - hematuria with urinary incontinence & dual flow of blood, type 3 - urinary incontinence and vaginal bleeding<sup>3</sup>. Various causes have been listed for the formation of VUF like high vaginal forceps-aided delivery, external cephalic version, curettage or manual removal of the placenta, placenta percreta, myomectomy, uterine rupture due to obstructed labor, uterine artery embolization, perforation of an intrauterine device, and brachytherapy for carcinoma of cervix. LSCS is the single most common cause of VUF due to a rising trend in repeat LSCS<sup>1, 5</sup>. While HSG remains the gold standard for diagnosis of VUF, cystoscopy and cystography have also been useful in identification, delineation of the tract and to check the involvement of ureters<sup>1, 6</sup>. MRI being a non-invasive technique is safer for diagnosis and better delineation of the tract. Both, conservative and surgical methods have been employed for the treatment of VUF, with surgical methods providing a definitive treatment. Non-surgical methods like prolonged catheterization and progesterones to keep the patient amenorrhic, so as to allow for healing of the fistulae; have been tried and found to have a 5% success rate. Surgeries include, but are not limited to laparotomy, laparoscopy, cystoscopic fulguration and robotic surgery<sup>3, 7-9</sup>. Various approaches via transabdominal like extraperitoneal, transvesical and retrovesical routes have been tried and found to be successful. In our case we approached the fistula by transvesical route by laparotomy.

## Conclusion

Youssef's syndrome is an uncommon vesicouterine fistula. In today's era, when dealing with a case of cyclical hematuria in a patient after caesarean section, one should keep in mind of Youssef's syndrome as a differential. Meticulous practice of obstetric and surgical principles during LSCS and other obstetric and gynaecological interventions can prevent formation of these fistulae.

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**Conflict of interest:** None. **Disclaimer:** Nil.

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