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## Case Report

# Post-caesarean vesicouterine fistula: Youssef's syndrome a case report

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### ABSTRACT

Vesicouterine fistula is an uncommon urogynecological fistula, typically induced following lower uterine segment cesarean section, especially an emergency one. The classical clinical features of Youssef's syndrome are cyclical hematuria, amenorrhea, and urinary continence. The index case reported was a 30 year-old multipara who had two previous lower segment cesarean sections, underwent a third-time cesarean section and developed urinary incontinence and hematuria on the second week of the post-operative period. CECT urography confirmed vesicouterine fistula. The fistula was managed conservatively.

**Keywords:** Cesarean section, Menouria, Vesicouterine fistula, Youssef's syndrome

### INTRODUCTION

A vesicouterine fistula (VUF) refers to abnormal or pathological communication between the epithelial surfaces of the bladder and the uterus or cervix.<sup>1-4</sup> Its incidence is about 1-4% of all cases of urogynecological fistula.<sup>5</sup> 83-93% of cases occur due to lower uterine cesarean section, especially an emergency one.<sup>2-4</sup> Other least common causes of vesicouterine fistula include surgical management of abortion (D and C), obstructed labour, vaginal birth after cesarean section, placenta percreta, forceps delivery, brachytherapy and on rare occasions, migrated intrauterine contraceptive devices.<sup>6</sup> Patients with vesicouterine fistula classically present with cyclic hematuria (menouria), absence of vaginal bleeding (amenorrhea), and complete urinary continence, that is no urine leak per vagina.<sup>7</sup> On rare occasions, urinary incontinence may even be a clinical presentation in patients with vesicouterine fistula, particularly with the involvement of the cervix or the fistula below the isthmus.<sup>1,8</sup>

Diagnosis of VUF is quite tricky. Various methods are obtainable that including methylene blue test, cystoscopy,

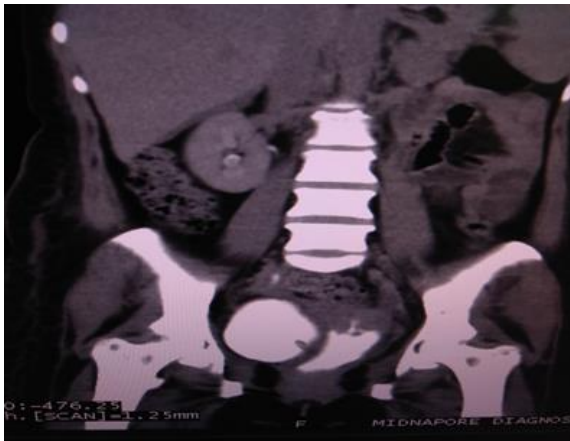
retrograde cystography, hysterosalpingography, contrast-enhanced computed tomography, and Magnetic resonance imaging.<sup>9</sup> However, conservative management may be acceptable in some cases, but the definitive mode of management is surgery. Numerous approaches are followed for surgery like open transabdominal, laparoscopic, and robot-assisted laparoscopic repair.<sup>1</sup>

The index case of vesicouterine fistula after cesarean section presented with a complaint of urinary incontinence and hematuria.

### CASE REPORT

A 30-year-old P2L1 patient underwent a 3rd time lower segment cesarean section at 32±2 weeks' gestation for PPRM and tachycardia with scar tenderness of mother at 2 cm dilatation. Delivery was uneventful although left-sided uterine scar dehiscence was noted, and the bladder was adherent to the lower uterine segment. A Foley catheter was inserted at the time of the cesarean and removed the subsequent day. Ten days post-operation she presented with urinary frequency, urgency, hematuria, suprapubic discomfort and urinary incontinence.

On examination, the patient was afebrile. On per speculum examination, the cervix appears to be healthy. A urine leak has been seen during the speculum examination. Per vaginal examination seems to be normal with red-stained serous lochia. The routine hematological, hormonal and hormonal investigations were normal. Catheterization was done for free drainage and urine was sent for RE/ME/CS. A provisional identification of genitourinary fistula was created. Ultrasound of the abdomen and pelvis showed normal findings. Methylene blue was injected into the bladder through a catheter and leakage of the dye was observed with speculum examination. Intravenous urography demonstrated the fistula and an unknown duplex system on the left, with no proof of hydronephrosis or ureteric injury. CECT scan of urography shows a vesicouterine fistula with extravasation of contrast noted from the left lateral wall of the bladder to the lower uterine canal and adjacent cervix (Figure 1).



**Figure 1: CECT Urography showing Vesicouterine fistula with extravasation of contrast.**



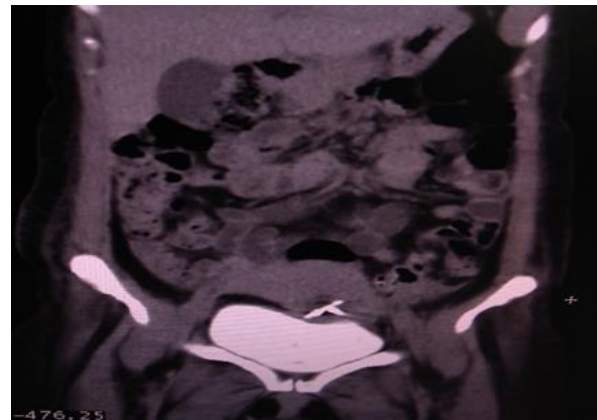
**Figure 2: HSG showing extravasation of dye from lower uterine segment.**

HSG showed extravasation of dye from the lower uterine segment (Figure 2). These findings corroborated with 3D CT urography (Figure 3) where there is a contrast in bladder and lower uterine cavity and cervix but not in the

upper uterine cavity where the IUD (CuT380A) is visible, clearly interpreting vesicouterine fistula. A urethral catheter was left on free drainage for 21 days. As wound swabs grew multi-resistant *Klebsiella pneumoniae*, appropriate antibiotics were started and a repeat evaluation was delayed for a further 21 days. Repeat CECT urography after 4 weeks showed no fistula (Figure 4).



**Figure 3: 3D CT Urography showing dye coming via ureter filling bladder, lower uterine segment and cervix but not upper uterine segment with IUD.**



**Figure 4: CT Urography showing contrast only in bladder not in uterus, only IUD is visible.**

## DISCUSSION

Classical history, cystoscopy, and HSG are sufficient to make the diagnosis. The raised rate of cesarean delivery could presently be the principal explanation for vesicouterine fistulas.<sup>10</sup> The triad of menouria, absence of vaginal leakage of urine and amenorrhea is mentioned as Youssef's syndrome. Youssef hypothesized that the cervical isthmus acts as a sphincter to forestall vaginal leakage of urine. Menouria with apparent amenorrhea may present late, usually a few weeks postpartum, once menses recover. Per the routes of menstrual blood flow, ureterovesical fistulas may present with menouria (type I), with twin flow through both the bladder and vagina (type II) and with traditional vaginal menses (type III).<sup>11</sup>

Menouria should be differentiated from endometriosis of the bladder.

The protrusion of uterine endometrial mucosa hinders the fistulous gap and additionally plays a role in the continence mechanism.<sup>12</sup> The menstrual blood could also be forced to flow into the bladder once the internal uterine orifice is obstructed by cervical fibrosis, which may also prevent urine flow into the vagina. The clarification of the absence of urinary incontinence might be found in the pressure gradients between these two hollow organs. Normally, the pressure in the filling phase of the bladder, in females, rarely exceeds 20 cm of water (5-15 cm H<sub>2</sub>O).<sup>13</sup> Steep rise in detrusor pressure during filling that persists after filling but rarely exceeds 50 cm water. Normally, the intravesical pressure during voiding in females is between 30 and 50 cm water. Intrauterine pressure fluctuates during the phase of the menstrual cycle. It is highest during menstruation when pressures between 130 and 160 cm H<sub>2</sub>O have been recorded. In the intermenstrual phase pressures are well lower; as low as between 35 and 70 cm H<sub>2</sub>O in the proliferative phase and in the secretory phase slightly higher pressure of 55-100 cm H<sub>2</sub>O have been reported.<sup>14</sup> Therefore, the pressure in the uterine cavity appears to be higher than the pressure in the bladder except during micturition.

In 5% of cases, spontaneous healing of uterovesical fistula occurs if early recognition is made and treatment of urinary tract infection and bladder catheterization for at least 3 weeks.<sup>15</sup> Generally, the healing takes place soon after the involution of the uterus, as a result of the recovery of the myometrial tone.

Surgical management is definite for Youssef's syndrome; however, conservative measures such as prolonged catheterization to aid in fistula healing, and medical management like the use of oral contraceptives, progestational agents, local estrogen and gonadotropin-releasing hormone analogue for the induction of amenorrhea, have been reported in the literature with successful outcomes.<sup>16-19</sup>

Small fistulas (<1 cm) may resolve if continuous drainage of the bladder is maintained. In 1985, Zimmern concluded that fistulas have a high spontaneous cure rate with a 3-week trial of Foley drainage. He also noted that if no improvement is observed after 30 days, a VVF is not likely to resolve spontaneously and in that case, prolonged catheterization only increases the risks of infection.<sup>20</sup> In this case, spontaneous closure of the fistula occurred after 3 weeks of continuous drainage. Estrogen therapy might help with optimizing tissue vascularization and healing and NSAID therapy might help to minimize early inflammatory changes at the fistula site in this case.

## CONCLUSION

Youssef's syndrome is a rarely encountered urogynecological fistula. However, with the increasing

rate of repeat cesarean deliveries, doctors got to remember this complication as a vital risk associated with repeat cesarean deliveries. They would conjointly need to discuss this risk once patients consent for repeat cesarean deliveries. Furthermore, it might be affordable to make sure that the foremost senior doctor who has been trained to proficiency performs repeat cesarean deliveries as a precaution to prevent VUF and other morbidities associated with this procedure.

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