

# Youssef Syndrome: A Case Report with Review of Literature

Short Running Title: Utero-vesical fistula

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**Abstract:** *Uterovesical fistula, a least common type of urogenital fistula is a rare complication following caesarean delivery. With patient history and selected investigations the diagnosis of uterovesical fistula is relatively easy. The surgical repair of these fistulae is standard treatment. Meticulous practice of obstetric and surgical principles will prevent the formation of these fistulae. Here we report a case of uterovesical fistula in a 38 year old woman following caesarean section.*

**Keywords:** Utero-vesical fistula, Youssef Syndrome, Caesarean section, cyclical haematuria, menuria

## 1. Introduction

Vesico-uterine fistula is the least common of all the urogenital fistulae, accounting for 1% to 4% (Lenkovsky 1988). They are most frequently caused by repeated caesarean sections, which are increasing. Till date around 800 cases have been reported in the world literature. Majority of patients present with urinary incontinence in the early post operative period. However, a few may present months or years after their caesarean sections with recurrent cyclical painless haematuria with or without vaginal leakage of urine. We report a case of uterovesical fistulas following caesarean section which was repaired successfully by surgery followed by brief review of literature.

## 2. Case Report

Mrs. X, a 38 year old lady attended our gynaecology Outpatient department with complaints of cyclical haematuria (menouria) for the past 4 years. She had undergone two Caesarean sections in the past; last one was 4 years ago. Both caesarean sections were done for cephalopelvic disproportion in a private hospital. Intraoperative details were not available. She did not give any other significant medical or surgical history. General examination and abdominal examination did not reveal any abnormality. On Speculum examination, Cervix was drawn up behind pubic symphysis and could not be visualised properly. Vaginal leakage of urine could not be demonstrated. Pelvic examination showed a retroverted uterus deflected to right side.

Routine laboratory tests revealed nothing abnormal. Urine culture and sensitivity did not reveal any growth. Ultrasonography showed uterus and ovaries to be normal with bilateral renal calculi and no hydronephrosis. Cystoscopy revealed a fistulous opening in the posterior wall of bladder above the trigone of the bladder (figure 1).

Injection of methylene blue into the uterus revealed the dye coming out through the bladder through the fistulous opening confirming the diagnosis of uterovesical fistula (figure 2). CT Abdomen (contrast) was done to delineate the fistulous track but no communication was seen in both initial and delayed films.

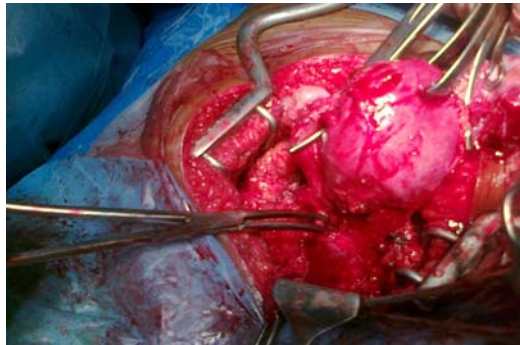


**Figure 1:** Cystoscopic picture showing fistulous opening in the posterior wall of the bladder



**Figure 2 :** Cystoscopic picture showing the dye coming out through the fistulous opening after injecting methylene blue in the bladder

She was planned for laparotomy and proceed. Intraoperatively dense adhesions were seen between bladder and anterior wall of uterus. A fistulous opening was seen in the posterior wall of the bladder just above the inter ureteric ridge and just above the level of isthmus in the uterus (figure 3). She successfully underwent transvesical transperitoneal repair of fistula (Cohenn's fistulectomy) and hysterectomy was done as patient was insisting and due to extensive adhesions.



**Figure 3:** Fistulous opening just above the isthmus in the uterus

She was on continuous bladder drainage for 14 days. Her postoperative period and follow-up were uneventful.

### 3. Discussion

Uterovesical fistula, a rare type of urogenital fistula was first reported in 1908. In 1957, Youssef described a syndrome characterized by cyclic hematuria, amenorrhea without vaginal leakage of urine (Youssef 1957). Our patient satisfied all the triad features of Youssef syndrome.

In earlier days uterovesical fistula resulted due to difficult vaginal deliveries, neglected obstructed labour and high delivery by forceps. But the recent increase is due to increase in caesarean section. Other less common causes are placenta percreta with bladder invasion (Majeed & Subhani 2007), manual removal of placenta in a previous caesarean patient, pelvic irradiation and cervical malignancies. Cases have also been reported following uterine artery embolisation for leiomyoma (Sultana et al 2002) and migration of IUCD (Schwartzwald et al 1986). Predisposing factors during caesarean may be due to inadequate mobilisation of bladder or accidental inclusion of bladder in the suture while closing the uterine incision.

**Józwik and Józwik** (2002) classified uterovesical fistula based on the location of fistula on the uterus and the route of menstrual flow. In pre-isthmus fistula (Type I) there will be only menouria with no regular menstrual flow and no urinary incontinence. In fistulas located at isthmus (type II) there will be coexistence of menouria, menstrual flow and incontinence. In Postisthmus fistula (Type III) there will be urinary incontinence, regular menstrual flow and no menouria. Other Atypical presentations include recurrent UTI, secondary infertility, first trimester abortions, urge incontinence and urethral passage of lochia.

Intraoperative diagnosis is the gold standard in detecting vesicouterine fistulas for allowing immediate repair.

Cystoscopy helps in identifying the fistula, determining the size and its location in relation to trigone and ureteric orifice. Other modalities for diagnosing uterovesical fistula are intravenous urography, hysterosalpingogram and Colour doppler USG / contrast helical CT (Kilinc et al 2003). It is important to differentiate this condition from endometriosis of the bladder.

Though surgery is the mainstay of treatment conservative management have been reported in few small fistulas which were immediately diagnosed such as continuous bladder catheterisation for 4-8 weeks, hormonal therapy and cystoscopic fulguration of the tract (Molina et al 1989, Ravi et al 2003). Spontaneous closure has been reported in 4%. Among the surgical approaches, transvesical transperitoneal approach has the lowest relapse rate (Porcaro et al 2002).

To conclude, Vesicouterine fistulae, despite being infrequent, are no longer a rare diagnosis. Prevention is better than cure. Hence, meticulous practice of surgical principles at caesarean section such as downward retraction of the bladder, correct identification of the anatomical landmarks while suturing and proper evaluation of intraoperative hematuria will help in preventing the formation of these fistulas.

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