A Case Report of Post Cesarean Section Vesico-Uterine Fistula (Youssef’s Syndrome) and Review of the Literatures

Belaynew Keleb Lake (belk8247@gmail.com)
Bahir Dar University

Getu Dinku Heyi
Bahir Dar University

Kassahun Alamrew Tsega
Bahir Dar University

Meseret Ahunem Workneh
Bahir Dar University

Aklilu Tesega Ayicheh
Bahir Dar University

Zelalem Ayichew Workineh
University of Gondar

Case Report

Keywords: Vesico-uterine fistula, Youssef’s syndrome, fistula repair, laparotomy

Posted Date: January 2nd, 2024

DOI: https://doi.org/10.21203/rs.3.rs-3763990/v1

License: This work is licensed under a Creative Commons Attribution 4.0 International License.
Read Full License

Additional Declarations: No competing interests reported.
Abstract

**Background:** Vesico-uterine fistula is one of the rare types of urogenital fistula that accounts for only 1-4%. Repeat cesarean section is a known cause of vesico-uterine fistula. Patients with Youssef’s syndrome had a classic triad of absence of incontinence, amenorrhea, and cyclic hematuria.

**Case presentation:** A case of surgically managed post-cesarean section vesico-uterine fistula (Youssef’s syndrome) patient after she presented with triads of cyclic hematuria, amenorrhea, and absence of urinary incontinence of 4 years duration which had managed by laparotomy with repair of fistulous tracts.

**Conclusion:** Patients with Youssef’s syndrome can present at any time following cesarean section, and diagnosis needs a high index of suspicion. Surgical repair of the fistulous tract is the mainstay of management.

Introduction

Vesicouetrine fistulas (VUF) are abnormal communications between the bladder epithelium and the epithelial layer of the uterus or cervix. These are very rare types of urogenital fistulas that occur as a rarest complication of cesarean section. The prevalence is 1-4% (1) of all urogenital fistulas. Clinical presentation of patients may vary depending on the type of vesico-uterine fistula (2). Depending on the type of vesico-uterine fistula patients may present with classic triads of symptoms of continence, cyclic hematuria, and amenorrhea (3) to only urinary incontinence (4). Because of the atypical presentation high index of suspicion is paramount and appropriate diagnostic modalities should be done to confirm the diagnosis (5). Management can be either conservative by bladder catheterization for at least 4 weeks when the fistula is diagnosed immediately after delivery (6) or surgical is definitive and the mainstay of management (7, 8).

Case presentation

36-year-old Para II woman both deliveries were via cesarean section 9 and 4 years back. The 2nd cesarean section was done 4 years back at a primary hospital after she was allowed to have a trial of labor. Intraoperative bladder injury was identified and repaired and the catheter was kept for 10 days. Following catheter removal she failed to control urine. She had repeated hospital visits and for complicated urinary tract infection treated with antibiotics. She had also repeated visits to private clinics and for recurrent urinary tract infections variety of antibiotics was given. 4 years after the surgery she presented to our Urogynaecology clinic complaining of cyclic hematuria, amenorrhea, recurrent lower urinary tract infections, and a history of infertility but no urinary incontinence. Following cyclic hematuria she had difficulty with urination and a burning sensation during urination.

Examination revealed stable vital signs, speculum examination revealed unremarkable findings. Her CBC and organ function tests were normal. Abdominopelvic Ultrasound showed deficient anterior myometrium
at the lower segment with communicating endometrium with the bladder neck. Transvaginal ultrasound revealed anterior uterine wall defect in direct communication with the bladder. Contrast-enhanced pelvic CT scan delayed phase revealed a 1.0cm sized posterior bladder wall defect through which contrast passes and communicates to lower parts (2cm cranial to ectocervix) of the endometrial cavity in delayed phase post-contrast study (Image 1). Before laparotomy cystoscopy was done and there was a communicating band-like structure seen at the bladder mucosa (image 2).

Upon laparotomy there was no significant intrabdominal adhesion; the pelvic organ looks healthy except there is dense adhesion over vesicouetrine reflection (image 3). Upon sharp dissection, both bladder and uterine defects (fistulas) were identified fistula mobilized, catheter tip visible, and scarred tissue trimmed (images 4 & 5). After identification of bilateral ureteric orifices that were far from the fistula site bladder and uterine defects were repaired separately with vicryl 2/0 and 1 respectively. Postoperatively she was on continuous drainage for 3 weeks with a catheter. She had been on follow-up at the outpatient clinic for 6 months and she is continent for urine and there was no menouria.

**Discussions**

Historically the first patient presented with hematuria as a result of vesico-uterine fistula was first reported by Machado 1935 (4). Later in 1957 Youssef coined the term Menouria (9) and termed as Youssef’s syndrome (10). The prevalence of vesico-uterine fistulas is rare 1-4% of all urogenital fistulas (1) its prevalence is increasing due to increment in cesarean rate along the globe (7, 11).

Vesico-uterine fistulas can be classified in to three based on the clinical presentations (2, 12). Type I (Youssef’s syndrome) had amenorrhea, cyclic hematuria, and absence of urinary incontinence. 90% of Vesico-uterine fistulas are Youssef’s syndrome (3). Type II vesico-uterine fistula is defined when there is urinary continence, cyclic hematuria, and regular menses. The presence of urinary incontinence with the absence of Menouria and regular menses is Type III Vesico uterine fistula.

Majority of the Youssef’s syndrome occurred following lower segment Cesarean section similar to our case (4, 13-18) in approximately 83-93% (19). Other causes of Youssef’s syndrome can be sloughing of an intrauterine catheter into the bladder (20), uterine curettage (21), vaginal lower segment operations (22), and forceps delivery (22), Gossypiboma (2).

Inadvertent sutures applied to bladder while uterus is repaired, abnormal vascular bed and blood supply to the bladder from multiple dissections during repeat cesarean section (23), hysteroscopic dilation and curettage, use of forceps or vacuum, placenta previa totalis and morbidly adherent placenta, history of repeated abortion, uterine rupture, inadequate mobilization of the bladder during cesarean section (3, 13) and infectious causes like tuberculosis (13). Vesico-uterine fistulas can be prevented by emptying the bladder and by careful dissection of the lower uterine segment.

Classically patients with Youssef’s syndrome occur late after months to years of the incident. The presence of cyclic hematuria, amenorrhea, and absence of menses is a classic triad of this syndrome (9,
History suggestive of Vesico-Uterine fistula should undergo confirmatory diagnostic modalities. Diagnosis can be confirmed with cystoscopy, hysterosalpingography, intra-venous pyelography, sonography (24), sonohysterography (25), CT and MRI (5). Our case had a typical clinical presentation and diagnosis was confirmed with cystoscopy, ultrasound, and pelvis CT scan.

Management of Vesico-uterine fistula can be conservative management (6, 26), fulguration, hormone therapy (26), and surgical management (27). Surgical management timings can be immediate repair within 48 hours or repair after 2-3 months of diagnosis (27). Depending on the site and the number of fistulas surgical approaches can be vaginal, trans-vesical-retroperitoneal, trans-peritoneal, or combined abdominal and vaginal (8, 28). Surgical repair of the bladder with hysterectomy is the best management option for patients who completed fertility or had uterine pathologies (26). Preservation of uterine function with successful pregnancy outcome is reported in 31.25% and term deliveries in 25% (8, 29). After surgical repair, it is generally recommended to have a repeat cesarean section because the risk of fistula recurrence is high (8). Our case undergoes laparotomy followed by fistula resection, bladder, and uterine repair. Patient clinical presentation may not be straightforward and a high index of suspicion and confirmatory imaging should be done once the diagnosis is made management should be discussed with the patient and appropriate intervention should be given.

**Conclusion**

Patients with Youssef's syndrome can present at any time following cesarean section and diagnosis needs a high index of suspicion. Surgical repair of the fistulous tract is the mainstay of management.

**Declarations**

**Consent for picture**

Taken from the patient for picture in native language Amharic and attached

**Consent for online publication**

Consent taken from the patient for an online publication in native language Amharic and attached

**Funding**

No funding sources

**Conflict of interests**

We have read and understood BMC's policy on declaration of interests and declare that we have no competing interests

**Ethical approval**
Not required but if needed we can have

**Accessibility on online data base**

There is no raw data because it is a case report but if needed all authors agree to open accessibility

**References**


**Figures**
Figure 1

Contrast enhanced Pelvic CT scan delayed phase sagittal reconstructed image showing 1.0cm sized posterior bladder wall defect communicating to lower parts of the uterus
Figure 2

Cystoscopy showing band like structure at the interior of the bladder
Figure 3

Intraoperative adhesion between bladder and lower uterine segment
Figure 4

Bladder fistula after mobilization

Figure 5
Bladder fistula with visible catheter tip before mobilization