Case Report

Incidentally Diagnosed Type 2 Youssef’s Syndrome with Migrated Postcesarean Postpartum Intrauterine Contraceptive Device

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Abstract

Vesicouterine fistula (VUF) also called Youssef’s syndrome is a rare and the least commonly encountered entity among genitourinary fistulae. Treatment options range from excision of the fistulous tract with hysterectomy and primary closure of the urinary bladder to fistula repair. Migrated postpartum intrauterine contraceptive device is one of the rarest causes for VUF. Here, we are describing a case of incidentally diagnosed VUF with Type 2 Youssef’s syndrome with migrated postplacental (postcesarean) intrauterine contraceptive device, treated with uterus-sparing laparoscopic VUF closure.

Keywords: Genitourinary fistula, hysteroscopy, laparoscopy, vesicouterine fistula

Introduction

Classical Youssef’s syndrome consists of cyclical hematuria, amenorrhea, menouria, and urinary incontinence due to vesicouterine fistula (VUF) developing after cesarean section (CS).1–3 Clinical presentations may vary with the absence of one or more symptoms. It is the least commonly encountered of all the genitourinary fistulae. Various causes are CS, vaginal birth after cesarean section, obstructed labor, placenta percreta, brachytherapy, traumatic bladder catheterization, uterine artery embolization, dilatation and curettage, and manual removal of placenta. Migrated intrauterine contraceptive device, inflammatory bowel disease, bladder tuberculosis, endometriosis, and congenital lesions are among the rare causes.

Postpartum intrauterine contraceptive device (PPIUCD) is a reversible, highly effective, long-acting contraceptive that can be initiated in the early postpartum period. As it has been used more frequently, more of its complications are getting known and one of these is migrated intrauterine contraceptive device (IUCD).

Case Report

A 30-year-old female, P3L1, reported chief complaints of cyclical hematuria and irregular menses for 4–5 months. She did not take any treatment for that. The patient had three previous CSs 8, 4, and 2 years back, respectively. Postplacental PPIUCD (copper-T [Cu-T]) was placed during the last CS. She requested the removal of Cu-T as she attributed her symptoms to it. Her general examination was normal. On per speculum examination, Cu-T thread was not seen. Per vaginal examination was unremarkable. Ultrasound showed Cu-T in the endometrial canal. Her routine investigations were normal. She was planned for hysteroscopic Cu-T removal.

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The vertical limb and half arm of Cu-T got removed, however, one limb of the horizontal arm could not be removed. During the procedure, a suprapubic bulge was noticed. Due to suspicion of bladder perforation, per urethral catheterization was done which drained 300 cc of blood-tinged urine. A urologist was called for an opinion, and cystoscopy revealed bilateral normal ureteric orifices along with a rent of around 1 cm × 1 cm in bladder mucosa at 7 o’clock position on the right posterolateral wall with part of Cu-T seen projecting through it. Rent was epithelized showing smooth margins without evidence of fresh bladder perforation. Diagnosis of VUF with migrated PPIUCD was made. The procedure was withheld for further work-up. Contrast-enhanced computed tomogram abdomen and pelvis with urogram was done showing fistulous communication between uterus and urinary bladder with embedded Cu-T arm [Figure 1]. The patient was planned for laparoscopic exploration and repair of VUF as she wanted to preserve the uterus.

Posterior cystotomy was done onto the fistulous opening laparoscopically [Figure 2a]. The Cu-T arm was removed intact [Figure 2b]. The fistula was excised and the uterine rent was repaired with 3-0 polyglactin suture. Urinary bladder was closed in layers with 3-0 polyglactin suture with interposed appendices epiploicae. Per urethral, 20 Fr two-way Foley’s catheter was left in situ. 20 Fr abdominal drain was also placed. Postoperative recovery was uneventful. Abdominal drain was removed on the third postoperative day. Foley’s catheter was removed 4 weeks postsurgery after cystographic confirmation of healing.

**DISCUSSION**

VUF is the least common of urogynecological fistulae, formed most commonly after CS. It is more common in patients undergoing repeat CS. Mechanisms proposed are undetected bladder rupture during emergency CS, usually with fetal presentation already engaged, caused by insufficient dissection and/or insufficient drainage of the organ, instrumental delivery, manual removal of placenta, or history of abortion. Less common causes include endometriosis, inflammatory bowel disease, and migration of IUCD. Józwik and Józwik classified VUF into three types. Type 1 includes patients with amenorrhea with menouria (cyclical hematuria), the classical Youssef’s syndrome. Type 2 includes patients with hypomenorrhea and menouria. In Type 3, patients have no menstrual complaints and no menouria. Maximum patients fall under Type 1 category (90%). Our patient had Type 2 VUF.

Contraceptive options in early postpartum period are very limited. PPIUCD is one of the long-acting, highly effective contraception initiated by the Government of India. PPIUCD insertion can be postplacental (inserted within 10 min of delivery), intraccesarean, and within 48 h postpartum. Common side effects of IUCD are increased menstrual bleeding, spotting, and abdominal cramps. Migration of IUCD can also occur rarely. In our case, possibly, a part of the bladder was taken during uterine closure and the IUCD migrated to incision (due to uterine involution) before it healed completely leading to VUF formation with Cu-T arm projecting into the urinary bladder. Our patient presented with intermittent spotting and menouria, which she attributed to PPIUCD. Her IUCD was found malpositioned, i.e. being present inside the uterus, but its placement was eccentric as indicated by nonvisualization of thread and difficulty encountered during its removal. There was a possibility of its being embedded partly or fully in the myometrium.

The incidence of lost string of PPIUCD varies from 5.3% to 24%. Majority of the lost strings are lying just curled inside the cervical canal (91.8%). Reason could be a large uterine cavity and process of involution which involves uterine contraction. USG or X-ray should be done to know the position. Hysteroscopic Cu-T removal is a good option.

![Figure 1: Contrast-enhanced computed tomography of pelvis ([a] axial, [b] oblique sagittal reformat) shows part of intrauterine contraceptive device extending through the anterior myometrium piercing through the posterior urinary bladder wall and protruding into bladder lumen (yellow arrow), (c) Delayed (after 15 min) oblique sagittal reformats reveal contrast in urinary bladder lumen and thin streak of contrast within endometrial cavity (red arrow) suggesting vesicouterine fistula](image-url)
Various diagnostic modalities for VUF include cystoscopy, voiding cystourethrography, and pelvic Computed Tomogram (CT)/magnetic resonance imaging (MRI) scan. Pelvic MRI shows high diagnostic accuracy, especially in confusing clinical picture.[6,7]

Small fistula can be treated conservatively if detected early with a reported success rate of 5%.[8] For large and/or nonhealing fistula, surgical approach is preferred. Options include open and minimally invasive (laparoscopic/robotic) procedures. Hysterectomy is an option if the patient has no further desire for fertility,[9] otherwise fistula repair is essential and success rate is high. Even pregnancy has been reported after repair.

As our patient was young and with only one live issue and wanted uterus preservation, transabdominal laparoscopic repair of fistula was successfully done.

**Conclusion**

VUF is a rare complication of PPIUCD. It might be difficult to discriminate intermittent menouria associated with VUF from intermittent spotting associated with PPIUCD. High index of suspicion should be there in case of irregular cycles with menouria after CS and PPIUCD insertion. History and high index of suspicion are key to diagnosis. Treatment can be conservative, medical, or surgical. Hysterectomy is preferred if family is completed, but uterus-sparing procedure can be done successfully in expert hands.

**Ethical approval**

This report has been approved by Institutional Ethics Committee of All India Institute of Medical Sciences, Rishikesh (approval no. AIIMS/JDH/OBG/2020/287). The informed patient consent was obtained.

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**Conflicts of interest**

There are no conflicts of interest.

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