Successful endoscopic management for early manifested postcesarean section uretero-uterine fistula: A case report and literature review

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INTRODUCTION

Obstetric and gynecological procedures associated with iatrogenic urinary tract injuries account for 0.4%–2.5% for benign conditions. Uretero-uterine fistula (UUF) is a rare presentation that constitutes 1.7%–5.1% of all urogenital fistulas.[1]

To our knowledge, there are 54 cases of postobstetric/gynecological UUF reported in the literature up to date.

Iatrogenic UUF was reported postcesarean section in most cases. Management depends mainly on surgical repair as reported in more than two-thirds of cases; however, there is still a role for minimally invasive procedures such as Ureteroscopy (URS) and internal JJ ureteric stenting.

We report a rare case of postcesarean section uretero-uterine fistula (UUF) in a 36 years old female who presented on a postoperative day 5 with paradoxical urine incontinence and occasional normal urethral voiding. After a complete evaluation, Cystoscopy and right internal JJ ureteric stent insertion was done; the patient had a dramatic response; she was completely dry until the stent was removed three months later with complete healing of UUF. To our knowledge, the present case is the 4th reported with successful endoscopic management for post-obstetrics and gynecological procedures Uretero-Uterine fistula.

CASE REPORT

A 36-year-old female patient p3 + 1 underwent emergency cesarean section for uterus rupture on the 36th gestational week and delivered a live boy weighing 2810 g.
The postoperative period was uneventful until postoperative day 5 when the patient experienced paradoxical urine incontinence with normal urethral voiding; the abdominal examination was unremarkable; pelvic examination showed a defect in the anterior fornix and urine pooling in the posterior fornix. KUB ultrasound of kidney, ureter, and bladder showed mild right hydronephrosis, so computed tomographic (CT) urography was done and showed mild right hydronephrosis and contrast extravasation at the lower right ureteric segment with right UUF [Figure 1a]; we excluded vesicovaginal and vesicouterine fistula after three swab tests.

Hysteroscopy showed urine coming from cervical Os. Cystoscopy and Right retrograde ureteropyelography showed a medial deviation of the lower Right ureteric segment (fistula site) [Figure 1b], and the diagnosis of the right UUF was confirmed.

Using cystoscopy, right JJ ureteric stent was inserted successfully [Figure 1c]. Day 1 poststent fixation, the patient had a dramatic response, and she was completely dry till the stent removed 3 months later. At that time, right Retrograde Pyelography (RGP) showed normal course and caliper of the right ureter [Figure 1d]. Follow-up CT urography done 4 weeks poststent removal revealed no more backpressure with complete healing of UUF [Figure 1e].

**DISCUSSION**

Ureteric injury during obstetric and gynecological procedures is rare and accounts for less than 0.5%–1% and may rise to 2% in radical hysterectomy. There are many factors suggested predisposing for iatrogenic ureteric injury during obstetric and gynecological procedures, including previous cesarean section due to fibrosis and ureteric displacement, dextro-rotation of the uterus explaining the more frequent injury of the left ureter, and also prolonged labor with disproportion that might result in ureteric wall edema and necrosis. The lower third segment of the ureter can be injured through a low uterine transverse incision or by too far lateral incision extension, inadvertent suture ligation, or hemostatic clip that might result in a hematoma, infection, and fistulous tract formation between the uterus and injured ureter.

UUF can be presented early days postoperative as in our case or a few weeks later. It can be explained by injury mechanism, which would present early in patients with direct ureteric injury, or later with ischemic injury. UUF usually presents with paradoxical urine incontinence and normal urethral voiding.

Diagnosis can be confirmed with urine dribbling from cervical uteri orifice on speculum examination; a three-swab test, and cysto-panendoscopy to rule-out vesicovaginal and vesicouterine fistulae. Sheen et al. modified the swab test by phenazopyridine administration every 8 hours followed by installation of 200 cc of methylene blue into the urinary bladder after 24 hours; yellow urine from the vagina could confirm UUF and exclude vesicovaginal fistula. Intravenous urography and CT urography can identify the location of the ureteric injury, extravasation, or even the fistulous tract and give an idea about renal function. Retrograde ureteropyelography can delineate the lower ureteric segment that not-enhanced in the contrast study and assess ureteric continuity to help in decision making.

UUF management’s rationale depends on the early minimal invasive procedures’ intervention that can guarantee the continuity of the injured ureter, preserve renal function, and prevent urine leakage with subsequent infection and tissue necrosis. Percutaneous nephrostomy can ensure urine diversion with complete healing of UUF as reported in two cases, but the surgical repair was mandatory later in six patients. On the other hand, internal JJ ureteric stent for UUF was successful as a treatment option for UUF in reported four cases, including the current case, and failed in three cases.

Surgical repair is the mainstay in treating uretero-uterine fistula, as reported in > 68% of cases, especially in delayed presentation.
and failed PCN or endoscopic management. The surgical approach depends mainly on UUF location, length of ureteric segment involved, the extent of fibrosis, and surgeon preference. It includes uretero-ureterostomy, ureteroneocystostomy with psoas hitch and boari flap if indicated, and human dura matter allograft ureteroplasty. Surgical intervention could be open, laparoscopic, or robot-assisted laparoscopic repair.

We reviewed the previously reported cases (n=54) of post-obstetrics and gynecology procedures UUF as shown in Tables 1 and 2; we found that 80% of patients followed caesarian section, 10% post-hysterectomy, and 10% post-abortion (D&C). UUF was left side in 64% of reported cases. Presentation ranged between one day and six months postoperative (median=20 days). The treatment options included spontaneous healing in one case, percutaneous nephrostomy was sufficient for UUF healing in only 25%, endoscopic intervention and JJ stent insertion was successful in 57%, and surgical repair was the gold standard treatment option offered to most patients (n>30) with a success rate > 96%

The present case of right UUF presented early days postcesarean section. It was managed successfully with cystoscopy and internal JJ ureteric stent for 3 months, obviating the need for percutaneous nephrostomy and/or surgical repair with its morbidity and complications.

**CONCLUSION**

Endoscopic management for postobstetric and gynecological UUF is a feasible and less invasive option and can be offered before surgical repair, especially to early presented cases.

**Consent**

We obtained written informed consent for publication from the patient; a copy is available for review.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name

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**Table 1: The previously published cases of post-obstetric and gynecology procedures Uretero-Uterine Fistula**

| Author            | Cases number | Etiology          | Time to present | Diagnosis                  | Side  | Management                         | Outcome      |
|-------------------|--------------|-------------------|-----------------|----------------------------|-------|------------------------------------|--------------|
| De Aguiar, 1959   | 1            | LSCS              | -               | -                          | -     | -                                  | -            |
| Zeliński, 1962    | 1            | -                 | -               | -                          | -     | -                                  | -            |
| Claret al., 1964  | 2            | -                 | -               | -                          | -     | -                                  | -            |
| Truc et al., 1969 | 1            | -                 | -               | -                          | -     | -                                  | -            |
| Mahgoob and Zeniny, 1971 | 1 | LSCS              | -               | IVU, dye test, HSG         | Left  | UNC                               | Cured        |
| Suhler and Saout, 1971 | 1       | LSCS              | -               | -                          | -     | -                                  | -            |
| Dukhovnikov et al., 1973 | 1 | LSCS              | -               | -                          | -     | -                                  | -            |
| Jequier and Piper, 1973 | 1 | LSCS              | 2 weeks         | Dye test, CPE, IVU, RGP  | Left  | PCN for 7 weeks                  | Cured        |
| Barton et al., 1978 | 1           | Abortion, D and C | -               | IVU, RGP                   | Right, left | UNCP                              | Cured        |
| Oumachigui et al., 1980 | 3 | LSCS              | 9 days 15 days 23 days | IVU, dye test, CPE         | Right, left | UNCP                              | Cured        |
| Moussu et al., 1980 | 1           | LSCS              | -               | IVU, RGP                   | Left  | UNC + psoas hitch                 | Cured        |
| Keegan and Forkowitz, 1982 | 1 | D and C           | 1 week          | IVU                        | Right | PCN, UNC + psoas hitch            | Cured        |
| Docquier, 1986     | 1            | LSCS              | -               | -                          | -     | -                                  | -            |
| Gorrea et al., 1986 | 1        | LSCS              | -               | IVU, RGP                   | -     | Spontaneous healing                | Cured        |
| Pernin et al., 1993 | 1           | -                 | -               | -                          | -     | -                                  | -            |
| Natta et al., 1993 | 1            | LSCS              | -               | -                          | -     | -                                  | -            |
| Sharfi et al., 1994 | 8           | LSCS, hysterectomy| -               | IVU, dye test, cystoscopy, RGP | -     | UNC + psoas hitch (3 cases) + 3 boari flap (3 cases) | All cured |
| Fernandez et al., 1994 | 1         | AH                | -               | -                          | -     | -                                  | -            |
| Sallutti et al., 1994 | 1         | LSCS              | 20 days         | IVU, CT, cystoscopy        | Left  | PCN, UNCP                          | Cured        |
| Lodh et al., 1996  | 1            | Abortion, D and C | 6 months        | IVU, RGP                   | Right | UNCP + boari flap                  | Cured        |
| Lazarevski and Badiev, 1996 | 1 | LSCS              | 2 weeks         | IVU, dye test, HSG         | Left  | UU, PCN, UNCP                       | Cured        |
| Wang and Hung, 1997  | 1          | Abortion, D and C | -               | IVU, CT                     | Right | PCN, UNCP, UNCP + boari flap       | Cured        |
| Kajbafzadeh, 1997 | 1            | LSCS              | -               | Cystogram, IVU, RGP, dye test | Cystography | Ureteric dilatation and DJS       | Cured        |
| Sheen et al., 1998 | 1            | LSCS              | 4 weeks         | IVU, dye test, HSG         | Left  | UNC (Politano–Leadbetter technique)| Cured        |

LSCS: Lower-segment cesarean section, AH: Abdominal hysterectomy, IVU: Intravenous urography, HSG: Hysterosalpingogram, CPE: Cystopyopendoscopy, RGP: Retrograde pyelogram, CT: Computerized tomography, UNC: Ureteroneocystostomy, PCN: Percutaneous nephrostomy, UU: Ureteroureterostomy, DJS: Double-J stent
Table 2: The previously published cases of post-obstetric and gynecology procedures Uretero-Uterine Fistula

| Author            | Cases number | Etiology | Time to present | Diagnosis | Side | Management | Outcome   |
|-------------------|--------------|----------|-----------------|-----------|------|------------|-----------|
| Nabi et al., 2000 | 1            | LSCS     | Week            | IVU, CPE, TST | Left | UNC        | Cured     |
| Nouira et al., 2000 | 1        | LSCS     | -               | -         | -    | URS, exploration of the ureter | -        |
| Singh et al., 2001 | 1            | LSCS     | 2 weeks         | IVU, CPE, dye test, Moir’s test | Left | URS, balloon dilatation, DJT | Cured     |
| Billmeyer et al., 2001 | 1        | LSCS     | -               | IVU, cystoscopy, TST | Left | Repair of VVF, UNC | Cured     |
| Kozlak et al., 2004 | 1            | LSCS     | -               | IVU       | -    | Repair with human dura mater allograft for stricture ureter | Cured     |
| Adhikary et al., 2005 | 1            | D and C  | 1 day           | IVU, CPE, RGP | Left | UNC + psoas hitch | Cured     |
| El-Tabeel et al., 2006 | 1            | LSCS     | -               | IVU, cystoscopy | Left | UNC + boari flap | Cured     |
| Eze, 2007          | 1            | LSCS     | -               | -         | -    | -          | -         |
| Lanary et al., 2008 | 1            | LSCS     | 18 days         | CT/IVU, cystoscopy, URS | Right | PCN, UNC | Cured     |
| Kumar et al., 2011 | 1            | LSCS     | Week            | IVU, cystoscopy, CT nephrostogram | Left | PCN, laparoscopic UNC | Cured     |
| Levy et al., 2012  | 1            | LSCS     | 2 days          | CT, CPE   | Right | PCN        | -         |
| Shetty et al., 2016 | 1            | -        | -               | CT, cystoscopy, RGP | Left | DJT for 6 weeks | Cured     |
| Lo et al., 2016    | 3            | ELSCS    | -               | IVU, CT, dye test, cystoscopy | Left | UNC        | Cured     |
| Yamamoto et al., 2017 | 1            | LSCS     | -               | IVU, CT, RGP, dye test, CPE | Right | UNC + psoas hitch, hysterectomy | Cured     |
| Sharma and Ratkal, 2019 | 3            | LSCS     | 2–3 weeks       | IVU, CT, RGP | Right | UNC        | Cured     |
| Selvaraj et al., 2020 | 1            | LSCS     | 2 months        | CT, CPE, RGP | Right | Robot-assisted laparoscopic bilateral ovariopreserving hysterectomy, UNC + boari flap and psoas hitch | Cured     |
| Current case       | 1            | LSCS     | 5 days          | CT, TST, CPE, RGP | Right | DJT stent for 3 months | Cured     |

LSCS: Lower-segment cesarean section, ELSCS: Emergency LSCS, CPE: Cystoscopy, TST: Three-swab test, HSG: Hysterosalpingogram, RGP: Retrograde pyelogram, UNC: Ureteroneucystostomy, PCN: Percutaneous nephrostomy, UU: Ureteroureterostomy, URS: Ureteroscopy, DJS: Double-J stent, IVU: Intravenous urography, CT: Computerized tomography, VVF: Vesicovaginal fistula

and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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