International Journal of Clinical Obstetrics and Gynaecology

ISSN (P): 2522-6614
ISSN (E): 2522-6622

www.gynaecologyjournal.com
2019; 3(1): 196-198
Received: 16-11-2018
Accepted: 22-12-2018

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Pregnancy with neglected Youssef syndrome a case report

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DOI: https://doi.org/10.33545/gynae.2019.v3.i1d.32

Abstract
A rare complication of cesarean section (CS), vesicouterine fistula in a 29 year female is presented. She was a case of Youssef Syndrome but with additional features in pregnant state. She had 18 weeks pregnancy with VUF with previous 3 CS manifesting as hematuria, and in non pregnant state as cyclic hematuria and amenorrhoea. Patient was treated with hysterotomy with bladder repair while saving the uterus. She regained normal vaginal menses.

Keywords: Cesarean section, transabdominal repair, vesicouterine fistula, Youssef's syndrome

Introduction
Vesicouterine fistula (VUF) is a rare form of Urogenital fistula, with pathological communication between the epithelial surfaces of the bladder and the uterus or cervix.
It has incidence of 1-4%. The common reason for the fistula is the bladder injury during lower Segment Caesarean Sections (LSCS) [1, 2].
Incidence of bladder injury during caesarean is 1:1000 as per RGOG [3]. Chances of such injury increase to 3 folds i.e. 0.6% v/s 0.19% in repeat caesarean section (CS) [4].
VUF was first reported in the literature in 1908 by Knipe [5]. However, it was described by Youssef in 1957 as a clinical syndrome consisting of cyclic hematuria (Menouria) amenorrhea, and complete urinary incontinence in a patient who had LSCS. Youssef was also the first to coin the term "Menouria" resulting from VUF [6].
The VUFs, which are observed after caesarean delivery, are not accompanied by urinary incontinence, instead they have amenorrhoea and cyclic hematuria, and are called Youssef's syndrome [7].
This is a of case of Youssef Syndrome but with additional features in pregnant state. She had 18 weeks pregnancy with VUF with previous 3 CS manifesting as hematuria, and in non-pregnant state as cyclic hematuria and amenorrhoea.

Case Report
A 29 year female attended our gynaecology outpatient department with 18 week pregnancy with previous 3 CS with hematuria. 1st CS was in 2007 for obstructed labour, 2nd was in 2010 and 3rd was in 2014 for previous two CSs. After her last CS she developed hematuria at the expected time of resumption of menses.
The hematuria was with pain but no incontinence and no bleeding per vagina Later she developed hematuria of 3-4 days every cycle with cramping pain in lower abdomen.
For this she went to an urologist, cystoscopy was done, but was inconclusive and the procedure was very painful for the patient.
She continued to have hematuria till she became pregnant again in 2017.During her pregnancy at 4th month of gestation she again complained of hematuria. She consulted our clinic for the same.
An ultrasound was performed showing 18 weeks live pregnancy with fundoposterior placenta and an anechoic fluid containing sac communicating with the bladder on the left side of uterus.

Provisional diagnosis of VUF was made
Cystoscopy was advised again to find out the lesion. She refused because of previous experience. Therefore, MRI was done.

MRI showed linear tract extending from the posterosuperior part of urinary bladder to the lower uterine cavity at left parasagittal region. Surgery was planned. After counseling cystoscopy was preformed under IV anaesthesia.

Firstly the cystoscopy appeared normal except a few tags of blood but while withdrawing the scope, on reduced pressure a glistening white buldge appeared on the trigon. This was a 1x2cm fistula at the posterosuperior wall of bladder containing membranes communicating with the uterus. The membrane disappeard on distension of the bladder and reappeared on evacuating the bladder, but no leak of saline per vagina was seen. These blood tags manifested as hematuria.

Thus this patient had menouria for 4 years in the form of hematuria during mensturation. Her amenorrhoea was also false as bleeding was through the urethra and not through the vagina.

Hemorrhage also manifested as hematuria.

MTP formalities were done and she was taken for laparotomy with hysterotomy and bladder repair. Infraumbilical midline vertical incision was taken. Suprapubic access was made for extraperitoneal dissection of the reteropubic space to dissect the bladder then sagittal cystotomy was done until fistula was reached. Posteriorly pubocervicovaginal fascia was dissected beyond the uterus cervix and vagina.

The rent in the uterus was extended vertically for hysterotomy.

Uterus was closed in single layer with vicryl number 1. Followed by two layer closure of bladder after tissue transposition with vicryl no 2-0 in a single running, locking manner. (O'Connor's technique) Tubal ligation was done by modified Pomeroy's method. An omental flap was utilized between the bladder and uterus. A drain was placed in the uterovesicle gap and abdomen was closed.

Drain was taken out on 3rd post-operative day, and bladder catheter after 3 weeks. No complication in the form for urinary incontinence and hematuria was noticed.

Discussion

The World Health Organization (WHO) reports that 130000 new urogenital fistula developed each year as a result of difficult deliveries. However, the real incidence maybe higher as many women do no seek treatment in developing countries.8

In earlier days main cause of VUF used to be difficult deliveries, obstructed labour, instrumental deliveries, placenta percreta with bladder invasion but now the major cause is related to surgery, usually CS due to inadequate mobilization of bladder or accidental inclusion of bladder in the suture while closing the uterine incision.

Jozwik and Jozwik (2002) classified VUF based on the location of the fistula on the uterus and the route of menstrual flow.9

In pre-isthmic fistula (type I) there will be only menouria with absent menstrual flow and no urinary incontinence.

If fistula is at Isthmus (type II) there will be combination of menouria, menstrual flow and incontinence.

In post isthmic fistula (type III) there will be no menouria instead there will be regular menstrual flow and urinary incontinence The type I VUF feature was first described a century ago. In 1957, Youssef described a syndrome characterized by cyclic hematuria (menouria), apparent amenorrhoea without vaginal leak of urine after a CS.6

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

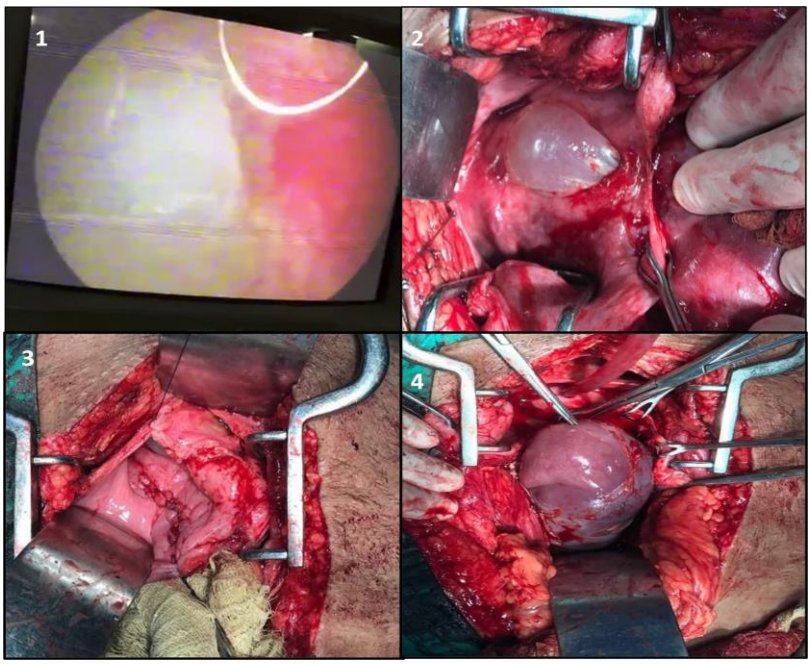

Fig 1: 1. Cystoscopy showing bulging of sac on reduced pressure.
2. Rent in bladder in connection with uterus seen after opening the urinary bladder
3. Urinary bladder rent sutured.
4. Product of conception delivered through uterus.

Cystoscopy helps in identifying the fistula, determining the size and its location in relation to the trigone and ureteric orifices. Other modalities for diagnosis are intravenous urography, hysterosalpingogram, saline infusion sonohysterography, ultrasound, and contast helical CT. Cystography maybe incompetent for the diagnosis as intera uterine pressure is higher than the intra vesicle pressure.

All the methods of diagnosis have some limitations but diagnosis with MRI can be made with 100% accuracy. It has the advantage of being non-invasive, does not contain ionizing radiation and regarded as the gold standard for the diagnosis and planning of the treatment [10].

In our case the pelvic ultrasound gave a clue. The diagnosis was confirmed by MRI, and surgical plan was made after

cystoscopy. Under the pressure of the circumstances we had to terminate the pregnancy to save the mothers life and to avoid grave injury to urinary bladder as the rent in the caesarean scar and the protruding amniotic sac into the bladder could have given way at point of time.

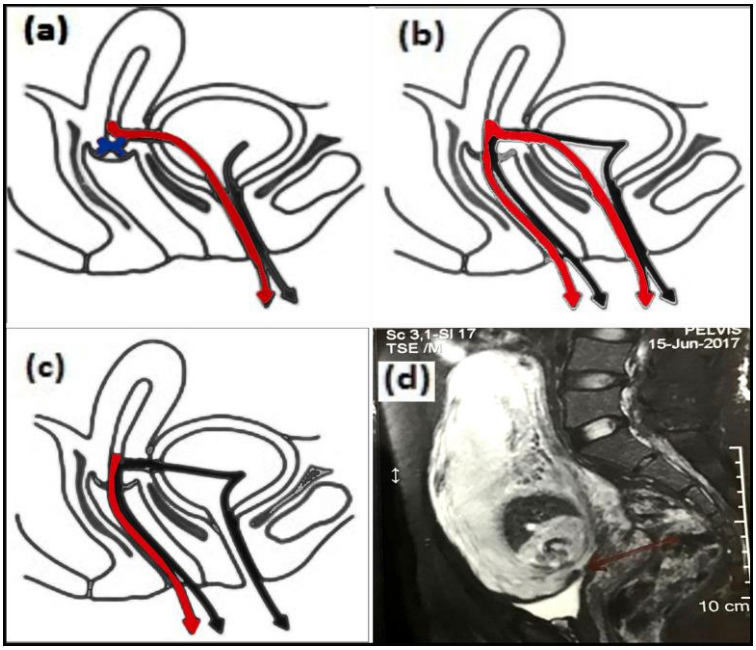

Fig 2: Jozwik and Jozwik (2002) classified VUF based on the location of the fistula on the uterus and the route of menstrual flow.
a) In pre-isthmic fistula (type I) there will be only menouria with absent menstrual flow and no urinary incontinence.
b) If fistula is at Isthmus (type II) there will be combination of menouria, menstrual flow and incontinence.
c) In post isthmic fistula (type III) there will be no menouria instead there will be regular menstrual flow and urinary incontinence.
d) MRI depicting the communication between bladder and uterus.

Conclusion

This patient had menouria for 4 years in the form of hematuria during mensturation. Her amenorrhoea was also false as bleeding was through the urethra and not through the vagina. Ante partum hemorrhage also manifested as hematuria.

In this case the pelvic ultrasound gave a clue. The diagnosis was confirmed by MRI. And surgical plan was made after cystoscopy Hysterotomy with tubal ligation and bladder repair thus saving the uterus and helped her regain normal vaginal menses.

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