

Case Report

SPONTANEOUS CLOSURE OF VESICOUTERINE FISTULA: A CASE REPORT

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ABSTRACT:

Vesicouterine fistula is a rare variety of urogenital fistula, representing about 1-4% of all cases, and caesarean section is the most common cause. The clinical presentation is variable and mainly includes a triad of cyclical hematuria (memoria), amenorrhoea, and urinary continence termed as Youssef's syndrome. The mainstay of treatment is surgical excision, and only less than 5% of patients respond to conservative therapy. Laparoscopic and robotic-assisted surgery is also gaining popularity. The case reported here was a young multiparous woman with previous four lower segment caesarean sections and had her elective fifth caesarean section and bilateral tubal ligation. The bladder was high up and badly adherent to the previous scar. She developed hematuria in the immediate postoperative period. Retrograde cystography confirmed the diagnosis of small vesicouterine fistula. The Patient responded to conservative management by catheterization for six weeks. Healing of fistula confirmed by repeat cystogram, and the Patient is having normal menstruation with no urinary problem.

Key Words: Uterus, Urinary bladder, Fistula

INTRODUCTION:

Vesicouterine fistula is a rare variety of urogenital fistula representing 1-4% of all urogenital fistulas.¹ Vesicouterine fistula is a pathological communication between epithelial surfaces of bladder and uterus or cervix.²⁻⁴ Only a few cases are reported in the literature, and most common cause is caesarean section.⁵⁻⁶ Almost 83-88% of cases of vesicouterine fistula are due to caesarean section.^{3, 7} The incidence is almost double after repeat caesarean section.⁷

Causes of vesicouterine fistula are mainly divided into four categories. Surgical, obstetrical, radiation necrosis and malignancy.⁸ Obstetrical causes include caesarean section, obstructed labor, dilatation and curettage, induced abortion, forceps delivery, placenta percent, and vaginal birth after caesarean section.

The classical clinical presentation is termed as Youssef's syndrome as first described by Youssef in 1957.⁹ It included triad of cyclical hematuria (menouria), absence of vaginal bleeding (amenorrhoea), and complete urinary continence. The first case of the vesicouterine fistula was described by Knipe in 1908.¹⁰ In 2000, Jozwik and Jozwik proposed another classification of Vesicouterine fistula (VUF), based on the route of menstrual flow.¹¹ According to Jozwik, there are three types of Vesicouterine fistula. Type 1 is a triad of amenorrhoea and menouria and complete continence of urine (Youssef's syndrome). Type 2 is a dual menstrual flow via both bladder and vagina. Type 3 is normal vaginal menses and a lack of menouria. In some patients, the only presentation of Vesicouterine fistula is urinary incontinence.¹²

Diagnostic modalities include Cystoscopy, Cystography, and Hystrogram.⁸ Treatment modalities include conservative, medical, and surgical management.

Here we report a case of the iatrogenic vesicouterine fistula, which healed spontaneously by bladder catheterization.

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CASE SUMMARY:

My Patient, 27 years old, G6P4A1, previous four caesarean sections, and three alive issues. The last section was done two years back. Her expected date of delivery was Aug 22, 2019. Her pregnancy was uneventful. She was admitted on Jul 24, 2019, at 35 weeks and four days for elective lower segment caesarean section. During surgery, it was noticed that the bladder was high up and badly adherent to the previous scar. Dissection of bladder tried, but bleeding started so bleeding points diathermic and incision given on lower segment of uterus 2cm above bladder adhesion. The outcome was a 2.8 kg baby boy with a good Appearance, Pulse, Grimace, Activity and Respiration (APGAR) score. Uterus was closed in two layers, bilateral tubal ligation was done, and the abdomen was closed. The Patient had hematuria in the immediate postoperative period, so it was decided to retain a foleys catheter for seven days. Her hematuria got settled in 48 hours with smooth recovery, so the foleys catheter was removed on the 7th postoperative day (Aug 1, 2019). She again started hematuria after removing the foleys catheter, and there was no vaginal bleeding. The urologist was consulted, and keeping in mind the possibility of vesicouterine fistula; the Foleys catheter was inserted on the same day for six weeks. Her baselines investigations were repeated, and abdominopelvic ultrasound was done to rule out any collection, and broad-spectrum antibiotics were started. To confirm the diagnosis of vesicouterine fistula, a retrograde cystogram was done, and it showed small vesicouterine fistula. (fig 1) Her hematuria got settled after two days, and stitches were removed on the 10th postop day with a healthy wound. She was discharged with Silicon Foley's catheter 18FR, oral analgesics, and antibiotics. She suffered from amenorrhoea after her caesarean section. She presented with urinary tract infection twice afterward, and antibiotics were given after culture and sensitivity reports. Her Foley catheter was

changed after two weeks and removed on Sep 16, 2019. She remained asymptomatic after that with no hematuria. As she was suffering from amenorrhoea, so progesterone was given for withdrawal bleeding to rule out menouria. She had vaginal bleeding with no menouria. She had regular cyclical bleeding after that. Her retrograde cystogram was repeated on Jan 14, 2020, and it confirmed the spontaneous healing of the fistula. (Figure 2)



Anteroposterior view Lateral view

Fig-1: Retrograde cystogram showing spillage of dye from bladder into uterus (Vesicouterine fistula)



AP view Lateral view

Fig-2: Retrograde Cystogram showing healed Vesicouterine fistula

DISCUSSION:

Vesicouterine fistula is a rare variety of urogenital fistula. In developing countries, it can occur after previous caesarean section, prolong and obstructed labor, forceps

delivery, uterine rupture, manual removal of placenta, abnormal implantation of placenta, inflammatory bowel disease and pelvic radiations.^{8, 13} In developed countries lower segment caesarean section is the main iatrogenic cause of this condition.

Clinical presentation is variable, including the clinical triad of Youssef's syndrome (menouria, amenorrhoe and urinary continence).⁹ Patient can also present with recurrent urinary tract infections and some with urinary incontinence.²

Diagnostic modalities include cystoscopy, cystography, hysterosalpingography, contrast-enhanced CT Scan, Magnetic Resonance Imaging (MRI) and Transvaginal ultrasound.¹²⁻¹⁴

Management options include conservative, medical, and surgical. If the fistula is small and diagnosed early, then bladder catheterization for 4-6 weeks helps in spontaneous healing of fistula, and this conservative management was successful in our case study. The overall success rate of conservative management is less than 5%.¹⁵ In some patients, such lesion present with lochiuria in puerperium and resolve spontaneously before diagnosis.^{16,17}

In 1999, Jozwik and Jozwik reported a case of spontaneous closure of the vesicouterine fistula. This was 41st Patient with spontaneous closure. He reviewed literature and found out that the total 796 cases reported until December 1997. Out of these, he identified 40 cases of spontaneous resolution of Vesicouterine fistula making success rate of less than 5%.¹⁸

Medical management includes the use of oral contraceptive pills, Progestogens, and GnRH analogs for induction of amenorrhoea, and there are case reports of a successful outcome with medical management.^{19, 20}

Surgical management is the treatment of choice in most of the cases. Various surgical approaches include vaginal, transvesical, transperitoneal, laparoscopic, and robotic procedures.²¹ It is recommended to delay fistula repair surgery for three months after iatrogenic surgery. It will help in

spontaneous closure of small fistulas and in the resolution of inflammation before surgical repair of large fistulas. Although there are case reports of a successful outcome with early surgical repair of Vesicouterine fistula.²²

Many case series and case reports are published in literature with satisfactory results of surgical repair of Vesicouterine fistula. In 2013, Rajamaheswai N et al. reported a 100% success rate of treatment of Vesicouterine fistula in 17 patients.¹⁴ Rao MP and colleagues reported excellent surgical outcomes in 12 patients.²³ Transvaginal repair of Vesicouterine fistula is less preferred route because of the higher location of the fistula. Recently Milani R et al managed a case of Vesicouterine fistula successfully with transvaginal repair and stressed that this route should be used by only experienced surgeons.¹³

Laparoscopic and robotic repair of Vesicouterine fistula is gaining popularity due to reduced blood loss, less postoperative pain, and shorter hospital stay. In 2013, Abdel Karim et al. reported the largest series of laparoscopic Vesicouterine fistula repair.²⁴ He managed 11 females with no complications. In 2009, Hemal AK et al. reported the first successful case series of robotic repair of Vesicouterine fistula in 3 cases.²⁵ Overall pregnancy rate after Vesicouterine fistula surgical repair is from 25% to 37.5%.^{15,23} In our case bilateral tubal ligation of the Patient was done.

CONCLUSION:

The incidence of caesarean sections is increasing worldwide, so doctors need to be aware of this possible complication, especially in cases of repeated caesarean sections. Patients should be informed about the risk of Vesicouterine fistula before repeat caesarean section, and informed consent should be taken. Repeat caesarean section should be performed by a senior obstetrician. Women with genitourinary fistulas lead a miserable life. Not only they have personal hygiene problems but are shunned by family and society. Women do

not come forward for treatment and continue to suffer in silence. Hence, there is a need to create awareness not only among masses but also in the the medical community.

AUTHOR'S CONTRIBUTION:

FF: Conception of idea
DN: Data collection
YA: Data analysis
SJR: Editing

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