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Vesicouterine Fistulae: Incidence, Symptoms, Outcome: An Institutional Study from Tertiary Care Hospital

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Abstract: ***Aim: A study of vesicouterine fistulae managed from 2020 to 2022, analysed the incidence, symptomatology, diagnosis and surgical outcome. Patients and methods: During the study period, 5 patients were managed, all underwent abdominal repair. Mean age of this study is 30.6. Results: Vesicouterine fistulae resulted following cesarean section in 4 patients and vaginal delivery in one patient. All patients had successful outcomes. Conclusion: The majority of vesicouterine fistulae occur following caesarean section, and it is feasible to achieve 100% successful repair.***

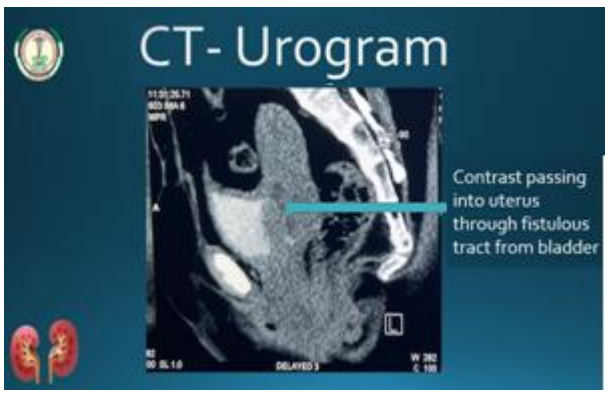
Keywords: Vesicouterine fistulae, Youssef syndrome, Menouria, Abdominal repair

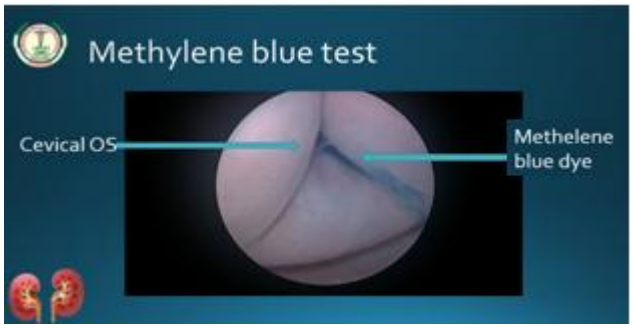
1. Introduction

A vesicouterine fistula is an abnormal pathway between the bladder and the uterus. The first case was reported by Knipe and colleagues in 1908. Vesicouterine fistula is the least common of all the urogenital fistulas, representing 1– 4% of all cases [1]. The vast majority of vesicouterine fistulae are secondary to iatrogenic causes, the most common being lower segment Cesarean section [2]. The less frequent causes include induced abortion, dilatation and curettage, vaginal birth after previous Cesarean section, obstructed labor, forceps delivery, placenta percreta, migrated intrauterine contraceptive device, and brachytherapy [2]. Youssef's syndrome classically presents as a triad of cyclical hematuria, amenorrhea and urinary continence, that is, there is novaginal leakage of urine [3]. Vesicouterine fistula should be suspected in cases presenting with urinary incontinence even years after Cesarean section. Hysterography and cystography are commonly used to diagnose this condition. The majority of VUF require definitive surgical repair, as only 5 % of patients respond to conservative management [4]. This article describes our experience with VUF in the last 30 months and presents a review of the literature.

Patients and methods

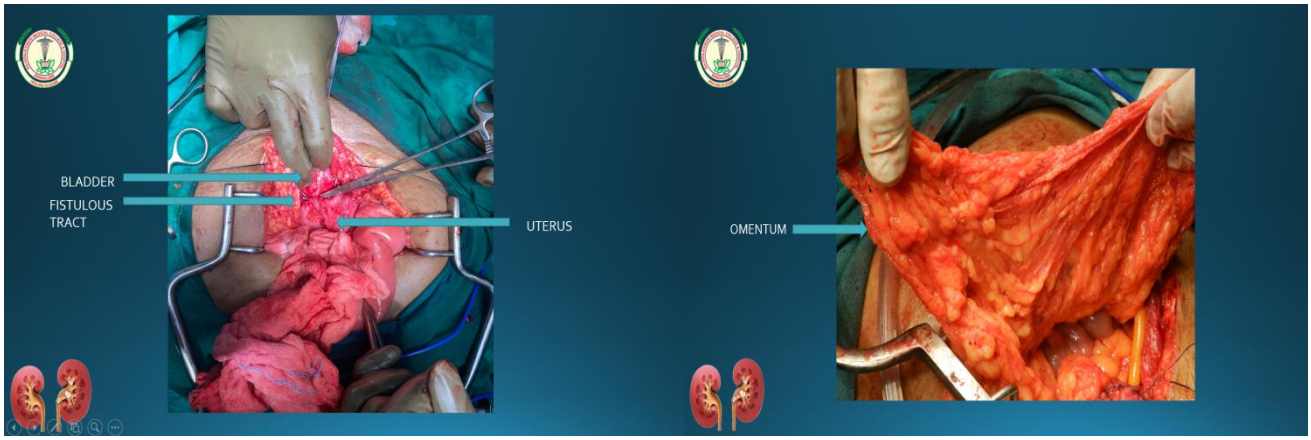
We performed a retrospective study of VUF managed from 2020 to 2022 and analyzed incidence, symptoms, and management after obtaining written and informed consent from patients. There were 5 vesicouterine fistulae managed during the study period. Patientmean age at the time of presentation was 30.6 (23–40) years. Patient data was reviewed to study the etiology, presenting features, surgical details, and outcome of fistula repair. All the patients were evaluated by detailed history and physical examination. Intravenous urography was done for all patients to rule out other urogynecological fistula. All patients were evaluated by cystoscopy and examination under anesthesia, including use of the methylene blue test to confirm VUF. All patients underwent surgical management by abdominal approach. All repairs were done by the same surgeon. Abdominal repair was done by the O'Connor method. The bladder was bivalved up to the fistula enabling mobilization of the bladder end of the fistulous tract from the uterus. Closure of the uterine rent was followed by bladder reconstruction in two layers. An omental flap was interposed between the bladder and uterus.





2. Results

There were 5 vesicouterine fistulae managed during the study period. Of these 5 fistulae, 4 followed cesarean section, 1 following vaginal delivery. One patient presented following the first cesarean section, three after their second, and one after vaginal delivery. Of the 4 cesarean sections, two were emergency sections for obstructed labor and the remaining two were elective. Among the one patient who developed fistula after forceps assisted vaginal delivery due to prolong and difficult delivery. All the patients presented with features of classical Youssef syndrome (menouria, amenorrhea, and urinary continence). Cystoscopy identified the supratrigonal location of the fistula in all patients, and methylene blue test was positive in all 5 patients. CT urogram is done in all cases which showed persistent fistulous tract between uterus and bladder, contrast extravasation seen into the uterus. All cases were managed surgically, 4 patients presented later and one who presented earlier had undergone cesarean section for obstructed labor. Abdominal repair was done in all 5 patients. Hysterectomy was combined with repair in one patients, whois postmenopausal. Omental Flap interposition was done in all cases. During a follow - up of 1 year 4 months all patients were relieved of their symptoms.



3. Discussion

Uterovesical fistula, a rare type of urogenital fistula was first reported in 1908. In 1957, Youssef described a syndrome comprising of cyclic hematuria, amenorrhea, menouria, and complete urinary incontinence in a patient who had lower segment Cesarean section (LSCS) [5]. The VUFs are among the least common urogynecological fistulas. The VUF also occurs following high vaginal forceps - aided delivery, external cephalic version, curettage or manual removal of the placenta, placenta percreta, myomectomy, uterine rupture due to obstructed labor, uterine artery embolization, perforation of an intrauterine device, and brachytherapy for carcinoma of cervix. The LSCS is the single most common cause of VUF [6]. Amenorrhea, cyclic hematuria without urinary incontinence in combination with a history of LSCS, has been described as pathognomonic of VUF [2]. A classification of VUF based on the routes of menstrual flow has been proposed by Jo´zwik and Jo´zwik [7] that divides VUF into three types. Type I, characterized by the triad of amenorrhea, menouria and complete continence of urine has been known as Youssef's syndrome. Type II is associated with dual menstrual flow via both the bladder and vagina. Type III is associated with normal vaginal menses and lack of menouria [7].

As with vesicovaginal fistulae, immediate presentation occurs when there is direct injury to the bladder during surgery [8]. Delayed presentation can occur when there is an ischemic necrosis and progressive devitalization of the posterior wall of the bladder [9, 10]. The disease may manifest with cyclical hematuria or urinary leakage or both. Urinary incontinence appears in the early postoperative period and may be associated with low - grade pyrexia or urinary sepsis or be completely asymptomatic [8]. Menouria with apparent amenorrhea may present late, usually a few weeks postpartum, when menstruation is restored [11]. Youssef hypothesized that the sphincteric action of the cervical isthmus prevents leakage of urine and menses through the internal os [5]. It has been postulated [12] that the pressure difference between the uterine cavity and the bladder cavity governs the flow of urine or menstrual blood through the fistula. Normally, the intrauterine pressure is higher than that of the bladder most of the time, except occasionally during micturition, which can account for the sole menouria and intermittent urinary incontinence in many patients with VUF. The uterine endometrium may occlude the fistulous opening and play a role in the continence mechanism [13]. Urinary incontinence without any relationship to voiding or the filling phase occurs if the VUF level is at or below the internal os or if the os is incompetent [5, 12]. VUF should be suspected when there is macroscopic hematuria and/or urinary leakage early in the postoperative

period of cesarean section or other gynecological procedures [14]. Methylene blue dye test reveals the leakage of blue through the cervical os and identifies VUF; however, this test does not delineate or document the course of the fistulous tract, and it can be negative in the presence of long and tortuous tract [6, 15].

The diagnosis of VUF is often confirmed by imaging studies and cystoscopy. Cystoscopy, even when repeated can fail to confirm the fistula [5]. Methylene blue instilled into the uterine cavity or through the urethra or through catheterization of a visible lesion in the bladder wall can confirm the fistula. This test, however, does not show directly the fistulous tract and its specific location. Moreover, this test can be negative in patients with a long and tortuous tract [5]. In radiological studies, both cystography and hysterography have been used in the diagnosis of VUFs. In Tancer's review of published reports, he found that hysterography was the most reliable diagnostic technique [2]. Intravenous urography can show the fistula when contrast medium enters the vagina, but distinguishing vesico - vaginal and vesico - uterine fistulae is difficult. Although VUFs are difficult to diagnose using USG, Park et al. reported that sonography can demonstrate the fistulous tract as double echogenic lines between the endometrium of the anterior wall of the uterine body and the mucosal layer of the posterior wall of the bladder [6]. However, sonography has inherent difficulty in differentiating the VUF tract from different patterns of a noncomplicated Cesarean scar [16]. Also transvaginal sonography is recently being used for diagnosis [17]. Magnetic resonance imaging has now become the first choice in the investigation of fistulas [18, 19].

Management:
Conservative management with continuous bladder drainage for 4 - 8 weeks along with antibiotics and anticholinergics is recommended in cases where a small fistula is detected in the early or immediate postpartum or postoperative phase. The success rate of conservative management is less than 5% [9]. Trans vesical fulguration with continuous catheterization and hormonal treatment can be successful for the treatment of VUF with a small, well epithelialized orifice, though the results in the presence of extensive scarring are poor and recurrence is high [20]. Oral contraceptives, progestational agents, and gonadotropin releasing hormone analogs have been used to induce amenorrhea [21]. Though the ideal duration of hormonal treatment is unknown, the majority of authors specify a period of 6 months [14]. This management technique is less likely to be successful in women with a mature fistulous tract (≥6 weeks after the inciting event) [22].

Cystoscopic fulguration of the VUF has also been reported for managing small fistulae [22, 23, 24]. It is a simple procedure with low postoperative morbidity rates [24]; however, it is seldom successful. Most of the fistulae warrant surgical closure [13, 25].

Surgery is the definitive method of treatment. It can be performed trans abdominally, endoscopically, and robotically. The transvaginal approach is not preferred because of the higher location and complexity of the fistulas. Surgical repair of VUF is performed more frequently by the abdominal approach [12 - 14, 26]. An extraperitonealtransvesical approach appears to be advantageous, as it avoids peritoneal contamination and enables direct access to the mouth of the fistula [12, 27, 28]. However, it is almost impossible to complete the repair without entering the peritoneal cavity. Moreover, the transperitoneal route is effective, as it allows satisfactory mobilization of both bladder and uterus [12, 14, 29]. In our series, all abdominal repairs were done by the transperitoneal route, and the 100 % successful outcome rate is heartening. The presence of uterine pathology warrants hysterectomy [28], which is acceptable in multiparous women of perimenopausal age. Even in its absence, fistula surgery when combined with hysterectomy permits surgical space for thorough repair and reconstruction.

In our study, fistula focus between the bladder and lower segment of uterus was found using O'Connor technique through extraperitoneal abdominal incision and mucosa of uterus and bladder were repaired using long - term absorbable suture materials. Extraperitoneal drainage catheter placed in abdomen was removed after 5 days and urethral Foley catheter was removed after 21 days following operation. No urine leakage was detected and spontaneous urination was normal.

Recently successful laparoscopic repair of VUF has been described. The advantages of the laparoscopic technique are quicker convalescence, shorter hospital stay, better cosmetic results, and success rates similar to open abdominal surgery [30 - 33].

In the majority of patients, surgical outcome is favorable, resulting in resolution of symptoms after initial surgery. In the largest case series, reported by Benchekroun et al., restoration of urinary continence was observed in 80 % of patients. Of 30 patients managed over a period of 25 years, 20 % resulted in residual fistula [34]. Our study reported here attained 100 % successful outcome for VUF, which is reassuring to the patient and the surgeon. All abdominal repairs were done using the transperitoneal transvesical technique, whereas Benchekroun et al. repaired eight cases using the extraperitonealtransvesical technique, a factor that might have accounted for our higher success rate, as the technique we used allows satisfactory mobilization of bladder and uterus [11, 29].

VUF are associated with infertility and first - trimester spontaneous abortions [35]. Fertility potential is considered poor, even after reconstruction [36]. However, Lotocki et al. reported a pregnancy rate of 31.25 % after VUF repair, with a term delivery rate of 25 % [37].

4. Conclusion

The majority of VUF are associated with previous lower segment cesarean section, and it is feasible to achieve 100 % successful outcome with surgical repair of the VUF.

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