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CASE REPORT

Intravesical Ectopic Pregnancy in Vesicouterine Fistula Patient: A Very Rare Case Report and Literature Review

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Abstract: Vesicouterine fistula (VUF) is a rare form of urogenital fistula, accounting for approximately 4% of cases. It commonly arises as a complication of uterine interventions, especially previous cesarean sections (C-sections). Intravesical ectopic pregnancy complications are infrequent. We discuss a 39-year-old female with severe lower abdominal pain spreading to the flank, dysuria, hematuria, and urinary hesitancy one week after laparotomy for fetal demise at 17–18 weeks of gestation. A dead fetus in the urinary bladder along with the VUF was revealed in the imaging. The surgical operation identified a fistula between the posterior bladder wall and the lower uterine segment. Fetus evacuation and fistula repair were performed with symptom resolution achieved. A literature review was conducted and nine articles were extracted. These articles discussed the risk factors and complications of VUF. The articles, selected for relevance and quality, highlighted cesarean sections as the most common risk factor, with hematuria being the most frequent complication. Only a small number of cases had fetal migration into the bladder documented, showcasing its rarity. This case represents a rare complication of VUF: intravesical ectopic pregnancy into the bladder. Surgical intervention led to favorable outcomes and is consistent with findings in the reviewed literature. The rare complication of VUF such as intravesical ectopic pregnancy requires proper diagnosis and treatment. This case emphasizes the importance of surgical management in achieving successful outcomes for VUF and its complications.

Keywords: vesicouterine fistula, intravesical ectopic pregnancy, cystography

Introduction

Vesicouterine fistula (VUF) refers to a defect resulting in communication between the uterus and the bladder. VUF constitutes a relatively small proportion of all urogenital fistula cases, accounting for only approximately 4%.[1,2] The majority of VUF cases result from iatrogenic injuries during medical procedures such as cesarean sections, the use of vaginal forceps, cervical dilatation, or uterine curettage for abortion.[3] Additional etiologies include uterine artery embolization, vaginal delivery after a prior cesarean section, intrauterine device migration, placenta percreta, and endometriosis.[4–6]

The primary clinical manifestations of VUF include urinary incontinence, cyclic hematuria (also referred to as menouria), amenorrhea, and recurrent urinary tract infections. These symptoms frequently present in a delayed manner, occurring weeks to years following the precipitating event.[7]

While pregnancy in the context of VUF has been documented, the occurrence of fetal movement through the fistula tract is exceptionally rare.[3,8] Here, we present a case of spontaneous migration of a 17–18-week gestational fetus from the uterine cavity into the bladder through a VUF.

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Case Report

A 39-year-old female was admitted to our hospital with complaints of severe lower abdominal pain radiating to the flank, as well as difficulty urinating, dysuria, and hematuria one week ago. Abdominal pain was felt after the patient underwent laparotomy surgery at the referring hospital.

The patient underwent a laparotomy because she was in her fourth pregnancy with a gestational age of 17–18, an ultrasound examination revealed the fetus had died (Figure 1) and needed a curettage. After curettage was performed at the referring hospital, perforation occurred and it was decided to laparotomy. During the operation, there was no intrauterine fetus but a urinary bladder leak was found. The urinary bladder was sutured. One day after surgery, the patient complained of hematuria again. Ultrasound and X-ray were re-examined, and the dead fetus was found in the urinary bladder. Then the patient was referred to us further treatment.

The patient had undergone C-sections in 2006 and 2012. She did not complain of any problems after said surgeries. She had a history of miscarriage in 2020 at 4–5 weeks of gestation, but no curettage was performed.

Through physical examination, she was stated as hemodynamically stable Her abdomen was slightly distended and tender, the surgical wound was covered with bandages, and there was no blood leakage. Vaginal examination within normal limits.

A follow-up examination was carried out. Ultrasound examination revealed the fetus in the lumen of the vesica urinaria (Figure 2). The patient also underwent cystography, which discovered a dead fetus in the lumen of the vesica

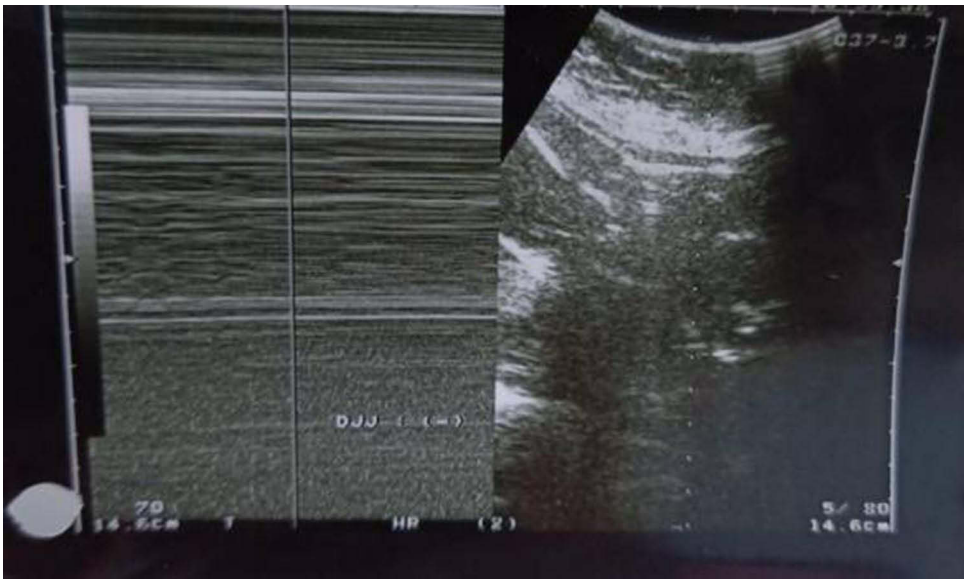

Figure 1 Ultrasound examination found that the fetal heartbeat was absent.

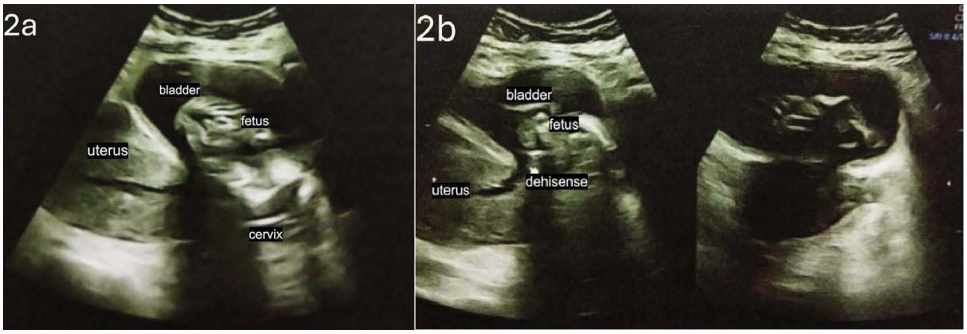

Figure 2 Ultrasound revealed fetus in the lumen of the vesica urinaria. (**a**) trigonum of the vesica urine; (**b**) dehiscence and the fetus.

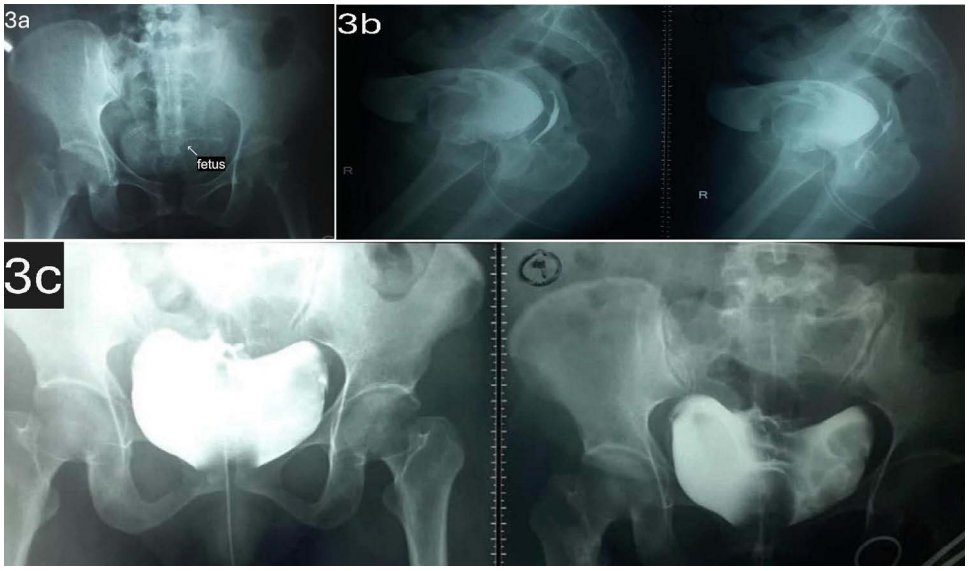

Figure 3 Cystography revealed a dead fetus in the vesica urinaria and the cranial vesica surface of the urinary bladder appears. (**a**) Plain pelvic X-ray; (**b**) lateral view with contrast; and (**c**) anteroposterior view with contrast.

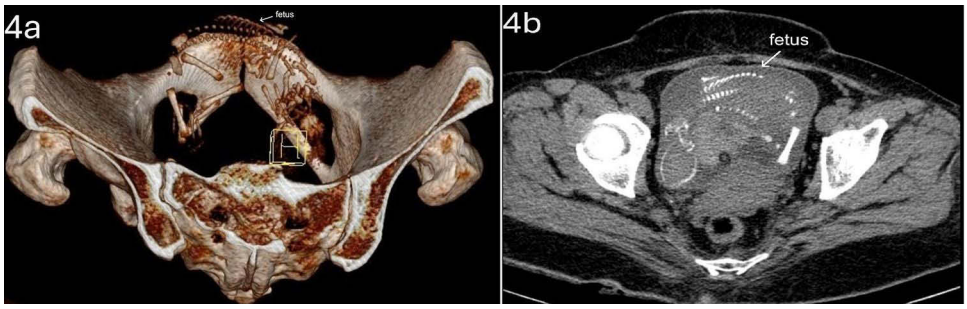

Figure 4 Abdominal CT scan showed that there was a fetal skeleton in the bladder. (**a**) 3D CT scan imaging of the pelvic region; (**b**) Axial CT scan of the pelvic region.

urinaria and the cranial vesica surface of the urinary bladder appears irregular (Figure 3). An abdominal CT scan showed that there was a fetal skeleton in the vesica urinaria (Figure 4). The vesicouterine fistula was evident. A diagnosis of fetal death intravesical and vesicouterine fistula was made for the patient. Management in this patient is bladder repair, evacuation of the fetus, and repair of vesicouterine fistula.

In intraoperative urethrocystoscopy findings, a macerated fetus and its structure were found, the ureter opening was identified as normal, and it was decided to explore the bladder. In the exploration of the bladder procedure, the bladder with normal shape and size was found, the structure of the fetus was destroyed, and the evacuation of the fetus was successful. Uterovesical fistula found on the posterior wall of the bladder. The fistula repair procedure was successful, then 2 layers of the bladder wall were sutured. A dower catheter (size 22) was inserted and the operation was continued by an obstetrician.

The postoperative course was unremarkable. After the urinary catheter was withdrawn in two weeks, her symptoms completely resolved and her postoperative follow-up was uneventful.

Methods

A thorough search of Google Scholar was performed using the terms “vesico-uterine fistula”, “fetal death intravesical”, and “case report”. The initial search revealed 470 studies, which were then filtered based on relevance and abstract content to match the study’s aims. Articles published in English or Bahasa Indonesia that satisfied the inclusion criteria, which included discussions about intravesical fetal death and vesicouterine fistula, were screened and selected. Exclusion

criteria included cohort studies, literature reviews, unavailable articles, and publications in languages other than English or Bahasa Indonesia. Finally, nine cases were selected for consideration. Data was extracted from all selected studies, with an emphasis on author names, publication year, study design, patient age, underlying causes of the fistula, and related complications.

Results

Extraction of Articles

Based on the extraction of the article (Table 1) above, several risks of fistula formation were found, including a history of Caesarean section which is one of the most common risk factors. In addition, other obstetric procedures such as curettage and forceps-assisted delivery are also risk factors.

Complications that occur from the presence of a fistula include the movement of physiological contents from each compartment to the connected compartment. In one case, the fetus was found in the bladder, but this complication is very rare. The most common complication is characterized by the presence of hematuria.

Discussion

A vesicouterine fistula (VUF) is an abnormal connection between the posterior wall of the urinary bladder and the anterior wall of the uterus. The first case of vesicouterine fistula in the English literature was reported by Knipe in 1908.[1] VUF is a sporadic type of urogenital fistula, accounting for 1% to 4% of all cases. Fistula can occur immediately after C-section, in the late puerperium, or after repetitive surgeries.[2]

While performing a cesarean section the vesica urinaria may be damaged by direct damage, abnormal sutures, or inadequate downward mobilization. Delayed formation of a vesicouterine fistula may lead to devascularization, infection, hematoma, or compression of the urinary bladder. After a cesarean section, the bladder becomes closely supported by the uterus, making it vulnerable to significant tension during a subsequent vaginal delivery. The thinning of the lower segment during labor may result in vesicouterine fistula formation. Researchers reported that progressing devitalization and damage of the base of the bladder may result from repeat cesarean sections for injuring its vascular network.[1,2]

The fundus and posterior surface of the uterus appear normal but the vesicouterine fossa is obliterated by dense adhesions. Fistulous communication is usually reported between the posterior supratrigonal part of the bladder and the anterior lower segment of the uterus or, more rarely, the cervix. Both openings are surrounded by dense fibrosis. The defect is lined by a gray hemorrhagic tissue that communicates with the cervix and extends to the internal ostium which may be narrowed.[1,2]

In this case, the patient was complained of severe lower abdominal pain radiating to the flank, as well as difficulty urinating, dysuria, and hematuria. Abdominal pain was felt after the patient underwent laparotomy surgery because the

Table 1 Extraction of Articles

Author, Year	Design of Study	Age of Patient	Causal/Risk Factors of Fistula	Complication of Fistula
Armstrong et al (2019)[9]	Case report	38 y.o	History of caesarean section 20 years earlier	Suspected fetus in urinary bladder
Sun et al (2018)[10]	Case report	31 y.o	Recent cesarean section	
Ogunlaja et al (2020)[11]	Case report	36 y.o	Cesarean section	Hematuria and infertility
Caraman et al (2022)[12]	Case report + literature review	28 y.o	Textiloma consisting of the suture stitches (cesarean section 7 years earlier)	Hematuria
Nouira et al (2005)[13]	Case report	68 y.o	Forceps delivery	Moderate urine leak
Guruvare et al (2004)[14]	Case report		Cesarean section 18 months earlier	Massive hematuria
Banale et al (2013)[3]	Case report	29 y.o	Curettage	Gross hematuria and abdominal pain
Gupta et al (2014)[15]	Case report	34 y.o	Cesarean section	Prolapse of amniotic sac
Junior et al (2018)[16]	Case report	38 y.o	Recent cesarean section	Heavy hematuria

Note: Data from these studies.[3,9–16]
Abbreviations: y.o, years old.

patient had a history of perforation after curettage with an ultrasound examination revealed the fetus had died. During operation, there was no intrauterine fetus but a urinary bladder leak was found. The urinary bladder was sutured. One day after surgery, the patient complained of hematuria again. She had a history of cesarean sections in 2006 and 2012, respectively. She had a history of miscarriage in 2020 at 4–5 weeks of gestation, but no curettage was performed. Through physical examination, her abdomen was slightly distended and tender, the surgical wound was covered with bandages. Ultrasound and X-ray were re-examined, and the dead fetus was found in the urinary bladder. Ultrasound examination revealed the fetus in the cavity of the urinary bladder. The patient also underwent cystography, which discovered a dead fetus in the cavity of the urinary bladder and the cranial vesica surface of the urinary bladder appears irregular. An abdominal CT scan showed that there was a fetal skeleton in the bladder. The vesicouterine fistula was also well visualized. The patient was diagnosed with Fetal Death Intravesical and Vesicouterine Fistula. This was in line with the explanation that clinical manifestations of a VUF are vaginal leakage of urine, cyclic hematuria (menouria), amenorrhea, infertility, and first-trimester abortion. Imaging investigations for detecting VUF include cystography, hysterography, intravenous pyelography, computed tomography (CT), and magnetic resonance imaging (MRI). Cystography usually shows, after filling the bladder with 200 cc 10% sterile iodide solution, an abnormal connection between the uterus and the posterior wall of the bladder.[7]

Numerous reports cover this subject.[1,3–5,7,8,17] Nevertheless, only four cases of fetal or embryonic migration into the lumen of vesica urinaria have been documented to date. Guruvare et al reported a patient with gross hematuria and thrombi in the bladder. The patient had a history of a cesarean delivery 18 months ago. Ultrasonography indicated the presence of a large mass within the urinary bladder, and the histological examination revealed a thrombus containing embryo remains.[14]

Management in this patient is bladder repair, evacuation of the fetus, and repair of vesicouterine fistula. A patient, as reported by Banale et al, had a history of two cesarean deliveries and presented with abdominal discomfort and gross hematuria one day before undergoing a termination of pregnancy at 17 gestation weeks. Ultrasonography indicated appearance of fetus inside the lumen of vesica urinaria. Injury to the posterior bladder wall and the anterior uterine wall was caused by cervical dilatation or curettage of the uterine cavity, enabling migration of the fetus into the vesica urinaria. The authors reported finding an inanimate fetus with a 20 mm fistula on the back wall of the vesica urinaria during cystoscopy. Bladder and uterus defects were repaired and removal of the fetus from the vesica urinaria was removed separately.[3]

Gupta et al published a case of severe abdominal tenderness and urinary retention. It was found that at 22 weeks of gestation, prolapse of the amniotic sac had happened through a VUF. The patient underwent two cesarean deliveries prior. The amniotic sac bulged through the urethra and its position was adjustable either manually or by positioning the patient in the Trendelenburg position. It was managed with a C-section and the vesicouterine fistula was sutured.[15]

A recent case in 2019 was also reported by Armstrong et al regarding a patient who was concerned about the excess clear vaginal discharge since 8 weeks of gestation. The patient had a history of C-sections 20 years ago. An ultrasonography examination was done, revealing an inhomogeneous echogenic mass in the vesica urinaria with the appearance of a fetal femur and cranium. An exploratory laparotomy was performed. Through the bladder, an intact 20-week-sized fetus was removed. The placenta was removed from the fistula site via the VUF.[9]

Lesovoy et al published a unique case report in which the VUF was formed more than two years before the pregnancy. In this case, the 17-week fetus migrated through the fistula tract.[8] This is like studies by Armstrong et al and Banale et al, which removed a 20-week and 17-week-sized fetus.[3,9] The patient had previously undergone two Cesarean deliveries. It is unique to some extent, as the symptoms remained unremarkable and no complications arose prior to 17 weeks of gestation. As the size of the fistula rose with the development of the uterus, the fetus was able to move into the bladder. Although the fetus's migration occurred precisely one day prior to the procedure, the morphologic abnormalities were consistent with the fetus's longer-term demise. Probably by the time of migration, the fetus had already died, and following its transfer, the fistulous tract showed a significant reduction.

Treatment options for the condition encompass both conservative management and surgical repair. Considering the possibility of the VUF resolving spontaneously, bladder catheterization is indicated when the fistula is discovered just

after delivery. It is used in conservative management for at least 4–8 weeks. Women with Youssef's syndrome is eligible for hormonal management. Surgical intervention serves as the definitive treatment for VUF following a C-section.[1]

Prospective studies evaluating vesicouterine fistula treatment and outcomes are currently unavailable due to the low incidence of the disease. Various surgical techniques have been employed to treat VUF. Chapple and Turner-Warwick proposed well-vascularized omental or peritoneal flaps between the bladder and uterus using vaginal, trans-vesica, and transperitoneal techniques. A comparable laparoscopic method for repairing bladder fistulas was also published by Das Mahapatra et al.[18] In a selected number of cases, Spruch et al performed hysterectomy to remove the uterus while repairing the bladder wall defect, with positive results.[19] Most cases are resolved with initial surgery, and surgical outcomes are positive. The post-repair pregnancy rate is 31.25% with a term deliveries rate at 25%.[1]

Conclusion

Pregnancy can still occur despite the presence of a long-term VUF. Migration of the fetus from the uterine cavity into the bladder is a problem that can happen to pregnant individuals with VUF.

Consent

Publication of this case has obtained patient consent and ethical clearance from Arifin Achmad Regional Hospital.

Disclosure

The authors report no conflicts of interest in this work.

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