

Fetal limb in bladder: A rare complication of vesicouterine fistula

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ABSTRACT

Vesicouterine fistula (VUF) is a rare urogenital fistula that is even rarer during pregnancy. Even if pregnancy occurs, the outcomes appear to be very poor. The most of the cases are related to iatrogenic bladder injury during cesarean section. There is very limited literature on the pregnancy with VUF associated with herniation of fetus or fetal part(s) into the bladder cavity. Here, we report a case of pregnancy in a known case of untreated VUF. She presented at 23 weeks of gestation with features of premature rupture of membrane and herniation of fetal left lower limb inside the bladder cavity. Her pregnancy ended up with hysterotomy and the removal of a non-viable fetus along with the repair of the fistula. Hence, regardless of the severity of the signs and symptoms associated with VUF, clinicians should convince the patients for the repair of the fistula especially if the future pregnancy is contemplated.

Key words: Bladder injury, Caesarean section, Pregnancy outcomes, Vesicouterine fistula

Vesicouterine fistula (VUF) is a rare anomaly accounting for 1–4% of all the genitourinary fistula [1]. The most of the cases are due to iatrogenic bladder injury that occurs during cesarean section [2]. Afflicted women may present with the classical triad of Youssef's syndrome-cyclical hematuria, amenorrhea, and urinary continence [3]. There is limited literature regarding the presence of VUF in pregnancy and its outcomes.

Here, we report a rare case of pregnancy with the presence of VUF which was associated with herniation of the amniotic sac containing one of the fetal lower limbs inside the urinary bladder. We also want to highlight the complications of pregnancy in untreated VUF and the importance of its timely management. We believe reporting our case will also help the clinicians to address some of the questions regarding the natural course of pregnancy in the presence of VUF.

CASE REPORT


A 33-year-old female in her third pregnancy presented at 23 weeks of gestation with features of premature rupture of membrane. She had a cesarean section for delivery of the second baby 3 years back. She also gave the history of diagnosis with VUF 2 years back during evaluation for occasional cyclic hematuria and deferring of the repair of the fistula. She was advised for medical termination of pregnancy at about 12 weeks of gestation by a local obstetrician whom she refused.

She presented to us at 23 weeks of gestation with lower abdominal pain and leaking per vagina. Ultrasonography was taken and revealed herniation of the amniotic sac containing a fetal lower limb inside the bladder through a defect in the posterior bladder wall with severe oligohydramnios as shown in Fig. 1a. Cystoscopic examination showed ballooning of the amniotic sac into the bladder cavity through a defect measuring about 4 cm × 3 cm located in the posterior wall of bladder about 4 cm away from trigone as shown in Fig. 1b.

Laparotomy was done using an infra-umbilical midline incision. With sharp dissection, the fistula site was identified and hysterotomy was done just above the site of the fistula (Fig. 2a). The herniated fetal left lower limb inside the bladder was retrieved back into the uterus and the non-viable fetus was removed (Fig. 2b). The fistula tract was excised; the uterine defect was joined to the hysterotomy wound making a single uterine wound and was closed in two layers with polyglactin 1-0 sutures. The bladder wall defect measuring about 4 cm × 3 cm was then closed in two layers using polyglactin 2-0 sutures followed by omental interposition. Foleys catheter was kept in place for 2 weeks and removed after confirmation of healing of the repair with cystogram.

DISCUSSION

VUF is a rare urogenital fistula. Although there are a few reports of pregnancy in the presence of VUF, herniation of the fetus or its

Access this article online	
Received - 20 December 2021 Initial Review - 06 January 2022 Accepted - 17 January 2022	Quick Response code 
DOI: 10.32677/ijcr.v8i1.3188	

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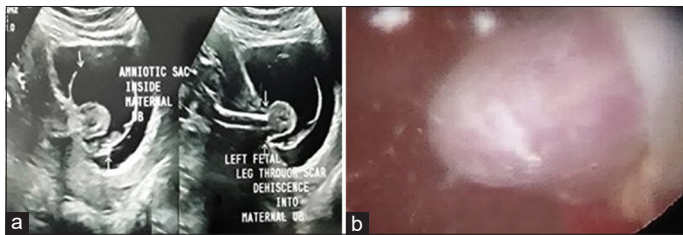


Figure 1: (a) Ultrasound images showing amniotic sac containing one fetal limb inside the bladder cavity. (b) Cystoscopic image showing ballooning of amniotic sac inside the bladder

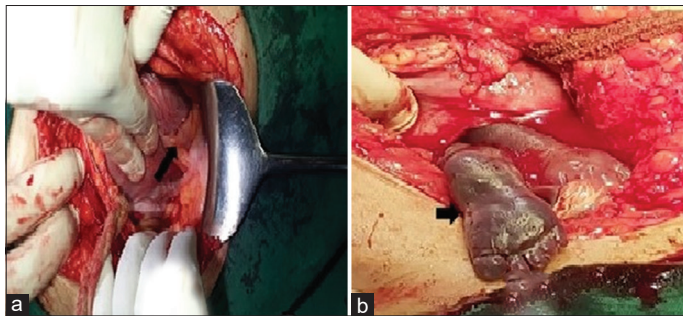


Figure 2: (a) Images showing location of vesicouterine fistula and (b) removal of fetal limbs by hysterotomy

parts into the bladder is very limited in the literature. The most of the cases are due to iatrogenic bladder injury during the cesarean section that occurs when a knuckle of the bladder is partially or completely incorporated into the suture during the closure of the lower uterine segment incision or during sharp or blunt dissection of the bladder away from the lower uterine segment. Hystero-graphy and cystoscopy remain the “gold standard” in the diagnosis [4].

The clinical presentation varies from a mere occasional cyclic hematuria as in our case to a spectrum of other symptoms such as amenorrhea, urinary incontinence, recurrent urinary tract infection, and secondary infertility. In our case, the patient had no bothersome symptoms and it was the reason for deferring the fistula repair by the patient. Apparently, part of the amniotic sac might have herniated into the bladder through the fistula tract in the early part of pregnancy. As the gravid uterus increases its size, the size of the fistula tract might also have increased allowing the fetal parts also to get herniated into the bladder cavity.

Management of VUF can be organ-preserving or hysterectomy. In our case, the patient wanted to preserve fertility so we did only VUF repair.

Similar to our case, Lesovoy *et al.* reported the case of a 39-year-old female with untreated long-standing VUF following cesarean section who presented with severe lower abdominal pain and difficulty in micturition. The further evaluation revealed an 11-week size fetus inside the bladder [5]. Another publication by Guruvare *et al.* reported the presence of an embryo inside the bladder in a patient who presented with gross hematuria [6]. Kives *et al.* also reported the case of a 28-year-old female in her second pregnancy who was diagnosed to have VUF with a ballooning

amniotic sac inside the bladder at 23 weeks of gestation. While waiting for the fistula repair, the patient had premature rupture of membrane and precipitous vaginal delivery at 25 weeks of gestation [7]. Sapre *et al.* describe a case in which a dead fetus was found inside the urinary bladder after migration through a VUF [8]. Another publication by Sandro *et al.* presented a report of the presence of VUF in a 36-years-old pregnant woman who presented to the emergency with complaints of pelvic pain and vaginal bleeding requiring surgical termination of pregnancy [9]. A similar case of vaginal bleeding in early pregnancy with VUF necessitating urgent surgical intervention was also reported by Gomez *et al.* [10].

Our overall observation is that pregnancies do occur even in the presence of VUF but with unfavorable outcomes. Had the surgical repair of fistulae be done in time, there could have been reduction in the prevalence of eventful pregnancies. Hence, regardless of the severity of the signs and symptoms associated with VUF, the patient should be convinced of VUF repair especially when pregnancies are contemplated in the future.

CONCLUSION

Pregnancy in the presence of VUF is rare and most of the time with unfavorable outcomes. Hence, clinicians should convince the patient for surgical repair of the VUF if the future pregnancy is contemplated.

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Funding: None; Conflicts of Interest: None Stated.

How to cite this article: Singh KA, Singh KT, Singh KS. Fetal limb in bladder: A rare complication of vesicouterine fistula. *Indian J Case Reports*. 2022;8(1):7-8.