

Uterus didelphys with double vagina diagnosed during third cesarean section: A case report

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Abstract

Uterus didelphys is a rare uterine anomaly. It is associated with fetal breech presentation, preterm delivery, and cesarean section. Longitudinal vaginal septum may be associated with uterus didelphys, which may cause dyspareunia and labor obstruction. We report a case of 28-year-old woman whose uterus didelphys and longitudinal vaginal septum were diagnosed during third cesarean section. This case report highlights the importance of routine examination of adnexal structures during cesarean section, so that any unsuspected pathologies in the uterus, tubes, and ovaries are diagnosed.

Keywords

Uterus didelphys, longitudinal vaginal septum, cesarean section

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Background

In the general population, the true incidence of Müllerian defect is not accurately known. The mean incidence of uterus didelphys is 8.4%.¹ In a review of 152 pregnancies in 114 patients with untreated uterus didelphys, the mean ectopic rate was 1.3%, abortion rate was 32.2%, preterm delivery rate was 28.3%, term delivery rate was 36.2%, and live birth rate was 55.9%.¹ There are case reports of different modes of deliveries including successful vaginal birth after cesarean (VBAC) in patients with uterus didelphys^{2–4} as well as ruptured uterus in a woman whose didelphys uterus was not diagnosed during previous cesarean section (CS).⁵

Uterine anomaly is a well-known risk factor for cervical and interstitial ectopic pregnancies.⁶ Cornual ectopic pregnancy in women with underlying uterus didelphys has been reported.⁷ Longitudinal vaginal septum is typically associated with septate uterus or uterus didelphys. It causes dyspareunia, infertility, and labor obstruction.^{4,8–10} Herein, we report a case of 28-year-old woman whose uterus didelphys and vaginal septum were diagnosed during third CS with the aim of creating awareness of routine examination of adnexal structures during CS, and the future obstetrics risks in cases of undiagnosed uterus didelphys.

Case presentation

A 28-year-old, G₃P₂, was presented to the emergency room (ER)—Jigme Dorji Wangchuck National Referral Hospital

(JDWNRH), Thimphu, Bhutan—at 38 weeks pregnancy with intermittent lower abdominal pain which started about 5 h. The intensity and frequency of the pain was gradually increasing with pain lasting for about 30–40 s and occurring every 3–5 min. She complained of prevaginal blood-stained mucoid discharge.

Her previous two children were delivered by CS. Her first child was delivered in 2012 by emergency CS due to primi breech in labor. Her second child was delivered in 2018 by elective repeat CS due to previous scar with fetal breech presentation. While reviewing the previous medical records maintained in the record section of JDWNRH, details about her uterine anomaly and vaginal septum were not mentioned, meaning that the anomaly was not recognized. Review of medical documents of her first CS revealed that the emergency CS was done for breech presentation. There is no mention about uterine anomaly in the operation note. Per-vaginal examination was performed before CS, but there was no mention about vaginal septum. Review of records of her second CS showed that there was extension of uterine incision to cervix, and it was repaired

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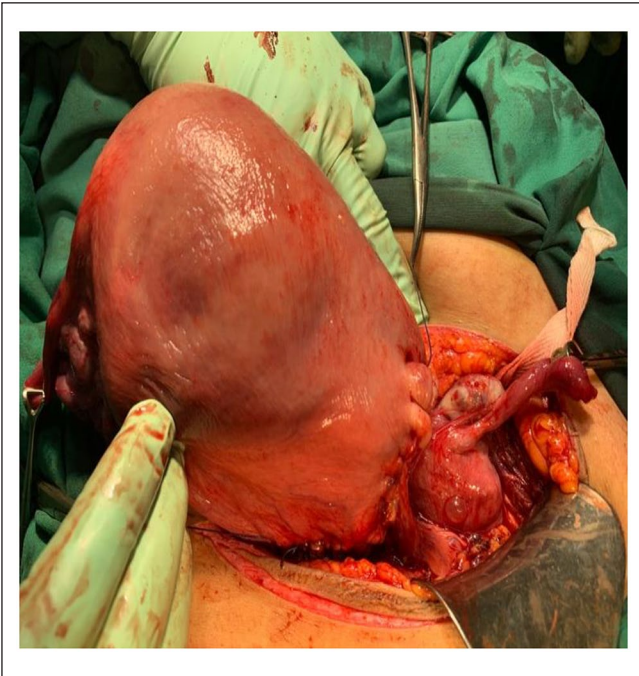


Figure 1. Per-operative image after closure of uterine incision showing uterus didelphys with separate fallopian tubes and ovaries. The abdominal incision scar shows excision of previous scar.

by consultant obstetrician-gynecologist. The uterine anomaly was not mentioned in this document too.

She attained menarche at 17 years of age. Her menstrual cycle was regular at 28 days lasting for 3–4 days with moderate dysmenorrhea. She experienced occasional dyspareunia since her first sexual debut. On examination, her vitals were stable, single life fetus with fundal height corresponding to period of gestation with cephalic presentation. Per-vaginal examination was not done in the ER as the mode of delivery will be repeat CS. Emergency CS was decided as she was in labor pain with history of previous two CS. The hospital protocol is to perform elective CS at 38 completed weeks of gestation in pregnancies with history of two or more CSs.

After obtaining written informed consent for emergency CS and bilateral tubal ligation (BLTL) under spinal anesthesia (SA), emergency CS with Pfannenstiel skin incision excising the previous scar under SA was performed. A live female baby weighing 3.4 kg in cephalic presentation was delivered at 6:57 am on 5 April 2021. APGAR score¹¹ was 7 and 9, at 1 and 5 min, respectively. There was fresh meconium-stained amniotic fluid.

After closure of uterine incision, while looking for left fallopian tube to do tubal ligation, the left horn of didelphys uterus with separate fallopian tube and ovary was noted. The pregnancy was noted in the right horn of uterus didelphys (Figure 1). BLTL with Pomeroy's technique¹² was performed. In Bhutan, BLTL is usually performed during third CS irrespective of woman's age if she requests for it, and written informed consent is provided.

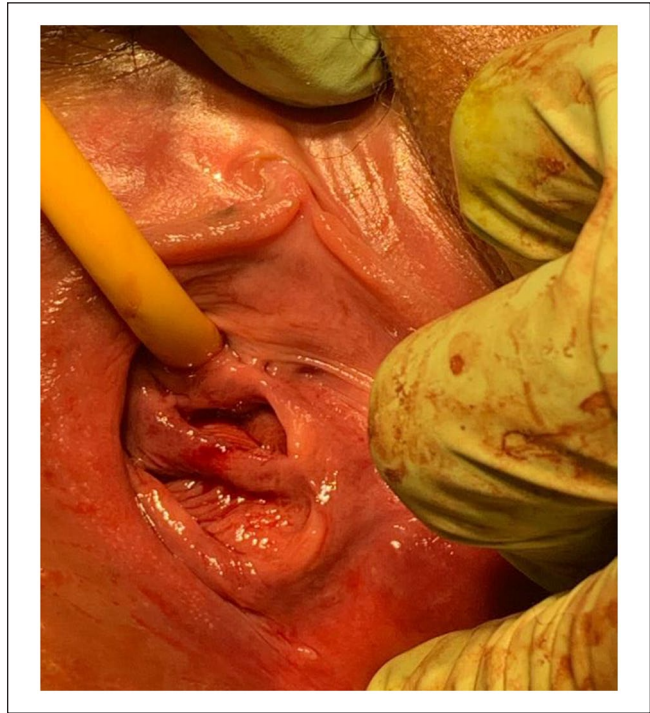


Figure 2. Double vagina with a longitudinal vaginal septum with urethral catheter in-situ.

Per-vaginal examination revealed double vagina with a longitudinal vaginal septum (Figure 2) with two cervixes. There was defect in the proximal end of vaginal septum (Figure 3). The option of vaginal septoplasty was offered and explained about the procedure, benefits, and complications, but the patient denied as she has occasional dyspareunia only and she is afraid of another surgery.

Postpartum ultrasound scan of kidney-ureters-bladder (KUB) revealed normal anatomy. Her serum creatinine was 0.9 mg/dL (within normal range) on second postoperative day.

The mother and baby had an uneventful postoperative recovery, and they were discharged home on third postoperative day.

Discussion

Uterus didelphys has term pregnancy rate of about 45%.¹ Breech presentation, vaginal birth, CS, and successful VBAC are reported in women with uterus didelphys.^{2,4,13} Breech presentation was noted in 43% and CS in 82% of 26 women with uterus didelphys.¹⁴ Successful external cephalic version in women with uterus didelphys has been reported.¹⁵ Women with uterus didelphys with twin pregnancies one in each horn were diagnosed during CS.^{2,16} Undiagnosed uterus didelphys has ruptured during a trial of vaginal birth after CS.⁵

Our case does not fall in any one of the two classifications according to the new classification of Herlyn–Werner–Wunderlich Syndrome (HWWS)¹⁷ as she has no

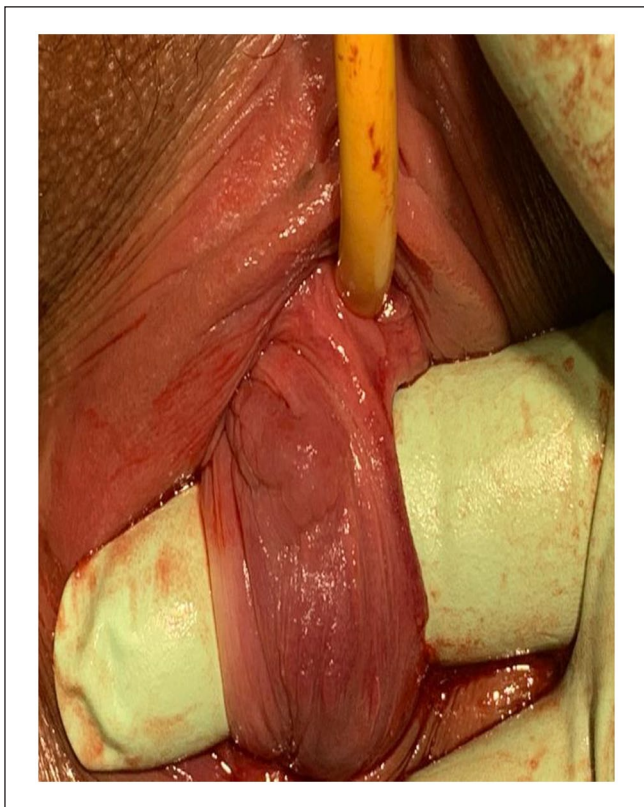


Figure 3. Demonstration of septal defect at the proximal end of vaginal septum.

vaginal obstruction. There are reports of association of HWWS with pelvic endometriosis.¹⁸ Among 16 cases of obstructing malformations of the uterus and vagina operated at The Children's Hospital, Harvard Medical School, between 1970 and 1983, one case had her kidneys in normal position.¹⁹

Similar to our case, there are case reports of successful term singleton as well as twin pregnancies elsewhere.^{3,20–23} Uterine anomaly is detected during infertility work up,²² during second vaginal delivery which was missed in first vaginal birth,²⁰ and cases of delayed-interval-delivery of twins complicated with chorioamnionitis in uterus didelphys.²³ Our case is unique and special one: The uterine anomaly and vaginal septum were not diagnosed prior to conception or antenatal ultrasound scan; she had three term pregnancies without any antenatal complications; and uterine anomaly and vaginal septum were diagnosed during third CS only.

Resection of vaginal septum relieves dyspareunia.^{9,21} Despite presence of symptoms such as dyspareunia and dysmenorrhea, vaginal examination was not done. In cases of dyspareunia, routine vaginal examination might reveal vaginal septum which can be excised with complete relief of symptoms.⁹ In our case, vaginal examination which was performed prior to the first CS has not recognized vaginal septum. This probably could be due to the lack of

knowledge about the vaginal anatomy and its anomalies among health workers.

The uterus didelphys was not recognized during previous two CS, probably the obstetrician did not examine the adnexal structures during CS. Despite repairing the extension of uterine scar to the cervix, uterine anomaly was missed. The standardization of CS procedure including routine examination of adnexal structures, documentation of findings, and clear communication with the patient will be very important. There are cases where uterine anomalies were missed during laparotomy.^{18,23}

Undiagnosed uterus didelphys risks uterine rupture in future if a trial of vaginal birth is planned. As reported in the literature, pregnancy in one horn of the uterus may be confused with cornual ectopic pregnancy,⁷ and it might mimic ovarian tumor.²⁴

Conclusion

During CS, adnexal structures should be assessed routinely, findings should be documented, and the information should be clearly conveyed to the patient. Routine vaginal examination should be performed in women with dyspareunia as the vaginal septum may be the cause. Diagnosis of unsuspected uterine anomalies and adnexal pathologies would help in counseling and managing future obstetrics and gynecological problems.

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Declaration of conflicting interests

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Ethical approval

Our institution does not require ethical approval for reporting de-identified individual case report.

Informed consent

Written informed consent was obtained from the patient for her anonymized information to be published in this article. This informed consent is available with the principal author.

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