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Case Study

UTERUS DIDELPHYS IN NULLIPAROUS AND MULTIPAROUS WOMEN – A RARE ENTITY

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ABSTRACT

Uterus didelphys is a rare congenital uterine abnormality in which the embryogenetic fusion of the Mullerian ducts fails to occur. It will lead to the formation of a double uterus with two separate cervices and most often a double vagina with a longitudinal septum as well. Here, we present two different cases of uterus didelphys with varied presentations. The first case is a nulliparous woman presented with post-coital bleeding. On examination, two cervical openings with a longitudinal complete vaginal septum were found, conservative management was done. Findings of didelphys uterus were confirmed on USG. The patient was counseled and discharged. The second case is a multiparous woman with previous cesarean delivery, rupture of membranes, and meconium in this pregnancy with term pregnancy taken up for emergency cesarean section. Dense adhesions and a mass on the right side of uterus were found intraoperatively, which on further inspection confirmed to be patent right horn of uterus. Diagnosis of uterus didelphys was made after doing per speculum and per vaginal examination post-cesarean.

Keywords: Uterus didelphys, Mullerian ducts, Nulliparous, Multiparous.

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INTRODUCTION

During embryogenesis, the uterus is formed by the fusion of the two paramesonephric ducts (Mullerian ducts), which normally fuse to form the single uterine body. Mullerian duct anomalies (MDAs) are congenital defects of the female genital system and result from abnormal embryological development of the Mullerian ducts. These abnormalities may encompass failure of development, fusion, canalization, elongation, reabsorption, or differentiation, which normally occur between 6 and 22 weeks *in utero*. The cause of the fusion failure is unknown and will lead to the formation of a double uterus with two separate cervices and most often a double vagina with a longitudinal septum as well, that is, a didelphic uterus. The vagina, renal system, and less commonly the skeleton may be affected with associated defects. Incidence of uterine anomalies is calculated to be 0.5–6% in all women, 13.3% in women with recurrent pregnancy loss (RPL), and 24.5% in women with infertility [1-4].

Septate uterus is the most common uterine anomaly with a mean incidence of ~35% followed by bicornuate uterus (~25%) and arcuate uterus (~20%). Among all Mullerian duct anomalies (MDAs), uterus didelphys was found to be the second least common (8.3%) [4]. These anomalies may result in infertility, poor reproductive outcome, and menstrual abnormalities. Most women with a didelphys uterus remain asymptomatic; but some, in whom variable degree of longitudinal vaginal septum is there, may present with dyspareunia or dysmenorrhea.

Post-coital bleeding is bleeding or spotting that occurs during or after sexual intercourse and is not related to menstruation. It mainly comes from surface lesions of the genital tract which mostly includes cervical causes such as cervical polyps, cervicitis, ectropion, cervical intraepithelial lesion (CIN), or carcinoma [5]. Sexual abuse and foreign bodies are also other causes of post-coital bleed. Here, we discuss two different rare cases of uterus didelphys with different presentation: One who presented as post-coital bleeding and other who was diagnosed during cesarean section.

CASE PRESENTATION

Case 1

A 28-year-old nulliparous recently married woman came at night in emergency at PGIMS, Rohtak, with complaints of excessive bleeding per vaginum after coitus. Initially, she went to a nearby government hospital where she received some injectables to stop blood and some pain killers (according to patient) and then referred to our institute for further management. There was a history of soakage of six pads in 7 h. On further history, it was found that it was not her first intercourse with her husband. On examination, the patient was conscious and well oriented with stable vitals. There was mild pallor (may because of blood loss). On per abdomen examination, it was soft but bladder was full which was emptied by K-91 urethral catheter. On local examination, applied pad was superficially soaked and no active bleeding was found. When per speculum (P/S) examination was done, a vertical complete transvaginal septum with around 1 cm tear on junction of vagina and septum on posterior side was found which was not bleeding at that time. Two different cervical openings visualized in each half of vagina. Above findings were confirmed on per vaginum (P/V) examination and uterus was found deviated toward the right side.

Conservative management was done. Findings of uterus didelphys confirmed on ultrasonography. The patient and her attendants were counseled as they were unaware of the patient condition and she was discharged and asked to follow up in OPD.

Case 2

A 26-year-old female gravida 4 para 1 with no live issue and two abortions with one previous cesarean section presented in our emergency at term gestation with complaints of leaking per vaginum. She was conscious and her vitals were normal. On per abdomen, there were a low transverse scar, term size uterus which was deviated to the left side, fetal heart was irregular, and contractions were mild. On per vaginum examination, os was admitting tip of finger, cervix seems to be deviated in S-shaped manner, presenting part was high-up, leaking was present which was meconium stained. Her pelvis was adequate in size and shape. The patient was taken up for emergency cesarean section

after consent. Intraoperatively, there were adhesions of omentum to bladder peritoneum and on to the uterine wall. Adhesiolysis was done and the delivered as vertex. While examining the patient for conditions of adnexa, a mass was found in the right side of uterus, and on further inspection, it was confirmed to be the patent right horn of the uterus with normal tube and ovary. The right horn also found to have a scar which is supposed to be of previous cesarean, as this time, pregnancy was in the left horn. The patient was closed after achieving complete hemostasis. To confirm the diagnosis of didelphys uterus, repeat P/S and P/V examination was done after the cesarean. It was found that there were two cervical openings with the vaginal septum in between. This confirmed our diagnosis of didelphys uterus. Post-operative period was uneventful and the patient was told about her uterus and discharged with stable vitals.

DISCUSSION

A didelphys uterus remains a very rare Mullerian duct anomaly when compared to other anomalies described in the Buttram and Gibbons classification. Here, we are adding uterus didelphys with vaginal septum as a rare cause of post-coital bleeding. Hence, one should keep in mind this condition while examining a case of post-coital bleed.

There is a significant risk of preterm labor and malpresentation in women with uterus didelphys when compared with normal uteri, so these patients may need special attention. Surgery is rarely indicated only in selected patients. Heinonen had performed cesarean section in 82% of patients [1]. While performing cesareans, we should always look for the uterine contour, cavity, and conditions of adnexa, not to miss any malformations associated. Some studies showed uterus didelphys to be associated with higher rate of infertility, spontaneous abortion, intrauterine growth retardation, and postpartum bleeding [6]. Herlyn-Werner-Wunderlich (HWW) syndrome has also been discovered to be associated with uterus didelphys and is also named as obstructed hemivagina and ipsilateral renal anomaly (OHVIRA). This is a very rare congenital anomaly of the urogenital tract which includes Mullerian ducts and Wolffian structures, and is portrayed by the triad of didelphys uterus, obstructed hemivagina, and ipsilateral renal agenesis [7]. As there is obstruction at one-half of vagina, this condition can lead to hematocolpos or hematometrocolpos on the side of the obstructed hemivagina, because of which patient will present with a mass effect and subsequent lower abdominal pain [8-10]. Most cases present after menarche with intense lower abdominal pain and/or with a protruding mass over the vaginal introitus [8-10]. A preliminary pelvic ultrasound is to be done followed by an MRI to confirm the diagnosis.

Heinonen *et al*. [1] in his study described a total of 26 cases which were studied retrospectively over 21 years. Out of 26 cases, five reported during pregnancy, five during routine examination, and remaining presented with abdominal pain (four cases), dyspareunia (four cases), bleeding (two cases), leukorrhea (two cases), pelvic tumor (two cases), and primary infertility (two cases). A total of 18 patients conceived and 40 pregnancies were reported out of which 12 were abortions and 28 deliveries. In our case series, we reported one case of post-coital bleeding and another during pregnancy. Rezai *et al*. [11] reported one case of didelphys uterus which was successfully conceived, presented with term pregnancy, and delivered live baby vaginally without any complication, similarly as ours. Although in our case, the baby was delivered through cesarean section. In both case reports, there were earlier abortions. Hence, obstetricians should have high index of suspicion of uterine anomaly with repeated abortions and also in pregnancies presenting in third trimester complications to make early diagnosis of uterus didelphys so that obstetric complications can be smoothly handled [11].

Slavchev *et al*. [12] also reported three cases where in all three presented with full-term pregnancy, favorable birth outcomes of live full-term infants. Two of which were delivered through cesarean section mainly indicated for labor activity lack and other due to oxytocin stimulation response lack.

Being a rare entity and on extensive review of literature and as per above studies, to conceive among uterus didelphys depends on case to case and highly debatable issue. Surgical correction in terms of metroplasty not usually indicated as the data are insufficient. Only when women are symptomatic, surgical excision may be indicated. During pregnancy, cesarean section is indicated only on case-to-case basis especially when, vaginal septum is thick and inelastic resulting in risk for vaginal dystocia [11].

CONCLUSION

Pregnancy in a uterus didelphys deserves early diagnosis of the anomaly, and meticulous care in pregnancy and delivery to avert the associated adverse outcomes. Clinicians should have high index of suspicion of uterine anomaly to make early diagnosis of uterus didelphys, especially while handling obstetric complications.

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