Case Report

Uterus Didelphys presenting with ruptured uterus in third pregnancy: A rare case report

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Abstract

Uterus Didelphys "double uterus" is a congenital malformation that results from complete failure of fusion of the ipislateral paramesonephric ducts during embryonic development. Ideally, diagnosis should be made before pregnancy and labor to prevent adverse outcomes. Here we report a case of uterus didelphys in 30 year old female Gravida4 Para3 Living2 at 31st week of pregnancy admitted with prolonged rupture of membrane, hypovolemic shock and intrauterine fetal death. Postoperative recovery of the mother was uneventful. Clinicians should have high index of suspicion of uterine anomaly when assessing cases of dysfunctional labor to avoid delayed diagnosis and the associated adverse outcome.

Key words

Gravida4 Para3 Living2, 31st week of pregnancy, Prolonged rupture of membrane, Hypovolemic shock and intrauterine fetal death.

Introduction

Mullerian duct anomalies are congenital anomalies of the female genital tract resulting from non-development or non-fusion of the mullerian ducts or failed resorption of the uterine septum [1]. Duplication of the uterus results from the lack of fusion of the paramesonephric ducts in a local area or throughout their normal line of fusion [2]. In uterus didelphys, individual horns are fully developed and are normal in size, with two cervices inevitably present. Each individual horn is associated with one fallopian tube. Women with didelphic uteri may asymptomatic and unaware of having a double uterus. They may present with complaints of dysmenorrhea and dyspareunia [3]. The true prevalence of this anomaly is unknown as it may be discovered in the later part of a patient's life when presenting with infertility. The incidence of mullerian duct anomalies in the literature ranges from 0.5 to 5.0%. Approximately 11% of uterine malformations are didelphic uterus [4].

The chance of seeing a pregnancy continuing to term is significantly reduced, down to only 20%, with $1/3^{rd}$ of pregnancies ending in abortion and over half in premature deliveries. Only 40% of pregnancies resulted in living children [5].

Case report

A 30 year old female Gravida4 Para3 Living2 at 31st week of pregnancy admitted in our hospital for prolonged rupture of fetal membrane, hypovolemic shock and intrauterine fetal death. She had seven and half month amenorrhea with complaints of pain abdomen since morning with sudden giddiness. She attained menarche at the age of 13 years and had a regular 28 days cycle with 4 days normal menstrual bleeding. There was neither history of infertility nor dysparenuia. She had past history of single post-term vaginal delivery with intrauterine death. Her 2^{nd} and 3^{rd} pregnancy was full term and delivered by caesarean section. Other systemic examination uneventful. The transvaginal was ultrasonography (TVU) showed bicornuate uterus with double cervix.

She planned for hysterectomy and uterus along with cervix came for histopathological examination. On gross examination, the formalin fixed specimen consists of two uterus along with two cervix fused together however, lumen of both cervix were different and unremarkable (**Figure - 1**). Larger ruptured uterus measures

18x9x3.5 cm with endomyometrium measures 3.5 cm in thickness. Cut surface shows haemorrhage and is grey white to grey brown in color with ruptured scar identified measuring 4x4 cm (Figure - 2). On serial cutting no fibroid is seen. Smaller uterus measures 12x6x2.5 cm with endomyometrium measures 2 cm in thickness. Cut surface is grey white slightly hemorrhagic and on serial cutting no fibroid is seen. On microscopy, various sections studied from larger rupture uterus and smaller uterus which shows chronic cervicitis with retention cyst formation (Figure - 4). Endometrium shows marked decidual type changes (Figure - 5). Myometrium of larger ruptured uterus shows marked hypertrophy and ruptured site shows hemorrhagic congested vessels (Figure - 3).

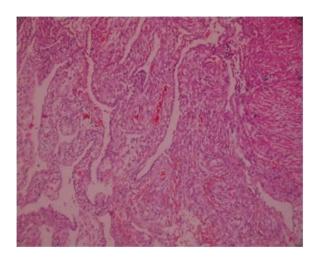
Figure – 1: Double cervix.



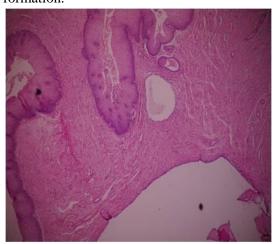
<u>Figure -2</u>: Double uterus with ruptured scar.



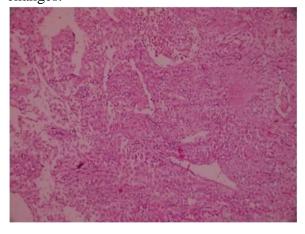
<u>Figure – 3</u>: H&E stained 10X view of ruptured scar showing haemorrhagic congested vessels.



<u>Figure -4</u>: H&E stained 10X view of cervix showing chronic cervicitis with retention cyst formation.



<u>Figure – 5</u>: H&E stained 10X view of endometrium showing marked decidual changes.



Discussion

Uterus Didelphys is rare and sometimes not even diagnosed. It occurs in 0.1% -0.5% of healthy fertile population [6]. Heinonen PK (2000) [7], evaluated the long-term clinical consequences, and reproductive performances of 49 women with uterus Didelphys that were followed up to 6.3 years. He found obstructed hemi vagina in 9 (18%) with 8 (16.3%) having ipsilateral renal agenesis. Five (13%) had primary infertility. Thirty four out of 36 (94%) women who wanted to conceive became pregnant, 21% miscarriage while 2% were ectopic pregnancy. The fetal survival rate was 75%, prematurity 24%, fetal growth retardation 11%, perinatal mortality 5.3%, and caesarean delivery rate 84%. Pregnancy was located in the right uterus in 76% cases.

Here, we report a rare case of 30 year old Gravida4 Para3 Living2 at 31st week of pregnancy complaints of ruptured fetal membrane, hypovolemic shock and intrauterine fetal death. The patient was diagnosed as uterus didelphys. The aim of this case report is to make clinicians to have high index of suspicion of uterine anomaly when investigating cases of dyspareunia, dysmenorrhea, infertility, spontaneous abortion, preterm labor, fetal malpresentation, intrauterine growth restriction, premature rupture of membrane and renal agenesis. Early diagnosis, meticulous follow up can avert most of these complications.

Conclusion

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

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