Failure of intrauterine contraception in a multigravida lady with an undiagnosed uterus didelphys: a case report

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ABSTRACT
Uterus didelphys remains one of the rarest uterine anomalies partly because; majority women experience no symptoms with an uneventful reproductive life. Despite the cost-effectiveness and efficacy paired with fewer side effects and convenience of using an intrauterine contraceptive device, it is most likely to prove futile in an unsuspected case of didelphic uterus resulting in an unintended pregnancy. Hence, intrauterine contraception is generally considered a contraindication in cases of uterine malformations. We present a case of a multigravida mother (G7 P3 L3 A3) with a history of three term vaginal deliveries, with an undiagnosed uterus didelphys, carrying a single live intrauterine pregnancy of approximately 6 weeks period of gestation, in the right uterine horn and a copper containing intrauterine contraceptive device in the left uterine horn, willing to undergo medical termination of pregnancy. A high index of suspicion, on the part of the gynaecologist as well as the radiologist, is required to investigate concurrent pregnancy with a history of an intrauterine contraceptive device insertion, in order to rule out rarer uterine malformations like uterus didelphys. Thorough history taking and clinical examination accompanied by improved imaging techniques should be performed at the time of first pregnancy in order to avoid an unsuitable placement of an intrauterine contraceptive device.

Keywords: Uterus Didelphys, Intrauterine pregnancy, Intrauterine contraceptive device

INTRODUCTION
Uterus didelphys is a congenital anomaly of the uterus, consisting of two hemi-uteri with separate endometrial cavities, two uterine cervices along with a longitudinal vaginal septum in most cases. With an incidence of 1 in 3000, uterus didelphys is presently the second least common of all uterine malformations at 8.3%.¹ About 15-30% of non-obstructed uterus didelphys may be associated with renal anomalies of which renal agenesis is the most frequent.²

The female reproductive system develops from the paired Mullerian ducts. Mullerian ducts develop lateral to Wolffian ducts at around 6-8 weeks’ gestation and then migrate medially to fuse in the midline at around 10th week of gestation to form the uterovaginal canal after incorporating with the urogenital sinus.³ Lateral fusion defect of paired Mullerian ducts may result in a ‘double’ uterus with two functioning endometrial cavities.

Full term pregnancies occur in approximately 45% of women with uterus didelphys.⁴ The remaining women, report either infertility or recurrent miscarriages or preterm labour due to smaller size of the uterine horns and cervical incompetence.

Failure of a single intrauterine contraceptive device in an unsuspected uterus didelphys is probably due to the presence of a ‘double’ uterus with two functioning endometrial cavities.
CASE REPORT

A 37 years old, G7P3A3L3 patient with a history of three previous vaginal deliveries and living issues accompanied with a history of three previous induced abortions presented with a will to undergo a fourth induced abortion. Her last menstrual period was recorded to be on 10th January 2020.

On taking elaborate history, she recalled to have undergone the third medical termination of pregnancy on 22nd September 2019, while carrying 18 weeks period of gestation. She displayed a discharge certificate confirming this with an additional documentation of an intrauterine contraceptive device inserted following the procedure. She had no medical comorbidities and no past surgical history. General and systemic examination did not reveal any abnormality. On per vaginal examination, thread of intrauterine contraceptive device was felt through a bulky cervical os.

On reviewing her previous imaging reports related to the previous pregnancy, no evidence of a mention of any uterine malformation was found. Following a positive urine pregnancy test on 18th February 2020, she was advised to get a transabdominal and transvaginal ultrasonography performed due to pregnancy following a failed contraception. On transabdominal ultrasonography, two uterine horns and a bulky cervix were noted with an intrauterine contraceptive device in the cervical cavity and single live intrauterine pregnancy corresponding to 6 weeks period of gestation in the right uterine horn.

The findings were verified on a transvaginal ultrasonography which additionally revealed two separate cervical cavities with an associated longitudinal vaginal septum and the presence of an intrauterine contraceptive device in the left cervical cavity.

Responding to the rising anxiety of the couple to terminate the unwanted pregnancy, medical termination of pregnancy was performed without delay, with oral mifepristone and sublingual misoprostol. She reviewed a week later with a complete abortion and an expelled copper containing intrauterine contraceptive device. Due to better delineation of uterine malformations and associated renal and urinary tract anomalies, an abdominal magnetic resonance imaging (MRI) scan was later performed which corroborated the ultrasound findings. Coronal STIR images revealed two widely spaced uterine corpora, each with a single fallopian tube. Separate divergent uterine horns were noted with a large fundal cleft. The normal uterine zonal anatomy was preserved. T2W axial images revealed two separate endocervical canals opening into separate fusiform endometrial cavities, with no communication between the two horns. The presence of two separate vaginal canals with a longitudinal vaginal septum was confirmed.

She was advised effective contraceptive choices in her follow up visit, 3 weeks following the medical termination, in order to avoid the risks associated with repeated induced abortions. By GATHER approach, the couple agreed upon the use of oral contraceptive pills.
Figure 4: MRI STIR axial images at the level of upper third of vagina, showing two separate vaginal canals (white arrows) with intervening longitudinal vaginal septum (red arrow).

DISCUSSION

Uterus didelphys represents a very rare mullerian duct anomaly which usually escapes suspicion as most women remain asymptomatic. There are just a handful of case reports on uterus didelphys with obstetric complications, in the literature with more research being done on commoner malformations with poorer reproductive outcomes like septate and bicornuate uterus. The use of intrauterine contraception in a case of didelphic uterus is far more rarely reported in the literature.

Despite most cases being asymptomatic, rarely women with didelphic uterus may present with dyspareunia and dysmenorrhoea coupled with haematocolpos or haematometrocolpos, especially with an obstructing vaginal septum. There are even reports of genital neoplasms and endometriosis in a didelphic uterus.

Fertility chances related to uterus didelphys, though being poorer than with a normal uterus, still remains better than with a septate or a bicornuate uterus.

A long term retrospective follow up study of 49 women with uterus didelphys revealed no deterioration in fertility rates but a decreased rate of spontaneous abortion and an increased rate of premature association with the anomaly when compared with septate and bicornuate uterus as reported in other studies. The increased risk of preterm labour and associated fetal growth restriction may be attributed to cervical incompetence and smaller sized uterine horns.

Intrauterine contraceptive devices though being fairly popular due to their long term reversible contraception, safety, efficacy and suitability for a wide range of population including in breastfeeding women, they are relatively contraindicated in presence of uterine malformations. Where cases of failed contraception, uterine perforation, expulsion, bleeding and pain with intrauterine contraception in such women have been reported, there are some reports of uncomplicated and successful intrauterine device insertion in such anomalous uteri. Comparatively there are even fewer reported cases of failure of intrauterine contraceptive device in a didelphic uterus indicating that in women with a ‘double uterus’, use of two intrauterine contraceptive devices in both uterine horns might be more justifiable to prevent unwanted pregnancy in the event of contraindications or unacceptability to hormonal or barrier contraceptives.

Additional research is needed in the form of case control and cohort studies with larger number of women with uterine anomalies, with special interest in rarest ones like uterine didelphys to provide information about the safety and efficacy of an intrauterine contraceptive device.

CONCLUSION

A high index of suspicion on the part of the gynaecologist as well as the radiologist, is required to investigate concurrent pregnancy with a history of an intrauterine contraceptive device insertion. Thorough history taking and clinical examination accompanied by improved imaging techniques should be performed at the time of first pregnancy in order to avoid an unsuitable placement of an intrauterine contraceptive device. The need to pursue larger studies on the safety and potential use of intrauterine contraceptive device in uterus didelphys, should be met with, before counselling women against their use.

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