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Case Report

AN UNUSUAL CASE OF TERM PREGNANCY IN A COMPLETE SEPTATE UTERUS WITH CERVICAL AND VAGINAL SEPTATION

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ABSTRACT
The exact incidence and prevalence of mullerian duct anomalies remains unknown. Although being the most common type, septate uterus is associated with the poorest reproductive outcomes such as recurrent spontaneous miscarraiges, preterm labour, malpresentations, fetal growth restrictions and even infertility. We present a case of a 19 years old primigravida mother carrying 39 weeks period of gestation, admitted with premature rupture of membranes in breech presentation. She was not previously diagnosed with complete septate uterus ultrasonologically during her routine antenatal care. A successful emergency caesarean section was performed and a complete septate uterus was diagnosed intraoperatively. Associated longitudinal vaginal septum was noted during clinical examination. Considering reports of septate uterus having worst reproductive outcomes among all the uterine anomalies, this case represents a rare one with an undiagnosed but uneventful term pregnancy.

Keywords: *Septate Uterus, Term Pregnancy, Mullerian Duct Anomalies*

INTRODUCTION
Uterine anomalies commonly remain undiagnosed at birth only to be reported during the childbearing age when reproductive malfunctions occur. Many patients remain asymptomatic during their reproductive years with a couple of them undergoing an incidental diagnosis later on. The incidence of mullerian duct anomalies range widely from as low as 0.001% to as high as 10% (Brown and Badawy, 2013). The mullerian ducts constitute a set of paired embryologic structures which differentiate to form the fallopian tubes, uterus, uterine cervix and upper four-fifth of the vagina. The lower one-fifth of the vagina develops from the urogenital sinus. Normal development of the female reproductive tract involves the fusion and resorption of the mullerian ducts in-utero and any disruption in the process can result in the formation of mullerian duct anomalies. A uterine septum is a result of failure of resorption of the tissue connecting the two mullerian ducts prior to the 20th embryonic week. Septate uterus (Class U2, ESHRE Classification), (Grimbizis *et al.*, 2013) is reported to have a mean incidence of 35%, thus forming the most common uterine anomaly (Sawai *et al.*, 2017). Although the true prevalence of septate uterus is unknown, it appears to range between 1 to 2 per 1000 to as high as 15 per 1000. (Valle and Ekpo, 2013) But it is associated with the poorest reproductive outcomes with nearly 20-25% women experiencing obstetrical complications like recurrent miscarraiges, preterm labour, malpresentations and fetal growth restrictions, among which the first two are the most common (Perino *et al.*, 2014). Substantial evidence suggests the association of renal anomalies with septate uterus, warranting a consideration of renal imaging is such women. (Broughton et al., 2016) 3D ultrasound, sonohysterography and MRI successfully clinch the diagnosis and are preferred diagnostic tools over invasive procedures like laparoscopy and hysteroscopy.

CASE
A 19 years old primigravida mother carrying 39 weeks period of gestation got admitted in the obstetric ward of our hospital with the complaint of leaking per vagina for last 4 hours. Thorough history taking did not reveal the presence of any medical co- morbidities like diabetes mellitus, hypertension or thyroid disorders. During routine antenatal care, she did not experience any obstetrical complications like bleeding, leaking or excessive pain abdomen. She did not complain of any labour pains on admission.

Overall, she had an uncomplicated antenatal period following a spontaneous conception. General examination and examination of other systems did not reveal any abnormality. On per abdominal examination, uterus was term size with probable breech presentation. No associated contractions were palpable abdominally. Clinically liquor volume was less than adequate. She perceived adequate fetal movements and fetal heart sound was recorded at 134 beats/min. On confirming the above findings on per vaginal examination, a longitudinal vaginal septum was noted extending through the entire length of the vagina starting from the external cervical os, which was dilated to just a tip of finger. Active leaking with clear liquor was noted. Membrane status could not be determined due to insufficient dilatation and thick tubular nature of the cervix. As a result, cervical septation could not be delineated separately. On reviewing her antenatal ultrasound reports, no record of any uterine anomaly was found. An ultrasound was repeated to determine the presence of any associated uterine malformation related to the longitudinal vaginal septum incidentally diagnosed per vaginally, along with determination of fetal presentation and the liquor volume. Breech presentation with oligohydramnios (AFI=4 cm) was confirmed but no mention of any uterine abnormality was recorded. No gross fetal anomalies were seen. Placenta was recorded to lie anteriorly over the fundus with no retroplacental collection. The patient was scheduled for an emergency caesarean section indication being primigravida at term with breech presentation with premature rupture of membrane and oligohydramnios. On giving a transverse incision over the lower segment of uterus, a thick septum was identified and cut delivering a healthy male baby by breech extraction. The uterine septum was found extending from the fundus to the internal cervical os.

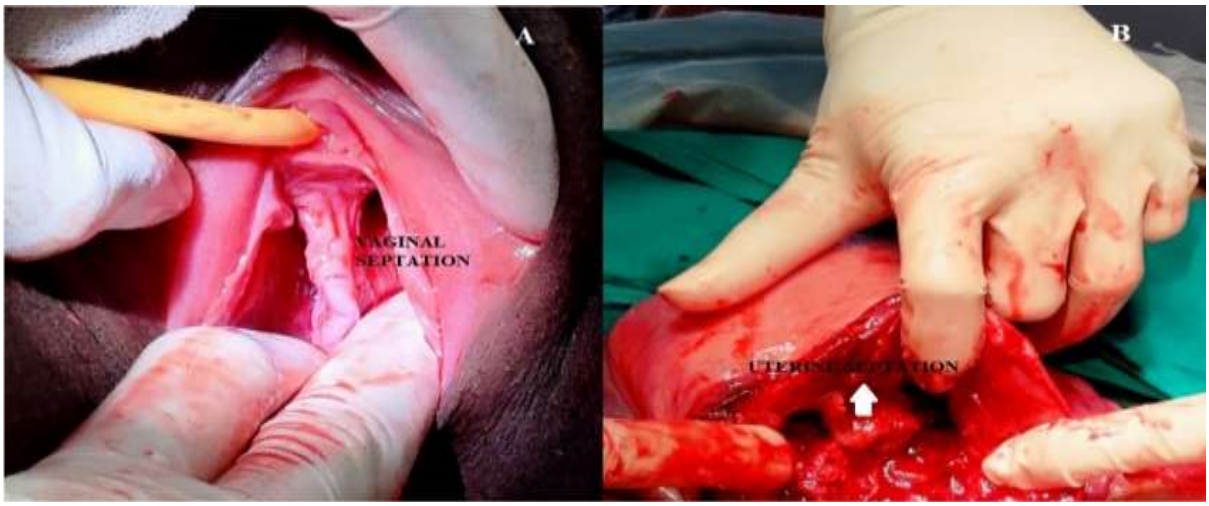

Figure 1: Vaginal septation seen pre-operatively **(A)**; intraoperative image of septate uterus **(B)**

Placenta and membranes were separated from the left uterine cavity. Intraoperatively, cervical septation couldnot be separately identified. The baby's weight was recorded to be 2650 grams with no congenital anomaly and an Apgar score of one minute and five minutes to be 7 and 10 respectively. The operation was completed with minimal blood loss and the patient was haemodynamically stable postoperatively. Both the mother and the baby were discharged in a healthy condition after a week. At the time of discharge, both the patient and her husband were adequately educated about septate uterus and the probable risks in future pregnancies. Contraceptive advice relevant to her situation was discussed and the reason for ruling out the use of any intrauterine contraceptive device was explained. By GATHER approach, both the patient and her husband agreed upon the use of oral contraceptive pills. The patient was counselled about the need for follow-up and 6 weeks post-delivery she was followed up with an additional imaging, MRI pelvis, to confirm the presence of a complete septate uterus. T2W axial images on MRI, revealed two endometrial canals, completely separated by low signal intensity fibrous tissue with

extension into endocervical canal and the entire vaginal canal. The external uterine fundal contour was convex with closely placed two uterine cavities (angle of separation < 75 degree).

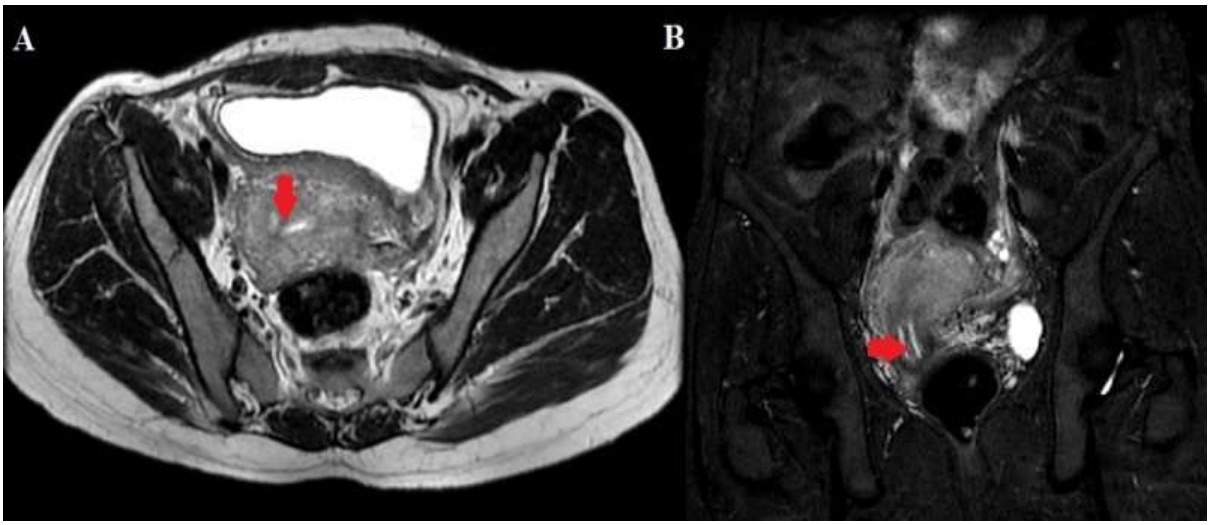

Figure 2: MRI images- T2W axial image shows two endometrial canals, completely separated by low signal intensity fibrous tissue (red arrow) with extension into endocervical canal **(A)**; coronal STIR image shows two cervical canals , completely separated by low signal intensity fibrous tissue (red arrow) **(B)**

DISCUSSION

Even with reported cases of poor reproductive outcomes, mullerian duct anomalies are relatively common and may be asymptomatic to the point of incidental diagnosis. The etiology of mullerian duct anomalies is complex and not completely understood. The most common types among them are the septate uterus, bicornuate uterus and arcuate uterus. Septate uteri include the presence of a complete or a partial septum. Uterine septa are often diagnosed at the time of evaluation for infertility but there is insufficient data to conclude that uterine septum is associated with infertility (Uterine septum: a guideline. Practice Committee of the American Society for Reproductive Medicine. Vol 106, September 1, 2016.)

Although septate uteri constitute the commonest uterine anomaly, numerous studies have shown that a uterine septum is associated with an increased risk of adverse pregnancy outcomes like recurrent miscarraiges, malpresentation, preterm birth, placental abruption and fetal growth restriction eventually increasing the risk of caesarean section and subsequent maternal and perinatal morbidity and mortality. (Uterine septum: a guideline. Practice Committee of the American Society for Reproductive Medicine. Vol 106, September 1, 2016.)

Even though both 3D ultrasound and MRI have proved to be highly accurate in the diagnosis of uterine anomalies with an additional advantage of accurately diagnosing associated renal and urinary tract abnormalities, 3D ultrasound is superior in cost effectiveness with comparable results to MRI. (Graupera et al. 2015) In this case report, an ultrasound could not diagnose complete septate uterus during routine antenatal care probably due to poor delineation of uterine malformations using 2D ultrasound used in our hospital. With a known increased risk of recurrent spontaneous abortions and preterm pregnancies, term pregnancies are relatively uncommon in complete septate uterus. It is essential to diagnose the condition early in pregnancy so that the woman can be made aware of the possible outcomes of her pregnancy and proper counselling can be done by the obstetricians for preparing her for any subsequent pregnancy. The mother in this case report, may experience secondary infertility, placental abnormalities like morbidly adherent placenta over the septum, placenta praevia or abruptio placentae in subsequent pregnancies, inspite of having an uneventful term first pregnancy. As evident from recent published data, she might

benefit from hysteroscopic uterine metroplasty and resection of the cervical septum to avoid such life threatening complications and simultaneously increasing the probability of a live birth pregnancy (Wang *et al.,* 2019). On avoiding hysteroscopic metroplasty, cervical cerclage operations may be required in subsequent pregnancies to reduce the risk of miscarriage or preterm labour, but with uncertain guarantee. In case of slightest suspicion of an anomalous uterus, radiologists should confirm using more accurate imaging techniques like a 3D ultrasound or a MRI for early diagnosis of both uterine and associated renal abnormalities. Proper history taking from both the woman and her partner is also invaluable especially in the presence of a vaginal septum.

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