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Case Report

Obstetrics and Gynaecology Section

Pregnancy in Bicornuate and Couvelaire Uterus: A Case Report

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ABSTRACT

In the general population, the incidence of uterine malformations is estimated to be 3-5%. Pregnancy in a bicornuate and couvelaire uterus is a rare anomaly. Pregnancy in a bicornuate uterus, a congenital Müllerian duct anomaly caused by incomplete fusion of the uterine horns, is associated with an elevated risk of adverse obstetric outcomes due to the altered uterine morphology, abnormal placental implantation dynamics and limited cavity volume. Women with a bicornuate uterus have been shown in large population-based studies to have increased risks of malpresentation, preterm delivery and notably placental abruption compared with pregnancies in normally shaped uterus, with abruption risks reported up to three times higher in this group. Hereby, the authors present a case of a 23-year-old female, gravida 2 para 1 living 1, previously preterm delivered, who came to the labour room with features of obstructed labour. The patient was posted for caeasarean section. Intraoperatively, there was a bicornuate couvelaire uterus with a broad ligament haematoma, with stillbirth of the baby. Obstetric hysterectomy was done in view of an atonic uterus with failure of conservative management. Regular check-ups, timely diagnosis, early ultrasound and delivery planning in a well-equipped hospital improve both maternal and fetal outcomes.

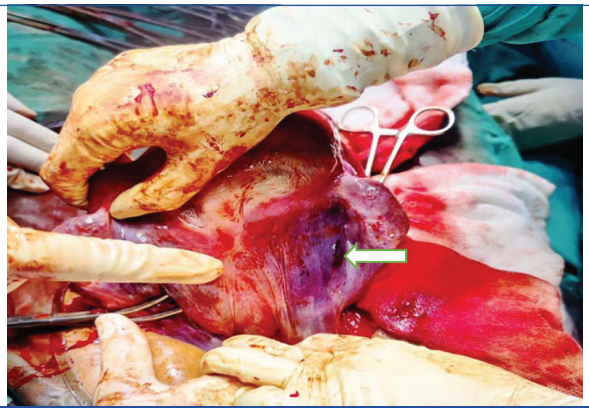
Keywords: Haematoma, Obstetric hysterectomy, Stillbirth, Uterine malformation, Uteroplacental apoplexy

CASE REPORT

A 23-year-old female with gravida 2, para 1, living 1 was referred to our centre from the Primary Health Centre (PHC) at 37 weeks two days. She had no antenatal visits {Antenatal Care (ANC)} at the hospital. The past medical and obstetric history of this patient showed that she had a previous preterm delivery in the seventh month of gestation with a baby birth weight of 800 grams. The cause for previous preterm delivery was suspected to be genitourinary infection, maternal stress or uterine anomalies. On examination, she was found to be in acute pain, afebrile, and pallor was present. She was vitally stable with a pulse of 86/min and blood pressure of 110/74 mmHg. On abdominal examination, uterine size was full-term, a tonic uterus was present, cephalic presentation and fetal heart sounds were variable on doppler. On per vaginal examination, the Os was fully dilated, station was zero, and the membranes were absent.

On investigations, haemoglobin was 8 g/dL, platelet count was 138000/cmm, blood group was A positive, prothrombin time, international normalised ratio, bleeding time and clotting time were within normal limits, Human Immunodeficiency Virus (HIV) and Hepatitis B surface Antigen (HBsAg) were negative. D-dimer was 1050 ng/mL, and fibrinogen was 600 mg/dL. The first ultrasound done at 14 weeks four days showed a single live intrauterine foetus, anterior placenta, normal amniotic fluid and baby weight 101 gm. An anomaly scan was not done. The second ultrasound was done at 27 weeks and four days of gestation, which showed single live intrauterine foetus, baby weight being 1.12 Kg, uteroplacental insufficiency present, amniotic fluid index 18.6 cm and single loop of cord around neck. The patient was asked to follow-up with 32 weeks and 36 weeks of ultrasonography, but did not follow up at Primary Health Care (PHC). The patient came to our hospital at 37 weeks and two days of gestation in labour. Obstructed labour was diagnosed with clinical features of maternal exhaustion and dehydration, a rigid, tender uterus, high riding non descending fetal head with significant moulding and oedema of vulva.

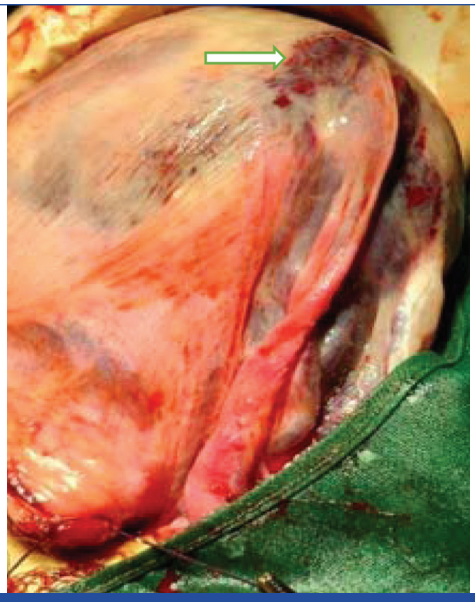
She was posted for emergency Lower Segment Caesarean Section (LSCS) in view of obstructed labour. Intraoperative findings showed that there was a broad ligament haematoma extending into pelvis [Table/Fig-1]. Incidental finding showed the uterus had uteroplacental apoplexy [Table/Fig-2]. The baby was delivered by modified Patwardhan's manoeuvre. The birth weight of baby was 2.65 kg and it was stillborn. Evidence of retroplacental clot measuring approximately 100 grams was noted. Two Packed Cell Volume (PCV), along with four Random Donor Platelets (RDP) were given. Estimated blood loss was 1500 mL. In view of an atonic uterus, uterotonics were given along with inj. tranexamic acid 1 g intravenously. Systemic devascularisation was done. Obstetric hysterectomy was done in view of an atonic uterus after obtaining written informed consent, as above stated conservative management failed. Specimen of a bicornuate uterus after obstetric hysterectomy is shown in [Table/Fig-3]. Patient was referred to a higher centre after initial stabilisation due to non-availability of an Intensive Care Unit (ICU) immediately postoperatively.



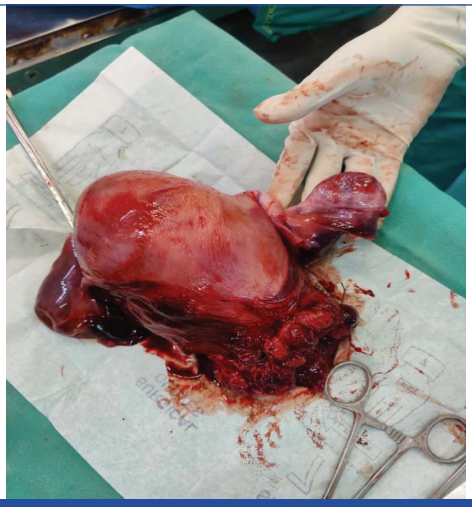
[Table/Fig-1]: White arrow showing broad ligament haematoma.

DISCUSSION

Unicornuate uterus, bicornuate uterus, didelphys, septate uterus, arcuate uterus and agenesis of uterus are several uterine anomalies [1]. A bicornuate uterus is a very rare anomaly and its incidence is estimated to be 0.1%-0.6% [2]. A bicornuate uterus is caused by impaired fusion or non-fusion of paramesonephric



[Table/Fig-2]: White arrow showing couvelaire uterus.



[Table/Fig-3]: Specimen of bicornuate uterus post-obstetric hysterectomy.

ducts. Paramesonephric ducts, also known as Müllerian ducts, are important embryological structures for the development of the urogenital system. Instead of a normal uterine shape, a bicornuate uterus is heart-shaped [3].

A bicornuate uterus has non-specific clinical manifestations. Most of the women remain asymptomatic and diagnosis is possible only during an examination. Management consists of careful monitoring and caesarean section being preferred mode of delivery [4]. As it is more likely that the reproductive outcome is not favourable in these patients, they are categorised under high risk pregnancy. Recurrent pregnancy loss or preterm labour in pregnant women is commonly encountered [5]. Bicornuate uterus associated with Couvelaire uterus is rare and the incidence is difficult to estimate because it is diagnosed by intraoperative direct visualisation. Couvelaire uterus, also known as uteroplacental apoplexy, is a rare complication of severe placental abruption in which haemorrhage from ruptured placental vessels infiltrates the decidua basalis and extends into the myometrium. The extravasated blood may dissect through the uterine muscle fibers and occasionally reach the serosal surface or broad ligament, resulting in a bluish or purplish discolouration of the uterine wall [6,7].

According to Cobec IM et al., Couvelaire uterus was associated with pre-eclampsia, amniotic fluid embolism placenta previa and placenta accreta [8]. In our patient, Couvelaire uterus was associated with bicornuate uterus. As the uterine cavity is abnormally shaped, those patients with full-term pregnancies have their babies settling for breech presentation rather than cephalic position [9]. But in our case, it was cephalic presentation.

Ultrasound, magnetic resonance imaging, hysteroscopy, hysterosalpingography or laparoscopy can be used for the diagnosis of a bicornuate uterus. However, when the pregnancy is advanced, it rarely provides a diagnosis [9]. The management of a bicornuate uterus will be preventive in the form of rest, Ultrasonography (USG) monitoring of fetal growth, cervical competence and lung maturation when its diagnosis is made at the start of pregnancy. At term, the increased frequency of obstructed labour makes caesarean section the preferred route of delivery [3]. In the patient, despite a prior preterm normal delivery, we preferred to perform a caesarean section. Intraoperatively, there was a broad ligament haematoma extending into the pelvis, with signs of Couvelaire uterus, decision of doing obstetric hysterectomy was made by the operating surgeon.

CONCLUSION(S)

A bicornuate uterus with a Couvelaire uterus is a rare combination. Bicornuate uterus does not always lead to obstetric complications and pregnancies can be carried to term. Early registration, regular check-ups, timely ultrasound and delivery planning in a well-equipped hospital improve both maternal and fetal outcomes.

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