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Case Report

Atypical presentation and complications of term pregnancy with Mullerian duct anomaly: two case reports

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ABSTRACT

Mullerian duct anomalies present as vague and overlapping features with other gynaecological conditions or may remain asymptomatic. These can lead to various obstetrical complications such as miscarriage, fetal growth restriction, preterm birth, abnormal placental implantation, malpresentation, increased risk of cesarean section, retained placenta and others. We hereby presented two cases with term pregnancy in women with Mullerian duct anomaly and the obstetrical complications they had. The first case landed up in a cesarean section at term due to breech presentation as a result of MDA. The second case had a vaginal delivery followed by entrapped retained placenta due to MDA, which had to be removed in piece meal. We hereby discussed the possible pathophysiology leading to these obstetrical complications in MDA pregnancies. MDA can lead to complications in pregnancy and previously undiagnosed women when detected with MDAs should be informed about complications and treatment options before and during pregnancy, for better maternal and neonatal outcomes.

Keywords: Mullerian duct anomalies, Pregnancy, Bicornuate uterus

INTRODUCTION

Mullerian duct anomalies are caused by abnormal interruption during development of female reproductive tract during fetal life.[1] In many cases, detection is difficult due to lack of awareness and common overlapping features with other gynaecological conditions.[1] These anomalies may lead to increased prevalence of various gynaecological and obstetrical complications. We hereby presented two cases in which Mullerian anomalies were detected during pregnancy and parturition and the anomalies led to obstetrical complications.

CASE REPORT

Case 1

26-year old primigravida, who conceived spontaneously within first year of married life, was referred to our centre at 28 weeks gestation due to bicornuate uterus mentioned on ultrasonography. Pregnancy was well supervised and uncomplicated. On serial sonography, gestational sac was seen in right uterine horn, then foetus was transverse initially and became breech towards term. She went into spontaneous labour at 38 weeks 2 days gestation with breech presentation. An emergency cesarean section was

done for her in view of breech in labour in primigravida. Intraoperatively, transcurvilinear incision was given over lower segment. Breech and lower limbs were in left horn while back and head were in right horn. Delivery of a baby boy weighing 2.7 kg with good Apgar score was carried out uneventfully. Uterus was heart shaped as in Figure 1. Bilateral fallopian tubes and ovaries were normal looking. Post-operatively, patient and baby remained stable.

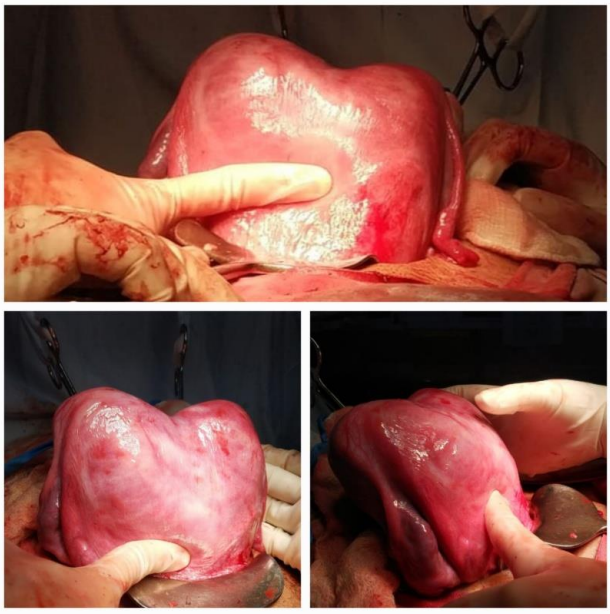

Figure 1: Heart shaped bicornuate uterus.

Case 2

20-year old P1021 referred to our centre on day 0, 20 hours following full term vaginal delivery with retained placenta. Baby was boy, alive and well. Her antenatal period was well supervised and uncomplicated. Time from spontaneous term rupture of membranes to delivery was almost 11 hours. Misoprostol and oxytocin were given at referring centre but placenta didn't separate. Patient had been married for 3 years. Previously, patient had two missed abortions at approximately 10-12 weeks that were managed by evacuation and curettage, both times. So, patient was referred to our centre with a possibility of adherent placenta.

On admission, she was hemodynamically stable with no active bleeding per vaginum with hemoglobin 6.6 g/dl. Uterine height was approximately 28 weeks gravid size. Bedside ultrasound was done, showing whole placenta in uterine cavity, anterior myometrium well delineated, posterior myometrium seemed thinned. Patient was taken up for removal of placenta under anaesthesia after explaining the patient and attendant about the high risk of bleeding and need for peripartum hysterectomy in case placenta was adhered.

Intraoperatively, under general anaesthesia, the cervix and uterine cavity could not admit the fist of hand freely. The cavity also seemed to be deviated towards right side of abdomen. There was no plane of separation found between uterus and placenta, despite tracing through cord, due to difficulty in insertion of hand. Above findings suggested presence of two cavities or a septum in utero, with placenta entrapped in one cavity/compartment, with no space or entrance into the opposite cavity/compartment. Placenta removal was then attempted in piece-meal under ultrasound guidance. Placenta was removed successfully without any heavy episode of bleeding intraoperatively. Two packed cells were transfused. Broad spectrum antibiotics were given. Immediate post-operatively, retracted uterus showed a heart shape abdominally suggesting a right unicornuate uterus with non communicating non functioning left uterine horn as in Figure 2. Patient remained stable in post-operative period.

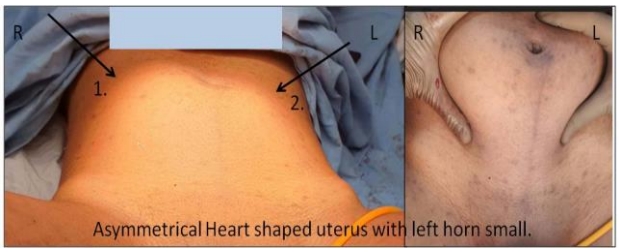

Figure 2: Unicornuate uterus with non communicating non functioning left uterine horn after evacuation of retained placenta: arrow 1: right unicornuate horn that contained retained placenta; arrow 2: left sided small horn palpable after evacuation of placenta.

DISCUSSION

Mullerian duct uterine anomalies (MDA) result from defective formation, fusion or reabsorption of Mullerian ducts during foetal life. These anomalies were present in 1 to 11% of the general population, 3 to 9% of women with infertility and 11 to 28% of women with a history of miscarriages.[2] The true prevalence of MDA, also known as congenital uterine anomalies (CUA), was uncertain, because of invasive diagnostic techniques and many of them remained unnoticed as they may cause no dysfunction to the woman. Depending on the failure of organogenesis and varying degrees of fusion or absorption defects, MDA can be divided into unification defects of the Müllerian ducts (unicornuate, bicornuate, or didelphys uterus) and canalization defects from incomplete resorption of the midline septum (subseptate or septate uterus).[3] The dysfunction of endometrial cavity may lead to pelvic pain, dysmenorrheal, abnormal vaginal bleeding, infertility, ectopic pregnancies, recurrent miscarriages and adverse pregnancy outcomes.[4] The obstetrical complications were high in incidence in women with MDA such as miscarriage, fetal growth restriction, preterm birth, abnormal placental

implantation, malpresentation, increased risk of cesarean section and retained placenta.[2,5] They were commonly associated with renal anomalies. Also, they were associated with anorectal and vertebral malformations and syndromes like Klippel-Feil syndrome, VACTERL. Previously, the gold standard diagnostic method was a combination of laparoscopy and hysteroscopy. However, imaging techniques, 2D-ultrasound and hysterosalpingogram were helpful in screening for CUA, while 3D-ultrasound and magnetic resonance imaging were suitable for categorizing CUA accurately.[6]

The pathophysiology of the obstetrical complications in women with MDA was still unclear and unproven. The implantation process of embryo was definitely affected by the endometrial thickness and vasculature and morphology of the endometrial cavity.[7] Kim et al had quoted in their recent systematic review, abnormalities of space in the uterine cavity, arrangement of uterine musculature, and impaired ability to distend are likely to have a negative effect on pregnancy maintenance. In addition, increased muscle mass and decreased connective tissue in the malformed cervix can cause asymmetric uterine cavity pressure, impairing the ability of distention and growth of the uterine cavity, which also leads to late miscarriage and preterm birth.[1] In our first case, the MDA bicornuate uterus, led to malpresentation of the foetus and a cesarean section, as obstetrical complications.

In our second case, the MDA led to retention of placenta post vaginal delivery. As the pathophysiologies of retained placenta were atonicity or abnormal contractility of uterus, a separated placenta that might get entrapped due to closure of cervix prior to placental delivery, placental hypoperfusion disorders and in other spectrum, abnormally invasive placenta (placenta accrete spectrum).[8,9] In our case, postulated mechanism could had been a combination of: abnormal contractility of uterus, premature closure of cervix and placental hypoperfusion (abnormal vasculature of uterus).

Many of MDA cases remained undiagnosed due to absent clinical features and radiological characteristics. But their knowledge and subsequent imaging investigations was important to prevent and manage both gynaecological and obstetrical complications.

CONCLUSION

Mullerian duct anomalies can present as and cause gynaecological and obstetrical complications. Previously undiagnosed women when detected with MDAs should be informed about complications and treatment options (however, optimal management approach cannot be definitely stated), before and during pregnancy, for better maternal and neonatal outcomes.

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