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Case Report

A unique case of successful twin pregnancy reaching term in a patient with uterus bicornis unicollis

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ABSTRACT

Abnormal fusion of the Mullerian ducts or failure of absorption of the septum causes varying degrees of congenital uterine malformation. Twin gestation in a case of bicornuate uterus is rare. We are reporting this case because of its rare presentation, where a case of undiagnosed twins managed to reach full-term after spontaneous conception. We report here a 30 year old gravida four para one with no live issues who presented at term in labour. Per Abdomen examination suggested a twin gestation which was confirmed by ultrasonography. Both the twins were delivered by an emergency caesarean section. Intraoperatively the uterus was found to be bicornuate uterus (bicornis unicollis) with both the babies in two different horns.

Keywords: Twin pregnancy, Uterus bicornis unicollis, Malformation, Mullerian Ducts abnormalities, Uterus abnormalities

INTRODUCTION

Congenital abnormalities of the female genital tract are defined as deviations from the normal anatomy due to abnormal development of the Mullerian and paramesonephric ducts. Congenital abnormalities of the uterus are often asymptomatic and unrecognized; may present as pain during menarche in very few young women or affect a woman's obstetric and/or gynaecological health.

It is difficult to determine the incidence as many women with uterine anomalies go undiagnosed. In one study, uteri of 679 women with a normal reproductive outcome were evaluated and the incidence was determined to be 3.2 percent.¹ However, in women with adverse reproductive outcomes, the incidence can be 5-10 percent in women with recurrent first trimester miscarriages and may approach as high as 25 percent in women with late first trimester/second trimester miscarriages or preterm delivery.² The prevalence rate of uterine anomalies is

4.3% in fertile women and general population, 3.5% in infertile women and 13% in women with recurrent miscarriages. Uterine malformations in order of incidence are: septate uterus (35%), bicornuate uterus (25%), arcuate uterus (20%).³

Uterine anomalies present with variable reproductive outcomes complicated by factors such as recurrent pregnancy loss, preterm labour (46.2 percent), malpresentation, uterine rupture and caesarean section (76.9 percent). However, they don't interfere with implantation and conception with similar pregnancy rates when undergoing IVF as compared to women with normal uteri undergoing IVF.^{4,5}

Spontaneous twin pregnancies in case of uterus bicornis unicollis have been reported rarely. We present an extremely rare occurrence of an undiagnosed spontaneous twin pregnancy in a woman with uterus bicornis unicollis.

CASE REPORT

A thirty year old, gravida 4 para 1 abortion 2 dead 1 (G4P1A2D1) presented to the hospital emergency with complaints of pain abdomen. She had no previous follow-up at our hospital. She gave no relevant history of any other complaints. Her LMP was 9/8/2013 according to which she was thirty weeks and three days by dates. Her previous obstetric history was significant for two early first trimester abortions with one second trimester fetal death. Her last antenatal follow-up was in the early second trimester. She also had an ultrasonography report at 13 weeks and routine blood investigations. Her previous ultrasonography was reported as a single live intrauterine pregnancy with variable lie and presentation. From the previous USG, her gestational age was calculated to be approximately 36 weeks. Previous antenatal investigations revealed a haemoglobin of 8.3 gm%, TLC of 9900 cells/cumm, B positive blood group and seronegative profile. Urine routine microscopy and blood sugars were within normal limits.

On examination, vitals were stable with a pulse of 92 beats/min and BP of 110/80 mm of Hg. Respiratory and cardiovascular examination was normal. Per abdomen examination showed an overdistended uterus with multiple fetal parts on palpation. On Doppler examination, two distinct fetal heart sounds were heard with different heart rates. Two to three uterine contractions were elicited every ten minutes, each lasting five to ten seconds. Per speculum examination showed a healthy cervix and vagina. On per vaginal examination, cervical os was 3 cm. Dilated, 70% effaced with non-cephalic presentation and -2 station.

An urgent sonogram was requested in view of suspicion of a twin pregnancy with non-cephalic presentation. Ultrasound showed intrauterine twin pregnancy of diamniotic monochorionic type with both twins in breech presentation with head of the first fetus in right hypochondrium and that of the second fetus in left hypochondrium (Figure 1-3). The latest scan showed a gestational age corresponding to approximately thirty-two weeks for both fetuses. A complete blood profile was ordered including complete blood count, coagulation profile, renal function and blood group and cross match.



Figure 1: Ultrasound showing the two cornuas in transverse section.

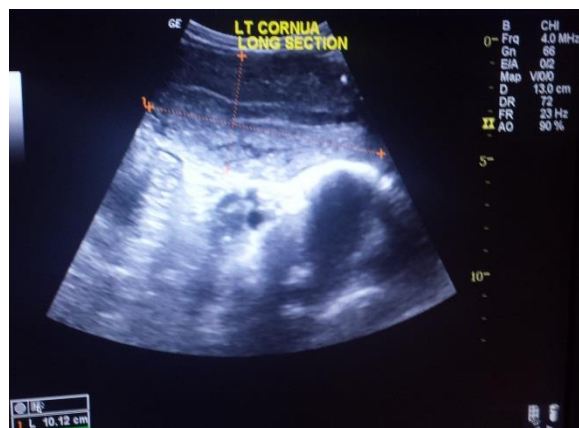


Figure 2: Ultrasound showing the left cornua on longitudinal section.



Figure 3: Ultrasound showing the right cornua on longitudinal section

Patient was taken up for an emergency LSCS. She was induced with spinal anaesthesia and the caesarean section was performed without any complications. She was given one unit of packed red blood cells post-operatively. Intraoperatively findings were as follows: twin pregnancy with bicornuate unicollis uterus. The first baby and the second baby were delivered with shoulder presentation and breech presentation respectively. First baby was a female child of 1.9 kg (APGAR 8/10) and the second baby was a male child of 1.6 kg (APGAR 8/10). Both babies were admitted in the neonatal ICU for observation.

The pregnancy was that of diamniotic dichorionic type. Two separate placentas were removed from two different cornuas (Figure 4). The histopathology report of the placentas showed two vessel umbilical cord in the placenta of the second baby. However, the baby had no congenital anomaly. Patient was discharged on day eight in a stable condition. She was followed up with a post-operative abdomino-pelvic sonogram on day twelve which was reported as bicornuate unicollis uterus with no renal anomalies.



Figure 4: Double placenta in the bicornuate uterus.

DISCUSSION

The embryological development of the uterus is briefly discussed. The epithelium of the uterus develops from the fusion of paramesonephric ducts. The myometrium is derived from the surrounding mesoderm. As the thickness of the myometrium increases, the unfused horizontal parts of the two paramesonephric ducts come to be partially embedded within its substance, and help to form the fundus of the uterus. The cervix can soon be recognised as a separate region. The uterine tubes develop from the unfused parts of the paramesonephric ducts. The original points of invagination of the ducts into the coelomic epithelium remain as the abdominal openings of the tubes. In majority of the cases the presence of an upper genital tract anomaly escapes attention. In some, the detection is made accidentally during investigation of infertility or repeated pregnancy losses, diagnostic D & E procedures, manual removal of placenta or during caesarean section.

Different types of fusion anomalies include septate uterus (septum or partition), arcuate uterus (dip in uterine fundus within the cavity), uterus didelphys (complete lack of fusion of müllerian ducts with a double uterus) and bicornis uterus. Uterine bicornis includes varying degrees of fusion of the muscle walls: uterus bicornis bicollis has two uterine cavities with double cervix with or without vaginal septum. Uterus bicornis unicollis have two uterine cavities with one cervix.

The Müllerian duct anomaly in the case reported was uterus bicornis unicollis (uterus with deep notching to give the appearance of double uterus, but with single cervix). The obstetric significance of these malformations has a direct relationship to the status of muscle mass of the organ. The outcome of pregnancy depends on the capability of the uterine fundus to expand and contract and on the dilating capacity of the cervix.

Obstetric difficulties arise from the abnormal fundus and the abnormal cervix in proportion to their variation from the normal. Cervical cerclage usually improves the fetal survival rate,⁶ but it was not done in our case. The obstetric performance and gestational capacity of a bicornuate uterus is inversely proportional to the degree

of muscular distortion produced by the forking. This results in high incidence of abortion and premature labor. Cases of twin pregnancy in each side of bicornuate uterus are rare. Around 12 cases of twin pregnancy delivering at varying period of gestation with bicornuate uterus have been reported.⁷

There are no specific guidelines for the management of pregnancy in a bicornuate uterus. It is decided according to the specificities of an individual case. The case presented is a rare case of a twin pregnancy in bicornuate uterus resulting from spontaneous conception, where the twins managed to survive till term.

CONCLUSION

Although cases of twin pregnancy in bicornuate uterus have been reported worldwide, further studies need to be undertaken to set specific guidelines for management, worldwide register needs to be maintained to report cases and consistently improve pregnancy outcomes and fetal survival rates.

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