International Journal of Reproduction, Contraception, Obstetrics and Gynecology
Sureddi PK et al. Int J Reprod Contracept Obstet Gynecol. 2021 May;10(5):2125-2129
www.ijrcog.org
pISSN 2320-1770 | eISSN 2320-1789

DOI: https://dx.doi.org/10.18203/2320-1770.ijrcog20211553

Case Report

Successful deliveries in a case of spontaneous pregnancy in 'uterus bicornis unicollis' with dicavitory twin gestation: a rare case report

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Received: 12 March 2021
Accepted: 12 April 2021

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ABSTRACT

Congenital anomalies of the uterus cervix and vagina are caused by alterations in development or fusion of the Mullerian ducts and are associated with multiple obstetric problems. A 32-year-old primigravida presented with spontaneous conception of dichorionic diamniotic twin with one twin in the right horn and another in the left horn of a bicornuate uterus. At 31.4 weeks of gestation the patient presented with preterm premature rupture of membranes followed by preterm labour, and progressed to deliver vaginally, resulting in the successful delivery of a live twin male and female baby. Spontaneous twin pregnancies in case of uterus bicornis unicollis are reported rarely. Here we discuss an extremely rare occurrence of spontaneous dicavitatory twin pregnancy in uterus bicornis unicollis and its outcome.

Keywords: Uterine malformations, Twin gestation, Uterus bicornis unicollis, Dicavitatory twin gestation, Vaginal delivery in twin gestation

INTRODUCTION

Congenital anomalies of the female genital tract involve the uterus, fallopian tubes, cervix and vagina are caused by alterations in organogenesis, fusion or canalization of the Mullerian ducts.

In the fertile group, incidence of Mullerian anomalies was reported as 3-4%, 5–10% in women with recurrent early pregnancy loss, and up to 25% in women with late first or second trimester pregnancy loss or preterm delivery.[1,2]

Further the incidence of twin pregnancies in patients with uterine malformations is estimated at 2.7–3.1%; however, given the limited incidence, studies are small.[3,4]

Twin pregnancy is associated with an increased risk of a range of complications such as infertility, recurrent miscarriage and preterm birth, to variable degrees. Patients with a uterine malformation and a twin pregnancy are at significant risk of preterm birth and low birth weight, prolonged NICU care, respiratory distress.[5,6]

CASE REPORT

A 32 years old primigravida with spontaneous conception of dichorionic diamniotic twin pregnancy with one twin in right horn and another in the left horn of a bicornuate uterus presented in the outpatient department at 12+2 weeks of gestation. She was first diagnosed with bicournate uterus unicollis on her NT NB scan. Her routine antenatal investigations were within normal limits and had no history of medical or surgical high-risk factors. Her first-trimester screening (Dual marker test) indicated a low risk pregnancy. She was regularly followed up in light of the increased risks in DCDA twin pregnancy in a bicornuate uterus, the patient was put on daily oral progesterone and weekly injectable progesterone support. An early malformation scan at 16 + 2 weeks of gestation was performed; along with measuring cervical length by

trans-vaginal scan. While quantitative fetal fibronectin testing allows risk stratification of pregnant women with a shortened/shortening cervix, this was not tested in her. In view of her high-risk pregnancy, decision for a prophylactic cervical encerclage was taken. Prior to the procedure, a high vaginal swab was done to rule out any infection. Procedure was uneventful and was prophylactically started on a 7-day course intravaginal pessaries to prevent infection. The patient was continued on daily progesterone pessaries to reduce the risk of preterm delivery.

Figure 1: Ultrasonography showing dicavitatory gestation in uterus bicornuate unicollis at 8 weeks of gestation.

Figure 2: 3-Dimensional ultrasonography (8 weeks of gestation).

Her subsequent follow up at 26 and 28 weeks demonstrated normal fetal morphology, normal and concordant fetal growth, normal placental blood flow and amniotic fluid levels. At 26.3 weeks of gestation she was given two doses of betamethasone, 24 h apart for fetal lung optimization. During her routine follow up at 28 weeks, she complained of itching all over the body. Her liver function tests and bile acids were done which revealed a picture of obstetric cholestasis. She was started on ursodeoxycholic acid three times a day with an antihistamine for symptomatic relief.

She presented in the emergency department at 31.4 weeks of gestation with preterm premature rupture of membranes (PPROM) with presence of uterine contractions, frank leak was confirmed on vaginal speculum examination.

On per vaginal examination, cervix was 3cm dilated 50% effaced with a first twin on maternal right presenting with vertex pesentation. She was managed as an inpatient for PPROM initially with intravenous antibiotics, rescue dose of betamethasone intramuscular steroid injections for fetal lung maturation and intravenous magnesium sulfate infusion was given for fetal neuroprotection. The cervical knots were cut. A bedside ultrasound was done and cephalic presentation was confirmed for both twins.

Figure 3: Ultrasonography at 12 weeks of gestation.

Figure 4: Ultrasonography at 16 weeks of gestation.

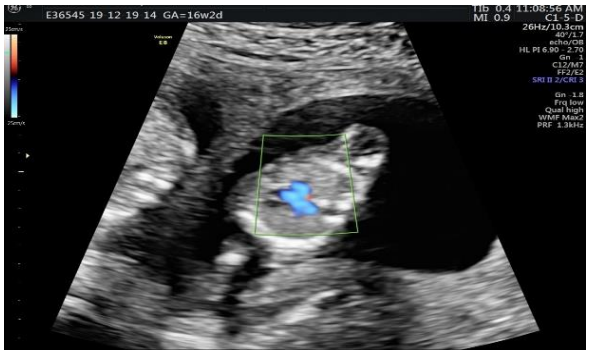

Figure 5: Ultrasonography at 16 weeks of gestation. Doppler flow in one of the fetuses.

Decision to give a trial of labour was taken with strict CTG (cardiotocograph) monitoring; with possible need for emergency caesarean section. Partograph was maintained. Labour progressed, contractions were augmented with low dose oxytocin every half hour. Thereafter twin 1 on maternal right was spontaneously delivered vaginally after 5 hours since onset of leaking, followed by twin 2 after 20 minutes of first twin, and both placentae were delivered with gentle controlled cord traction. Measures for prevention of PPH were taken. APGAR scores of twin 1, male and twin 2, female were 8/10 and 7/10 and weighed 1440 g and 1249 g respectively. Both babies were transferred to NICU in view of preterm delivery. Patient was continued on antibiotics for another five days. Both babies had an uneventful course in the NICU and were discharged on day 10 after establishment of breastfeeding.

DISCUSSION

This case describes a rare presentation of twin pregnancy occupying separate uterine cavities in a woman with known bicornuate unicollis uterus and highlights the challenges involved in antenatal management as well as delivery.

In order to classify these congenital anomalies, The American Society for Reproductive Medicine (ASRM) developed the first classification system for female genitourinary congenital malformations in 1988.[7] A similar and more objective criteria was then developed by the European Society of Human Reproduction.[7]

A systematic review by Chan et al identified that canalization defects, such as septate uteri, were associated with reduced conception rate and increased rate of first-trimester miscarriage when compared with unification defects such as arcuate, bicornuate and didelphys uterus.[8]

There are few studies of women with uterine malformations and twin pregnancies, particularly in the bicornuate uterus.[3] Twin pregnancies individually increase the risk of complications; however, there are limited data to illustrate the effect when coupling twin pregnancy with a uterine malformation.

Bicornuate uterus results from incomplete fusion of 2 Müllerian ducts leading to varying degrees of separation between the cavities. Complete bicornuate uterus has two separate uterine cavities without any communication.

Bicornuate uterus carries an increased risk of miscarriage (1.2 times the risk), preterm delivery (1.9 times) and malpresentation (2 times), when compared with the diagnosis of a didelphys uterus.[8,9] Uterine anomalies have been associated with an increased incidence of spontaneous abortion, malpresentation, placental abruption, preterm delivery, intrauterine growth restriction, and the need for operative delivery.[2,10,11] Various theories which have been used to explain adverse pregnancy outcomes are the following diminished muscle mass, abnormal uterine blood flow, and cervical insufficiency.[12]

As there are few documented cases, the query that arises is how to best manage these patients. Although both vaginal progesterone, pessaries and prophylactic cerclage was done in this patient; The use of vaginal progesterone pessaries and cervical cerclage in twin pregnancy has been shown not to prevent preterm delivery, however, the use of progesterone did not have any adverse outcomes.[3,13,14] Fox et al investigated the use of cervical cerclage in twin pregnancies in the abnormal uterus.[3] Their study did not show any benefit but this may relate to the small sample size.[3]

Various factors that have to be considered regarding the mode of delivery for women with congenital uterine anomalies and these should be discussed antenatally with the patient. Factors such as previous pregnancy mode of delivery, multiple pregnancy, fetal presentation, other pregnancy related complications, pre-existing medical conditions. The reported increased risk of caesarean section delivery is related to increased incidence of fetal malpresentation and fetal distress secondary to labour dystocia, rather than bicornuate uterus itself being an indication for caesarean section and successful vaginal births have been previously reported in cases of anomalous uterine twin gestation.[15,16]

In this case, the patient presented with frank leak and in active labour with a good bishops score, cephalic presentation of both twins and hence it was considered suitable to aim for vaginal delivery for this pregnancy with immediate access to an operation theatre if the situation required operative intervention.

There are several reports of pregnancies in bicornuate uterus. Henry et al and Cruceyra et al among others few have reported dizygotic twin gestation in a septate bicornuate uterus reaching 37-38 weeks gestation and then requiring elective caesarean section.[17,18] Arzu et al reported spontaneous DCDA twin pregnancy in bicornuate unicollis uterus, in which there was spontaneous onset of labour at 35 weeks subsequently requiring emergency cesarean section.[19]

These caesarean sections were addressed as ‘bilateral lower segment caesarean sections’, in order to retrieve babies from different cavities. There is one case report from Kenya by Elias and Amisi in which first twin from one cavity delivered spontaneously in preterm labour and other twin was delivered with caesarean section due to fetal distress.[20]

In our case successful spontaneous vaginal delivery was achieved and subsequent neonatal outcomes were good. Successful vaginal delivery from bicornuate unicollis uterus is being reported in the literature for the first time from this present case report, as per best of our knowledge.

Several studies have also shown that it is possible for dicavitary twin gestations to have independently functioning uteri and cervices, i.e., in didelphys uterus and hence delayed-interval delivery may be possible.[21] In a case reported by Nohara et al with a delivery delay of 10 weeks after emergency caesarean section for fetal distress at extreme prematurity, enabling the second twin to continue to 35 weeks of gestation.[22]

CONCLUSION

Thus, in conclusion, the primary goal is to establish a correct diagnosis of a uterine malformation, as management is dependent on it. Due to the lack of data and paucity of such cases there are no strict clinical guidelines or protocols. Ante-natal management, Management of labour and Mode of delivery must be individualized in each case keeping best interests of mother and baby in mind. This case report also emphasises on the vaginal route for delivery with appropriate monitoring as a confident and successful mode of management and delivery.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Sureddi PK, Dalmia RD, Tambvekar SE, Athavale UV. Successful deliveries in a case of spontaneous pregnancy in 'uterus bicornis unicollis' with dicavitory twin gestation: a rare case report. Int J Reprod Contracept Obstet Gynecol 2021;10:2125-9.