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Successful delivery of spontaneously conceived twins in a single horn of a bicornuate uterus: A case report

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ABSTRACT

Uterine malformations are associated with infertility, recurrent miscarriage and preterm birth, to variable degrees. Accurate classification is crucial to management. A 30-year-old primigravida presented with spontaneous conception of monochorionic diamniotic twins in the right horn of a bicornuate uterus, previously diagnosed as a didelphys uterus. At 28 + 5 weeks of gestation, the patient had preterm prelabour rupture of membranes followed by preterm labour, resulting in the patient undergoing an emergency caesarean section, with the successful delivery of live twin males.

1. Introduction

Uterine malformations occur as a consequence of abnormal organogenesis, fusion or canalization of the Müllerian ducts during fetal development [1,2]. This occurs in approximately 5.5–6.7% of the general population [1,2]. Accurate clinical classification of uterine malformations is imperative, as infertility, recurrent miscarriage and preterm birth are associated with such malformations to variable degrees [1,3]. A pregnancy that originates in one horn of a didelphys uterus has a better outcome than one that originates in one horn of a bicornuate uterus, with studies illustrating an increased prevalence of miscarriage, preterm delivery and malpresentation in the latter [3].

Twin pregnancy is associated with an increased risk of a range of complications [4,5]. Patients with a uterine malformation and a twin pregnancy are at significant risk of preterm birth (<28 weeks) and low birth weight [4,5]. 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 [4,5]. There is little published evidence comparing different classifications of uterine malformations in twin pregnancies, and further investigation is required [5]. We present a rare case of twins in the right horn of a bicornuate uterus, which had originally been diagnosed as a didelphys uterus prior to conception.

2. Case Report

A 30-year-old primigravida with spontaneous conception of monochorionic diamniotic (MCDA) twins in the right hemi-uterus presented to the maternity outpatient department at 14 + 1 weeks of gestation. Aged 23, the patient had been diagnosed with a didelphys uterus after undergoing a dilatation, curettage, hysteroscopy, cystoscopy and vaginal septum removal. This surgery was the consequence of an incidental finding of two cervices and a vaginal septum on routine pap smear. The patient was referred to the maternal fetal medicine department for tertiary care.

In light of the increased risks in MCDA twins (e.g. twin-to-twin transfusion syndrome), the patient underwent monitoring with ultrasound scans every 2–3 weeks and measurements of cervical length. The first-trimester screen indicated a low-risk pregnancy, with a normal morphology scan at 20 + 2 weeks. Ultrasound scan at 24 + 1 weeks revealed a cervix shortened to 19 mm. Fetal fibronectin testing indicated a level of 372 ng/mL. Quantitative fetal fibronectin testing allows risk stratification of pregnant women with a shortened/shortening cervix [6,7]. A level of 372 placed the patient at a 29% chance of delivery within the next 14 days [8]. The patient was commenced on daily progesterone pessaries to reduce the risk of preterm delivery. At 25 + 6 weeks the patient presented to hospital with cramping abdominal pain and scant vaginal blood loss. The patient was admitted and steroid loaded with two doses of betamethasone 11.4mg, 24 h apart for fetal lung optimization. Repeat ultrasound demonstrated a cervical length of 10 mm.

At 28 + 5 weeks, the patient had preterm premature rupture of membranes, with mild irregular contractions. On speculum examination, the cervix appeared closed. The patient was commenced on intravenous benzylpenicillin, magnesium sulphate and received a rescue dose of betamethasone in anticipation of preterm labour. Mild contractions persisted. Repeat speculum examination showed her cervix was dilated 2 cm. Modes of delivery were discussed with the patient with the decision for a caesarean section at 28 + 5 weeks.

A transverse uterine incision was made in the formed lower uterine segment. Two live male babies were delivered. On admission to the

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intensive care nursery, the first twin weighed 1067 g with Apgar scores of 7^1 and 9^5 and the second twin weighed 1156 g with Apgar scores of 5^1 and 9^5. Intraoperative findings identified a single uterus with distinct uterine cavities, separated by a septum to the level of the lower uterine segment. An indentation was identified at the uterine fundus, with non-symmetrical enlargement of the right uterine horn in comparison with the left. A vaginal examination following the caesarean section revealed a single dilated cervix with a thin cervical septum dividing two halves from the external os. It was determined that the uterus was actually a bicornuate uterus with a uterine septum to the level of the lower uterine segment with a separate cervical septum dividing one cervix. The external uterine shape did not resemble a didelphys uterus (as previously diagnosed). The patient recovered well and was discharged 3 days post-operatively.

Discussion

The classification of uterine malformations has long been debated [9]. The American Society for Reproduction Medicine (ASRM) produced the first classification system for female genitourinary congenital malformations in 1988 [9]. In response to the subjective nature of the ASRM classification system, the European Society of Human Reproduction and Embryology-European Society for Gynaecological Endoscopy developed their own criteria in 2012 [9]. Studies have attempted to compare the two classification systems, and have highlighted the importance of accurate diagnosis, in both under- and over-treatment of malformations [9]. Uterine malformations individually and variably affect patients, depending on their classification [3]. The diagnosis of a bicornuate uterus carries an increased risk of miscarriage (1.2 times the risk), preterm delivery (1.9 times the risk) and malpresentation (2 times the risk), when compared with the diagnosis of a didelphys uterus [3,10].

There are few studies of women with uterine malformations and twin pregnancies, particularly in the bicornuate uterus [4]. 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 [4]. Fox et al. [4] investigated this relationship, and showed an increase risk of preterm birth (<28 weeks), lower birth weight and cerclage in twin pregnancy in a malformed uterus compared with twin pregnancy in a normal uterus. However, to our knowledge there are only two case reports involving a twin pregnancy in one half of a bicornuate uterus, as in our case study. Maagard and Langhoff-Roos [11] report a case of spontaneously conceived twins in a single horn of a bicornuate uterus delivered at 26 + 4 weeks via caesarean section due to preterm labour, whilst Narlawar et al. [12] report a case of spontaneously conceived twins in one horn of a bicornuate uterus resulting in spontaneous abortion at 22 weeks.

With few documented cases, the question arises of how best to manage these patients. The use of vaginal progesterone pessaries and cervical cerclage in twin pregnancy has been shown not to prevent preterm delivery [4,13,14]; however, the use of progesterone did not have any adverse outcomes [13]. Furthermore, the effect of pessaries in uterine malformation twin pregnancies has not been investigated. Fox et al. [4] investigated the use of cervical cerclage in twin pregnancies in the abnormal uterus. Their study did not show any benefit but this may relate to the small sample size [4].

In conclusion, it is important that the correct diagnosis of a uterine malformation is made. Incorrect diagnosis may impact significantly on the outcome and on the management of the pregnancy. Ongoing publication of case reports with respect to this topic is needed to provide a larger body of data for what is a rare presentation.

Contributors

Katherine Adams contributed to the conception of the study, acquisition of data, and drafting and revising of the article.

Lee Minuzzo supervised the study, revising of article and gave final approval for the publication of the manuscript.

Both authors approved the final manuscript.

Conflict of Interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

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Patient Consent

The patient provide written informed consent for the publication of the case report.

Provenance and Peer Review

This case report was peer reviewed.

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