Retained second twin secondary to an undiagnosed bicornuate uterus in a poorly supervised labour: A case report

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Abstract

Congenital anomalies of the uterus may result from maldevelopment, abnormal fusion or failure of recanalisation of the paramesonephric (Müllerian) ducts. They are uncommon and are associated with various fertility and pregnancy outcomes. Uterine anomalies have been associated with infertility and pregnancy-related complications. Some cases of successful pregnancies among women with a bicornuate uterus have been reported. However, successful twin pregnancy in a bicornuate uterus is very rare. We report the case of a 24-year-old primigravida who presented with a retained second twin secondary to an undiagnosed bicornuate uterus. An abdominal examination revealed an enlarged abdomen with the uterus tilted to the right and also a palpable firm mass on the left iliac fossa. The retained foetus was presenting cephalic, and the foetal heart rate was 118 bpm. A diagnosis of a retained second twin secondary to a suspected uterine anomaly and suspected foetal distress was made. An emergency caesarean section revealed two horns of the uterus, each having a fallopian tube, an ovary and a cervix. Both cervices opened into one vagina. A 3.0 kg live male baby was successfully delivered through a transverse incision on the lower segment of the right horn of the uterus. The client had an uneventful recovery and was discharged home together with her babies after 4 days. In conclusion, congenital anomalies of the uterus should be considered in cases of a retained second twin. A prompt and accurate diagnosis followed by appropriate management will go a long way in ensuring a good outcome, as was had in this case.

Keywords
Bicornuate, retained, second twin, case report

Introduction

Congenital anomalies of the uterus may result from maldevelopment, abnormal fusion or failure of recanalisation of the paramesonephric (Müllerian) ducts. They are uncommon and are associated with various fertility and pregnancy outcomes. The prevalence of congenital uterine anomalies has been recorded as 5.5% among an unselected population. The arcuate uterus is the commonest uterine anomaly among women in the general population, accounting for 3.9% of cases, while the bicornuate uterus accounts for 2.3%. Uterine anomalies have been associated with infertility and pregnancy-related complications, including recurrent pregnancy losses, preterm labour and delivery, malpresentation, placenta previa, pre-labour rupture of membranes, intrauterine growth restriction, retained placenta and postpartum haemorrhage. Some cases of successful pregnancies among women with a bicornuate uterus have been reported. However, successful twin pregnancy in a bicornuate uterus is very rare. Using the CARE guidelines for case reports, we present the case of spontaneous twin pregnancy that was carried to term in a woman with a bicornuate uterus, who had caesarean delivery for a retained second twin.
A 24-year old housewife who was a primigravida at a gestational age of 38 weeks +2 days presented to the labour ward on account of inability to deliver the placenta of the leading twin as well as the second twin 3 hours after the delivery of the first twin. She had a spontaneous onset of labour and chose to continue to labour at home. The whole labour process lasted for 6 hours, and she was subsequently delivered of her first twin at her home, with the assistance of a neighbour (traditional birth attendant). The baby cried at birth and weighed 2.4 kg. However, there was difficulty in the delivery of the placenta of the leading twin and of the second twin. She was not given any injection following the delivery of the first twin, and there was no obvious associated excessive bleeding per vaginam. Her pregnancy was spontaneous, and the antenatal period had been uneventful. She received antenatal care at a primary health centre, and no ultrasound scan was done during her pregnancy.

A quick examination showed that she looked healthy but was anxious. She was in occasional painful distress of labour. She was anicteric and not pale, and there was pedal oedema up to the mid-leg. Her chest was clinically clear, and her pulse was 84 beats per minute (bpm), full volume and regular, while her blood pressure was 130/80 mmHg. Her heart sounds were normal. The abdomen was enlarged, with the uterus tilted to the right. There was also a firm palpable mass on the left iliac fossa. The foetus was presenting cephalic, in a longitudinal lie and right occipito-anterior position. The foetal heart rate was 118 bpm, strong and regular. A vaginal examination was done during her pregnancy.

There were no palpable contractions noted after 10 minutes of presentation, and the state of the cervix remained unaffected by the ARM. She was quickly transferred to the theatre for an emergency caesarean section under general anaesthesia. Two units of blood were grouped and cross-matched for her. The operation findings revealed a clean peritoneal cavity and bilateral cones of the uterus, with the right uterus gravid separated from a left uterine cavity which was contracted and about twice the size of the patient's fists. Each uterine cone had a normal fallopian tube and an ovary. Each cone opened through the cervix into a common pouch (vagina). A live male baby in cephalic presentation, with Apgar scores of 9/10 and weighing 3.0 kg, and with a posterior-fundal placenta which weighed 400 g were also observed. The client received general anaesthesia, and the abdominal cavity was accessed through a Pfannenstiel incision. A curvilinear incision which was made more to the right side of the right uterine horn was used to deliver the retained second twin, placenta and membranes. The uterine incision was repaired in three layers using Vicryl 2 for the first two layers and Vicryl 2-0 for the serosa. The anterior abdominal wall was repaired in layers using Vicryl 2 for the rectus sheath and Vicryl 2-0 for the subcutaneous tissue and skin.

Postoperatively, she was managed with intravenous antibiotics (augmentin and metronidazole), intravenous fluids (5% dextrose water) and analgesics (pentazocine and piroxicam). After 48 hours, all her intravenous drugs were converted to oral medications. She was also placed on haematinics. The patient had an uneventful recovery and was discharged home with her babies after 4 days. Figures 1–3 show the pictures of the intraoperative findings. She was counselled to present early (immediately after a missed period) in her next pregnancy and was advised to book her pregnancy at a hospital that truly understands her risks.

**Discussion**

A bicornuate uterus occurs when there is an incomplete fusion of the paramesonephric ducts. Following this incomplete fusion, varying degrees of separation between two uterine cavities can occur. This could result in an arcuate uterus, partial bicornuate uterus or complete bicornuate uterus. The case reported had a complete bicornuate uterus in which two uterine horns were divided down to the internal os of the cervix, with no communication between the two uterine cavities (two cervixes present).

A systematic review showed that unification defects, to which the bicornuate uterus belongs, do not appear to reduce fertility but are associated with aberrant outcomes throughout pregnancy. Women with bicornuate and unicorarute uteri have an increased risk of miscarriage, preterm birth and fetal malpresentation, while women with uterus didelphys seem to have only a modestly increased risk of preterm
labour. A retained second twin can also occur as a complication following delivery of the first twin, as seen in the case presented. The case of a retained second twin was reported in a patient who had uterus didelphys. However, the second twin was dead on delivery.

Due to their uncommon occurrence, there is no consensus on the management principles of pregnancies in patients with congenital uterine anomalies. 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. The mode of delivery could be via caesarean section or vaginal delivery.

Challenges to vaginal delivery may include cervical dystocia, malpresentation and possible risk of uterine rupture. Almost all cases of twin pregnancy in a bicornuate uterus reported in literature were delivered through caesarean section. Our patient had a vaginal delivery for the first twin and a caesarean delivery for the second twin. Dystocia might have accounted for the inability of the second cervix to achieve full dilatation. The client lived in a town where home delivery was fashionable, possibly due to the unavailability of many hospitals owned by both the government and the private sector in that region of the state. Poverty and a lack of education may equally contribute to many women delivering their babies at home compared to in hospital. The hospital where this index patient was managed was a mission hospital that was functional within that vicinity. Hospital bills are paid out-of-pocket and are generally high in that region, and this may discourage many women from using hospitals for their deliveries. Despite being a high-risk pregnancy, she received poorly supervised antenatal care in a primary health-care centre and delivered the first twin at home. She risked developing major complications that could lead to maternal morbidity and mortality which did not happen in this index case.

Management should be individualised to have a good outcome. Early-trimester ultrasound especially three-dimensional ultrasonography may be useful in identifying pregnancies co-existing with uterine anomalies for proper follow-up. Where the diagnosis is made before pregnancy and facilities for corrective surgery exist, this would be the preferred option to reduce adverse pregnancy outcomes. Early diagnosis of uterine anomalies also offers obstetricians the opportunity to be ready for eventualities in the management of the pregnant woman and to plan for the best option of delivery. Our patient was counselled to book early with doctors who truly understand her risks, and her management will depend on the findings at pregnancy as discussed previously.

Conclusion

A retained second twin can occur as a complication of a bicornuate uterus in pregnancy, and a high index of suspicion is required for its diagnosis. A prompt and accurate diagnosis followed by appropriate management will go a long way in ensuring a good outcome. There is therefore the need for an early first-trimester ultrasound for every pregnant woman immediately after her missed period for the identification of pregnancies co-existing with uterine anomalies for appropriate follow-up and management.

Acknowledgements

None.
Authors’ contributions
J.T.E., C.A.O. and E.K.C. conceived this study and researched the literature used in this study. J.T.E. wrote the first draft of the paper. C.A.O. and E.K.C. obtained the necessary consent for this study and contributed to the clinical content of this paper. All the authors reviewed, edited and approved the final version of this paper.

Availability of data and materials
There were no major data generated and analysed for this case report. Therefore, data sharing is not applicable to this article.

Conflict of interest
The authors declared no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

Ethical approval
Catholic Maternity Hospital, Moniaya-Ogoja, Cross-River State, does not require ethical approval for reporting individual cases or case series.

Funding
The authors received no financial support for the research, authorship and/or publication of this article.

Informed consent
Written informed consent was obtained from the patient for publication of her anonymised information in this article.

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