Torsion In A Complete Bicorporeal Gravid Uterus: A Case Report

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Abstract

Purpose

Bicorporeal uterus has a prevalence of 0.3% and can have significant implications in pregnancy. We encountered a patient in her third pregnancy with a bicorporeal uterus, having had two previous caesarean sections. This pregnancy was in the left hemi-uteri which was damaged during her previous delivery. We discuss the antenatal and intraoperative considerations undertaken. We also share her antenatal imaging and intraoperative photos of her pelvic anatomy.

Methods

A comprehensive literature search was undertaken on PubMed revealing that uterine torsion in the context of a bicorporeal uterus in pregnancy is a rarely documented occurrence. We used the patient's electronic obstetric notes for all her pregnancies as well as the computerised records of her imaging. Her caesarean section was electively planned and led by two experienced obstetric consultants and a senior registrar. Postnatally she was contacted for permission to publish the images from her scans and caesarean section.

Results

Her previous obstetric history was important and allowed a high index of suspicion for obstetric complications. Prediction of surgical complexities on antenatal screening and imaging was important in her case. Finally anticipation of possible intraoperative complications allowed for pre-operative planning.

Conclusion

This case encompassing a series of learning points and surgical tips which has educational value as well as clinical interest. In the presence of a uterine abnormality, placental complications, extensions of incisions and vascular injury should be anticipated. A safe incision site must be identified before entering the uterine cavity. Therefore involvement of senior clinicians is essential.

Introduction

Congenital anomalies of the genital tract arise from embryonic malformations during development of the mullerian duct. The abnormalities which are prevalent in 0.5-5% of the population arise due to failure of development, fusion, cannulisation or reabsorption of the organs. [1]

According to the ESHRE/ESGE classification system described in 2013 there are 6 classes of uterine malformations, 4 classes of co-existent cervical malformations and 4 classes of co-existent vaginal malformations (Fig. 1).[2] Uterine didelphys, now known as complete bicorporeal uterus has a prevalence of 0.3%.[3]

Clinically, complete bicorporeal uterus has implications in pregnancy including miscarriage, preterm labour and birth, malpresentation in labour resulting in caesarean section, decreased live births compared to a normal uterus. [1]

Uterine torsion is defined as rotation of more than 45 degrees along the long axis of the uterus.[4] It is a rare condition in humans however is commonly described in veterinary studies. It is suggested that the gravid uterus is more predisposed to torsion when compared to non-gravid uterus. Additionally the risk is increased when combined with uterine malformations. [5]

The following case report describes an unusual presentation of a complete bicorporeal uterus (didelphys) found to have uterine torsion during caesarean section. This is an interesting case to discuss the surgical management in this scenario.

Literature Review

A comprehensive literature search was undertaken on PubMed from database inception to May 2021 using the terms 'uterine torsion', 'didelphys torsion', 'bicorporeal torsion'. This revealed 900 results. Of these papers involving animals, torsion of adnexal organs and torsion of fibroids were excluded. Resulting in 172 articles on uterine torsion. 36 articles were excluded as they did not specify a pregnant or gravid uterus. Of the remaining articles only 6 featured gravid bicornuate or didelphys uteri. Two of these articles, from 1986 and 1954 published in the Journal of the Indian Medical Association and Akusherstvo i Ginekologiya respectively could not be sourced.

In review of the existing evidence (Table 1), torsion in the context of a bicorporeal uterus in pregnancy is a rarely documented occurrence. There is limited literature describing the scenario and we intend to undertake the novel endeavour to discuss the surgical implications in this case report.

Table 1
Literature review of torsion of the gravid bicorporeal uterus

Title	Author, Year, Journal	Findings
Uterine torsion and subsequent rupture in a gravid bicornuate uterus associated with an elevated alpha-fetoprotein.[6]	LaHood J et al., 2018, BMJ Case Reports	Case report featuring a patient with intermittent pain, intrauterine growth restriction and a raised alpha-fetoprotein. On emergency laparotomy at 24 weeks gestation she was found to have torsion and rupture of her right uterine horn. Authors concluded that her clinical picture could be explained by placental ischaemia secondary to uterine torsion.
Case report: torsion of a gravid horn of a didelphic uterus.[7]	Achanna S, Monga D, Hassan MS. J 1996, The Journal of Obstetrics and Gynaecology Research	Case report featuring a patient attending with acute abdominal pain after abdominal massage by traditional birth attendants during early labour. Patient had a placental abruption and fetal demise, followed by failure to progress and malpresentation leading to a caesarean section. During the operation uterine atony and post-partum haemorrhage resulted in hysterectomy of the gravid horn leaving the non-gravid horn intact.
Uterine torsion and ischaemia of one horn of a bicornuate uterus: a rare cause of failed second trimester termination of pregnancy. [8]	Oláh KS. 2002, BJOG	Case report of a woman with known bicornuate uterus who had 2 previous pregnancies in the right horn resulting in an elective caesarean section for breech presentation, and a missed miscarriage. At 21 weeks gestation in her 3rd pregnancy she attended with abdominal pain and was found to have a placental abruption with fetal demise. After 5 days of attempted induction of labour failed, she had a laparotomy and was found to have a torsion of the right uterine horn which had distended to the size of a term pregnancy. Resection of the right horn and adenexa was performed.
Torsion of a gravid horn of a bicornuate uterus.[9]	Jain U, et al. 1986, Journal of the Indian Medical Association	Unable to source.
Uterus didelphys. Report of a case with torsion of the uterus occurring during labour. [10]	Legarth J et al., 1982, Acta Obstetrica et Gynecologica Scandinavica	30 year old patient in her first pregnancy underwent a caesarean section due to torsion of the right uterine body blocking the birth canal.

Title	Author, Year, Journal	Findings
Torsion of the uterus didelphys in pregnancy at term[11]	PALII ET. 1954, Akusherstvo i Ginekologiya	Unable to source.

Case History

This patient was a 39 year old white European woman who was in her third pregnancy. She had a complete bicorporeal uterus, two cervices and a vaginal septum (Fig. 2–4).

Her first baby was a male infant born at 40 weeks gestation in May 2014 weighing 3205g. During this pregnancy it was not known that she had a complete bicorporeal uterus. She went into spontaneous labour and had an emergency caesarean section at 9cm dilatation for pathological CTG and abnormal fetal blood sample. She was diagnosed with complete bicorporeal uterus and double cervix at the time of caesarean section and her baby was in the right uterine body. She had a blood loss of 1.5 litres due to uterine atony and extension of the angle of the incision. She required blood transfusion postnatally.

After her first pregnancy she had further investigations with a transvaginal ultrasound scan confirming bicorporeal uterus, double cervix and two normal ovaries, as well as normal renal anatomy.

Her second pregnancy proceeded uneventfully and she opted to try for a vaginal birth after caesarean section. In that pregnancy her fetus was in the right uterine body. She went into labour at 40 weeks gestation in February 2017 and delivered a female infant born weighing 3450g by a Category 1 caesarean section under general anaesthesia for fetal distress at 9cm dilatation with the clinical suspicion of placental abruption. Postpartum haemorrhage of 2.2 litres due to a vaginal tear extending to the posterior wall of the non-gravid (left) uterine body was diagnosed. A vaginal septum was identified during this operation. She was counselled postnatally to have a caesarean section in subsequent pregnancies.

In her third pregnancy the fetus was in the left uterine body. She was started on cyclogest 200mg daily until 34 weeks and had cervical length monitoring. She also had an MRI scan at 34 weeks gestation for placental localisation, demonstrating a high placenta with no evidence of abnormal placental adherence (Fig. 5). Due to the previous uterine tear she was given steroids at 36 + 6 weeks and had an elective caesarean section at 37 + 6 weeks gestation. On entry into the peritoneal cavity, presenting bowel loops as well as one of her ovaries and fallopian tube (Fig. 6) were identified. On exploration it was noted that the uterus was rotated 180 degrees with the posterior aspect of the left uterine body presenting. The left round ligament, fallopian tube and ovary were stretched across the uterus to the right side. The uterus was manually dextrorotated 180 degrees to correct torsion. The bladder was reflected and a transverse lower segment incision was made. A male infant weighing 2895g was delivered in good condition.

Uterotonics were administered and placenta delivered by controlled cord traction. The uterine cavity was explored and found to be singular and empty. The incision was closed in two layers and required one further haemostatic suture to ensure good haemostasis. The right uterus was identified on the right side in pelvis and exteriorised. The right ovary and fallopian tube were visualised and found to be normal. On exteriorisation the left uterus was re-inspected and the incision found to be high in the lower segment, extending to the right (Fig. 7). Her abdomen was then closed and she had a total blood loss of 550ml. Postnatally, she was advised that in a future pregnancy she should have a caesarean section by a senior obstetrician.

Discussion

This is an unusual case encompassing a series of learning points and surgical tips so we believe it has an educational value as well as clinical interest.

Important practice points:

- Previous obstetric history

In this case the obstetric history was important. The knowledge of this patient having uterine didelphys helped increase suspicion of anatomical distortion in the form of uterine torsion. It could be proposed that her previous labours were obstructed by a partially torted uterus, this would be supported by a case report by Sachin et al. which described an asymptomatic torsion of a uterus by 180 degrees.[12]

- Prediction of surgical complexities on antenatal screening and imaging

Uterine torsion is more common in pregnancy where there is an anatomical malformation. Numerous malformations have been associated with uterine torsion including presence of fibroids, abnormal fetal presentation, and uterine malformations. [4][5] Considering the events after the delivery of the second child (vaginal tear extending to posterior wall of left uterine body), and this pregnancy being sited in the left uterine body, antenatal screening and imaging was of paramount importance. This was warranted to identify placental site and rule out morbidly adherent placenta prior to delivery. Imaging also enabled preoperative planning and anticipation of possible complications. This approach of imaging is highly useful in evaluation of patients during preoperative planning. [13]

- Anticipation of possible intraoperative complications

Considering the anatomical alterations caused by torsion, caution must be taken to make the uterine incision in the correct site. In torsion there is the risk of incising the lateral or posterior uterine walls. This could lead to damage to the uterine vasculature and significant post-partum haemorrhage. These risks need to be pre-empted by the presence and supervision of a senior obstetrician and by ensuring a senior gynaecologist is available to attend should a hysterectomy be necessary. This is especially important as

uterine malformations are sometimes associated with abnormalities of the urinary tract, so surgical experience is vital in order to avoid injuries to surrounding organs such as ureters and bladder.[14]

Conclusion

What we have learnt from this case is a high index of suspicion antenatally following suggestive previous obstetric history. Consideration of anatomical distortion in cases of delay in delivery and fetal distress should not be overlooked. In the presence of a uterine abnormality, placental complications, extensions of incisions when operating leading to vascular injury should be anticipated. The need to involve a senior obstetrician early and identification of landmarks, repositioning of torted or malpositioned organs, will result in lower risks of injury to vascular bundles, bladder, and ureters. A safe incision site must be identified before entering the uterine cavity. It is also important to ensure the uterine cavity is emptied completely before closure of the uterine incision. A thorough inspection must be performed before closing the abdomen to look for extensions including posterior uterine wall, ensure bowels are intact especially in the presence of adhesions, and there is optimal uterine tone and haemostasis is secured.

Declarations

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Conflicts of interest/Competing interests The authors declare that they have no conflict of interest.

Availability of data and material ESHRE/ESGE contacted and confirmed that image is open access that does not require specific written permission for use.

Code availability Not applicable

Authors' contributions

Kimme Khan – Manuscript writing, Data collection, Obtaining patient consent

Abdullatif Elfituri – Manuscript writing, Other (taking photographs), Data collection

Christina Legit – Manuscript editing

Stergios K. Doumouchtsis – Manuscript editing

Ethics approval not applicable

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Figures

Figure 1

ESHRE/ESGE Classification of female genital tract congenital anomalies.[2]

Figure 2

Transverse MRI demonstrating left and right uterine bodies demarcated by X

Figure 3

Transverse MRI demonstrating double cervix visible below X

Figure 4

Sagittal MRI demonstrating vaginal septum demarcated by X.

Figure 5

Sagittal MRI at 34 week gestation demonstrating a high placenta, fetus in left uterine body (X) and empty right uterine body (Y)

Figure 6

Entry into peritoneal cavity demonstrating A: Rotated uterus, B + C: Left ovary and round ligament stretched over to the patient's right, D: Bowel in the uterovesical fold, not usually seen during a caesarean section.

Figure 7

Complete Bicorporeal uterus after delivery of baby and placenta and closure of uterine incision.