CASE REPORT

Accidental discovery of Unicornuate uterus during laparotomy for ruptured uterus: A case report

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Abstract

Background: Unicornuate uterus is one of the congenital anomalies that results from hypoplasia or agenesis of the Mullerian ducts. Its incidence is not well established due to varied classification systems. It is commonly diagnosed during evaluation for recurrent pregnancy losses and dysmenorrhoea. Some are diagnosed accidentally during Caesarean sections and laparotomies. This case is unique as she had previous recurrent pregnancy losses and a failed cervical cerclage in her 4th pregnancy. She also had a Caesarean section in this facility in the past but proper diagnosis of her condition had not been made prior to the index pregnancy.

Case presentation We report a unicornuate uterus accidentally diagnosed during laparotomy for a ruptured unicornuate uterus in a 29 year ol3d G7P3+32A at a gestational age of 35weeks and 2days. She had laparotomy with delivery of a fresh still born. There was an oblique rupture in the lower segment of the unicornuate uterus involving part of the previous incision. She had repair of the unicornuate uterus because she insisted on her desire for future reproduction.

Conclusion Clinicians should always consider congenital uterine anomalies as one of the causes of recurrent pregnancy losses especially where cervical cerclage fails. Exhaustive evaluation of patients like this with 3D-ultrasonography and hysterosapingography in the preconception period will help to identify this anomaly. There is need for meticulous intraoperative examination of the pelvic and abdominal organs to forestall missing anomalies such as this.

Key words- Accidental discovery, Unicornuate uterus, Laparotomy, ruptured uterus

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Introduction

The female genital tract originates from the Mullerian duct. This involves a complex interplay of endocrine, genetic and molecular factors.[1]

Unicornuate uterus results from hypoplasia or agenesis of one of the Mullerian ducts. It is subclassified into those with and those without rudimentary horn. The rudimentary horn may be communicating, non-communicating, or with no cavity.[2] Other congenital anomalies of the uterus may result from abnormal fusion, resorption, canalisation of the Mullerian ducts.[3] According to the ASRM (American Society of Reproductive Medicine) unicornuate uterus is a type II Congenital Uterine Anomaly (CUA).[2] The classification scheme is the most commonly used in the past 3 decades and this provides a reliable system for clinicians to group cases.[3]

The true prevalence of CUA is difficult to establish due to lack of a standardised classification system.[4] A systematic review revealed a prevalence of 5.5% among an unselected population.[5] The prevalence appeared to increase in women being evaluated for recurrent miscarriages and infertility.[5] In South West Nigeria the prevalence of CUA in infertile women was 2.3% of which unicornuate uterus accounted for 3.8%.[6]

CUA is commonly diagnosed accidentally during evaluation for miscarriages, infertility or menstrual disorder.[3] There has however been accidental discoveries of unicornuate uterus during caesarean sections.[7,8] A 3-D ultrasound is a common method of diagnosing this anomaly and considered a standard. Magnetic Resonance Imaging is sensitive and specific in delineating endometrium and uterine horn. Combined laparoscopy and hysteroscopy are also considered valuable in the classification and diagnosis of genital tract anomalies.[9]

Unicornuate uterus is associated with a poor reproductive outcome.[3,4] Miscarriages and preterm births are reported complications.[3,10] A ruptured pregnancy located in the rudimentary horn of a unicornuate uterus has been reported.[11] Successful pregnancies have however been reported in women with unicornuate uterus.[12]

We report this case which was managed in Jos University Teaching Hospital (JUTH) to highlight the need to consider the possibility of a unicornuate uterus in women with recurrent pregnancy wastages with failed cervical cerclage.

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Case Report

Mrs. MF, was a 27 year old hausa seamstress who was $G7P3^{+3}$ (2 alive), at 35weeks and 2 days and brought to the delivery suite of JUTH by her husband, with the complaint of generalized continuous abdominal pains of 2 hours duration. Four hours prior to presentation she had intermittent abdominal pain which became continuous 2 hours prior to presentation. She also had vaginal bleeding and dizziness but no fainting spells.

The index pregnancy was booked for antenatal care in our facility at a gestational age of 30 weeks. She did not have cervical cerclage in this pregnancy.

She had miscarriages in her first, second and third pregnancies at gestational ages of 16, 8 and 16 weeks respectively. She had a cervical cerclage inserted in her 4^{th} pregnancy at 16 weeks gestation and had a preterm spontaneous vaginal delivery at 32 weeks following preterm rupture of fetal membranes. The neonate died 2 weeks after delivery.

She also had preterm labour in her 5^{th} and 6^{th} pregnancies at 31 and 30weeks respectively for which she had emergency caesarean section and vaginal birth after caesarean section respectively both in our facility. The babies from the 5^{th} and 6^{th} deliveries were all nursed in the incubator after birth, they are alive and doing well. She did not have cervical cerclage in those pregnancies.

She was neither a known hypertensive nor diabetic. She had no known drug allergies. She had a secondary level of education and was married in a monogamous setting to a 40 year old commercial driver. She neither drank alcohol nor used tobacco in any form.

Examination revealed a young woman that was restless and not febrile with a temperature of 36.2°C and had palor. Cardiovascular system revealed tachycardia of 114 beats per minute and blood pressure of 80/40mmhg. She also had tachypnoea with a respiratory rate of 24 cycles per minute, her breath sounds were vesicular.

The abdomen was uniformly enlarged with generalized tenderness with rebound tenderness. The fetal parts were easily palpable. Fetal heart tones were not heard. There was fresh vaginal bleeding and the cervical os was 3cm dilated and no presenting part was felt.

A working diagnosis of hypovolaemic shock due to rupture scared uterus was made. Intravenous access was secured with a wide bore cannula, blood samples were obtained for urgent packed cell volume which was 20%. Four units of whole blood were grouped and cross matched. Resuscitation was commenced with crystalloids and intranasal oxygen. She was informed of the diagnosis and counseled with her spouse on the options of management-uterine repair with bilateral tubal ligation, subtotal hysterectomy or uterine repair alone. She however opted for uterine repair alone because she expressed her desire for future conception. She was asked to sign an informed consent. She was immediately taken for laparotomy and uterine repair. Intraoperative findings included haemoperitoneum of 2 litres. The placenta and a lifeless fetus were found in the abdominal cavity which were immediately delivered. The still born neonate was male and weighed 2.05kg. A left unicornuate uterus with about 8cm long oblique rupture in the region of the lower uterine segment involving part of the previous caesarean scar and a grossly normal looking left fallopian tube and ovary were seen. There was a right rudimentary hemi-uterus with a grossly normal looking right fallopian tube and ovary. She had a repair of the ruptured uterus with vicyl number 2 sutures. She had a unit of blood transfused intra-operatively and 3 units during the postoperative period. She also had post-operative antibiotics- (Ampicillin-cloxacillin and metronidazole) and analgesics (diclofenac and pentazocine).

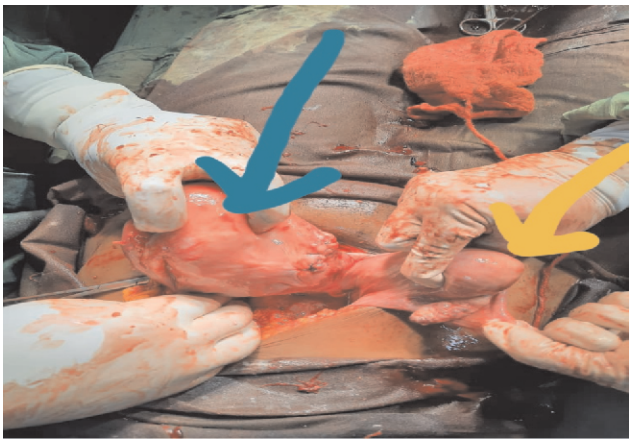

Figure 1- Repaired ruptured left unicornuate uterus (green arrow) and a right rudimentary horn with Fallopian tube and ovary (yellow arrow)

She had an uneventful postoperative course. Her post transfusion packed cell volume was 35% on post-operative day 2. She was counselled on the need for contraception. She opted for implants. She was also informed of the need for excision of the rudimentary horn before the next pregnancy to prevent pregnancy in it which may rupture. She was also informed of the need for early booking in her next pregnancy and admission for bed rest and monitoring from 28 weeks of gestation. She was discharge home on the 5^{th} post-operative day on oral antibiotics and haematinics. She was seen in the out-patient clinic on the 2nd and 30^{th} of June, 2020, she didn't have any complaints. Examination revealed normal vital signs with good operation wound healing. On her clinic visit of June 21, 2020 she was sent to the family planning unit where she was counselled again and had implanon inserted. She was reminded of the need for excision of the rudimentary horn of the unicornuate uterus and the

need for early booking for antenatal care in the next pregnancy. She was told to come when ready for excision of the rudimentary horn.

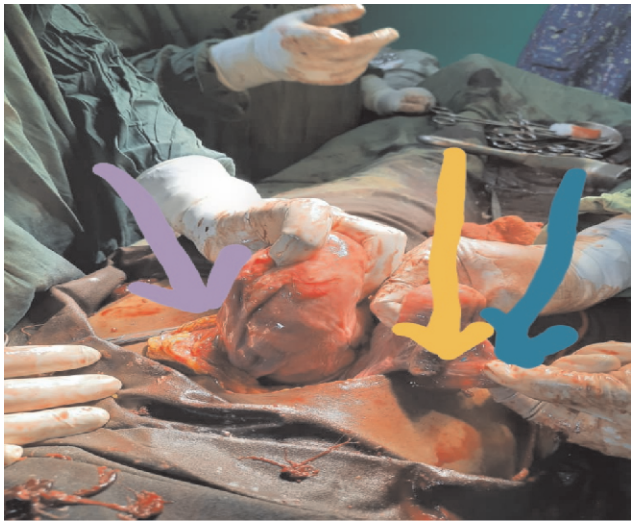

Figure 2: Left unicornuate uterus with Fallopian tube depicted with a gray arrow; a right rudimentary horn with Fallopian tube and ovary depicted with green and purple arrows respectively.

Discussion

Mrs MF had a ruptured unicornuate uterus with a history of previous caesarean section. She was initially thought to have cervical insufficiency because of the history of 3 miscarriages and 3 preterm births. She had a cervical cerclage that eventually failed. The diagnosis of cervical incompetence in this environment is commonly made using clinical features[13], as in the case of this patient. The suspicion of a congenital uterine anomaly was not entertained at any point. She had a caesarean section in the past but the CUA was not discovered intraoperatively. This may be because the uterus was not exteriorised and examined before repair. Unicornuate uterus with a rudimentary horn may present with menstrual problems like haematocolpus, haematosalpinx or dysmenorrhoea.[14] It may also present with miscarriages, preterm births or still births,[4,14] as seen in this patient. Abnormal or absent ovarian or uterine arteries resulting in decreased blood flow to the uterus, and decreased muscle mass in the unicornuate uterus have been suggested as possible mechanisms.[14]

The diagnosis is not commonly made due to the low index of suspicion among clinicians and radiologist.[15] This patient had a non-cavitary rudimentary horn as she never had features of gynacologic problems like dysmenorrhoea, haematometria nor haematosalpinx. Another factor that may have contributed to this missed diagnosis is the fact that she only had 2-D ultrasound scans. In the hand of the inexperienced sonographer diagnosis may be difficult.[16] It has been stated that detection with a 2-D ultrasound is better during the second half of the menstrual cycle when the endometrium of the rudimentary horn will be thicker and hence easily visualised.[16] Diagnostic accuracy is higher with the 3-D ultrasound because it offers the possibility of obtaining reconstructed images which clearly depicts the deviation of the unicornuate uterus and characteristic appearance of the endometrium.[3]

Sato et al reported a case of a ruptured unicornuate scarred uterus during trial of labour.[17] Bodur et al reported unicornuate uterus with rudimentary horn diagnosed for the first time during the third scheduled caesarean section.[8]

Surgery is the main stay of managing uterine rupture generally. The surgery adopted should be the quickest surgery that is lifesaving.[18] This patient had a laparotomy with repair of the ruptured uterus. The rudimentary horn was not excised because she was not stable enough for an additional procedure.. For women with unicornuate uterus, it is recommended that routine laparoscopic excision of the rudimentary horn be done during the non-pregnant state.[3,4] She was told of the need for a bilateral tubal ligation as future pregnancies may be catastrophic but the couple expressed the desire for further reproduction as they had only 2 children alive. There is a high cultural premium placed on child bearing in this environment and it is seen as a stabilizing factor for marriages.[19]

The repair of uterine rupture alone preserves a woman's fertility but leaves her with a scar that has a high risk of repeat rupture in a future pregnancy.[19] This patient was counselled severally on the need for a very close monitoring during her next pregnancy. It is recommended that pregnancy in a unicornuate uterus should be monitored with serial ultrasound for features of intrauterine growth restriction and cervical length measurement to determine onset of preterm labour.[4] It is important to note that despite the poor pregnancy outcomes in women with unicornuate uterus like in this patient, successful pregnancies have been reported.[12]

In conclusion, clinicians should always consider congenital uterine anomalies as one of the causes of recurrent pregnancy losses especially where cervical cerclage fails. Exhaustive evaluation of patients like this with 3D-ultrasonography and hysterosapingography in the preconception period will help to identify this anomaly. Meticulous examination of the pelvis and abdomen is needed intraoperatively to forestall missing similar anomalies and this may include delivering the uterus from the pelvic cavity for proper examination.

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