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Pregnancy and Congenital Uterine Anomalies: Case Series

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Abstract

Background: Congenital Uterine Anomalies are malformations of the Uterus which occur during embryonic life and result from the abnormal formation, fusion or resorption of the Mullerian ducts. Most of them are asymptomatic and diagnosis is done incidentally or during examinations performed for other purposes. We report three cases of women with pregnancies in malformed uteri. **Aim:** To depict the diagnostic challenges and therapeutic aspects of management of pregnancies in congenitally malformed Uteri. **Case Presentation:** The first case was a 22-year-old student who came to consult for a second opinion on the management of an ectopic pregnancy. A 2D Ultrasound done prior revealed an ectopic pregnancy but failed to specify its location in a rudimentary uterine horn. Management with a Multi-dose Methotrexate regimen was initiated but progress was not favorable. She came to us for a second opinion on management. Diagnostic laparoscopy was done and revealed an ectopic pregnancy in a rudimentary uterine horn. This was confirmed by histopathology. Management consisted of resection of the rudimentary horn and a right total salpingectomy. The second case was that of a woman who presented with spotting in early pregnancy. An Obstetric 2D ultrasound done revealed a bicornuate Uterus with a Gestational sac in one horn. She was placed on progesterone supplementation for 2 weeks, with regular antenatal contacts. She had an elective cesarean section at 39 weeks. Surgery revealed a complete Bicornuate Uterus. The post-operative period was uneventful with no complications. The third case was that of a woman with a past history of five successive spontaneous abortions, who presented with spotting at about 8 weeks of gestation. An Obstetric Ultrasound done revealed a Bicornuate Uterus and an embryo in

one cornus. She was admitted, given her poor obstetric history, for about 14 days and placed on progesterone supplementation till 20 weeks of gestation. Antenatal contacts were regular and she had an emergency cesarean section at 36 weeks. There were no complications in the post operative period. **Conclusion:** The high degree of diagnostic accuracy makes 3D ultrasound the diagnostic modality of choice. Nevertheless, 2D and Hysterosalpingography can be used as well. Management of pregnancies in women with congenital Uterine anomalies varies per case as presenting symptoms and outcomes with pregnancies are not alike. When diagnosed out of pregnancy, and depending on the type of anomaly, surgical management may be recommended.

Keywords

Congenital Uterine Anomalies, Diagnosis, Pregnancy, Management

1. Introduction

Congenital uterine anomalies (CUAs) are malformations of the uterus which develop during embryonic life and result from the abnormal formation, fusion or resorption of the mullerian ducts. Congenital abnormalities result primarily from embryological maldevelopment of the paramesonephric ducts and have been associated with pregnancy complications, reduced fertility and other adverse fetal outcomes [1]. Most congenital uterine abnormalities are asymptomatic and are discovered incidentally or during examinations performed for other purposes. In as much as most women with uterine anomalies have a normal reproductive outcome, some experience adverse reproductive outcomes [2]. Some adverse reproductive outcomes include recurrent pregnancy loss, low birth weight, preterm birth, and preterm rupture of membranes, fetal malpresentation, and pregnancy miscarriage. Certain studies reveal that some causes of CUAs could be births with Assisted Reproductive Technologies (ART). The Mullerian ducts form the fallopian tubes, uterus, cervix and upper two-thirds of the vagina. The lower third is formed by the urogenital sinus. Together these make up the female genital tract. The association of pregnancy and uterine malformation is not very frequent and clinical manifestations are non-specific hence diagnosis is difficult in countries with weak technical facilities. It is therefore not unusual to discover a bicornuate uterus or rudimentary uterine horn during a first pregnancy checkup or during a cesarean section or Laparoscopy as we see in the cases below. Ultrasound is necessary for early diagnosis. Management consists of close monitoring, symptom management in pregnancy and Fetal lung maturation if the need arises.

We report the diagnostic challenges and therapeutic aspects in this case series of ectopic pregnancy in a rudimentary uterine horn, a case of pregnancy in a Partial bicornuate Uterus and another in a complete Bicornuate Uterus.

2. Cases

2.1. 1st Case: Ectopic Pregnancy in a Rudimentary Uterine Horn

A 26-year-old female, G2P0010, student, LMP 21/03/2022, Gestational Age: 12 weeks.

She presented in June 2022 for a second opinion on the management of an ectopic pregnancy.

History revealed a month prior to consultation; the patient missed her period and did a home urine pregnancy test which was positive. Onset of spotting prompted consultation and an obstetric ultrasound was requested on 30/05/2022 which revealed a "non-ruptured right adnexal gestational sac of 23 mm (7 weeks 5 days), no visualized embryo". BHCG was done and was at 6941 mUI/ml. This was in favor of an ectopic pregnancy.

She was started on methotrexate 100 mg, multidose protocol which she took on Days 1, 3, 5 and 7 and Folinic acid on days 2, 4, 6 and 8. BHCG done 4 days later was 5647 (18.6% decrease) and 6 days later was at 4226 (39.1% decrease).

Onset of pelvic pain prompted request of a second ultrasound, 2 weeks after the first which revealed 'Right non-ruptured adnexal gestational sac of 70 mm and no visualized embryo'. Despite treatment with methotrexate, the gestational sac was increasing. Faced with this, she came for a second opinion on her management.

Her obstetric past history is relevant for a voluntary abortion in 2019 at 9 weeks by manual uterine aspiration with no complications. She had her first menses at 12 years, bleeds for 4 days with a regular cycle of 28 days.

On systems review there was mild lower abdominal pain.

On physical examination she was conscious, conjunctivae colored, abdomen was flat, speculum examination revealed a macroscopically normal cervix. Vaginal examination, uterus was not enlarged, no palpable adnexal mass or tenderness.

We concluded on a non-ruptured right ectopic pregnancy with failure of medical treatment at 12weeks of gestation.

She was admitted and prepared for operative laparoscopy. Preoperatory workups as well as anesthetic consultation were done.

Per operatory findings were:

- Right non-ruptured enlarged rudimentary uterine horn as seen in **Figure 1**.
- Left adnexae (ovaries and fallopian tubes) macroscopically normal.
- Pelvis free, no adhesions
- No visualized malformations in the urinary system

Procedure: Resection of the rudimentary uterine horn and right total salpingectomy were done as seen in **Figure 2**.

Complications: Uterine perforation with hysterometer around the left uterine insertion of the round ligament as seen in **Figure 3**. There was no bleeding.

Progress: was favorable and patient was discharged on day 2 post operatory.

2.2. 2nd Case: Pregnancy in a Complete Bicornuate Uterus

Mrs. FZ, 34-year-old, married, teacher. She presented to our clinic on the 11th July 2019 to start Antenatal consultations, she had missed her period and had a positive urine pregnancy test. She was G2P0010, with a spontaneous miscarriage in January 2019, for which Manual vacuum aspiration was done in another health facility and patient returned home. There were no further complications.

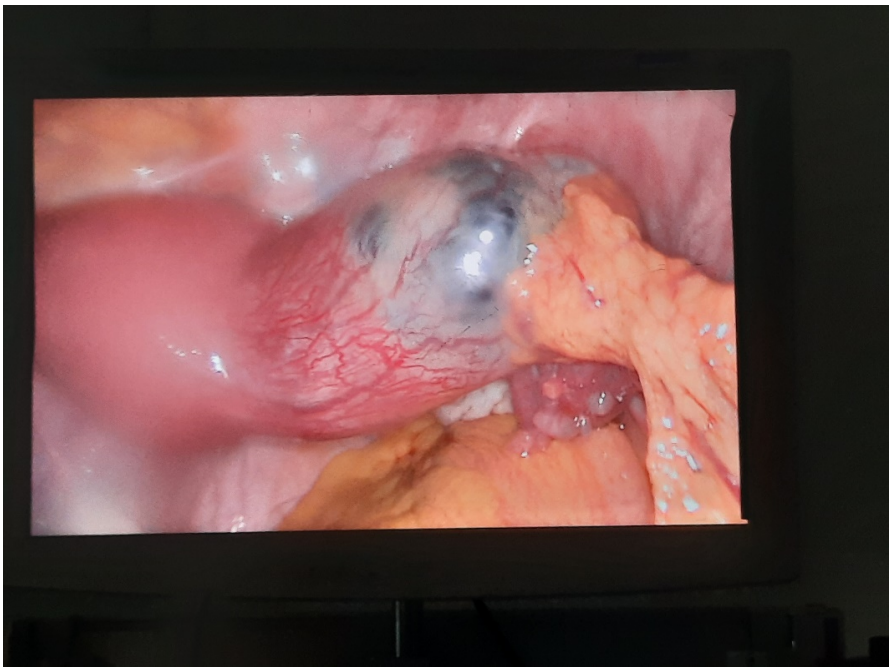

Figure 1. Laparoscopic view of rudimentary horn, hyper vascularized an enlarged.

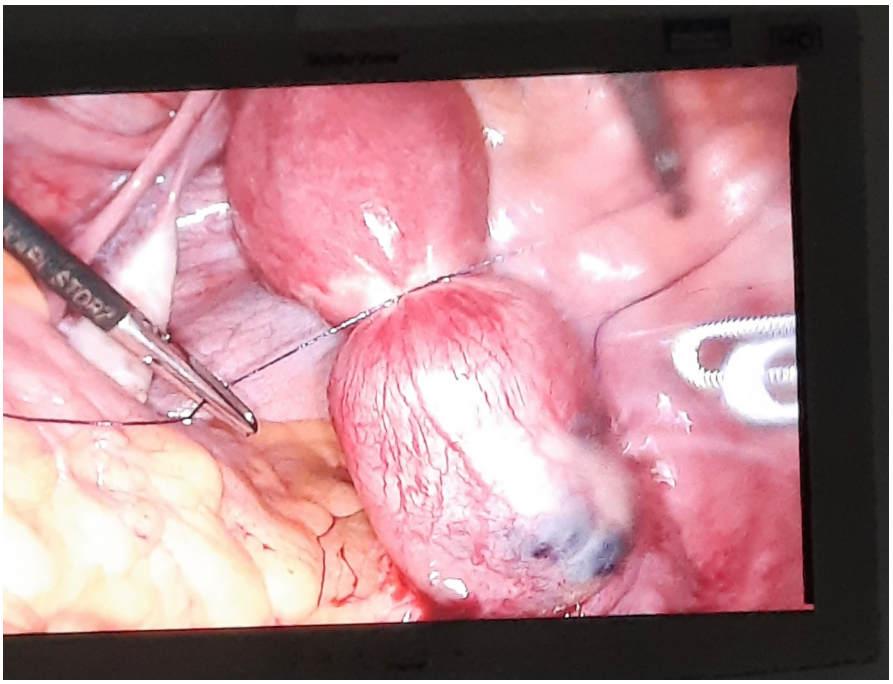

Figure 2. Laparoscopic per operatory view of rudimentary uterine horn.

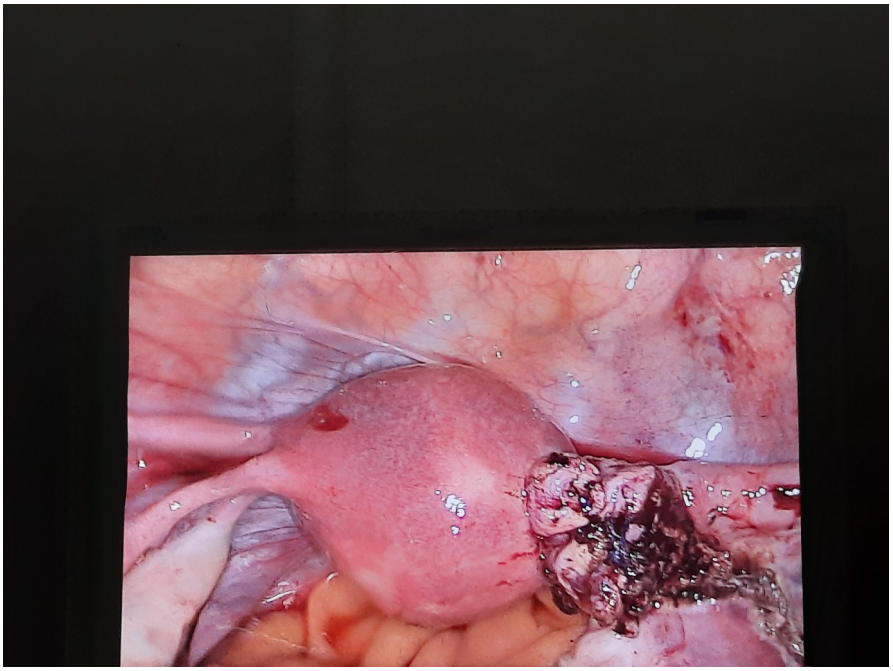

Figure 3. Laparoscopic view after excision of rudimentary uterine horn and right total salpingectomy.

She had no history of dysmenorrhea, sexually transmitted infection or abnormal uterine bleeding. She neither had a past history of surgery nor any other comorbidities. At the first antenatal contact, she complained of mild pelvic pain and spotting.

Physical examination was unremarkable. An ultrasound done revealed a bicornuate uterus. She was placed on progesterone supplementation: 200 mg twelve hourly for 14 days, folic acid 5 mg daily and symptoms subsided. She continued progesterone 200 mg daily till 13 weeks. Antenatal contacts were regular (monthly) and she was supplemented with iron, calcium, intermittent preventive treatment of malaria at the appropriate gestational age. She was scheduled for an elective cesarean section at 39 weeks. C section was carried out and a complete bicornuate uterus was seen. The post operative period was uneventful and there were no complications. The patient was discharged 4 days post operatory and progress was favorable (**Figures 4-6**).

2.3. 3rd Case: Pregnancy in a Partial Bicornuate Uterus

Mrs. LL, 27 years old, midwife, G6P0050, presented in July 2019 with amenorrhea of 8 weeks 2 days, spotting and mild lower abdominal pains. It's worth noting that she's had 5 previous spontaneous miscarriages with increasing gestational ages, which were managed medically (03) cases and manual vacuum aspiration was done (02) cases and progress was favorable with both management methods. She had no medical pathology and had never been operated on before. Blood group rhesus was O positive and Hemoglobin electrophoresis was AA. On physical examination, she was worried, and conjunctivae colored. On speculum

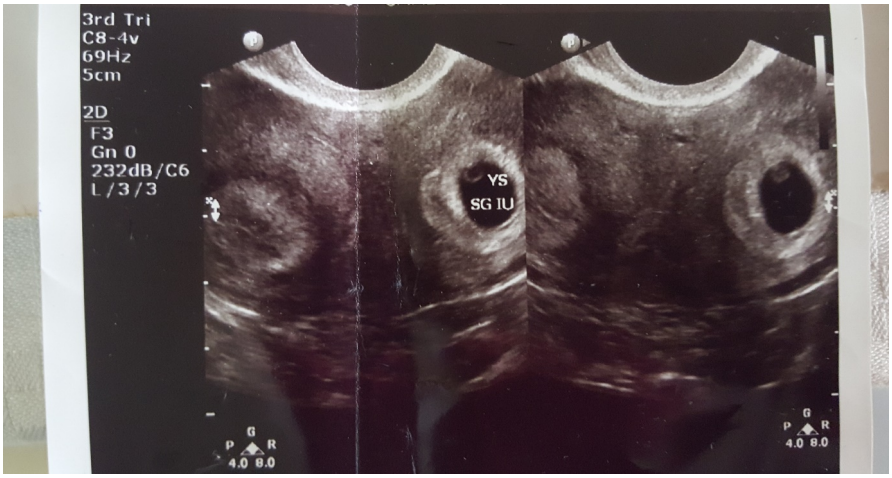

Figure 4. 2D Ultrasound image of a Bicornuate Uterus with an Intra uterine gestational sac in one horn at 5 weeks of gestation.

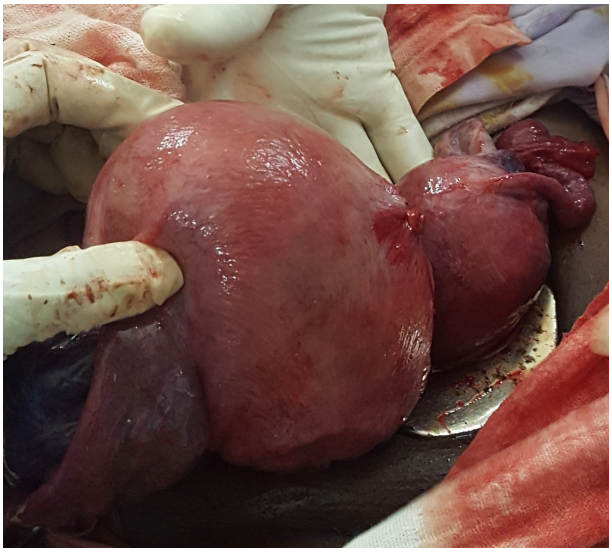

Figure 5. Per Operatory (Cesarean section) image of a Bicornuate uterus. One Cornus is bigger (that which contained the fetus). Fallopian tube and ovaries attached to each cornus.

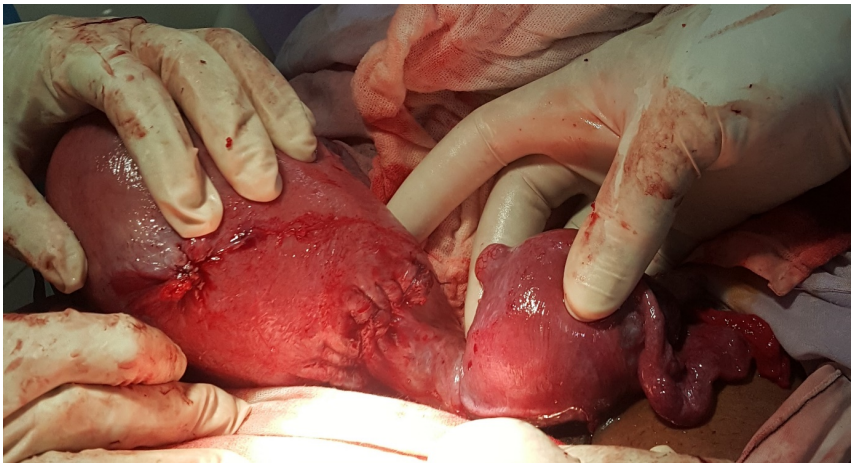

Figure 6. per Operatory image of complete Bicornuate Uterus.

examination, conjunctivae were pink. Her cervix was long, median, and closed, and fingers were soiled with blood. A diagnosis of threatened miscarriage was made. Workup was done (High vaginal swab, Urine culture and sensitivity, both came back sterile, malaria thick film was negative as well as C Reactive protein). An ultrasound done revealed a gravid uterus with a live embryo and gestational sac, fetal heart tones present at 8 weeks 3 days. It also revealed a bicornuate uterus. The embryo was in the left horn as seen below. She was put on bedrest, progesterone ovules 200 mg twice daily and folic acid. She went home, spotting and Lower abdominal pain persisted so she was admitted and put on injectable progesterone (500 mg), 1 ampoule (amp) daily, intramuscular, phloroglucinol daily, 3 ampoules in 500 cc of Glucose, 12 hourly for 72 hrs and bed rest. She was discharged on day 14. She continued injectable progesterone, 1 amp IM/48 hours for 1 week, them 1 amp /72 hours till she was 12 weeks, then she was discharged. She was seen in the hospital 1 week after she was discharged and symptoms had regressed. She then continued IM Progesterone /72 hours till 20 weeks when symptoms resolved completely. She was supplemented with iron, calcium and intermittent preventive treatment at the appropriate gestational ages. Pregnancy evolved normally. At 33 weeks, fetal lung maturation was done when she presented with preterm labour. There was spontaneous loss of liquor at 36 weeks. A cesarean section was done and she was delivered of a life male fetus, Birth weight 2500g, Apgar 8,9 10. Patient was discharged on the 4th day post operatory and there were no complications (**Figures 7-9**).

3. Discussion

Congenital Uterine anomalies (CUAs) are incidental findings. Most congenital uterine abnormalities are asymptomatic and are discovered incidentally or during

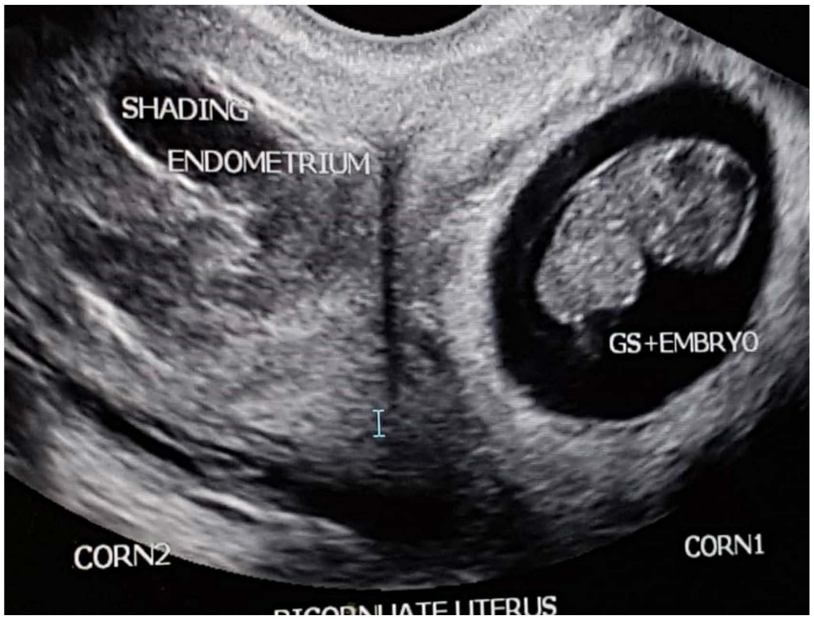

Figure 7. 2D Ultrasound image of bicornuate Uterus with an embryo in the left Horn.

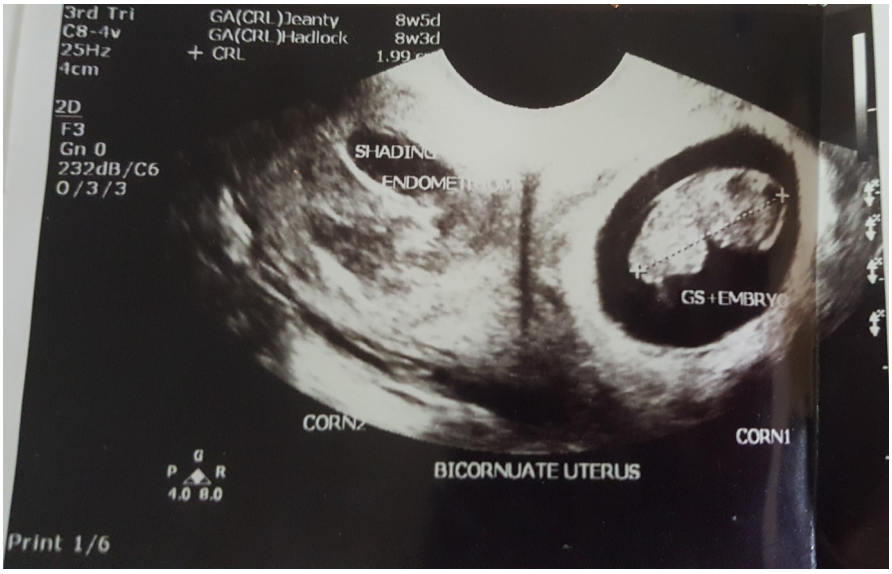

Figure 8. 2D ultrasound image of a Gravid bicornuate Uterus with an embryo at 8weeks 5 days in the left Horn.

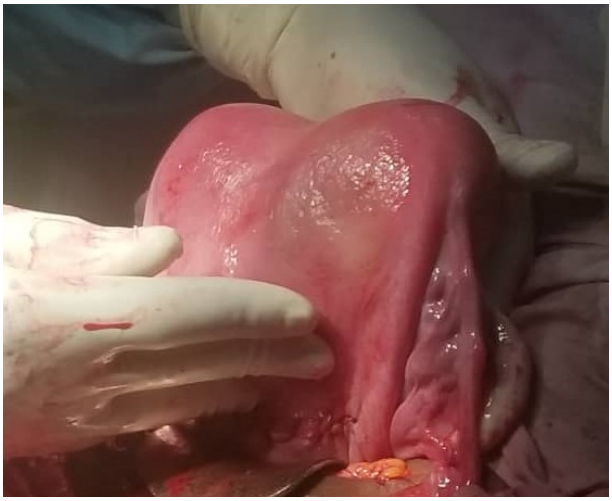

Figure 9. Per cesarean section image of a partial bicornuate Uterus.

examinations performed for other purposes. In the general population, a prevalence of 5.5% of CUAs was reported in a recent systematic review by Chan *et al*, with 8.0% in infertile women, 13.3% in those with a history of miscarriage and 24.5% in those with miscarriage and infertility, and a prevalence rate of 0.2% - 0.6% for Bicornuate uterus (BU) [3]. These ratios vary with others in literature due to assessment of different study populations and the use of different diagnostic techniques. BU arises from an incomplete lateral fusion of the two mullerian ducts at about the 10th week of intrauterine life. At the embryological level, the organogenesis of the genitourinary tract makes it possible to distinguish four phases: the first, urinary phase (3rd, 4th and 5th weeks) comprises the formation of Wolff's ducts and their progression towards the cloaca, the development of ureteral buds in the direction of renal blastemas; the second phase, genital and uri-

nary (6th, 7th, 8th and 9th weeks) involves the completion of the urinary tract by the ascent and rotation of the kidneys. From the 9th week begins the formation of the Müllerian ducts and their progression towards the genital sinus; the third phase (10th, 11th and 12th weeks), genital, involves the joining of the two Müllerian canals; the fourth and last phase is that of resorption of the wall adjoining the Müllerian canals (13th to 17th weeks). The type of malformations is linked to the date of onset of the teratogenic agent during organogenesis: thus, between ten and thirteen weeks, the two Müllerian ducts approach the midline. The anomalies observed are a fusion defect of the two Müllerian ducts, at the origin of the two-horned uteri [4].

There are various classifications for CUAs. The American Fertility Society (AFS) classification (1988) has been the most commonly used. This was later revised by the American society of reproductive medicine (ASRM) due to the limitations of the former. The Bicornuate uterus as in our cases presented were Class IV of the ASRM and the rudimentary uterine horn type IIa. The European Society of Human Reproduction and Embryology (ESHRE) has recently developed a new updated classification system.

Clinically CUAs in pregnancy are asymptomatic or have nonspecific symptoms. In our cases of BU, the patients presented with mild pelvic pain and spotting, symptoms which also occur in normal pregnancies without CUAs. Diagnosis generally is incidental or during an examination performed for another purpose or during a cesarean section for obstructed labour [4] or any other indication. Ultrasound is essential for diagnosis. Accurate diagnosis and correct classification ensure appropriate counselling for women. In the cases of the BU diagnosis was possible as Ultrasound was done early in pregnancy 5 weeks and about 8 weeks of gestation respectively and by experienced medical personnel, who had in mind the occurrence of such pathology. A 2D Ultrasound done revealed an ectopic pregnancy as presented in the 1st case, but failed to specify its location in a rudimentary uterine horn. This could be because it was a 2D ultrasound or because the personnel who carried out the ultrasound didn't think of such a diagnosis. Hence it was missed on regular 2D Ultrasound and diagnosis of the rudimentary uterine horn was made per operatory (Laparoscopy).

Our low index of clinical suspicion owing to the scarcity of data on the epidemiology and clinical presentation of this anomaly in Sub Saharan Africa, coupled with sub-optimal diagnostic testing (two-dimensional ultrasonography) illustrate the frequent diagnostic challenges of congenital uterine anomalies encountered in resource-limited settings as in the case of the Ectopic Pregnancy (EP) in rudimentary uterine horn. Our findings were similar to that of a study published by Dohbit *et al.*, wherein the diagnosis of a Bicornis Bicollis Uterus in a 13-year-old premenarchal non-virgin was missed on 2D ultrasound and made per operatory (exploratory Laparotomy) [5] as well as another study done in rural Kenya [6] where the diagnosis was missed on regular 2D Ultrasound which the pregnant woman did at 22 and 27 weeks of gestation respectively, which were normal. During delivery, faced with retained second twin, an emergency

cesarean section was done and revealed a Bicornuate uterus. It's worth noting that diagnosis of a uterine malformation is difficult with advanced gestational ages and even more so with 2D Ultrasound. In our study, diagnosis was made quite early; 5 weeks and 8 weeks of gestation respectively. Hamidou *et al.* [4] had to carry out an exploratory laparotomy on a 20 weeks pregnancy because a 2D ultrasound carried out revealed an abdominal pregnancy meanwhile it was a Bicornuate Uterus with a viable intra uterine pregnancy. This goes to further depict the difficulties with diagnosis and management of such cases.

Alternatively, patients may be asymptomatic in case the uterine horns are lined with non-functional endometrium. In Other cases of non-communicating rudimentary uterine horn, a cyclical pelvic pain may be non-responsive to analgesia and severe enough to alter the patient's quality of life, as reported by Dohbit *et al.* [5]. Symptoms may be non-specific, leading to late diagnoses and a high incidence of complications. If for example there was a rupture of the rudimentary uterine horn with the ectopic pregnancy it could have caused a massive internal hemorrhage.

Pregnancy in a rudimentary horn of a uterus is a rare clinical condition with a reported incidence of 1 in 100,000 to 140,000 pregnancies [7]. Quamariya *et al.* reported a case of fetal demise at 23 weeks in a gravida 2 para 1 patient. Faced with failed attempts at induction of labour, magnetic resonance imaging was done and the diagnosis of abnormally located pregnancy was made. Patient underwent laparotomy, which found the pregnancy was located in a rudimentary uterine horn and the horn was excised. This is similar to our 1st case where in the patient was diagnosed with an ectopic pregnancy with a 2D Ultrasound, but this ultrasound missed the diagnosis of the rudimentary horn in which this pregnancy was found. She was started on methotrexate multi-dose regiment and faced with failure of medical management of the ectopic pregnancy, she came to us for second opinion and it was per laparoscopy that the diagnosis of a rudimentary horn containing the pregnancy was made. Surgical excision of the horn was done associated with a right total salpingectomy.

Nevertheless, cases have been reported in literature of patients who carried pregnancies till term in rudimentary uterine horns [8]. Only 8% of rudimentary horn pregnancies are diagnosed before symptoms appear [8]. Pregnancy outcomes are usually torsion of horn, abortions, uterine rupture, secondary abdominal pregnancies and very rarely full-term pregnancies. The rudimentary horn musculature and hypertrophic ability are related to the time of rupture [8]. There are definitely still some full-term rudimentary horn pregnancies and they benefit from intently close follow up. In any case, when these abnormalities are diagnosed before pregnancy, it is beneficial to do a resection of the rudimentary horn for fear of menstrual blood retention and obstetric complications [8]. For patients diagnosed during pregnancy, immediate surgical management is recommended as reported in our case.

The key to correctly making the diagnosis and classifying a uterine anomaly is the evaluation of the internal and external contours of the Uterus. With this in

mind, the gold standard test had been combined laparoscopy and Hysteroscopy, although invasive [2]. However 3D ultrasound has now become the gold standard for uterine anomalies due to its high degree of diagnostic accuracy, less invasive nature and considerably less expensive [2]. MRI and combined Laparoscopy and Hysteroscopy are reserved for diagnosing complex mullerian anomalies.

Regarding management, when the diagnosis of Bicornuate Uterus is made at the onset of pregnancy, treatment is generally preventive with prophylaxis done as in all pregnant women, regular follow up, treatment of pathologies if they occur, ultrasound monitoring of Fetal growth, Fetal lung maturation if and when necesary. When diagnosis is made during pregnancy as reported by Hamidou *et al.*, treatment is still preventive. In our cases of pregnancy on a BU, management was preventive. Fetal lung maturation was done in the case of preterm labour but not in the other case. Our management was similar to that of Hamidou *et al.* wherein the patient was seen every two weeks till term and cesarean section done with favorable outcome for both mothers and children.

4. Conclusion

Due to the risks associated with pregnancies in malformed uteri, such as bicornuate or unicornuate with rudimentary uterine horns, and the diagnostic and therapeutic challenges in resource-limited settings, we highlight the need for a high index of suspicion and the use of 3D ultrasound in patients with risk factors like recurrent pregnancy loss, chronic pelvic pain, or failure of regular first line management plans. Encourage documentation and sharing of rare cases such as these.

Informed Consent

Obtained from all three patients for publication of cases and accompanying images.

Authors' Contributions

AAM, ASN, MS, BE: Management of the patients, acquisition of data, manuscript writing and revisions; MP, NNCC manuscript writing and revision. All the authors read and approved the final manuscript.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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