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CASE REPORT

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Ectopic pregnancy in noncommunicating horn of unicornuate uterus: 3D-ultrasound and primary laparoscopic management

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Abstract

Unicornuate uterus with pregnancy in the noncommunicating rudimentary horn is extremely rare. Diagnosis requires awareness, high suspicion index, 3D ultrasound, and MRI. If missed, it can be catastrophic. Treatment varies across literature. We present a case where detection was done by 3D ultrasound and primary laparoscopic surgery done for treatment.

KEYWORDS

3D ultrasound, laparoscopy, rudimentary horn pregnancy, unicornuate uterus

1 | INTRODUCTION

Prevalence of unicornuate uterus is 0.1% in the population, with 74% of unicornuate uterus having a rudimentary horn, which is present due to partial development of one mullerian duct. Rudimentary horn can be communicating or noncommunicating depending on their fusion with the larger horn. When rudimentary horn fuses, it has a communication with the larger uterine horn, and when fusion fails, there is no communication between the rudimentary horn and the larger horn, with 70%-90% of the times, rudimentary horn not communicating with the main horn.[1] Mullerian anomalies lead to multiple problems like infertility, endometriosis, dysmenorrhea, recurrent mis-carriages, preterm birth, fetal growth restriction, placental abnormalities, increase cesarean section rates which affect morbidity and mortality of women.[1-3] Sometimes, unicornuate uterus has been associated with other malformations like musculoskeletal malformations, auditory defects, Hirschprung disease, absent gall bladder,[3]z and VACTERL anomalies.[4]

Diagnosing uterine anomalies early, if possible during adolescents, and later during early pregnancy period will help to reduce morbidity and mortality with uterine rupture being as high as 80%,[5] most uterine horn rupture occurring at 10-15 weeks of gestation.[6,7] There have been reports of pregnancy in rudimentary horn up till term, live [8,9] and with fetal demise [2] but such conditions are extremely rare and should be avoided. If the clinician has an ectopic mind and is suspicious that he's dealing with ectopic pregnancy, he should keep in mind that he may be dealing with a uterine anomaly, else diagnosis may be missed.

Detecting it is of utmost importance, because if not detected in time, pregnancy will progress to a stage where rupture is inevitable, and this will be detrimental for the woman.

We present a case where we detected pregnancy in the rudimentary horn of unicornuate uterus early by 3D vaginal ultrasound. Treatment was primarily surgical with laparoscopy being the mode of surgery.

2 | CASE REPORT

23 years old, married woman, primigravida, walked into the outpatient clinic with right-sided lower abdominal pain for a day. No other positive complaints. She had missed her periods that month. Per abdomen was soft, with mild tenderness on the right lower abdomen. Per speculum examination showed a single normal looking cervix, with no discharge or bleeding. Per vaginal examination, cervix was closed, with mobile uterus, portio-sliding pain absent, and no palpable vaginal septum. A 2D ultrasound was done, which showed an enlarged uterus with thickened endometrium. Extrauterine pregnancy was seen with live embryo of 6 weeks and 4 days, which was completely surrounded by a thick wall. An ectopic pregnancy was suspected, but the thick wall did not look like a fallopian tube. Hence, in 3D vaginal ultrasound, two uterine horns could be separated with both the horns connected only with a small strip of tissue. There was no endometrial connection of the pregnant horn with the cervix, or the main horn (Figure 1). Both ovaries appeared normal. There was no free fluid in the pouch of Douglas. We were expecting her Bhcg to be of normal pregnancy range which was found to be 47371 IU/l.

After thorough counseling of the couple, and preanesthetic checkup and consent, she was posted for explorative laparoscopy and removal of the ectopic pregnancy. After placing all the ports, thorough examination of the abdomen was done, which showed two horns of the uterus, with a noncommunicating horn being vascular, two fallopian tubes arising from each horn, with two normal ovaries. Both the horns were connected with a fibrous band. A schematic diagram illustrates a better understanding of the condition (Figure 2). We were dealing with an unruptured live ectopic pregnancy of rudimentary noncommunicating horn of unicornuate uterus. No free fluid present Rest of the abdomen looked normal (Figure 3).

The fibrous band, along with the base of the pregnant horn cauterised using bipolar, and cut with scissors laparoscopically. Ipsilateral salpingectomy was done. Both the horn and the ipsilateral tube were removed using endobag (Video S1). There was no hemorrhage during surgery. Patient was discharged the next day with advice to follow-up with renal ultrasound. Macroscopically, the uterine horn was about size of 4*4cm, which was dissected. It had a myometrium, and the products of conception well implanted into the endometrium. Histology revealed the ectopic pregnancy in the rudimentary horn, and not in a fallopian tube (Figure 4 A,B).

3 | DISCUSSION

Among mullerian anomalies, unicornuate uterus accounts for 2.4%-13%.[3,6] Pregnancy in rudimentary horn of unicornuate uterus is rare, incidence being 1 in 76.000-1 in 150.000.[5] Here, we report pregnancy in ASRM classification type IIb of unicornuate uterus/ ESHRE-ESGE classification U4a.[10] The pregnancy in noncommunicating horn which has no connection with cervix, or the main horn bears the growing fetus. It is due to transperitoneal migration of sperm to the contralateral rudimentary horn, fertilizing the ova on that side,[1] or migration of fertilized ovum, which probably could have fertilized in the pouch of Douglas.[5] Diagnosis of such a case requires high suspicion index. Diagnosis can be made on 2D ultrasound with accuracy being only 26%.[3] Other reported literature mentions ultrasound sensitivity to be around 29%-33%.[11] It should be supplemented with 3D ultrasound, which improves accuracy rates. MRI also confirms the diagnosis, it is an excellent tool for diagnosing uterine anomalies and any other anomalies associated like urological anomalies, but it is expensive and not available globally under emergency circumstances. It can be done when expert ultrasound imaging is not present.[3] Tsafrir suggested a criteria to diagnose early pregnancy in the rudimentary horn via ultrasound: pseudopattern of assymetrical bicornuate uterus, absent visual continuity between cervical canal and lumen of pregnant horn and presence of myometrial tissue around the gestational sac, hypervascularisation typical of placenta accrete.[12] Similar criteria were proposed by Marvelos and it requires identification of empty uterus with single interstitial portion of fallopian tube, a

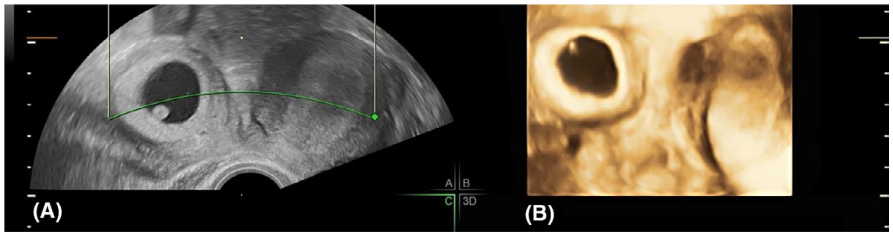

FIGURE 1 A, 2D ultrasound showing transverse view of unicornuate uterus on left side, pregnant rudimentary horn on right side. B, 3D configuration of the unicornuate uterus and pregnant rudimentary horn

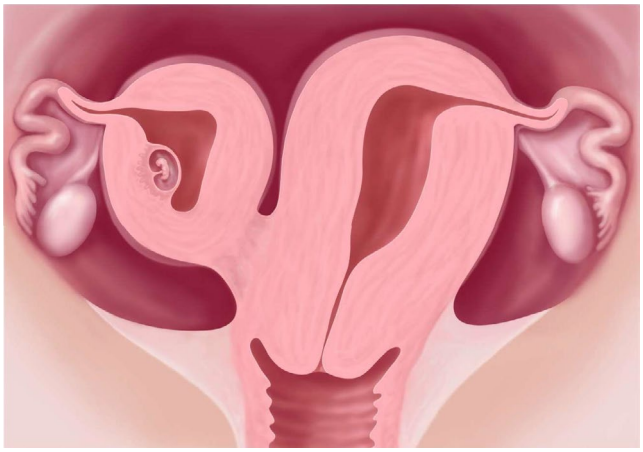

FIGURE 2 Schematic diagram representing unicornuate uterus on left side, pregnant rudimentary horn on the right side with no connection to the cervix, and a small band connecting both

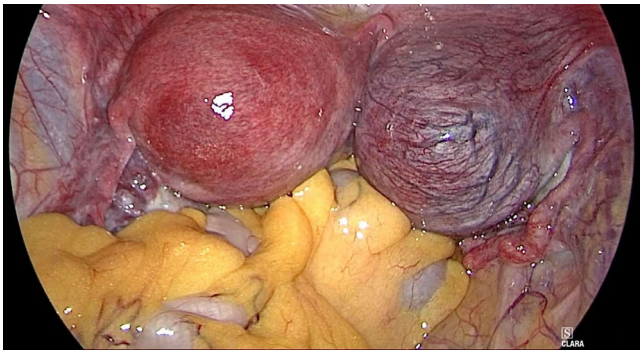

FIGURE 3 Laparoscopic overview of the abdomen, rudimentary pregnant horn on the right, and the unicornuate uterus on the left

gestational sac surrounded by myometrial tissue separate from uterus, and a vascular pedicle connecting the unicornuate uterus to the G-sac.[13] Our patient, was first subjected to 2D USG, pregnancy was confirmed, and when we suspected that we were dealing with a uterine anomaly, 3D configurations were made to confirm the uterine anomaly. There was one cervix, which was communicating with the left uterine horn, the right horn had no communication with the cervix or the left horn. There was a live embryo, and G-sac was surrounded by myometrium. Differential diagnosis of ectopic in fallopian tube and pregnancy in an anomalous uterus should be ruled out, because treatment differs. 3D ultrasound configurations help in differentiating when in doubt. Transverse sections of the normal uterus and anomalous uterus with ectopic pregnancy may look the same, but with 3D configurations, they can be easily differentiated (Figure 1). A table to help differentiate between them by ultrasound has been made (Table 1).

Management will depend upon the hemodynamic condition of the woman, her gestational age. Essentially, removal of uterine horn is the line of management. Earlier days, or in places where access to health care is difficult, when diagnosis is a problem, women often come with rupture of rudimentary horn with unstable hemodynamic condition. Emergency open surgery with multiple blood transfusions is the only option.[14,15] But with advent of better diagnostics, and more women being diagnosed in first trimester scans, medical line of management with surgical removal being done on a later date has become possible.[5] There are no fixed guidelines to manage such ectopics, but like management of other ectopic pregnancy, in early hemodynamically stable pregnancy,

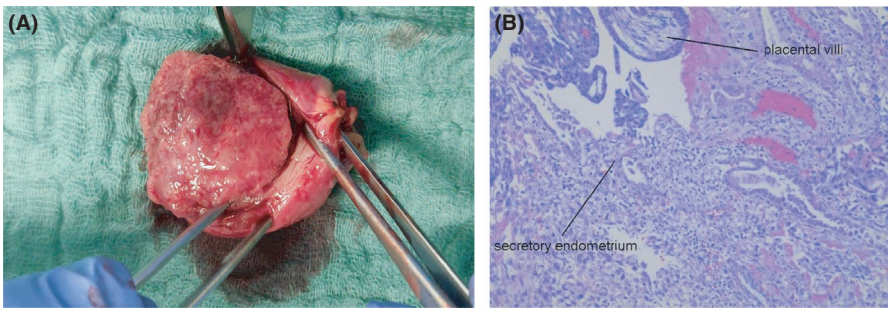

FIGURE 4 A, Macrosopic picture of the excised rudimentary horn with the embryo. B, Histology of the rudimentary horn showing the secretory endometrium with placental implanatation

TABLE 1 Ultrasound features to help differentiate between fallopian tube ectopic pregnancy, bicornuate uterus pregnancy and rudimentary horn pregnancy

USG features	Fallopian tube ectopic pregnancy	Bicornuate uterine pregnancy	Unicornuate uterus with rudimentary horn pregnancy
Two uterine horns	Absent	Present	Present
Ring of myometrium around G-sac	Absent, present in interstitial ectopic pregnancy <5mm	Present	Present
Visual continuity between G-sac and cervix	Absent	Present	Absent

intrauterine or intramuscular methotrexate, or intrauterine KCL can be injected. She is followed up with Bhcg. Once pregnancy completely resolves, she is advised to get the rudimentary horn and ipsilateral fallopian tube removed to prevent further ectopic pregnancy.[5] This method improves operative morbidity and chances of intraoperative hemorrhage but delays definitive management.

There are case reports in literature wherein they have removed the rudimentary pregnant horn successfully without prior medical management[16] similar to ours. We did a primary surgical excision of the uterine horn with live fetus in situ. No complications arose. For direct laparoscopic treatment, if one has fear of excessive bleeding during surgery, reversible occlusion of uterine arteries can be done. It has been done before during cornuostomy for interstitial ectopic pregnancy successfully.[17] In our case, medical line of management did not seem appropriate, because we had a live fetus of 6 weeks 4 days with high Bhcg values (47 371 IU/mL), which could lead to intravenous methotrexate treatment failure. Most literature predicts successful methotrexate treatment with Bhcg values less than 5000 IU/mL.[18] Hence, we recommend primary laparoscopic surgical management, because it removes the chances of methotrexate failure all together, does not depend upon Bhcg values and is a causal treatment and not just a treatment to remove the pregnancy. Complete management with removal of pregnancy and correction of anomaly is done at one shot with complications being almost nil. Our patient and her husband were satisfied with treatment option given and hope to get pregnant again soon.

Reproductive outcomes of ectopic pregnancies after treatment have been studied earlier. For tubal ectopic pregnancies, the cumulative incidence for intrauterine pregnancy was 65.3% for expectant management, 55.3% for methotrexate, and 39.5% for surgery.[19] In cases of tubal surgeries, salpingectomy and salpingostomy have similar post-treatment intrauterine pregnancy rates (56% and 60%, respectively), but salpingostomy has more recurrent ectopic pregnancy rates than salpingectomy (18.7% vs 5.3%).[20] Reproductive outcome after laparoscopic excision of unicornuate horn is similar to reproductive outcomes of mullerian anomalies.[21] Every patient should be screened carefully, past laparoscopic surgical notes should be reviewed and based on patients history and surgical outcome, plan for delivery should be made. Vaginal delivery is possible if patient is carefully selected, monitored, and counseled well.[21,22]

4 | CONCLUSION

Pregnancy in rudimentary horn of unicornuate uterus is not something we see every day in our clinics, requires high suspicion to diagnose, and often increases morbidity of the patient if missed. But with prior knowledge of such cases and an alert mind, this can be detected early with a good ultrasound scan and adequate treatment can be done. We recommend laparoscopic surgical excision of the rudimentary pregnant horn for a one time proper definitive treatment of the patient.

5 | FUTURE PERSPECTIVE

A hypothetical treatment option could be this that product of conception is removed from the rudimentary horn laparoscopically, and the rudimentary horn and the main horn united by Laparoscopic Strassman's Metroplasty. This could increase the uterine cavity area for pregnancy and probably restore better reproductive outcome. But feasibility of surgery, and obstetrical morbidity of the patient will have to be seen and will require great deal of counseling of the couple. Such procedures have been done before for treatment of hematometra and chronic pelvic pain with fairly good outcome.[23]

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CONFLICT OF INTEREST

The authors declare that there is no conflict of interest regarding the publication of this article.

AUTHOR CONTRIBUTION

Author 1: researched the subject, collected data, drafted and wrote and revised the manuscript. Author 2: researched the subject, collected data, and edited the case video and figures. Author 3: main pathologist, involved in the case, provided the histopathological pictures. Author 4, Author 5, and Author 6: researched the subject, collected data, and revised the manuscript. Author 7: treating surgeon, researched the subject, did final editing of the manuscript. All authors have read the final version of the manuscript and approve the same.

ETHICAL APPROVAL

Informed written consent was taken from the patient according to the institutional ethics committee guidelines.

DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no data was generated or analyzed during the current study.

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SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section.

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