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Case report

Uterine rupture of a patient with rudimentary horn pregnancy at 26th gestational weeks

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ABSTRACT

Introduction and importance: Pregnancy of Rudimentary Horn is a type of ectopic pregnancy, that is recognized almost always during surgical treatment of a rupture of the rudimentary horn. This is an obstetric case diagnosed preoperatively by magnetic resonance imaging (MRI).

Case presentation: We report the case of a 19-years-old primigravida patient with rupture of rudimentary horn in 26th gestational week. The patient presented with sudden onset severe abdominal pain in the emergency room. Intraabdominal free fluid is detected. To evaluate the etiology of free fluid and location of the gestational sac, an abdominal magnetic resonance imaging (MRI) scan was planned. The decision of emergent laparotomy is made because of sonographic detection of abdominal excessive -concentrated- free fluid, abdominal tenderness, and 2 points decrease of hemoglobin value in the control hemogram. A rudimentary horn pregnancy and fundal rupture of the rudimentary part of the uterus are diagnosed during the surgical procedure. A baby -live- weighing 450 g was delivered. The ruptured rudimentary horn and same-sided tuba uterina were surgically removed.

Clinical discussion: Rudimentary horn pregnancy is a rare ectopic pregnancy. Diagnosis is difficult clinically, even with diagnostic imaging modalities. Identifying both cornuas systematically in all patients increases the detection rate. The absence of continuity between the gestational sac's lumen and the cervical canal on imaging is an important finding.

Conclusion: Due to the serious maternal and fetal complications, its detection at an early week may be life-saving.

1. Introduction

The unicornuate uterus is formed as a result of failure in the development and elongation of the müllerian duct during embryogenesis. As a result of this defect, it usually results in a functional uterus, normal cervix, and fallopian tube with agenesis on the contralateral side or an abnormal müllerian development such as a rudimentary horn [1].

The majority of patients are asymptomatic. The rudimentary horn may or may not be connected with the normal cavity. It may have functional endometrial cavity [2]. Pregnancy is rare in the rudimentary horn and is a form of ectopic pregnancy with an incidence of between 1/76.000 to 1/140.000 [3]. Despite advances in imaging methods, pregnancy may progress before diagnosis. Pregnancy in rudimentary horns can reach different gestational weeks in patients, depending on the muscular structures of the rudimentary horns. The patient usually applies to the hospital with clinical symptoms of abdominal hemorrhage and acute abdominal pain due to rupture of the uterine wall in the second trimester.

In this case report, a pregnancy that developed in a non-communicating rudimentary horn and resulted in rupture in the late 2nd trimester is presented.

This case report has been reported in line with the SCARE Criteria [4].

2. Case presentation

A 19-year-old patient with gravida 2 abortion 1 para 0 came to routine pregnancy controls and was referred last week - because of smaller femur length according to gestational week- from a peripheral hospital to perinatalogy department for Level 2 Ultrasound Scan. Level 2 Ultrasonography of the patient is reported as no skeletal dysplasia was detected, 45x50mm submucous myoma uteri was detected in the lower

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uterine segment. One week later the patient was applied to the emergency room with complaints of abdominal pain, nausea, and vomiting.

As a result of detection of guarding and rebound tenderness in the abdomen during the abdominal examination of the patient, abdominal sonographic examination was performed by a radiologist (M.N.A.) with 4 years of experience (for the pre-diagnosis of acute appendicitis). Appendix vermiformis was in normal appearance. There was -predominantly in lower quadrants- excessive free fluid in the abdominal cavity. A fetus -compatible with BPD 25 weeks and 2 days- with positive cardiac activity is detected in the gestational sac in the pelvic area. Except for the gestational sac, a left-deviated uterine structure -related to the cervix and vagina- was detected in the pelvic area. There was no connection between the gestational sac and the cervix. To evaluate the etiology of free fluid and location of the gestational sac, an abdominal magnetic resonance imaging (MRI) scan was planned. By the evaluation in comparison with current MRI scan without contrast agent (Fig. 1) and pre-pregnancy (dated 05.03.2020) abdomen MRI scan with contrast agent (Fig. 2); it is understood that there is an anomaly compatible with unicornuate uterus with a rudimentary horn and current 2nd-trimester gestational sac is located in this rudimentary horn (ectopic pregnancy). Findings were evaluated in favor of a rupture of ectopic pregnancy.

The patient had an abdominal tenderness in the abdominal examination, diffuse free fluid in the abdominal cavity. The first hemoglobin value was 11,2 mg/dl, one hour later was 9,1 mg/dl. An emergency laparotomy was performed by a gynecologist (S.S.) with 4 years of experience. During the laparotomy in the abdomen appr. 1000 cc of hemorrhagic fluid and coagulated blood was obtained. It was detected that the pregnancy was placed in the rudimentary horn and was ruptured from the left upper wall of the fundus (Fig. 3A). By performing the hysterectomy on the rudimentary uterus, a live fetus was delivered and given to the pediatrician in the operation room. It was observed that the placenta was completely invaded to the rudimentary horn myometrium, the placenta and its appendages could not be separated from the myometrium (placenta percreta) (Fig. 3B). The diagnosis of placenta percreta is confirmed histopathologically. The right rudimentary horn and its fallopian tube were surgically removed. The ureter of the same side was checked due to the high probability of accompanied urinary tract anomalies. Endometriotic foci -probably due to the retrograde menstruation- were observed on Douglas and peritoneal surfaces.

After controlling for intraabdominal bleeding, the abdominal cavity was irrigated with appr. 1000 cc of warm saline. The operation is completed after the replacement of drainage in the abdomen. The abdominal layers were closed. After the closure of abdomen, the patient was placed in lithotomy position and uterine cavity -that associated with cervix- was evacuated with vacuum curettage. During the operation 1 unit of erythrocyte suspension and 1 unit of fresh frozen plasma were infused to the patient. During the postoperative follow-up, a total of 150 cc sero-hemorrhagic fluid drainage was observed. Due to the low hemoglobin value and symptoms of anemia by the patient, 2 units of erythrocyte suspension and 1 unit of fresh frozen plasma were infused to the patient. On the 2nd postoperative day, drainage was withdrawn. The patient was discharged on the postoperative 3rd day without any complication.

3. Discussion

According to our knowledge; this is the first publication in the literature reporting pre- and post-pregnancy magnetic resonance imaging findings in the rudimentary horn. Unicornuate uterus is associated with appr. 90% non-communicating rudimentary horn [2]. In the uterus with rudimentary horns, the rudimentary part is usually located on the

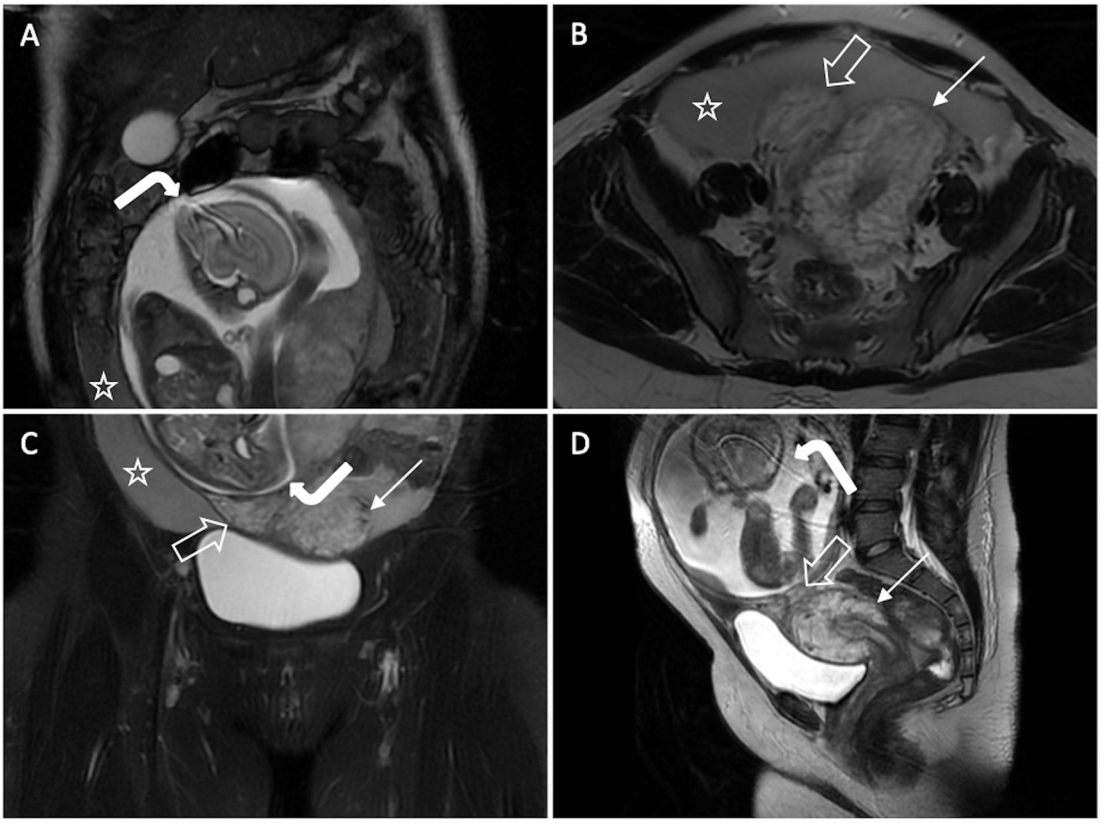

Fig. 1. Coronal (A, C), axial (B), and sagittal (D) T2WI MRI show a large amount of intraperitoneal hemorrhage (stars), a fetus (curved arrows). There was suspected to have a unicornuate uterus that shows a single banana-shaped left uterine horn (thin arrows). There is also a right side pelvic structure located between the uterine fundus and gestational sac (thick open arrows). Note the structure is the same intensity as the myometrium.

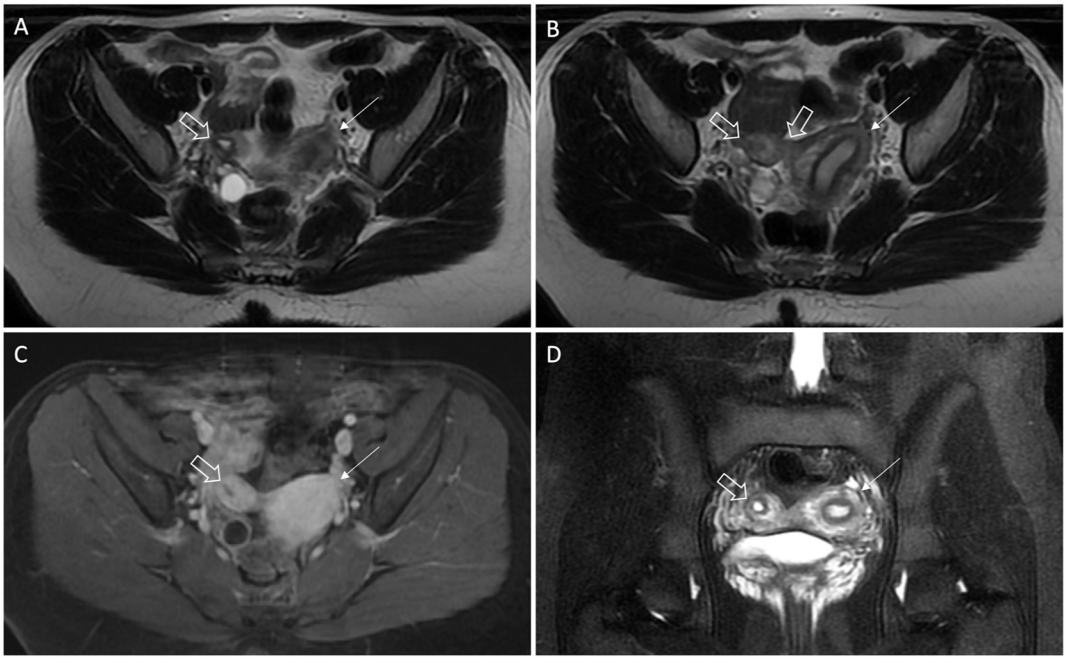

Fig. 2. Axial T2WI MRI (A, B), axial contrast-enhanced FS T1WI MRI (B), and coronal FS T2WI MRI (D) show a normal-appearing left uterine horn (thin arrows) and a right side pelvic structure connected with the myometrium (thick open arrows). This is a rudimentary horn that contains zonal anatomy and endometrium. Axial T1WI C+ FS MRI (C); dominant and rudimentary horns show normal myometrial enhancement.

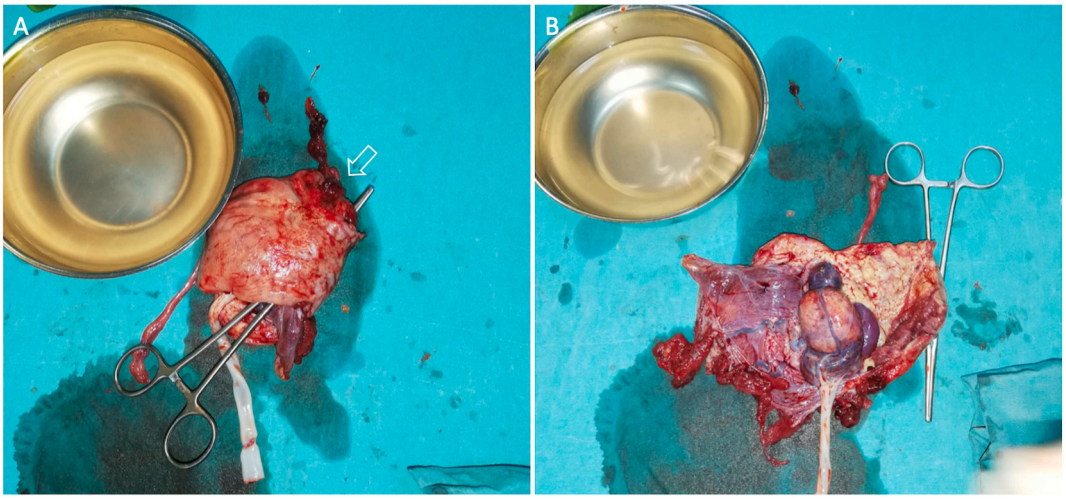

Fig. 3. Macroscopic specimen of ruptured (arrow) rudimentary horn (A) and placenta adhesion anomaly area (B).

right side. The reason for this is that the left müllerian canal moves caudally than right part [5]. In our case too, the rudimentary horn was located on the right. The rudimentary horn, which is not connected to the cavity-containing uterus, can cause retrograde menstruation, causing hemosalpinx and endometriosis from the same-sided fallopian tubes [6]. In our case, endometriotic foci were observed on the pouch of Douglas and peritoneum. The majority of rudimentary horn pregnancy patients were admitted to the hospital -between the gestational weeks of 10th and 20th- with acute abdominal pain as a result of uterine rupture [7]. In our case too, rudimentary horn pregnancy could not be diagnosed during routine follow-up and routine second-trimester ultra-sonoghraphic scan, and the patient was admitted to the hospital with acute abdominal pain at 26th gestational weeks. The rudimentary horn was diagnosed by comparing the pre-operative and pre-pregnancy MRI scans, and the diagnosis was confirmed during the surgical procedure.

The rudimentary horn is hard to identify, especially if not suspected, and may be misdiagnosed as pelvic mass or cervix [8]. When pregnancy develops in a rudimentary horn, the absence of continuity between the gestational sac's lumen and the cervical canal on ultrasound is an important finding [9].

MRI allows accurate classification of unicornuate uterus. At MRI, the small, curved unicornuate uterus is typically displaced off midline. This appearance is named “Banana” configuration. It has normal myometrial zonal anatomy, with normal endometrial-to-myometrial width and ratio. The appearance of the rudimentary horn varies by subtype. If there is no endometrium present, zonal anatomy is absent and the entire horn may demonstrate diffuse low signal intensity. A rudimentary horn without endometrium and the absent rudimentary horn subtype present minimal risk and do not usually require surgical intervention. However, the presence of endometrium in a rudimentary horn is an important finding and should be reported. A non-communicating rudimentary horn with endometrium may manifest as a large uterine mass,

endometriosis, and ectopic pregnancy. If endometrial tissue is present, there may be preserved zonal anatomy. After gadolinium injection; dominant and rudimentary horns show normal myometrial enhancement [10].

In our case, in pre-pregnancy ultrasound and MRI examinations; the rudimentary horn was followed with misdiagnosis as myoma. When we re-evaluated the old MRI images, rudimentary horn and zonal anatomy were clearly observed, especially in the coronal FS T2WI sequence (Fig. 2D). During pregnancy follow-ups, it was not noticed that the gestational sac was not continuous with the cervical canal on ultrasound. Preoperative MRI images revealed that the gestational sac was located separately from the normal uterine cavity (Fig. 1).

During surgery, same-sided salpingectomy -to avoid following ectopic pregnancies-, excising of the rudimentary horn is recommended. Oophorectomy is not recommended. Laparoscopy can be performed in non-ruptured cases [11]. However laparotomy was preferred because our patient was with ruptured-rudimentary horn, was in an advanced gestational week, and was hemodynamically unstable. In addition, this anomaly is highly associated with - mostly same sided- urinary tract abnormalities (incidence found to be 36%) [12]. The incidence of major renal anomalies associated with incomplete uterine duplication with non-communicating rudimentary horn varies between 31% and 100%. The most common anomaly is renal agenesis on the same side with non-communicating rudimentary horn, while the same-sided pelvic kidney is 2nd most common one. In our case, no urinary tract anomalies were found.

4. Conclusion

Rudimentary horn pregnancy is a rare ectopic pregnancy. Diagnosis is difficult clinically, even with diagnostic imaging modalities. Identifying both cornuas systematically in all patients increases the detection rate. The absence of continuity between the gestational sac's lumen and the cervical canal on imaging is an important finding. Due to the serious maternal and fetal complications, its detection at an early week may be life-saving.

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Ethical approval

This is a case report study and ethical approval not required.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Research registration

Not applicable.

Guarantor

Muhsin Nuh Aybay.

CRediT authorship contribution statement

MNA conducted the literature review. SS analyzed and interpreted the patient data for the case presentation. MNA and SS wrote the manuscript together. All authors read and approved the final manuscript.

Declaration of competing interest

The authors of this manuscript declare no relationships with any companies, whose products or services may be related to the subject matter of the article.

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