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Case Series

Spectrum of rudimentary horn pregnancy: a case series

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ABSTRACT

Uterine malformations are the results of abnormal mullerian duct development -fusion, canalization, and septal defects. A unicornuate uterus with a rudimentary horn is one such anomaly of the uterus occurring due to fusion defects. It is associated with a high rate of spontaneous abortion, preterm labor, intrauterine growth retardation, intra-peritoneal haemorrhage, and uterine rupture. We have three cases of rudimentary horn pregnancy of which: the first one was in the first trimester and diagnosed before rupture and managed by excision of the rudimentary horn; the second one was diagnosed after a rupture in the first trimester and managed by surgical excision; and the third one was diagnosed as an obstetric emergency as a second-trimester rupture of rudimentary horn pregnancy. It was managed by initial resuscitation followed by complete excision of the rudimentary horn. Despite advances in ultrasound technology, the antenatal diagnosis of a rudimentary horn pregnancy remains difficult for inexperienced physicians. A high index of clinical suspicion for uterine malformations early in gestation can reduce the mortality rate, along with early intervention. When a rudimentary horn pregnancy is diagnosed, the excision of the horn with ipsilateral salpingectomy is the recommended surgical treatment for the best prognosis. This case series highlights the need for high clinical suspicion of this rare condition.

Keywords: Rudimentary horn pregnancy, Ectopic pregnancy, Laparotomy

INTRODUCTION

Uterine malformations are the results of abnormal mullerian duct development -fusion, canalization, and septal defects.

A unicornuate uterus with a rudimentary horn is one such anomaly of the uterus occurring due to fusion defects.[1] The incidence of Mullerian duct malformations in the general population is estimated to be 4.3% while that of the unicornuate uterus is about 0.4%. Rudimentary horn pregnancy occurs in approximately 1/76,000 to 1/150000 pregnancies.[2]

Conception in the rudimentary horn is very rare and arises either from small communication with the uterine cavity or by transperitoneal migration of the ovum from the contralateral side. The presence of a functional cavity in the contralateral part is the only clinically important factor for complications, such as hematometra or ectopic pregnancy, and treatment (removal) is always recommended even if the horn is communicating. The associated renal anomalies are reported to be as high as 50 to 80%.[1] It is associated with a high rate of spontaneous abortion, preterm labor, intrauterine growth retardation, intraperitoneal haemorrhage, and uterine rupture.[3] Rudimentary horn pregnancy in a non-communicating rudimentary horn is very difficult to diagnose before it ruptures. A criteria for diagnosing pregnancy in rudimentary horn was outlined by Tsafir et al.[4] They are (A) a pseudo pattern of asymmetrical bicornuate uterus, (B) absent visual connective tissue surrounding the gestation sac and the uterine cervix, and (C) the presence of myometrial tissue surrounding the gestation sac. However, only a few cases are diagnosed before rupture and mostly present as an emergency with hemoperitoneum.[4]

We presented a case series of three cases of rudimentary horn pregnancy.

CASE SERIES

Case 1

Pre-rupture diagnosis of rudimentary horn pregnancy

A 28-year-old female G3P2 patient presented at 8.4 weeks gestation and was referred to a tertiary care hospital with a diagnosis of ectopic pregnancy which has been detected during laparoscopic sterilization at a peripheral referral center. She had amenorrhea of 2 months. There is no history of abdominal pain or vaginal blood loss at any time. The patient was gravida 3 para 2 with two previous cesarean sections with the Last childbirth 3 year back. There was no significant intrapartum or postpartum history in a previous pregnancy. She had a normal menstrual period with no history of dysmenorrhea. On admission patient's general condition was good and her vital signs were stable. A physical examination of the abdomen revealed a relaxed and non-tender abdomen. On per vaginal examination revealed uterus 6-8 weeks with cervical motion tenderness. Per abdomen uterus was non-palpable. Transabdominal ultrasound shows a well-defined extrauterine gestational sac with the live fetus of CRL 19.9 mm and 8 weeks 4 days in the left adnexa. Fetal heart rate was 132/min. Fetal movement is seen. She was diagnosed with an ectopic pregnancy in the left adnexa. A complete blood count and coagulation profile were normal. The decision was made for laparotomy. The patient underwent a laparotomy through a Pfannenstiel incision. Intra-operative findings included a normal uterus with a normal ovary and fallopian tube on the right side. The pregnancy was in the rudimentary horn of the left side, with a normal ovary and fallopian tube attached to it. The horn was attached to the uterus at the left corner of the fundus reaching up to cervix. A small incision was made over the pregnant horn and a dead fetus with unidentified sex was delivered followed by excision of the rudimentary horn. The post-operative period was uneventful and the patient was discharged on the 5th day. Histo-pathological diagnosis confirmed the diagnosis.

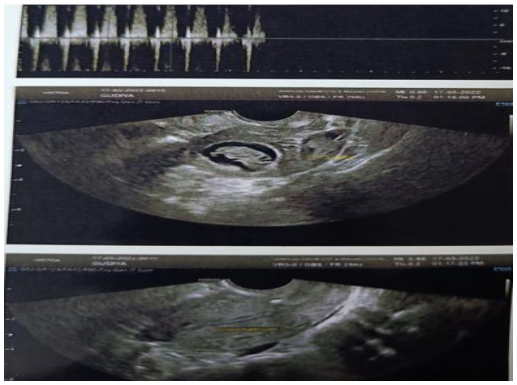

Figure 1: Ultrasound showing empty uterine cavity and extrauterine fetus in left side.

Figure 2: Showing fetus.

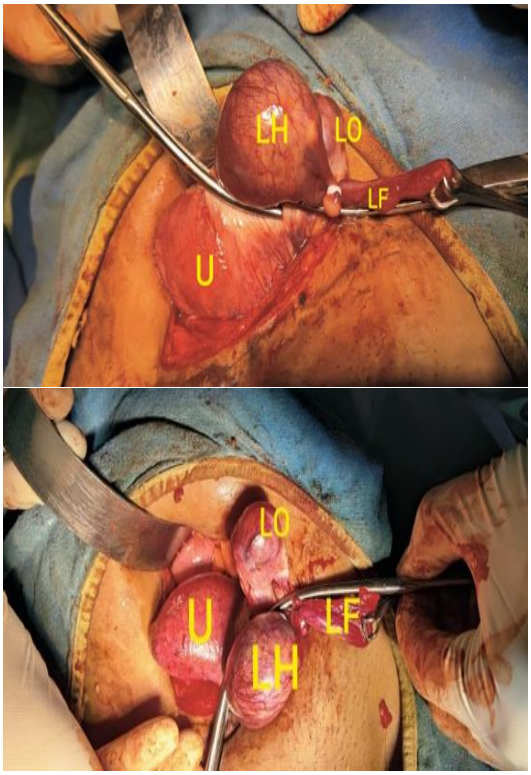

Figure 3: Unicornuate uterus with a non-communicating rudimentary horn in the left side.

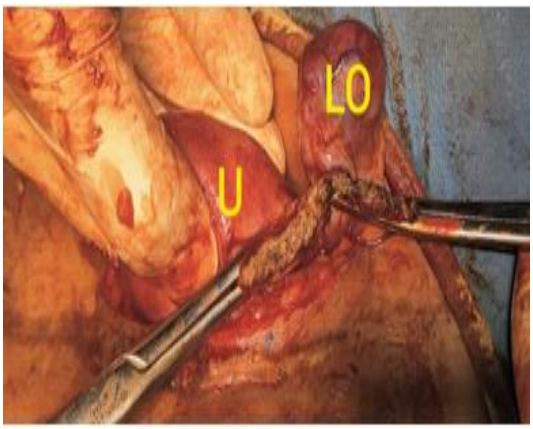

Figure 4: Cut end after excision of left rudimentary horn.

Case 2

First-trimester rupture of rudimentary horn pregnancy with secondary abdominal pregnancy

A 33-year-old female G6P5 patient presented at 12.3 weeks gestation and was referred from the peripheral hospital in view of a ruptured ectopic. She had three months of amenorrhoea and was complaining of pain abdomen for the last 24 hours. The patient was gravida 6, parity 5 with all normally delivered babies. Her last childbirth was 1.5 years back. She had no significant intrapartum and postpartum history with a normal menstrual period. On admission, the patient's general condition was poor with unstable vitals. On physical examination of the abdomen revealed a soft but tender abdomen in the right side mainly the flanks. On per vaginal examination, the uterus seems of 12 weeks with cervical motion tenderness. Ultrasound shows a single, extrauterine fetus of 11 weeks 4 days without cardiac activity and moderate free fluid in the peritoneal cavity possibly due to a unicornuate uterus with ruptured ectopic pregnancy in rudimentary horn. Her hemoglobin was 6.8 with a normal coagulation profile. The decision of laparotomy was taken. Two units of PCV were transfused intra-operatively. Patient underwent laparotomy by Pfannenstiel incision. The intra-operative finding included a normal uterus with a normal ovary and fallopian tube on the right side; ruptured ectopic in the rudimentary horn of the left side with a normal ovary and fallopian tube attached to it; there was a hemoperitoneum of 500 ml which was suctioned out; embryo was found in the peritoneal cavity and was extracted out; the left horn was connected to the uterus just above the cervix by a thick fibrous band. Horn was excised along with the right fallopian tube. Left-sided tubal ligation was done. Post-operative patient was stable and discharged on 7th day.

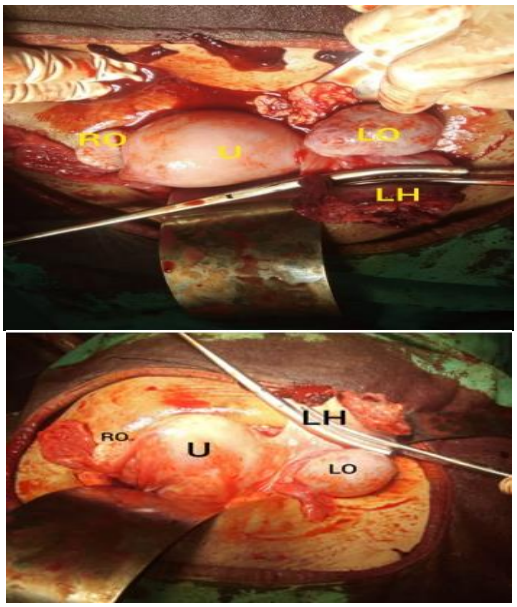

Figure 5: Left ruptured rudimentary horn pregnancy.

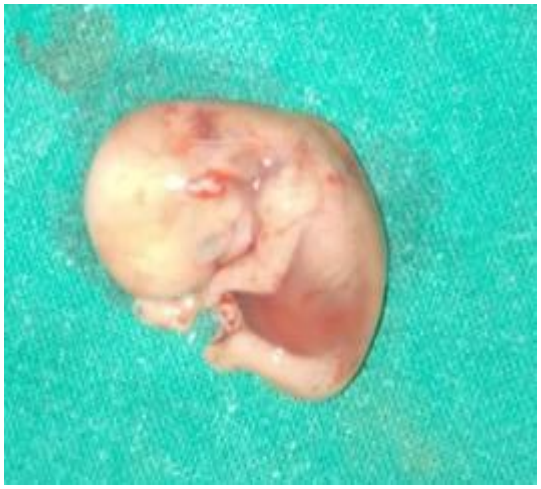

Figure 6: Fetus.

Case 3

Ruptured rudimentary horn pregnancy in the second trimester- an acute obstetric event

A 22-year-old primi patient was referred from the peripheral center in view of ectopic pregnancy. She had five months of amenorrhea and complained of acute pain in the lower abdomen.

On examination general condition was poor, with blood pressure 80/50 mmHg and pulse rate 120/min with severe pallor. Per abdomen examination it seems of 22 weeks period of gestation with tenderness in the right flank region.

Investigation

Hemoglobin 6 g/dl with normal coagulation profile. She had an old ultrasound report showing single live intra-uterine pregnancy of 14 weeks.

Now bedside urgent ultrasound was done which reveal a gestational sac of 17-18 weeks in the right side of the lower abdomen with absent cardiac activity and hemoperitoneum. Thus the decision of laparotomy was taken.

Intra-operative two-unit PCV was transfused. Patient underwent laparotomy by pfannenstiel incision. Intra-operative findings included- around 600 ml hemoperitoneum present, the baby was lying in the abdominal cavity which was delivered, unicornuate uterus present with rudimentary horn towards the right side, there is a rupture of the posterior wall of the right rudimentary horn leading to secondary abdominal ectopic pregnancy.

Rudimentary horn excised and hemostasis achieved.

Post-operative patient was stable and discharged on 6th day.

Figure 7: Fetus delivered from abdominal cavity.

Figure 8: Ruptured right rudimentary horn pregnancy from the posterior wall.

DISCUSSION

Uterine anomalies result from the failure of complete fusion of the Müllerian ducts during embryogenesis. The incidence in the general population is estimated to be 4.3%.[1] A unicornuate uterus with a rudimentary horn is the rarest anomaly and results from the failure of one of the Müllerian ducts to develop completely and an incomplete fusion with the contralateral side.[5] The incidence of this anomaly is approximately 0.4%.[1]

In the majority (83%) of cases, the rudimentary horn is non-communicating.[5] The anatomical variations of a rudimentary horn serve as the basis for the classification of a unicornuate uterus by the American Society of Reproductive Medicine (ASRM).

Acién et al performed a systematic review to analyze the classification systems for uterine anomalies and concluded that an embryological clinical classification system seemed to be the most appropriate.[7] This paper presents a case from class 1, and would be classified as class IIB according to the ASRM (Table 1).

Table 1: Embryological-clinical classification for female genito-urinary malformations.

Unilateral genito-urinary agenesis or hypoplasia
Cases of unicornuate uterus with contralateral renal agenesis due to agenesis or hypoplasia of an entire urogential ridge.
Uterine duplicity (bicornuate or didelphys uterus) with a blind hemivagina (or unilateral cervicovaginal atresia) and ipsilateral renal agenesis
This includes the Herlyn-Werner-Wunderlich syndrome and there can also be cases of resorption partial of the intervaginal septum.
Isolated or common uterine or uterovaginal anomalies
This includes the anomalies in the Müllerian development processes (also included in the classification of the American Fertility Society) without other associated anomalies; and also the transverse vaginal septum.
Accessory uterine masses with an otherwise normal uterus, and other possible gubernaculum dysfunctions
Anomalies of the urogenital sinus
These include cases of imperforated hymen, vesicovaginal fistulas, persistent urogenital sinus, cloacal anomalies, and other external gastrointestinal or urinary anomalies.
Malformative combinations

The reported incidence is 1 in 100,000 to 140,000 pregnancies.[2] The most accepted explanation is the transperitoneal migration of the sperm cells or a fertilised ovum.[1,5] This explanation was supported by the observation of the corpus luteum in the contralateral ovary. It is extremely uncommon for such cases to result in a viable baby. These cases usually result in the rupture of the horn in the second or third trimester. Only 10% of cases such as these reach term, and the fetal salvage rate is only 2%.[8] The rupture occurs because of the underdevelopment of the myometrium and a dysfunctional endometrium.[8] A rudimentary horn pregnancy can be further complicated by placenta percreta due to the poorly developed musculature and the small size of the horn; the reported incidence is 11.9%.[8] The key to diagnosis prior to the rupture is a high index of clinical suspicion. A history of severe dysmenorrhoea may be a clue for diagnosis. However, the rudimentary horn may be underdeveloped and its endometrium non-functional, so dysmenorrhoea may be absent. A careful pelvic examination in the first trimester showing a deviated uterus with a palpable adnexal mass should provoke suspicion of a Müllerian anomaly. It can be confirmed by an ultrasound or MRI. Tsafrir et al suggested the following criteria for diagnosing pregnancy in the rudimentary horn: (1) a pseudo pattern of the asymmetrical bicornuate uterus; (2) absent visual continuity between the cervical canal and the lumen of the pregnant horn, and (3) the presence of myometrial tissue surrounding the gestational sac.[1,4,5] Ultrasound sensitivity remains only 26%.[8] The enlarging horn with the thinned

myometrium can obscure the adjacent anatomical structures and the sensitivity further decreases as the gestation progresses. MRI has proven to be a very useful diagnostic tool.[4] The differential diagnosis includes a tubal, corneal, or intrauterine pregnancy in a bicornuate uterus. Ultrasonographical features may help to reach a diagnosis, as in the following examples. A tubal pregnancy will not show a ring of the myometrium surrounding the gestational sac. A variation in the thickness of the myometrium in two horns and a marked distance between them favour the diagnosis of a rudimentary horn pregnancy. The continuity between the endometrium lining the gestational sac and the other uterine horn is typical for a pregnancy in a bicornuate uterus.[4] It is recommended by most that immediate surgery be performed whenever a diagnosis of pregnancy in a rudimentary horn is made even if unruptured.[9] However, conservative management until viability is achieved has been advocated in very select cases with larger myometrial mass, if emergency surgery can be performed anytime and the patient is well-informed.[10] Pregnancy in a rudimentary horn carries a grave risk to the mother. There is a need for increased awareness of this rare condition and to have a high index of suspicion, especially in developing countries where the possibility of early detection before rupture is unlikely the first case: laparotomy was done in view of ectopic pregnancy which turned out to be a rudimentary horn ectopic pregnancy intraoperative; second case: patient landed up into shock and bedside USG shows rudimentary horn pregnancy; third case: patient initially have pregnancy in the rudimentary horn which leads to rupture and presented as secondary abdominal pregnancy.

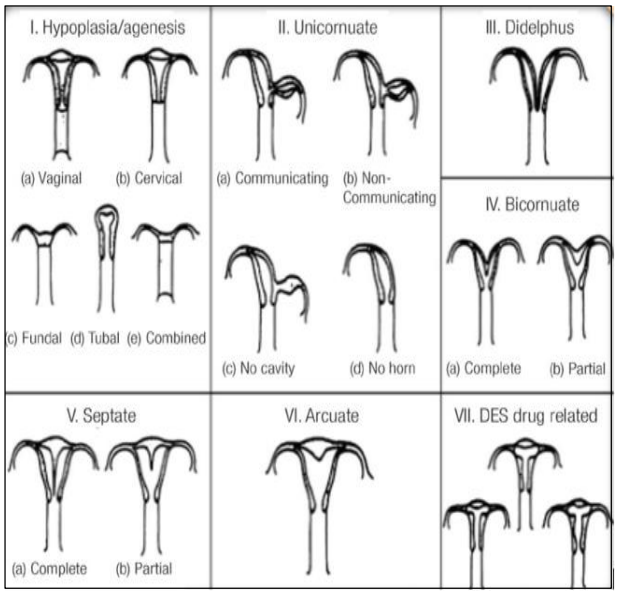

Figure 9: Embryological-clinical classification for female genito-urinary malformations.

Immediate surgery is recommended whenever a diagnosis of a pregnancy in the rudimentary horn is made. The traditional treatment is a laparotomy and the surgical removal of the pregnant horn to prevent rupture and recurrent rudimentary horn pregnancies. In recent years, several cases have been treated successfully by laparoscopies using various techniques.[12] Some authors have described systemic methotrexate administration or feticide with intracardiac potassium chloride as alternatives or adjuncts to surgery in early gestation.[12] Conservative management, until viability is established, has been advocated in selected cases with large myometrial masses. Emergency surgery can be performed at any time. In all such cases, the patient should be informed of the risks of the condition as well as their management options.

CONCLUSION

Despite advances in ultrasound technology, the antenatal diagnosis of a rudimentary horn pregnancy remains difficult for inexperienced physicians. A high index of clinical suspicion for uterine malformations early in gestation can reduce the mortality rate, along with early intervention. When a rudimentary horn pregnancy is diagnosed, the excision of the horn with ipsilateral salpingectomy is the recommended surgical treatment for the best prognosis. These case series highlight the need for high clinical suspicion of this rare condition.

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