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Uterine Myoma in a Rudimentary Horn Miming an Adnexal Neoplasm in Pregnancy, Successful Delivery after Laparoscopic Resection at 14+5 Weeks Pregnancy: A Case Report

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Abstract

Congenital uterine anomalies are rare, but a correct diagnosis and treatment during pregnancy are essential for successful clinical outcomes for both mother and child.

Keywords: Uterine anomalies; Miscarriage; Laparoscopy; Gynecological surgery; Surgery during pregnancy; Unicornuate uterus

Introduction

Congenital uterine anomalies result from the abnormal formation, fusion or resorption of Müllerian ducts during embryological development [1,2]. This results in various degrees of rudimentary horns connected or not to the opposite horn. The prevalence of these anomalies in unselected population is 5.5%, 13.3% in those with history of miscarriage, 8% in infertile women and highest than 24.5% in infertile women who also had a history of miscarriage [1,2]. A unicornuate uterus presents the 0.1% of women [3] and it constitutes approximately 20% of Müllerian duct anomalies [4]. The rudimentary horn is recognized in about 74%-90% of these cases [5]. Anomalies are more common in infertile women as in women with repeated miscarriage and obstetric adverse outcomes [6,7]. True prevalence is difficult to assess, partly because different classification systems are used and partly because best diagnostic techniques are invasive and rarely applied to low-risk population [1,2,8].

Müllerian anomalies are frequently asymptomatic and are often missed in routine gynecological examination [9]. Methods for the assessment of uterine morphology are three-dimensional ultrasound, hysterosalpingography, hysteroscopy, laparoscopy and RMI [10]. In most cases, unicornuate uterus is incidentally discovered when pelvis is imaged but sometimes it can even be missed at time of laparotomy or laparoscopy by inexperienced surgeons.

Various classification systems have been developed; the latest is ESHRE-ESGE system that includes anatomical variation of uterus, vagina and cervix in different classes [11]. This classification is not in widespread use and does not allow comparison, the most common classification of American Fertility Society [12,13] AFS classification defines seven classes, unicornuate uterus is represented in class II divided in four subclasses: with communicating rudimentary horn, non-communicating, no cavity horn and no horn.

Clinical presentations for Müllerian anomalies are different; when symptoms are present, range from amenorrhea, dysmenorrhea, pelvic pain, hematometra, hematosalpinx and adverse reproductive and obstetric outcomes as infertility, spontaneous abortion, preterm delivery, malpresentation, intrauterine growth restriction, placental abruption and intrauterine fetal demise [14,15].

It is theorized it is due to diminished muscle mass abnormal uterine blood flow and cervical incompetence [6,7].

Diagnosis and management of such types of anomalies are often a challenge for the gynecologist.

We present a case report of a successful delivery in an undiagnosed unicornuate uterus after laparoscopic resection of a rudimentary horn at 14+5 weeks' gestation thought to be an ovarian neoplasm.

Case Report

A 34-year-old Caucasian woman GII P0, with a spontaneous pregnancy at 4 weeks of amenorrhea and positive urine pregnancy test, comes to our observation because of vaginal bleeding. She has a history of mild dysmenorrhea and menorrhagia. In 2015 she experienced a twin pregnancy miscarriage managed with a D&C (dilatation and curettage). Transvaginal ultrasound revealed no gestational sac, normal right ovary and left adnexal mass of 60*60 mm of mixed echogenicity and increased vascular pattern. The image was suspect for solid ovarian neoplasm.

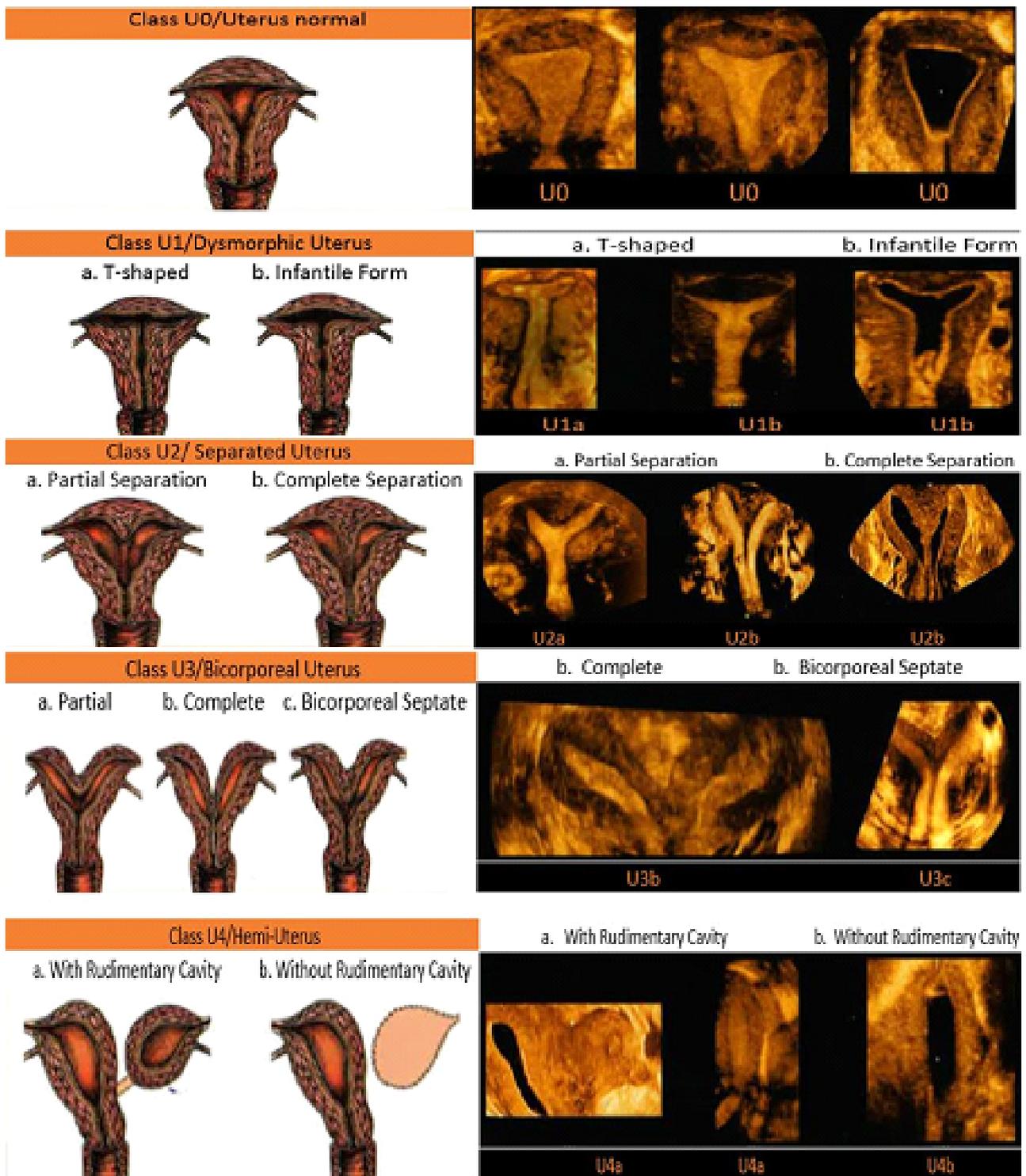


Figure 1 New classification and terminology of uterine malformations & contribution of the frontal view Echographic.

A two week follow up was scheduled and a vaginal ultrasound showed a yolk sac, without embryo. The evaluation confirmed the left adnexal mass, tumor markers were negative (CA 125=29.7 U/ml, CA 15-3=21.6 U/ml, CA 19-9= 3.8 U/ml, CEA=0.9 ng/ml, AFP 1.1 ug/l, LDH= 156 U/l) (**Figures 2 and 3**).

The patient was referred to our oncological multidisciplinary team discussion (MDM) and the board decided for a follow up by bi-weekly scan.

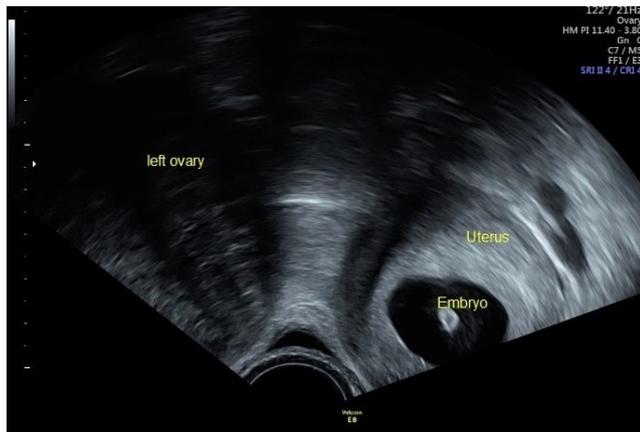


Figure 2 Echographic appearance of uterus and pelvic mass.



Figure 3 Echographic appearance of pelvic mass.

The first-trimester screening scan at 12 weeks' gestation revealed ongoing pregnancy with a low risk for fetal trisomy (T21, T13 eT18), but an increased risk of preeclampsia, IUGR and preterm birth, the patient started prophylaxis with acetylsalicylic acid from 12 to 36 weeks gestation. Because of the increased size of the left adnexal mass, now measuring 73*74*80, the patient was referred again to the oncological multidisciplinary team discussion and the board decided for an explorative laparoscopy with adnexectomy.

At 14+5 weeks' gestation the patient underwent laparoscopic examination under general anesthesia. The pelvic status was the following: uterus in the midline, the right fallopian tube, the right ovary, the right round ligament and the utero-ovarian ligament were normal. The left aspect of the uterus appeared smooth and without any adnexal landmark (**Figure 4**).

The left part of the pelvic cavity was occupied by a bulky structure that was laterally connected with a normal ovary, a fallopian tube was visible and of normal aspect, no left round ligament was identified (**Figure 5**).



Figure 4 Laparoscopic appearance of uterus and pelvic mass.

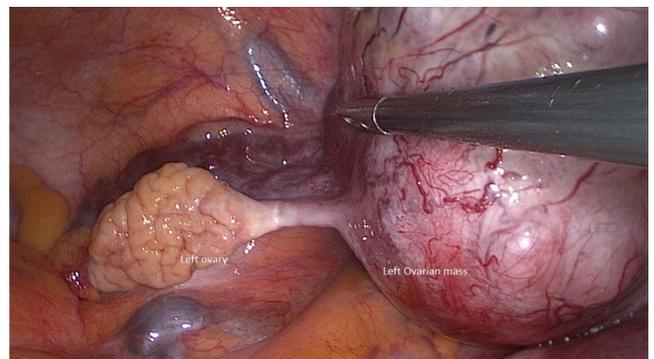


Figure 5 Laparoscopic appearance of left adnexa.

The mass appeared connected to the left lateral aspect of the isthmic uterine region by a retroperitoneal cord. After careful inspection of the anatomy and after identifying the ureters course bilaterally, the resection of the mass was made. We started with opening peritoneum, then transection of the retroperitoneal cord in regard to the isthmic region of uterus was made ultracision and bipolar forceps. Thereafter coagulation and resection of left uterus-ovarian ligament, followed by coagulation and resection of rudimentary broad ligament. After careful control of hemostasis and ureters position, the surgical piece was extracted in a plastic bag (endocatch). The macroscopic appearance was suggestive for myoma. At the end of the surgery the ultrasound examination showed a viable fetus with a normal fetal heartbeat. The postoperative course was uneventful, and the patient was discharged after 4 days.

Laparoscopic findings and hysto-pathological report brought to a diagnosis of right unicornuate uterus with left fibromatous rudimentary horn. According to American Fertility Society Classification this malformation comes under U4a/U4b. At 21 week's gestation, the second trimester screening scan showed a normal fetal anatomy, the placenta appeared to be correctly inserted, no major signs for abnormally adherent placenta were seen. The echographic measurement of the cervical length showed a cervix of 25 mm. According to national guidelines an intra-vaginal 200 mg progesterone

treatment started, and sonographic measurement of cervical length every two weeks was scheduled. The first sign of intrauterine growth restriction appeared at 30 weeks' gestation: BPD and HC were at the first percentile, Doppler evaluation of umbilical artery, middle cerebral artery and ductus venosus were normal. The fetal presentation was breech.

At 32 weeks' gestation fetal growth remained stable at the same percentile. Blood pressure was normal, no clinical signs of preeclampsia and cervical length was stable at 25 mm. Urinary proteins value was 0.54 gm/l and protein/creatinine ratio 12.9 mg/mmol.

After 3 weeks there were an increase of these values: urinary proteins were 0.67 gm/l and protein/creatinine ratio were 56.7 mg/mmol, daily blood pressure readings remained normal, fetal weight estimation was at the 1st percentile, Doppler evaluation of umbilical artery, middle cerebral artery and ductus venosus were still normal. We arranged an elective cesarean section at 37 weeks' gestation after pulmonary maturation with two intramuscular injections of dexamethasone 12 mg. At 37+3 weeks' gestation a healthy boy was delivered by cesarean section, the post-operative appearance of the uterus confirmed the diagnosis of unicornuate uterus (**Figure 6**).



Figure 6 Laparotomic appearance of uterus after cesarean section.

The baby weight was 2470 gr (5th percentile), 46 cm length, Apgar score was 9-10-10. Both baby and mother were discharged in good condition.

Discussion

In case of unicornuate uterus pregnancies can be either in the primary uterine cavity or in the rudimentary horn. Pregnancies developing in the rudimentary horn have to be removed. This is the first report of laparoscopic resection of rudimentary horn, during ongoing pregnancy in the main horn; several case reports described laparoscopic approach for removal of the rudimentary horn, but no one during pregnancy in unicornuate uterus.

Uterine anomalies should therefore be suspected in pregnancies with atypical ultrasonographic features and more investigations done to confirm the diagnosis, with a view to improving management of clinical care. The first trimester screening scan has not to focus only on fetus, it is also important to observe thoroughly adnexal region and to investigate possible unclear or suspected images in order to reduce associated morbidity and mortality. Prophylactic resection of a noncommunicating uterine horn with a cavity should be considered in an asymptomatic, reproductive-age patient with this rare Mullerian anomaly.

It would be safer to remove the rudimentary horn outside pregnancy to better manage future pregnancy, but this clinical presentation did not allow us to make the proper diagnosis. As a fast-increasing ovarian mass can represent a harmful event for a young girl, it was mandatory to proceed with the surgery in order to exclude ovarian cancer. It is interesting that an accurate first trimester screening scan allow the clinician to diagnose the high-risk cohort for intrauterine growth restriction and preeclampsia. Moreover, the absence of the left uterine artery during the scan should have brought our attention toward the presence of uterine anomalies. The balance of risks and benefits of results of serial ultrasound, cervical length, blood pressure and urine protein follow-up guided us to the choice of 37 weeks' gestation caesarean section.

Conclusion

Unicornuate uterus is present in only 0.1% of population, facing such challenging cases of anomaly, physician skills and decisions are very important to manage pregnancy complications at time of appearance because of a lack of literature for evidence-based decision making. Though rare, uterine Müllerian anomalies as unicornuate uterus should be included in differential diagnosis facing pelvic unrecognised mass, dysmenorrhea, and first trimester miscarriage: that is essential to provide better care to patients. Prophylactic resection should be considered in an asymptomatic, reproductive-age, because surgical resection of rudimentary horn may improve obstetric outcomes in selected cases such as ours. Future reports will define optimal management approach.

Conflict of Interest

The authors have no conflicts of interest to report with this research.

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