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Case Report

Unusual pregnancy in the rudimentary horn of the uterus

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ABSTRACT

Mullerian anomalies are rare to be diagnosed, most of the females with Mullerian anomalies go undiagnosed, as they remain asymptomatic. It is a rare clinical scenario that surprises clinicians since it poses difficulty in diagnosis and treatment. Hereby, we describe a case of a 24-year-old, gravid-2-abortion-1 woman who complained of increased abdominal pain and vaginal bleeding with 9 weeks pregnancy. During past laparoscopic surgeries, she was diagnosed of having a bicornuate-uterus. The first sonography reported a bicornuate uterus with a missed-abortion of 9 weeks. As she was taken up for suction-evacuation, suspicion of a unicornuate-uterus with rudimentary horn raised as no products of conception were evacuated. Repeat ultrasound and MRI demonstrated a gestation sac in the non-communicating rudimentary horn attached to the unicornuate uterus. Further diagnostic hysteroscopy and laparoscopy confirmed the diagnosis of pregnancy in the non-communicating rudimentary horn of the unicornuate uterus with a right-sided ovarian cyst not separately seen from the right-sided fallopian tube and ovary for which the decision of laparoscopic resection of the rudimentary horn and right salpingo-oophorectomy was made. Patient was later discharged without any complications and was advised contraception for the next 6-12 months. Further consultations for infertility and urinary tract abnormalities were organized. Though rare, cases of Mullerian anomalies need to be addressed with utmost care because usually they might present with symptoms of pain or infertility, but rarely they might have catastrophic consequences such as rupture of the rudimentary horn. Hence, prompt diagnosis and management are necessary.

Keywords: Mullerian anomalies, Rudimentary horn, Non-communicating, Unicornuate uterus, European classification

INTRODUCTION

The prevalence of congenital uterine malformations is about 1 in 200 women, this prevalence only includes those who are diagnosed as having malformations whereas several women remain undiagnosed. Defects in the developmental process lead to various uterine malformations. The uterus, cervix, and the upper part of the vagina are formed by the fusion of two paramesonephric or Mullerian tubes, developmental defects lead to various uterine malformations. The European Society of Human Reproduction and Embryology suggests the classification of uterine malformations based on their anatomical deviation.

A unicornuate uterus is a rare uterine malformation with an incidence ranging from 2.5-13%.[1,2] These are associated with various gynecological or obstetric complications and diagnosing Mullerian anomalies is often difficult. It may cause a delay in the fertile period or pregnancy.[3,4] Patients may present with dysmenorrhea, dyspareunia, menorrhagia or malformations of the upper urinary tract, which are commonly inherited with a unicornuate uterus. Pregnancy in the non-communicating rudimentary horn can also lead to a life-threatening complication such as rupture of the rudimentary horn.[5,6]

We report a case of a unicornuate uterus with a cavitated rudimentary horn having a gestation sac that did not communicate with the normal uterine cavity.

CASE REPORT

A 24-year-old, gravida 2 abortion 1 at 9 weeks pregnant attended the early pregnancy unit with blood spotting per vagina and pain in the abdomen. She had a spontaneous abortion at 4 months which took place 4 years ago. She had undergone 2 laparoscopic procedures one for aspiration of ovarian cyst and the other for excision of the chocolate cyst (endometrioma). During laparoscopy, she was diagnosed of having a bicornuate uterus. She appeared well and was hemodynamically stable. The ultrasound scan confirmed a missed miscarriage corresponding to 9 weeks of pregnancy with no cardiac activity in an apparent bicornuate uterus. There was no adnexal mass or free fluid. The vaginal examination revealed bulky uterus with normal cervix.

She opted for the surgical method of evacuation but on suction-evacuation, no products of conception were obtained. The tip of the suction cannula could not reach the product of conception. The suspicion of a rudimentary horn was then raised.

She was sent for a repeat ultrasonography (USG) the very next day where she was diagnosed of having a hemiuterus with a non-communicating right horn with a gestational sac and a right-sided ovarian cyst. Magnetic resonance imaging (MRI) confirmed the diagnosis of hemiuterus with moderate hematometra in the right-sided non-communicating horn of the uterus with gross right-sided hydrosalpinx with a right-sided ovary not well visualized.

On the subsequent day, she consented to a diagnostic hysteroscopy and laparoscopy.

Under general anaesthesia and all aseptic measures, per vaginal examination showed a bulky uterus with a right adnexal mass. On hysteroscopy, there was evidence of a single normal cervix with a single uterine cavity. Later laparoscope was inserted, revealing a unicornuate uterus with a normal left-side horn attached to which, was a right-sided rudimentary horn with an intact round ligament and fallopian tube (Figure 1). This right part of the uterus held the pregnancy, was dilated, inflamed, and without signs of rupture. The right-sided ovarian cyst was not separately seen from the right-sided fallopian tube and the right ovary had e/o bowel adherent to it, which was released then the right-sided cyst along with the ovary and fallopian tube was removed. Later the right-sided uterine rudimentary horn was resected (Figure 2). After the separation of the uterovesical fold, the uterine artery on the right side was coagulated. Left-sided uterine cavity, fallopian tube, and ovary appeared normal, and diffuse mild pelvic endometriosis was noted.

The cut section of the specimen contained products of conception. The patient recovered without complications and was later discharged, was advised contraception for 6 months to 1 year. Further consultation about fertility issues and the exclusion of urinary tract abnormalities was organized.

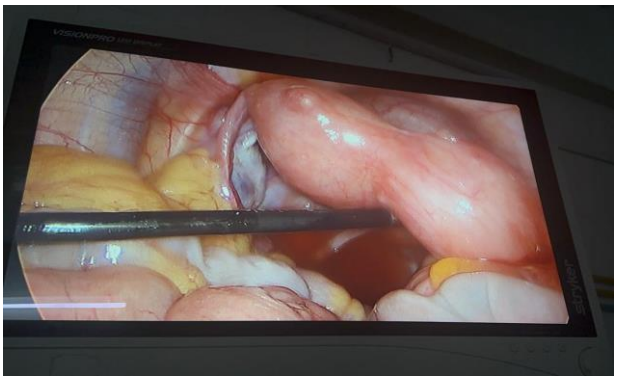

Figure 1: Right pregnant rudimentary uterine horn attached to the left side of the unicornuate uterus.

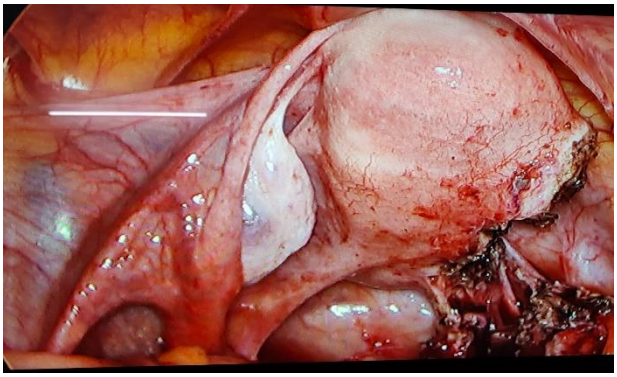

Figure 2: After the excision of the right rudimentary horn, which was linked to the uterus.

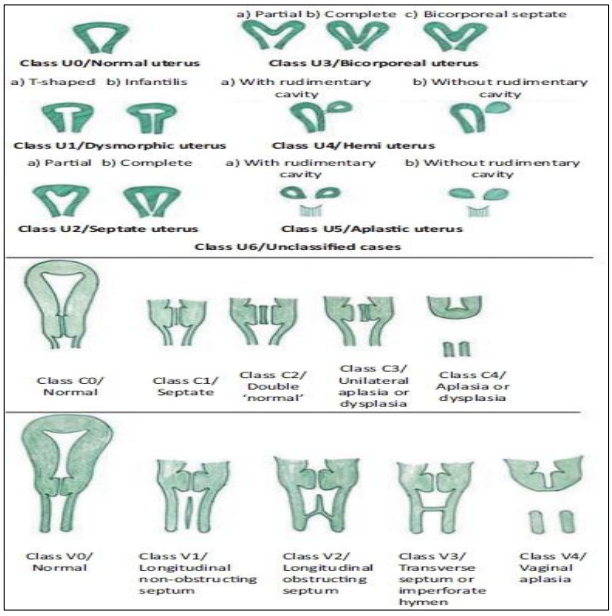

Figure 3: The European Society of Human Reproduction and Embryology and the European Society for Gynecological Endoscopy classification.

DISCUSSION

Unicornuate uterus with pregnancy in a rudimentary horn is an extremely rare clinical scenario that often surprises the gynecologist or obstetrician. The exact incidence of unicornuate uterus is unknown and is difficult to determine since many women with such anomalies are not diagnosed, especially if they are asymptomatic or nonpregnant.[7]

Complete formation and differentiation of the Mullerian duct into the female reproductive system depends on the completion of 3 phases of development as follows: organogenesis, fusion, and septal resorption. Development of the reproductive system is closely associated with the development of the urinary system; hence many malformations have associated urinary tract abnormalities. Organogenesis defects may cause aplasia hypoplasia or unicornuate uterus. Fusion defects if lateral may cause didelphys or bicornuate or arcuate uterus likewise if vertical fusion is defective, it may cause TVS or imperforate hymen. Septal resorption defect may cause a septate uterus.

In the ESHRE/ ESGE classification system, anomalies are sorted in the classes and sub-classes of the system according to the increasing severity of the anatomical deviation (Figure 3).[8,9]

Main uterine anomaly classes: class U0 or normal uterus; class U1 or dysmorphic uterus incorporates all cases with normal uterine outline but with an abnormal shape of the uterine cavity excluding septa, e.g. T-shaped uterus, infantile uterus, and other minor uterine deformities; class U2 or septate uterus incorporates all cases with normal fusion and abnormal absorption of the midline septum (partial or complete); class U3 or bicorporeal uterus incorporates all cases of fusion defects with an abnormal fundal outline; class U4 or hemi-uterus incorporates all cases of unilateral formed uterus, the contralateral part could be either incompletely formed or absent; class U5 or aplastic uterus incorporates all cases of uterine aplasia; and class U6 is kept for still unclassified cases.

Co-existing cervical anomalies: sub-class C0 normal cervix; sub-class C1 or septate cervix; sub-class C2 or double cervix; sub-class C3 or unilateral cervical aplasia; and sub-class C4 or cervical aplasia, also severe cervical formation defects.

Co-existing vaginal anomalies: sub-class V0 or normal vagina; sub-class V1 or longitudinal non-obstructing vaginal septum; sub-class V2 or longitudinal obstructing vaginal septum; sub-class V3 or transverse vaginal septum and/or imperforate hymen; and sub-class V4 or vaginal aplasia.

Our case is classified under ESHRE/ESGE: U4aC0V0. That is hemiuterus with an incompletely formed contralateral part. Further, the incompletely formed part may be communicating or non-communicating with the main uterine cavity.

There are various complications in such a malformed uterus. Gynecological complications such as infertility, dyspareunia, dysmenorrhea, and menorrhagia and obstetric complications such as mid-trimester abortion, malpresentation, preterm labour, prolonged labour, obstructed labour, retained placenta, and postpartum hemorrhage. And most life-threatening complication in such a uterus is the rupture of the rudimentary horn which carries pregnancy either due to a thinned muscular wall or an unphysiological implantation.

The incidence of a unicornuate uterus with a rudimentary horn is quite rare. Usually, ipsilateral ovaries are normal as it is not developed from paramesonephric ducts but may be abnormally located. Our patient had a significant history of endometriosis which can be explained by retrograde menstruation theory proposed by Sampson. The theory proposes that endometriosis occurs due to sloughed endometrial cell debris via the fallopian tubes into the pelvic cavity during menstruation.[10]

Conception occurs either from the main uterine cavity or from trans-peritoneal migration of the spermatozoa from the contralateral fallopian tube or the fertilized ovum from the ipsilateral or contralateral fallopian tube.[11] These pregnancies are characterized by a high risk of miscarriage. When the pregnancy exceeds the first trimester there is an increased risk of uterine rupture. Over 89% of such pregnancies end up experiencing rupture in the 2nd trimester and only 1% may succeed with live birth. Uterine rupture in such cases is a serious life-threatening situation complicated by maternal hemorrhagic shock.[12]

Surgery is recommended whenever a diagnosis of a pregnancy in the rudimentary horn is made immediately. The traditional treatment is a surgical removal of the pregnant horn to prevent rupture and recurrent rudimentary horn pregnancies which can either be done by laparotomy or laparoscopy. The laparoscopic approach is better for resecting these horns as it is safe and has more advantages than laparotomy.[13,14] Some authors have described systemic methotrexate administration or feticide with intracardiac potassium chloride as alternatives or adjuncts to surgery in early gestational period.[15] In all such cases, the patient should be informed of the risks of the condition as well as their management options.

CONCLUSION

Pregnancy in the rudimentary horn of the uterus places diagnostic and therapeutic challenges. Despite ultrasonography advances, the antenatal diagnosis of rudimentary horn pregnancy is difficult for an inexperienced physician. When a rudimentary horn pregnancy is diagnosed, the excision of the rudimentary horn with ipsilateral salpingectomy is recommended for

the best prognosis, it can either be done laparoscopically or by exploratory laparotomy.

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