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A Case of Pre−Rupture Diagnosis and Laparoscopic Management of Non−Communicating Rudimentary Uterine Horn Pregnancy

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Abstract

Pregnancy in a rudimentary uterine horn is extremely rare and difficult to diagnose before rupture. We report a case of non−communicating rudimentary horn pregnancy diagnosed before rupture and removed laparoscopically. A 31−year−old primiparous woman with a suspected ectopic pregnancy was referred to our hospital at 8 weeks' gestation. Transvaginal ultrasonographic tomography（TV−UST）revealed a mass containing a gestational sac and live fetus on the left side of the uterus. The mass was connected to the uterus via a thick pedicle；a thin myometrium−like echo image surrounded the gestational sac. The mass did not connect with the endometrium. Magnetic resonance imaging confirmed TV−UST findings. We diagnosed non−communicating rudimentary horn pregnancy. The rudimentary horn was removed with laparoscopic surgery. The recommended treatment for rudimentary uterine horn pregnancy is surgical removal of the rudimentary horn, similarly to other ectopic pregnancy. Laparoscopy is a useful and safe method for treating rudimentary horn pregnancy.

Key words：Rudimentary uterine horn pregnancy, Laparoscopic surgery, ectopic pregnancy

Introduction

Uterine anomalies result from complete or partial failure of development of one of the embryonic Müllerian ducts and incomplete fusion with the contralateral side in the seventh or eighth week of gestation. Unicornuate uterus, which accounts for 0.1% of all uterine anomalies, is classified as a type II Müllerian anomaly according to the American Fertility Society classification system[1)]. Approximately 75%−90% of unicornuate uterus cases are accompanied by a rudimentary horn. Rudimentary horns are subclassified as communicating or non−communicating with the uterine cavity and can contain a functional or non−functional endometrial cavity.

Pregnancy in a rudimentary horn is extremely rare, with an incidence of approximately 1 in 76,000−150,000 pregnancies and constituting approximately 0.24%−0.6% of ectopic pregnancies[2)]. Most rudimentary horn pregnancies involve a non−communicating rudimentary horn, and most do not reach term；80%−90% rupture during the first or second trimester. When rudimentary horn pregnancy is diagnosed, excision of the pregnant horn is recommended to avoid the serious risk of massive bleeding. However, it is difficult to diagnose rudimentary horn pregnancy before rupture because most patients are asymptomatic.

In this paper, we report an extremely rare case of a non−communicating rudimentary horn pregnancy diag-

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nosed before rupture and successfully treated with laparoscopic resection.

Case report

A 31-year-old primiparous woman with no relevant past medical, surgical, or family history presented to her local hospital for evaluation of amenorrhea lasting 5 weeks since her last menstrual period. She had a history of an irregular menstrual cycle, consistently about 7 days' duration, with no dysmenorrhea. The patient had no relevant gynecologic history, such as pelvic pain or infertility. A urine pregnancy test was positive. However, a gestational sac was not detected in the uterus on transvaginal ultra-sonographic tomography (TV-UST). The patient returned to the hospital for recheck at 8 weeks' gestation. An intrauterine gestational sac was again not detected, raising the possibility of ectopic pregnancy. The patient was referred to our hospital for consultation. At initial examination, we observed no intrauterine gestational sac, but a mass with a gestational sac and live fetus was seen on the left side of the uterus with TV-UST (Figure 1). The mass was connected to the uterus via a thick pedicle and there was a thin myometrium-like echo image surrounding the gestational sac. Bicornuate uterus pregnancy was suspected. However, because continuity of the endometrium was not seen, we suspected rudimentary horn pregnancy. Magnetic resonance imaging (MRI) confirmed the findings of TV-UST : connection between the mass and the uterus with no endometrial continuity (Figure 2). No urinary anomaly was detected

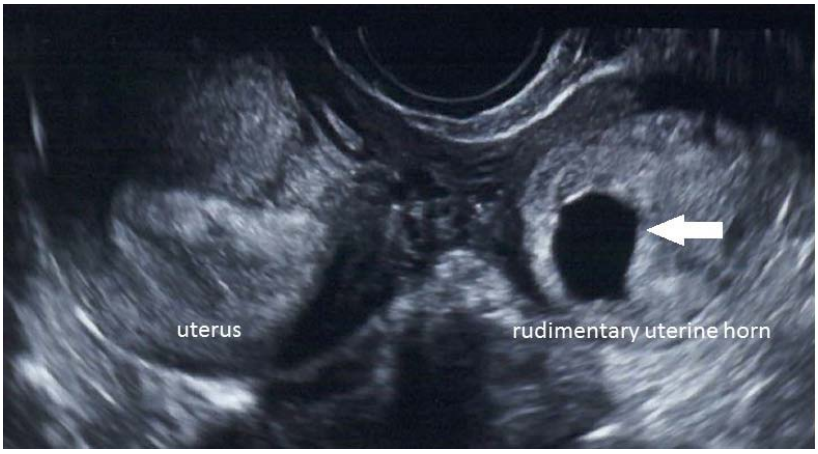

Figure 1. Left rudimentary uterine horn pregnancy is seen. A viable 8-week embryo is seen in the left rudimentary uterine horn, surrounded by a thin myometrial wall. Solid arrow ; gestational sac.

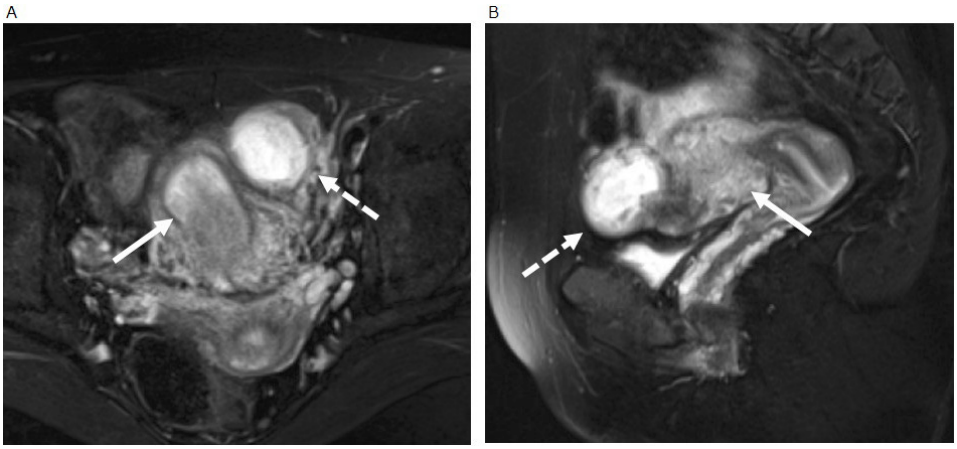

Figure 2. MRI T2-weighted image. A) Horizontal cross-section. B) Sagittal section. Solid arrow ; Uterus. Dotted arrow ; rudimentary uterine horn pregnancy (gestational sac).
Mass with a gestational sac and live fetus was seen on the left side of the uterus. Connection between the mass and the uterus with no endometrial continuity

by MRI. We diagnosed non-communicating rudimentary horn pregnancy and laparoscopic surgery was performed. On initial evaluation, a thick fibro-muscular pedicle connecting the uterus to a left rudimentary horn was observed (Figure 3). A normal ovary, fallopian tube, and round ligament were attached to the left rudimentary horn. The right ovary and fallopian tube appeared normal. The band connecting the rudimentary horn to the uterus was dissected with the use of the LigaSure™ vessel sealing system. The ovarian ligament, round ligament, and fallopian tube attached to the rudimentary horn were resected. The rudimentary horn and fallopian tube were removed via an endobag. The uterine stump and round ligament were sutured for reinforcement. The postoperative period was uneventful, and the patient was discharged healthy on postoperative day 4. The pathology report showed decidua and villi in the smooth muscle tissue of the rudimentary horn, confirming ectopic pregnancy in the rudimentary uterine horn (Figure 4).

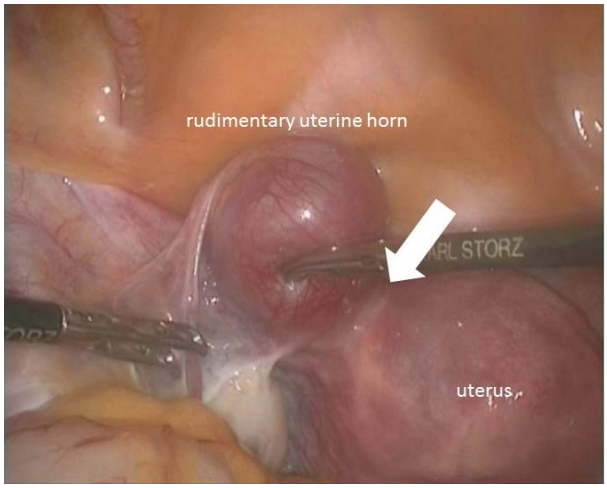

Figure 3. Left rudimentary horn is attached to the uterus with a thick fibro-muscular band (white arrow). The left ovary, fallopian tube, round ligament, and ovarian ligament are attached to the rudimentary uterine horn and appear normal.

Discussion

Rudimentary uterine horn is caused by failure of development of the Müllerian duct. Rudimentary horns are classified as communicating or non-communicating with the uterus. Non-communicating uterine horns are further classified as cavitated or not, with or without a functional endometrium. In relatively young women who have a functional endometrium in a rudimentary uterine horn cavity, rudimentary uterine horn may be diagnosed by menstrual disorders, including dysmenorrhea, endometriosis (due to reflux of menstrual blood from the rudimentary uterine horn into the oviduct), hematometra (retention of menstrual blood), and pyosalpinx[3]. However, very few cases present with such symptoms. In cases with non-functional endometrium, there are no symptoms of menstrual disorder. In our patient, there were no symptoms of menstrual disorder before diagnosis of pregnancy. Therefore, diagnosis of the condition often occurs only after rudimentary horn pregnancy. In all cases of uterine anomaly, the possibility of renal or urinary tract malformation must be considered. Concurrent uterine and urinary anomalies are reported in 38% of cases[4], but this complication was not found in our patient. Careful attention to the location of the ureter is necessary to avoid ureteral injury during dissection of the rudimentary uterine horn.

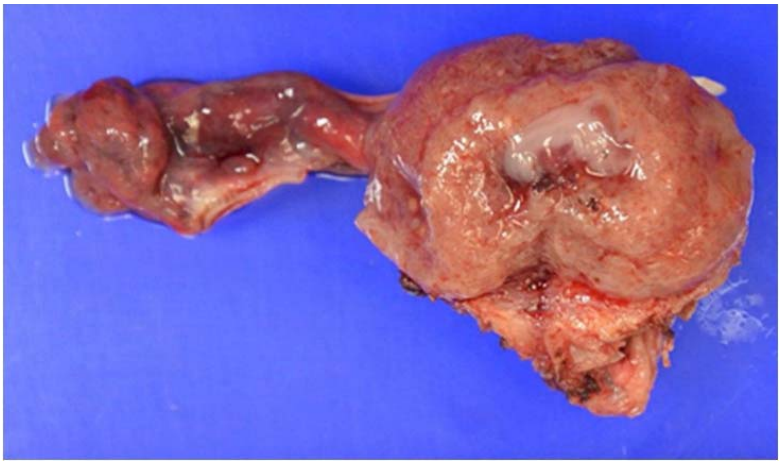

Figure 4. Macroscopic appearance of the resected specimen. Decidua and villi in the smooth muscle tissue of the rudimentary horn

For a safe resection, presurgical assessment of anatomical relationships and the arrangement of adjacent tissues and organs with MRI or computed tomography angiography is very important.

Non-communicating rudimentary uterine horn pregnancy is thought to result from transperitoneal migration of the fertilized ovum or the zygote from the contralateral oviduct[5].

Early diagnosis of rudimentary uterine horn pregnancy is essential, but diagnosis is difficult before rupture and most cases are diagnosed during laparotomy for acute abdomen. Pre-rupture diagnosis of rudimentary uterine horn pregnancy is difficult and rare. As a result, rudimentary uterine horn pregnancy is sometimes misdiagnosed as other uterine malformation, such as bicornis, uterus duplex, or mediastinum uterus.

In other uterine malformations, the gestational sac should be continuous with the cervix, which was not true in our case. Rupture of rudimentary uterine horn pregnancy frequently results from late or incorrect diagnosis.

There are a few reports of rudimentary uterine horn pregnancy reaching atterm delivery[2], in which the rudimentary uterine horn is discovered during caesarian section for failure of transvaginal delivery. However, because of the relatively small vascular volume and anomalous blood supply resulting from the reduced expansibility of the rudimentary uterine horn, a malformed fetus, fetal growth restriction, oligohydramnios, and fetal malpresentation have been reported[6]. Therefore, it is necessary to consider termination of pregnancy and horn excision when a rudimentary uterine horn pregnancy is confirmed. Recently, the availability of ultrasound and MRI has improved the diagnosis of rudimentary uterine horn pregnancy at an early gestational age. The sensitivity of ultrasound in diagnosing rudimentary uterine horn pregnancy can be as low as 30%[7]. Recently, criteria for the early diagnosis of rudimentary uterine horn pregnancy have been proposed, including ⑴ gestational sac surrounded by myometrium adjacent to a normal empty uterus, ⑵ non-communication of the gestational sac with the endometrial cavity and cervix, and ⑶ pseudo-pattern of an asymmetrical bicornuate uterus[8]. In cases where ultrasound findings raise suspicion of rudimentary uterine horn pregnancy, it may be necessary to confirm the condition with MRI.

MRI in these cases shows a gestational sac in the rudimentary uterine horn, surrounded by myometrium that has isointensity with the uterine muscle layer. In our case, these findings were present.

Previously studies have reported that rupture occurs in 80% of rudimentary uterine horn pregnancies, with most occurring during in the first or second trimester, before 20 weeks' gestation[9]. As in the case of non-pregnant rudimentary uterine horn, surgical removal of rudimentary horn pregnancy is recommended. The attachment between the uterus and the rudimentary horn may be fibrous or fibro-muscular band. Rudimentary uterine horn surgery has generally been performed via laparotomy. The presence of a thick fibro-muscular band between the uterus and the rudimentary uterine horn make laparoscopic removal more difficult. However, new energy devices have been developed that facilitate the laparoscopic removal of rudimentary uterine horns. Furthermore, there have been some reports of successful laparoscopic surgery during not only first trimester but also the middle trimester of pregnancy[10].

In conclusion, laparoscopic removal of a rudimentary uterine horn pregnancy before rupture was safe and successful in this case. Rudimentary uterine horn pregnancy should be considered in patients with ectopic pregnancy ; early and precise diagnosis before rupture is important.

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（平成31．4．1受付，令和 1．5．20受理）

「The authors declare no conflict of interest.」