

# Rudimentary uterine horn pregnancy in 15 cases: case series with a two-year follow-up

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## Summary

**Purpose:** The aim of this study was to understand and investigate the reproductive outcomes of patients with rudimentary horn pregnancies after surgical removal of the rudimentary horn. **Materials and Methods:** The clinical records, including age, obstetric history, gestational age (weeks), presenting feature, preoperative diagnosis, intraoperative diagnosis, blood loss, and surgical management of women who were diagnosed with pregnancy in the rudimentary horn from 2006 to 2010 were reviewed in this retrospective study. The reproductive outcomes of these women that underwent surgical removal of the rudimentary horn were also recorded during the two-year follow-up period. **Results:** Fifteen women diagnosed with rudimentary horn pregnancies were included in this study and non-communicating horn was accounted for 87.3% of these cases. The gestation periods of them varied from six to 26 weeks. Rupture of the uterine wall occurred in three cases (20%) and no maternal deaths were observed. Eleven patients underwent laparotomy for rudimentary horn removal and four underwent laparoscopic removal. During the two-year follow-up period, ten out of 15 patients (66.7%) became pregnant and seven of them delivered babies, whereas two asked for medical abortion, and only one had spontaneous abortion during the first trimester. Among the seven women giving live births, three of them were vaginally delivered and four were by cesarean section, five women were term pregnancy, and the other two women gave birth prematurely. **Conclusion:** This study indicates that the early diagnosis, proper consultation, and quick surgical treatment in patients with rudimentary horn pregnancies are necessary.

**Key words:** Rudimentary horn pregnancy; Non-communicating horn; Rudimentary horn removal; Reproductive outcome.

## Introduction

Unicornuate uterus is a type II Müllerian anomaly caused by an arrest in the development of one Müllerian duct and incomplete fusion with contralateral side [1]. Unicornuate uterus associated with various degrees of rudimentary horn can be classified into unicornuate uterus with communicating horn or non-communicating horn and with no cavity. Previous study has shown that the non-communicating horn is the most frequent type accounting for approximately 83% of total unicornuate uterus [2]. However, pregnancy in a rudimentary horn, a special form of ectopic pregnancy, is a rare condition with the incidence rate of 0.01% to 0.007% [3]. Pregnancy in rudimentary horn often results in the rupture of uterine wall, manifesting as acute abdominal pain with intraperitoneal hemorrhage and causing high risks of both maternal and fetal morbidity even mortality [4]. Management of rudimentary horn pregnancy is still a challenge.

It is meaningful for reporting the cumulative experience of the diagnosis, treatment, and follow-up for women with rudimentary horn pregnancy. Therefore, a retrospective study was necessary to report the present authors' clinical experience regarding 15 women with rudimentary horn pregnancy and also present the reproductive outcomes of them after surgical removal of the rudimentary horn.

## Materials and Methods

A total of 15 women who were diagnosed with pregnancy in rudimentary horn in the Women's Hospital, School of Medicine, Zhejiang University were reviewed from 2006 to 2010 in this retrospective study. The inclusion criteria were: 1) preoperative work-up for patients included evaluation of pelvis and urinary system using ultrasonography and intravenous pyelography, 2) all patients underwent surgical removal of a rudimentary horn pregnancy either by exploratory laparotomy or laparoscopy, 3) rudimentary horn and ipsilateral fallopian tube were excised in all cases, and 4) these women did not receive any other fertility treatment postoperatively. The clinical records of these women were collected, including age, obstetric history, gestational age (weeks), presenting feature, preoperative diagnosis, intraoperative diagnosis, blood loss, and surgical management. Meanwhile, their reproductive outcomes by gynecological examination and ultrasonography were recorded at one and three-months follow-up, then a telephone follow-up was conducted during the two-year follow-up period. This study was approved by the Ethical Committees of aforementioned hospital and all the patients had accepted the informed contents that their medical records could be reviewed within the scope of this retrospective study.

## Results

A total of 15 women with rudimentary horn pregnancy were managed at the Women's Hospital, School of Medicine, Zhejiang during years 2006-2010. The mean age of

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Table 1. — *Characteristics of the women with pregnancy in rudimentary horn.*

Case	Age	Obstetric history	Gestational age (weeks)	Presenting feature	Preoperative diagnosis	Intraoperative diagnosis (blood loss, ml)
1	38	G <sub>3</sub> P <sub>1</sub> A <sub>1</sub> L <sub>1</sub>	8	Hypotension	Rudimentary horn pregnancy	Non communicating rudimentary horn, left (50 ml)
2	23	G <sub>1</sub>	12	Pelvic mass	Rudimentary horn pregnancy	Non communicating rudimentary horn, left (50 ml)
3	27	G <sub>2</sub> A <sub>1</sub>	8	Hypotension	Rudimentary horn pregnancy	Non communicating rudimentary horn, left (100 ml)
4	33	G <sub>2</sub> L <sub>1</sub>	6	Bleeding per vaginam	Rudimentary horn pregnancy	Non communicating rudimentary horn, right (10 ml)
5	24	G <sub>7</sub> A <sub>6</sub>	10	Pelvic mass	Rudimentary horn pregnancy	Non communicating rudimentary horn, left (30 ml)
6	27	G <sub>3</sub> P <sub>1</sub> A <sub>1</sub> L <sub>1</sub>	8	Pain abdomen, pelvic mass	Rudimentary horn pregnancy	Non communicating rudimentary horn, right (50 ml)
7	25	G <sub>1</sub>	26	Pain abdomen, pelvic mass	Rupture ectopic pregnancy	Non communicating rudimentary horn, right (100 ml)
8	24	G <sub>3</sub> P <sub>1</sub> A <sub>1</sub> L <sub>1</sub>	9	Hypotension	Rudimentary horn pregnancy	Communicating rudimentary horn, right (20 ml)
9	38	G <sub>3</sub> P <sub>1</sub> A <sub>1</sub> L <sub>1</sub>	26	Pelvic mass	Rudimentary horn pregnancy	Non communicating rudimentary horn, left (50 ml)
10	37	G <sub>5</sub> P <sub>1</sub> A <sub>3</sub> L <sub>1</sub>	9	Pain abdomen, syncopal attack, pelvic mass	Ruptured ectopic pregnancy	Non communicating rudimentary horn, left (50 ml)
11	25	G <sub>1</sub>	13	Pain abdomen, pelvic mass	Ruptured ectopic pregnancy	Non communicating rudimentary horn, right (50 ml)
12	17	G <sub>1</sub>	7	Hypotension	Rudimentary horn pregnancy	Non communicating rudimentary horn, right (20 ml)
13	26	G <sub>2</sub> A <sub>1</sub>	6	Pain abdomen, pelvic mass	Rudimentary horn pregnancy	Non communicating rudimentary horn, right (20 ml)
14	21	G <sub>1</sub>	11	Hypotension	Rudimentary horn pregnancy	Non communicating rudimentary horn, left (20 ml)
15	29	G <sub>1</sub>	12	Pelvic mass	Rudimentary horn pregnancy	Non communicating rudimentary horn, right (10 ml)

G: gestation; P: production; A: abortion; L: live.

these women was  $27.6 \pm 6.3$  (17–38) years. Periods of gestation at diagnosis varied from six to 26 weeks. Three women (20%) presented to emergency with acute abdomen, while another 12 patients were symptomless until they were diagnosed with rudimentary horn pregnancies. Prior to admission in this hospital, nine patients (60%) had more than once pregnancy before, six women underwent vaginal deliveries, and one underwent cesarean delivery before.

Characteristics of included patients with pregnancy in rudimentary horn are presented in Table 1. In this study, ten out of 15 women were diagnosed with rudimentary horn pregnancy by the first ultrasonography during the first trimester in this hospital. Three women had 3D ultrasound examination due to diagnostic dilemma in conventional ultrasound. MRI was performed to confirm the location of the pregnancy in a rudimentary horn for the other two women. On admission, hemoperitoneum had been described in one case (6.7%) and intrauterine adhesions had been found by ultrasonography in two cases (13.3%). In addition, one case had already been diagnosed with uterine

malformation (6.7%). Vaginal examination showed an adnexal mass in eight women (53.3%). Rupture of the rudimentary horn was found in three women (20%). Non-communicating horn accounted for 86.7% of the included cases. Excision of the rudimentary horn and ipsilateral fallopian tube were performed in all patients, with conventional exploratory laparotomy (73.3%) in 11 cases and laparoscopic surgery (26.7%) in the remaining four cases. Eight women (53.3%) had a rudimentary horn in right side and seven women (46.7%) had a rudimentary horn in left side. In addition, six rudimentary horns were broadly attached to the unicornuate uterus (40%) and other nine horns were attached through a fibrous band (60%).

There were no maternal deaths, while rupture of the uterine wall occurred in three cases (27.3%) at the time of late first, early second, and late second trimester during the gestation, respectively. Moreover, the earliest onset of rupture occurred at nine weeks of gestation, which demonstrated that the risk of rupture also existed in early gestation. Among these three cases with rudimentary horn pregnancy, one case had hemoperitoneum (2000 mL) at 26 weeks of

Table 2. — *Pregnancy outcomes after surgical management of the women with rudimentary horn pregnancy.*

Case (N)	Surgical management	Pregnancy outcome after surgical management
1	Exploratory laparotomy	Medical abortion in early pregnancy once
2	Exploratory laparotomy	Cesarean delivery at 35 weeks after pregnancy
3	Exploratory laparotomy	Uterogestation with cesarean delivery
4	Laparoscopic surgery	Medical abortion in early pregnancy once
5	Exploratory laparotomy	No pregnancy
6	Laparoscopic surgery	Contraception and no pregnant
7	Exploratory laparotomy	Recurrent abortion in early pregnancy for three times
8	Exploratory laparotomy	Contraception and no pregnant
9	Laparoscopic surgery	Uterogestation with vaginal delivery
10	Exploratory laparotomy	Contraception and no pregnant
11	Laparoscopic surgery	Uterogestation with cesarean delivery
12	Exploratory laparotomy	Contraception and no pregnant
13	Exploratory laparotomy	Uterogestation with cesarean delivery
14	Exploratory laparotomy	Uterogestation with vaginal delivery and abortion in early pregnancy for once
15	Exploratory laparotomy	Vaginal delivery at 34 weeks after pregnancy

gestation on admission, one case had slight bleeding in the abdominal cavity during operation at nine weeks of gestation and was considered as a rupture of the uterine wall after admission, one special case was suspected with rupture at 13 weeks of gestation, owing to abdominal pain for a few days prior to her admission, and during the exploratory laparotomy, the authors found the entire amniotic sac and parts of placenta thickly sticking to the opening of rupture, and thus there was no bleeding at all.

No intraoperative or postoperative complications were encountered in all these 15 cases. Postoperative hospitalizations ranged from three to nine days. Pregnancy outcomes after surgical management of the women with rudimentary horn pregnancy are shown in Table 2. During the two-year follow-up period, ten out of 15 patients (66.7%) became pregnant with a total number of 13 pregnancies, and seven of them delivered babies; two of them asked for medical abortion and one had spontaneous abortion during the first trimester. Among the seven women giving live births, three of them were vaginally delivered and four by cesarean section, which suggested that the uterus of these women had well recovered and the uterus on the other side was normal. It is worth mentioning that five women were term pregnancy (38-41 gestational weeks) and the other two women (patients 2 and 15) were preterm (34 and 35 gestational weeks). Among the five non-pregnant patients, contraceptive was adopted by four patients and only one woman who wanted a baby did not become pregnant during two years post-treatment (6.7%).

## Discussion

Although sporadic cases of producing a live infant have been reported, the prognosis of rudimentary horn pregnancy is always poor for the pregnancies [5, 6]. Thus, it is meaningful to report the cumulative experience of diagno-

sis, treatment, and follow-up of rudimentary horn pregnancy. In the present study, the basic characteristics and the pregnancy outcomes of 15 cases of rudimentary uterine horn pregnancies were described. Non-communicating horn accounted for 87.3% of 15 cases. Rupture of the uterine wall occurred in three cases (20%) and no maternal deaths were observed. Eleven patients underwent laparotomy for rudimentary horn removal and four underwent laparoscopic removal. As a result, during the two-year follow-up period, ten out of 15 patients (66.7%) became pregnant and seven of them delivered babies, whereas two asked for medical abortion and only one had spontaneous abortion during the first trimester.

The main reason for the rarity of rudimentary horn pregnancy is non-communicating horns, which accounts for approximately 80% of cases [6]. Another retrospective study also reported that the prevalence of non-communicating horn was about 75% in the unicornuate uterus [4]. In the present study, there was also a high proportion of non-communicating horn (86.7%). Nevertheless, the present findings showed that the number of cases with non-communicating horn locating at the left side of the unicornuate uterus was as many as the right one, which clashed with the previous literatures with the tendency of the right-sided location of the rudimentary horn [7, 8].

Because women with rudimentary horn pregnancy often have a history of previous normal pregnancies, it is difficult to make the confirmed early diagnosis for this ectopic pregnancy [9]. Abdominal pain and collapse with hemoperitoneum can occur suddenly [10]. Nowadays, prenatal diagnosis is mainly conducted with ultrasonography since it is accurate, simple to learn and to apply, as well as with low cost [11]. However, the sensitivity of ultrasound examination is low. 3D ultrasound appears to be sufficiently accurate for the diagnosis and differentiation of congenital uterine anomalies, but its accuracy becomes inferior dur-

ing pregnancy [12]. MRI has been widely used and it contributes to confirm the location of the pregnancy in rudimentary horn if ultrasonography is unable to certify that the pregnancy is intrauterine [10, 13]. In the present study, MRI was performed to confirm the location of the pregnancy in rudimentary horn for two women, indicated that MRI and 3D ultrasound might be a more wise choice when a dilemma existed in early diagnosis of rudimentary horn pregnancy. Unfortunately, it is still different to distinguish rudimentary horn pregnancy with other abnormal pregnancy, and it is easy to be misdiagnosed as interstitial tubal pregnancy, ectopic pregnancy or abdominal pregnancy owing to the similar sonographic [14, 15]. Thus, improving the accuracy of diagnosis is still a challenge and it is critical for taking timely and proper treatment for patients.

For rudimentary horn pregnancy, there is a high risk of miscarriage, ectopic pregnancy, preterm labor, intrauterine growth retardation and malpresentation, and rupture of the uterine wall is the most significant threat [16]. Maternal mortality in the 19<sup>th</sup> century is reported as high as 88% due to rupture and 90% of deaths occurred within the first ten to 15 minutes of the onset of the symptoms [17]. With the development of the obstetric and gynecological medicine, the most recent data revealed that maternal mortality was less than 0.5% despite rupture rates of 50% [10]. Although there were no maternal deaths in the present study, morbidity remains substantial with high rates of placenta accrete and postpartum hemorrhage, which is still a challenge for the future development of medicine.

Traditional treatment of rudimentary horn pregnancy is surgical removal by laparotomy. If patients desired to preserve their fertility, excision of the rudimentary horn and ipsilateral salpingectomy was recommended [4]. In recent years, laparoscopy has been widely used, which was confirmed to be safe and effective procedure to remove a cavitated non-communicating rudimentary horn in the unicornuate [18, 19]. Meanwhile, laparoscopy has been reported to have a significant advantage in effective surgical management in the congenital uterine anomalies [20]. Additionally, Medeiros *et al.* demonstrated that operative laparoscopy could be used as an excellent alternative to laparotomy for administrating the unicornuate uterus with non-communicating rudimentary horn [21]. In this present study, a case with rupture of rudimentary horn underwent laparoscopic removal. Considering the above results and several other advantages of laparoscopy including smaller abdominal scar, less adhesion formations, diminishing postoperative pain, and shortening hospital stay, this minimally invasive surgery would be a more ideal choice in removing the non-communicating rudimentary horn.

The impacts of unicornuate uteri on reproductive outcomes after surgical treatment were also elucidated. In this present study, only one woman who wanted to have babies was not pregnant and other patients who had baby-plan be-

came successfully pregnant during the two years of follow-up. In total, ten out of 15 patients (66.7%) became pregnant and seven of them delivered babies, which suggested that patients had well postoperative recovery. In addition, one patient (6.7%) had experienced recurrent abortion in the early pregnancy for three times and two women (13.3%) had preterm deliveries. The adverse effects of surgical excision were within the accepted limits. Surgical interventions of the rudimentary horn resection before conception can preserve fertility and improve obstetrical outcomes. Based on the present data, it might be speculated that the rates of adverse outcomes have been historically overestimated.

In conclusion, the present case-series might provide useful data which could facilitate the consultation of patients with rudimentary horn pregnancy. However, small sample size and short follow-up time were the main limitation of this study and a larger sample size and long-term follow-up is still needed. Obviously, the early diagnosis is still a challenging, which is the key to successful management. Meanwhile, a proper consultation and a quick surgical treatment in these severe and rare cases are necessary.

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