Successful Term Delivery after Laparoscopic Resection of a Non-communicating Rudimentary Horn in a Patient with a Unicornuate Uterus: A Case Report

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Unicornuate uterus accompanied by a non-communicating rudimentary horn is a rare uterine malformation. If an embryo is implanted into the rudimentary horn, continuation of pregnancy is difficult due to the risk of uterine rupture. We recently performed laparoscopic resection of the right rudimentary horn after two right rudimentary horn pregnancies in a woman, in whom a normal pregnancy occurred in the left unicornuate uterus, leading to successful delivery of a baby. This case is presented herein.

The diagnostic procedures leading to identification of this rare malformation were prompted by inability to remove uterine contents during surgery performed after a diagnosis of missed abortion. A right rudimentary horn pregnancy, which had occurred twice, was treated with methotrexate. To prevent further pregnancy in the right rudimentary horn, resection of this rudimentary horn was planned and successfully implemented under laparoscopic guidance. This surgical procedure is usually difficult, but fertility could be preserved by employing minimally invasive surgery, involving the use of a LigaSure™ Vessel Sealing System to avoid ligation and assure virtually no bleeding. Soon after surgery, a natural pregnancy in the left unicornuate uterus was confirmed. Intrauterine fetal growth was normal, and transvaginal delivery at term was possible.

Key words: Unicornuate uterus, Rudimentary horn, Pregnancy, Methotrexate, Laparoscopy

INTRODUCTION

Uterine malformations often arise from anastomotic failure of the right and left Müllerian ducts or hypoplasia of these ducts, with a prevalence of 4% reported by Raga et al. [1]. The prevalence of unicornuate uterus is 0.1% [2]. A unicornuate uterus is associated with a rudimentary horn in 74%-90% of cases; such horns may or may not communicate with the unicornuate uterus and may or may not have a functional endometrium. Overall, approximately 25% of horns are both functional and non-communicating (ASRM Müllerian Anomaly Classification II [3]).

The incidence of rudimentary horn pregnancy is extremely low (1 in 76000-150000 pregnancies). Most of these pregnancies involve a non-communicating rudimentary horn [4]. In these cases, fertilization takes place via transperitoneal migration of sperm.

Pregnancies within a rudimentary horn can cause serious maternal morbidity and mortality. The reported rupture rate is approximately 50%, with 87% occurring in the first trimester [5]. If rudimentary horn pregnancy is detected, fetal materials are often resected by open or laparoscopic surgery. However, resection during pregnancy carries a high risk of bleeding. Furthermore, cutting the rudimentary horn from the unicornuate uterus may be extremely difficult depending on the degree of adherence, possibly having an adverse affect on fertility preservation. We recently managed a patient in whom rudimentary horn pregnancy had occurred twice. She was conservatively treated with drug therapy, followed by successful laparoscopic surgery, with minimal stress, to resect the rudimentary horn using a LigaSure™ Vessel Sealing System (LVSS) (Valleylab, Colorado, USA) during a non-pregnant period. This patient later became pregnant naturally in the unicornuate uterus, followed by transvaginal delivery of a healthy baby. This case is presented herein.

CASE

The patient was a 29-year-old, nulliparous woman whose medical and family histories were unremarkable. She had a 50-day menstrual cycle and showed no signs of irregularity or dysmenorrhea. At another hospital, she was diagnosed as having spontaneously aborted a twin pregnancy (gestational age, 7 weeks). Removal of uterine contents was attempted at that hospital but was found to be impossible. For this reason, she was referred to our hospital. Transvaginal ultrasonography at the first presentation to our hospital revealed what appeared to be a gestational sac in the right-sided uterus (2 nonviable fetuses, each 35 mm in size). Left-sided uterus, 7-cm long with a hyperechoic endometrium was revealed. A missed abortion of a right rudimentary horn pregnancy was suspected. Four days after the first visit, laparoscopic examination was performed. Her blood human chorionic gonadotropin...
Laparoscopic findings allowed a diagnosis of right rudimentary horn pregnancy (Fig. 1). No signs of rupture were noted. According to the revised-ASRM classification, endometriosis was stage I (score 2), without intraperitoneal adhesions.

Puncture aspiration of the fetal sacs under transvaginal ultrasound guidance was carried out the same day, for the following reasons: (1) there was no evidence of threatened rupture; (2) the blood hCG level was low; and (3) the patient and her family desired conservative treatment. Because normalization of the blood hCG level tended to be delayed postoperatively, methotrexate (MTX) was additionally injected at a dose of 50 mg intramuscularly 4 weeks after surgery. The blood hCG level gradually decreased, and hCG was negative 19 weeks after surgery.

Hysterosalpingography, performed later, revealed contrast material filling only in the left unicornuate uterus cavity and fallopian tube with no filling in the right fallopian tube (Fig. 2). Pyeloureterography revealed no malformations of the kidney or urinary tract. Based on a general assessment of these findings, the patient was diagnosed as having left unicornuate

Fig. 1 Laparoscopic findings: Left unicornuate uterus with right rudimentary horn pregnancy

Fig. 2 Hysterosalpingography: The uterin corpus was deviated to the left side (Left unicornuate uterus)
uterus with a non-communicating right rudimentary horn free of renal or urinary tract malformations (AFS IIb).

She became pregnant 14 months after surgery, but the pregnancy occurred again in the right rudimentary horn. At gestational age 8 weeks, a fetal heart beat was detected, but there was no sign of rupture. Because the patient and her family would not consent to resection of the rudimentary horn, conservative treatment was again selected. Under transvaginal ultrasound guidance, MTX 50 mg was injected locally. Ten weeks later, the blood hCG level showed a tendency to rise, and MTX 50 mg was again injected intramuscularly. Thereafter, her blood hCG level decreased.

Four years and 5 months after the first visit to our hospital (3 years and 3 months after the second rudimentary horn pregnancy), consent to resect the right rudimentary horn was obtained, and laparoscopic surgery was carried out.

**LAPAROSCOPIC SURGERY FINDINGS**

Laparoscopic surgery was carried out under general anesthesia with tracheal intubation and abdominal insufflation. A 5-mm incision was made over the upper edge of the umbilical ring. After abdominal insufflation using a closed technique, a 5-mm trocar was inserted, followed by insertion of an endoscope and observation of the peritoneal cavity. Twelve-mm trocars were then inserted into the right and left hypogastric regions, followed by manipulations for laparoscopic surgery with 3 holes.

The left unicornuate uterus was 7 cm in size. The fallopian tube and left ovary were normal. The right rudimentary horn was 4 cm in size, and the right fallopian tube and right ovary were normal. The right rudimentary horn was connected with relatively sparse tissue to the left unicornuate uterus (Fig. 3a). There were no adhesions within the peritoneal cavity. Endometriosis was mild (revised-ASRM classification: stage 1, score 2). Left tubal patency was revealed with Chromopertubation.

Using the LVSS, the right round ligament and the right ovarian ligament were carefully dissected so as to preserve the right ovary, followed by dissection of the linking sparse connective tissue and resection of the right rudimentary horn and right fallopian tube (Fig. 3b, 3c). The Douglas’ pouch was fenestrated and removed from the body.

The operative time was 60 minutes, and the blood loss volume was 18 ml. The resected right rudimentary horn and left unicornuate uterus were examined in detail, but no communication between them was de-
tected. The resected right rudimentary horn contained endometrium and showed histological signs indicative of endometrial proliferation. The postoperative course was uneventful, without complications. The patient was discharged from the hospital 3 days after surgery.

Postoperatively, she was followed as an outpatient. Four months after surgery, she became pregnant, and the embryo was confirmed to be in the left unicornuate uterus.

Due to threatened premature delivery, she was hospitalized at the 30th gestational week and discharged in good condition in week 36 of her pregnancy. At 39 weeks and 6 days’ gestation, labor pain developed and a baby was delivered transvaginally on 40 weeks gestation. The newborn was a boy weighing 3770 g (APGAR score 8/1/9/8). Both mother and the baby were discharged in good condition.

DISCUSSION

There are two patterns of unicornuate uterus with a non-communicating rudimentary horn. In one pattern, the endometrium of the rudimentary horn functions due to absence of communication between the rudimentary horn and major horn (unicornuate uterus), and retention of menstrual blood leads to hematometra, causing intense menstrual pain as the blood volume increases. This pattern is often seen in relatively young women.

It has also been reported that patients with this condition often have endometriosis [21% [2] to 33% [6]] due to reflux of menstrual blood from the rudimentary horn into the oviduct (transplant theory of Sampson). In the present case, there was no history of dysmenorrhea, and endometriosis was mild. With the other pattern, the diagnosis is made during management of pregnancy, as in the present case. Because the incidence of rupture is not low during rudimentary horn pregnancy, [7-9] risk to the mother is high, necessitating early treatment. The present patient had a history of two rudimentary horn pregnancies. To our knowledge, no other cases with two consecutive implantations in the rudimentary horn have been reported to date. The most reliable standard treatment may be excision of the pregnancy and rudimentary horn. In the present patient with rudimentary horn pregnancy, conservative treatment with MTX was selected for the following reasons.

1. Her systemic condition was stable, with no signs of rupture;
2. The safety of rudimentary horn resection during pregnancy is unknown;
3. The possibility of rudimentary horn resection adversely affecting fertility of the residual unicornuate uterus could not be ruled out;
4. The patient did not consent to rudimentary horn resection during pregnancy, despite being informed of the aforementioned factors.

MTX is on occasion selected as a conservative treatment for ectopic pregnancy and has been considered to be excellent in terms of preserving the fertility, i.e., a patent tube, in cases with a tubal pregnancy [10, 11]. The usefulness of MTX in cases of rudimentary horn pregnancy, such as our present case, was reported by Edelman et al. [12] and Cutner et al [13]. Subsequently, several case reports described a successful laparoscopic approach for rudimentary uterine horn [14-18]. Reports have begun to be published on laparoscopic surgery during not only the first trimester of pregnancy [19] but also mid-trimester [4]. Keeping these observations in mind, we planned laparoscopic resection of the rudimentary horn in the present case after obtaining fully informed consent. When laparoscopic resection of a rudimentary horn is performed, care is needed regarding the status of communication between the unicornuate uterus and the rudimentary horn and the arrangement of the ureter. This condition is reportedly complicated by renal or urinary tract malformation in 38% of all cases [20], but no such complication was noted in our present case. Falcone et al. [21] reported that the communication assumes the form of either a loose connection or a firm connection. In the former type, operative manipulation is relatively easy, while the latter type requires manipulation of blood vessels, etc., making the operation more complex. Regarding ureter arrangement, it is essential to take adequate care to avoid ureter injury in cases in which the renal system and urinary tract are free of abnormalities. Detailed assessment of anatomical relationships and the arrangement of adjacent tissues and organs in the operative field by means of magnetic resonance imaging or 3-D computed tomography angiography before rudimentary horn resection reportedly ensures a safe operation [18, 22, 23].

In cases with comorbid endometriosis, freeing of adhesions may be difficult, and abnormal ureter arrangement can make the operation more difficult, raising the risk of complications. In the present case, laparoscopy revealed loose communication between the unicornuate uterus and rudimentary horn, with no signs of adhesion associated with endometriosis. Intraoperatively, the ureter arrangement below the broad ligament could be identified under direct vision. In this way, safe, rapid, and easy resection was possible using the LVSS, with minimal blood loss. The LVSS is a device recently introduced in general and abdominal surgery [24 25]. Applying a precise amount of bipolar energy and pressure to the tissue being dissected allows vessels to the sealed. This sealing process is achieved by changing the nature of the vessel wall itself. With this device, collagen and elastin within the vessel wall fuse into a single structure that obliterates the lumen, thereby reducing the risk of hemorrhage. A feedback-control mechanism, with an automatic “off” switch when the impedance reaches a critical level, ensures that tissues are not damaged due to excessive coagulation. The seal integrity in 3-4-7 mm vessels approximates the burst strength of a ligatures and clip. Moreover, this device resists dislodgment and because the need for dissection before application is minimal, it facilitates dissection. Furthermore of course, no foreign material can be inadvertently left behind. Laparoscopic surgery with LVSS may be viewed as a simple, minimally stressful operative procedure. To date, there have been very few reports describing pregnancy after laparoscopic resection of a rudimentary horn. One report [26] describes a term pregnancy with cesarean delivery necessitated by breech presentation a few months after the procedure.
Women hoping to have children have the expectation of achieving pregnancy in the residual unicornuate uterus. However, since a unicornuate uterus can precipitate abortion, premature labor, and intrauterine growth retardation, the live birth rate from such a uterus is reportedly low (43.7%) [1]. The present case became pregnant naturally in the left unicornuate uterus 4 months after right rudimentary horn resection. We can thus say that, in this case, resection of the rudimentary horn prevented a third rudimentary horn pregnancy. However, it is difficult to demonstrate that it contributed to the subsequent left unicornuate uterus pregnancy. We cannot rule out the possibility that this type of operative manipulation adversely affects the environment of the unicornuate uterus.

However, the unicornuate uterus pregnancy in this case could be maintained, and the fetus grew normally. Despite signs of premature labor, the infant was eventually delivered transvaginally at term. Therefore, surgical resection of a rudimentary horn may be beneficial in selected cases such as ours. Future reports of similar cases will delineate appropriate clinical management.

REFERENCES