CATASTROPHIC HEMORRHAGE AFTER RUPTURE OF RUDIMENTARY HORN PREGNANCY WHICH ONCE MISDIAGNOSED AS UTERUS BICORNIS

(Rudimentary horn pregnancy as if uterus bicornis)

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SUMMARY

A 22-year-old, primigravida, at 18 weeks' gestation presented with an acute onset of abdominal pain, severe hypotension and tachycardia. It has been revealed in her previous medical history that she had been operated with a presumptive diagnosis of ectopic pregnancy and due to the intraoperative misdiagnosis of uterus bicornis, the intact right uterine pregnancy in the rudimentary horn was left in place to continue in another hospital. After the operation, prophylactic cerclage was performed. She was urgently brought to a private hospital and emergency laparotomy was performed for the presumptive diagnosis of intraperitoneal hemorrhage. A significant hemoperitoneum was encountered with the fetus floating freely with the placenta in the peritoneal cavity. The right rudimentary uterine horn had a fundal rupture that necessitated performing a rudimentary uterine and ipsilateral tubal excision. The patient recovered uneventfully.

KEY WORDS: Rudimentary horn, ectopic, rupture

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INTRODUCTION

In a series reported by Heinonen and associates, 11 out of 13 patients with a unicornous uterus had a rudimentary horn and two did not¹. The rudimentary anlage may communicate with the unicornous uterus. Most rudimentary horns are noncommunicating.

There is a risk of pregnancy developing in

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the rudimentary horn from transperitoneal migration of the sperm or ovum from the opposite side. O'Leary and O'Leary found the corpus luteum to be on the side controlateral to the rudimentary horn containing a pregnancy in 8% of cases2. Signs and symptoms of an ectopic pregnancy will develop with eventual rupture of the horn if the pregnancy is not detected early. Rupture through the wall of the vascular rudimentary horn is associated with severe intraperitoneal hemorrhage and shock. Therefore, removal of the rudimentary horn is required as soon as a pregnancy is confirmed. Most pregnancies in a rudimentary horn rupture in the first or second trimester. Fedele and associates have found ultrasonography useful in determining the presence of a rudimentary horn³.

CASE REPORT

A 22-year-old, primigravida, at 18 weeks' gestation presented with an acute onset of abdominal pain, severe hypotension and tachy-

cardia. It has been revealed in her previous medical history that she had been operated with a presumptive diagnosis of ectopic pregnancy. At that time, the patient had a positive test result for pregnancy on the commercially available urinary β-hCG measurement kits. She presented with the complaint of minor pain in the lower abdomen with no signs of hemoperitoneum on an outpatient basis. Upon ultrasonographic examination, a living embryo was detected in an adnexial localization with an empty uterine cavity. As a result, she underwent laparotomy with the presumptive diagnosis of ectopic pregnancy. On surgical exploration, two uterine bulbs with similar sizes were found with intact fallopian tubes. As a result, the patient was thought to be misdiagnosed to have an ectopic pregnancy and the true diagnosis was thought to be uterus bicornis. Moreover, in order to improve the reproductive performance of the patient, a cerclage was performed on the single cervix that the patient had. As a result, the patient was allowed to go on with her rudimentary horn pregnancy left in place. These procedures were performed around 6-7 weeks' of pregnancy. Upto 18 weeks' of gestation, her pregnancy course was uneventful.

In 18 weeks' of gestation, the patient had an acute onset of abdominal pain and loss of consciousness so she was urgently brought to our hospital. Her general condition was poor upon admission. She was confused and almost unconscious. The systolic blood pressure was 40 mmHg and the diastolic was almost 0 mmHg. Her pulse was filiform, weak and 122 bpm. Her extremities were cold and sweaty. So all her symptoms and signs were indicative of an intraperitoneal hemorrhage and shock necessitating an emergency laparotomy.Blood product replacement was urgently initiated and emergency laparotomy was performed for the presumptive diagnosis of intraperitoneal hemorrhage. Upon laparotomy, significant hemoperitoneum was encountered with the fetus floating freely with his placenta in the peritoneal cavity. A unicornous uterus and a ruptured right rudimentary horn were detected. The right rudimentary uterine horn had a fundal rupture and it necessitated performing a rudimentary uterine and ipsilateral tubal excision. The figure shows the pathologic specimen. In the post-operative period, the patient recovered uneventfully and was dismissed in three days after the operation.

DISCUSSION

Rudimentary uterine horn pregnancy is a very rare condition. According to Holden and Hart, some 350 cases of pregnancy in a rudimentary horn have been reported since the original case report by Mauriceau in 16694. Signs and symptoms of an ectopic pregnancy will develop with eventual rupture of the horn if the pregnancy is not detected early. Soundararajan and Rai reported a case of rudimentary uterine horn that presented during pregnancy and mimic an ectopic pregnancy⁵. In this case, the horn was removed laparoscopically. Fedele and associates have found sonography useful in determining the presence of not only rudimentary horn but also a cavity within3. Also in 1998, Yahata et al. reported the laparoscopic management of uterine horn pregnancy mimicing an ectopic pregnancy but in this case a rudimentary horn pregnancy was suspected from transvaginal ultrasonography⁶. Kriplani et al. reported a case of pregnancy in the rudimentary horn of a unicornuate uterus, suspected on routine pelvic examination and confirmed by sonography as early as at 8 weeks of gestation7. Laparotomy and surgical management of ipsilateral



Figure 1: The ruptured rudimentary horn with the fetus and its placenta.

adnexa at the time of excision of rudimentary horn were represented.

In our case, the patient did not have the signs of hemoperitoneum on first presentation but the positive pregnancy test and an empty uterine cavity led to the misdiagnosis of ectopic pregnancy. Ultrasonographic examination did not help for the diagnosis of rudimentary horn pregnancy. Since rudimentary horn pregnancy is a very rare condition, it is not easy to gain experience in diagnosing this entity on ultrasonographic investigation. Although ultrasonography is reported to be a useful tool in diagnosing rudimentary horn pregnancy, this may not be the case in unexperienced hands.

Rupture of the uterus in nulliparous patients is generally associated with müllerian anomalies. A unicornuate uterus with a rudimentary horn is a müllerian anomaly associated with endometriosis and pregnancy complications, including miscarriage, ectopic pregnancy, uterine rupture, preterm labor and malpresentation. Therefore, the horn is removed if it is thought to contain functional endometrium. This is usually done by laparotomy in the nonpregnant state. Unfortunately and generally, the condition becomes overt in the course of the first pregnancy due to the onset of related complications. Our case also presents a nulliparous woman presenting with spontaneous rupture of the rudimentary horn pregnancy. No symptoms and signs were indicative of her müllerian anomaly before her pregnancy. Besides, since müllerian anomalies are also rare entities, it seems possible to have some misdiagnoses. In this case, since the rudimentary anlage reached nearly to the size of the unicornous uterus, the patient was misdiagnosed to be having a uterus bicornis. We also know that there's some debate on whether the unicornous uterus with a communicating horn may represent a hypoplastic side of a bicornuate uterus. In these kind of cases, the only way for differential diagnosis is vaginal examination both for the presence of vaginal septum and the number of colli. In our patient, eventhough a prophylactic cerclage was performed on the single collum that the patient had, the definitive diagnosis was still uterus bicornis. The only explanation for this misdiagnosis may be its rarity.

In 1998, Dicker et al. reported the case of a woman who benefited from laparoscopic surgery of a rudimentary horn pregnancy. Laparoscopy, in this exceptional case, was said to be the most accurate diagnostic tool that carries significant advantages in effective surgical management, thereby avoiding laparotomy. So laparoscopy seems to be a reasonable option both for diagnostic and therapeutic purposes in patients suspected to have an ectopic or rudimentary horn pregnancy.

Although ultrasonography seems to be a useful tool for the diagnosis of unicornous uterus with a rudimentary horn, it may not be so due to inexperience and its rarity. It's possible to conclude that in any patient presenting with the symptoms of unruptured or ruptured ectopic pregnancy, rudimentary horn pregnancy should always be sought upon ultrasonographic examination. The differential diagnosis is utmost important both for the management and reproductive life of the patient.

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