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Case Report

Rudimentary Corneal Rupture of A Unicornuate Uterus in A 23-Week Pregnancy: About A Case

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Abstract

The rudimentary uterine horn belongs to the group of major uterine malformations according to the classification of Musset's classification. This rudimentary horn may have a cavity with endometrial lining of the uterus, and thus can serve as a site of implantation during pregnancy. The presence of a pregnancy in a rudimentary horn is a rare event, which can be revealed by a uterine rupture. It leads to an emergency fetal extraction and it is then recommended to perform an exeresis of this rudimentary horn [6, 7]. If it is not performed, it theoretically exposes the risk of recurrence of uterine rupture in case of a new pregnancy in this rudimentary horn. There is a need to increase awareness of this disease, especially in developing countries where the possibility of detection of the disease is limited. We report a case of ruptured rudimentary horn pregnancy in shock at 23 weeks of gestation misdiagnosed as intrauterine pregnancy. The pregnancy was located in a rudimentary left uterine horn ruptured on its posterior surface and communicating with the right horn. A hemi hysterectomy was then performed with the removal of the rudimentary horn and the homolateral adnexa. The fetus was deadborn. The postoperative course was simple and the patient was discharged on the fifth postoperative day.

Keywords: Uterine Rupture, Uterus unicornis, Rudimentary Uterine Horn.

INTRODUCTION

Congenital anomalies of the uterus are caused by failure of fusion of the two Mullerian ducts, complete or partial persistence of the septum between them, or failure of one side to develop. The incidence of uterine congenital anomalies because of Mullerian defects in the normal fertile population is 3.2% [1].

The rudimentary uterine horn belongs to the group of major uterine malformations according to the classification of Musset's classification [2].

This rudimentary horn may have a cavity with endometrial lining of the uterus, and thus can serve as a site of implantation during pregnancy [3, 4]. The occurrence of a rudimentary horn pregnancy is estimated to be 1/100 000 à 1/140 0002. This pregnancy most often leads to rupture of the rudimentary horn, especially in the second trimester, and is responsible for a maternal mortality of <0.5%1 and a very high neonatal mortality [5]. It may be also be associated with gynecological and obstetric complications like infertility, endometriosis, hematometra, urinary tract anomalies, abortions, and preterm deliveries.

The presence of a pregnancy in a rudimentary horn is a rare event, which can be revealed by a uterine rupture. It leads to an emergency fetal extraction and it is then recommended to perform an exeresis of this rudimentary horn [6, 7]. If it is not performed, it theoretically exposes the risk of recurrence of uterine rupture in case of a new pregnancy in this rudimentary horn.

We report a case of ruptured rudimentary horn pregnancy in shock at 23 weeks of gestation misdiagnosed as intrauterine pregnancy.

CASE REPORT

We report the case of patient F., 25 years old, G2 P1, with a live child born by vaginal delivery, with no notable medical or surgical history. Pregnancy follow-up was unremarkable, a first trimester ultrasound performed at 12 weeks of amenorrhea was considered normal.

The patient presented to the emergency room of the Souissi Maternity Hospital in Rabat with acute abdominal and pelvic pain during a pregnancy of 23 weeks of amenorrhea, with nausea and vomiting, as well as mucocutaneous pallor. The clinical symptomatology had started a few days before her admission with diffuse abdominal pain.

The hemodynamic state was unstable with dyspnea, a blood pressure of 08/06 mm Hg, a pulse of 110 beats per minute and cutaneous-mucosal pallor; the clinical examination revealed a painful abdomen with defense, a uterus of normal volume, painful to mobilization, with a perceived latero-uterine mass, without other associated signs.

The patient was rushed to the operating room, where an obstetrical ultrasound was performed at the same time as the conditioning process, which showed a right latero-uterine mass, probably ruptured, containing a fetus with no cardiac activity and a small to medium peritoneal effusion (Fig 1). The diagnosis of suspected uterine rupture at 23 weeks of amenorrhea was therefore retained.

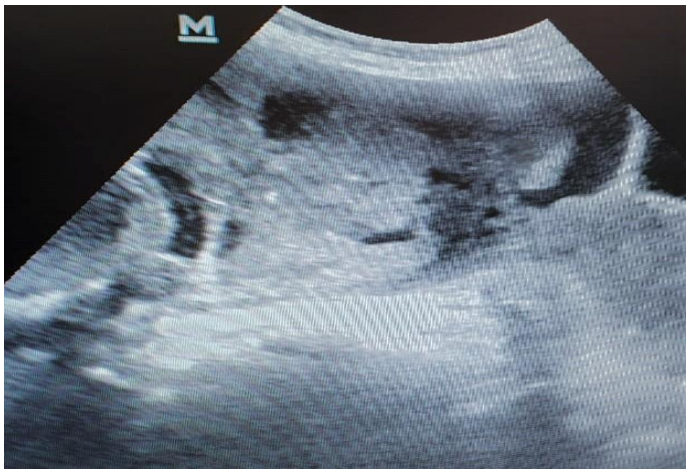

Fig 1: Ultrasound image of a ruptured rudimentary horn pregnancy at 23 weeks of gestation

In front of this state of hemorrhagic shock, it was decided to perform an emergency laparotomy. This revealed a moderate profusion of hemoperitoneum.

The pregnancy was located in a rudimentary left uterine horn ruptured on its posterior surface and communicating with the right horn. The left tube and the left ovary, macroscopically normal, were implanted on this rudimentary horn (Fig 2). A hemi hysterectomy was then performed with the removal of the rudimentary horn and the homolateral adnexa. The fetus was deadborn. It was female, weighed 600 grams and had no visible morphological abnormalities.

The postoperative course was simple and the patient was discharged on the fifth postoperative day.

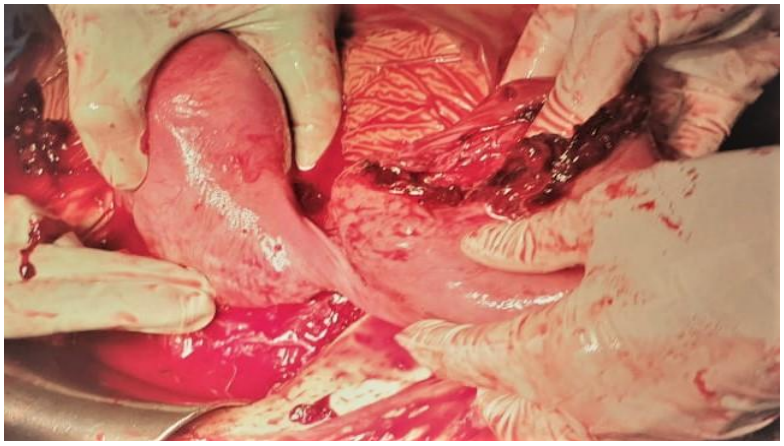

Fig 2: Preoperative image of a ruptured rudimentary horn of a unicornuate uterus at 23 weeks

DISCUSSION

Uterine malformations affect 0.5% of women. Five percent of these are unicornuate uterus [3]. The incidence of rudimentary horn pregnancies is estimated to be 1/100,000 to 1/140,000 [4]. Embryologically, pseudo-unicornuities result from a developmental defect of one of the two Müllerian ducts. They are not common and represent about 12% of uterine malformations [2, 8]. Only some of them have a rudimentary canaliculated horn lined with a functional endometrium allowing the implantation of an egg [2].

In 90% of cases, there is no communication with the normal horn according [9]. The absence of menstrual retention can be explained by the aplasia of the isthmus, which is necessary for the menstrual period to start.

There is a slight predominance of this rudimentary horn on the right side probably due to the fact that the left Müllerian duct progresses more caudally than the right [7].

This major uterine malformation is associated in 10% of cases with urinary malformations such as homolateral often-unilateral renal agenesis but sometimes also a pelvic kidney or horseshoe kidney [8] and sometimes with bone malformations and/or ovarian malformations.

Outside of pregnancy, pseudo unicornuate uterus is the most difficult uterine malformation to detect by ultrasound and it is laparoscopy that formally determines this type of uterine malformation. Our patient presents an agenesis of the right kidney as shown on the MRI images (Fig).

MRI can certainly be a significant help in the diagnosis [10]. This radiographic workup is performed in the presence of dysmenorrhea, unexplained infertility or recurrent abortions, or as an incidental finding during an operation. In the first three months of pregnancy, one may be alerted by a latero-uterine mass corresponding to the small gravid horn next to the normal horn. This mass suggests an ectopic pregnancy or an ovarian cyst, and laparoscopy is proposed when there is doubt about the location of the pregnancy or the nature of the uterine malformation [10, 11].

The first case of uterine rupture associated with a rudimentary horn was reported in 1669 by Mauriceau [12]. The time of rupture varies from 5 to 35 weeks depending on the musculature of the horn and its ability to hypertrophy and dilate.

Hemorrhage is greater in rudimentary horn pregnancy rupture because the uterine wall is thicker and more vascular [13]. Rudimentary horn rupture is the most significant threat to the pregnancy and a life-threatening situation [14].

Early diagnosis of this pregnancy is critical and can be difficult. Ultrasound, hysterosalpingography, hysteroscopy, laparoscopy and MRI are diagnostic tools [15].

The sensitivity of ultrasound is only 26% and decreases as the pregnancy progresses [16]. Rudimentary horn pregnancy is often mistaken for tubal, horn, intrauterine or abdominal pregnancy [17].

The non-specific symptomatology, mainly severe abdominal pain in the second trimester, makes the diagnosis of pregnancy in a uterine horn difficult, often associated with intraperitoneal hemorrhage, which can be revealed by ultrasound.Rupture often leads to emergency surgery, during which the diagnosis is usually made [6]. Earlier detection of uterine malformations can be helpful in preventing this devastating obstetrical event that can result in maternal mortality [18].

Outside of pregnancy, the discovery of a bicornuate uterus with a rudimentary horn should lead to excision of the rudimentary horn, when possible. This procedure can be performed by a laparoscopic approach [19, 20].

If a pregnancy in a rudimentary horn is diagnosed in the first trimester, it may be reasonable to continue the pregnancy with ultrasound monitoring until close to 28 weeks to allow fetal lung maturation. Surgical intervention may be performed when the estimated fetal mass is greater than 1000 grams or if the myometrial thickness at any point in the wall is less than 5 mm [21]. But immediate surgery is recommended by most physicians after diagnosis, even in unruptured cases [16]. Medical management with methotrexate and its resection by laparoscopy are also reported [22].

These pregnancies are also at risk for placenta accreta and percreta [23, 24].

CONCLUSION

Prenatal diagnosis of pregnancy on rudimentary horn remains difficult to establish despite the progress of ultrasound and other diagnostic modalities, the diagnosis of confirmation being laparotomy. Early diagnosis of a rudimentary horn pregnancy is usually accidental.

Reanimation, surgery and timely blood transfusion are necessary to save the patient. Appropriate diagnostic methods and early referral to level 3 hospitals are necessary to reduce patient morbidity and mortality.

There is a need to increase awareness of this disease, especially in developing countries where the possibility of detection of the disease is limited.

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