



Case Report

Recurrence of Uterine Rupture in a Pseudo-Unicornuate Uterus at 17 Weeks of Amenorrhea: Case Report and Literature Review

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Received for publication on 29 October 2008. Accepted in revised form 13 November 2008

Key words: Rudimentary uterine horn, Pregnancy, Uterine rupture, Pseudo-unicornuate uterus, Recurrence

ABSTRACT

Pregnancy in a rudimentary horn is a very rare condition. It is responsible for several complications. Prognosis is reserved because the natural evolution generally leads to a cataclysmic uterine rupture at the beginning of the second trimester. Classically, the treatment after foetal extraction consists of ablation of the rudimentary horn and associated fallopian tube. We report the obstetric outcome of a patient with history of rudimentary uterine horn rupture, the treatment of which was ablation of the rudimentary horn.

INTRODUCTION

Pregnancies occurring in a malformed uterus are relatively rare [1]. Even more rare are those occurring in a rudimentary uterine horn. The frequency of a pregnancy in a rudimentary uterine horn is about one in 150,000 pregnancies [2]. This leads to an urgent foetal extraction and it is then recommended to carry out a rudimentary horn excision. If the latter is not realized, it exposes the patient to the risk of a repeated uterine rupture in the event of a new pregnancy in the rudimentary horn.

CASE REPORT

A 20 year old female, G2P0, presented to the emergency department at the end of her 17th week of amenorrhea with violent abdominal pain. Her first pregnancy was marked by the appearance of an intense pelvic pain occurring at 18 weeks in the context of hemorrhagic shock.

During laparotomy, the amniotic membranes were discovered bulging into the abdominal cavity from a breach in the ruptured left rudimentary uterine horn in what was discovered to be a pseudo-unicornuate uterus. Treatment consisted of ablation of the left rudimentary horn, with excision extending to its junction with the right hemi-uterus. The current pregnancy was unbooked at presentation. Its

evolution was marked by the appearance of pelvic pain at 17 weeks gestation. There was no bleeding but the patient was noted to have malaise and fatigue. On clinical examination, the blood pressure was 90/50 mm/Hg, pulse was 100 bpm, and there was mucocutaneous pallor. The abdominal examination showed pelvic guarding, and both flanks were dull to percussion. The gynaecological exam found a bulky uterus with cervical excitation and pelvic peritonism. Physical assessment revealed a hemoglobin of 6 g/dl. Ultrasound scan showed a uterus of normal structure with an empty cavity, placenta, and a non-viable pregnancy of 17 week's gestation outside the myometrium. They also noted a large intra-peritoneal effusion, suggesting an abdominal pregnancy (**figure 1**).

An urgent lower midline laparotomy was carried out together with simultaneous resuscitation of the patient. The surgical findings included a hemoperitoneum of two liters, a full thickness rupture of the uterine wall measuring 6 cm involving the remaining hemi-uterus, and a dead foetus with a placenta showing no sign of placental insertion in the abdomino-pelvic cavity (**figure 2**). After extraction of the foetus and placenta, repair of the hemi-uterus was carried out by way of interrupted stitches in two layers. The post

operative course was uncomplicated and the patient was discharged on the 7th post-operative day.

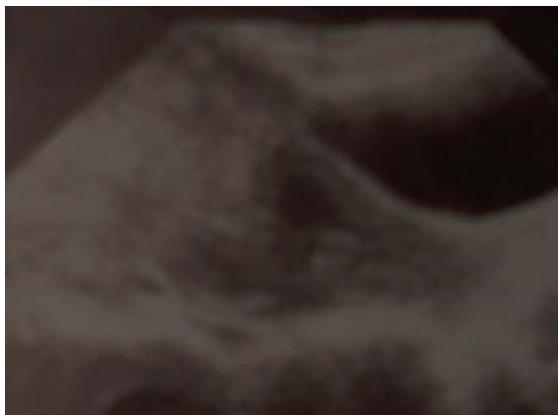


Figure 1: Ultrasound scan showing a uterus of normal structure with an empty cavity, a placenta, and a non-viable foetus of 17 week's gestation outside the myometrial structure.

Discussion:

The embryological origin of pseudo-unicornuate uteri is a lack of development of one of the two Müllerian tubes. This entity accounts for only 10% of uterine malformations [2,3] and only a proportion of these have a patent rudimentary horn lined with functional endometrium [2]. Co-existing urinary tract abnormalities are frequent in these patients and are dominated by unilateral renal agenesis, homolateral to the side of the rudimentary horn [2]. The incidence of the unicornuate uterus, although difficult to specify, is estimated at 1/1000 women [2]. Outside pregnancy, the rudimentary horn is the most difficult uterine malformation to detect. The occurrence of a hematometrium is infrequent, and this lack of menstrual retention in the rudimentary horn could be explained by isthmic aplasia, or by the paucity of bleeding from the often hypoplastic endometrium [4]. The diagnosis should be considered in cases of severe dysmenorrhea, unexplained



Figure 2: Full thickness uterine rupture of 6cm involving the remaining hemi-uterus, and a dead foetus with a placenta presenting no sign of placental insertion in the abdomino-pelvic cavity

infertility, or miscarriages with repeated premature births, intra-uterine growth restriction, and hypertension in pregnancy, which all merit radiological assessment.

The clinical diagnosis of rupture of a hemi-uterus is difficult. The only constant clinical sign is an intense, brutal abdominal pain which may or may not be associated with a hemoperitoneum. Sometimes, there is hemorrhagic shock imposing an urgent laparotomy for maternal and possibly foetal rescue depending on the gestation. Pelvic examination during the first trimester can raise the alert, with the finding of a latero-uterine mass corresponding to the small gravid horn beside the normal horn [5]. Ultrasound confirms the diagnosis by showing a pregnancy surrounded by a variable thickness of myometrium which is usually absent in the event of tubal pregnancy.

The diagnosis can also be refined by the use of MRI [6]. Laparoscopy allows formal confirmation of this type of uterine malformation if there is doubt about the localization of the pregnancy, or the nature of uterine malformation [9]. The prognosis of

rudimentary horn pregnancies remains reserved. Indeed, the muscle of the rudimentary horn is particularly delicate because it is so thin. Moreover, non-functional endometrium usually provokes pathological placentation. Thus, the pregnancy generally evolves towards rupture in the beginning of the second trimester bringing about a hemorrhagic acute abdominal syndrome similar to a cataclysmic ruptured ectopic pregnancy [7,8].

The treatment of the rudimentary horn pregnancy, whether complicated or not, classically involves foetal extraction, followed by resection of the rudimentary horn and of its homolateral tube. Certain authors even propose to carry out this procedure on diagnosis of a pseudo-unicorn uterus with rudimentary horn in the non-gravid state, to avoid an obstetrical accident. Future pregnancies would then require extremely close follow up. The patient should be informed and aware of the incurred risks. Obstetricians caring for these patients should be aware of the serious risk of uterine rupture during pregnancy. A Caesarean section prior to labour is strongly recommended [10].

CONCLUSIONS

The rudimentary horn pregnancy is a very serious condition which may put the maternal life in danger. Improving the outcome is based initially on the diagnosis and pre-conceptual care for this malformation. The treatment will consist of an urgent laparotomy with foetal extraction and ablation of the ruptured horn in order to avoid a repeated, potentially more severe incident.

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To cite this article: Errarhay S, Mahmoud S, Bouchikhi, *et al.* Recurrence of Uterine Rupture in a Pseudo-Unicornuate Uterus at 17 Weeks of Amenorrhea: A Case Report and Literature Review. *Libyan J Med.* 2009; 4:53-55. AOP:081115