

ASYMPTOMATIC HORN RUDIMENTARY PREGNANT UTERINE RUPTURE WITH A VIABLE FETUS.

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RUPTURE ASYMPTOMATIQUE D'UNE CORNE RUDIMENTAIRE AVEC FOETS VIABLE SUR GROSSESSE

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R É S U M É

Prérequis : la rupture d'une grossesse sur corne utérine rudimentaire entraîne un hémopéritoneum abondant et grave, lors du 2^{ème} trimestre de la gestation.

L'objectif de ce travail est de décrire les éléments de diagnostic et les concepts actuels de la prise en charge de la grossesse abdominale avancée secondaire à une rupture de corne rudimentaire.

Cas : Les auteurs décrivent un cas de grossesse abdominale avancée évolutive secondaire à une rupture asymptomatique d'une corne rudimentaire chez une deuxième gémelle adressée pour anamnios sur un terme estimé à 30 semaines. Le diagnostic a été évoqué devant la présence d'un fœtus en présentation oblique avec perception de la tête dans l'hypochondre gauche et la présence à l'échographie d'un utérus vide contigu à une masse contenant le placenta. Une laparotomie a été programmée permettant l'extraction d'une fille pesant 1250g avec Apgar 5/9 à la 1^{ère} et 5^{ème} minute. Une hémihystérectomie emportant la corne rudimentaire avec le placenta était réalisée sans difficultés.

Conclusion : L'association à un anamnios d'une présentation irrégulière avec aspect inhabituel du placenta doit faire évoquer le diagnostic de grossesse abdominale avancée. Une laparotomie programmée permet d'augmenter les chances de survie materno-fœtale.

M O T S - C L É S

grossesse abdominale - corne rudimentaire - rupture utérine - diagnostique - traitement

S U M M A R Y

BACKGROUND: Rupture of pregnant rudimentary horn in a pseudo-unicornuate uterus is usually resulting in severe and dramatic haemoperitoneum at the beginning of the second trimester of gestation. Advanced abdominal pregnancy due to horn rudimentary uterine rupture with delivery of a viable fetus is exceptional.

AIM OF THE STUDY: To analyse obstetrical entailments, diagnosis and current concepts of management of advanced abdominal pregnancy secondary to rudimentary horn rupture.

CASE: An asymptomatic ruptured rudimentary horn pregnancy in a 31-year-old, second gravida, is reported. She was referred to our maternity for anhydramnios at estimated gestational age of 30 weeks. An advanced abdominal pregnancy was diagnosed with sonographic features suggestive of horn rudimentary uterine rupture. Elective laparotomy was performed and a healthy infant was delivered. Excision of the rudimentary horn was done and an uneventful recovery followed.

CONCLUSION: An unusual sonographic appearance of the placenta with anhydramnios must first lead to consider the diagnosis of advanced abdominal pregnancy in time to save the surgeon from an unpleasant and dangerous surprise and to increase the chance of materno-fetal survival.

K E Y - W O R D S

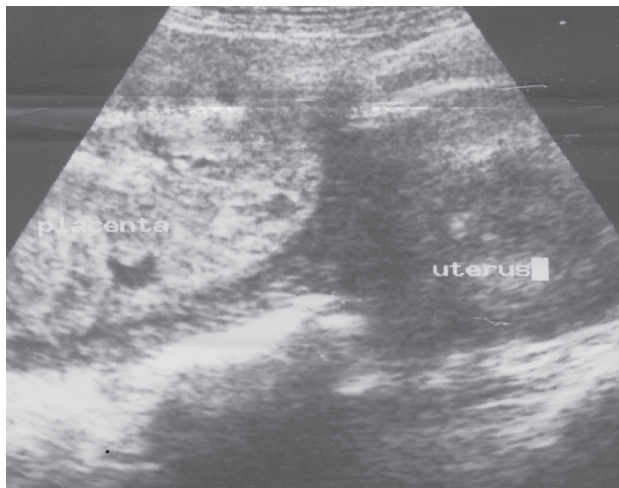
advanced abdominal pregnancy; rudimentary uterine horn; rupture; diagnosis; management.

Advanced abdominal pregnancy (AAP) is defined as a pregnancy of over 20 weeks' gestation with a foetus living, or showing signs of having once lived and developed, in the mother's abdominal cavity[1, 2]. It is encountered in 1 in 25 000 births [1]. Advanced abdominal pregnancy secondary to horn rudimentary pregnant uterine rupture is extremely rare, as this rupture usually results in acute and severe haemoperitoneum at the beginning of second trimester of gestation. Though relatively rare, this situation can have dramatic and catastrophic consequences for the foetus and the mother especially when underdiagnosed.

CASE

A 31 year-old woman, gravida 2 with one living child, was referred to our maternity center for anhydramios with breast feeding amenorrhea. She had a history of term breech delivery without diagnosing any uterine malformation at uterine revision. She had had no antenatal visits nor scan during this pregnancy. Moreover, interview didn't retrieve any history of abdominal pain or vaginal bleeding. On examination she was in no evident pain or distress. An abdominal mass presumed to be the uterine fundus was palpated at 27 cm above the symphysis pubis. The fetal presentation was not clear. No foetal heart sounds were heard. Ultrasound examination revealed a female viable fetus in an oblique lie, the fetal head next to the mother's stomach, and the parameters corresponded to 30 weeks of gestation. There was an irregular mass, adjacent to the empty uterus (fig.1), in which was recognized a placenta. No visible

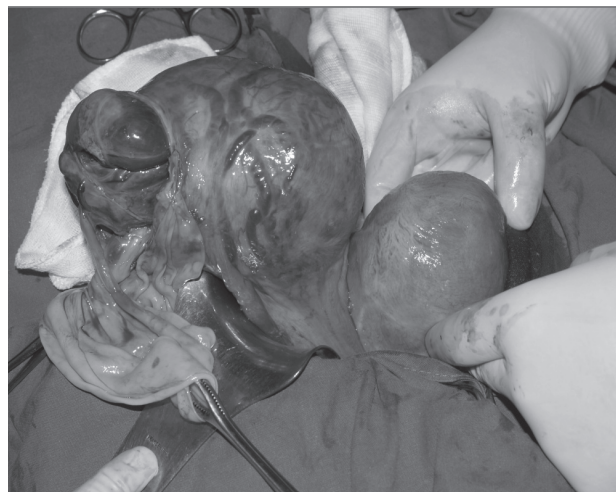
Figure : 1: Mass containing placenta adjacent to an empty uterus.



amniotic sac was seen. The estimated fetal weight was 1300g. The diagnosis of advanced abdominal pregnancy secondary to horn rudimentary rupture was made and magnetic resonance imaging was thought to be unnecessary. Fetal corticotherapy was administered and laparotomy was planned 48 hours later, in the presence of experienced members of our team. At the exploratory laparotomy, performed under general anaesthesia through a sub-umbilical median incision that extended 3 cm

above the umbilicus, the peritoneal cavity was occupied by a lying viable fetus in oblique position. The fetal head was in contact with the maternal stomach; the breech was astride a healthy placenta protruding out of the horn rudimentary uterine rupture. The placenta was attached to the ruptured rudimentary horn deriving blood supply from it. No putrid odour was noted. There was no active bleeding from the edges of the rupture (fig.2). The foetus was removed without difficulty. The baby

Figures : 2: Placenta protruding out of the horn rudimentary uterine rupture.



was weighing 1250g with Apgar scores of 5 at 1 minute and 9 at 5 minutes and had no evident deformity. Omentum was discharged from adherent membranes by manual separation without hemorrhage and hemihysterectomy with homolateral salpingectomy was performed without difficulties, allowing complete removal of rudimentary horn, placenta and membranes (fig.3). Blood loss was estimated to be 200 mL. No

Figure : 3: Hemihysterectomy with homolateral salpingectomy.



transfusion was given. The mother was given ceftriaxone for 7 days, she was discharged well on post-operative day 8. Mother

and baby were in excellent health when last seen for routine follow up two years later.

In summary, this was an interesting case of advanced secondary abdominal pregnancy due to an asymptomatic rupture of a horn rudimentary uterus that resulted in a viable extrauterine pregnancy.

DISCUSSION

The incidence of advanced abdominal pregnancy is 1 in 25000 births [1]. Few cases of advanced pregnancy secondary to horn rudimentary uterine rupture were reported. Rupture of horn rudimentary pregnant uterus occurs usually at the beginning of the second trimester and results in severe acute haemoperitoneum.

The interesting aspect of this case was that the rupture was postulated to have occurred at the beginning of the second trimester but the patient didn't complain of abdominal pain or clinical signs of hemoperitoneum. The review of reported cases of advanced abdominal pregnancy secondary to horn rudimentary rupture revealed constant presence of abdominal pain at the presumed moment of rupture [2, 3, 4, 5, 6]. Asymptomatic rupture in our case can only be explained by a chronic disruption in the distended wall of the rudimentary horn without significant hemorrhage. Maternal face of placenta that remained implanted in the rudimentary horn made the growing of the fetus possible. Although we didn't have an accurate estimated age in our case, we supposed absence of growth retardation and examination of the neonate confirmed prenatal gestation age of 30 weeks gestation. Moreover, despite the uterine rupture had resulted in sac amniotic rupture, fetal growth was good and malformation or deformity were absent. This is not surprising because the fetus was still deriving its blood supply from the uterus and the peritoneal cavity made fetal movements possibles. However, the chance of fetal survival is reported to be dismal in secondary advanced abdominal pregnancy, with the mortality ranging from 40% to 95% [7]. There is also a high rate of congenital malformations of 30-90% [7]. Moreover, maternal mortality has been reported to be about 0.5 -18% in such cases. This poor reported outcome is mainly due to high diagnostic error in this type of case, and the literature has shown that the diagnosis was missed in approximately 50-90% of cases [7]. The ability to make the correct diagnosis is based on the level of skill of the medical care provider and a high index of suspicion including history, physical examination and imagery. So that, the diagnosis should be clinically suspected when a history of bleeding or excessive abdominal pain during the beginning of second trimester, a history of previous abortion or breech delivery, abnormal fetal lie or abdominal mass palpated apart from the fetus are present. Abnormal relationship among the fetus, placenta and uterus, oligo-anhydramnios and fetal malpresentation especially transverse lie should alert sonographer. Magnetic resonance imaging could have been of help in the diagnosis, localizing the area of implantation of the placenta and most importantly, its vascular supply and relationship to the pelvic organs [8, 9]. In our case collegial decision was to not perform MRI because

diagnosis was evident and mobilization of woman could expose her to haemorrhagic complications.

Because perinatal death may result from either prematurity or prolonged gestation in a compromised environment, the decision about when to intervene in the case of a live baby remains unanswered. Once the foetus has reached a viable age, there is little reason to delay delivery [10, 11]. Regardless of timing, the mother's own safety will be best assured by careful monitoring, foresight and pre-operative preparation. The principal controversy concerning management of AAP is whether or not to remove the placenta. Because the abnormally implanted placenta's blood supply is diffuse and often unidentifiable, attempts to remove it can incite catastrophic haemorrhage. Measures taken to control this haemorrhage during surgery risk compromising the blood supply of other organs. A placenta left in situ might resorb spontaneously but if it does not, the risk of infection, necrosis, and the need for a second surgery is considerable. Most authors agree that the placenta should be removed provided its blood supply is identified and can be ligated without damaging other organs [10, 11, 12, 13]. If the blood supply cannot be identified and safely ligated, the placenta should be left in place and the patient followed for possible complications. Whether adjuvant methotrexate or selective arterial embolization is beneficial remains uncertain [11, 14].

In this particular case, fortunately, there was no dilemma with regard to the management of the placenta because it was located still within the uterine cavity and membranes were fairly attached to omentum.

The diagnosis of pseudo-unicornuate uterus should have been made after the first delivery and preventive hemihysterectomy (with homolateral salpingectomy) by laparoscopy should have been performed. Some authors suggest simple unilateral tubal ligation in cases of non communicating rudimentary horn [15].

CONCLUSION

Advanced abdominal pregnancy secondary to horn rudimentary rupture is extremely rare. Pre-operative diagnosis is especially important; it gives the medical team time to review management and possible complications of AAP. As obstetricians, one must always bear in mind that when a pregnant patient, with history of breech delivery, presents with oligo-anhydramnios especially with the fetus in an abnormal lie, it is important for an experienced sonographer with good equipment to search for an empty uterus with adjacent mass containing placenta around the fetus to minimize the risks to both mother and fetus.

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