Novel diagnostic procedure

The value of hysteroscopy in the diagnostic approach to a rudimentary horn pregnancy

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Summary

A 33-year-old woman presented with an ectopic pregnancy without any complaints. Laparoscopy was performed since a tubal pregnancy was expected. However, both fallopian tubes appeared normal and it was not possible to differentiate accurately between a pregnancy in a non-communicating horn and a pregnancy in a bicornuate uterus. We therefore performed MRI which showed a thin myometrium around the pregnancy. In order to differentiate between a communicating and a non-communicating uterine horn the authors performed a hysteroscopy. Since there was only one cervical os, and an entrance to the second uterine cavity was not seen along the cervical canal, it was concluded that this pregnancy was situated in a non-communicating rudimentary horn. The non-communicating uterine horn, with the pregnancy in situ, was completely removed. Since a pregnancy in a bicornuate uterus is viable in contrast to a pregnancy in a non-communicating horn, accurate diagnosis is important.

BACKGROUND

A unicornuate uterus with a rudimentary horn is a rare congenital müllerian anomaly which arises as a result of the arrested development of one of the two müllerian ducts. Rudimentary horns are found in 74% of unicornuate uteri and 70–90% are non-communicating horns.¹ The overall incidence of a unicornuate uterus with a rudimentary horn is 1:5400.¹ A pregnancy in a rudimentary horn of the uterus is a rare condition which is assumed to occur through intra-peritoneal migration of either sperm (ipsilateral ovulation) or a fertilised ovum (contralateral ovulation).

The incidence of a pregnancy in a rudimentary uterine horn is estimated to range from 1:74 000 to 1:140 000 pregnancies.^{1–2} This type of ectopic pregnancy has a poor outcome with rupture rates estimated to be around 80–90% in the second trimester of pregnancy.^{3–5} Because of this significant threat, excision of the rudimentary horn is necessary to prevent maternal morbidity, preferably before pregnancy but otherwise whenever the diagnosis is made. The prenatal diagnosis of a pregnancy in a rudimentary horn is, however, very difficult, even when the combination of diagnostic techniques that is advocated in the literature (ultrasound, MRI and laparoscopy) is utilised.⁵ Only 5% of rudimentary horn pregnancies in the literature were diagnosed preoperatively.⁴ In this case report, we discuss diagnostic considerations and present our prenatal approach with the use of hysteroscopy in order to differentiate between a pregnancy in a rudimentary horn and a pregnancy in a bicornuate uterus. Hysteroscopy has only been described in the pregestational diagnosis of a rudimentary horn in combination with hysterosalpingography and laparoscopy.^{4–5}

CASE PRESENTATION

A 33-year-old woman presented without complaints at 9 weeks and 2 days amenorrhoea for her first antenatal check up. This was her third pregnancy: she had had one spontaneous abortion and her second pregnancy was uneventful. She had delivered at term by caesarean section because of breech presentation of the fetus. During the caesarean section there appeared to be a bicornuate uterus with the pregnancy in the left uterine horn. After delivery no further analysis to confirm the diagnosis or to investigate related anomalies (eg, urinary system anomalies) had been performed. At the time of her visit she had no complaints, in particular no signs of vaginal bleeding or abdominal pain.

INVESTIGATIONS

Vaginal ultrasound showed an intact extra-uterine pregnancy with crown–rump length according to duration of amenorrhoea. Therefore a laparoscopy was performed since a tubal ectopic pregnancy was expected. However, the right horn of the bicornuate uterus showed an increased blood vessels and livid colour, indicating that the pregnancy was situated in this horn (figure 1). Since the myometrium appeared to be very thin, the possibility of a rudimentary horn was postulated and a pelvic MRI was performed which showed that the myometrium around the pregnancy was very thin. Moreover the cervical canal of the uterine horn with the pregnancy could not be seen (figure 2). This combination of findings is highly suggestive of a pregnancy in a rudimentary horn of a bicornuate uterus. The patient was referred to a university clinic for further analysis and treatment.

Figure 1 The rudimentary uterine horn with the pregnancy as observed with laparoscopy. Note the dominant vessels, resembling a placenta percreta, on the serosa and the livid red colour of the myometrium.

Figure 2 MRI showing no cervical canal between the uterine horn with the yolk sac and the cervical canal of the other uterine horn, suggesting a pregnancy in a rudimentary horn of a unicornuate uterus.

Figure 3 The rudimentary uterine horn after resection.

Figure 4 The rudimentary uterine horn after resection, opened at the fundus showing the fetus in the yolk sac.

Since the pregnancy was very welcome and desired by the patient and her husband, the diagnosis had to be definite before excision of the rudimentary horn with the pregnancy. Therefore a diagnostic hysteroscopy was performed in order to differentiate between a communicating and a non-communicating uterine horn. During the hysteroscopy (vaginoscopic approach under general anaesthesia), only one cervix with a single external cervical os was observed. The left uterine cavity was visualised without any difficulties and the left tubal ostium was seen. The endometrium appeared to be decidualised and no entrance to the right uterine horn was observed, either in the left uterine cavity or along the cervical canal. The results of the laparoscopy and the pelvic MRI excluded the possibility of a viable pregnancy in a bicornuate uterus.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis was pregnancy in a non-communicating rudimentary horn or a viable pregnancy in a bicornuate uterus.

TREATMENT

The rudimentary horn with the pregnancy and fallopian tube were removed by laparotomy. Laparoscopy was not performed because of the need for intra-abdominal morcellation and the consequent risk of spill of trophoblast and persistent trophoblastic disease.

OUTCOME AND FOLLOW-UP

Pathological examination showed a uterine horn with a thin (12 mm maximum thickness) myometrium with neither endocervical tissue nor an internal cervical ostium. The fallopian tube as well as the fetus were normal (figures 3 and 4). Postoperative recovery was uneventful and the patient was pregnant again 2 months after surgery.

DISCUSSION

A unicornuate uterus is a result of aplasia of one of the müllerian ducts. In 75% of such cases there is incomplete fusion of the müllerian ducts and this anomaly is associated with various degrees of rudimentary horns connected to a unicornuate uterus. As in our case, the majority of rudimentary horns contain a functional endometrial cavity without connection to the unicornuate uterus.^{1 5 6} There is a predominance of right-sided rudimentary horns which may be caused by the slightly advanced development of the left müllerian duct.¹ Renal anomalies may also occur in 38% of women with rudimentary horns.⁷

Haematometra with dysmenorrhoea is a common symptom in women with a rudimentary horn although 40% of cases are asymptomatic.^{1 6} In asymptomatic women, the presence of a rudimentary horn is most frequently diagnosed during the evaluation of fertility.⁶ Due to the thin and poorly developed myometrium, the most significant threat is rupture of a pregnancy in a rudimentary horn; 80–90% of the horns rupture between 10 and 20 weeks of amenorrhoea.^{3 4 5} Less than 10% of the pregnancies in a rudimentary horn reach term, with a fetal survival rate of between 0% and 13%.^{3 5} Despite this threat, recent estimates of maternal mortality are low at <0.5%.^{1 5}

Consequently, even if the rudimentary horn is detected during pregnancy, intervention with excision of the rudimentary

horn and the usually highly desired pregnancy, is mandatory in order to prevent maternal morbidity. The ipsilateral fallopian tube has to be removed as well to prevent a future tubal ectopic pregnancy.³ Although laparoscopic procedures are presented in the literature,^{1 3 4} we decided for laparotomy in order to prevent persistent trophoblastic disease as a possible complication of intra-abdominal morcellation. In addition to immediate surgical excision, successful treatment with methotrexate is also described,^{2 8} although the rudimentary horn needs to be excised in a later stage to prevent recurrence.

Although there is no discussion about the necessity of intervention and this rare type of ectopic pregnancy is well known in the literature with approximately 150 cases described, the diagnosis of a rudimentary horn pregnancy is usually very challenging. Ultrasonic symptoms are a thin decidualised endometrium around the yolk sac and, in particular, a thin myometrium. Moreover, a big distance between the horn with the yolk sac and the rest of the uterine cavity (with dense desidualised endometrium) may be the most prominent finding. Finally, the absence of a cervical canal connecting the horn with the yolk sac to the other uterine horn can be observed. This symptom may be missed if not explicitly checked for.⁵ Pelvic MRI can be of additional value in evaluating the connection to the cervix. Since this connecting cervical canal is key in differentiating between a rudimentary horn and a bicornuate uterus, laparoscopy alone can never be conclusive. In case of persistent doubt, hysteroscopy can easily determinate the absence of a cervical channel to the uterine horn and consequently discriminate between a rudimentary horn and a bicornuate uterus.

CONCLUSION

Pregnancy in a rudimentary uterine horn is a rare condition which needs early diagnosis and adequate treatment in order to prevent rupture of the horn and consequent maternal morbidity. Excision of the rudimentary horn, preferably before but, if necessary during, pregnancy is mandatory. In the latter situation, extensive diagnostic techniques, including MRI and hysteroscopy, may be needed to differentiate accurately between a pregnancy in a bicornuate uterus, which is viable, and a rudimentary horn pregnancy which needs immediate treatment.

Learning points

- ▶ A pregnancy in a rudimentary horn is a rare type of ectopic pregnancy which is difficult to diagnose, even with laparoscopy.
- ▶ Hysteroscopy is of value when diagnosing this rare type of ectopic pregnancy.
- ▶ Pregnancies in a communicating horn of a bicornuate uterus are viable.
- ▶ Excision of the non-communicating rudimentary horn is mandatory to prevent uterine rupture.

Acknowledgements Professor Dr F W Jansen, gynaecologist, was involved in the diagnosis and treatment of this rare condition. He was of crucial importance in the diagnostic and therapeutic decisions taken in this case. We also thank WH den Hartog, resident in pathology, who kindly supplied the pictures displayed in figures 3 and 4.

Competing interests None.

Patient consent Obtained.

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Please cite this article as follows (you will need to access the article online to obtain the date of publication).

van Esch EMG, Lashley ELO, Berning B, de Kroon CD. The value of hysteroscopy in the diagnostic approach to a rudimentary horn pregnancy. *BMJ Case Reports* 2010;**10**:1136/bcr.08.2010.3229, date of publication

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