CASE REPORT

Case report and review of the literature: primary twin ovarian pregnancy


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A case report of primary twin ovarian pregnancy is presented. A 36 year old woman, gravida 4 para 3, was admitted to the hospital for suspected ectopic pregnancy, with vaginal bleeding at 11 weeks after her last menstrual period, associated with pelvic pain. An endovaginal ultrasonography led to the diagnosis of twin ovarian right ectopic pregnancy with two dead fetuses associated with a compartmentalized haemoperitoneum. Unilateral oophorectomy was carried out by laparotomy. Histological studies confirmed an uni-ovular di-amniotic ovarian pregnancy. Seven cases of ovarian twin pregnancies are reviewed in the literature. This case is the first one where diagnosis has been made by endovaginal sonography.

Key words: ovarian pregnancy/twin pregnancy

Introduction

Primary ovarian pregnancy is an uncommon form of ectopic pregnancy constituting <3% of all cases (Marcus and Brinsden, 1993; Riethmüller et al., 1996). The incidence varies between 1:6000 and 1:40 000 pregnancies (Marcus and Brinsden, 1993; Tuncer et al., 1994). There appears recently to have been an increase in ovarian pregnancy because of the improvement in diagnosis ability. Sonography and β-human chorionic gonadotrophin (β-HCG) have made it easier for the early preoperative diagnostic of ectopic pregnancy. Ovarian twin pregnancy is very rare and only seven cases have been reported in the literature. Another case is presented here, with the hope of increasing the establishment of early preoperative diagnosis by endovaginal ultrasonography. These eight cases are reviewed.

Case report

A 36 year old woman, gravida 4 para 3, was referred to our hospital for suspected ectopic pregnancy, with vaginal bleeding, at 11 weeks after her last menstrual period, associated with pelvic pain. She denied any past history of pelvic inflammatory disease, ectopic pregnancy or intrauterine device use. She had just felt, 3 weeks before, a drastic pelvic pain with feeling of faintness which had disappeared in a few hours. Her pelvic examination noted a bulging painful cul-de-sac and uterine spotting. There was no acute distress and no abdominal pain. β-HCG was measured at 10 500 mIU/ml and transabdominal and endovaginal ultrasonography were performed. The ultrasound revealed a thickened endometrial cavity with an empty uterus, a cystic haemoperitoneum in the pouch of Douglas cul-de-sac, and a right twin ectopic pregnancy. Vaginal ultrasonography confirmed a uni-chorionic, di-amniotic twin pregnancy which had ceased devlopment at 8 weeks of pregnancy, located in the right ovarian region. Ectopic pregnancy was suspected to be ovary because the right ovary was not seen and because the peripheral region of the mass seemed to have an ovarian cortex including follicles (Figure 1). The appearance of a corpus luteum cyst was noticed on the left ovary.

Because of the enormity of the ovarian mass, with a significant haemoperitoneum, which contraindicated laparoscopy in our university hospital, a laparotomy was performed, and revealed a ruptured right ovarian pregnancy with a cystic haemoperitoneum. The other pelvic structures, especially the right tube, were entirely normal, the left ovary presented a corpus luteum. There was no evidence of endometriosis or chronic inflammation in the pelvis. A unilateral oophorectomy was carried out and the mass sent for histological examination.

The patient was judged to have fulfilled all of Spiegelberg’s criteria. No corpus luteum was seen on the right ovary.

The patient had an uneventful postoperative course and was discharged on the seventh day. β-HCG control was normal (<10 mU/ml) 3 weeks later.

Discussion

Spiegelberg described, in 1878, four criteria for the diagnosis of ovarian pregnancy: the tube has to be entirely normal, the gestational sac has to be anatomically sited in the ovary, the ovary and the gestational sac have to be connected to the uterus by the utero-ovarian ligament, and placental tissue has to be mixed with ovarian cortex. Our case fulfilled those criteria. Macrosopically, ovarian pregnancy can take the appearance of an ovarian haematoma, clear ovum, embryonized ovum <3 months and placenta with fetus aged >3 months. Histology alone can confirm the diagnosis and distinguish the four forms: intrafollicular, juxtafollicular, juxtacortical and interstitial.
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Figure 1. Ultrasonographic appearance: uni-chorionic, di-amniotic twin pregnancy in the right ovary.

Figure 2. Right oophorectomy opened for histological examination: showing two thick cysts which each contained a small embryo measuring 15 mm in crown–rump length. Ovarian stroma, blood clots and chorionic villi were seen in continuity; the peripheral region of the ovary consisted of ovarian cortex.

pregnancy. The present report concerns an interstitial embryonized ovarian pregnancy. Ovarian pregnancy with a contralateral corpus luteum is a very rare form. Check and Chase (1986) have previously described such a case.

The ratio of ovarian pregnancies to all ectopic gestations is 1–6% (Marcus and Brinsden, 1993; Riethmüller et al., 1996). The estimated incidence of ovarian pregnancy ranges from approximately 1 in 6000 to 1 in 40 000 pregnancies (Marcus and Brinsden, 1993; Tuncer et al., 1994).

Ovarian pregnancy is in itself an uncommon type of ectopic pregnancy and primary ovarian twin pregnancy is an extremely rare condition, of which only seven cases have been reported in the English literature. These are summarized in Table I, which also includes our case.

Environmental conditions favouring tubal ectopic gestation such as pelvic inflammatory disease, previous surgery, and history of infertility are very rare in ovarian pregnancies. Recurrence is also exceptional and as the fertility of these women is conserved, the next pregnancy is usually intrauterine. However, a few risk factors seem to be present for ovarian pregnancies: endometriosis and intrauterine device usage are reported to contribute in the majority of cases. Three of the reported patients with twin ovarian gestations had endometriosis and one had intrauterine device usage, which were considered as risk factors. Transtubal reverse flow of menstruation may explain the flow back of the ovum (Riethmüller et al., 1996). Ovarian implantation may occur secondarily in the corpus luteum or extrafollicularly. This theory has been demonstrated by in-vitro fertilization. A few cases of ovarian pregnancies have been previously described after embryo transfer (Philippe et al., 1987; Marcus and Brinsden, 1993). However, ovarian pregnancy may occur without these factors since Caesarean section was the only factor found in our patient and in two of those reported in the literature. We have no explanation for this association. It fact, all the primary twin ovarian pregnancies were observed in primiparas or multiparas except one which occurred in a nulliparous woman; most occurred in women ~30 years old except for one in a very young woman aged 20 years.

Preoperative diagnosis is still difficult for ectopic pregnancy and especially ovarian pregnancy. In recent years, β-HCG and transabdominal ultrasound scanning have been well established
for diagnosis of ectopic pregnancies. The distinguishing characteristic of this case is that the diagnosis was made by transvaginal ultrasound scanning. This form of scanning leads to an early diagnosis when there are only suspicious symptoms like antenatal bleeding, and pelvic pain present. Ruptured ectopic pregnancy with circulatory collapse (Panda, 1990) or wrong diagnosis of malignant ovarian tumours producing HCG (Ohba et al., 1992) may also decrease the accuracy of diagnosis. An emergency situation with haemoperitoneum can result in an emergency laparotomy and sometimes blood transfusions. We hope early preoperative endovaginal ultrasonography will reduce the frequency of such emergency situations. Early preoperative diagnosis based on vaginal ultrasonographic findings has resulted in laparoscopic surgery and conservative treatment in singleton ovarian cases (Russel and Cuttler, 1989). However, twin ovarian pregnancies have been treated by laparotomy with at least oophorectomy. This was due to the gestational size, and haemoperitoneum, and because four out of the eight cases were treated in the 1960s and 1970s, when pelviscopy was not yet established. In singleton primary ovarian pregnancy, Shamma and Schwartz (1992) proposed a medical treatment such as methotrextate. Such conservative treatment, as well as laparoscopy, might be proposed in the early diagnosis of ovarian twin pregnancies in order to avoid a laparotomy.

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References


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