Unruptured retroperitoneal pregnancy implanted in the left broad ligament: A case report

Nerupturirana retroperitonealna trudnoća u levom širokom ligamentu

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Abstract

Introduction. Retroperitoneal ectopic pregnancy is extremely rare, but potentially fatal condition due to possible massive hemorrhage, representing a great challenge to clinicians. Case report. We presented early retroperitoneal pregnancy in a patient with previous caesarean section, diagnosed at the sixth gestational week, located in the left broad ligament, primary treated by laparoscopy, which had to be converted to laparotomy due to massive intraoperative bleeding from the implantation site. Conclusion. High index of suspicion, combined with carefully interpreted clinical and ultrasound findings are crucial for the timely diagnosis of retroperitoneal pregnancy, before the occurrence of severe bleeding. The rising, even plateau of serum β-human chorionic gonadotropin (β-HCG) levels without identification of uterine or ectopic (tubal) pregnancy should cause suspicion on ectopic pregnancy in unusual location.

Key words: pregnancy, ectopic; retroperitoneal space; laparoscopy; intraoperative complications; gynecologic surgical procedures.

Introduction

Retroperitoneal pregnancy is a very rare form of ectopic pregnancy. It could be the result of primary retroperitoneal implantation with enigmatic pathogenesis or secondary following tubal rupture in the broad ligament. In modern literature there are less than 25 well-documented cases of primary retroperitoneal pregnancy.1,2

We presented a primary retroperitoneal pregnancy implanted in the left broad ligament.

Case report

A 21-year-old gravida 1, para 1, was admitted into our clinic due to a 6-week history of amenorrhoea, lower abdominal pain and vaginal bleeding. Ectopic gravidity was suspected. Before 18 months the patient had term caesarean delivery performed due to breech presentation, oligoamnion and dystocia followed by an uneventful postoperative course. The women was otherwise healthy, had no history of pelvic inflammatory disease and use of intrauterine devices. Her menarche occurred at 12 and menstrual cycles were regular.

On admission the patient was hemodynamically stable. Examination of her cardiac and respiratory systems was unremarkable. Her abdomen was soft, but with mild inguinal tenderness on the left side. A speculum examination indicated the presence of a single cervix with scarce bleeding from external os and no other pathological findings. Bimanual pelvic examination revealed the slightly enlarged soft uterus...
and a tender palpable mass about 4 cm in diameter, on the left adnexal region. Transvaginal ultrasound examination (Toshiba Nemio XG, 6 MHz) showed the empty uterus with 5 mm endometrial strip (Figure 1). A round cystic mass, 4 × 3 × 2 cm in diameter, filled with heterogeneous content and hypoechogenic structure inside, like gestational sac without fetal pole, 1 cm in diameter, was seen just behind the uterine corpus, on the left side (Figure 2). Color Doppler examination revealed reach vascularisation, with the typical “ring of fire”, low resistance blood flow around the described mass (Figure 3). Both ovaries appeared sonographically normal with corpus luteum on the left ovary. There was no intraperitoneal fluid in the pouch of Douglas. Her laboratory results were as follows: white blood cells (WBC) 12.8 ×10^9/L, red blood cells (RBC) 4.48 ×10^{12}/L, Hb 123 g/L, hematocrit (Ht) 37.5, platelets (PLT) 368 ×10^9/L. Serum electrolytes, coagulation profile and liver function tests were all within physiological limits. At the day of admission her serum β-human chorionic gonadotropin (β-HCG) level (Abbott test; Architect-Total-β-HCG) was 28.643 mU/mL and the next day quantitative β-HCG level decreased to 27,000 mU/mL. Ectopic gravidity was suspected and diagnostic laparoscopy was performed after the written informed consent was obtained.

Surgery was conducted under general anesthesia, induced by means of propofol as induction agent, fentanyl as an analgesic and rocuronium as a muscle relaxant. Anesthesia was maintained with 1–1.5% end-tidal sevoflurane in 50%:50% O_2/N_2O mixture at 6 L/min flow. The lungs were ventilated to maintain end-tidal carbon dioxide concentration 30–35 mmHg. Laparoscopy revealed that the omentum was in slight adhesions with anterior abdominal wall (adhesiolysis was immediately undertaken), the uterus was slightly enlarged and the round retroperitoneal mass, 3 cm in diameter, located in the left broad ligament, behind the left round ligament and the left Fallopian tube was superior (Figure 4). The overlying peritoneum was intact. The pouch of Douglas was empty. Both ovaries and Fallopian tubes macroscopically appeared normal in size and shape and without any pathological fin-

Fig. 1 – Ultrasound image of early retroperitoneal pregnancy in the left broad ligament, just behind the uterine corpus. The uterine cavity is empty.

Fig. 2 – Ultrasound image of early retroperitoneal pregnancy in the left broad ligament: a round cystic mass, filled with a heterogeneous content and the hypoechogenic structure inside, like a gestational sac without fetal pole.

Fig. 3 – Retroperitoneal pregnancy in the left broad ligament: color Doppler examination revealed reach vascularisation with the typical “ring of fire”, low resistance blood flow around the described mass.

Fig. 4 – Laparoscopic finding of retroperitoneal pregnancy in the left broad ligament at the 6th gestational week: a) round retroperitoneal mass with the intact overlying peritoneum; b) the left Fallopian tube is superiorly, macroscopically normal in size and shape; c) the uterus is slightly enlarged; d) the part of the left ovary, without any pathological findings; e) the pouch of Douglas is empty.
The first report of retroperitoneal pregnancy was the ca-
dominal pregnancies with trophoblast invasion as retroperito-
pregnancy is unknown mainly due to false recognizing of ab-
exceptionally rare. The true incidence of retroperitoneal
ct pregnancy, as a subcategory of abdominal pregnancy, is
histopathology report confirmed ectopic gravidity (Figure 5).

There were no macroscopic signs of communication or fistu-
lar between the described mass and the uterine cavity or the
left Fallopian tube. Hemostasis was completed with hemosta-
tic sutures and the abdomen was closed with drainage placed
in the pouch of Douglas. Postoperative course was unevent-
ful. Serum β-HCG levels decreased to 750 mIU/mL two days
after the surgery and became negative after seven days. The
histopathology report confirmed ectopic gravidity (Figure 5).

**Fig. 5 – Histopathology report confirmed ectopic
gravidity: chorionic villi (haematoxylin-eosin, ×100).**

**Discussion**

The incidence of ectopic pregnancy is 0.25–1% of all
pregnancies. More than 95% of all ectopic pregnancies are tu-
abal pregnancies. The incidences of extratubal ectopic pregnan-
cies are as follows: abdominal in 1.3%, ovarian and cervical in
less than 1% of all ectopic pregnancies 1. Retroperitoneal ecto-
pic pregnancy, as a subcategory of abdominal pregnancy, is
exceptionally rare. The true incidence of retroperitoneal
pregnancy is unknown mainly due to false recognizing of ab-
dominal pregnancies with trophoblast invasion as retroperi-
toneal 1. The first report of retroperitoneal pregnancy was the
case of the broad ligament ectopic pregnancy described almost
two hundred years ago by Loschge 8. Our own review of the
literature (Medline data base, through electronic searches
without language restriction) showed a total of 65 reported
cases of retroperitoneal pregnancies during the last 57 years,
with 26 well documented cases during the last 15 years (Ta-
ble 1). The largest series of the broad ligament retroperitoneal
pregnancies (62 cases) was reported by Champion and Tesis-
tore 8 with the incidence of one in 183,900 pregnancies.

The sites of retroperitoneal ectopic implantation include
the broad ligament, obturator fossa, areas around large retrope-
ritoanal blood vessels, even the upper retroperitoneal space –
attached to the head of pancreas and major blood vessels 7–10.
There have been also reports on broad ligament twin preg-
nancies 11,12 and heterotopic pregnancies involving the broad
ligament and the uterus 13 or broad ligament and interstitial
pregnancy 14. Occurrence of partial hydatiform molla was also
reported in intraligamentous pregnancy 15.

The most often among retroperitoneal pregnancies is
the one located in the broad ligament or intraligamentous
pregnancy. The original anatomical relationships for diagno-
sing broad ligament ectopic pregnancy are: location of the
uterus medially, the pelvic side walls laterally, the pelvic
floor inferiorly and the Fallopian tube superiorly 6. Recently,
original criterions are fulfilled with the statement that
overlying peritoneum should be intact in order to confirm the
diagnosis of true retroperitoneal implantation 1. Our reported
case fulfills all the mentioned criteria.

Retroperitoneal ectopic implantation could appear after
spontaneous conception 1,4,8,11,16–26, intrauterine inseminati-
on 27 or after in vitro fertilization/pre-embryo transfer
(IVF/ET) 2,7,28 (Table 1). Intrauterine device (IUD) in situ
was found in 8% of abdominal pregnancies, so it is specula-
ted that IUD could be a factor contributing to the develop-
ment of abdominal pregnancy 29.

Retroperitoneal pregnancy could be the result of
primary retroperitoneal implantation or secondary following
tubal rupture or trophoblast invasion in the broad ligament.
There is also the possibility of primary interstitial and
secondary retroperitoneal pregnancy 20.

The pathogenesis of primary retroperitoneal pregnancy
is quite obscure. It seems that in the majority of cases,
primary retroperitoneal implantation could appear after ute-
rine or tubal surgery that could develop a communication in
the retroperitoneal space resulting with the passage of fertili-
zation ovum after spontaneous conception or after IVF/ET (Ta-
ble 1). The fistulous tract could be developed after termal
injury during laparoscopic salpingectomy 1, after classical
salpingectomy or salpingooophorectomy or due to inapprop-
riate heating of the uterine wall after cesarean section 16,30.
Spontaneous migration of the embryo from the uterus to the
retroperitoneal space through these communications could
result in retroperitoneal pregnancy.

There is also the possibility of false passage during em-
bryo transfer and placement of embryos into the retroperi-
toneal space in cases of retroperitoneal pregnancies after
IVF/ET 7,28.

The theory of spontaneous migration of the embryo
from the uterus to the retroperitoneal space along lymphatic
channels was based on findings of trophoblast surrounded by
lymphatic tissue 10,31. The contrast-enhanced computed
tomography was used to demonstrate the route of embryo
migration in retroperitoneal ectopic pregnancy providing fur-
ther evidence in support of the proposed embryo migration
mechanism via lymphatic vessels 22.
### Table 1

**Summarized cases of retroperitoneal pregnancies reported in the last 15 years**

| Authors | History (previous surgery) | Duration of amenorrhoea / conception | Site of implantation | Treatment |
|---------|-----------------------------|--------------------------------------|----------------------|-----------|
| Rams et al. (2015) | caesarean section before 18 months | 12 weeks / spontaneous | right broad ligament | laparotomy |
| Pratopapas et al. (2014) | laparoscopic right salpingectomy for EP | 6 weeks / spontaneous | right broad ligament | laparoscopy |
| Salomon et al. (2013) | laparoscopy | 15 weeks live / spont. | right broad ligament | laparoscopy |
| Saghi et al. (2013) | curettage | 20 weeks / spontaneous | broad ligament | MTX / laparotomy |
| Roda et al. (2013) | caesarean section before 4 years | 39 weeks non viable fetus | right broad ligament | laparotomy |
| Singh et al. (2012) | bilateral tubal ligation | 36 weeks live birth | right broad ligament | planned cs |
| Yoon et al. (2012) | no surgery | 5 weeks 2 days / spont. | between the IVC and ureter. | laparotomy |
| Martinez-Vara et al. (2012) | no surgery | 6 weeks / after IUI | next to left uterine lig. | laparoscopy |
| Sackin et al. (2011) | unknown | 20 weeks live birth / spont. | broad ligament | planned cs |
| Persson et al. (2010) | no surgery | * / spontaneous | ** | laparotomy |
| Milicic et al. (2009) | no surgery | 6 weeks 5 days / spont. | close to the blood vessels | laparotomy |
| Okovic et al. (2008) | right salpinge-oophorectomy | 22 weeks / spontaneous | left iliac fossa | laparotomy |
| Abdul et al. (2008) | no surgery | 6 months / spontaneous | right iliac fossa | laparotomy |
| Abdul et al. (2008) | right salpinge-right colostomy | 7 weeks 5 days | anterior aspect of IVC | laparoscopy |
| Lee et al. (2009) | appendectomy | 7 weeks / spontaneous | right obturator foramen area | laparotomy |
| Bae et al. (2009) | no surgery | spontaneous | left paracolic sulcus | laparoscopy |
| Chang et al. (2008) | left salpinge-popote | 18 weeks / spontaneous | right intra-ovarian | laparotomy |
| Holzhauser et al. (2008) | no surgery | following IVF | broad ligament | laparotomy |
| Puppon et al. (2006) | bilateral salpingectomy | spontaneous | left broad ligament | laparotomy |
| Aparaku et al. (2006) | left salpinge-ophorectomy | spontaneous | left pericolic region | laparotomy |
| Corno et al. (2006) | no surgery | 6 weeks / spontaneous | right broad ligament | laparoscopy |
| Lee et al. (2005) | unknown | 10 weeks / spontaneous | right broad ligament | laparoscopy |
| Slow et al. (2004) | bilateral salpingectomy for EP | 53 days / after IVF | bilateral of the common IA | laparotomy |
| Reid and Steel (2003) | two caesarean section | 11 weeks | right broad ligament | laparotomy |
| Pruppon et al. (2003) | bilateral salpingectomy | 11 days after IVF/ET | the head of pancreas | laparotomy |
| Pruppon et al. (2001) | bilateral salpingectomy | 11 weeks / spontaneous | left broad ligament, twin | laparotomy |

**Abbreviations:** * discovered in the 19th week, laparotomy in the 39th week, viable fetus; ** primary interstitial and secondary intraligamentous; EP - ectopic pregnancy; MTX - methotrexate; IA - iliac artery; IVC - inferior vena cava; IVF - in vitro fertilization; ET - embryo transfer; IUI - intruterine insemination; spont. - spontaneous; planned cs - planned caesarean section.
In the case we reported here, lymphatic tissue was not found around the trophoblast, even after careful histopathological examination, so we speculate that the development of the described left broad intraligamentous pregnancy could be explained by spontaneous migration of the embryo from the uterus to the retroperitoneal space through the microscopic fistulous tract caused by inappropriate healing of the uterine wall after the previous caesarean section. Still, we could not exclude with the certainty the possibility of embryo migration via lymphatic vessels, taking into account the localization of described retroperitoneal pregnancy.

The preoperative diagnosis of retroperitoneal pregnancy represents the challenge for clinicians. In fact, in the most cases, the diagnosis is made during surgery.

Maternal morbidity and mortality associated with abdominal, especially retroperitoneal, pregnancies could be reduced by early diagnosis. Transvaginal ultrasound examination is the main tool in the diagnostic of an early abdominal (and retroperitoneal) pregnancy. The proposed criteria are: the absence of an intruterine gestational sac; the absence of tubal dilatation or complex adnexal mass; a gestational sac surrounded by loops of the bowel and separated from the uterus; and a wide mobility of the gestational sac 33, 34. In fact, sonographic appearance of an early retroperitoneal pregnancy depends on its location. Usually it is fixed deep within the pelvis and not mobile as pregnancy in the non-communicating horn of the unicornuate uterus (cornual pregnancy) 35. The absence of communication between gestational sac and endometrial cavity differentiates the retroperitoneal broad ligament pregnancy from the pregnancy in non-communicating horn of the unicornuate uterus (cornual pregnancy) 35. The absence of communication between gestational sac and endometrial cavity differentiates the retroperitoneal broad ligament pregnancy from the pregnancy in non-communicating horn of the unicornuate uterus (cornual pregnancy) and interstitial ectopic pregnancy 35, which was also the truth in the case we reported here. The absence of myometrial layer around this retroperitoneal broad ligament pregnancy differentiates it from interstitial pregnancy. If early retroperitoneal pregnancy is located outside the pelvis, transvaginal ultrasound examination is helpless, and other diagnostic tools, as magnetic resonance imaging (MRI) and other imaging techniques, must be applied.

The suspicion is crucial for the timely diagnosis of retroperitoneal ectopic pregnancy. Rising β-HCG levels, or plateau, without identification of uterine or ectopic (tubal) pregnancy should cause suspicion on ectopic pregnancy in unusual location. In case we reported here, the diagnosis of ectopic pregnancy was made when the patient was still hemodynamically stable, so we opted for laparoscopic treatment during which the definitive diagnosis of left broad ligament pregnancy was confirmed.

The treatment of retroperitoneal pregnancy also represents a great challenge for clinicians. The most of retroperitoneal pregnancies are diagnosed and removed during the early stages of gravidity, but there are reports on broad ligament pregnancies with viable term fetuses even post term 42. The great majority of such cases are discovered on surgery for caesarean section.

In spite of many reports on abdominal pregnancies with viable fetuses advanced to term, the risk for the mother is still very high, especially in cases of retroperitoneal pregnancies with the close proximity to large vessels. Immediate surgery is indicated for abdominal pregnancies prior to 23 to 24 weeks because of the high incidence of maternal morbidity and a poor prognosis for the fetus 43.

Fetal anomalies or deformities (facial and cranial asymmetry, joint deformities, CNS anomalies) are associated problem in such pregnancies 41. However, there is a reported case of successful secondary retroperitoneal pregnancy in which the diagnosis had been suspected during the 18th week, discarded due to lack of symptoms and advanced to term with normal course 20.

The main concern with retroperitoneal pregnancy is associated with possible fatal bleeding due to the proximity of large blood vessels. This is also the possibility during the surgery after the attempt to remove the ectopic pregnancy. In the most of the reported cases laparotomy was the treatment. Nowadays, it seems that most unruptured early nontubal pregnancies could be managed laparoscopically. Laparoscopic treatment of ectopic pregnancy is minimally invasive procedure associated with lower cost, shorter hospital stay and faster recovery. However, the minority of reported cases of retroperitoneal pregnancies are treated laparoscopically. Laparoscopy is suitable for hemodynamically stable patients. Laparoscopic surgery has limitations as unnatural hand-eye coordination and impossibility to palpate the organs, especially retroperitoneal, but with improved skills that should not be a problem.

Hemorrhage during surgery is the most serious complication. Laparoscopically, it could be controlled by instillation of vasopressin 1 or with bipolar electrodes, monopolar scissors and laparoscopic bowel grasper applied across the corneal edge of the uterus 5, or with temporary occlusion of the right hypogastric artery by removable vessel clips to diminish the risk of bleeding complications 39. There is also the possibility to apply stitches and close the implantation site inside the broad ligament to achieve hemostasis 44. In spite of that, there is still the risk of massive intraoperative hemorrhage, which was also happened in the case of our patient, so laparoscopy had to be converted to laparotomy. It seems logical that the extent of intraoperative bleeding depends on the viability and vascularisation of retroperitoneal pregnancy. In the case of our patient, color Doppler ultrasound examination revealed reach vascularisation and the level of serum β-HCG before the surgery was high for ectopic pregnancy at 6th gestational week (over 20 000 mU/mL), both suggesting the vitality of pregnancy which could explain massive intraoperative hemorrhage. Histopathological examination excluded gestational trophoblast disease in the reported case, already extremely rare in ectopic pregnancy 43, with the prevalence of 0.16 : 1000 deliveries 46.

Preoperative use of methotrexate in cases of nonruptured retroperitoneal ectopic pregnancies could diminish intraoperative blood loss 47. Medical treatment of retroperitoneal pregnancy with methotrexate was reported with variable success 17, 21. In cases of ectopic tissue adherent around the major vessels, not removed in toto, methotrexate treatment is possible 2.
Conclusion

Retroperitoneal ectopic pregnancy is rare, but potentially fatal condition due to possible massive hemorrhage, representing a great challenge to clinicians. The early diagnosis and appropriate surgery are conditio sine qua non for successful treatment.

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