Heterotopic twin gestation – abdominal and intrauterine pregnancy

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Abstract

A very rare case of spontaneous heterotopic intrauterine and abdominal twin pregnancy has been presented. The first fetus was born by caesarean section. After 3 weeks, because of the symptoms of appendicitis the abdomen was re-opened. During re-operation appendicitis was found out, and the presence of terminated abdominal pregnancy at 18th week of its duration. Further course of puerperium was uncomplicated.

Key words: heterotopic twin pregnancy, intrauterine/abdominal pregnancy

Abdominal pregnancy is a type of ectopic pregnancy in which the trophoblast of fetus growing outside the uterine cavity has no contact with the oviduct, ovary and broad ligament of the uterus. It is assumed that in the U.S. 1.09:10 000 deliveries or 9.2:1000 pregnancies occur to be ectopic [1]. Even rarer phenomenon is the coexistence of intrauterine and ectopic pregnancy. In spontaneous pregnancies, such coexistence does not occur more often than 1:30 000 births [2]. However, a heterotopic twin pregnancy in which one fetus develops intrauterine and another in the abdominal cavity occurs very rarely. According to most authors, such coexistence occurs with an incidence of less than 1:100 000 births [3]. The first such case was probably described in 1708, when during the autopsy performed on a woman deceased during childbirth, the presence of a second fetus in abdominal cavity was detected.

We describe a case of a heterotopic twin pregnancy in which, after cesarean delivery of a fetus developing inside the uterus, the presence of another fetus was detected in abdominal cavity during the re-operation.

Case report

A 39 year old Gravida III Para III was admitted into ZOZ Hospital in Kutno in 36th week of gestation because of increasing blood pressure and reduced intrauterine fetus growth. Her first pregnancy ended in childbirth through natural passages in 40th week of pregnancy, whereas the other one – cesarian section in 29th week because of premature rupture of fetal membranes and breech lie.

At the moment of admission the following was noted: BP 180/110 mm Hg, pulse 72/min. Obstetric examination: lack of contraction activity, normal myometrial tone with its fundus reaching 3 finger-breadths above the naval. Fetal heartbeat ± 136/min. Internal examination: vaginal part formed, cervical canal closed, presenting part of fetal head pressed to plane of inlet, amniotic fluid remained, lack of uterine bleeding. CTG – reactive tracing.

In additional examinations the following was found out: blood group O Rh positive, peripheral blood morphology: Ht – 37.7%, Hb – 12.3 g%, E – 4.18 × 10⁵/µl, L – 8.25 × 10⁵/µl, platelets 146 ×10³/ul. Uric acid – 6.4 mg/dl. Other exam results within normal limits.

Ultrasonography: single fetus in cephalic longitudinal lie, presumable body mass 2000 g, placenta in fundus and posterior wall of uterus, III grade of maturity according to Granuum, deepest amniotic fluid pocket 10 mm. S/D 3.0 RI 0.6, FHR 132/min.

In spite of the treatment, the next day blood pressure rose to 200/120 and headaches appeared. In CTG trace – temporary bradycardia in the fetus. Due to the danger of eclampsia and exponents of intrauterine hypoxia, the gravida was qualified to cesarean section.

Male fetus was born alive with weight 2280 g/47 cm, estimated as having a score of 9 on the Apgar scale. The patient’s condition after surgery – good.

Patient three weeks after childbirth again came to the hospital because of temperature increased to 39.5°C. Abdomen flatulent. Pain on pressure and spontaneous in the right iliac fossa. Signs of peritoneal irritation. Ultrasound: in the underbelly a solid formation with blurry borders visible, measuring approximately 10 × 7 cm, surrounded by fluid. The presence of free fluid between intestinal loops and Douglas’ cavity.

Abdominal computed tomography showed „around the lower abdomen and pelvic minor pathological solid-
liquid mass present, with natural gas inclusions, size 10 × 6 cm. Around the change, in the immediate vicinity, small bowel loops visible. Also in the area, and between bowel loops, lower abdomen and pelvic minor free fluid visible. The image of the change non-characteristic – inflammatory infiltration? The presence of foreign body cannot be excluded” (Fig. 1). The patient was qualified to re-operation.

Ultrasound taken 4 months after the operation did not show any placental tissue that had been remained in abdominal cavity. The β-HCG concentration fell to indeterminable values.

Histopathological examination result „Purulent appendicitis. Fragments of necrotic and inflamed placental tissue, membranes and umbilical cord”.

Abdominal pregnancy is a grave threat to the life of both mother and fetus. According to world literature, maternal mortality ranges between 0 to 30% [4]. Risk of death of a pregnant because of complications associated with the development of abdominal pregnancy is 7.7 times higher than the fallopian tubes pregnancy and 90 times higher than in the case of intrauterine pregnancy [5]. The most common cause of death is massive hemorrhage caused by partial or total placental separation, which can occur anytime during pregnancy. Potential sites of trophoblast implantation are: omentum, pelvic wall, posterior wall of the bladder, abdominal organs such as spleen cavity, bowels, liver, large pelvic vessels, diaphragm and uterine peritoneum. The fetal outcomes seem to be worse than the mother’s. The perinatal mortality proportion ranges from 40% to 95% [2]. Moreover, there is a very high percentage of malformations. Fetal outcome tends to be better if there is normal volume of amniotic fluid as well as higher gestational age at the moment of diagnosis [5].

The diagnosis of abdominal pregnancy is relatively difficult. Although Allibone et al. [4] presented basic guidelines for the ultrasonography diagnostics, percentage of diagnostic failures, described in literature, ranges from 50% to 90% [5, 6]. Even in cases in which clinical examination was coupled with ultrasound evaluation, the percentage of diagnostic failures exceeded 50% [1]. The main reason is attributed to the fact that the person performing ultrasonography does not take into account the possibility of abdominal pregnancy. Performance of MRI may be useful.

The described case is particularly unique, because it presents unusual obstetrical complication, initially wrongly diagnosed. On the basis of clinical examination, ultrasound and computed tomography it was found out that the cause of bowel obstruction and peritonitis was a surgical towel which had been left during the cesarean section performed three weeks before and possibly acute appendicitis. The presence of abdominal pregnancy was not taken into consideration at all.

Placental management in abdominal pregnancy is still controversial. If possible, it is recommended to remove the entire placenta [1]. Placenta probably regres-
sessed as soon as fetal-placental circulation is interrupted. This allows to avoid an uncontrolled hemorrhage in future. However, if this proves difficult and combined with the risk of excessive blood loss, the prevailing opinion is to leave it in whole or in part in the abdominal cavity. It is connected with the risk of secondary hemorrhage, infection, bowel obstruction, preeclampsia/eclampsia, bowel fistulas and sepsis caused by infection of degenerated and susceptible to disintegration tissues. The risk of re-intervention, often in a critically ill patient, should be taken into consideration [5].

Placental involution can be supervised by serial ultrasound examinations and evaluation of β-HCG concentration in a woman’s serum. Some authors recommend the use of methotrexate, but with varying degrees of success.

In the described case, the placenta was removed only in part. Lack of abundant bleeding during the attempt of its separation can be explained by the presence of regressive changes in the afterbirth which appeared in the labour terminated a few weeks before. After the surgical operation the patient felt well. Also, one year later no disturbing symptoms were observed.

Since in our case the abdominal pregnancy was developing till about 18th week, it is extremely difficult to answer the question whether the abdominal pregnancy was caused by primary or secondary implantation. It seems to have been a primary abdominal pregnancy. This results from the fact that during the operation no signs of early or inveterate damage to the fallopian tubes and ovaries were found, and the entire placental insertion was located within the small bowel and its mesentery. In addition, in the early weeks of pregnancy the patient had no signs suggesting the presence of tubal pregnancy.

In conclusion, although abdominal pregnancy is a very rare obstetrical complication, it should always be taken into account in diagnostically difficult cases. The earliest diagnosis may in fact contribute to reducing perinatal morbidity of fetuses’ mothers and morbidity of neonates associated with pregnancy development.

References

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