Spontaneous rupture of malignant ovarian cyst in 8-gestation-week pregnancy – a case report and literature review

Ljiljana Mladenović Segedi[1], Aljoša Mandić[2], Dimitrije Segedi[1], Tatjana Kozarčić[1]

Arch Oncol 2011;19(1-2):39-41.
UDC: 618.11-006:618.2:616.071:615-085
DOI: 10.2298/AOO1102039M

[1]Clinic of Gynaecology and Obstetrics, Novi Sad, Serbia, [2]Oncology Institute of Vojvodina, Sremska Kamenica, Serbia

Correspondence to:
Ljiljana Mladenović Segedi, MD., PhD.,
Clinic of Gynaecology and Obstetrics,
Branimira Ćosića 37,
21000 Novi Sad, Serbia
dimseg@open.telekom.rs

Received: 26.09.2010
Provisionally accepted: 13.10.2010
Accepted: 09.11.2010

SUMMARY

Ovarian cancer takes the second place in the incidence of all genital carcinomas occurring in pregnancy. In spite of a low overall incidence of ovarian cancer in pregnancy and, in most cases, asymptomatic clinical picture, the routine use of ultrasonographic examination in early pregnancy has led to a more frequent detection of adnexal masses in pregnant women. The authors presented a case of a 35-years-old patient with the diagnosis of unilocular cyst in the left ovary detected by ultrasonography at gestational week 8, with the subsequent sinistral adnexectomy administered for the rupture of the cystic tumor. Based on the histopathological examination we established the diagnosis of a serous cystadenocarcinoma stage I. The patient refused any proposed modality of oncologic diagnostics and therapy, and delivered a healthy female newborn of 3630g/49 cm, 10 days after the probable term of delivery. The control MRI, 8 months post partum did not confirm the presence of any pathological changes of either genital organs or any other organ of the small pelvis. The patient is in good general condition, under continual medical observation.

Key worsd: *Ovarian Neoplasms; Rupture, Spontaneous; Cystadenocarcinoma, Serous; Pregnancy*

INTRODUCTION

Cancer is the second most common cause of death in the women during their reproductive years, complicating between 0.02% and 0.1% of all pregnancies (1). Ovarian tumors are evidenced in about 1/1000 pregnancies out of which 3% to 6% malignant ones. Ovarian malignancies are found to be the second most common gynecological cancer diagnosed during pregnancy except for cervical carcinoma (2). The overall incidence of ovarian cancer is still low, 1/12 500–25 000 pregnancies (3,4). Adnexal masses, which are detected during physical examination, are usually asymptomatic in significant number of pregnant women. However, they cause certain complications such as ovarian torsion and rupture of cysts. The routine use of ultrasound in pregnancy has led to more frequent detection of adnexal masses. Most of the common tumor markers for ovarian neoplasms, such as CA125, α-fetoprotein, human chorion gonadotropin, lactate dehydrogenase and inhibin, are elevated and fluctuate with gestational age but have limited diagnostic value during pregnancy (4,5). Magnetic resonance imaging (MRI) gives significant information and helps to reduce the need of surgical intervention. As an imaging modality without ionizing radiation exposure, it provides the additional information on a pelvic mass detected during ultrasonography (6). The treatment modalities in this group of patients have never been investigated in any randomized studies because of ethical reasons. Management of ovarian cancer in pregnancy is dependent on gestational age at diagnosis, stage of disease, future childbearing desires, and the mother's wishing to keep the pregnancy despite the malignant disease.
All diagnostic and therapeutic modalities have to be overviewed in decision to choose the best option for pregnant patient considering the effects of these modalities on the disease, mother, and fetus.

A CASE REPORT

A patient was 35 years old, pregnant for the first time, last period was on April 20, 2008. Unilocular ovarian cyst of 11.5 cm was diagnosed on a routine ultrasound examination at gestational week 8 (GW). A day after the diagnosis she had an operation for the ovarian cyst rupture. Left oophorosalpingectomy was performed. There was no macroscopic suspicion for neoplasm and a frozen section was not obtained. The histopathological examination confirmed serous cystadenocarcinoma stage I without infiltration of the spontaneously ruptured capsule. The material was sent for control analysis to another hospital and the diagnosis was confirmed except for histological stage described as stage II. The patient was referred to our Institute with her desire to continue the pregnancy not wanting another operation despite a suggestion of her surgeon for radical operation and termination of pregnancy. The patient was aware that she could be a biological mother probably only this time and she was not willing to lose the pregnancy. The patient was offered few modalities as she did not agree to undergo surgical staging: to keep the pregnancy with the following regular fetal and oncological monitoring and MRI examination at GW 18-20 and in case the disease was not detected, to wait for the delivery, and six weeks after normal delivery to undergo a complete operation with surgical staging. If MRI showed any changes in the ovary, the option was a cesarean section and complete operation. The patient was also informed about increasing possibilities for the recurrent disease. A neoadjuvant chemotherapy administered in the second trimester was offered as another modality because the rupture of malignant cyst was a bad prognostic factor. The patient refused chemotherapy as an option and continued pregnancy without any adjuvant treatment.
After 41 weeks and 3 days of pregnancy (according to the time of her last menorrhea) the patient was admitted to the Department of obstetrics and gynecology with irregular contractions and spontaneous rupture of membranes. Because of weak contractions occurring every l5 minutes, the patient was stimulated by oxytocin infusion and at the cervix dilatation of 4 cm she was administered epidural anesthesia. After 6 hours of regular, coordinated, and effective uterine contractions the patient delivered a healthy female newborn, 49 cm of length and 3630 gr of

weight, and Apgar score 8/10 (vaginal delivery associated with episiotomy). The patent was suggested to undergo a laparoscopic operation with complete staging six weeks following the delivery, but she did not agree with another surgery. She was discharged five days post delivery together with her baby.

The control MRI examination of the small pelvis performed 8 months following the delivery did not detect any pathological changes of the uterus, right ovary or any other organ of the small pelvis. The patient was in good general condition, subjectively with no physical disorders.

DISCUSSION

The presence of ovarian cancer in the women who want to preserve pregnancy despite of the diagnosis opens a new challenge for patients and doctors regarding the aspects of ethics, psychology, clinical approaches in these "special cases", and medical-legal issues.

Effects of the disease on pregnancy and fetus such as influence of pregnancy on the disease itself and effects of therapy in these cases leave clinical practice with too many opened questions.

Ovarian cancer is recorded in 3% to 6% of all ovarian tumors and has still been very rare according to Sayedur's study, (incidence 0.08/1000 deliveries) (7). Similar incidence was found in Zhao's study (8). The routine use of ultrasound in prenatal care improves diagnostics of adnexal masses and becomes the ideal method of detection and surveillance of adnexal masses. The use of ultrasound can also contribute to diagnostics of their structural characteristics (9).

Ultrasonographycally detected adnexal masses greater than 5 cm, bilateral masses, those growing or persisting to the second trimester, and presenting with papillary projections, solid compartments, being multicystic or septate, need further investigation. Despite a good diagnostic equipment and sensitivity of ultrasound in detection of adnexal masses, the diagnostic incidence of detection of malignant ovarian cysts is still relatively not very high (10). The MRI could be helpful in increasing the accuracy in diagnostics of malignant adnexal masses (6).

Histopathological findings of ovarian cancer during pregnancy are similar to these of nonpregnant women in the corresponding reproductive-age group.

The germ cell tumors comprise 45% of ovarian malignancies diagnosed during pregnancy; epithelial tumors are found in 37.5%; sex cord–stromal tumors in 10%; and miscellaneous pathologies in 7.5% of all cases (11-13).

The invasive serous tumors are the most common invasive epithelial ovarian neoplasms that appear as cystic structures (10).

Surgical intervention is often necessary either for the symptoms or for the suspect malignancy. Ideally, this is done after 14 to 16 weeks' gestation when the placenta has taken over hormonal support of the pregnancy from the corpus luteum (9).

Surgical exploration using a midline incision is recommended to limit uterine manipulation and to enable adequate staging and debulking.

In nonpregnant patients, the standard treatment for invasive epithelial ovarian cancer is initial debulking surgery followed by adjuvant chemotherapy. Surgery typically consists of laparotomy with total abdominal hysterectomy, bilateral salpingo-oophorectomy and meticulous staging omentectomy and lymphadenectomy (14). This type of surgical approach during pregnancy is not possible unless the patient decides to terminate the pregnancy.

Administration of adjuvant chemotherapy after surgery has more and more been accepted as the treatment procedure in the patients with malignant disease in pregnancy. In invasive epithelial cancer of FIGO stages greater than IA and IB, the poorly differentiated or the non-staged ones, the adjuvant chemotherapy could be taken into account equally in pregnant and nonpregnant patients.

The risk of teratogenesis from chemotherapy is high in the first trimester, at about 10% (15). After organogenesis is completed at the start of the second trimester, the risk of fetal abnormality is very low (16, 17). There is, however, a risk of intrauterine growth retardation and premature labor. Long-term outcomes are still unknown, with little published data (15).

The data about performed chemotherapy in epithelial ovarian cancer during pregnancy are still very rare and usually are presented as case reports.

Sayar et al. presented nine reports on chemotherapy applied for invasive epithelial ovarian tumors during pregnancy. In seven patients, unilateral salpingo-oophorectomy was performed as an initial surgery during pregnancy, and total abdominal hysterectomy with contralateral salpingo-oophorectomy was postponed until after delivery. All authors used platinum-based chemotherapy during pregnancy after the initial surgical intervention. In most cases, the delivery was completed by cesarean section. All infants were born with adequate maturity, in good condition, and without gross congenital anomalies (10, 15, 19).

In the late second or third trimester and advanced disease in the patient who still wants to preserve pregnancy, neoadjuvant chemotherapy could be considered (18).

There is no indication that pregnancy decreases overall survival rate in the patients with epithelial ovarian cancer (10).

This limited number of case reports on epithelial ovarian cancer in pregnancy points to partial surgery with platinum-based chemotherapy in the second trimester as an option for the patients who want to preserve pregnancy. After the delivery, surgical intervention and application of several cycles of chemotherapy are necessary to complete the therapy procedure. In advanced stages of the disease during the early period of the first trimester, an initial complete staging and debulking surgery without preservation of pregnancy followed by conventional chemotherapy is still the most reasonable approach (20).

Conflict of interest

We declare no conflicts of interest.

REFERENCE

1 Lishner M. Cancer in pregnancy. *Ann Oncol*. 2003:14 Suppl 3:iii31–iii36.

2 Zanotti KS, Belinson JL, Kennedy AW. Treatment of gynecologic cancers in pregnancy. *Semin Oncol*. 2000;27:686–98.

3 Whitecar MP, Turner S, Higby MK. Adnexal masses in pregnancy: a review of 130 cases undergoing surgical management. *Am J Obstet Gynecol*. 1999;181:19–24.

4 Oehler MK, Wain GV, Brand A. Gynaecological malignancies in pregnancy: a review. *Aust N Z J Obstet Gynaecol*. 2003;43:414–20.

5 Spitzer M, Kaushal N, Benjamin F. Maternal CA-125 levels in pregnancy and the puerperium. *J Reprod Med*. 1998;43:387–92.

6 Curtis M, Hopkins MP, Zarlingo T, Martino C, Graciansky-Lengyl M, Jenison EL. Magnetic resonance imaging to avoid laparotomy in pregnancy. *Obstet Gynecol*. 1993;82:833–6.

7 Sayedur Rahman M, Al-Sibai MH, Rahman J, Al-Suleiman SA, El-Yahia AR, Al-Mulhim AA, et al. Ovarian carcinoma associated with pregnancy. A review of 9 cases. *Acta Obstet Gynecol Scand*. 2002;81:260–4.

8 Zhao XY, Huang HF, Lian LJ, Lang JH. Ovarian cancer in pregnancy: a clinicopathologic analysis of 22 cases and review of the literature. *Int J Gynecol Cancer*. 2006;16:8–15.

9 Boulay R, Podczaski E. Ovarian cancer complicating pregnancy. *Obstet Gynecol Clin N Am*. 1998;25:385–99.

10 Sayar H, Lhomme C, Verschraegen CF. Malignant Adnexal Masses in Pregnancy. *Obstet Gynecol Clin N Am*. 2005.32;(4):569-93.

11 Partridge EE, Phillips JL, Menck HR. The National Cancer Data Base report on ovarian cancer treatment in United States hospitals. *Cancer*. 1996;78:2236–46.

12 Dgani R, Shoham Z, Atar E, Zosmer A, Lancet M. Ovarian carcinoma during pregnancy: a study of 23 cases in Israel between the years 1960 and 1984. *Gynecol Oncol*. 1989;33:326–31.

13 Copeland LJ, Landon MB. Malignant disease in pregnancy. In: Gabbe SG, Niebyl JR, Simpson JL, editors. Obstetrics, normal and problem pregnancies, 3rd ed. New York, NY: Churchill Livingstone; 1996. p. 1155–81.

14 Cannistra SA. Cancer of the Ovary. *N Engl J Med*. 2004;351:2519-29.

15 Cardonick E, Iacobucci A. Use of chemotherapy during human pregnancy. *Lancet Oncol*. 2004;5:283–91.

16 Peres RM, Sanseverino MT, Guimarães JL, Coser V, Giuliani L, Moreira RK, Ornsten T, et al. Assessment of fetal risk associated with exposure to cancer chemotherapy during pregnancy: a multicenter study. *Braz J Med Biol Res*. 2001;34:1551–9.

17 Reidenbach F. Chemotherapy safe in pregnancy. *Lancet Oncol*. 2002;3:128.

18 Picone O, Lhommé C, Tournaire M, Pautier P, Camatte S, Vacher-Lavenue MC, et al. Preservation of pregnancy in a patient with a stage IIIB ovarian epithelial carcinoma diagnosed at 22 weeks of gestation and treated with initial chemotherapy: case report and literature review. *Gynecol Oncol*. 2004;94:600– 4.

19 Tabata T, Nishiura K, Tanida K, Kondo E, Okugawa T, Sagawa N. Carboplatin chemotherapy in a pregnant patient with undifferentiated ovarian carcinoma: case report and review of the literature. *Int J Gynecol Cancer*. 2008;18:181–4.

20 Amant F, Van Calsteren K, Halaska MJ, Jos Beijnen T, Lagae L, Hanssens M. Guidelines of an International Consensus Meeting. *Int J Gynecol Cancer*. 2009;19:S1-S12.