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CASE REPORT

DYSGERMINOMA IN PREGNANCY

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Summary

Introduction: Malignant germ cell tumors (MGCTs), as a subtype of rare non-epithelial ovarian cancers (NOEC), are most commonly found in pregnancy. Of all MGCTs, 38% are dysgerminoma. Considering the rarity of these entities, the aim of this paper is to show a rare case of ovarian dysgerminoma presented in pregnancy and its influence on course and outcome of the pregnancy.

Patient Review: Patient aged 26, gravida 2, para 1, with one vaginal delivery five years before, was admitted to the Clinic for Gynecology and obstetrics in term pregnancy because of uterine contractions accompanied by left thigh pain and tingling sensation in the left leg. Solid hypoechogenic mass with regular borders, 125x90 mm in diameter adjacent to the left side of the uterus was seen by ultrasound, without free fluid in pelvic cavity. Since regular uterine contractions started, the decision was made to terminate pregnancy by Caesarean section (CS) because of tumor previa. Histopathological examination confirmed ovarian dysgerminoma, but after staging operation which was performed two months after CS, following imaging diagnostics, ovarian dysgerminoma was confirmed with FIGO stage IA, meaning that patient's specific oncological treatment was finished.

Conclusion: Diagnosis of ovarian dysgerminoma is in general challenging since up to 50% are asymptomatic or symptoms are non-specific. The management of ovarian cancer in pregnancy should be multidisciplinary and individualized in the best interest of the mother and the fetus. The overall five-year survival rate for ovarian dysgerminoma is favorable in more than 90% of cases. Women diagnosed with dysgerminoma in pregnancy are young and in general have good feto-maternal outcome.

Key words: Ovarian dysgerminoma, feto-maternal outcome, dysgerminoma in pregnancy, ovarian cancer, cancer in pregnancy

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INTRODUCTION

The incidence of cancer in pregnancy is approximated to be 1 in 1000 deliveries [1]. Gynecological cancers are among the most frequently diagnosed cancers during pregnancy with ovarian malignancies complicating 1% to 6% of pregnancies [1,2]. Malignant germ cell tumors (MGCTs), as a subtype of rare non-epithelial ovarian cancers (NOEC), are most commonly found in pregnancy. Of all MGCTs, 38% are dysgerminoma [3].

In general, MGCTs present as unilateral rapidly growing tumors in adolescence and early adulthood [4,5]. Most of them are diagnosed at an early stage and have very high survival rate. Considering young age of patients, fertility sparing surgery is treatment of choice [6].

Adnexal mass in pregnancy is not only a challenge for diagnosis and treatment, but can also lead to feto-maternal complications [7]. Hence, each patient demands individualized approach in order to achieve an adequate and timely diagnosis and provide appropriate treatment both for the mother and the fetus.

Since this group of ovarian cancers are rare entities, especially in pregnancy, the aim of this paper is to show a rare case of ovarian dysgerminoma presented in pregnancy and its influence on the course and outcome of the pregnancy.

CASE REPORT

Patient aged 26, gravida 2, para 1, with one vaginal delivery five years before, was admitted to the Clinic for Gynecology and obstetrics in term pregnancy because of uterine contractions accompanied by left thigh pain and tingling sensation in the left leg. At presentation, abdominal and ultrasound examination were performed, as well as basic laboratory tests. The uterine fundus height was 33 cm and abdominal circumference was 95 cm. A firm painless mass along the left isthmic side of the uterus with reduced mobility was palpated. Ultrasound examination revealed a vital term fetus with cephalic presentation, adequate amount of amniotic fluid for gestational age and normal placental insertion. Solid hypoechogenic mass with regular borders, 125x90 mm in diameter adjacent to the left side of the uterus was seen by ultrasound, without free fluid in the pelvic cavity. Basic laboratory test results were within normal range for term pregnancy.

Upon admission, the patient also reported that on initial prenatal ultrasound at 13 weeks of gestation hypoechoic lesion measuring 5 cm on the left side was found and that the finding was highly suspected to be fibroid. She also reported that this finding persisted only with minor size enlargement throughout trimesters. Tumor markers (CA-125, HE4, CEA, CA 19-9) were evaluated during the first trimester of pregnancy and were within normal range. Amniocentesis proved normal fetal male karyotype. This procedure was performed in the second trimester because the mother has 47, XX karyotype with small supernumerary marker chromosomes (sSMC). Otherwise normal course of pregnancy was noted with adequate fetal development.

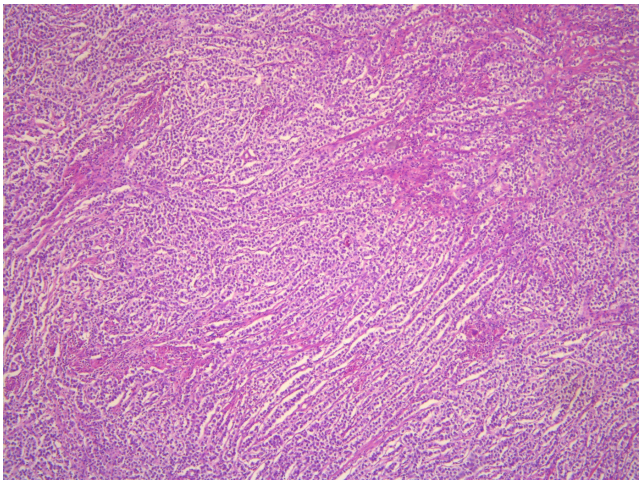

Figure 1. Background lymphocytes surrounding sheets of tumor cells.

Since regular uterine contractions started, the decision was made to terminate pregnancy by Caesarean section because of tumor previa. The patient delivered healthy baby with Apgar score 9 in the fifth minute. Intraoperatively, the left ovary was transformed into a large (140x95x80 mm) homogeneous tumor, with yellowish-white intact capsule and softer consistency. There were no signs of disease spread or free fluid in the abdominal and pelvic cavity. Microscopically cut section showed tumor composed of solid, confluent beaches and bands of large, polygonal, indistinct border cells with large nucleus and light pink, watery plasma **(Figure 1 and 2)**. A scant lymphoid infiltrate and sparse fibrous stroma were present. Final histopathological examination confirmed ovarian dysgerminoma, possibly FIGO IA stage. Imaging was done postoperatively. Neither abdominal and pelvic MRI nor chest X-ray showed signs of disease spread. All the analyzed tumor markers (CA-125, CA 19-9, CEA,

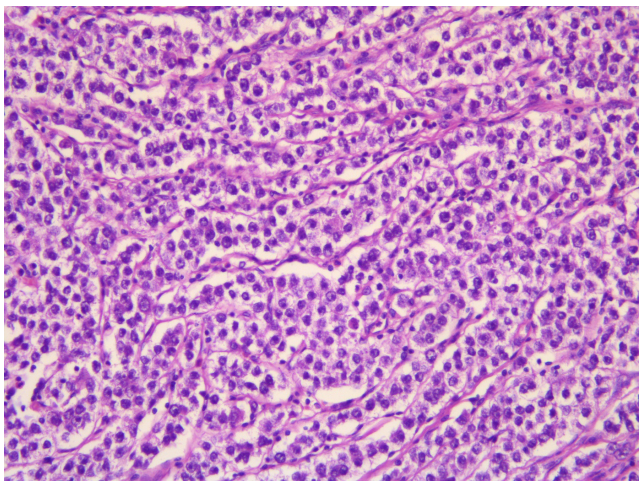

Figure 2. Tumor cells with large clear cytoplasm

LDH, beta hCG) were within normal range. Staging operation was performed two months after CS confirming ovarian dysgerminoma FIGO stage IA, meaning that patient's specific oncological treatment was finished. During the seven-year follow-up with regular pelvic ultrasound, MRI imaging and tumor markers no sign of recurrence has been noticed. The patient was discharged from hospital with a guideline for regular follow-ups. After eight-year follow-up, there has been no sign of disease relapse. Also, normal psychomotor and social development of the child has been reported.

DISCUSSION

This paper presents a case of pregnancy terminated by term CS and complicated by a large adnexal mass that was later proven to be ovarian dysgerminoma.

Although malignant germ cell tumors are rare, their peak incidence is in women of reproductive age [7]. Because of this, one fifth of all ovarian malignancies in pregnancy are MGCT and ovarian dysgerminoma prevail with almost 40% of cases [7]. Overall incidence of ovarian dysgerminoma is less than 1 per 100 000 pregnancies, so most of the literature data are based on case reports and small case series [8].

Generally, diagnosis of ovarian dysgerminoma is challenging as in up to 50% of cases it is asymptomatic or its symptoms are non-specific [9]. Diagnosing these malignancies in pregnancy is even harder because common symptoms of MGST such as abdominal pain and distension may occur in normal pregnancy leading to misdiagnosis. Also, as pregnancy progresses, the growing uterus with fetus interferes with adnexal or uterine masses [7]. This is the reason why ovarian dysgerminoma happens to be an incidental finding during CS as was our case [9].

Although diagnosis is usually made by ultrasound, data show that up to 40% of cases could be missed on ultrasound examination [7]. The numbers are probably higher in the second and the third trimester, meaning that the first trimester exam is important not only for fetal anatomy scan but also for the pelvic inspection. Second line examination is MRI of the pelvis which is safe throughout pregnancy and is more sophisticated in adnexal mass diagnosis and provides more information that ultrasound.

Although in this case adnexal mass was ultrasonographically seen in the first trimester, it was mistaken for fibroid. Dysgerminoma is most commonly misdiagnosed as uterine fibroid, especially pedunculated uterine fibroid with focal cystic degeneration [10]. Beside ultrasound and pelvic MRI, additional diagnostic tool that could be helpful in differentiation of adnexal mass are elevated tumor markers such as serum lactic dehydrogenase and AFP which could be elevated in up to 86% cases, as well as serum beta hCG [9], but these tumor markers are also elevated in pregnancy itself. Histopathology stays the gold standard of ovarian malignancy diagnosis.

The management of ovarian cancer in pregnancy should be multidisciplinary and individualized in the best interest of the mother and the fetus. When making a decision, patients age, gestational age, parity, stage of the tumor, desire for present pregnancy, and future fertility should be taken into consideration [9]. For most early-stage ovarian cancers, unilateral oophorectomy or adnexectomy with appropriate staging should be the surgery of choice and it is safest to perform in the second trimester [6]. Ovarian dysgerminoma is highly chemosensitive to platin-based chemotherapy and is reserved as adjuvant therapy for patients except stage IA [6,11].

The overall five-year survival rate for ovarian dysgerminoma is favorable in more than 90% of cases [10]. Recurrence rate for stage IA is approximately 20% [10].

The possible rate of complications of adnexal mass in pregnancy such as torsion, incarceration, hemorrhage and rupture has increased [9]. Intrauterine growth restriction (IUGR) is the most common (22,8%) complication in neonates [9].

According to a systematic review of literature by Kodama el al. which included 102 ovarian MGCT-complicated pregnancies, the majority of cases resulted in live birth (77.5%) at term (56.6%) via Cesarean section. IUGR was present in 22.8%. During the pregnancy course, obstructed labor, tumor rupture and torsion occurred in 2.9%, 8.8%, and 1.0%, [12].

In our case we had a normal pregnancy course without influence on the neonate and mechanical symptoms of tumor that manifested at the term of delivery. Histopathological analysis did not show necrosis or hemorrhage of the tumor. After eight-year follow-up, our oncological, perinatal and pediatric outcome is good.

CONCLUSION

Women diagnosed with dysgerminoma in pregnancy are young and in general have good feto-maternal outcome. Fertility sparing surgery can be offered in women desirous of pregnancy. The treatment strategy must be discussed and structured individually.

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DISGERMINOM U TRUDNOĆI: PRIKAZ SLUČAJA

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Sažetak

Uvod: Maligni tumori germinativnih ćelija (MGCT), kao podtip retkih neepitelijalnih karcinoma jajnika (NOEC), su najčešći maligni karcinomi jajnika koji se mogu javiti u trudnoći a od kojih se disgerminom javlja u svega 38%. Obzirom na retkost ovih entiteta, cilj ovog rada je da prikaže redak slučaj disgerminoma jajnika koji se javio tokom trudnoće, kao i njegov uticaj na tok i ishod trudnoće.

Prikaz slučaja: Pacijentkinja starosti 26 godina, drugorotka, primljena je na Kliniku za ginekologiju i akušerstvo u terminu trudnoće zbog bolova po tipu kontrakcija, koji su praćeni bolom u levoj butini sa širenjem ka levoj nozi. Ultrazvukom je uočena čvrsta hipoehogena masa pravilnh kontura, prečnika 125x90 mm uz levu stranu materice, bez slobodne tečnosti u maloj karlici. S obzirom na spontano započinjanje porođaja, doneta je odluka da se trudnoća završi carskim rezom zbog predstojećeg tumora. Histopatološkim pregledom potvrđen je disgerminom jajnika, ali nakon operacije preduzete da se utvrdi stadijum tumora koja je obavljena dva meseca nakon carskog reza, i prethodne imidžing dijagnostike, disgerminom jajnika je potvrđen kao FIGO stadijum IA, što znači da je specifično onkološko lečenje pacijentkinje završeno.

Zaključak: Disgerminom jajnika u trudnoći predstavlja veliki izazov u dijagnostici jer je u do 50% slučajeva asimptomatski ili su simptomi nespecifični. Lečenje karcinoma jajnika u trudnoći treba da bude multidisciplinarno i individualizovano, te u najboljem interesu majke i ploda. Ukupna petogodišnja stopa preživljavanja kod disgerminoma jajnika je povoljna u više od 90% slučajeva. Žene sa dijagnozom disgerminoma u trudnoći su mlade i generalno imaju dobar feto-maternalni ishod.

Ključne reči: Disgerminom jajnika, feto-maternalni ishod, disgerminom u trudnoći, karcinom jajnika, karcinom u trudnoći

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