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Case Report / Приказ случаја

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Dysgerminoma and pregnancy

Дисгермином и трудноћа

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SUMMARY

Introduction Dysgerminomas are germ cell ovarian tumors. They affect young females, prevalently during childhood. The problem arises when dysgerminoma is diagnosed in women of reproductive age who have never given birth and require a surgical procedure.

Case outline A 28-year-old patient was admitted to hospital in week 26 of her first pregnancy. The reason for patient hospitalisation was the growth of the istmic myoma diagnosed by her ObGyn in the primary care unit. By examining the medical hystory of the patient, the following was revealed: A year and a half before pregnancy, she was diagnosed with left ovary dysgerminoma. The patient's medical history led us to conclude that uterine myoma was a misdiagnosis and that the actual diagnosis was dysgerminoma of the right ovary. The surgery was performed after the fetal viability had been achieved.

Conclusion Malignant ovarian tumours may occur in young women during pregnancy and increase in size significantly in a short period of time, although their recurrence is not expected in such a short period of time after surgical treatment.

Keywords: dysgerminoma; malignant; ovrian germ cell tumor; pregnancy

САЖЕТАК

Увод Дисгерминоми су тумори герминативних ћелија јајника. Најчешће се јављају код млађих особа и то у дечјем добу. Проблем настаје када се постави дијагноза дисгерминома у репродуктивном периоду и када је потребно лечити га хируршки а пацијенткиња још увек није остварила своје потомство.

Приказ случаја Примигравида, 26 година стара, примљена је у клинику у 26-ој недељи гестације због сумње на експанзиван раст истмичног миома откривеног ултразвуком од гинеколога у примарној здравственој заштити. Код болеснице је годину и по дана раније урађена левострана аднексектомија због оваријалног дисгерминома. Дијагноза миома материце је била погрешна и радило о дисгерминому десног јајника. Хируршко лечење дисгерминома обављено је након постизања феталне одрживости.

Закључак Малигни тумори јајника могу се јавити код младих жена током трудноће убрзо после хируршког лечења и значајно увећати у кратком временском периоду.

Кључне речи: дисгермином; тумори герминативних ћелија оваријума; трудноћа

INTRODUCTION

Dysgerminomas are germ cell ovarian tumors. About 20% of all ovarian tumors originate from germ cells whereas only 3% of them are malignant. Dysgerminomas account for about 1% of all germ cell tumors but they are frequently malignant [1]. They affect young females, prevalently during childhood and the vast majority of them need and respond well only to chemotherapy. The problem arises when dysgerminoma is diagnosed in women of reproductive age who have never given birth and require a surgical procedure. A more serious situation occurs if dysgerminomas develop in young patients during pregnancy, which poses many medical and ethical dillemas. We present a case of a young patient with a previous history of a dysgerminoma which was managed by left adnexectomy. The patient conceived two years after the surgery. However, a dysgerminoma of the right ovary was diagnosed in the sixth month of pregnancy. The surgery was performed after the fetal viability had been achieved.

CASE REPORT

A 28-year old patient was admitted to hospital in week 26 of her first pregnancy. The reason for hospitalisation was the growth of the istmic myoma diagnosed by her obstetrics and gynecologist in the primary care unit. Ultrasonography revealed a viable pregnancy at 24 weeks gestational age and a

solid mass of uncertain origin, with the size of 100×100mm, detected on the right side of the uterus. An ultrasound examination performed four weeks earlier showed the mass of 60×60mm.

By examining the medical history of the patient, the following was revealed: A year and a half before pregnancy, the patient was diagnosed with a left ovary dysgerminoma. She had undergone left adnexectomy and the partial resection of the right ovary. Dysgerminoma with negative immunoprofile (alpha fetoprotein, inhibin and ema) and positive immunoprofile of the tumor (reticulin + and high level of Ki 67) was confirmed by histopathology. The pathology of a part of the right ovary showed only corpus luteum. The surgery was the only management option. The control ultrasound as well as tumor markers 6 month before pregnancy were normal. The patient's hystory led us to conclude that uterine myoma was a misdiagnosis and that the actual diagnosis was dysgerminoma of the right ovary. The laboratory findings were as follows: D-dimer 5022 ng/mL, LDH 12715 IU/L, AST 95 U/L, ALT 174 U/L. The magnetic resonance imaging (MRI) finding in week 27–28 of pregnancy showed a giant tumor in the pelvis, 200×200mm in size, ascites, lymphomegaly and bilateral hydroureteronephrosis (Figure 1). The pregnancy was terminated at week 31/32 of gestation by Caesarean section and a viable preterm female baby was born with birth weight of 1630 gr and apgar score 7/10, 8/10 at 1 and 5 minutes of life. Afterwards, the total abdominal hysterectomy with right adnexectomy, omentectomy, para-aortic, iliac and obturator lymphadenectomy was done. The distal part of the right ureter was also resected and the ureterocystostomy was performed. The patient subsequently underwent chemotherapy with bleomycin, etoposide and platinum (BEP × 4 cycles). She is currently free of disease at 2 years post-treatment with a healthy baby.

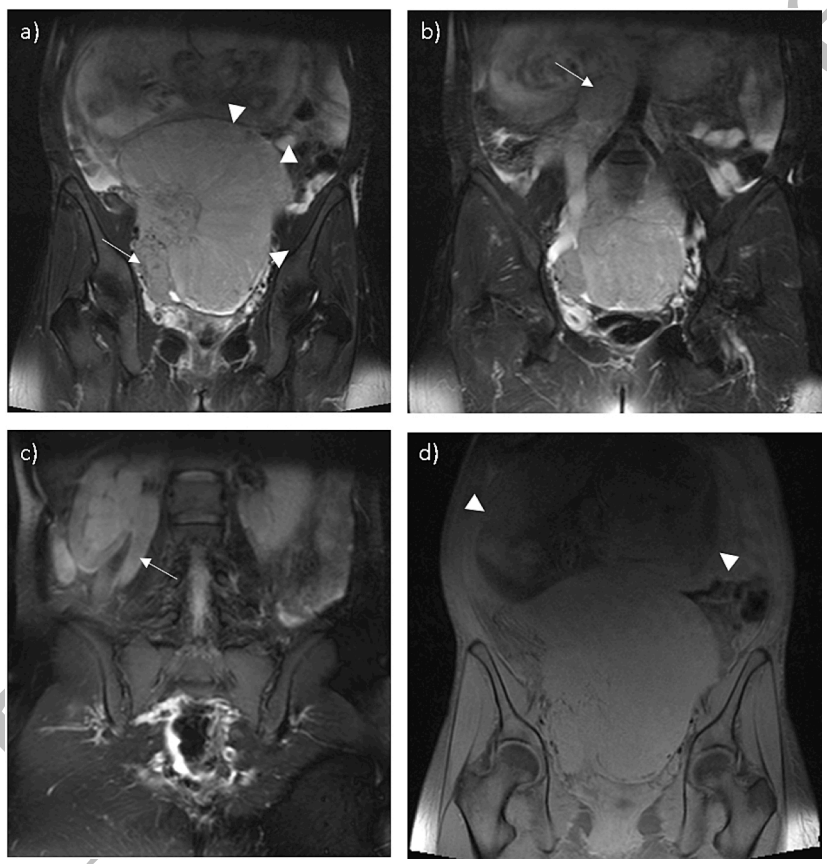

Figure 1. Pelvic MRI. Coronal reconstructed T2-weighted images show: a) a large intermediate signal intensity solid mass with prominent fibrovascular septa, occupied pelvic cavity with upward extension into abdominal cavity (arrowhead); in the caudal part of the mass another lobulating mass corresponding to conglomerate of lymph nodes is showen (arrow); b) para-aortic lymph node mass (arrow); c) mass effect on adjacent structures with consequent hydroureteronephrosis; the diameter of the ureter measured up to 13 mm (arrow); d) Coronal fat-saturated T1-weighted image shows displacing of fetus cranially (arrowhead).

DISCUSSION

As mentioned above, dysgerminomas are tumors originating within the primordial ovarian germ cells. Dysgerminoma has a classic correlation with seminoma of the testis, having an identical histological structure. Germ cell tumors account for about 70% of ovarian neoplasm cases during the first decades of life, and are rarely found after this period [2]. Approximately 80% of cases are reported in patients under 30 years of age (mean age: 21 years), which is a finding consistent with our case.

The incidence of adnexal masses associated with pregnancy varies from 1 in 80 to 1 in 8000 pregnancies, based on different studies. The frequency of ovarian tumors in such adnexal masses is between 1 in 80 and 1 in 2200 pregnancies [3]. The reported rate of malignant tumors in the total number of ovarian tumors associated with pregnancy was from 1.3% to 7.9%. In fact, in a study by Ueda M. at al. among 106 cases of ovarian tumors discovered during pregnancy only five (4,7%) were malignant [4]. The most common diagnosis was dermoid and dysgerminoma was noted in only one case.

Only two cases of dysgerminoma were diagnosed during pregnancy in our clinic, in a 10-year period, which represented less than 1%. This fact my lead to a conclusion that the rate of the malignant ovarian tumors associated with pregnancy is very low. This discrepancy of ovarian malignancy incidence between pregnant and non-pregnant women can be explained by the age difference among women [5]. Most patients with malignant ovarian tumor were over 40 years old and those patients were rarely pregnant. Thus, dysgerminoma may be considered the only malignant ovarian tumor to be kept in mind when detecting adnexal mass during pregnancy.

Taking into account the rarity of this tumor, a misdiagnosis during pregnancy is not uncommon, as it was the case here. A literature review reveals that it is not unusual to misdiagnose dysgerminoma by an ultrasound examination and diagnose uterine fibroids instead. Not only ultrasound but also MRI misdiagnosed the dysgerminoma as a fibroid uterus in our case. MRI has a sensitivity of about 98% for detecting the origin of an ovarian tumour. However, there have been reports of mistaking a malignant ovarian tumor for pedunculated uterine fibroid with areas of cystic degeneration, as in our case [6]. Ovarian tumors generally remain asymptomatic, until they are discovered due to their large size or related complications.

In the current case, dysgerminoma was diagnosed as a result of the enlargement of the pelvic mass which was thought to be a uterine fibroid. The 14-week obstetric ultrasound showed a corpus luteum cyst in the enlarged right ovary, but all diameters were within the normal range. The 20-week obstetric ultrasound showed a pelvic mass diagnosed as uterine fibroid with a diameter of 60 mm and, 4 weeks later, the mass was twice as large. The specialist literature indicates that certain neoplasms may undergo geometric growth of up to 20% of their original size in a very short period of time (1–2 months). The structure, consistency and contiguity with the uterus all pointed to uterine fibroid, as

was demonstrated by ultrasound examination. For this reason, the above finding was perceived as uterine myomatosis and if there is a suspicion of a uterine myom in pregnancy, the diagnostic procedure is not the same as in case of a malignant ovarian tumor, when testing for tumor markers, pelvic MRI and other diagnostic methods are performed. Although dysgerminoma is highly suspected when a patient has phenotypic signs of certain syndromes associated to the states like Cowden's syndrome, ataxia telangiectasia syndrome, Swyer syndrome (pure gonadal dysgenesis associated with the XY 46 karyotype) and Apert syndrome (an autosomal dominant disorder), in this particular case the suspicion should be based on the previous medical history of the patient [7, 8, 9, 10].

Our patient was diagnosed with the left ovary dysgerminoma 1.5 years before actual pregnancy and she underwent surgery. No precise recommendations for further outpatient follow up are known based on any randomized controlled trials. However, follow-up should maximize the ability to identify recurrences while minimizing risks. Follow-up care depends on the stage of a disease, which is typically predictive of recurrence risk. Ovarian dysgerminomas tend to recur most often in the first 2-3 years after treatment. Therefore, most authors suggest follow-up observation and a physical examination every 3–4 months for the first 3 years, every 6 months during the fourth and fifth year, and annual surveillance thereafter. Typically, the authors do not recommend any adjuvant chemotherapy for stage Ia dysgerminomas as was the case with the first surgery. Although 10–15% of stage Ia tumors may recur, essentially all of them are salvaged with chemotherapy [11]. This patient underwent all postoperative checkups. However, since she conceived afterwards the patient was probably not provided with an adequate follow up. It may also be hypothesized that pregnancy induces rapid growth of tumor, although further studies are needed to confirm the hypothesis.

By publicizing this case, we aim to raise awareness of malignant ovarian tumors possibly affecting young females in pregnancy, the volume of which may rapidly increase within a very short period of time although the recurrence of previous malignant disease is not expected in such a short period after surgery. This poses a great challenge for obstetricians.

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