

Mucinous Adenocarcinoma Transformation in Ovarian Mature Cystic Teratoma Complicating Pregnancy: A Case Report

Hajar Abbasi¹, Salomeh Mohammadi², Atefeh Moridi¹, Athena Behforouz¹

¹ Preventative Gynecology Research Center, Shahid Beheshti University of Medical Sciences, Tehran, Iran

² Department of Pathology, Mahdiah Hospital, Shahid Beheshti University of Medical Sciences, Tehran, Iran

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Abstract- Most adnexal masses found in pregnancy are simple benign cysts less than 5 cm in diameter. On the other hand, the majority of persistent adnexal masses 5 cm or greater in diameter are mature teratomas. Malignant transformation occurs in 0.2 to 2 percent of mature teratomas, and squamous cell carcinoma arising from ectoderm is the most common secondary neoplasm. The mucinous cystadenocarcinoma transformation is very rare. We presented a 38-year-old G2Ab1 woman at 32 weeks of gestation with a huge, rapid growth adnexal mass who came to our emergency department due to severe preeclampsia, stage 1 of IUGR, and borderline AFI. After 48 hours of expectant management, a cesarean section associated with unilateral salpingo-oophorectomy was done, and the histopathology of the cyst revealed mature teratoma that was focally involved with mucinous cystadenocarcinoma. The complete surgical staging was done, and due to the stage of disease (IC1), she received adjuvant chemotherapy, and she has remained disease-free in her last follow up after one year.

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Introduction

The incidence of adnexal masses complicating pregnancies varies from 0.05 to 2.4 percent, of which 1 to 6 percent of these are malignant (1-4). Many of these masses are asymptomatic and recognized in the first half of pregnancy incidentally (5). Most adnexal masses identified in pregnant women are simple benign cysts less than 5 cm in diameter, but the majority of persistent adnexal masses 5 cm or greater in diameter are mature teratomas (1). Malignant transformation occurs in 0.2 to 2 percent of mature teratomas, and squamous cell carcinoma arising from ectoderm is the most common secondary neoplasm (6-10). The mucinous adenocarcinoma transformation is very rare, and as far as we explored the sources, there was just one case of mucinous adenocarcinoma arising in mature teratoma in pregnancy (5).

We reported a third-trimester pregnant case of

mucinous adenocarcinoma arising in mature cystic teratoma.

Case Report

We presented a 38-year-old G2Ab1 woman who came to our emergency department with the complaint of headache, abdominal pain, and high blood pressure (170/90 mmHg) at 32 weeks of gestation.

We managed severe preeclampsia and controlled her blood pressure. In her recent documents, the fetus was stage 1 of IUGR (Intra Uterine Growth Retardation) with borderline amniotic fluid index. In addition, she had a left ovarian cystic lesion in favor of a dermoid cyst measured 25*53 mm at first-trimester sonography. For better evaluation, another sonography was done after stabilizing the patient's status. It showed a large cystic lesion measuring 151*140 mm containing 23*25 mm echogenic nodule in favor of dermoid cyst in the left ovary. Tumor

Corresponding Author: A. Behforouz

Preventative Gynecology Research Center, Shahid Beheshti University of Medical Sciences, Tehran, Iran
Tel: +98 2122259685, Fax: +98 215506626, E-mail address: behforouz.a@gmail.com

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markers were normal. Cesarean section was done with midline skin incision after 48 hours of severe preeclampsia management. In advance, intraperitoneal fluid was sent for cytology. An appropriate for gestational age, 1400 gr baby, was delivered. The uterus was repaired. The whole abdomen, pelvis, and omentum were explored systematically. A 150*150 mm cystic lesion with hair and fat component was in the left side of the pelvis, which ruptured during surgery. The contralateral ovary was macroscopically normal without any lesion. Neither pathological point was found in exploration. Left salpingo-oophorectomy was performed, and the specimen was sent for frozen section pathology. The result was mature teratoma that was focally involved with mucinous adenocarcinoma. Before surgery, we discussed with the patient about the possibility of hysterectomy and bilateral salpingo-oophorectomy according to frozen's result. The patient wanted definitive treatment and did not intend to become pregnant again. Thus appendectomy with a total abdominal hysterectomy and right salpingo-oophorectomy and staging was done. The histopathology report revealed other parts were free of tumor. She received six cycles of CARBOPLATIN-TAXOL following surgery and was disease free at her last follow-up after 1 year.



Figure 1. Cut section of ovarian mass showing cystic teratoma and adjacent multiloculated solid cystic mass

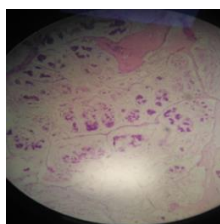


Figure 2. Mucinous carcinoma

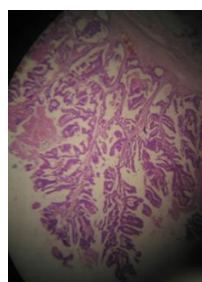


Figure 3. Mucinous carcinoma with papillary projections

Discussion

Currently, the incidence of adnexal masses complicating pregnancies varies from 0.05 to 2.4 percent, of which 1 to 6 percent of these are malignant (1-4). Many of these masses are asymptomatic, but nowadays, they are recognized in the first half of pregnancy incidentally due to routine antenatal ultra-sonographies, which are performed for obstetrical indications (1,5,11-13). Most adnexal masses identified in pregnant women are simple benign cysts less than 5 cm in diameter, but the majority of persistent adnexal masses 5cm or greater in diameter are mature teratomas (1). Malignant transformation occurs in 0.2 to 2 percent of mature teratomas, and squamous cell carcinoma arising from ectoderm is the most common secondary neoplasm (6-10). Other possible malignant neoplasms include basal cell carcinoma, melanoma, adenocarcinoma, sarcoma, and thyroid carcinoma. Management of these malignant transformations must be tailored to transformed histology (6,10).

Malignant transformations arising in mature cystic teratoma in pregnancy is extremely rare, and as far as we explored the sources, most of them were reported as squamous cell carcinoma except one case, which was reported as mucinous adenocarcinoma arising in mature teratoma in pregnancy (5).

Although teratomas are mostly seen in younger women, teratomas with malignant transformation usually occur in significantly older women (14). Due to the limited number of cases that were reported as a malignant transformation on mature cystic teratoma, the prognosis is undetermined. Risk factors of malignant transformation arising in mature cystic teratoma are age over 45 years, tumor diameter greater than 10 cm, rapid growth, and low resistance flow on Doppler sonography of cyst (6,10). If a pregnant patient has the suspicious findings of malignancy in ultra-sonography of her adnexal mass, the optimal time for semi elective surgery is after the first trimester, and the optimal route of surgery is laparotomy (15).

Our reported case had a huge cystic lesion with a diameter of more than 15 cm and rapid growth during pregnancy course from 5cm to 15 cm in approximately 25 weeks, which guided us to the probability of malignancy over mature teratoma.

In addition, due to the pregnancy status of our patient, the value of serum tumor markers decreased because the interpretation of several tumor markers varies with gestational age and comorbid conditions due to oncofetal antigens (16). But as we suspected for malignant

transformation, appropriate tumor markers were checked. Alpha-fetoprotein (AFP), human chorionic gonadotropin (hCG), carcinoembryonic antigen (CEA), and cancer antigen 125 (CA125) are tumor markers involved in biological functions associated with fetal development, differentiation, and maturation. These levels are normally elevated during pregnancy and fluctuate with the course of gestation and some placentation factors. CA125 may be useful as a tumor marker of epithelial ovarian cancer between 15 weeks of gestation and delivery. In addition, a CA125 in the range of 1000 to 10,000 is likely related to cancer (16). Also, maternal serum levels of AFP (MSAFP) normally rise during pregnancy but AFP levels of more than 1000 ng/ml represent ovarian germ cell tumors such as endodermal sinus (yolk sac) tumors which can be associated with AFP levels of more than 10,000 ng/ml. Some authors suggested a MSAFP levels above 9 mom represent probability of germ cell tumors of either gonadal or non-gonadal origin in the absence of fetal abdominal wall or neural tube defects (17).

Despite normal results of all evaluations, our patient's cyst frozen section showed malignant components on mature cystic teratoma and due to the intraoperative rupture of cyst, the patient's stage of cancer upgraded to 1C1. The impact of ovarian tumor capsule rupture on the prognosis is controversial (18,19). Preoperative rupture associates with a worse prognosis but data regarding intraoperative rupture are inconsistent (20). In the current staging system, surgical spill known as stage 1C1 while preoperative rupture known as stage 1C2. Patients with disease limited to one or both ovaries who have intraoperative cyst rupture are upgraded from 1A or 1B to 1C1 which is typically the threshold for chemotherapy treatments, as our patient (20).

So our patient received adjuvant chemotherapy and she has remained disease free for over 1 year.

In conclusion, most adnexal masses less than 5 cm in diameter are benign cysts in pregnancy but persistent masses over 5 cm in diameter are often teratomas. Malignant transformation can occur in mature cystic teratomas and the alarm signs and symptoms are age over 45-year-old, tumor diameter over 10 cm, rapid growth and low resistance flow on Doppler sonography. Some of serum tumor markers can be helpful for adnexal mass evaluation during pregnancy with higher ranges. Although the malignant transformation of mature cystic teratoma during pregnancy is rare, especial mucinous adenocarcinoma, but early detection and surgical management as soon as possible after first trimester can help to improve the prognosis.

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