Spontaneous Pregnancy and Live Birth after Fertility-Sparing Treatment for a Yolk Sac Tumor: A Case Report

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ABSTRACT

A 17-year-old female adolescent presented to the gynecology department and was diagnosed with a yolk sac tumor (YST). The patient received neoadjuvant chemotherapy followed by fertility-sparing surgery and then adjuvant chemotherapy. The patient, who was married 6 years after the diagnosis of YST (endodermal sinus tumor) and who wanted pregnancy became pregnant spontaneously in 2020 and had a healthy, term, vaginal delivery in 2021 (birth weight 3040 g, Apgar score of 8–9). There were no complications during her pregnancy. There are not enough studies about the management of such patients during pregnancy. The increase in this and similar case reports will shed light on the approach and management of these patients. It is possible to achieve spontaneous pregnancy and healthy vaginal delivery after fertility-sparing surgery for the YST.

Keywords: Endodermal sinus tumor, fertility preservation, pregnancy

INTRODUCTION

Malignant ovarian germ cell tumors (MOGCTs) are a rare histological subtype of ovarian cancers and constitute approximately 2–3% of all ovarian cancers. These tumors mainly occur in adolescents and young women of reproductive age and are seen in 70% of this age group. Survival rates have increased dramatically after the introduction of platinum-based chemotherapies. Nowadays, fertility-sparing surgery in MOGCT treatment includes the preservation of the uterus and contralateral ovary. Before the establishment of systemic chemotherapy, MOGCTs had a very poor prognosis. However, after the introduction of chemotherapy, the prognosis dramatically improved. Yolk sac tumors (YSTs) are highly malignant germ-cell tumors characterized by differentiation toward yolk sac structures, with a characteristic expression of alpha-fetoprotein (AFP). These tumors are highly aggressive malignancies and show early intra-abdominal dissemination and metastasis. After the MOGCT surgery, the fertility and pregnancy ratio has not been clarified yet.

CASE REPORT

A 17-year-old patient admitted to the hospital in October 2012 with sudden onset of abdominal swelling, and severe abdominal pain revealed a giant mass of solid and cystic necrotic areas of approximately 10x16x21 cm in the pelvic region in the computed tomography of the upper and lower abdomen. Axial and coronal sections view of the abdomen computed tomography taken before preoperative neoadjuvant chemotherapy are shown in Figures 1 and 2. The measured
AFP value in the blood of the patient was 72538 ng/mL (0–9 ng/mL), and the human chorionic gonadotropin (B-hCG) value was found to be normal. YST was considered because the patient’s blood AFP value was high, and the B-hCG value was 0.5 mIU/mL (0–5 mIU/mL). Because of the involvement of the lymph nodes of the patient, it was accepted as stage 3 according to the Children’s Oncology Group/Germ Cell Tumor staging. It was decided to take neoadjuvant chemotherapy and then be operated on. No appearance compatible with metastasis was observed in thorax and brain computed tomography. Other tumor markers were determined: carcinoembryonic antigen was 1.88 ng/mL (0–3 ng/mL), CA 125 was 42.56 U/mL (0–35 U/mL), CA-19-9 was 23.59 U/mL (0–33 U/mL), and CA15-3 was 25.53 U/mL (0–30 U/mL). The patient was given two cycles of neoadjuvant chemotherapy (Cisplatin, etoposide, ifosfamide). Computed tomography performed after two cycles of chemotherapy revealed 80% reduction in the tumoral mass. AFP value in blood after chemotherapy was 2727 ng/mL. Then, it was decided to operate the patient. The pathology report of the patient, who underwent laparotomic left oophorectomy in January 2013, was reported as a YST. The patient received four more cycles of adjuvant chemotherapy (etoposide, ifosfamide, cisplatin, last May 2013). The patient continued her routine follow-up after chemotherapy. The patient was evaluated with AFP level at every visit, and there was no finding compatible with recurrence in her imaging. The patient menstruated regularly. The anti-Mullerian hormone value of the patient, who got married in 2019 and wanted pregnancy, was found to be 0.76 ng/mL (1.2–4 ng/mL). The patient was referred to the infertility center and could not go because of the pandemic. During this period, the patient got pregnant spontaneously. A gestational follow-up of the patient, who was 25 years old when got pregnant, was done in our hospital. Routine blood and tumor markers requested were normal at the first visit. The first trimester combined screening test was interpreted as a low-risk group. No fetal structural anomaly was found in the 20-week scan. An oral glucose tolerance test performed with 75 g of glucose at week 26 resulted in the normal range. The patient was admitted to the delivery room when the delivery did not begin at the 41st week. As the Bishop score was not good, labor induction was performed with a dinoprostone insert. A live female baby weighing 3040 g was delivered with normal spontaneous vaginal delivery. The Apgar score was 8–9. The baby was given to the mother. No problem was encountered in the neonatal period.

**DISCUSSION**

YSTs at the ovary are relatively rare neoplasms, usually diagnosed in the early stage of life, that are characterized by a high potential for malignancy but also by high chemotherapy sensitivity. YSTs occur in young women, and it is now possible to achieve cure and fertility preservation. Therefore, reproductive outcomes are considered important. The median age in patients diagnosed with MOGCT...
was 19–22 years, and the patient was 17 years old at the
time of diagnosis. The most common symptom of MOGCT
is abdominal distention. Some patients have ascites and
peritonitis, and secondary to torsion, fever and infection
may be observed.[6] The duration of symptoms varies from
2 days to 6 months, but the mean duration of symptoms
is 4 weeks. The patient had mild abdominal swelling and
pain that started in the last 3 months. Increased AFP values
are valuable for diagnosis, and in the patient, the AFP value
was greater than the normal range.[7] As MOGCT occur in
young patients, the treatment strategy should ideally be
considered in a fertility-sparing and long-term survival ap-
proach. The treatment approach is the complete removal
of the tumor and the combination of adjuvant chemother-
opy and platinum-based chemotherapy. In the patient,
after neoadjuvant chemotherapy, fertility-sparing surgery
was applied and left oophorectomy was performed, and
then adjuvant chemotherapy treatment was applied. In
the study performed by Mitchell et al., regular menstrual
bleeding was observed in 24 patients after 26 MOGCT
treatments, and successful pregnancy was achieved in 11
patients.[8] In another study, Nishio et al. observed regular
menstrual bleeding in 28 patients after 30 MOGCT treat-
ments. Twelve patients had pregnancy requests, and 8 of
these patients ended with a successful pregnancy.[9] In the
study conducted by Low et al., 43 of 47 MOGCT patients
had regular menstrual bleeding, 20 of these patients de-
sired pregnancy, and 19 of them had a successful pregnan-
cy.[10] In the patient, there were regular menstrual periods
after MOGCT treatment, and a healthy and trouble-free
pregnancy was followed after. The patient was managed
successfully in giving birth to a healthy female baby.

CONCLUSION
Currently, there is no well-established guideline toward pa-
tients who have a history of overcoming MOGCT, yet plan-
ing a future pregnancy or follow-up treatments. This case
report has been a good example of addressing this issue
more closely. Well-organized randomized controlled trials
with larger patient groups will enlighten this issue.

Disclosures
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the patient for publication of this case report and any accompa-
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