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## 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



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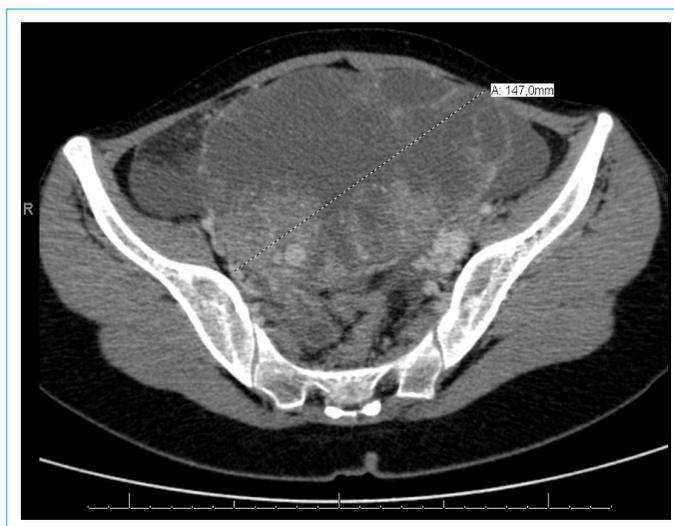
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### INTRODUCTION

Malignant ovarian germ cell tumors (MOGCTs) are a rare histological subtype of ovarian cancers and constitute approximately 2–3% of all ovarian cancers.<sup>[1]</sup> These tumors mainly occur in adolescents and young women of reproductive age and are seen in 70% of this age group.<sup>[2]</sup> Survival rates have increased dramatically after the introduction of platinum-based chemotherapies.<sup>[3]</sup> Nowadays, fertility-sparing surgery in MOGCT treatment includes the preservation of the uterus and contralateral ovary.<sup>[4]</sup> 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).<sup>[5]</sup> 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 10×16×21 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



**Figure 1.** Axial section view of the abdomen computed tomography taken before preoperative neoadjuvant chemotherapy.



**Figure 2.** Coronal section view of the abdomen computed tomography taken before preoperative neoadjuvant chemotherapy.

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.<sup>[5]</sup> 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.<sup>[6]</sup> 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.<sup>[7]</sup> As MOGCT occur in young patients, the treatment strategy should ideally be considered in a fertility-sparing and long-term survival approach. The treatment approach is the complete removal of the tumor and the combination of adjuvant chemotherapy 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.<sup>[8]</sup> In another study, Nishio et al. observed regular menstrual bleeding in 28 patients after 30 MOGCT treatments. Twelve patients had pregnancy requests, and 8 of these patients ended with a successful pregnancy.<sup>[9]</sup> In the study conducted by Low et al., 43 of 47 MOGCT patients had regular menstrual bleeding, 20 of these patients desired pregnancy, and 19 of them had a successful pregnancy.<sup>[10]</sup> 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 patients who have a history of overcoming MOGCT, yet planning 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

**Informed Consent:** Written informed consent was obtained from the patient for publication of this case report and any accompanying images. The study adhered to the tenets of the Declaration of Helsinki.

**Conflict of Interest:** The authors declare that they have no conflicts of interest.

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