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Juvenile Granulosa Cell Tumor: Successful Pregnancy After Recurrent Mass

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ABSTRACT

Juvenile Granulosa Cell Tumor (JGCT) is a rare ovarian tumor found in children and adolescents. Although the prognosis in early stages is excellent, recurrence can occur within two years post treatment. The mainstay of treatment is surgery, and long-term surveillance is essential to detect early recurrence. This case report of a 26-year-old woman diagnosed with JGCT presenting as a right ovarian mass. The patient underwent right unilateral salpingo-oophorectomy. Histopathology confirmed the JGCT without vascular invasion. Following surgery close monitoring was implemented and the patient successfully conceived and delivered a healthy child. During cesarean delivery, a small mass was found in the omentum and completely excised. The patient's stable clinical status and complete tumor resection chemotherapy were deferred. In early-stage JGCT, close surveillance and an individualized therapeutic approach can manage recurrences without chemotherapy, allowing patients to maintain fertility. The successful pregnancy outcome underscores the importance of careful management in young patient JGCT.

Keywords: Juvenile granulosa cell tumor, Pregnancy, Recurrent tumor

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1. Introduction

Juvenile Granulosa Cell Tumor (JGCT) is a variant of granulosa cell tumor originating from gonadal stromal cells. This tumor is included in a group of rare tumors, with a prevalence of about 5% of all granulosa cell tumors **(1)**. Most cases are found in children and adolescents before menarche, with a mean age of diagnosis below 20 years. Symptoms are often nonspecific, such as abdominal pain, abdominal distension, or abnormal uterine bleeding. This often leads to a delay in diagnosis, especially in young patients who are not considered at high risk for ovarian tumors **(2,3)**.

Histologically, JGCT shows a typical morphological pattern, including Call-Exner follicles, which are the main feature of this tumor. Although JGCT has a good prognosis, especially in the early stages, there is the risk of recurrence, especially in the first two years after primary therapy **(4-6)**. In stage I, surgery in the form of unilateral salpingo-oophorectomy is often considered sufficient to treat the disease. However, the therapeutic approach must be tailored to the patient's clinical condition, especially if the patient still wants to maintain reproductive function **(7)**.

This report highlights the challenges in the management of JGCT, especially in young patients who wish to preserve fertility. In addition, this report highlights the role of long-term surveillance in detecting early recurrences, which can contribute to the success of therapy and the patient's quality of life.

2. Case Presentation

A 26-year-old woman presented with chief complaints of irregular menstrual cycles and abdominal fullness for the past six months. The patient also complained of weight loss of approximately 5 kg in a period of three months without any clear cause, although appetite remained normal. There were no additional complaints such as fever, sharp abdominal pain, or changes in bowel patterns.

In further anamnesis, the patient revealed a history of a slowly enlarging abdominal mass without pain. Family history did not indicate any malignancy or genetic disorders. Physical examination showed abdominal distension in the right pelvic region with a palpable mass, well-defined, solid-cystic consistency, and non-tender when pressed. No lymphadenopathy or signs of ascites were found.

Transabdominal ultrasonography revealed a multilocular cystic mass measuring 2.0×2.7 cm and 2.1×1.5 cm originating from the right ovary, with hyperechogenic characteristics in some of its walls. Laboratory examination showed CA-125 levels within normal limits. Endometrial curettage cannot be performed because the patient was not married. Magnetic Resonance Imaging (MRI) of the abdomen showed a mass with well-defined boundaries without infiltration of surrounding organs, confirming the suspicion of granulosa cell tumor.

Then, the patient underwent laparoscopic surgery in the form of a right unilateral salpingo-oophorectomy. The procedure also involved removal of the mass identified in the right ovary along with a biopsy of the omentum to ensure no metastasis. Histopathology results showed Juvenile Granulosa Cell Tumor (JGCT) with low mitosis without vascular invasion.

Postoperatively, the patient was recommended to undergo close monitoring every three months for the first two years, including physical examination and abdominal ultrasonography. During the first year after the surgery, the patient remained asymptomatic and reported a return to normal menstruation.

At the next visit, the patient reported her first pregnancy. The pregnancy progressed without any complications, and the patient underwent a caesarean section due to oligohydramnios at 39 weeks of gestation. The patient underwent routine check-ups every 2 months and underwent physical examinations and abdominal ultrasonography during November 2019- September 2022. Then the patient had a second pregnancy. During the second caesarean section, a 2 cm mass was found in the omentum which was removed. The histopathology results again confirmed recurrent JGCT. After discussion with the medical team, the patient decided not to undergo chemotherapy because the recurrent mass was completely removed, and there were no other signs of metastasis. The patient was followed up for 1 year with MRI monitoring and the results showed no residual mass, normal left ovary with normal follicles. A year later the patient became pregnant for her third child.

3. Discussion

Juvenile Granulosa Cell Tumor (JGCT) has different clinical characteristics compared to adult granulosa cell tumor **(8)**. This tumor is often diagnosed at an early stage because symptoms appear more quickly due to the large size of the tumor or complications such as ovarian torsion. The main treatment for JGCT is surgery **(7,9)**. In the present case, unilateral salpingo-oophorectomy was sufficient to treat the primary tumor because the patient was in stage I without metastatic involvement.

It is important to note that although the prognosis of early-stage JGCT is excellent, the risk of recurrence remains. Studies show that approximately 10-20% of patients with stage I JGCT experience recurrence, especially within the first two years after primary therapy **(3)**. The recurrence found in the current patient in the form of a small omental mass suggests that long-term surveillance is essential to detect recurrent lesions at an early stage.

According to the stable clinical conditions and histopathology results showing good tumor differentiation, the patient decided not to undergo chemotherapy after detection of a recurrent mass. The literature shows that chemotherapy is usually recommended in cases with advanced stages or extensive recurrences **(10,11)**. In this patient, surgical removal of the recurrent mass provided adequate disease control without the need for additional chemotherapy. In addition, this approach allowed the patient to maintain reproductive function, as evidenced by three successful pregnancies after initial diagnosis.

In the management of JGCT, it is important to pay attention to the biological characteristics of the tumor. JGCT has a relatively indolent nature, with slow growth and low metastatic potential in the early stages. However, this tumor still has the possibility of recurrence, especially if not treated optimally. Therefore, early detection of recurrence is important to optimize therapy results **(12)**.

A conservative approach, as applied in this case, requires close surveillance. This surveillance involves regular clinical monitoring, imaging, and measurement of tumor markers, such as inhibin B or AMH (anti-Mullerian hormone). The use of these tumor markers has high sensitivity in detecting residual or recurrent tumor activity, allowing for early intervention **(6)**.

In addition to clinical aspects, psychological considerations of the patient also play an important role. The decision to forego chemotherapy and only undergo surveillance is a complex decision, requiring a thorough understanding of the risks and benefits of each therapeutic option **(13,14)**. The current patient demonstrated a good understanding of her clinical condition, which was the basis for successful conservative management.

In the context of fertility, the management of JGCT in young patients requires a multidisciplinary approach involving oncologists, gynecologists, and fertility specialists. Surgery aimed at removing the tumor without damaging healthy ovarian tissue is a challenge **(15-17)**. In the present case, unilateral salpingo-oophorectomy successfully preserved the function of the contralateral ovary, allowing the patient to conceive naturally.

The literature shows that pregnancy after JGCT therapy does not increase the risk of recurrence, so patients with preserved fertility can safely undergo pregnancy. However, pregnancy should be closely monitored, especially if there is a history of recurrence, as seen in our patient. Cesarean delivery performed in the first pregnancy also allows early detection of recurrent omental masses, which are then surgically treated **(14,15)**.

In the management of the recurrent JGCT, surgery remains the primary modality, especially if the recurrent lesion is local and can be completely removed. Chemotherapy may be considered in cases with extensive metastases or if the tumor cannot be completely resected. However, in patients with small recurrent lesions, as our case, surgical approach alone often provides satisfactory results **(7,16)**.

Overall, the current case emphasizes the importance of individualized management of JGCT. Considering the patient's age, desire to preserve fertility, and the clinical characteristics of the tumor, a conservative approach can provide optimal outcomes without compromising quality of life. This case also shows that with close long-term surveillance, recurrences can be effectively managed, allowing patients to have a productive and quality life.

4. Conclusion

Juvenile granulosa cell tumor has a good prognosis if diagnosed early and treated appropriately. As the present case demonstrated, close surveillance without chemotherapy may be a safe therapeutic option in cases with small recurrent masses and stable clinical conditions. Successful pregnancy in this patient underscores the importance of an individualized therapeutic approach to maintain quality of life.

5. Declarations

Acknowledgments

None.

Ethical Considerations

Written consent to publish this report for academic purposes was obtained from the patient.

Authors' Contributions

Mahida El Shafi contributed to the conceptualization, design, analysis, collecting data, and writing the paper. Brahmana Askandar Tjokroprawiro performed conceptualization, writing and reviewing the paper.

Conflict of Interest

The authors declare no conflict of interest.

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