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Pregnancy and Childbirth After Sertoli-Leydig Cell Tumor Resection: A Case Study and Literature Review

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Abstract: *Objectives:* To explore the clinical manifestations and pathological features in the biopsy of ovarian Sertoli-Leydig cell tumor, as well as to improve the clinical understanding of the disease. *Methods:* A case of pregnancy and childbirth after Sertoli-Leydig cell tumor resection was retrospectively analyzed. The patients' clinical data were collected, including the clinical manifestations, postoperative biopsy results, auxiliary examination results, immunohistochemical results, treatment, and prognosis of the patient. *Results:* (1) SLCT occurred unilaterally; (2) according to the International Federation of Obstetrics and Gynecology (FIGO), the clinical staging was stage IA; according to the pathological classification of malignant tumors, it was grade II (moderately differentiated); (3) a healthy female live baby was delivered. *Conclusion:* Such tumors are rare low-grade malignancies and are even rarer in pregnancy. An increase in preoperative testosterone levels with positive ultrasonography results can be used to assist diagnosis; however, postoperative biopsy pathology remains the "gold standard" for the diagnosis of SLCTs. The definite diagnosis of SLCTs is of great significance for surgical planning and prognostic evaluation.

Keywords: Ovarian tumor; Sertoli-Leydig cell tumor; Pathology; Diagnosis; Testosterone; Women

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1. Introduction

Ovarian Sertoli-Leydig cell tumor, also known as androblastoma or male blastoma, occurs mainly in young women and is usually presented as a unilateral solid or cystic-solid mass [1]. Having endocrine functions, its clinical manifestations include defeminized features, and later masculinized signs. The incidence of this disease is low, and there are limited literature reports on this disease at present. This paper discusses a case of natural pregnancy and delivery of a healthy female live baby after the removal of an androblastoma in the right ovary. Based on literature, data were reviewed and summarized for the diagnosis, treatment, and prognosis of the disease.

2. Case report

The patient is a 23-year old lady, married, and not pregnant. The main reason is that there was a cystic-solid mass in her right ovary for more than one month upon physical examination. She had amenorrhea for 5 years, with irregular menstruation in the past. Intermittent oral administration of Diane-35 (cyproterone

acetate and ethinylestradiol) was ineffective. On admission, the patient was obese, with a hoarse, deep voice. On physical examination, thick, black hair was noted all over her body (2 cm × 1.5 cm × 1.5 cm); her abdomen was distended, and a mass of about 5 cm × 5 cm in size, rubbery in nature, with clear boundaries, was palpable at her right adnexal area; there was mild tenderness upon palpation; the left adnexal area was normal.

Investigations for sex hormone determination: testosterone (T) 6.53 nmol/L, estradiol (E2) 42 pg/mL, progesterone (P) 0.4 ng/mL, prolactin (PRL) 610.14 ng/mL, luteinizing hormone (LH) 3.41 mIU/mL, follicle stimulating hormone (FSH) 4.51 mIU/mL, and dehydroepiandrosterone sulfate (DHEAS) 217.5 μg/dL. There were no obvious abnormalities in tumor markers and thyroid function. Gynecological ultrasound showed a cystic-solid mass of 5.3 cm × 4.9 cm × 4.8 cm in the right ovary. In January 2020, a laparoscopic right ovarian cystectomy was performed in a hospital in Shaanxi Province. The right ovarian tumor specimen was round, smooth, and rubbery, and the cut surface was uniformly yellow. Intraoperative quick freezing pathological results: no tumor cells were found in the peritoneal washings, normal ovarian tissue on the left, sex cord stromal tumor on the right, and testicular cells were seen; the specific type could not be determined. Postoperative pathology showed right ovarian androblastoma, with low-grade malignancy. Postoperative chemotherapy was recommended, but the patient refused. One month after the surgery, her menstruation returned, and subsequently, she was found pregnant. Her last menstrual period (LMP) was on February 17, 2020, and she was diagnosed with intrauterine pregnancy five weeks later. In the first trimester, she was treated for "threatened abortion" for half a month, and her thyroid function test about two months in pregnancy indicated subclinical hypothyroidism, which eventually resolved. On November 17, 2020, a 3,300 g female live baby was delivered via cesarean section, and the baby's development was normal. Peritoneal lavage fluid was collected during the surgery; right adnexectomy, left ovary biopsy, omentum biopsy, peritoneal biopsy (left and right paracolic peritoneal tissues, as well as rectum uterine lacuna peritoneal tissue), pelvic cavity sampling biopsy, pelvic diaphragm, liver, and spleen exploration, as well as tumor cytoreduction were done. On postoperative review, testosterone was more than 60 ng/mL, human chorionic gonadotropin was more than 10,000 mIU/mL, alpha-fetoprotein was 121 ng/mL, carbohydrate antigen 125 (CA-125) was 14.30 U/mL, human epididymis protein was 52.81 pmol/L. The pathological diagnosis was as follows: the frozen section showed no evidence of malignancy, and the paraffin section (right side) showed no obvious abnormalities in the ovary; (greater omentum) there were fat liquefaction and necrosis with foreign body granuloma inflammation; there was no tumor tissue involvement in the left ovarian tissue, bilateral paracolic inguinal peritoneal tissue, uterine-rectal lacuna, and right pelvic lymph node; no tumor metastasis was found in the left pelvic lymph node (0/2); (peritoneal lavage fluid) no cancer cells; (peritoneal washing fluid cell mass) no cancer cells were found. One month after being discharged from the hospital, the patient received chemotherapy as prescribed by the doctor and is still being followed up.

3. Discussion

3.1. Clinical features of ovarian stromal cell tumors

Ovarian Sertoli-Leydig cell tumor [2] originates from the sex cord and stromal tissues in the primordial gonad and consists of a mixture of different cells. The main symptoms are endocrine disorders, such as amenorrhea, acne, abnormal menstruation, and so on. With high androgen performance [3], more than 30% of patients have amenorrhea, increased acne, hirsutism, Adam's apple, deeper voice, and enlarged clitoris. Abnormal uterine bleeding or postmenopausal vaginal bleeding occurs in about 50% of patients. In this case, the patient felt a mass in her lower abdomen, which was rubbery, well-demarcated, and non-tender. A small number of patients will experience abdominal discomfort, such as bloating and abdominal pain. When the tumor ruptures or undergoes torsion, the abdominal pain will be aggravated, characterized by a tear-like or knife-like pain, radiating to the anus [4]. As the disease progresses, ovarian tumors may rupture, bleed, or undergo torsion, resulting in acute abdomen, or even life-threatening conditions. This patient presented with endocrine disorders; she had prominent hyperandrogenism, amenorrhea for five years,

obesity, thick, black hair over her body, thick vulvar, male distribution of pubic hair, clitoral hypertrophy, as well as high testosterone level.

3.2. Pathological features of ovarian stromal cell tumors

Microscopically, the tumor cells are mainly composed of two cellular components [5]: one is well-differentiated Sertoli-type cells, which are arranged in solid or hollow tubules; they are columnar cells, with sparse or lightly stained cytoplasm; they have small, oval or spherical nuclei, in which mitotic figures are rare; the other is Leydig-type cells, which are distributed in the interstitium singly or in sheets, especially around tubules; they are round granular cells, with abundant eosinophilic cytoplasm, having round nuclei; in the center or on one side, the fibrous mesenchymal tissue is relatively abundant, composed of closely arranged spindle cells, accompanied by varying amounts of collagen fibers. According to the degree of tubular differentiation of Sertoli cells and the proportion of primitive gonads, ovarian Sertoli-Leydig cell tumors can be divided into three pathological grades (Grade I, II, and III): well-differentiated, moderately differentiated, and poorly differentiated reticular type with heterologous elements [6]. Imaging examinations are mostly suggestive of solid, cystic-solid, or cystic masses. In this case, only the right ovarian androblastoma was excised. Rapid intraoperative pathological results revealed normal ovarian tissue on the left side and sex cord-stromal tumor on the right side. Testicular cells were seen, but the specific type could not be determined. Postoperative pathology revealed a right ovarian androblastoma, with low-grade malignancy. After explaining the condition to the patient's family, they refused to remove the affected ovary. Considering the patient's age and reproductive requirements, the bilateral appendages were retained following the patient's requirements; hence, only the tumor was removed. On re-examination after cesarean section, there was no recurrence, and the patient is currently receiving chemotherapy.

3.3. Treatment and prognosis of ovarian stromal cell tumors

Surgery is the mainstay of treatment for stromal cell tumors [7]. The purpose of surgery is to completely remove the tumor and achieve radical treatment. The resected ipsilateral appendage will be sent for pathological examination; if it is benign, the scope is sufficient [8], but if malignant germ cell components are reported, pelvic lymph node dissection, omentectomy, and para-aortic lymph node biopsy are required, in order to remove the diseased tissue to the maximum extent, so as to reduce recurrence and metastases. If the patient has fertility requirements, the contralateral appendage and its reproductive function can be preserved. The majority of patients with stromal cell tumors are curable with standard care; the recurrence rate is extremely low, and patients' quality of life is rarely affected [9], with the majority of cases having no impact on life expectancy. In this case, the patient had no recurrence after tumor resection, and she conceived naturally and gave birth to a healthy female live baby. Therefore, for young patients who desire to have children, if the uterus and contralateral appendages are normal, their reproductive function can be preserved.

Disclosure statement

The authors declare no conflict of interest.

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