CASE REPORT

Fertility-sparing Surgery in Sertoli-Leydig Cell Tumor of the Ovary: A Case Report

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Abstract

Sertoli-Leydig cell tumors are rare sex-cord stromal tumors. They arise from the sex cord sertoli cells and from the stroma of the genital ridge, the Leydig cells. They are generally of low-grade malignancy and rarely they can be poorly differentiated when they have a very aggressive course and poor prognosis. Majority of the tumors are characterized by androgen secretion. Most of the Sertoli-Leydig cell tumors occur in younger women less than 30 years of age, which necessitates the need for conservative management. A case of Sertoli-Leydig cell tumor of the ovary occurring in a 19-year-old girl, presenting with virilizing features is presented. She was treated with unilateral salpingo-oophorectomy and adjuvant chemotherapy.

Keyword: Case report, Fertility sparing, Polycystic ovarian syndrome, Prognosis, Sertoli-Leydig cell tumor, Sex cord tumor, Surgery, Surgical management.

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Case Description

A 19-year-old unmarried woman, presented with amenorrhea, voice change, and excessive hair growth in one year duration. She attained menarche at the age of 13 years, and her periods were regular with moderate flow. One year ago, her periods stopped abruptly and she noticed hair growth over the face, upper lip, chest, and trunk. The hair growth was progressive and rapid. She also noticed voice change which has become worse in the last 6 months. She has two elder sisters who are married and having children and no menstrual disturbances reported in them. There is no family history of malignancy. There is no history of weight changes or consuming medications for any ailments in the recent past. There is no bowel or micturition difficulties.

In her past history, six months ago she was investigated at the Government GH for secondary amenorrhea and the ultrasonography (USG) showed an enlarged ovary measuring 4 × 3 cm. The total testosterone was increased to 300 ng/dL and the 17(OH) progesterone levels were increased to 519 ng/dL. On the basis of the above findings, she was diagnosed as a case of congenital adrenal hyperplasia and was prescribed medications which she has not taken.

On examination, the body mass index was 18.5, there was no pallor or peripheral lymphadenopathy. There was excessive hair over the face, axilla, chest and trunk. The hair growth was dense from the pubic region to the umbilicus (Fig. 1). The Ferriman Galway score was 18. The breast examination was normal. On abdominal examination, no mass or ascites was made out. On local examination, there was clitoromegaly measuring 1.5 × 2 cm. As she had already undergone transvaginal USG, with prior permission, vaginal examination was carried out. The uterus was small in size and through the left fornix, a mobile soft cystic mass measuring 8 cm in diameter was felt. Right fornix was free. On rectal examination, there were no nodules felt in the pouch of Douglas (POD).

Her complete hemogram, liver function test (LFT), and renal function test (RFT) were normal. On repeat hormonal evaluation, total testosterone was 340 ng/dL, dehydroepiandrosterone (DHEA) level was 335 µg/dL and 17(OH) progesterone level was increased to 120 ng/dL. Cancer antigen 125 (CA 125), carcinoembryonic antigen

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Patient consent statement: The author(s) have obtained written informed consent from the patient for publication of the case report details and related images.

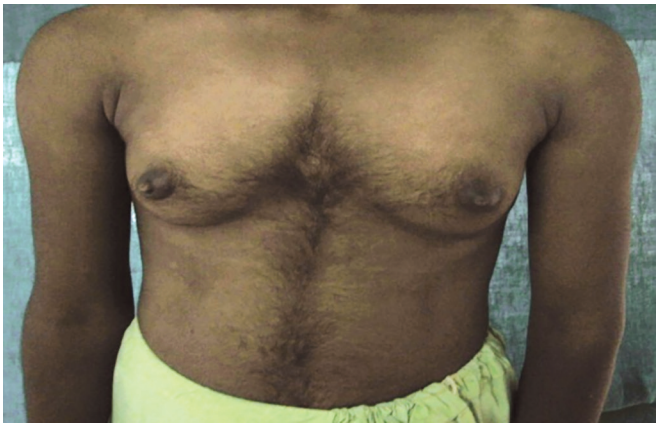

Fig. 1: Severe hirsutism over the abdomen and upper chest

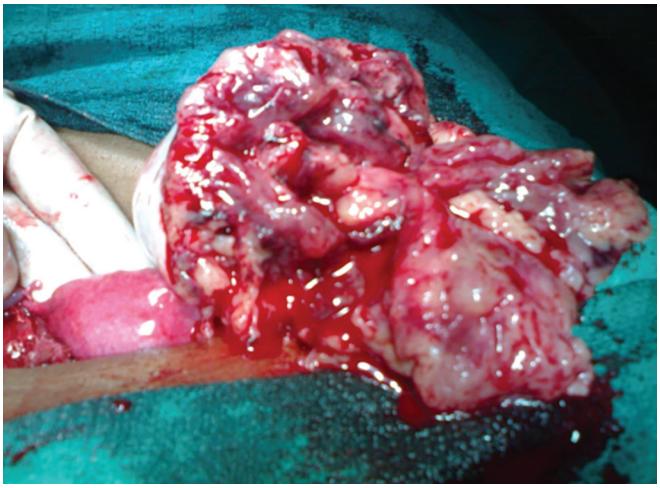

Fig. 2: Ruptured ovarian mass on the left side

(CEA), serum beta human chorionic gonadotrophin (serum β hCG), lactic dehydrogenase (LDH), and αfetoprotein were all negative. The thyroid stimulating hormone (TSH) level was 3.78. The chest X-ray was normal. On abdomen and transvaginal USG, the uterus measured 4.9 x 2.6 x 3.6 cm, a solid mass with cystic areas measuring 7.9 x 7 cm was seen arising from the left ovary. The right ovary was normal measuring 2.5 x 1.5 cm. There was no evidence of adrenal mass and other organs were normal. There was minimal fluid in the POD. On computed tomographic scan imaging, there was a heterogeneous adnexal mass with cystic areas arising from the left adnexa.

On the basis of the clinical findings and imaging studies, diagnosis of androgen-secreting tumor was made and laparotomy was proceeded with. Necessary consent was taken after discussing in detail about the fertility-sparing surgery. At staging laparotomy, there was hemorrhagic ascitic fluid measuring 200 mL which was taken for cytology. There was an left-sided ovarian mass measuring 10 cm in size which had already ruptured (Fig. 2). The right tube and the ovary were normal. There were no omental or peritoneal deposits. Left salpingo-oophorectomy was carried out along with infracolic omentectomy. Her post-operative period was uneventful. The histopathology was reported as moderately differentiated Sertoli-Leydig cell tumor showing hollow and solid tubules separated by fibrous stroma. There was no evidence of tumor cells in the ascitic fluid or the omentum. She was diagnosed as a case of Stage 1C malignant Sertoli-Leydig cell tumor of the ovary. She was referred to medical oncologists for further management and was given six cycles of chemotherapy with bleomycin, etoposide, and cis platin.

Her periods resumed after 3 months and she was getting periods once every 45–60 days at one year of follow-up. As the fertility-sparing surgery has been undertaken in a moderately differentiated Sertoli-leydig cell tumor, she was advised follow-up once in 3 months and early marriage and completion of the family at the earliest.

Discussion

Our case is presented for its rarity and for the conservative management undertaken in a young girl. Sertoli-Leydig cell tumors are also called androblastomas, and they constitute less than 0.5% of all ovarian tumors. Nearly 20% of Sertoli-Leydig cell tumors contain heterologous elements such as gastrointestinal epithelium, carcinoid, cartilage, and skeletal muscle.[1] Our case was a pure Sertoli-Leydig cell tumor.

In women presenting with hirsutism with or without virilizing features and minimally enlarged ovary, it may be mistaken for polycystic ovarian syndrome (PCOS) or congenital adrenal hyperplasia as in our case who was diagnosed as congenital adrenal hyperplasia 6 months earlier. However, rapidly progressive hirsutism with virilization should alert the clinician as to the possibility of androgen-secreting ovarian tumors or adrenal causes and should be evaluated thoroughly. Studies have shown that only in 40–50% of Sertoli-Leydig cell tumors, there are signs of androgen excess.[2] In the study by Guo et al.,[3] endocrine symptoms were seen only in 62% of patients diagnosed with Sertoli-Leydig cell tumors. Our case presented with rapidly progressive hirsutism and virilization. Conservative surgery is acceptable for young patients wishing to preserve fertility.[3] Majority of the Sertoli-Leydig cell tumors are unilateral and occur in young women. More than 90% of tumors are also diagnosed in stage 1 disease, therefore have a good prognosis. The prognosis depends on the degree of differentiation and the tumor stage.[4,5] Though our patient was also diagnosed in stage 1 disease, as the tumor had already ruptured, as well as it was a moderately differentiated tumor, she was given postoperative chemotherapy to prevent recurrences. Patients with Sertoli-Leydig cell tumors require lifelong surveillance. Besides symptoms, clinical findings and imaging studies, testosterone levels may be used as a useful tumor marker as this woman had elevated testosterone levels. In view of the possibility of recurrence, our patient was advised early marriage and to complete the family at the earliest.

The clinical significance of this case is rapidly progressive hirsutism with or without virilization should alert the clinician as to the possibility of androgen-secreting tumors.

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