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Case of a rare ovarian tumor: Sertoli - leydig cell tumor

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Abstract
Sertoli-Leydig cell tumors are germ cell tumor of ovaries. The cancer cells produce and release a male sex hormone which may cause the development of male physical characteristics (virilization), including facial hair and a deep voice. This type of tumor is sometimes called arrhenoblastoma of the ovary or a stromal tumor. Sertoli-Leydig cell tumors are rare tumors which account for less than 0.5% of all ovarian tumors. While they can be found in women of all age groups, they are most common in young women. Here is a case represented of a germ cell tumor in an 18year old patient and the line of management.

Keywords: germ cell tumor, arrhenoblastoma

Introduction

A 20year old unmarried nulligravida came to hospital with complains of amenorrhea since 6 months, associated with pain in abdomen and irregular menses since 1 year. Patient has complaints of virilization since last 6 months. No history of any PV discharge, fever, burning micturition or any other associated complain. No history of breathlessness, giddiness, cough, cold, bowel and bladder complaints.

Menstrual history: patient has menarche at 12 years of age, had normal menses till 17 years of age, now she has irregular menses since last 2 years with absent menses since 6 months.

Other significant history: Excess of facial hair. Voice change noticed since 6 months. No history of weight gain or reduction in last 1 year. No acanthosis or baldness. No history of blood in stools or hematuria. No medical or surgical history in the past. Personal: Appetite and sleep – normal bowel and bladder – normal Addictions - no history.

General examination: Height: 146 cms. Weight: 40kg
BMI: 19kg/m sq, Temperature – Afebrile, Pulse – 100/min BP - 100/68 mmHg, RR – 20/min Pallor - present (++), No history of cyanosis, edema, clubbing, icterus, lymphadenopathy, hydration.

Systemic Examination: Normal finding.
Per speculum and per vaginal examination not done. On inspection no abnormality detected.

Investigations: Routine blood investigations were done.
Significant findings: Hb- 7.4 g%; TLC- 7800/cumm; Plt- 2.3 lakh/cumm.

Table 1: Tumor markers values

Tumour markers	Normal ranges
CA 125 -15 U/ml	<35 U/ml
CA 19.9 - <2.0	0-37 U/mL
CEA 1.64ng/ml	<2.5ng/l (non-moker) <5ng/l (smoker)
Alpha Feto Protein -204.88ng/l	10-20ng/l
Free testosterone – 9.30 pg/ml	<40 Ugm/l
LDH – 206 U/l	150-320 U/l
Beta HCG - <1.20 IU/l	<5IU/l

Table 2: Radiological investigations

Ultrasonography	MRI pelvis
Well defined complex solid cystic lesion in left adnexa measuring 8 * 7.2 cm. left ovary not seen separately from the lesion. Solid component shows mild internal vascularity on color doppler images? Neoplastic ovarian mass. Uterus and right sided ovary normal with ET 6.1 mm No organomegaly or any other abnormality noted in the abdomen.	Well defined hyperintense solid cystic lesion noted in midline anterior to the uterus. It measures 6.2 * 4.4 * 3 cms. No evidence of fat density or calcification. Anteriorly it is closely abutting bilateral rectus abdominis muscle. Superiorly the lesion is moving the bowel loops. Posterolaterally it is in close relation with right common iliac vessels. Left ovary not seen separately from the lesion. S/o Sex cord stromal tumor. Right ovary is normal 26*16 mm. Uterus measures 4.2 * 2.0* 6.2 cm. appears normal in size shape and morphology.

Plan of management

1. Anemia of correction.
2. After all pre op evaluation and surgical fitness & pre-operative counselling done.
3. Plan exploratory laparotomy.
4. Frozen section will be sent.
5. Accordingly plan of unilateral or bilateral salpingo oophorectomy depending on the frozen report.
6. Histopathology report.
7. Follow up.
8. Chemotherapy or further management can be planned accordingly.

Intraoperative findings

- After proper fitness and all risks and consequences explained to the patient. Patient explained about the need of contralateral oophorectomy and oocyte banking explained to the patient. Patient not willing because of financial crunch.
- Patient explained about the need of further chemotherapy and radiotherapy.

Operative procedure

1. Intra operative findings

Left sided 5 * 6 cms tumor noted. Solid and cystic in consistency with vascularity present. Uterus and right sided ovary normal. No any other adhesions or any other finding. No ascites or any other finding.

2. Frozen specimen report

- Left ovarian mass shows features of sex cord stromal tumour.
- Peritoneal fluid: Cytospin prepared smear studied show pre dominantly hemorrhagic. Few clusters and sheets of round to oval cells are seen having mildly pleomorphic vesicular nuclei and moderate amount of eosinophilic cytoplasm.
- Final diagnosis will be based on histopathology finding.

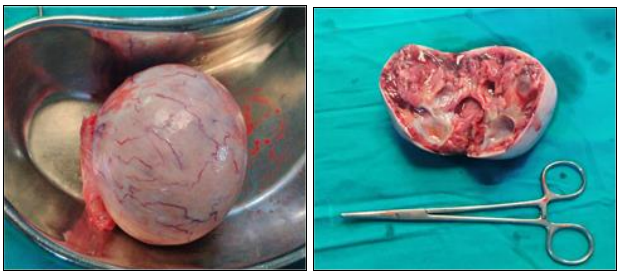

Fig 1: Surgically removed unilateral tumour with its cut section

3. Histopathological report

Sertoli leydig cell tumor– moderately differentiated with heterologous elements seen in – left ovarian mass.

- Left fallopian tube – unremarkable.
- Right ovarian biopsy - Stromal Hyperplasia.

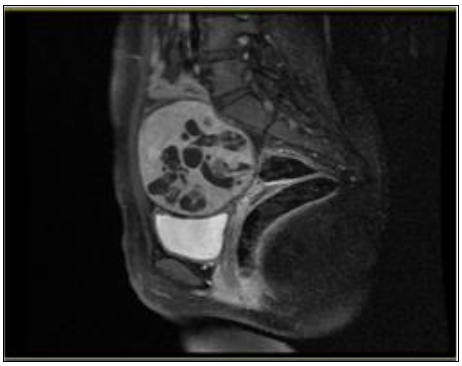

Fig 2: MRI pelvis- sagittal view of Sertoli Leydig cell tumor.

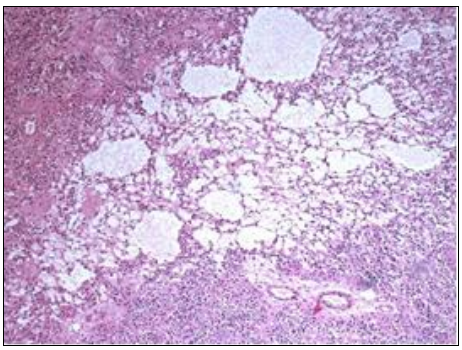

Fig 3: Well differentiated Sertoli- Leydig cell tumor: open Sertoli cell tubules (with blue columnar to oval nuclei) are admixed with numerous clusters of round, eosinophilic Leydig cells; Reinke crystal in the cytoplasm of Leydig cells.

4. Postoperative care

Patient stable post operatively. Check dressing of the patient d. .one on day 4 – wound healthy. Suture removal done on day 8 – wound healthy. Patient was explained about the further management to be given with chemotherapy.
Advise of onco physician reference given.

5. Follow up

Patient had menses in next month after the surgery with normal flow and no other associated complaints.

- Patient did not take chemotherapy and did not keep a follow up thereafter due to financial crunch.
- All risks and prognosis explained to the patient.
- Repeat counselling done but the patient did not keep any follow up.

Discussion

Here we are presenting a rare case of Sertoli Leydig cell tumors. These are very rare tumors composed of variable proportions of Sertoli cells, Leydig cells, and in the case of intermediate and poorly differentiated neoplasms, primitive gonadal stroma and sometimes heterologous elements [1]. Sertoli–Leydig cell tumor (a sex-cord stromal tumor), is a testosterone- secreting ovarian tumor and is a member of the sex cord-stromal tumor group of

ovarian and testicular cancers. Recent studies have shown that many cases of Sertoli–Leydig cell tumor of the ovary are caused by germline mutations in the DICER1 gene [2, 3]. These hereditary cases tend to be younger, often have a multinodular thyroid goiter and there may be a personal or family history of other rare tumors such as pleuropulmonary blastoma, Wilms tumor and cervical rhabdomyosarcoma. Closely related terms include arrhenoblastoma and androblastoma [4]. SLCT should always be considered in a young female patient who has symptoms of virilization and an ovarian mass on examination or investigation. Management issues mostly revolve around the histopathology of the tumor. Poorly differentiated tumors require aggressive management because the chances of them being malignant are high. Intermediately differentiated tumors need an individualized approach. The patient should be involved in all decision making. Our case emphasizes on the need for multidisciplinary management and importance of monitoring even after the operative procedure for better prognosis.

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