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RESEARCH ARTICLE

OVARIAN SERTOLI CELL TUMOUR: RARE CASE OF SEX CORD STROMAL TUMOUR

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ABSTRACT

Sertoli cell tumour is a sex cord gonadal stromal tumour of Sertoli cells. Sertoli cell ovarian tumours are very rare and constitute less than 0.5% of ovarian tumours. They differ from Sertoli leydig cell tumours in as they do not contain leydig cells or immature gonadal stroma. Most tumours are unilateral, confined to ovaries and are seen during second and third decades of life. These tumours are characterised by the presence of testicular structures that secrete androgens.

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INTRODUCTION

Sertoli cell tumour is a sex cord gonadal stromal tumour of sertoli cells.Sertoli cell ovarian tumours are very rare and and constitute less than 0.5% of the ovarian tumours. They differ from sertolileydig cell tumours in asthey do not contain leydig cells or immature gonadalstroma (Young and Scully, 1985). Most tumors are unilateral, confined to the ovaries, and are seen during the second and third decades of life (Zaloudek and Norris, 1984). These tumors are characterized by the presence of testicular structures that secrete androgens (Roth *et al*., 1981). Many patients have symptoms of virilization depending on the quantity of androgen production. Sertoli cell tumors occur most often in women of reproductive age group, but they occasionally arise in children and postmenopausal women. The average patient age is about 30 years (Young and Scully, 1985).

Case Report

A 24-years old, multiparous female presented to the surgery opd with complaints of pain in abdomen and lump in abdomen with intermittent episodes of constipation for 1 year duration. She had also noticed a gradual change in her voice for 1 year and excessive hair growth on her face, chest, and limbs for the last 6 months. In addition, she complained of abdominal discomfort due to abdominal lump.

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Patient has history of similar complaints in past for which she underwent laparotomy with excision of right ovarian mass with ovarian reconstruction 3 yrs back, histopathology report was suggestive of poorly differentiated sertoli cell tumour. Patient took chemotherapy inj. Paclitaxel and inj. Carboplatin 4 cycles.

On physical and clinical examination revealed pseudo precocity, hirsutismand hoarseness.

Per abdominal examination revealed a mass in left pelvic area 14 cm × 12 cm and ascites.

Ultrasonography: A cystic mass measuring 13 cm × 11 cm with ascitic fluid.

Contrast-enhanced computed tomography

Abdominal lump measuring 15 cm × 22 cm in size complex cystic mass is abutting anterior abdominal wall and displaces intestinal loops.

CA 125: 328 μ/ml.

Serum Calcium – 8.4 mg/dl

Serum Testosterone -10.98 ng/dl

Ultrasound guided fine needle aspiration cytology, which revealed the presence of benign cystic cells.

Routine Investigation

Hematological investigation: Hemoglobin - 14.5 gm%, Total leukocyte count - 9500/mm 3, Erythrocyte sedimentation rate - 20 mm at the end of 1 hour. Bleeding time and clotting time was within normal limit. Urine microscopy and biochemistry was normal.

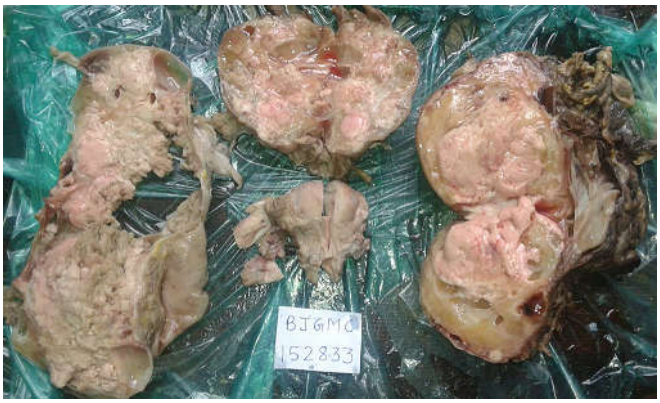
Her chest X-ray was clear and not shows any opacity.

The patient underwent exploratory laparotomy under general anesthesia because there was a strong suspicion of malignancy. INTRA-OPERATIVE FINDINGS – Midline laparotomy incision taken. Evidence of soft tissue mass measuring 16*14*6cm arising from the uterus and left ovary. Mass was predominantely solid but also with multiloculated cyst filled with hemorrhagic fluid. Mass could not be resected thoroughly as there was involvement of posterior wall of bladder and profuse oozing from the raw surface. Debulking of tumour done along with Hysterectomy with Left salpingo-opherectomy. Hemostasis achieved by abdominal packing. Sample was sent for histopathological examination.

Post-operative period was uneventful, patient discharged on postoperative 7th day and sutures were removed on postoperative day 14.

Pathologic Finding

Gross

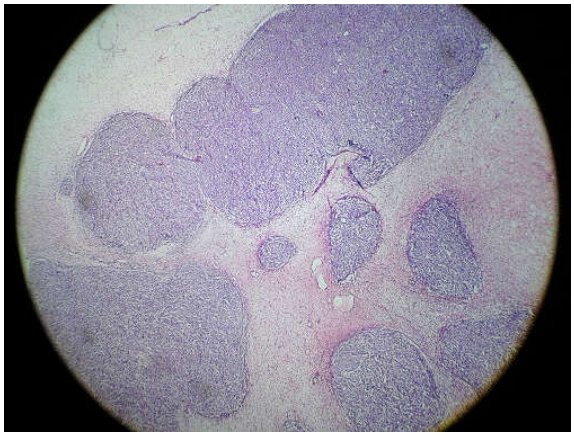


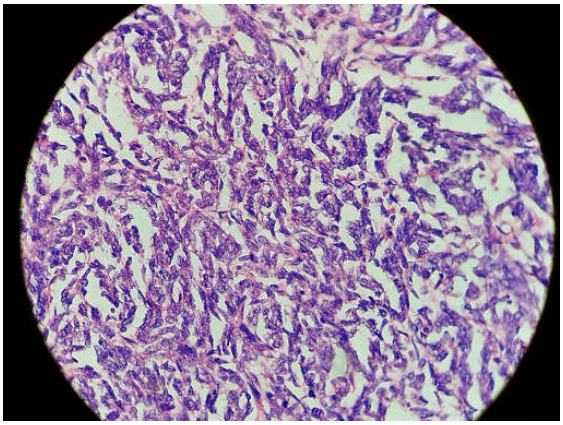
Uterus and cervix with adnexa large soft tissue mass measuring 16× 14× 6 cm. Externally mass has bosselated appearance on cut surface mass is solid yellowish in colour shows area with mucoid degeneration, cystic changes atperiphery and necrosis. Left sided ovary measures 5× 3 cm. Uterus with cervix measuring 6.5*5*3 cm.External appearance unremarkable. Endometrial thickness is 0.3cm.

- Myometrial thickness 1.2cm
- Endometrial canal 3cm
- Endocervical canal 2cm
- Left side fallopian tube measuring 4 cm in length.

Microscopy

Tumour tissue arranged in distinct islands and lobules separated by hypocellular areas.Tumour cells have oval to spindle shaped nuclei with. Marked nuclear pleomorphism and scantyeosinophilic cytoplasm. Few areas show tumour tissue arranged in glandular fashion.





Hypercellular areas with foci of leydig cell clusters are seen. Increased mitotic activity is noted, areas of hemorrhage and necrosis are seen. Impression: Sertolileydig cell tumour poorly differentiated. After 1 month of surgery patient was advised chemotherapy regimen comprising of inj. Paclitaxel and inj. Carboplatin. 2 cycles of chemotherapy were given 3 weeks apart. Patient came with recurrence and urinary complaints in the form of urinary obstructionwith dysuria after 3 months of chemotherapy. Pain relief achieved with superior hypogastric block under CT guidance.

DISCUSSION

Sertolicell tumour of ovary is rare neoplasm which belongs to group of sex cord stromal tumours of ovary and accounts for less than 0.5% of primary ovarian malignancy. It is characterized by uncontrolled proliferation of testicular structures in ovary (Young and Scully, 1985). Sertoli cell tumour of ovary canaffect any age group, however 75% occur during second and third decades of life. The neoplastic sertoli cells exhibit varying degrees of differentiation which include well differentiated, moderately differentiated, poorly differentiated and with heterologous elements (Weng *et al*., 2013). Degree of differentiation appears to be age related. Patients with poorly differentiated tumours appear to be 10 yrs younger than patients with well differentiated tumour. Majority of sertoli cell tumours are unilateral. At the time of presentation occurrence of extra ovarian spread is uncommon (2-3%) (Zanotti, 2002). Clinical signs and symptoms can be related to either hormonal production or the symptoms related to abdominal lump. Nearly half 50% of the patients show abnormal hormonal production mostly androgen excess or rarely estrogen excess (Osborn and Yannone, 1971). Clinical features of virilization can be seen in upto 30% of patients (Young and Scully, 1985; Zaloudek and Norris, 1984).

Symptoms due to androgen excess are hirsutism, acne, alopecia, hoarsenessof voice, breast atrophy, amenorrhea (Weng *et al*., 2013). Symptoms due to estrogen excess are precocious puberty, abnormal uterine bleeding, menstrual irregularities, uterine polyps (Prunty, 1967). Symptoms due to abdominal lump are feeling of heaviness, constipation abdominal pain, urinary complaints (Young and Scully, 1985; Zaloudek and Norris, 1984). Imaging studies can be utilized in the diagnosis of sertoli cell tumour .ultrasound of abdomen and pelvis is modality of preference for initial assessment. Ultrasound shows purely solid, purely cystic or mixed solid and cystic components. Mixed pattern is more common (60%) (Young and Scully, 1985) as in our in case.

Other imaging modalities such as CT, MRI, PETCT can be used for better characterization of tumour andextraovarian spread if any. Management of ovarian sertoli cell tumour is primarily surgical (Weng *et al*., 2013). As these tumours are most commonly found in reproductive age groupfertility sparing surgery can be considered in patients with well differentiated tumour (Young and Scully, 2002). Patients desiring fertility and having moderately or poorly differentiated tumour can be considered for unilateral salpingo-oophorectomy (Gui *et al*., 2012). Pelvic lymph node metastasis is extremely rare so widely accepted concept is that there is no need of pelvic lymphadenectomy (Sigismondi *et al*., 2012). Generally postoperative chemotherapy is considered for patients with poor prognostic factors such as advanced stage disease, moderate topoor differentiation, high mitotic index (Young and Scully, 1985). The first line chemotherapy regimen is bleomycin, etoposide and cisplatin (Young and Scully, 1985). Prognosis of sertoli cell ovarian tumours depends on degree of tumour differentiation and staging. The overall 5 year survival rate for well differentiated tumour is 100% whereas for moderately to poorly differentiated tumour is 80% (Young and Scully, 1985).

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