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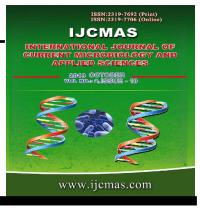

Original Research Article

A rare case of pure Sertoli Cell Tumour of ovary in a woman with Posthysterectomy status

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A B S T R A C T

Keywords

Sertoli cell tumour; ovarian neoplasm .

Sertoli-Leydig cell tumor (SLCT), also known as arrhenoblastoma, is a rare ovarian neoplasm which belongs to the group of sex cord-stromal tumors and accounts for less than 0.5% of all primary ovarian neoplasms. Very few case reports have been documented in the literature so far. Herein, we report a case of pure Sertoli cell tumor involving the left ovary in a 44-year-old woman who under went vaginal hysterectomy 9 years ago and presented with a 2-month history of a abdominal-pelvic mas, lower abdominal pain, and loss of appetite.

Introduction

Sertoli-Leydig cell tumor (SLCT) of ovary is extremely uncommon sex cord-stromal neoplasm. It occurs more frequently in third and fourth decades of life: 75% of tumours are seen in women younger than 40 years. It is mostly unilateral, and confined to the ovary and nearly 90% of the tumors presents in early stage .Occurrence of extra ovarian spread of SLCTs is extremely uncommon(2-3%). Bilateral ovarian SLCTs are exceptionally rare (1.5–2.0%).

Surgical resection remains the mainstay of management of ovarian SLCTs. The role of postoperative chemotherapy remains questionable and is only indicated in patients with poor prognostic factors.

Herein, we report a case of pure Sertoli cell tumor involving the left ovary in a 44-year-old woman who underwent vaginal hysterectomy 9 years ago and presented with a 2-month history of a pelvic abdominal mas, lower abdominal pain , and loss of appetite.

Case report

A 44 year old multiparous woman presented with 2 month history of abdominal mass, lower abdominal pain, and loss of appetite. She had undergone vaginal hysterectomy for dysfunctional uterine bleeding 9 yrs ago. Her Physical examination revealed midline mass of 20 weeks, firm, mobile, nontender, extending

from lower pelvis to umbilicus. Vaginal examination revealed cystic mass in the vault, more on the left side.

An ultrasound examination of the pelvis showed a 13cm x 5cm x 11cm heterogeneous solid cystic mass replacing the left ovary, with absent uterus (post hysterectomy status) and normal right ovary. Contrast-enhanced computed tomography (CT) scan of abdomen, and pelvis showed well defined, moderately enhancing solid mass of 15cmx3.5cmx13cm, extending from pelvis up to mid-abdomen just above umbilicus. Impression:? Ovarian mass/? broad ligament fibroid. There was no ascites. No significant lymphadenopathies in abdomen and pelvis were noticed. A chest radiograph (X-ray) showed no evidence of pulmonary nodules.

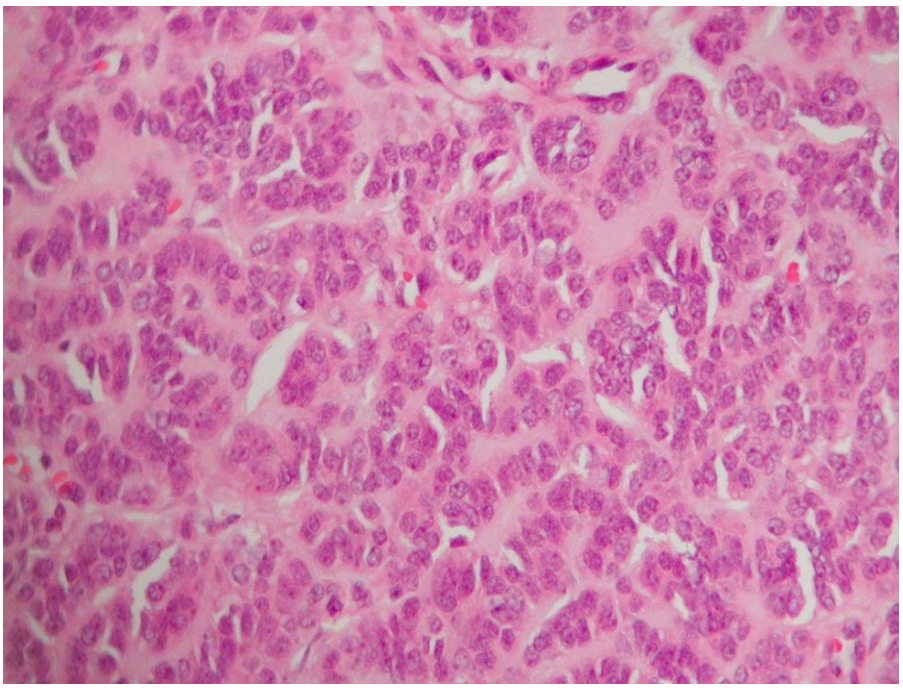
Laboratory tests showed, slightly increased levels of CA-125 of 65 U/mL (normal range <35 U/mL), and normal levels of carcino-embryonic antigen (CEA),alpha fetoprotein, and beta HCG. All other laboratory tests including complete blood count (CBC), renal, bone, hepatic and coagulation profiles, alkaline phosphatase,were within normal ranges.

Provisional diagnosis of ovarian neoplasm was made and staging laparotomy was done. Operative findings showed replacement of the left ovary by a 15x 3.5x 15-cm solid, grey-white, smooth-surface mass with intact capsule without spill and with no remnant healthy ovarian tissue. There was no spillage of tumor cells during surgery. The abdominal cavity was explored systematically but there were no deposits anywhere else in the cavity. The para-aortic lymph nodes were not enlarged. The right ovary was found to be normal. Left salpingo-oophorectomy and right ophorectomy was performed. Peritoneal washings were sent for cytologic examination for malignant cells. An omental biopsy was also done during the procedure. However, lymph node dissection was not performed.

Gross examination of the pathologic specimen showed well circumscribed ovarian mass measuring 15x3.5 x15 cm. The external surface was smooth. A cut section of the specimen revealed yellow nodules separated by thick bands of fibrosis with areas of heamorrhage. The histopathologic examination showed a tumor composed of columnar cells arranged in the form of tubules separated by dense fibrous septa. Also seen areas with papillary formation. Cells arranged around fibrovascular septa and solid sheets of cells. Cells contain oval nucleus with eosinophillic cytoplasm. (figure 1)Some cells contain vacuolated cytoplasm. Vascular invasion and capsular invasion were present . minimal atypia /mitosis seen . Right ovary, peritoneal washing, and omental biopsy did not reveal any abnormal cells. Immunohistochemistry revealed tumour cells positive for inhibin.

Based on the above findings, a final diagnosis of Sertoli cell tumour, stage IA, was made. The postoperative period was uneventful and the patient was discharged on the seventh postoperative day. In view of capsular invasion and vascular invasion, patient was advised follow up. CT scan done after six months of surgery revealed no recurrent /residual mass. And there no evidence of enlarged lymphnodes. Serum levels of testosterone and oestrogens were normal.

Figure.1 Cells contain oval nucleus with eosinophillic cytoplasm



Discussion

Sertoli-Leydig cell tumor (SLCT) of ovary is an uncommon sex cord-stromal neoplasm and accounts for less than 0.5% of all primary ovarian neoplasms. It is characterized by uncontrolled proliferation of naturally occurring testicular structures (Sertoli and Leydig cells) of varying degrees of differentiation in ovary. The neoplastic Sertoli and Leydig cells exhibit varying degrees of differentiation (grading) which include well differentiated, moderately differentiated, poorly differentiated, and with heterologous elements (Ahmed Abu-Zaid *et al*., 2013).

Clinical signs and symptoms of SLCT can be related to either hormonal production or presence of mass-occupying lesion. Clinical features of virilization is seen in more than one-third (33–38%) of patients Androgen-excess manifestations include virilism, hirsutism, acne, receding hairline, alopecia, hoarseness of voice, loss of subcutaneous tissue deposits, breast atrophy, clitoromegaly, oligomenorrhea and amenorrhea. Conversely, although rare, estrogen-excess manifestations include: precocious puberty, menstrual irregularities, generalized edema, weight gain, breast hypertrophy, endometrial hyperplasia, and endometrial carcinoma.

Elevated serum levels of testosterone and androstenedione can be often identified in approximately 80% of patients with ovarian SLCTs and virlizing manifestations.Testosterone serum levels >200 ng/dL (7 nmol/L) are generally associated with an androgen-secreting neoplasm from ovaries, adrenals, or elsewhere. Urinary 17-ketosteroid levels are often normal or slightly elevated in patients with SLCTs as opposed to patients with virilizing

adrenal tumors who often express extremely elevated levels of urinary 17-ketosteroid levels (Ahmed Abu-Zaid *et al*., 2013).

Nearly 50% of SLCT patients experience symptoms related to growing space-occupying lesions of ovary. These symptoms frequently manifest as abdominal/pelvic mass or pain. Pain is typically chronic and dull in nature, and occurs secondary to capsular expansion and possible subsequent compression of nearby visceral structures. Acute abdominal pain requiring prompt emergency intervention occurs in less than one-fifth of SLCT cases and can be attributable to ovarian torsion, capsular rupture, or bleeding (Ahmed Abu-Zaid *et al*., 2013).

Components of SLCTs can be purely solid, purely cystic, or mixed .Mixed (solid and cystic) components are most commonly seen in 60% of all ovarian SLCTs. Pure variants are rare. Well-and moderately differentiated SLCTs are the most frequently encountered histological variants. Mitotic figures are extremely rare.

Immunohistochemically, all SLCTs, including moderately and poorly differentiated variants, stain positive for inhibin and calretinin, and negative for epithelial membrane antigen (EMA) In addition, it has been shown that SLCTs stain positive for WT-1 and CD56 (Ahmed Abu-Zaid *et al*., 2013). Microscopically it shows a tubular growth pattern, but it may also have other growth patterns which can be extensive, making the correct diagnosis difficult. It consists of tubules lined by Sertoli cells and intervening nests of Leydig cells (Zizi-Sermpetzoglou *et al.,* 2010).

Management of ovarian SLCTs remains challenging owing to lack of standardized management protocol guidelines .Surgical resection remains the mainstay of management of ovarian SLCTs. Owing to rarity of ovarian SLCTs, limited number of documented case reports/series and lack of randomized clinical trials, effectiveness of post-operative chemotherapy remains questionable and requires further evaluation Generally, postoperative chemotherapy is considered for patients with poor prognostic factors such as: advanced disease staging, moderate-to-poor tumor grading, high mitotic profile, existence of heterologous elements and tumor rupture .The first-line and most frequently used chemotherapeutical regimen is bleomycin, etoposide, and cisplatin (BEP).Other regimens also exist such as (1) cisplatin, Adriamycin, and cyclophosphamide (CAP), and (2) cisplatin, vinblastine, and bleomycin (PVB) (Ahmed Abu-Zaid *et al*., 2013).

Prognosis of ovarian SLCTs is significantly correlated with degree of tumor differentiation (grading) and tumor extent (staging) Degree of tumor differentiation (grading) is related to age of the patient.Poorly differentiated SLCTs are seen in younger patients. (Ahmed Abu-Zaid *et al*., 2013).

Well-differentiated (grade 1) SLCTs are associated with zero malignant potential, whereas moderately (grade 2) and poorly (grade 3) differentiated SLCTs are associated with 11% and 59% malignant potential respectively . The overall 5-year survival rate for well-differentiated (grade 1) SLCTs is 100%, whereas for moderately (grade 2) and poorly (grade 3) differentiated SLCTs is collectively 80%. With respect to tumor staging, the overall 5-year survival rate for stage I is 95%

.whereas for stages III and IV is nearly zero percent . Long-term followup is highly advised in all patients (Ahmed Abu-Zaid *et al*., 2013).

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