

# AN UNUSUAL CYTOLOGICAL EXPERIENCE OF VIRILISING OVARIAN SERTOLI–LEYDIG CELL TUMOR – A RARE CASE REPORT

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## Abstract :

Sertoli–Leydig cell tumor of ovary is a gonadal tumour of the sex cord–stromal type. It is a rare tumor comprising 0.1 to 0.5% of all ovarian tumours. Peritoneal cytology has been well established as a diagnostic and staging tool in the management of the common epithelial tumours of ovary. Germ cell, mesenchymal, and sex-cord stromal tumours are much less frequently encountered in peritoneal specimens, often with cytologic features that may pose diagnostic difficulty and dilemma. We report a case of peritoneal fluid cytology of sertoli leydig cell tumor of ovary in a 20 year old female who presented with virilising symptoms. On removal of ovarian tumor, her virilising symptoms regressed, with regaining of menstruation.

**Keywords :** Ascitic fluid cytology, Sertoli-Leydig cell tumor, Virilising.

## Introduction :

Sertoli leydig cell tumors (SLCT) are rare sex cord-stromal neoplasms that account for less than 1% of ovarian tumors, occurring most commonly in young adults. A majority of the patients present with clinical features of virilization due to excessive secretion of testosterone by the tumor and more rarely estrogenization. <sup>[1]</sup> SLCT also known as arrhenoblastoma arise from specialized gonadal stroma capable of differentiating into both Sertoli- and Leydig-cells. <sup>[2]</sup> SLCT are commonly misinterpreted as yolk sac tumor, a serous neoplasm or as a malignant mixed mesodermal tumor where all of them show papillary configuration. Although Sertoli-Leydig cell tumors can be quite large, disease rarely extends beyond the ovary at

presentation and extra pelvic spread of tumor is exceedingly rare <sup>[2]</sup>. We report a case of SLCT presented with ascitis and diagnosis was made on cytological examination of ascitic fluid. Right

oophorectomy was done and histopathological examination confirmed the diagnosis. On removal of ovarian tumor and four cycles of chemotherapy with bleomycin, etoposide, and cisplatin regimen which were administered 3 times a week, her virilising symptoms regressed, with regaining of menstruation.

## Case report :

A 20 -year-old unmarried female presented with amenorrhoea since 1 year, excessive hair growth and hoarseness of voice since 5 months, loss of weight and appetite since 3 months. She attained menarche at the age of 16 years with regular cycles of 4/28 days. On physical examination there was excessive hair growth over the face and atrophy of both breasts. Per abdomen showed a mass of the size of a 28-week gravid uterus arising from the pelvis with moderate ascitis. Pervaginal examination showed clitoromegaly. USG abdomen showed mixed echogenic solid and cystic areas in the right adnexa. Computed tomography abdomen revealed a homogeneously enhancing solid and cystic mass arising from the right adnexa measuring 10x9x7 cm with moderate ascitis.

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Serum testosterone level was raised to 14.5 ng/dl. A low dose dexamethasone suppression test suggested an ovarian source. Ascitic fluid sent for cytology was hemorrhagic which showed cohesive clusters of small, relatively uniform cells with scanty cytoplasm and small rounded or blunt papillary fragments with hyalinised cores lined with small, mildly atypical cuboidal cells. (Figure 1). Cytological features in correlation with clinical and radiological findings were suggestive of virilising sertoli-leydig cell tumor. Right oophorectomy was done and sent for histopathological examination. Grossly the ovary measured 10x8x6 cms. Cut section showed greywhite solid and cystic tumor. Microscopy confirmed the cytological diagnosis of Sertoli–Leydig cell tumor. (Figure 2). In view of tumor stage I c, she was considered for chemotherapy. Four cycles of chemotherapy with bleomycin, etoposide, and cisplatin regimen were administered 3 times a week. She tolerated the chemotherapy well, with no major complications and is on regular follow up since six months. She has resumed her periods, with resolution of her virilisation symptoms. Testosterone levels were within normal range.

**Discussion :**

SLCT are of special interest because of the tendency to

occur in a younger age group with majority of cases presenting at an average age of 25 years. [3] There are marked differences in the frequency of ovarian tumors in the first three decades, where there is a greater percentage of germ cell and sex cord-stromal tumors and lesser frequency of surface epithelial neoplasms and great rarity of metastatic tumors. [4] The clinical characteristics associated with this tumor vary widely and are related to the degree of histologic differentiation and the presence of a retiform pattern and/or heterologous elements. [5] Although many SLCT are non-functioning, abnormal hormone production is found in greater than half of patients. [2] Hormonal manifestations due to SLCT generally occur over a relatively shorter period of time (months rather than years) [2]. They commonly present with androgenic manifestations characterized by defeminisation, or loss of female secondary sex characteristics. Rarely may they present with estrogenic features. [2] It is generally agreed that androgenic manifestations are less common in SLCT with retiform differentiation. They are called retiform variants because microscopically the growth patterns simulate those of the rete testis. [5] However our patient though histologically diagnosed as Retiform variant of SLCT presented with androgenic manifestations. Although SLCT can be quite large, disease rarely extends beyond the ovary at presentation and extra pelvic spread of tumor is exceedingly rare. [2] In peritoneal fluid cytology, SLCT are

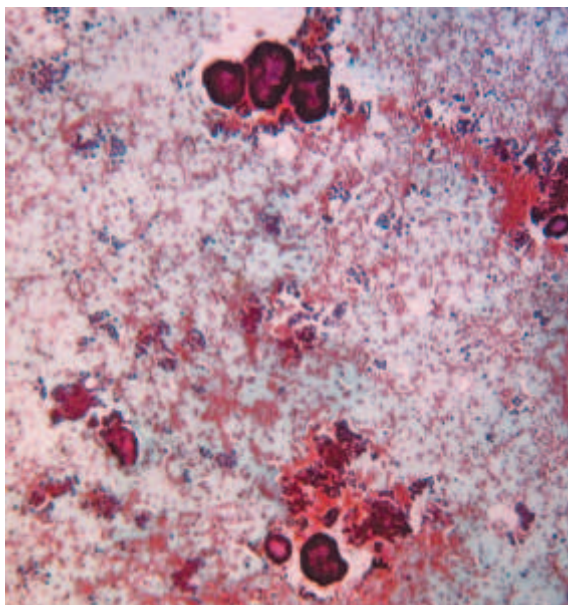


Figure 1) Ascitic fluid cytology showing cohesive clusters and papillary fragments in a hemorrhagic background. (PAP stain, 100 X).

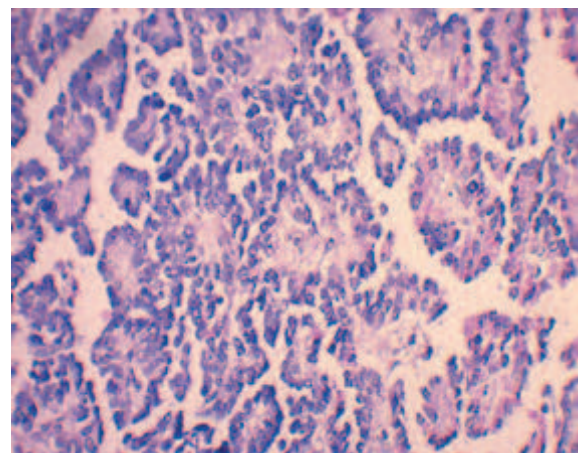


Figure 2) Microscopy showing predominantly retiform tumor areas in histopathology. (Hematoxylin & Eosin stain X100).

commonly misinterpreted as yolk sac tumor or a serous neoplasm. Because of the young age of the patient and frequent papillae, yolk sac tumor is often the most common differential diagnosis considered. The papillae in SLCTs do not have the central blood vessel of the classic papilla in a yolk sac tumor (Schiller-Duval body), but are cellular or are hyalinised. Resemblance to a serous borderline tumor may be imparted by the presence of cellular papillary clusters. The clefts, papillae and cellular atypicality may suggest a serous adenocarcinoma. Our patient presented with ascitis and diagnosis was made on

cytological examination of ascitic fluid which showed blunt papillary fragments with hyalinised cores lined with cellular mildly atypical cuboidal cells.

**Conclusion :**

SLCT are often misinterpreted as serous papillary cystadenocarcinoma or endodermal sinus tumor, which are more malignant neoplasms requiring different therapy, and hence recognizing this entity is important. This case report highlights the ascitic fluid cytological features of SLCT which helped in the preoperative diagnosis and proper management.

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