

# Bilateral Sertoli-Leydig Cell Tumor And Synchronous Adenocarcinoma Of The Colon In A Postmenopausal Woman Whose Left Ovary And Uterus Were In A Hernia Sac: Report Of A Case

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## Citation

Y ÖZDEMİR, S SÜCÜLLÜ, A FİLİZ, U BERBER, S ÖZDEMİR, E YÜCEL, M YILDIZ. *Bilateral Sertoli-Leydig Cell Tumor And Synchronous Adenocarcinoma Of The Colon In A Postmenopausal Woman Whose Left Ovary And Uterus Were In A Hernia Sac: Report Of A Case*. The Internet Journal of Surgery. 2009 Volume 22 Number 2.

## Abstract

Background: Sertoli-Leydig cell tumors are rare sex-cord stromal tumors, and seldom seen at postmenopausal women. It is an unfortunate situation when it is found near a colonic adenocarcinoma as a second primary tumor. Case presentation: A 76-year-old woman presented with anemia for a couple of years. She was operated for a left-sided hernia four years ago and she has a recurrent hernia on examination. Colonoscopy suggested an ulcero-vegetan tumor in the ascending colon. A right hemicolectomy with herniorraphy was planned. Laparotomy revealed that the hernia sac was containing the left ovary and uterus. The patient underwent right hemicolectomy, total hysterectomy and bilateral salphingo-oophorectomy. Pathological examination revealed the presence of a colonic adenocarcinoma coexistent with bilateral ovarian Sertoli-Leydig cell tumor. Conclusion: Clinical awareness and recognition of second tumors are important as they can change treatment strategies. Surgeons must be aware of the rare possibility of coexisting tumors.

## INTRODUCTION

Sertoli-Leydig cell tumor (SLCT) belongs to the group of sex cord-stromal tumors of the ovary and constitutes less than 0.5% of primary ovarian tumors<sup>1</sup>. The terms arrhenoblastoma, androblastoma, or gonadal stromal tumor of android type have all been used as synonyms for this cancer. Sertoli-Leydig cell tumors usually occur in young women and 70-75% of these tumors occur during the second and third decade of life<sup>2</sup>. Less than 10% of these tumors are seen either prior to menarche or after menopause<sup>2</sup>. They are usually unilateral and placed at the left ovary<sup>1</sup>. In two thirds of patients there is virilization, amenorrhea, hirsutism and gynecomastia caused by an elevated estrogen or androgen level.

The herniation of ovary and uterus into the inguinal canal is another rare situation and most of the reported cases are newborns and children<sup>3</sup>.

We presented the clinical and pathological findings of a patient with concomitant bilateral ovarian SLCT and colonic

adenocarcinoma in a 76-year-old postmenopausal woman whose left ovary and uterus were herniated into the inguinal canal. To our knowledge, the following description of a postmenopausal woman is the first case in the literature with the synchronous tumors of colonic adenocarcinoma and ovarian Sertoli-Leydig Cell tumor found in an inguinal hernia sac.

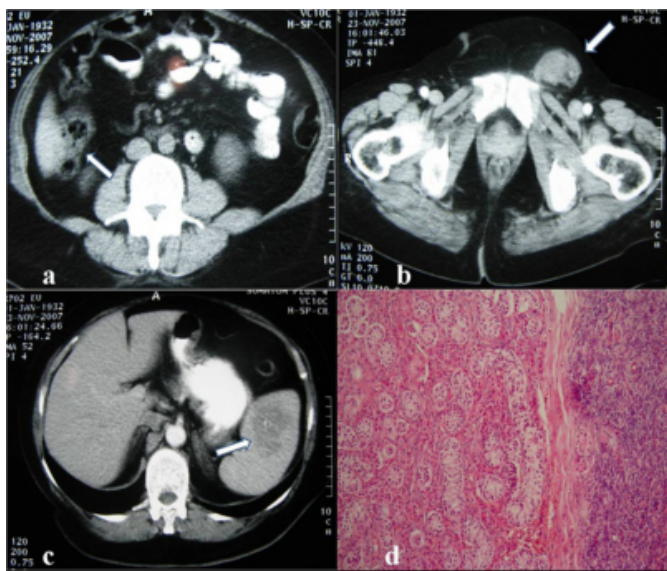
## CASE REPORT

A 76-year-old postmenopausal woman presented with anemia for a couple of years. Colonoscopy suggested an ulcero-vegetan tumor in the ascending colon. Endoscopic biopsy was reported as adenocarcinoma. She had been operated for a left-sided hernia four years ago and she had a recurrent hernia on physical examination. Biochemical examinations were normal except iron deficiency anemia. Tumor markers were normal. A computed tomography scan of the abdomen and the pelvis revealed a tumoral mass located in the ascending colon in 4x2cm dimensions, a lymphadenopathy located in the right iliac region, a left-sided inguinal hernia and a 6x5x5cm hemangioma at the

spleen.

### Figure 1

Figure 1: (a) Computed Tomography scan showing colonic adenocarcinoma located in the ascending colon, (b) Computed Tomography scan showing a left-sided inguinal hernia, (c) Computed Tomography scan showing an infarction area at the spleen, (d) High-power view of ovarian Sertoli-Leydig cell tumour



A right hemicolectomy with herniorrhaphy was planned according to the preoperative examination. Laparotomy revealed that the hernia sac was containing the left ovary and atrophic uterus, and both ovaries had irregular contours. After peroperative gynecology consultation, the patient underwent right hemicolectomy, total hysterectomy and bilateral salpingo-oophorectomy. Histological examination of the ovaries showed tumoral structure consisting solid tubules of Sertoli cells with surrounding Leydig cells. Immunohistochemical examination showed diffuse strong expression of inhibin and no EMA, Vimentin and S100 expression. The histologic examination of the right colon revealed a pT3, pN0, grade II adenocarcinoma. According to these findings, the diagnoses of colonic adenocarcinoma and bilateral SLCT of ovary were determined.

### DISCUSSION

Sex-cord stromal tumors of the ovary are uncommon ovarian malignancies, which account for less than 8% of all ovarian tumors<sup>4</sup>. Sertoli-Leydig cell tumors are rare, comprising less than 0.5% of all ovarian tumors, and contain Sertoli and/or Leydig cells in varying proportions. Reported cases vary between 2- and 76-year-old patients, but usually they occur in young women between 20-30 years<sup>5</sup>. Young and Scally

reported a review of 207 cases and bilaterality was seen only in 3 of these cases<sup>6</sup>.

Clinically, 2/3 of the patients may suffer loss of secondary sex characteristics and androgenic effects. However, less than 50% of the patients with Sertoli-Leydig cell tumors have no endocrine manifestations<sup>2</sup>. The diagnosis is usually determined after the evaluation of endocrinologic symptoms. In our case, there were no endocrine manifestations and SLCT was incidentally diagnosed, so the hormonal status was not evaluated preoperatively.

In bilateral tumors, the point that there are two primary tumors or one is the metastasis of the other was not investigated. A metastasis of a well-differentiated tumor is not very probable. Also, bilaterality of these lesions with both components makes us consider that these are neoplasms or developmental defects according to hormonal status.

The hernia sac may contain structures such as ileum, jejunum, colon, omentum, vermiform appendix, stomach, ovary, uterus, urinary bladder, acute appendicitis, Meckel's diverticulum or fallopian tubes<sup>7,8</sup>. Some of these are rarer than others. Although rarely encountered, most of the cases of inguinal hernia containing ovary and uterus were reported to be found in children<sup>3</sup>. Our case had a recurrent inguinal hernia and the first operation note could not be obtained. Constipation and obesity were thought to be the cause of the herniation.

These two rare situations were detected in our patient at the time of operation for colonic adenocarcinoma. Herniation of uterus and ovary at the same time makes our patient a research case.

Because of bilaterality of a rare tumor in a postmenopausal woman, the herniation of an ovary and the uterus, the absence of androgenic effects, good differentiation despite bilaterality, and coexisting colonic adenocarcinoma and splenic hemangioma, we thought that this case would be helpful in literature. Clinical awareness and recognition of second tumors are important as they can change treatment strategies. Surgeons must be aware of the rare possibility of coexisting tumors.

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