Ovarian Endometrioid Adenocarcinoma With a Yolk Sac Tumor Component in a Postmenopausal Woman: Case Report and Review of the Literature

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Clinical Practice Points

- Yolk sac tumors (YSTs) of the ovary are rare and highly malignant germ cell tumors of utmost importance, occurring in children and young adults. They are characterized by endodermal differentiation.
- YSTs coexisting with a variety of histologic patterns have been described but those with an epithelial malignant component are extremely rare, especially in postmenopausal women.
- We describe a rare case of ovarian endometrioid adenocarcinoma (EOC) with a YST component occurring in a 73-years-old woman (pT1aN0M0) that was treated with a combination of paclitaxel and carboplatin for 6 cycles. At 22-months' follow-up, the patient was free of recurrence. This is the longest disease-free survival seen when compared with other reported cases in the literature.
- No conclusions could be drawn from this case report; we hope that other authors describe their experiences to define the most appropriate approach to this rare tumor.

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Introduction

Malignant germ cell tumor is an uncommon type of ovarian cancer that accounts for fewer than 5% of total ovarian cancers.[1] Yolk sac tumors (YSTs) of the ovary, also called endodermal sinus tumors (ESTs), are rare and highly malignant germ cell tumors of utmost importance, occurring in children and young adults[2]; they are characterized by endodermal differentiation.[3] YSTs represent about 7.3% of malignant germ cell tumors.[4] YSTs coexisting with a variety of histologic patterns has been described but those with an epithelial malignant component are extremely rare.[5] We describe a rare case of ovarian endometrioid adenocarcinoma with a YST component occurring in a postmenopausal woman.

Case Report

We report a case of a 73-year-old woman. She observed an increase in abdominal girth. An ultrasonogram showed a large unilateral right ovarian mass, which was confirmed by subsequent computed tomography (CT). The patient underwent open abdominal surgical exploration: Bilateral salpingo-oophorectomy and omentectomy were performed. The pathologic examination resulted in a diagnosis of endometrioid adenocarcinoma (EOC) with a YST component in the right ovary (Figures 1, 2); cytologic examination of peritoneal fluid was negative for neoplastic disease. She was staged as having pT1aN0M0 disease. Before surgery, tumor markers (serum alpha-fetoprotein, cancer antigen 125 [CA125], and carcinoembryonic antigen [CEA]) were not elevated. After collegial discussion, considering the predominance of the endometrioid adenocarcinoma component, we decided on postoperative treatment consisting of 6 courses of paclitaxel (175 mg/m^2) and carboplatin (AUC4). The patient was treated with chemotherapy without any problems. At 22-months' follow-up, the patient was free of recurrence. CT results were normal and tumor markers (serum alpha-fetoprotein, CA125, and CEA) were not elevated.

Discussion

We describe a rare case of ovarian endometrioid adenocarcinoma (EOC) with a YST component occurring in a 73-year-old woman. Twenty-four cases of the coexistence of EOC and YST in postmenopausal women have been reported in the literature[6] and of them, our patient had the longest disease-free survival.[7,8] The clinicopathologic profile of YST arising in association with EOC differs from that of pure

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Figure 1 Endometrioid Adenocarcinoma With a Yolk Sac Tumor Component in Right Ovary

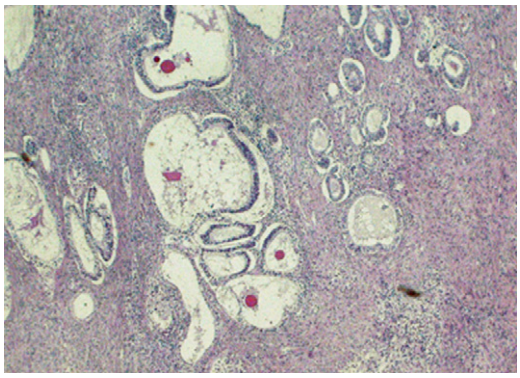
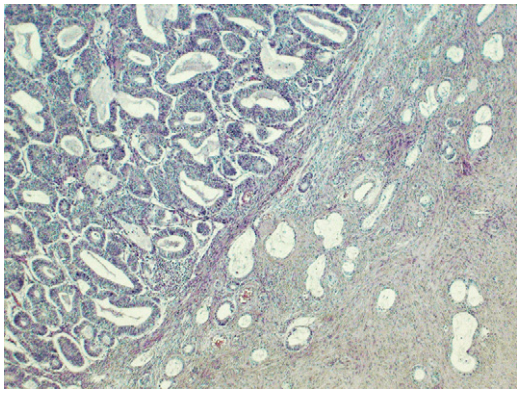


Figure 2 Endometrioid Adenocarcinoma With a Yolk Sac Tumor Component in Right Ovary



YST.[7] It is reported that the development of extraembryonal germ cell tissue from tumors of somatic origin is a rare event in the female genital tract and occurs mainly in the aerodigestive and urinary tracts.[9] There are 4 different theories for this event: the teratoma theory, retrodifferentiation, the collision theory, and the neometaplasia theory.[8,10,11] Neometaplasia, also called aberrant differentiation, is the most reasonable theory to explain the histogenesis of this tumor: somatic carcinomas have the capability for germ cell differentiation, and the germ cell component is thought to derive from somatic mesodermal cells and not from germ cells.[7] Clinically these tumors are characterized by rapid growth and extensive intraabdominal spread. The patients usually present with abdominal pain, a palpable mass, abdominal distention, and vaginal bleeding.[1] Because of its rarity, YST prognostic factors remain unclear; the stage, an ascites volume of $<$ 100 mL, and a residual tumor measuring $<$ 1 cm tend to affect the prognosis of YST positively.[12] Also, tumor markers such as alpha fetoprotein and human chorionic gonadotropin are considered in the prognostic factors.[13] There are no randomized studies relating to the management of YST of the ovary; there are only retrospective reviews and single case reports. Surgical diagnosis and treatment consists of laparotomy with bilateral salpingo-oophorectomy, omentectomy, multiple peritoneal and abdominal biopsies, peritoneal cytologic studies, and resection of all visible disease. BEP (bleomycin, etoposide, cisplatin) is the current standard therapy for ovarian germ cell tumors.[2,9] Chemotherapy is effective for pure YST;[14] we do not know if YST with EOC responds to chemotherapy[5] or which kind of chemotherapy.

After collegial discussion and considering that our patient's disease is more similar to EOC than to YST (seeing a preponderance of the EOC component), we chose a combination of paclitaxel and carboplatin for 6 cycles. In 1 study, it was believed that regimens for YST may be appropriate in these rare tumors;[8] other studies used a combination of BEP and paclitaxel and carboplatin, concluding that this particular tumor is more similar to EOC than to YST in clinical behavior and suggesting that a combination of paclitaxel and carboplatin should be compared with BEP.[7] Other authors also reported that neoplasms with a YST component in older women are less responsive to the chemotherapy used for ovarian germ cell tumors, and adjuvant therapy should include aggressive platinum-based chemotherapy designed to treat both epithelial ovarian cancer and germ cell tumors. These authors reported that 2 recent patients with stage 1 disease (as in our patient) treated aggressively with platinum-based chemotherapy are living and have been disease free $>$ 1 year after operation.[6] Finally, no conclusion could be drawn from this case report; we hope that other authors describe their experiences to define the most appropriate approach to this rare tumor.

Disclosure

The authors have stated that they have no conflicts of interest.

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