Unusual Collision Tumor of Ovary: Endometrioid Adenocarcinoma and Teratoma

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Introduction

Collision tumors represent the coexistence of 2 histologically distinct tumors adjacent to one another in the same organ, without admixture of the components of the tumors. Although uncommon, they have been described in almost all major organs, with a variety of component pathologic features. The majority are diagnosed postoperatively after histologic examination. The origin of these tumors has been debated. It is proposed that collision tumors could arise from 2 different cell lines growing at the same time side by side or a chance occurrence of 2 tumors in the same organ. The other commonly held view is that the origin is from a common precursor pluripotent stem cell. Cancer is considered to be a disease of the stem cell, and given the plasticity of the stem cell to differentiate, this theory is made more plausible. Ovarian collision tumors are rare. We report a case of collision tumor of the ovary composed of teratoma and endometrioid adenocarcinoma.

Case Report

A 71-year-old para I woman presented with a 4-month history of abdominal swelling and postmenopausal bleeding. She attained menopause at the age of 50 years and had no previous gynecologic problems. Her past medical history was unremarkable and she was on no medications. On examination, she had a large mass arising from the pelvis and reaching up to the umbilicus. CA125 levels were elevated at 256 U/mL and carcinoembryonic antigen levels were normal. Pelvic ultrasonography showed a large complex mass in the pelvis with cystic and solid components, measuring $27 \times 19 \times 15$ cm, and neither ovary was visualized separately. Computed tomography (Figure 1) confirmed the large complex mass occupying the pelvis and lower abdomen. There was no ascites or presence of disease elsewhere.

The risk of malignancy index (RMI) scoring system can be used to predict the risk of malignancy of adnexal masses. It incorporates ultrasonographic features, menopausal status, and preoperative CA125 level, and is calculated using the equation: RMI score = ultrasound score $\times$ menopausal score $\times$ CA125 level (in units per milliliter). Her RMI was high at 2304 ($3 \times 3 \times 256$) and she proceeded to surgery.

At surgery, a right ovarian mass measuring 40 cm was found to almost reach the inferior surface of the liver, with a cystic component within the abdomen and a solid component in the pelvis. The tumor was adherent to the right pelvic side wall, anterior surface of the rectum and sigmoid colon, and back of the uterus. The adhesions were lysed, and a total abdominal hysterectomy, bilateral salpingo-oophorectomy was carried out, along with right pelvic node dissection and infracolic omentectomy.

The gross specimen showed replacement of the right ovary by a solid lobular tumor with an adjacent cyst having a mural nodule with multiple small cysts (Figure 2A). The left ovary, fallopian tube, and uterus were grossly unremarkable. The microscopic findings were very interesting. There were 2 main histologic components. The first component was a teratoma with a variety of mature elements. Within
the teratomatous component there were foci of carcinoid, which were positive for chromogranin and CD56 (Figure 2B and D), and a yolk sac tumor that was positive for alpha fetoprotein (Figure 2C). There were also scattered foci of immature neuroepithelium consistent with immature teratoma (Figure 2E). The second major component was that of a poorly differentiated adenocarcinoma with both glandular and solid elements (Figure 2F). The solid elements were composed of cells with a rather spindled appearance. Diffuse positivity for EMA, Ber-EP4, and cytokeratin markers is consistent with the carcinoma and the morphologic features, and positive staining with CK7, ER, CA125, and vimentin is supportive of endometrioid adenocarcinoma.

In summary, this was a stage IA collision tumor of ovary composed of a teratomatous neoplasm with components of an immature teratoma, yolk sac tumor, and carcinoid tumor, and a poorly differentiated endometrioid adenocarcinoma.

The patient received 6 cycles of adjuvant carboplatin chemotherapy and is currently on surveillance.

Discussion

Collision tumors involving the ovary are quite rare, and various combinations have been reported. They include combinations of cystadenocarcinoma and dermoid cyst, teratoma and mucinous cystadenocarcinoma, carcinosarcoma and dermoid cyst, choriocarcinoma and cystadenoma, sarcoma and mucinous tumor, sarcoma and serous carcinoma, serous cystadenoma and Sertoli-Leydig cell tumor, and granulosa cell tumor and ovarian hepatoid carcinoma. Teratoma appears to be a more common component among the reported combinations of collision tumors of the ovary. The collision of teratoma and endometrioid adenocarcinoma described here has not been reported previously to our knowledge. Could the endometrioid adenocarcinoma have risen within the teratoma? Malignant transformation, although uncommon, is a well-recognized complication of teratoma. The majority of these transformations are squamous cell carcinomas. Other uncommon transformations described are carcinoid tumors and adenocarcinomas, as well as serous papillary adenocarcinoma and clear cell carcinoma.

We are not aware of an endometrioid adenocarcinoma arising from a teratoma being described in the literature. Ohishi et al reported a case of ovarian tumor in a premenopausal woman, composed of malignant germ cell tumor and epithelial tumor with components of endometrioid adenocarcinoma and clear cell adenocarcinoma. The germ cell component consisted of yolk sac tumor, immature teratoma with neuroectodermal and rhabdomyosarcomatous differentiation. However, they reported that the epithelial component arose from the endometriosis rather than from the teratoma.

We believe that our case is a collision tumor, although it is impossible to exclude the possibility of endometrioid adenocarcinoma arising directly from the teratoma. Molecular studies of the tumors can be helpful in defining the clonality and histogenesis of these tumors. Furlan et al used microallelotyping to define the clonal origin of collision tumors of the gut. The molecular analysis, although helpful to identify clonal origin of tumors, is a research method and to our knowledge is not done routinely. We have not carried out clonal analysis because we do not have such facilities at our center.

The collision tumors are diagnosed postoperatively because there are no specific features that aid their diagnoses preoperatively. Kim et al retrospectively analyzed radiologic findings in pathologically confirmed collision tumors associated with teratoma to identify features that might point to their existence before surgery. They found that 6 of 7 collision tumors had radiologic clues such as the presence of nonfatty fluid in the cyst and a large solid component in the ovarian mass, which pointed toward the presence of a collision tumor. Such clues, in addition to frozen section analysis, could help in deciding on further management, particularly the type and extent of surgery. In our patient, there were no specific radiologic or intraoperative clues...
to the presence of collision tumor; that was revealed only by histologic diagnosis postoperatively. It is therefore crucial to carefully sample the areas to diagnose collision tumors.

The management of collision tumors can be challenging, particularly if the components of the collision tumor are of diverse histologic types, affecting prognosis. The factors that need to be considered in deciding further management after surgery are the most aggressive component of the collision tumor and the stage, which will determine the prognosis. In our case, decision about further management of the collision tumor was guided by its dominant pathologic fea-
tures. Although the guidelines recommend extensive pelvic nodal sampling for germ cell tumors confined to the ovaries/pelvis, in this case there was no suggestion of tumor harboring germ cell components either preoperatively or at surgery. The tumor was staged surgically as FIGO stage I, and the multidisciplinary team felt that further surgical staging was not required given the good prognosis associated with the dominant components of the collision tumor. As far as the teratomatous component is concerned, there is evidence that a surveillance policy could be a reasonable choice in stage I malignant ovarian germ cell tumors, as these carry good prognosis and could be salvaged with platinum-based chemotherapy should they relapse. The other component of collision tumor, poorly differentiated endometrioid adenocarcinoma, on its own is an indication for platinum-based adjuvant chemotherapy. Ovarian endometrioid adenocarcinoma carries a much better prognosis compared with the other histologic variants, stage for stage. We discussed all the options with the patient, and on balance we decided to offer adjuvant chemotherapy with single-agent carboplatin with active surveillance, which was acceptable to the patient.

In summary, this is a unique collision tumor of the ovary, a teratoma coexisting with endometrioid adenocarcinoma, which, to our knowledge, has not been reported previously. It is possible that endometrioid adenocarcinoma could have arisen from a teratoma but such an event has not been reported.

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References