SHORT COMMUNICATION

SYNCHRONOUS OVARIAN ENDOMETRIOID ADENOCARCINOMA AND ENDOCERVICAL MUCINOUS ADENOCARCINOMA

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SUMMARY

Objective: We report a rare case of synchronous cancer consisting of ovarian endometrioid adenocarcinoma and endocervical mucinous adenocarcinoma. Related literature was reviewed and it appeared that no similar case had been reported previously.

Case Report: A 30-year-old (gravida 1, para 1, abortus 0) woman complained of abdominal fullness, chest tightness and dyspnea on exertion of several days’ duration. Gynecologic sonography showed a right complex adnexal cyst, 16 × 14 cm in size. Computed tomography showed an 18 × 16 cm right pelvic tumor, with both cystic and solid components, ascites and bilateral massive pleural effusion. Cytology of the pleural effusion showed no malignant cells. The patient underwent staging surgery. Histology showed moderately to poorly differentiated endometrioid adenocarcinoma of the right ovary with extensive lymphovascular permeation, as well as paraaortic and bilateral pelvic lymph node metastases. Extensive tumor thrombi were observed in the lymphovascular channels of the left ovary, bilateral tubes and uterus. Endocervical adenocarcinoma, <3 mm in depth, was also identified on the cervix. The final surgical-pathologic stage of ovarian endometrioid adenocarcinoma was stage IIIc and of endocervical mucinous adenocarcinoma was stage IA1. Adjuvant chemotherapy with carboplatin and paclitaxel was prescribed postoperatively, but the malignancy was not controlled due to lung, brain and vulva metastases. The patient died of respiratory failure.

Conclusion: The coexistence of primary neoplasms in the ovary and cervix is rare. Diagnosis should be based on histologic examination and requires appropriate treatment for both tumors. [Taiwanese J Obstet Gynecol 2006; 45(3):264-267]

Key Words: endometrioid adenocarcinoma, mucinous adenocarcinoma, synchronous tumor

Introduction

Multiple primary cancer is a rare condition. The concept of multiple malignant lesions has interested investigators since 1889, when Billroth first described the condition [1]. Synchronous and metachronous are terms used to describe the occurrence of second primary tumors. Synchronous tumors occur, or are diagnosed, at the same time; metachronous tumors occur at different times. Moore et al introduced the concept of synchronous tumors and defined them as secondary tumors that occur within 12 months [2]. If a new tumor develops many years after the first, then the previously administered radiotherapy or chemotherapy must be considered to be a possible carcinogen. It is often postulated that when two tumors occur in close succession, they may have been exposed to the same hormone or carcinogen. The most common synchronous gynecologic tumors are ovarian and endometrial; synchronous ovarian and endocervical tumors have rarely been reported in the past.
Case Report

A 30-year-old woman (gravida 1, para 1, abortus 0) with no significant past medical history visited our outpatient department on January 4, 2005 with the complaints of chest tightness, dyspnea on exertion and orthopnea of several days’ duration. She had a nontender abdominal mass, increased abdominal girth, nausea and decreased appetite over the past month. Gynecologic sonography showed a right complex adnexal cyst, measuring 16 × 14 cm in size, with ascites and bilateral pleural effusion. She was admitted for further evaluation the same day. Abdominal and pelvic computed tomography (CT) showed a large tumor with both cystic and solid parts in the pelvis, mainly on the right side, ascites and massive bilateral pleural effusion (Figure 1). Cytologic examination of the pleural effusion, performed twice, showed no malignant cells. The serum CA-125 level was 702 U/mL but the carcinoembryonic antigen (CEA) level was within the normal range. The results of preoperative examinations, including blood, urine, liver function, renal function, electrolytes, electrocardiogram and chest X-ray, were all within normal ranges except for bilateral pleural effusion.

The patient underwent laparotomy on January 7, 2005. Yellowish, watery ascites (~1,000 mL) was found in the abdominal cavity and was sent for cytologic examination. The cystic mass, 16 × 14 × 15 cm, had a dark red smooth surface with no adhesions to the peritoneum or intestines. The internal content comprised fragile soft tissue soaked in a cola-like fluid and a small daughter cyst containing chocolate-like fluid. Frozen section of the right ovary disclosed endometrioid adenocarcinoma with papillary architecture and massive tumor necrosis. The left ovary had a rough surface measuring about 4.5 × 4 × 4 cm in size. The internal content was a chocolate-like fluid. There was no enlargement of the retroperitoneal lymph nodes. Staging surgery was performed, including total abdominal hysterectomy, bilateral salpingo-oophorectomy, bilateral pelvic lymph node dissection, paraaortic lymph node sampling, bilateral infundibulopelvic ligament resection, appendectomy and omentectomy.

In the final pathology report, the section of the right ovary mass showed a picture of grade 2/3 endometrioid adenocarcinoma with tumor necrosis, papillary growth pattern, endometriosis and extensive lymphovascular permeation (Figure 2). Metastatic adenocarcinoma with tumor thrombus in the lymphovascular channels was found on the left ovary, bilateral tubes, uterus, cervix, bilateral pelvic lymph nodes and paraaortic lymph nodes. The cervix also presented with metastatic adenocarcinoma, about 18 mm in depth and 7 mm in width. Interestingly, according to the histochemical studies, endocervical mucinous adenocarcinoma was identified on the cervix (Figure 3), which

Figure 1. Computed tomography shows a large pelvic tumor with cystic and solid parts.

Figure 2. (A) Ovarian endometrioid adenocarcinoma: complex, branching glands with focal tumor necrosis; glandular lesion showing irregular budding but smooth surface; tall columnar cells with central nuclei and nuclear stratification (hematoxylin & eosin, 100×). (B) Tumor cell nonreactive to carcinoembryonic antigen (CEA).
was reactive for CEA and nonreactive for vimentin. The lesion was < 3 mm in depth. The tumor on the ovary was negative for CEA and vimentin. The cytology of the ascites was negative for malignancy. The final surgical-pathologic stage of ovarian endometrioid adenocarcinoma was stage IIc and of endocervical mucinous adenocarcinoma was stage IA1.

After surgery, adjuvant chemotherapy with carboplatin (area under the curve, 5) and paclitaxel (175 mg/m²) was prescribed monthly from January 14, 2005. Although CA-125 serum level declined to within the normal range, multiple nodules in both lungs, prevascular and pretracheal spaces were present on CT 2 months later. Gemcitabine 800 mg/m² was added. However, tiny nodules were apparent on chest X-ray, when compared to previous X-rays, and the CA-125 level was slightly elevated from 9.06 U/mL to 43.7 U/mL. After finishing six courses of adjuvant chemotherapy, 18F-2-fluoro-2-deoxy-D-glucose positron emission tomography (FDG-PET) was arranged and disclosed increasing FDG uptake in both lungs, liver, paraaortic region and pelvis, compatible with multiple metastases. Salvage chemotherapy with pegylated doxorubicin HCl liposome was continued but progressive dyspnea and cachexia developed. She was transferred to a hospice ward on August 8, 2005 and died of respiratory failure on September 1, 2005.

Discussion

In 1961, Moertel et al described 1,909 patients who were found to have multiple primary malignant neoplasms out of 37,580 patients with malignant neoplasms in a 10-year period, which represented an overall occurrence rate of 4.6% [3]. In 1984, the occurrence rate of synchronous gynecologic malignancies in patients with gynecologic tumors was 1.78%, and three cases of synchronous invasive cervical cancer and ovarian tumor were found among 2,362 patients by Axelrod et al in the Downstate Medical Center Tumor Registry [4]. In 1989, Eisner et al reviewed the histopathology of 3,863 patients with female genital malignancies in the UCLA Tumor Registry over a 30-year period [5]. Twenty-six patients (0.7%) with invasive synchronous primary cancers were identified. The most frequent synchronous genital lesions were ovarian and endometrial cancer in 11 patients (0.3%), with only a single case of synchronous ovarian and cervical tumors (0.025%) [5]. In 1983, LiVolsi reported four cases of endocervical adenocarcinoma coexisting with ovarian mucinous adenocarcinoma [6]. In 2004, two cases of synchronous ovarian and cervical cancer were reported out of 861 women with gynecologic tumors in Taiwan [7]. One of the cervical cancers was squamous cell carcinoma and the other was adenosquamous carcinoma. The ovarian cancers were both serous cystadenocarcinoma [7].

Our patient presented with rare synchronous primary gynecologic tumors of ovarian endometrioid and endocervical mucinous adenocarcinomas. Distinction between endometrial and endocervical adenocarcinomas may be made by using a small panel of antibodies, including CEA, estrogen receptor and vimentin [8]. Cells containing CEA are detected in approximately 80% of endocervical adenocarcinoma and < 10% of endometrial carcinomas [9]. The cervical tumor in this case had diffuse positivity to CEA and the ovarian tumor cells were nonreactive to CEA (Figures 2B and 3B). Recently, human papillomavirus (HPV) DNA was assessed to determine whether ovarian neoplasms were
metastatic or independent. The presence of HPV DNA in ovarian tumors confirms that they are metastatic endocervical adenocarcinomas [10].

Ovarian endometrioid carcinoma was reported to have the highest incidence of multiple primary tumors (21.3%) in 413 patients with primary ovarian tumors in Silverman et al's study [11]. Embryologic, hormonal, genetic or other phenomena may be associated with the development of malignancies arising simultaneously in genital tissues [11–13]. Ovarian and endometrial cancers are the most common synchronous primary tumors. Generally, synchronous double primary cancers have a more favorable prognosis than a tumor of a single metastatic lesion [7]. Patients with low stage and low grade synchronous ovarian and endometrial cancers have excellent prognoses. Detection at a relatively early stage suggests diagnosis may be facilitated by early symptoms from the endometrial carcinoma [5]. Our patient had an advanced stage of ovarian cancer when diagnosed. In this case, the reaction or nonreaction of immunohistochemical stain to CEA on the tissue slides determined the ovarian tumor cells from the endocervical ones. The treatment plan was mainly based on the protocol for advanced ovarian cancer since hysterectomy is adequate treatment for cervical lesions.

References


