Gynecologic Oncology Reports 2 (2012) 65–66

Contents lists available at SciVerse ScienceDirect

Gynecologic Oncology Reports

journal homepage: www.elsevier.com/locate/gynor

Case Report

Endodermal sinus tumor of the ovary in an 86 year old woman

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ARTICLE INFO

Article history:
Received 19 January 2012
Accepted 20 February 2012
Available online 27 February 2012

Keywords:
Endodermal sinus tumor
Germ cell malignancy
BEP chemotherapy

Introduction

Endodermal sinus tumor (Yolk sac tumor) is the second most common germ cell malignancy of the ovary accounting for 20–25% of malignant germ cell tumors. These are rare tumors noting that ovarian germ cell malignancies comprise only 2–3% of ovarian cancers. The most common presenting symptoms for patients with endodermal sinus tumors are a rapidly enlarging mass and pain. The median age of presentation in one series is 19 and the oldest reported patient is 75 years old (Bailey and Church, 2005). We report the oldest known patient with a pure endodermal sinus tumor.

Case report

An 86 year old gravida 4, para 4 woman with a history of lymphoma presented to her local oncologist with a 3 month history of abdominal pain and increasing vaginal prolapse. A computed tomography (CT) scan demonstrated a 5 cm pelvic mass. The patient underwent a laparoscopic biopsy of the mass at her local hospital which demonstrated a pathologic diagnosis of endodermal sinus tumor of the ovary. She was referred to our institution for gynecologic oncology consultation and management. She reported increasing pelvic pain, pessary use for vaginal prolapse, and urinary hesitancy. Her past medical history was significant for a hysterectomy. She had no other medical co-morbidities. Her family history was negative for cancer. At gynecologic oncology referral she was found to have a rapidly enlarging 8 cm pelvic mass, markedly larger than noted at time of laparoscopic ovarian biopsy. Repeat radiological imaging revealed a growing mass in the pelvis with intra-abdominal metastases. The patient underwent an exploratory laparotomy, bilateral salpingo-oophorectomy, omentectomy, pelvic peritoneal stripping and left retroperitoneal tumor resection, completing an optimal cytoreductive procedure. Surgical findings included a mass arising from the left ovary fixed to the pelvic sidewall, sub-centimeter tumor nodules on the colon and small intestine mesentery, and tumor involving the omentum, representing stage IIIC disease. Alpha-fetoprotein (AFP) level at diagnosis was noted to be 7010 ng/mL. Pathology confirmed pure endodermal sinus tumor of the ovary consistent with the previous biopsies. After an uneventful post-operative recovery the patient was treated with 4 cycles of bleomycin, etoposide and cisplatin (BEP) chemotherapy. Complications of chemotherapy included 1 episode of febrile neutropenia, grade 4 thrombocytopenia, grade 4 anemia, and grade 4 hypomagnesemia and hypokalemia. All toxicity was managed effectively with supportive care. At the completion of BEP therapy, a CT scan showed no residual tumor and the AFP level was 7.1 ng/mL.

Pathology

Pathology from the initial right ovarian biopsy showed a high-grade malignant neoplasm with complex pattern of anastomosing channels and spaces, papillary structures and scattered Schiller–Duval bodies. The tumor cells demonstrated immunoreactivity for sal-like protein 4, alpha-fetoprotein and glycipan-3 and absent immunoreactivity for cytokeratin 7, estrogen receptor and octamer-binding transcription factor 4. The histology and immunophenotype were diagnostic of pure yolk sac tumor (endodermal sinus tumor). Pathology from the second surgical procedure showed similar histology, pure yolk sac tumor, with involvement of right ovary, left ovary, colon epiploic tissue, omentum and small bowel.

Discussion

There have been five other documented cases of pure endodermal sinus tumors in women over 50 years of age, two of which were in their sixth decade, two in their seventh decade and one 75 year-old woman (Brown and Green, 1976; Ferracini et al., 1979; Kinoshita, 1990; Oh et al., 2001; Pliskow, 1993). Four of these women had pure endodermal sinus tumors of the ovary, and one was extra-ovarian in origin (Oh et al., 2001). There have been limited case reports of post-menopausal women diagnosed with pure endodermal

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2211-338X/$ – see front matter
doi:10.1016/j.gynor.2012.02.003

sinus tumor of the ovary. Rare cases of mixed endodermal sinus tumor within endometriosis, endometrioid carcinoma, or mucinous cystadenoma of the ovary have been reported (Lopez et al., 2003). These cases challenge the theory that pure endodermal sinus tumors originate only from germ cells, and present the possibility that they may originate from somatic mesodermal cells, a sort of retrodifferentiation or neometaplastic process. This is supported by the finding of positive AFP staining and elevated serum AFP levels in cases of carcinomas of somatic origin (Talerman and Vang, 2011).

Cases of pure or mixed endodermal sinus tumors of the ovary occurring in post-menopausal women are rare. There is limited data regarding the natural course of the disease, prognostic factors, or tumor response to therapy. In one retrospective analysis of premenopausal women diagnosed with endodermal sinus tumors, age was not a significant prognostic factor, nor was the presence or absence of additional histological elements or lymph node dissection (Nawa et al., 2001). Before the introduction of effective chemotherapy, the prognosis for patients with malignant germ cell (non-dysgerminoma) tumors was poor, with a 3 year survival rate of 13% for malignant endodermal sinus tumors. The establishment of BEP chemotherapy for malignant germ cell tumors has significantly improved outcomes. A Gynecologic Oncology Group (GOG) study treated 93 patients with malignant germ cell tumors with BEP chemotherapy. Ninety-one patients were disease-free at a median of 38 months. None of the 25 patients with endodermal sinus tumors developed recurrence (Williams et al., 1994). Additional series noted a 76% 5-year disease-free survival for non-dysgerminoma stage III and IV germ cell tumors treated with BEP (Bailey and Church, 2005). Ovarian endometrioid carcinomas with endodermal sinus tumor components have typically not responded to cisplatin-based chemotherapy regimens. These mixed tumors represent an aggressive variant of ovarian malignancy associated with a rapid growth pattern and high stage at diagnosis. The chemosensitivity of the component of the endodermal sinus tumor patterns has tended to determine pathological recurrence, and a majority of cases of ovarian endodermal sinus tumor associated with endometrioid or mucinous neoplasms in postmenopausal women have resulted in death secondary to disease (Nawa et al., 2001). Of the documented cases of pure endodermal sinus tumors in postmenopausal women, final outcomes were not consistently reported. Patients often experienced a laboratory response but subsequently died of disease several months after initial diagnosis. To our knowledge, this is the oldest case of an ovarian endodermal sinus tumor reported in the literature demonstrating a complete serologic and clinical response to BEP chemotherapy following surgical debulking.

Conflict of interest statement

No conflict of interest.

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