Squamous Cell Carcinoma Arising in an Ovarian Mature Cystic Teratoma: A Case Report

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Malignant transformation in a mature cystic teratoma of the ovary is rare. The most common malignancy is squamous cell carcinoma, which consists of about 75% of malignant transformations. In the present report, we describe a case of advanced-stage squamous cell carcinoma arising in a mature cystic teratoma.

A postmenopausal 63-year-old woman with squamous cell carcinoma arising in a mature cystic teratoma is presented. The initial investigation by ultrasound showed a left adnexal mass with mixed echo pattern, which aroused the suspension of malignancy. She underwent a laparotomy and left oophorectomy. Histopathology was compatible with squamous cell carcinoma arising in a mature cystic teratoma. After a few episodes of intestinal obstruction and colostomy, she underwent partial resection of the ileum and sigmoid colon four months after the initial oophorectomy. Histopathologic study showed metastatic poorly-differentiated squamous cell carcinoma. Subsequently, she underwent two courses of combination chemotherapy with cisplatin, leucovorin, and 5-fluorouracil with no response. She died from progression of the disease nine months after the initial operation.

Keywords: Malignant transformation • mature cystic teratoma • squamous cell carcinoma • ovary

Introduction

Mature cystic teratoma (MCT), composed of well-differentiated tissues derived from the three germ cell layers (ectoderm, mesoderm, and endoderm), is the most common tumor of the ovary and accounts for 10 – 20% of all ovarian tumors in women of reproductive age. Malignant transformation in an MCT of the ovary is rare, with an incidence rate of less than 3%. The most common malignancy is squamous cell carcinoma (SCC), which represents about 75% of malignant transformations, followed by adenocarcinoma and melanoma. In the present report, we describe a case of SCC arising in an MCT in an advanced stage.

Case Report

A postmenopausal 63-year-old woman presented with a two-week history of progressing abdominal pain particularly following eating with subsequent nausea, vomiting, and anorexia in recent months. In February 2002, she was admitted to Imam Khomeini Medical Center (IKMC), Tehran for further investigation. The abdominal pain preceded her recent chief complaints for one year without seeking any medical attention.

Ultrasound examination showed a well-outlined encapsulated mass in the left adnexa with a mixed
A cystic mass measuring 10 cm in diameter with smooth and intact external capsular surface. The mass contained hair, tooth, and a dense brownish fluid. The cyst wall thickness was from 0.2 up to 0.5 cm in most areas, but regions measuring 1.5 to 3.5 cm in thickness were also seen. Histopathologic examination showed features of MCT in which the dysplastic bronchial mucosa had undergone malignant transformation resulting in invasive keratinizing SCC of pulmonary type. Peritoneal fluid was negative for malignancy.

She refused any adjuvant therapeutic intervention and was discharged. After four months, the patient returned to our center complaining of constipation and abdominal pain. Further investigation revealed an intestinal obstruction. After a few episodes of intestinal obstruction and colostomy, she underwent laparotomy, and then partial resection of the ileum and sigmoid colon were performed four months after the initial oophorectomy. The pathology showed metastatic poorly-differentiated SCC, but astonishingly the peritoneal fluid cytologic study was negative for malignancy. She was staged IV in the International Federation of Gynecology and Obstetrics (FIGO) classification.

Subsequently, she underwent two courses of combination chemotherapy with cisplatin, leucovorin, and 5-fluorouracil in a four-week interval, but she resisted subsequent courses of treatment because of very poor general status and died from progression of the disease in December 2002, nine months after the initial operation.

**Discussion**

SCC arising from an MCT is a rare pathologic event and in most instances not diagnosed preoperatively. There are no particular signs or symptoms which are characteristic of malignancy arising in a dermoid cyst. The patient in this report suffered from abdominal pain for one year but she was neglected. She sought medical advice only after worsening of the pain and having extra complaints such as nausea and vomiting. The common symptom is abdominal pain followed by abdominal or pelvic mass, but the patients may be asymptomatic or have symptoms of abdominal distention or bloated abdomen, as those caused by benign cysts. In some other cases, various symptoms due to invasion of nearby organs are the presenting complaints, such as gastrointestinal symptoms of constipation or diarrhea, rectal bleeding, or urinary frequency. Other nonspecific signs of wasting disease such as weight loss or cachexia may be found in advanced cases.

Preoperative diagnosis of an MCT of the ovary is relatively easy due to the radiologic detection of bony tissues including teeth, bones, and cartilages. However, preoperative diagnosis of malignant transformation is very difficult clinically, because this tumor cannot be readily differentiated from an uncomplicated MCT or other ovarian tumors. Since MCT is a common ovarian neoplasm and is increasingly diagnosed as an incidental finding in patients, there has been growing emphasis on preoperative risk assessment of these tumors in order to optimize surgical management. There are evidences for some factors as risk factors for malignancy in MCT including age, tumor size, and serum tumor markers.

SCC arising in an MCT has historically been observed in relatively older patients particularly after menopause; although, it has been sometimes reported in young patients around 30 years or even younger. The patient's age in our case was 63 years, which is consistent with the usual age range of this disease. A couple of studies demonstrated a role for patient’s age in differential diagnosis as it is prudent to maintain a higher suspicion of malignancy in MCTs occurring in patients over the age of 45. Accordingly, it is considered helpful for prevention and early detection of the malignant transformation to have regular ovary examination through pelvic ultrasonogram in middle age.

Tumor size has also been noted to predict malignancy. Although MCT presents in a wide range of sizes, larger tumors correlate with an increased risk of malignant transformation. In our case, the tumor diameter was around 10 cm, which is larger than a typical benign cyst. Kikkawa et al. reported that a tumor diameter of larger than 9.9 cm was 86% sensitive for malignancy in their series. In general, it is recommended that a diameter equal or greater than 10 cm or a tumor...
demonstrating rapid growth should prompt suspicion.12

The prognosis for these tumors has been often reported to be very poor with a five-year survival of only 15 – 30%.1 Prognostic indicators of survival have been attempted to be identified in various studies. Most of them agree that higher FIGO stages would carry a worse prognosis.8,9,13,14

The other potential predictors reported include rupture or spillage,15 tumor grade, vascular involvement, and the mode of tumor infiltration.9,14 In addition, better prognosis has been reported when the malignant element is an SCC compared with adenocarcinoma or sarcoma.1 In Peterson's study reviewing a total of 190 cases, metastases were noted in 64% of the patients and large intestine (23%) was most frequently involved.16 In a recently published case series of 11 cases, metastasis was noted in only 18% of the patients (2 cases), and the sites were rectum and urinary bladder.7 The patients with metastases, such as the case in the current report, have very poor prognosis.16

The main therapeutic approach to an ovarian MCT with malignant transformation has been surgical.8,16 Conservative unilateral oophorectomy without further postoperative treatment may be justified for early stage IA disease, especially for nulliparous and young patients who desire future fertility; however, in the postmenopausal women, total removal of the genital organs would seem to be the procedure of choice.8 Postoperative treatments in the literature included single-agent or combination chemotherapy, radiotherapy, or a combination of these modalities. Results of these treatment regimens were variable and have not been systemically evaluated in an adequate number. Therefore, the optimal adjuvant therapy for SCC arising from an MCT has not been yet established.4,12,16

In conclusion, clinicians should keep this rare type of tumor in mind when faced with a dermoid cyst, especially in older patients or in larger than usual cysts. Lastly, although many authors have documented the use of a combination of chemotherapy and/or radiation, one must point out the different outcomes obtained. Therefore, the optimal treatment of this cancer should be individualized based upon clinical findings of the patients and experience of the care providers.

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