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Case Report

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

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ABSTRACT

Mature cystic teratoma of the ovary (MCTO) is the most common ovarian germ cell tumor. Its malignant transformation is a rare complication that occurs in almost 2% of the MCTs. MCTO is benign and usually appears between 30 and 40 years of age, but patients with malignant ovarian MCT are 10–15 years older than those with benign MCT. The most common malignant transformation in MCTO is Squamous Cell Carcinoma (SCC) which is rarely diagnosed with pre-operative imaging.

We report the case of a postmenopausal woman, presenting with severe abdominal pain and a large palpable mass in her abdomen. She was diagnosed postoperatively with SCC arising from MCTO which was confirmed histopathologically. The patient received postoperative chemotherapy and was well at 6-month follow-up after chemotherapy.

MCTO is benign, but can rarely become malignant in older ages. So MCTO-arising SCC should be considered in elderly women with abdominal pain and mass, and also some other evident features such as large tumor diameter, elevated serum markers, and solid components in Magnetic resonance imaging (MRI).

Introduction

Mature cystic teratoma (also known as dermoid cyst) of the ovary (MCTO) comprises 20-25% of all ovarian tumors and is generally the vast majority of germ cell neoplasm of the ovary [1, 2]. MCTO is benign, however, 0.17- 2% of MCTO may transform to malignancy. Among different histological forms of malignant transformation in MCTO, SCC transformation is the most common (80%) [3]. Age of the patient, tumor size, ultrasound findings, computed tomography (CT) features and rates of SCC and CA125 tumor markers are some risk agents and the clinical manifestations are correlated with MCT transformation into SCC [4].

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This article presents the case of a 65-year-old woman with SCC arising from dermoid cyst.

Case presentation

A 65-year-old multiparous woman presented to surgery department with severe abdominal pain and a mass detected in the left lower quadrant (LLQ) of the abdomen. She had abdominal pain from six months before that exacerbated in the last three days. The pain was consistent and non- positional with no radiation. She had loss of appetite, nausea, and vomiting, but had no history of fever or weight loss. Her bowel and bladder habits were unaltered.

In her past medical history, she was postmenopausal since the age of 50 years old and was receiving medication for hypertension. She reported a family history of breast cancer in her mother.

On physical examination, a significant abdominal distension and a huge palpable mass, approximately 25*15cm were found in LLQ and hypogastric area that was firm and immobile with tenderness in palpation.

Initial laboratory tests showed normal results. Ultrasound revealed a cortical simple cyst of 25mm in upper pole of the right kidney. A huge solid-cystic structure of 175*165mm was seen that extended from left side of the abdomen to the liver and right adnexa. Color Doppler ultrasound, showed no obvious vascularity in solid component, but limited free fluid was detected in pelvis.

CT scan of the abdomen and pelvis revealed a massive, heterogeneous mass with fat density in some areas and coarse classification originated from left abdomen and left adnexa extended to right lower quadrant (RLQ), right adnexa and epigastric area. An aseptate cyst (26mm) was seen in upper pole of the right kidney (bosniac1) and a similar cyst (10mm) was seen in lower pole of the left kidney (bosniac1).

Our differential diagnoses of the mass with probable adnexal origin included ovarian cancer, fallopian cancer, metastatic carcinoma or benign ovarian neoplasms (such as endometrioma or mature cystic teratoma).

To approach the mass, laparotomy was planned by an oncosurgeon. Exploratory laparotomy, left unilateral salpingo-oophorectomy, and resection of the mass (Figure 1), exploring the left ureter and ligation of gonadal artery were done. Intraoperative sample for frozen section wasn't sent.

Histopathology report showed left ovarian mass, and left salpingo-oophorectomy which was grossly, consisting of a green-brownish deformed cystic tissue (22*16*15cm) with a firm bulging area (8*7cm) and a cream-whitish discolored area (7*6cm) on the surface. On cuts of bulging area, keratinous material was seen. On opening of the cyst, hair tufts and cream-whitish sebaceous substance were observed. Furthermore, attached fallopian tube measuring 8*2cm was seen.

In microscopic examination, sections from left ovarian mass revealed a malignant epithelial neoplasm composed of sheets and nests of atypical squamid cells with enlarged hyperchromatic to vesicular and pleomorphic nuclei, visible nuclei, abundant cytoplasm, and keratin pearls. Moreover, frequent

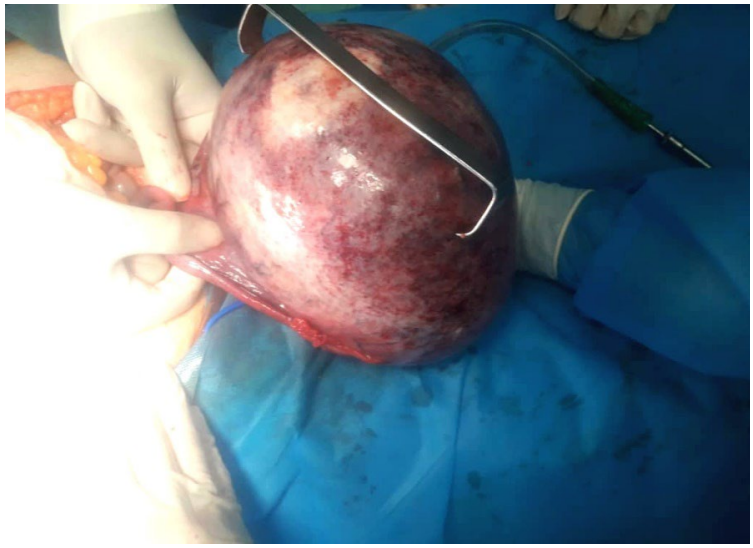

Fig. 1. operative finding, this large firm mass was resected.

mitotic figures and lymphatic invasion were present. Ovarian capsular invasion, vascular and perineural invasion were not identified. Adjacent tissue showed cystic structures lined by stratified squamous epithelium filled by keratinous material and hair shaft. Histopathological diagnosis was ovarian SCC, arising from dermoid cyst (somatic type), and a low-grade tumor (G1), which was well differentiated with lymphatic invasion but no blood vascular invasion. The fallopian tube was free of any invasion.

Immunohistochemical (IHC) staining of epithelial markers showed that the malignant cells were positive for P63, weakly positive for CK5/6, partially positive for CK 7, and about15% positive for Ki65. The cells were found to be negative for CK20 and PAX8. Final data were compatible with SCC.

After confirming the diagnosis of SCC, the patient did not consent to radical surgery and so the adjuvant chemotherapy was performed with a carboplatin regimen. She received 2 cycles of chemotherapy with Carboplatin regimen. At a 6-month follow-up after chemotherapy, she had no problem, and laboratory and imaging tests were normal.

Discussion

Mature cystic teratoma of the ovary (MCTO) is the most common germ cell neoplasm of the ovary and comprises almost 20-25% of all ovarian tumors [1, 2]. The incidence of MCTO is about 1.2–14.2 cases per 100.000 people per year. It can occur at any age, but the highest peak is between 30 and 40 years, whereas patients with malignant ovarian MCT are 10–15 years older than those with benign MCT. Malignant transformation occurs in almost 2% of the MCTs (0.8%–5.5%) and commonly consists of SCC [5].

SCC arising from MCTO is very rare, and often not diagnosed preoperatively on imaging workups [6]. The most common symptoms of presentation are abdominal pain (54%) and abdominal mass (31%), followed by constipation, menorrhagia, urinary frequency, weight loss, and fever [7].

Older age, large tumor diameter, elevated serum markers, and presence of solid components in MRI provide important clues to the suspicion of malignant transformation in MCT preoperatively. But the final diagnosis of malignant transformation in MCTO is based on postoperative histopathological findings [8].

Management of MCTO-arising SCC includes bilateral salpingooophorectomy, total hysterectomy and comprehensive surgical staging (peritoneal washing, omentectomy, appendectomy, peritoneal biopsies, and pelvic plus paraaortic lymphadenectomy) in early stages and optimal cytoreductive surgery, adjuvant therapy with platinum-based or paclitaxel-based chemotherapy, and concurrent whole pelvic radiation in advanced disease. Fertility sparing surgery can be done in young women with early-stage of the disease [2, 5]. Bleomycin/etoposide/cisplatin (BEP) and paclitaxel/carboplatin (TC) are the recommended first-line chemotherapy regimen for ovarian germ cell neoplasm and epithelial ovarian cancer. Chemotherapy can make better prognosis of patients with MCTO-arising SCC at advanced stage, nevertheless, today there is no first-line adjuvant therapy for this condition. According to some evidence, the mean age of patients with SCC arising from MCTO is 53.5 years old and the age ≥ 45 years is correlated with worse prognosis. The mean tumor size is reported to be 14.8cm and generally there is no difference in survival of tumors ≤10 cm and >10 cm (3).

Herein, our case was a 65-year-old woman whose age was older than the mean age for ovarian SCC. She was presented with abdominal pain and mass, and on physical examination there was a firm, immobile large (about 25*15cm) palpable mass in lower abdomen with tenderness which was considerably greater than the mean tumor size previously reported. The patient had mechanical prosthetic valve in the mitral and MRI was contraindicated in the patient. Thus, CT scan of the abdomen and pelvis was done which showed a massive, heterogeneous mass with fat density in some areas originated from left adnexa.

The patient age, size and characteristics of the mass on physical examination predicted malignant transformation. Therefore, exploratory laparotomy, unilateral oophorectomy, and resection of the mass were performed. Histopathological diagnosis was SCC, arising from dermoid cyst with well differentiated tumor (G1). IHC results, also, confirmed the diagnosis of SCC.

The patient received two cycles of chemotherapy with Carboplatin according to individualized and integrated treatment of platinum-based chemotherapy for adjuvant therapy in cases of MCTO-arising SCC. During a 6month follow-up after chemotherapy, the patient was well, the lab data and CT-scan were normal, and CA125 was 9.

Similar to our case, Balik et al. reported a 66-year-old woman with MCTO-arising SCC presented with abdominopelvic mass and abdominal pain that a

cystic mass of 20*18 cm with solid components and calcifications was seen extending from left adnexal region, towards the umbilical area. The patient underwent total hysterectomy and bilateral salpingo-oophorectomy, omentectomy, appendectomy, with bilateral pelvic, and paraaortic lymph node dissection. Contrary to histopathology results of our case that patient was found with malignant invasion of the uterus and appendix. The patient refused continuation of treatment after a single dose of cisplatin and died five months later [2].

Conclusion

We highly recommend suspicion to SCC arising from MCTO when an elderly woman is presented with abdominal pain and mass and also some other clues, such as large tumor diameter, elevated serum markers, and reports of solid components in MRI. So, clinicians should be aware that although MCTO is benign, itcan become malignant in older ages and is rarely diagnosed preoperatively on imaging tests.

List of abbreviations

MCTO: Mature cystic teratoma of the ovary

SCC: Squamous cell carcinoma

IHC: Immunohistochemistry

Ethical Considerations

Compliance with ethical guidelines

There were no ethical considerations to be considered in this article.

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Conflict of Interests

The authors have no conflict of interest to declare.

Authors' contributions

EZ analyzed and interpreted the patient data regarding the oncologic disease and chemotherapy, and also contributed in writing the manuscript. SH was a major contributor in writing the manuscript. SMG performed the surgery and interpreted surgical data. RA substantively revised the work. All authors read and approved the final manuscript.

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