Case Report

Primary ovarian squamous cell carcinoma occurring in mature cystic teratoma

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ABSTRACT

Mature cystic teratoma (MCT) is a common benign tumor of the ovary in females. Malignant transformation in a MCT of the ovary is a rare condition and accounts for only 1%–2% cases. The most common malignancy is squamous cell carcinoma (SCC), which comprises about 75% of malignant transformations. We report a case of a 52-year-old postmenopausal female who presented with complaint of pain and mass per abdomen for 4 months. She underwent bilateral salpingo-oophorectomy, and histopathological examination revealed primary ovarian SCC arising in a MCT. Later, she underwent chemotherapy, total hysterectomy, and omentectomy with iliac lymph node dissection.

Key words: Ovary, squamous cell carcinoma, teratoma

INTRODUCTION

Mature cystic teratoma (MCT) is the most common germ cell tumor of the ovary in adolescents and reproductive age group and accounts for about 30%–45% of all ovarian neoplasms. It comprises well-differentiated tissues derived from the three germ layers (ectoderm, mesoderm, and endoderm).[1] MCT is generally a benign tumor. However, malignant transformation is extremely rare and occurs only in 1%–2% cases.[2] The most common malignant transformation is squamous cell carcinoma (SCC) which accounts for 70%–80% of cases followed by adenocarcinoma and carcinoid tumors.[3] We reported a case of primary ovarian SCC arising in MCT at our institute with review of literature.

CASE REPORT

A 50-two-year-old postmenopausal, nulliparous woman presented to gynecology department with complaints of pain and mass per abdomen for 4 months. There was no history of weight loss, loss of appetite, and significant bowel- and bladder-related symptoms.

On per abdominal examination, the abdomen was uniformly distended, and a 20-week size mass with regular margins and smooth surface was palpable. Per rectal examination revealed fullness in the pouch of Douglas and a mass of 10 cm × 5 cm was palpable. Lymph nodes were not palpable, and a diagnosis of ovarian malignancy was made clinically.

Complete hemogram and biochemical investigations were within normal limits. Ultrasonography of the abdomen revealed left adnexal mass suggestive of dermoid cyst. Computed tomography scan of the abdomen showed a well-defined hypodense lesion measuring 11.3 cm × 9.5 cm × 4 cm with cystic, fatty, and solid enhancing components in the pouch of Douglas compressing the uterus. CA 125 tumor marker level was 32 U/ml which is toward the upper limit of normal, whereas carcinoembryonic antigen (CEA),

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alpha-fetoprotein, and lactic dehydrogenase were within normal limits.

The patient underwent laparotomy which revealed a voluminous abdominopelvic mass. Excision of mass was done with bilateral salpingo-oophorectomy and sent for histopathological examination. On gross examination, a partially cut open gray-white cystic mass with attached tube, measuring 10 cm × 9 cm × 3 cm, was received. External surface showed congested vessels. Cut section of the specimen showed cystic and solid gray-white area. Cystic part showed tufts of hair with pultaceous material and solid gray-white area measured 5 cm × 3.5 cm [Figure 1]. Microscopy revealed cyst wall lining of keratinized stratified squamous epithelium along with glandular epithelium, cartilage, neural tissue, hair follicles, and abortive glomerulus [Figure 2]. Sections from solid gray-white area showed malignant squamous cells in nests and cords with high nucleocytoplasmic ratio, round vesicular nucleus, and prominent nucleoli [Figure 3]. Many atypical mitotic figures (3–4/10 hpf) along with tumor giant cells were seen [Figure 4]. Lymphatic invasion by tumor cells was present. No keratin pearls were noted. A histopathological diagnosis of primary ovarian SCC arising in MCT was made. Immunostaining with CK5/6 was done which confirmed squamous differentiation [Figure 5]. Later, the patient was given six cycles of chemotherapy following which CA 125 was reduced to 8.8 U/ml. Then, the patient underwent total hysterectomy, omentectomy with iliac lymph node dissection. The patient was stable at the time of discharge.

Figure 1: Gross specimen of ovarian mass showing cystic area with tufts of hair and pultaceous material and solid areas

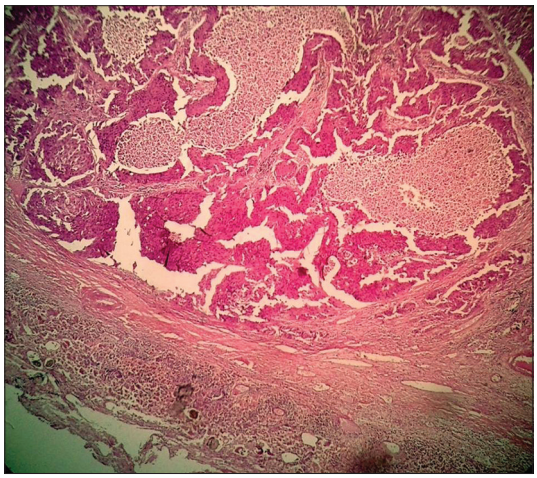

Figure 3: Invasive squamous cell carcinoma arising from cyst wall with areas of necrosis (×40)

DISCUSSION

Germ cell tumor of the ovary accounts for around 20%–25% cases of ovarian neoplasms. MCT is the most common germ cell tumor of the ovary occurring in perimenopausal age group.[4] The malignant transformation in these tumors is extremely rare and usually occurs in postmenopausal women. Preoperative diagnosis is difficult as there are no distinctive clinical and radiological features which are characteristic of malignancy arising in MCT. Tumor markers are also often within normal limits.

SCC arising in MCT has historically been observed in relatively postmenopausal patients with the median age

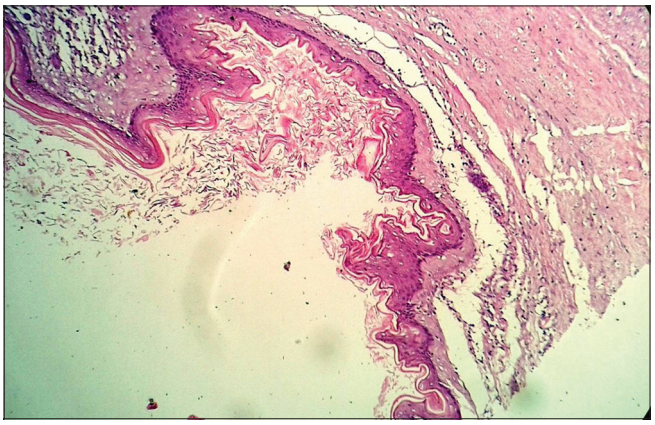

Figure 2: Cyst wall lining of keratinized stratified squamous epithelium (×100)

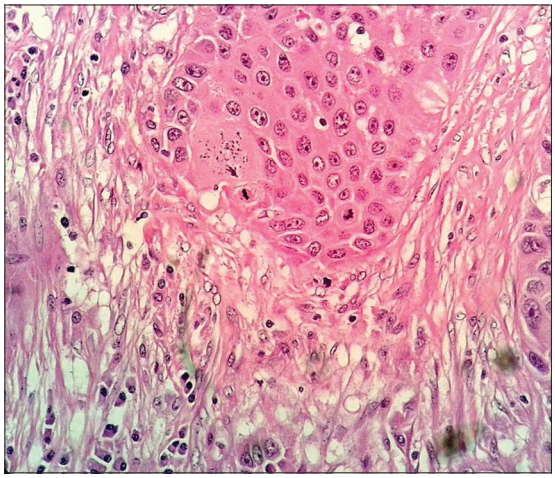

Figure 4: High-power view showing mitotic figures (×400)

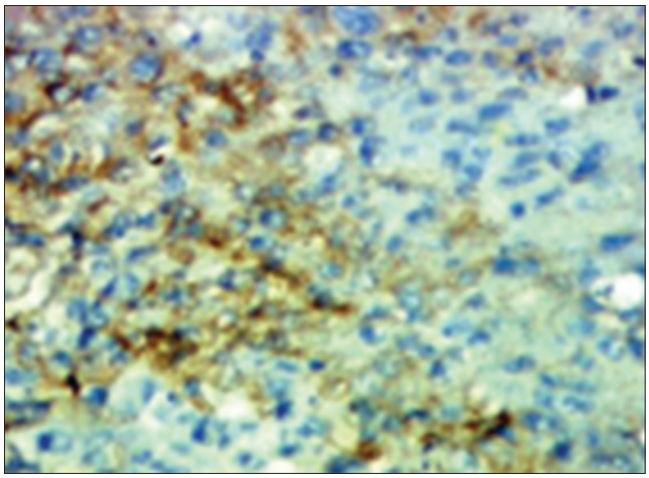

Figure 5: CK5/6 positivity

of diagnosis between 45 and 60 years. In our case also the patient's age is 52 years which is consistent with the usual reported age range of this disease. As reported by Tangjitgamol *et al*., a high suspicion of malignancy should be raised in MCT cases occurring in patients above 45 years of age.[5]

Spannuth *et al*. reported that the most common presenting symptom is pain and mass per abdomen. In advanced cases, weight loss and cachexia may be present. In our case also the patient presented with pain and mass per abdomen.[6]

Tumor size should be considered to predict malignancy. Although MCT can present with wide range of sizes, larger size, especially in postmenopausal women, correlates with high risk of malignancy. Kikkawa *et al*. and Yamanaka *et al*. established that larger tumor with diameter 9.9 cm may be related to increasing risk of malignant transformation.[7,8] In our case, the tumor size was 11.3 cm which is highly in favor of malignancy.

According to study done by Chiang *et al*., higher levels of serum CA 125 were associated with the survival rates and also concluded that patients with raised levels of CA 125 are associated with advanced stages and have poor prognosis.[9] In our case, CA 125 was 32U/ml which is within the normal range. Hirakawa *et al*. quoted that CEA and CA 19-9 have been recently suggested as important investigations in the evaluation of MCT.[10]

The disease carries very poor prognosis, especially once it spreads beyond the ovary. The 5-year survival rate of only 15%–30% has been reported. The other predictors reported include tumor grade, rupture or spillage, and vascular invasion. In patients with metastases, the prognosis is poor. However, when compared with adenocarcinoma or sarcoma, the malignant transformation of SCC carries better prognosis.[11]

The main therapeutic approach to an ovarian MCT with malignant transformation is surgical. Conservative unilateral oophorectomy without further postoperative treatment can be done for early stage IA disease. Postoperative treatments in the literature included single-agent or combination chemotherapy, radiotherapy, or a combination of these modalities. In the present study, the patient was given six cycles of chemotherapy following the surgery. The patient was stable at the time of discharge and is in the road to recovery.

CONCLUSION

Keeping in view the rarity and poor prognosis of SCC of the ovary arising in MCT, the clinicians should keep this entity in mind when faced with MCT, especially in postmenopausal patients or in patients with larger than usual cysts.

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Conflicts of interest

There are no conflicts of interest.

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