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CASE REPORT

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Mature cystic teratoma of ovary with squamous cell carcinoma arising from it

Sushna Maharjan

Department of Pathology, Chitwan Medical College Teaching Hospital, Chitwan, Nepal

Correspondence
Sushna Maharjan, Department of Pathology, Chitwan Medical College Teaching Hospital, Chitwan, Nepal.
Email: sushnamaharjan74@gmail.com

Key Clinical Message

Clinicians, pathologists, and radiologists should be aware of rare malignant ovarian tumors arising in mature cystic teratoma (MCT). Suspicion should be raised if the patient is elderly, the tumor is huge, and the tumor has large solid foci. However, malignant transformations in MCTs in younger women have been reported.

KEYWORDS

elderly, huge tumor, large solid foci, malignant transformations, mature cystic teratoma

1 | INTRODUCTION

Mature cystic teratoma (MCT) is the germ cell tumor originating from primordial germ cells and histologically consists of tissues arising from endoderm, mesoderm, and ectoderm.[1-3] MCT accounts for 30%-40% of all ovarian neoplasms. It is the most common (60%) tumor among the benign counterparts. The observed frequency of malignant ovarian tumors associated with MCTs was 0.17% to 1.4%.[5,6] Squamous cell carcinoma (SCC) is the frequent malignancy arising from the ectodermal component of MCT followed by adenocarcinomas and carcinoid tumors.[7-10]

Squamous cell carcinoma arising from MCT is frequently observed in postmenopausal women.[7] Preoperative diagnosis of malignant transformation is challenging because the signs and symptoms are nonspecific, and neither serum tumor markers nor imaging techniques such as ultrasound or CT scan can predict the specific diagnosis.[11] Prognosis of the patient is determined by surgical stage but it is usually very poor.[12] The objective of this case study was to present the rare case of ovarian malignancy arising from MCT which was not clinically suspected and diagnosed by histopathologic analysis. It signifies that clinicians should raise a suspicion of malignancy if the patient is elderly, the tumor is huge, and the tumor has large solid foci. However, malignant transformations in MCTs in younger women have been reported.

2 | CASE PRESENTATION

A 43-year-old woman with gravida 2, para 2, had complaints of pain and abdominal discomfort for 2 months. There was no history of constitutional symptoms such as loss of appetite and loss of weight. Bowel/bladder habits were normal. There was no other significant clinical or family history. Abdominal examination revealed a palpable mass on the left lower quadrant. Pelvic cavity did not show any free fluid collection. Abdominal ultrasound revealed a mass of about 10×7 cm^2 in diameter with cystic and solid densities. Serum CA125 was 10.7 IU/mL. ßHCG, AFP, and LDH were other serum tumor markers tested. These were also normal. Mature cystic teratoma of left ovary was the provisional diagnosis.

The patient underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy. The specimen was sent for histopathologic examination (HPE). On gross examination, a left ovarian mass of 10×7 cm^2 in diameter was identified. The surface of ovarian mass was uneven. Cut section showed a unilocular cystic mass with 0.2-cm maximal wall thickness, filled with pultaceous material and hair shafts. The cyst wall contained a solid, gray white nodule measuring 5.5×4 cm^2 (Figure 1). Uterus showed an intramural fibroid of size 1.8×1.5 cm^2. Both fallopian tubes and right ovary were normal.

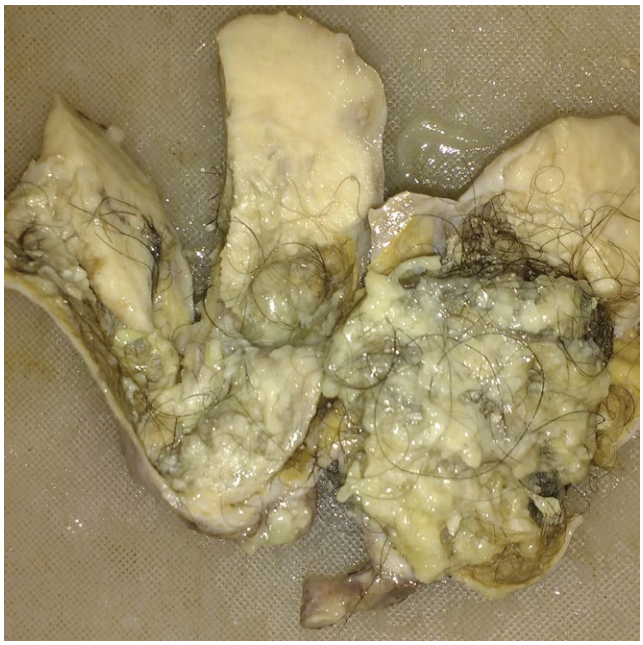

FIGURE 1 Gross photograph showing a cyst filled with pultaceous material and hair; and a gray white solid area

Microcopy revealed stratified squamous epithelial lining of the cyst with anucleated keratin debris and foreign body giant cell reaction. Sections from the solid nodule showed moderately differentiated squamous cells arising from the surface epithelium (Figures 2, 3, and 4). Ovarian surface was free of tumor. Lymphovascular and perineural invasion were not seen. Sections from right adnexa, left fallopian tube, and uterus showed no evidence of malignancy. Diagnosis was made as mature cystic teratoma of left ovary with squamous cell carcinoma arising from it (moderately differentiated). According to the International Federation of Gynecology and Obstetrics (FIGO), tumor stage was determined as stage IA.

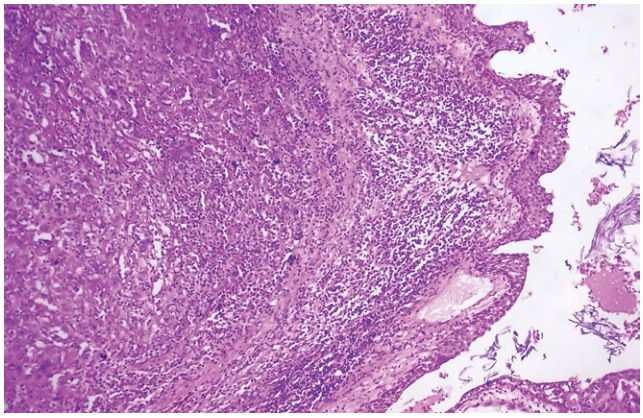

FIGURE 2 Microscopic photograph showing cyst lined by stratified squamous epithelium with sheets of malignant squamous cells in the underlying stroma (H&E stain, ×10). [H&E- hematoxylin and eosin]

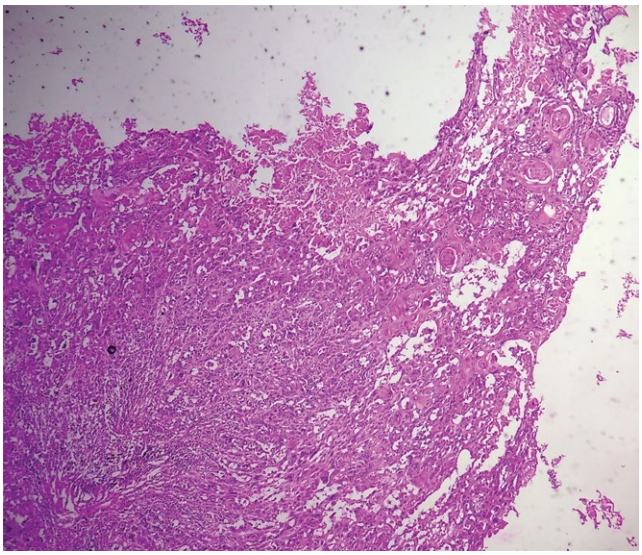

FIGURE 3 Microscopic photograph showing squamous cell carcinoma with sheets of malignant squamous cells and keratin pearls (H&E stain, ×10)

Intramural leiomyoma was also present. Adjuvant carboplatin-based chemotherapy was given to the patient. Her ultrasound, CT scan, and serum tumor markers were normal on follow-up after 6 months of treatment.

3 | DISCUSSION

The incidence of malignant ovarian squamous cell carcinoma arising in MCT is rare.[13] It is mostly observed in postmenopausal women with an average age of 58.2 years as compared to 37.5 years in MCT.[14] Some cases of malignant transformation of MCT have been reported in younger patients.[15-17] Preoperative detection of malignant

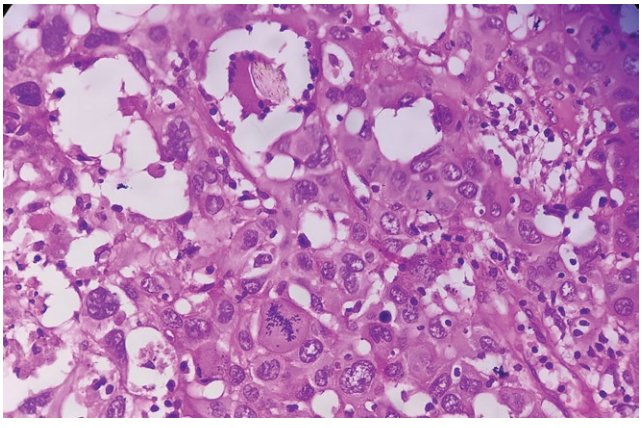

FIGURE 4 Microscopic photograph showing sheets of malignant squamous cells with pleomorphism and atypical mitotic figure (H&E stain, ×40)

transformation of MCTs is challenging due to nonspecific tumor markers and imaging findings.[18]The patients are usually asymptomatic; although symptoms like abdominal pain and abdomino-pelvic mass may be seen, similar to this case. It may be discovered accidentally during gynecologic examination due to mass effect.[19] Acute intestinal obstruction may occur when there is adherence of a part of small bowel to the mass.[20] Advance cases may develop constitutional symptoms such as cachexia and weight loss. A variety of symptoms may be present secondary to the invasion of nearby organs.[21]

Kikkawa et al[14] mentioned tumor size as the major factor for ruling out malignancy arising from MCT. They reported an average size of 152.3 mm in SCC arising in MCT in comparison to MCT with an average size of 48.2 mm; and the cutoff size between these was 99 mm. Maximal diameter of the mass was 10 cm (100 mm) in the present study. Development of malignancy in MCT is usually predicted by factors like old age, large tumor size, levels of serum tumor markers (SCC antigen, CA125, CA19-9 and CEA),[22] and larger solid component in MCT. If tumor spreads beyond the ovary, SCC arising from MCT has a poor prognosis.[23]

A 5-year survival rate is found in 95% of cases if tumors are limited to the ovary.[24] According to Chen et al,[8] 5-year survival rate is 48.4% among overall stages of disease, whereas individual stages had the 5-year survival rates of 75.7, 33.8, 20.6, and 0% for stages I, II, III, and IV, respectively. The appropriate treatment of choice for malignant ovarian tumors arising in MCT is complete surgical excision.[23] It is justifiable to perform unilateral oophorectomy if the patient is nulliparous or young woman who wants to preserve fertility, especially in stage IA disease; however, the choice of treatment in the postmenopausal women would be a total abdominal hysterectomy with bilateral oophorectomy.[25] Hackethal et al[7] found that malignant SCC arising in MCT in patients above 50 years old have a high concentration of CA 125 and tumor size of more than 10 cm. They also observed a better survival rate in stage Ia disease.

Literature review had shown various postoperative treatment modalities such as chemotherapy (single or combination), radiotherapy, or a combination of these two therapies. However, adequate outcome of these treatment modalities has not yet been established because of the rarity of this entity that has precluded patients from participating in large randomized trials.[21] Hackethal et al[7] stressed the role of chemotherapy based on alkylating agents. Overall survival of patients had improved to 57.1 months when alkylating agents were administered compared to 25.2 months for those who received non-alkylating regimens. A higher survival was associated with complete resection followed by adjuvant chemotherapy in an advanced disease but adjuvant radiotherapy did not improve survival of the patients in the same study. Cisplatin–ifosfamide–paclitaxel (TIP), a highly active chemotherapy regimen has proved to be efficient in SCC arising in MCT.[26] The combination of carboplatin–paclitaxel regimen in squamous cell carcinomas of MCT has also been reported.[27]

On review, quite limited cases of mature cystic teratoma of ovary with squamous cell carcinoma arising from it have published by Nepalese authors till now.[28-30] Bashyal et al[28] reported three cases of squamous cell carcinoma arising MCT; among them, one of the patients was a young patient aged 33 years. A malignant transformation from MCT was observed in seven cases (3%) in the other study conducted by Sherpa et al[29]; among these, five cases were SCC and remaining two cases were adenocarcinoma and carcinoid tumor.

4 | CONCLUSION

It suggests that a suspicion of malignancy arising from MCT should be raised if the patient is elderly, the tumor size is huge, and the tumor has a larger solid component. However, malignant transformations have been reported in younger age as well, similar to the present case.

CONSENT

Written informed consent was obtained from the patient.

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CONFLICT OF INTEREST

There is no conflict of interests regarding the publication of this paper.

AUTHOR CONTRIBUTIONS

SM: conceived of the research, collected the data, and compiled and revised the manuscript.

ORCID

Sushna Maharjan https://orcid.org/0000-0001-9611-6718

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