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A rare case of microinvasive squamous cell carcinoma arising in mature cystic teratoma of the ovary

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Summary

Malignant transformation of a mature cystic teratoma (MCT) of the ovary is rare, with squamous cell carcinoma (SCC) being the most common type. The authors report a novel case of microinvasive squamous cell carcinoma arising in a mature cystic teratoma of the ovary. A 56-year-old woman presented with a 12-cm mass, which was diagnosed as a left ovarian mature cystic teratoma preoperatively by ultrasonography. Subsequently, laparoscopic surgery for the ovarian tumor was performed. The pathologic diagnosis was microinvasive squamous cell carcinoma arising in a mature cystic teratoma of the ovary. Appropriate staging surgery was then performed, with no evidence of malignant tissue except for the removed left ovary. Microinvasive SCC arising in MCT of the ovary is extremely rare, and, to the best of the authors' knowledge, this has not previously been reported in the literature.

Key words: Malignant transformation; Mature cystic teratoma; Microinvasive squamous cell carcinoma.

Introduction

Mature cystic teratoma (MCT) is the most common germ cell tumor of the ovary, and accounts for 10–20% of all ovarian tumors. Malignant transformation occurs in 1–2% of MCTs, and the prognosis of patients with this disease is generally poor [1]. In comparison with benign MCT, malignant transformation occurs in older populations with a mean age of 45 to 60 years [2]. The most common malignant transformation is squamous cell carcinoma (SCC), accounting for 63.7 to 88.9% of all malignancies originating from MCT [3].

Squamous cell carcinoma in situ arising in MCT of the ovary have been reported in the literature [4], but, to the best of the authors' knowledge, microinvasive SCC arising in MCT of the ovary has not previously been reported in the literature. Herein, they present an extremely rare case of microinvasive SCC arising in MCT of the ovary.

Case Report

A 56-year-old woman was referred to the present institution due to lower abdominal pain. Upon physical examination, a pelvic mass was noted. Transvaginal ultrasound revealed a left adnexal mass (about 12 cm). Computed tomography (CT) showed a left adnexal tumor suggesting MCT of the left ovary (Figure 1). The serum CA 125, CA 19-9, and SCC levels were normal. With a tentative diagnosis of MCT of the left ovary, laparoscopic surgery was performed. During the surgical procedure, a large, white-yellow, well-circumscribed, mobile, cystic left adnexal mass with no adherence to other pelvic structures was found. The uterus, right adnexa, other pelvic and abdominal structures, and peritoneal surfaces were all grossly normal. No ascites was observed. She underwent laparoscopic bilateral salpingo-oophorectomy. During left salpingo-oophorectomy, the tumor ruptured. Histopathologic analysis revealed microinvasive squamous cell carcinoma arising in MCT of the left ovary (Figures 2 and 3). The woman then underwent additional surgery, involving total abdominal hysterectomy, partial omentectomy, and pelvic lymph node biopsy. The pathologic specimen showed no evidence of malignant tissue. Therefore, the final diagnosis was International Federation of Gynecology and Obstetrics (FIGO) Stage Ic microinvasive SCC arising in MCT of the left ovary. The patient did not receive any postoperative chemotherapy or radiation treatment. At the time, this manuscript was being prepared, the patient was well and disease-free, in the 14th postoperative month.

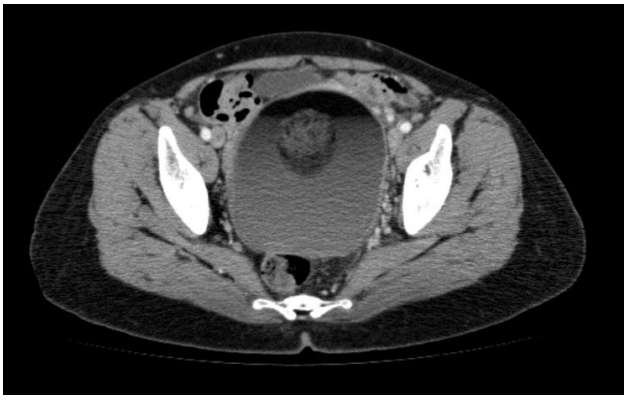

Figure 1. — Enhanced axial CT shows a 12-cm left ovarian mass.

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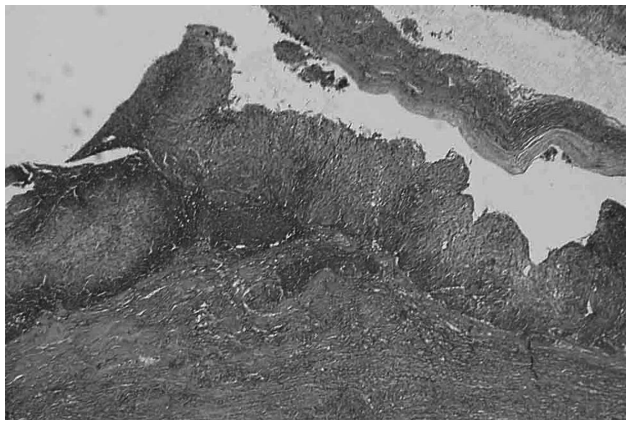

Figure 2. — The full thickness of the epithelium is composed of atypical cells with microinvasive foci (H&E, x20).

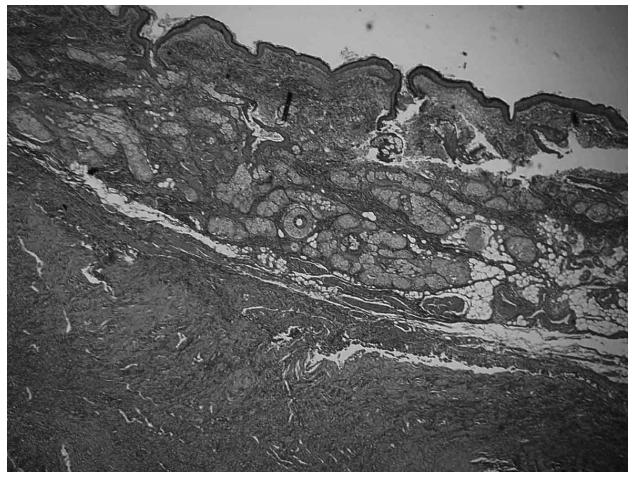

Figure 3. — The lining of the cysts is composed of skin with its associated structures (H&E, x20).

Discussion

MCT, which is composed of well-differentiated tissues derived from the three germ-cell layers (endoderm, mesoderm, and ectoderm), is the most common benign ovarian neoplasm. Malignant transformation of MCT, occurring with an average frequency of 1–2%, has been reported in isolated case reports and small case series [2, 5]. SCC is the most common transformation, although adenocarcinoma, adenosquamous carcinoma, sarcoma, carcinoid, and melanoma have also been reported [6-8]. In some cases, the diagnosis is not suspected until microscopic examination has been completed. In the present case, the authors did not expect malignant transformation of MCT because of the microinvasive cancer lesion.

Patient age, tumor size, imaging characteristics, and serum tumor markers are risk factors for malignancy arising from MCT [9]. Although malignancy occurs at any age, most patients are postmenopausal, and the median age of patients is 55 years (range: 37-75) [9]. Rim *et al*. [5] reported 11 cases of malignant transformation arising from MCT with a mean age of 50.6 years, which was much higher than the median age of 35 years in patients with MCT. In the present case, the patient's age was 56 years, which is included in the risk factor group. Tumor size has also been reported to predict malignancy. Although MCT presents in a wide range of sizes, larger tumors are related to an increased risk of malignant transformation. Kikkawa *et al*. [10] demonstrated that tumors in most cases with SCC arising from MCT are > 9.9 cm and commonly contain areas of hemorrhage and necrosis. In the present case, the tumor diameter was 12 cm, being markedly larger than a typical benign cyst. The most common pathological malignancy is SCC, which represents about 75% of malignant transformation and is diagnosed as invasive SCC. Zakkouri *et al.* reported a rare case of squamous cell carcinoma in situ arising in MCT of the ovary. Here, the authors present an extremely rare case of microinvasive SCC arising in MCT of the ovary. Fortunately, her tumor was found as an incidental microinvasive SCC. This may have progressed to invasive SCC after a prolonged period.

Due to the relative rarity of microinvasive SCC arising in MCT, there is no consensus regarding treatment. However, complete tumor excision and appropriate staging are integral to the prognosis, and optimal cytoreduction is significantly correlated with an improvement in survival [11]. Chen *et al.* [12] reported five cases of squamous cell carcinoma in situ arising in MCT, whose optimal debulking rate was 100% and the five-year survival rate in patients with this disease was 100%. However, there is no literature on microinvasive SCC arising in MCT of the ovary. The present patient underwent complete tumor excision and the pathologic specimen showed no evidence of malignant tissue except for the removed left ovary. She was well and disease-free at the 14th postoperative month, without adjuvant therapy.

In conclusion, microinvasive SCC arising in MCT of the ovary is extremely rare, and, to the best of the authors' knowledge, it has not previously been reported in the literature. As seen in the present patient, large ovarian tumors in patients with advanced ages, even if SCC markers have not been elevated, the probability of malignant transformation should always be kept in mind, even in patients whose pre- and intra-operative findings support MCT.

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