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## Case Report

## Urothelial carcinoma arising from an ovarian mature cystic teratoma



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## ABSTRACT

**Objective:** Ovarian mature cystic teratomas with malignant transformation are rare, and squamous cell carcinoma is the most common pathological entity. Among these malignant transformations, urothelial cell carcinoma is rare.

**Case Report:** We report a woman presenting with a huge pelvic cystic mass, favoring a right ovarian mature cystic teratoma with malignant transformation, based on magnetic resonance imaging, who was successfully treated with surgery.

**Conclusion:** The final pathology confirmed concomitant malignant transformation of urothelial carcinoma.

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## Introduction

Mature cystic teratoma (MCT) is one of the common benign ovarian tumors. The incidence of malignant transformation in MCTs is 1–3%. Among these malignant transformations, squamous cell carcinoma, adenocarcinoma, and carcinoid tumor account for most malignant transformations [1]; however, the co-existence of urothelial carcinoma within MCT is rare [2]. Here, we present the case of a woman with an incidental finding of a urothelial carcinoma arising from a large right ovarian MCT, who was successfully treated by surgery.

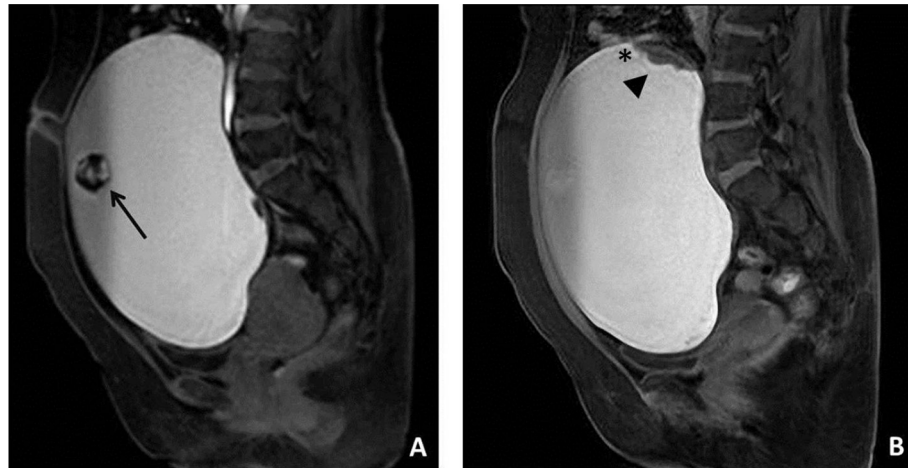
## Case Report

A 54-year-old nulliparous postmenopausal woman presented with abdominal distension and vaginal spotting. The initial pelvic examination disclosed a huge, soft mass in the pelvic cavity. Transabdominal ultrasonography revealed an 18 cm × 12 cm × 20 cm cystic mass with a smooth surface and a small fatty component within the right ovary. The tumor markers were as follows: carcinoembryonic antigen, 5.41 ng/mL (reference range < 5.0 ng/mL); carbohydrate antigen (CA)-125, 45.35 IU/mL

(reference range < 36.3 IU/mL); and CA 19-9, 928.41 IU/mL (reference range < 37.0 IU/mL). The other laboratory testing revealed no abnormalities. Pelvic magnetic resonance (MR) showed a 19 cm × 12 cm × 20 cm, lobulated cystic-solid mass with a fatty component in the right ovary (Figure 1A). The tumor had a small solid component inside the cystic lobulated component, and an enhancing papillary mural nodule projection into the cystic lumen (Figure 1B). The soft mural nodule was 4 cm in size and there was an obtuse angle between the soft component and the inner wall, favoring malignant transformation. A uterine myoma and mild ascites with borderline enlarged lymph nodes in the left internal iliac area were also noted. Colonoscopy and cystoscopy were normal.

A midline laparotomy was performed, with a provisional diagnosis of malignant transformation of MCT based on the large size, high serum CA19-9 level, and the specific finding of small solid components on pelvic MR. At the time of surgery, scant clear, yellow ascites was noted in the pelvis. A huge smooth-surfaced cystic tumor was palpated and ~2.3 L of clear, brown fluid was evacuated. The left ovary and uterus were grossly normal. Initially, a right salpingo-oophorectomy was performed. Grossly, a solid lesion was identified on the inner surface of the cyst, which measured 4.0 cm × 3.0 cm × 1.4 cm in size (Figure 2A) and the excised tumor was sent for frozen evaluation, which revealed a cystic teratoma with possible malignant transformation. Debulking surgery was then performed without incident.

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**Figure 1.** (A) Sagittal enhanced T1-weighted magnetic resonance image shows a huge cystic mass, measuring 19 cm × 12 cm × 20 cm, in the right ovary. A fatty lesion was found within the cyst (black arrow). (B) Enhanced papillary mural nodules (arrowhead), measuring 4 cm projection into the cystic lumen, made an obtuse angle (asterisk) between the soft component and the inner wall.

Microscopically, the right ovary revealed a cystic lesion consisting of mature skin appendages lined by squamous, respiratory, and urothelial epithelium (Figure 2B). Complex close-packed, irregular nests and papillary structure of urothelial cells infiltrating the stroma were noted in the solid mass area. The urothelial cells showed well-differentiated cytological features and the adjacent area showed benign urothelial epithelium. No extra-ovarian extension was identified. The final pathology report confirmed the co-existence of urothelial carcinoma from the ovarian MCT.

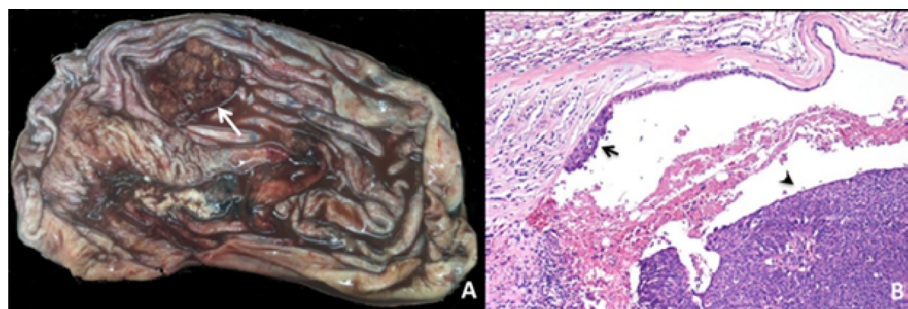
The patient had an uneventful postoperative course and was discharged 4 days later without sequelae. Subsequent 18-fluorodeoxyglucose positron emission tomography/computed tomography (18-FDG PET/CT) did not show distant metastasis or local recurrence 8 months later.

## Discussion

MCT is one of the most common ovarian tumors and accounts for 10–25% of ovarian tumors [1]. Malignant transformation, a rare pathological entity, occurs in 1–2% of MCTs and is rarely recognized preoperatively [1–3]. For patients with malignant transformation, squamous cell carcinoma is the most common malignancy arising from MCT, and accounts for 80% of all cases [4]. Other malignancies, such as adenocarcinomas, sarcomas, or malignant melanomas, have been reported [2,4]; however, urothelial carcinoma arising from

MCT is rare. Only five cases of urothelial carcinoma arising from MCT have been reported, including the patient reported here. An extensive review of the literature of urothelial carcinoma malignant transformation is presented in Table 1 [1,5–7]. The mean age of the five cases was 53 years. Only two of these were postmenopausal and the other two were premenopausal; this is not compatible with the previously published data [2,3,6]. Mostly, these tumor masses were > 10 cm in size at the time of diagnosis and malignant transformation of urothelial carcinoma was found in the early stages. All of these patients were treated primarily by surgery, including salpingo-oophorectomy. Only one patient underwent adjuvant chemotherapy, possibly for advanced stage disease [5].

The common clinical manifestations of malignant transformation of MCT include abdominal mass, abdominal pain, weight loss, abdominal distension, vagina bleeding, and urinary frequency [3,4,6]. Although malignant transformation can occur at any age, it seems that most patients have this condition during the postmenopausal period. The other risk factors of malignant transformation include older age (> 45 years), concomitant larger tumor size (> 9.9 cm), and elevated serum tumor markers (CA125, CA199, carcinoembryonic antigen, and squamous cell carcinoma antigen – SCC) [2,3,6]. No specific tumor marker is used to detect or screen concomitant malignant transformation of MCT, especially urothelial carcinoma, but CA125 and CA199 may be elevated in some cases (Table 1) [5].



**Figure 2.** (A) Gross photograph of right ovarian tumor. A mural solid tissue mass (white arrow) was noted inside the cystic tumor without serosa invasion. (B) Complex close-packed, irregular nests and papillary structure of urothelial cells infiltrating the stroma (black arrowhead) are noted. The urothelial cell shows moderate grade cytological features. The adjacent area shows benign urothelial epithelium (black arrow; hematoxylin and eosin, 10×).

**Table 1**  
Reported cases of urothelial carcinoma arising from ovary cystic teratoma.

Study	Age (y)	Menopause	Symptoms at diagnosis	Tumor size (cm)	Right/left	Tumor marker elevated	FIGO stage	Primary surgery	Further treatment	Follow-up
1 Lee et al [5], 1999	67	Post	voiding difficulty lower abdominal pain	14 × 7 × 5	left	CA-125 CA 19-9	1C	ATH + BSO + omentectomy	Chemotherapy: carboplatin etoposide	NED 5 mo
2 Kido et al [1], 1999	48	NA	NA	NA <sup>a</sup>	NA	NA	1C	NA	NA	NDA
3 Rayyan et al [6], 2009	45	Pre	Pain bleeding	8	Left	NA	1A	LSO	Surgical staging	NED >5 y
4 Lee & Lee [7], 2010	52	Pre	Abdominal mass	22 × 19 × 5	Right	NA	1A	RSO	NA	NED 15 mo
5 Present case	54	Post	Abdominal mass	20 × 13 × 21	Right	CA-125 CA 19-9	1A	Debulking (ATH + BSO + omentectomy + BPLA)	None	NED 8 mo

ATH = abdominal total hysterectomy; BPLA = bilateral pelvic lymphadenectomy; BSO = bilateral salpingo-oophorectomy; DOD = died of disease; FIGO = International Federation of Obstetrics and Gynecology; LSO = left salpingo-oophorectomy; NA = not available; NDA = no data available; NED = no evidence of disease; RSO = right salpingo-oophorectomy.

<sup>a</sup> The authors did not mention the size of this tumor, but they described that the five tumors ranged from 10 cm × 10 cm to 14 cm × 32 cm.

MCT is easily diagnosed via sonography, but malignant transformation is difficult to identify before surgery. MR is useful in the detection of occult malignancy arising from MCTs [1,3]. The appearance of MCTs can vary, but the surface of benign MCTs is always smooth. A soft tissue protuberance into the cystic lumen, known as Rokitansky nodule, is common in MCTs. The Rokitansky nodule never shows a transmural growth pattern, favoring malignant transformation [3]. MCT with malignant transformation is characterized by the specific MR findings: > 9.9 cm; presence of enhancing soft tissue components; obtuse angle between the soft tissue components and the inner cyst wall; transmural or extracapsular tumor growth with extension into adjacent structures; or disseminated metastasis [3]. It is reported that malignant transformation tends to have solid components inside the tumor that may exhibit transmural extension with direct invasion of neighboring organs. Moreover, 18-FDG PET/CT has been used for the initial diagnosis and further staging when malignant transformation is highly suspected [3,8]. Although we applied an 18-FDG PET/CT survey for this patient postoperatively, it still had its clinical significance for definite staging.

In conclusion, urothelial carcinoma arising from MCT is rare. For those patients with larger size, transmural-enhancing soft tissue components inside the teratoma, > 45 years of age, or elevated tumor markers, the possibility of malignant transformation should not be omitted. Moreover, an adjunct PET/CT might be considered for preoperative staging if malignant transformation is suspected.

## Conflicts of interest

The authors have no conflicts of interest relevant to this article.

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