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Case Report

A Rare Case of Malignant Transformation arising From Mature Cystic Teratoma into Squamous Cell Carcinoma of The Ovary With Poor Prognosis in a Young Woman

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Abstract

Background: Squamous cell carcinoma is rarely seen as the malignant transformation of mature cystic teratoma of the ovary. Although mature cystic teratoma mostly occurs in young women, the malignant transformation occurs after perimenopause in most cases. Therefore, we rarely encounter the case of young-onset malignant transformation. The risk factors for the development of the malignant transformation are unclear and the details of young-onset malignant transformation are unknown.

Case presentation: We present a case of a 38-year-old woman with 13cm large multilocular tumor containing fat and solid components on Pelvic MRI. SCC value was 2.1ng/ml. The tumor had the invasion into the uterus and the mesenteric surface of the sigmoid colon on laparotomy. The pathological diagnosis was Squamous cell carcinoma as the malignant transformation derived from mature cystic teratoma of the ovary, FIGO Stage IIb after the complete resection including total abdominal hysterectomy with bilateral salpingo-oophorectomy. She suffered from the repeated melena by the penetration to the rectum of the disseminated nodule one month later from her first adjuvant chemotherapy. She died 6 months after her operation.

Conclusion: However, there have been scattered case reports of young-onset malignant transformation arising from mature cystic teratoma, this is the first detailed and decent report of the malignant transformation with poor prognosis and very short time from initial diagnosis to death in a young woman. We need to accumulate the multidisciplinary date on young-onset malignant transformation to clarify the risk factors for the onset of the malignant transformation. Our task is to select and treat the patients with mature cystic teratoma progressing to the malignant transformation in the future as early as possible.

ABBREVIATIONS

MCT: Mature Cystic Teratoma; MT: Malignant Transformation; SCC: Squamous Cell Carcinoma; MRI: Magnetic Resonance Imaging; CT: Computed Tomography; FIGO: International Federation Of Gynecology And Obstetrics; TC: Paclitaxel and Carboplatin; PD-1: Programmed Cell Death-1

INTRODUCTION

Mature cystic teratoma (MCT) is the most common ovarian germ cell neoplasm and accounts for approximately 10-20% of all ovarian tumors [1,2]. The malignant transformation (MT) derived from MCT accounts for approximately 0.2-3.5% of MCT [1-5,19]. The histological type representing about 64-80% of MT is Squamous cell carcinoma (SCC) [1,3,6]. We generally find MCT

in young women, whereas MT mostly occurs after perimenopause [1-7,10-13,17-19]. Interestingly, there are some cases of young-onset MT within the past reviews [2-6,8,9,11,13,15,16,18,19]. In fact, 11-37.5% of MT were young 30s [2,8] and approximately 30% of young-onset MT followed the poor prognosis in advanced stage [8]. But we cannot find a comprehensive date on young-onset MT so far.

CASE PRESENTATION

The patient was a 38-year-old nulliparous woman with a chief complaint of low abdominal pain for about one month. She had anorexia nervosa and no family history. The multilocular and heterogeneous tumor 13cm large and the ascites in the Douglas' pouch were detected on the pelvic Magnetic Resonance Imaging (MRI) (Figure 1). The Computed Tomography (CT) examination revealed no distant metastasis and no local invasion. SCC value was 2.1ng/ml (within normal range is less than 1.5ng/ml). The cervical and the endometrial cytology showed no abnormalities. We suspected MCT with some kind of malignancy of the left ovary. We performed the complete resection including total abdominal hysterectomy and bilateral salpingo-oophorectomy. The abnormal findings on laparotomy were as follows: The left ovarian tumor 13cm large adhered firmly to the uterus and the mesenteric surface of the sigmoid colon, forming a cyst in the Douglas' pouch. The yellowish gray ascites was negative cytology. Macroscopically, the cystic tumor had the thickened wall, hyaline, hair, the fat component, and the necrotic material (Figure 2). Microscopically, we found moderately differentiated SCC (Figure 3A) in MCT (Figure 3 B,C) with the weak stromal reaction. However, the squamous epitheliums as the skin components were confirmed as shown (Figure 3B,C), we could not find the transitional areas from the normal squamous epithelium to SCC due to the severe necrosis.

The final diagnosis was SCC as MT arising from MCT, International Federation of Gynecology and Obstetrics (FIGO) Stage IIb. The multiple peritoneal disseminations appeared at the time of her first adjuvant Paclitaxel and Carboplatin (TC) therapy. She was forced into the palliative care because of the repeated melena by the rectal penetration of the disseminated nodule about one month after her first chemotherapy. She died 6 months after her operation.

DISCUSSION

The average onset age of MT was 53.5 years whereas the mean onset age of MCT was significantly younger 38.8 years and the average tumor size of MT was 15.5cm more than 5cm larger than that of MCT in a study of 527 cases of MCT [7]. Similar report has been made elsewhere [2]. Large MCTs after perimenopause may have already progressed to MT at the time of initial diagnosis [3-5,10,11,13,14,17]. The difference between our case and the previous reports was the largeness of the tumor size despite being young. We should keep in mind the possibility of malignancy and perform some surgery if we catch a deviation from the typical findings even a little in daily clinical practice.

Although there are some cases of young-onset MT in the previous reports of MT, we can find only the total number of patients and their mean, minimum, and the oldest ages without the specific age groups and its details [2-4,6-9,11,13,18,19]. For young cases of MT, the detailed case report is available for the successful pregnancy and delivery due to the good prognosis on the early stage [16]. The case of poor prognosis in a young woman was not SCC but Adenosquamous cell carcinoma as MT causing the death of just two months with no details [8]. This patient is the first detailed case of young-onset SCC as MT with very poor prognosis.

In this patient, MT and MCT were found simultaneously at young 30s. A possible reason for the young-onset MT like this case could be carcinogenesis by a mechanism close to de novo. On the other hand, multistage carcinogenesis accumulated by multiple genetic abnormalities over a long period of time can be considered as a reason of the MT in postperimenopausal women, since MT often occurs from MCT over the years after perimenopause [2-13,17]. Interestingly, Tamura et al. report that XCL-1 expression may be a novel biomarker for MT from MCT into SCC and a biomarker candidate for the therapeutic response to an anti-Programmed cell death-1 (PD-1) / PD-L1 therapy

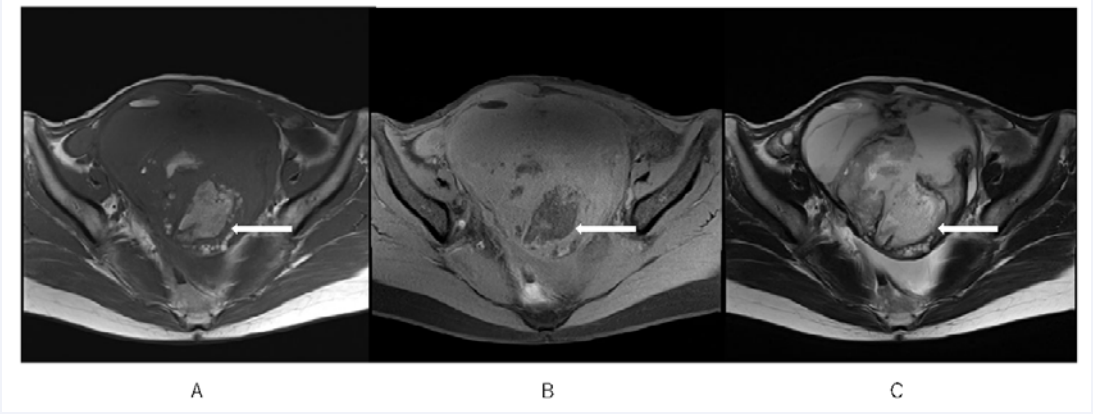

Figure 1 The MRI findings.
The tumor was revealed with the partial high signal intensities (arrows) in the T1-weighted image (A) and the areas of high signal intensity (arrows) in the T1-weighted image were suppressed in the fat-suppressed T1-weighted image (B). The multilocular and heterogeneous tumor with the markedly high signal intensity (arrows) was found in the T2-weighted image (C).

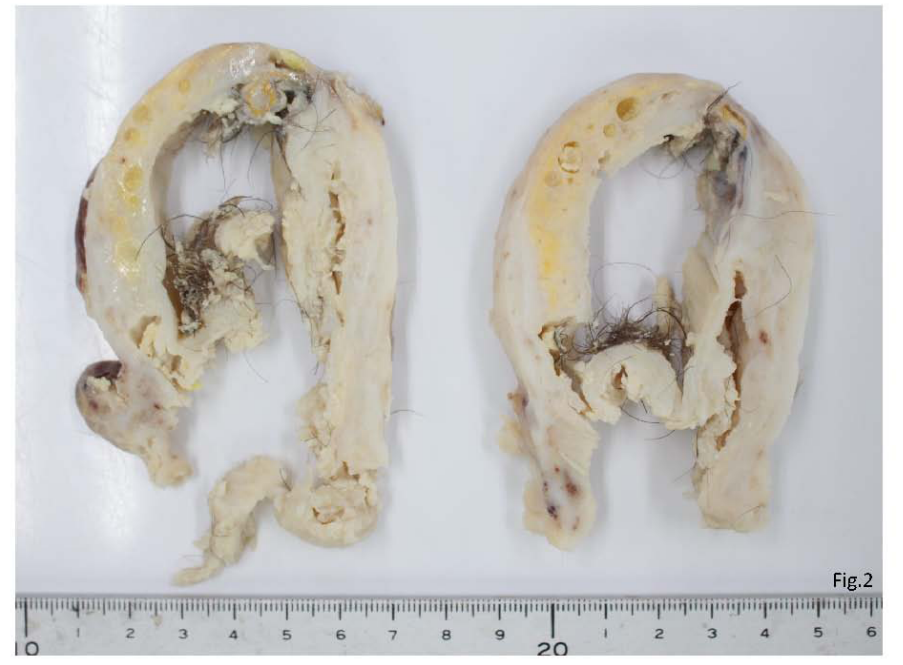

Figure 2 The macroscopic view of sebaceous glands, hair, and necrotic material in the cystic tumor with the thickened wall.

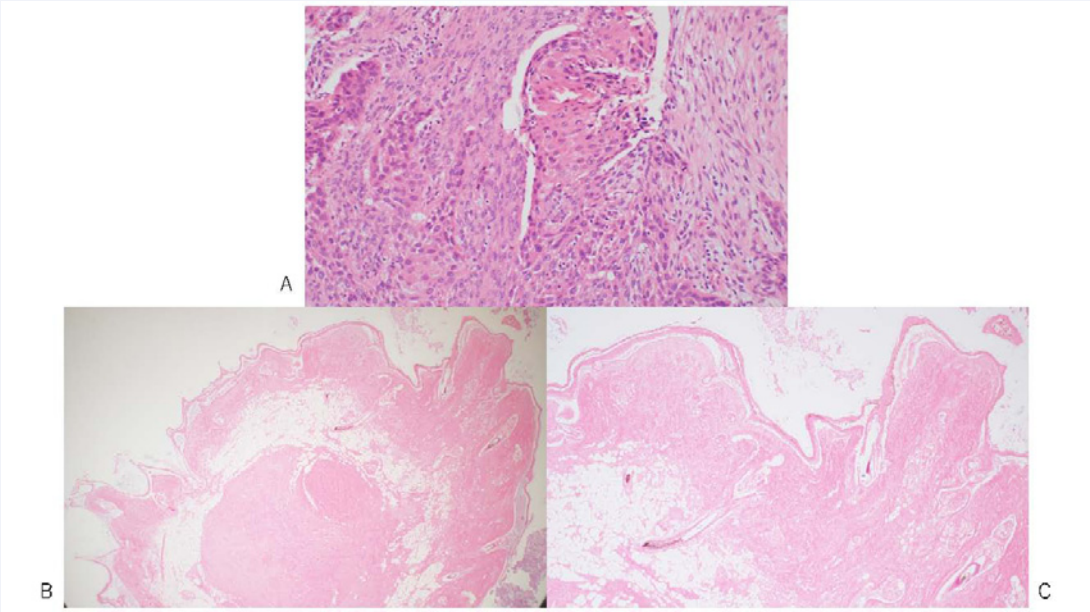

Figure 3 The microscopic view of moderately differentiated SCC (A) arising from MCT (B and C) of the left ovary (H & E stain under A 200x, B 20x, C 40x magnification).

through the immunohistochemistry analysis [8]. However,we unfortunately missed the opportunity to perform genetic testing in the present case, as a next step, genetic testing in such a young-onset MT will be the key to pinpointing the risk factors for progressing to MT in the future.

We also found a variety of young-onset MT between the medical stage and the prognosis. This patient was exposed to the sharp exacerbation even with the complete resection of the tumor and negative ascites cytology. No specific measures against young-onset MT patients in advanced stage or under rapid progression have been reported. But some cases of young-onset MT have been also reported in early stage and have a relatively good prognosis [8,12,14-16,18,19]. Among other things, fertility-preserving surgery is acceptable for Stage I MT and results in no postoperative recurrence [18,19]. The reason of the differences in these previous reports even though the young-onset MT originates from the same MCT remains elusive. Further

investigation whether there are some differences in progression in young-onset MT is our future task, too.

CONCLUSION

This is a first detailed and decent case report of SCC as MT and MCT found simultaneously at young 30s leading to poor prognosis.

In the present report, we propose the following new two viewpoints. First, since there is a time differences in the MT onset derived from MCT between young women and post-perimenopausal women, there may be some differences in the pathogenesis of MT between them, for example, in gene expression. Second, there are different prognostic patterns even with the same young-onset MT.

As a next step, when we encounter such a young-onset MT case, we should perform some genetic analysis to clarify the risk factors for MT. Our mission is to detect and treat earlier the patients with MCT at the risk for developing MT in the future.

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