Juvenile ovarian mature teratoma with malignant transformation
diagnosed after laparoscopic surgery: A case report

Nozomu Yanaihara¹, Chihiro Sato¹, Masami Iwamoto², Kazu Ueda¹, Aikou Okamoto¹

¹Department of Obstetrics and Gynecology, The Jikei University School of Medicine 3-25-8 Nishi-Shinbashi, Minato-ku, Tokyo 105-8461
²Department of Pathology, The Jikei University School of Medicine 3-25-8 Nishi-Shinbashi, Minato-ku, Tokyo 105-8461 (Japan)

Summary
Malignant transformation of an ovarian mature teratoma is rare, and it might be diagnosed following laparoscopic surgery in women of reproductive age. Here we report a rare juvenile case of squamous cell carcinoma arising in an ovarian mature teratoma. A 29-year-old woman with a preoperative diagnosis of ovarian mature teratoma underwent single-incision laparoscopic surgery, in which tumor resection was performed extracorporeally following fine-needle aspiration of the cystic contents. Histopathological examination revealed squamous cell carcinoma arising in a mature teratoma of the right ovary. Subsequent right salpingo-oophorectomy and partial omentectomy showed no evidence of a residual malignant component. Because the rate of tumor rupture is significantly higher with the laparoscopic approach than with the laparotomy approach, maximum efforts should be made to avoid intraoperative tumor spillage in cases of an ovarian mature teratoma considering the possibility of unexpected malignancy.

Key words: Laparoscopic surgery; Malignant transformation; Ovarian mature teratoma.

Introduction
Ovarian mature teratoma is the most common pathological type of benign ovarian tumor among women of reproductive age. It has been reported that secondary malignant transformation, in which squamous cell carcinoma accounts for approximately 80%, can occur in 1%–2% of ovarian mature teratoma, typically in postmenopausal women [1-3]. Here we present a rare case of a juvenile ovarian mature teratoma with malignant transformation that was diagnosed after laparoscopic surgery.

Case Report
A 29-year-old nulligravida woman was referred to our hospital with pelvic masses. She did not have relevant medical and family histories. She had been having regular menstrual cycles since menarche. Pelvic examination revealed a large mass at Douglas’ pouch, and transvaginal ultrasonography revealed a solid 8-cm mass in the right adnexal region and a 4-cm mass in the left adnexal region. Laboratory values were within normal limits, including those of tumor markers such as SCC, CA125, CA19–9, and AFP. Pelvic magnetic resonance imaging revealed two mixed-tissue masses, suggesting mature teratomas derived from both ovaries (Figure 1). A preoperative diagnosis of ovarian mature teratoma was made, and minimally invasive low-position single-incision laparoscopic surgery was performed [4]. A 3-cm incision was made at 1 cm from the pubis for the insertion of Lap Protector™ (Hakko, Tokyo, Japan) and EZ Access (Hakko). During the procedure, an apple-sized mass and a pigeon’s egg-sized mass with smooth surfaces were identified in the right and left ovaries, respectively (Figure 2a). Additionally, a small amount of serous ascites was observed. To avoid intraoperative spillage of tumor contents, the tumors were resected extracorporeally following fine-needle aspiration of the contents [4]. Histopathological examination revealed squamous cell carcinoma arising in mature teratoma of the right ovary and mature teratoma without malignant potential of the left ovary (Figure 2b). Next, the patient underwent right salpingo-oophorectomy and partial omentectomy, and no evidence of a residual malignant component was observed. Finally, the patient was diagnosed with International Federation of Gynecology and Obstetrics (FIGO) stage IC1 (pT1c1NxMx) disease. She did not receive further postoperative treatment, and was free of disease during 12 months follow-up.

Discussion
Because of the rarity of an ovarian mature teratoma with malignant transformation, which occurs in 1%–2% of the ovarian germ cell tumors, information on its clinicopathological features, treatment modalities, and prognostic factors is limited. Thus far, two systematic literature reviews that analyzed more than 200 cases of squamous cell carcinoma arising in an ovarian mature teratoma, which is the most common form of malignant transformation, have been reported [1, 2]. Patient age (mean age, 55 years), tumor burden (mean size, 148 mm), specific imaging features, and serum tumor markers (SCC, CEA, CA19–9, and CA-125) have been shown to be associated with the risk of malignant
transformation of an ovarian mature teratoma [1-3]. Recently, genomic abnormalities in squamous cell carcinoma arising in an ovarian mature teratoma indicated that this rare tumor has a specific mutation profile characterized by high rate of \( TP53 \) mutation, which is associated with a favorable prognosis [5]. Other factors associated with prognosis include clinical stage, tumor size, tumor markers, and tumor debulking [1-3, 6]. In particular, FIGO stage has been shown to be the most important prognostic indicator among these factors, and the 5-year survival rate has been shown to be > 95% for stage I disease [1-3, 6].

There is no clear guideline on the optimal treatment for this rare tumor type [1-3, 6]. It is known that accurate staging and complete cytoreduction at the initial intervention are clinically important. However, the benefit of either adjuvant chemotherapy or radiotherapy is uncertain. With regard to surgery, the adverse effects of tumor rupture remain debatable. Although rupture of the tumor capsule, which may cause tumor cell spillage, is not associated with poor patient prognosis [1], malignant cells can be rapidly disseminated owing to artificial spillage into the peritoneal cavity [7, 8]. Therefore, intraoperative spillage of tumor contents should be avoided as much as possible. There may be arguments about surgical approaches in young women with ovarian mature teratomas, who are at a relatively low risk for malignant transformation. Laparoscopic ovarian cystectomy is generally accepted in these patients despite a considerably high risk of tumor rupture during surgery. It was previously reported that the rate of tumor spillage was significantly higher with the laparoscopic approach than with the laparotomy approach [9]. However, in the present case, the possibility of tumor spillage was minimized by utilizing a novel laparoscopic technique, in which the cystic contents were suctioned directly by using a fine needle under the incision point before removal of the cystic wall. We previously reported that this novel approach of reduced port surgery has the same safety and efficacy as those of the conventional multiport procedure for the treatment of patients with ovarian cysts [4].

In conclusion, malignant transformation is a rare compli-
cation of ovarian mature teratomas in women of reproductive age, and it may be diagnosed after laparoscopic surgery. Maximum efforts should be made to avoid intraoperative tumor spillage in cases of an ovarian mature teratoma considering the possibility of unexpected malignancy.

Authors contributions

NY and KU designed the study. CS and MI provided help and advice on imaging studies. NY wrote the manuscript. AO supervised all of this study. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript.

Ethics approval and consent to participate

Informed consent was obtained from participant. The Ethics Review Committee of The Jikei University School of Medicine approved this case report.

Acknowledgements

The authors would like to thank Enago for the English language review.

Conflict of interest

There are no conflicts of interest to declare for any of the authors.

Submitted: November 16, 2018
Accepted: January 21, 2019
Published: June 15, 2020

References


Corresponding Author:
Nozomu Yanaihara
Assistant Professor, Department of Obstetrics and Gynecology
The Jikei University School of Medicine
3-25-8 Nishi-Shinbashi, Minato-ku, Tokyo 105-8461 (Japan)
E-mail: yanazou@jikei.ac.jp