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Presentation of a Patient who Underwent Fertility-Sparing Surgeries for Contralateral Recurrence of Ovarian Immature Teratoma with Gliomatosis Peritonei

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Abstract: We report a patient who has maintained a regular menstrual cycle despite undergoing cystectomy and chemotherapy for contralateral recurrence of ovarian immature teratoma with gliomatosis peritonei. We initially performed a fertility-sparing right salpingo-oophorectomy, omentectomy and peritoneal biopsy for immature teratoma with gliomatosis peritonei, with adjuvant chemotherapy; we performed a left ovarian cystectomy and peritoneal biopsy for mature cystic teratoma with gliomatosis peritonei 16 months after the first surgery, a fertility-sparing left ovarian cystectomy and peritoneal biopsy for contralateral recurrence of ovarian immature teratoma with gliomatosis peritonei 60 months after the first surgery, and a left ovarian cystectomy and peritoneal and external iliac lymph node biopsy for endometrial cyst with gliomatosis peritonei 71 months after first surgery. The peritoneal gliomatosis lesions gradually decreased through the 4 surgeries over 8 years. The patient has maintained a regular menstrual cycle and currently shows no evidence of disease.

Keywords: immature teratoma, conservative surgery, gliomatosis, recurrence, ovary

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Introduction

Immature teratoma is a type of ovarian germ cell tumor diagnosed in young women. It is commonly treated with conservative surgery; prognosis is rapidly improved by performing concomitant chemotherapy after the initial surgery.^{1,2} Gliomatosis peritonei is the metastatic implantation of mature glial tissue within the peritoneal cavity of patients with ovarian teratomas.³ As second-look or secondary debulking surgery is not generally performed for immature teratoma, as gliomatosis peritonei is not usually discovered in a patient who has completed chemotherapy and shows no evidence of disease.⁴ This is a rare case of a patient with ovarian teratoma recurrence in the contralateral ovary. The first and later occurrences were treated with conservative surgery, using local resections or cystectomies. We were able to follow the gliomatosis peritonei over eight years, using biopsies. We observed that the peritoneal implants have not become malignant and have slowly shrunk.

Case Report

A 27 year-old single woman—gravidia 0, para 0—was referred to us because of a lower abdominal mass; magnetic resonance (MR) imaging revealed a tumor 20 cm in size. Tumor marker tests revealed cancer antigen 125 = 279 U/mL, CA19-9 = 130.6 U/mL, squamous cell carcinoma = 3.9 ng/mL, and alpha-fetoprotein = 50.6 ng/mL. After providing informed consent, she selected conservative surgery.

We performed a right salpingo-oophorectomy, partial omentectomy and peritoneal biopsy, and found lint-like scattered mass lesions 2 mm in size at the vesicouterine pouch and the Douglas pouch. The tumor had progressed to the peritoneum of the Douglas pouch.

The tumor is a mass with cystic regions of many sizes, with solid parts and adipocyte-laden parts (Fig. 1). The diagnosis was stage II c(a) immature teratoma grade 2 of the right ovary with gliomatosis peritonei. Pathologically, the mass contained squamous epithelium, adipose tissue, bone and cartilage tissue, and immature neuroectodermal tissue, and was therefore classified as a grade 2 immature teratoma (Fig. 2A and B). Peritoneal lesions, which were composed of mature glial cells, were diagnosed as gliomatosis peritonei using hematoxylin and eosin (H&E) stain (Fig. 2C). Immunohistochemically, the glial cells

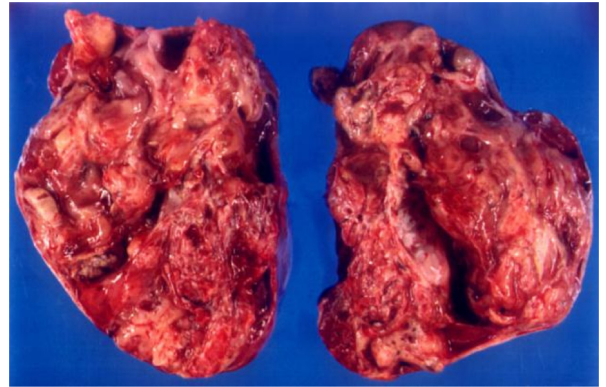


Figure 1. A tumor with cystic regions of many sizes with solid parts, and adipocyte-laden parts.

were positive for glial fibrillary acidic protein (GFAP), which is expressed by numerous cell types of the central nervous system, including astrocytes, ependymal cells and glial cells (Fig. 2D). Subsequently, the patient received cisplatin-based chemotherapy.

Sixteen months after the first operation, the patient underwent cystectomy of a left ovarian tumor and a Douglas pouch biopsy. Pathological diagnosis was mature cystic teratoma of left ovary and gliomatosis peritonei (Fig. 2E).

Sixty months after the first operation, MR imaging revealed a left cystic ovarian tumor with solid parts 7 cm in size. We recommended left salpingo-oophorectomy, but the patient selected further conservative surgery. Cystectomy of the left ovarian tumor and a Douglas pouch biopsy were performed. Pathologically, the mass contained immature neuroectodermal tissue, and so was classified as a grade 1 immature teratoma and gliomatosis peritonei (Fig. 2F and G). Subsequently, the patient received cisplatin-based chemotherapy again.

71 months after the first operation, cystectomy of a left ovarian tumor—5 cm in size—and biopsies of the Douglas pouch and left external iliac lymph node—8 mm in size—were carried out. The pathological diagnosis was endometrial cyst of left ovary, gliomatosis peritonei and glial implant in lymph node (Fig. 2H and I). The glial cells and GFAP expression in the peritoneal biopsy specimen had decreased.

Although this patient has never conceived a child, she has maintained a regular menstrual cycle, despite multiple surgeries for ovarian teratoma and chemotherapy.

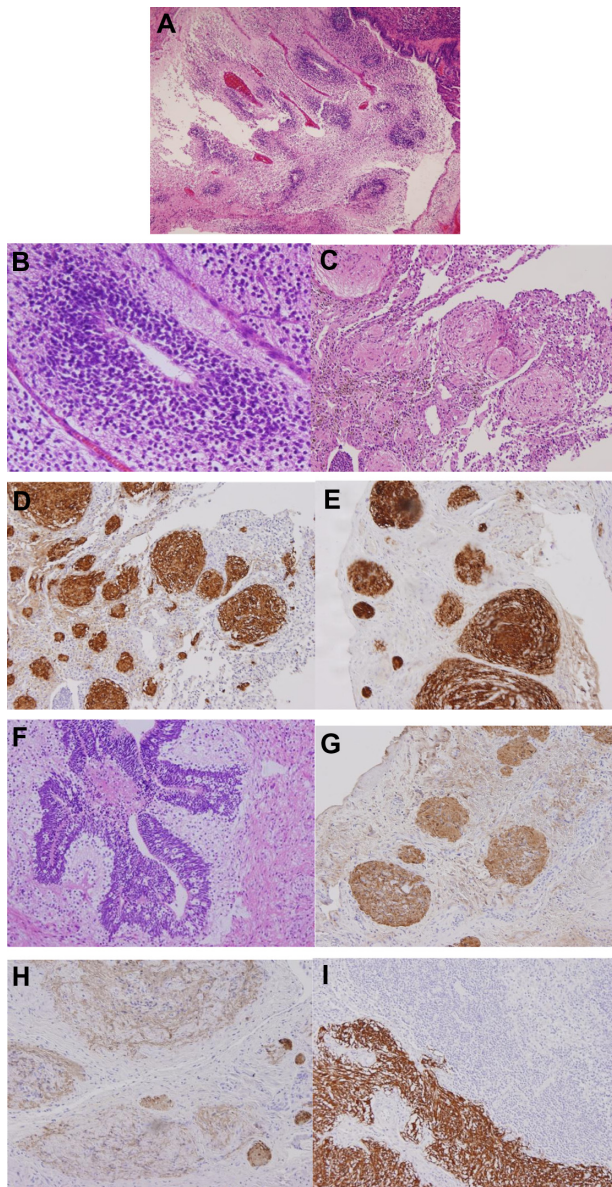


Figure 2. Pathological findings. (A) Immature neuroectodermal tissue in the first operation ($\times 40$ H&E). (B) Immature neuroectodermal tissue in the first operation ($\times 400$ H&E). (C) Gliomatosis peritonei in the first operation ($\times 100$ H&E). (D) GFAP expression in gliomatosis peritonei in the first operation. (E) GFAP expression in gliomatosis peritonei in the second operation. (F) Immature teratoma grade 1 in the third operation ($\times 100$ H&E). (G) GFAP expression in gliomatosis peritonei in the third operation. (H) GFAP expression in gliomatosis peritonei in the fourth operation. (I) GFAP expression in the glial implant in lymph node in the fourth operation.

She currently shows no evidence of disease.

Discussion

Immature teratoma of grades 1 and 2 are considered ovarian germ cell borderline tumors, according to the World Health Organization's classification. As they

occur predominantly in women in their teens and 20's, conservative surgery is performed as a standard; concomitant chemotherapy after the initial surgery has led to rapid improvements in prognosis.^{1,2} Jefferys et al produced a retrospective study of 47 patients with malignant ovarian germ-cell tumors who were treated by conservative surgery and adjuvant chemotherapy. During chemotherapy, 61.7% (29/47) of patients developed amenorrhea but 91.5% of these women resumed normal menstrual function upon completion of chemotherapy. Twenty patients (42.6%) attempted conception after chemotherapy, 19 of whom (95%) were successful; 14 healthy live births were recorded, with no documented birth defects.⁵

In patients with ovarian germ-cell tumors, a recurrence in the contralateral ovary still could be treated by a local resection or cystectomy followed by chemotherapy if fertility was desired, thereby preserving some normal ovarian tissue, if present.⁵ However, there is little information about recurrence in the contralateral ovary in patients with immature teratomas. In the present case, we re-performed conservative surgery for contralateral recurrence of an ovarian immature teratoma. As such cases are too uncommon to offer evidence for or against repeated fertility-conserving surgery, such surgery should be performed only after obtaining informed consent.

Histological grading of peritoneal implants, from grade 0 to grade 3, is essential for therapeutic and prognostic considerations. Grade 0 is defined by tumor consisting of only mature tissue with no mitotic activity. Gliomatosis peritonei is the metastatic implantation of the mature glial tissue in the peritoneal cavity of patients with ovarian teratomas.³ The prognosis for gliomatosis peritonei should be very good and no further therapy should be necessary. Peritoneal implants consisting of embryonic tissue and metastatic immature teratomas imply a poor prognosis.⁶ If no other teratomatous elements or malignant glial tissue can be found in the implants, the mature glial implants can be ignored and the methods of therapy should be judged only by the stage and grade of the primary ovarian teratoma.⁷ Progression of the peritoneal implants may be as follows: (1) transforming fibroblastic and eventual disappearance; (2) transformation to malignant tissue (glial or teratomatous); and (3) persistence



without morphological changes.⁸ In the present case, gliomatosis peritonei was initially classified as grade 0. Because the peritoneal implants were not malignant and shrank over the 8 years, we thought them to be of the first stage (1) described above.

Although the gliomatosis peritonei shrank, a lymph node glioma was detected in the fourth surgery. Presence of glial tissue in lymph nodes is rare, with very few reported cases. Patients with intraperitoneal and lymph node metastases of mature glial tissue do not need therapy for such metastases. The prognosis for these patients is excellent, but they require long-term follow-up.

Author Contributions

Conceived and designed the experiments: SS, YM. Analyzed the data: SS, YM. Wrote the first draft of the manuscript: YM, MT. Contributed to the writing of the manuscript: SS, YM. Agree with manuscript results and conclusions: TS, KW, OI. Jointly developed the structure and arguments for the paper: YM. Made critical revisions and approved final version: YM. All authors reviewed and approved of the final manuscript.

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Competing Interests

Author(s) disclose no potential conflicts of interest.

Disclosures and Ethics

As a requirement of publication the authors have provided signed confirmation of their compliance with ethical and legal obligations including but not limited to compliance with ICMJE authorship and competing interests guidelines, that the article is neither under consideration for publication nor published elsewhere, of their compliance with legal and ethical guidelines concerning human and animal research participants (if applicable), and that permission has been obtained for reproduction of any copyrighted material. This article was subject to blind, independent, expert peer review. The reviewers reported no competing interests.

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