

A Recurrence Case after Full-term Delivery of Endometrial Stromal Sarcoma with Fertility Conservative Treatment

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ABSTRACT Objective Endometrial stromal sarcoma, a relatively rare female tumor, was about 0.2% of uterus tumors. Operation is the chief treatment measure for the disease. The paper reports a case of recurrence of endometrial stromal sarcoma with fertility conservative treatment after full-term delivery. **Case** A 24-year-old unmarried patient with increasing amount of menstruation in the late six months had a mass in her lower-abdomen, which was confirmed by B-ultrasonic as myometrium mixed mass, she was given an enucleation of uterus tumor. The pathological examination of the tumor sample revealed that the tumor was a low-grade endometrial stromal sarcoma (LESS). And the patient underwent 6 cycles of chemotherapy over 8 months before she got pregnant and had a full-term delivery. But in 34 months after delivery, the disease recurred with metastasis of pelvic and peritoneal cavity and the patient had to receive comprehensive therapy. **Conclusion** The unmarried young patient with stage I low-grade endometrial stromal sarcoma has the chance to preserve uterus for her pregnancy and delivery. But the using of cesarean needs prognosis evaluation.

Key words Endometrial stromal sarcoma; Full-term pregnancy; Recurrence; Fertility conservation

Endometrial stromal sarcoma, a relatively rare female tumor, was about 0.2% of uterus tumors. Larson^[1] reported that the average age of the patients who got low-grade endometrial stromal sarcoma was about 47.2 years old, while those who got high-grade one was about 50.2 years. Operation is the chief treatment measure for this disease. The paper reports a case of recurrence of endometrial stromal sarcoma with fertility conservative treatment after full-term delivery.

CLINICAL MATERIALS

A unmarried patient with 24 years old was found to have a lower-abdominal mass for two months, because of her increasing amount of menstruation in the late six months and she was sent to the hospital for checking on 11th, October, 2001. The B-ultrasonic diagnosis revealed there was a myometrial mixed mass (7 cm × 6 cm × 5 cm). In gynecologic examination, the uterus of

the patient was found with a protrusion (7cm) at the right side wall but no abnormal bilateral oviduct and menstruation. Blood CA125, CEA, blood and urine routine, and the function of liver and kidney were all normal. The patient was pre-operative diagnosed as hysteromyom and undertook enucleation of hysteromyoma on 19th October, 2001. The patient was found with a protrusion (about 7 cm × 6.5 cm × 6 cm) in the middle and back wall of uterus but normal in bilateral oviduct and enlarged ovary (3 cm × 2 cm × 1.5 cm) with several vesicles, Omentum and intestine were all normal. The operation was successful with blood loss being 150ml. Autopsy reports showed it as low-grade endometrial stromal sarcoma. The immunohistochemistry method showed that SMA, Desmin and P53 were negative while Vimentin, ER, PR and CD10 were positive. Chemotherapy was given for 6 cycles with the following medicine: DDP(Cisplatin, DDP) 30mg d1-3, CTX (Cyclophosphamide, CTX) 0.4 d1-3, THP (Pirarubicin) 40mg d1 plus VCR(Vincristin, VCR) 2mg d1. The chemotherapy was over on 28th April, 2002 and the patient turned to take traditional Chinese medicine.

At the end of 2002, the patient got married and got pregnant in Jan, 2003 and on 25th September, 2003, she delivered a mature male infant about 3,800g with cesarean. During the delivering operation, uterus,

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oviduct, pelvic and abdominal cavity, liver, gallbladder and spleen were all found normal. On 15th November, 2005, the patient was found with hysteromyoma by B-ultrasonic diagnosis with its diameter being 1.5~2 cm. and doctor's suggestion about operation was again refused. On 7th June, 2006, the patient was hospitalized with abdominal distension. CT diagnosis showed that she got malignant uterus tumor with some pelvic effusions and lymph nodes metastasis. On 11th June, the patient received one cycle of the intravenous chemotherapy with Paclitaxel Liposomes (210mg) and Nedaplatin (100mg). On 16th June, the patient underwent the operation for hysterectomy, omental resection, appendectomy, pelvic lymph removal and enucleation of para-aorta lymph node. Exploratory surgery revealed that no ascites in abdominal was seen, liver, gallbladder, spleen, stomach, intestine and appendix were all normal, uterus was enlarged with its surface irregularity and relative hardness as if the patient were pregnant about 3 months, but the color of it was as normal as ever. Bilateral ovary and oviduct were normal, pelvic lymph node was enlarged and became stiff, military?granule? lesions were found in lymph node of accessory aorta; several scattering granule lesions were found in right side of abdominal peritoneum. Post-operative pathological report showed that endometrial stromal sarcoma (low-grade) was found with infiltration of multifocal tumor tissues around the myometrium; Cervical and vaginal stump and uterus were seen no tumor metastasis; and lateral peritoneal node, pelvic lymph node as well as other nodes were also seen no metastasis. Endometrium was of secretory-phase; chronic cervicitis was seen; right ovary luteum was bleeding; left ovary and bilateral tubal was normal.

After operation, the patient was given one cycle of the chemotherapy with paclitaxel liposome (210mg) and Nedaplatin (100mg) and then discharged. From 23th August, 2006 to 13th January, 2007, the patient received so many treatments as pelvic external radiotherapy(4400cGy), three cycles of combined chemotherapy with Oxaliplatin 150mg d1, VP16(Etoposide, VP-16) 0.1 d1-2, IFO(Ifosfamide, IFO) 2mg d2, Mesna 40mg every 4 hours for 3 times a day and THP 40mg d1. After operation, the patient underwent endocrinotherapy

with medroxyprogesterone and tamoxifen. Her son grows healthily with normal mind. The patient was asked to take chromosome examination without any response.

DISCUSSION

Uterus sarcoma is uncommon, which is about 3% of uterus tumor, while endometrial stromal sarcoma is more uncommon with only 10% of uterus sarcoma. According to the cell shape and amount of karyokinesis, endometrial stromal sarcoma is divided into two types: low-grade malignant one and high-grade malignant one. The clinic feature of the former is that the disease always happens to the young people who are found with irregular bleeding in vagina, rapidly growing tumor, local infiltration, distant metastasis and advanced recurrent. 5-year and 10-year survival rates of the sarcoma are as high as 100%^[2]. At present, the main treatment is hysterectomy and bilateral oviduct hysterectomy, and omental resection, appendectomy when necessary. But it is no good for those unmarried female, especially those with infertility, to undergo the hysterectomy because the operation prevents them from pregnancy. LESS is likely to recur, as one case in Styron's report which revealed that the disease recurred after treatment over 29 years^[3]. Stadvold once reported a myxoid endometrial stromal sarcoma in a 16-year-old nulliparous woman^[4]. And it is reported by Ma Shaokang that three patients with stage IV lung metastases respectively underwent three cycles of pre-operative chemotherapy with DDP, IFO and ADM with result of metastases being completely removed and pelvic tumors lessened. These three patients had continuous chemotherapy for 3 cycles after operation without recurrence during the observation periods of 5 years^[5]. The report showed that chemotherapy is effective for patients with LESS because post-chemotherapy may reduce local recurrence, and it will be better for patient to undergo surgical treatment or chemotherapy combined with anti-estrogen.

The patient mentioned in this paper strongly demanded fertility conservation. She preferred conservative operation to supplementary operation. In consider-

ation of the clinic feature of early blood metastasis and advanced recurrence of low-grade endometrial stromal sarcoma, the patient was given chemotherapy in hope to lessen tumor metastasis and recurrence after operation. The result showed that after 6 cycles of chemotherapy, the patient recovered better.

During cesarean, no abnormal symptoms were seen in the patient's uterus and bilateral oviduct. But after 36 months of operation, the disease recurred with extensive metastasis. The analysis of the case reveals that the unmarried young patient with low-grade endometrial stromal sarcoma should be aware of the risk of fertility conservation in operation and undergo post-operation chemotherapy to delay the recurrence of tumor and to control blood metastasis, and receive hysterectomy and bilateral oviduct hysterectomy to avoid recurrence after delivery. The influence of chemotherapy with DDP, CTX, THP and VCR on bearing quality needs further study with more clinical materials.

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