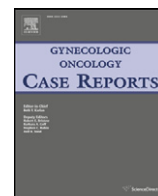


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Complete remission achieved by oophorectomy for recurrent endometrial stromal sarcoma after laparoscopic morcellation



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Introduction

Preoperative differentiation of endometrial stromal sarcoma (ESS) from myoma is challenged by the similarities in their clinical presentations and the unreliability of imaging studies. Often, ESS is diagnosed postoperatively after laparoscopic power morcellation of a presumed myoma (Amant et al., 2014; Park et al., 2011). In approximately 80% of ESS cases diagnosed postoperatively after morcellation, preoperative diagnosis was either leiomyoma or adenomyosis (Park et al., 2011). Laparoscopic morcellation of ESS with an electromechanical morcellator can result in the dissemination of the tumor (Park et al., 2011; Badia and Karini, 2010), but there has been no report of the spread of ESS after morcellation with a single incision on the uterus using a harmonic scalpel. Furthermore, if the ovaries are preserved at the time of the initial surgery, bilateral salpingo-oophorectomy (BSO) for recurrence is recommended (Amant et al., 2014; Yoon et al., 2014), but the effect of BSO for recurrent ESS has not been reported. Here, we present a case where morcellation with a simple incision in total laparoscopic hysterectomy for a presumed myoma led to the peritoneal dissemination of ESS; and complete remission was achieved and laparoscopically confirmed after BSO for recurrent ESS.

Case

A 39-year-old woman (gravidia-2, para-2) was presented with menorrhagia and anemia. Preoperative diagnosis by magnetic resonance imaging was a 5-cm myoma of the uterus and a 4-cm cystadenoma of the right ovary. Intraoperative findings were significant for an 8-cm uterus and a 4-cm cyst of the right ovary, and total laparoscopic hysterectomy and cystectomy of the right ovary were performed. The anterior wall of the uterus was laparoscopically incised with an ultrasonic scalpel (HARMONIC ACE®, Ethicon, United States) to be deformed for vaginal removal. The total weight of the uterus was 190 g. Pathological diagnosis was ESS, formerly referred to as low-grade ESS, and serous cystadenoma of the right ovary. Postoperative thoracic computed tomography (CT) revealed no evidence of lung metastasis; thus, the final diagnosis was ESS stage IA, according to the International Federation of Gynecology and Obstetrics (FIGO) staging for uterine sarcomas (2009).

The woman was followed up by a gynecologic oncologist, with vaginal ultrasound examination every 3 months and abdominal and thoracic CT every 6 months. Twenty months after the initial surgery, routine thoracic/abdominal/pelvic CT indicated the presence of multiple lung metastases (5 mm and 4 mm) and the absence of abdominal and pelvic disease, and she underwent video-assisted thoracic surgery. Pathological examination of the partial pneumonectomy confirmed the lung metastases of ESS, and immunohistochemical examination showed positivity for estrogen receptor and progesterone receptor (Fig. 1). We recommended BSO, to which she agreed after mature consideration, and four months after the thoracic surgery, she underwent laparoscopic exploration and BSO. Intraoperative findings were significant for the disseminations of tumor on the pelvic peritoneum (Fig. 2). Biopsy of the lesions and BSO were performed, but the disseminations persisted. Pathological examination confirmed the peritoneal dissemination of ESS.

Six months after the BSO surgery, laparoscopic exploration for the assessment of castration was performed, which revealed disappearance of peritoneal disseminations (Fig. 3). Thereafter, she has had no evidence of disease for 22 months. She received no hormonal therapy, chemotherapy, or radiotherapy throughout the entire course of her disease.

Discussion

Our case illustrates that a single incision using a harmonic scalpel could potentially lead to ESS dissemination which has been similarly

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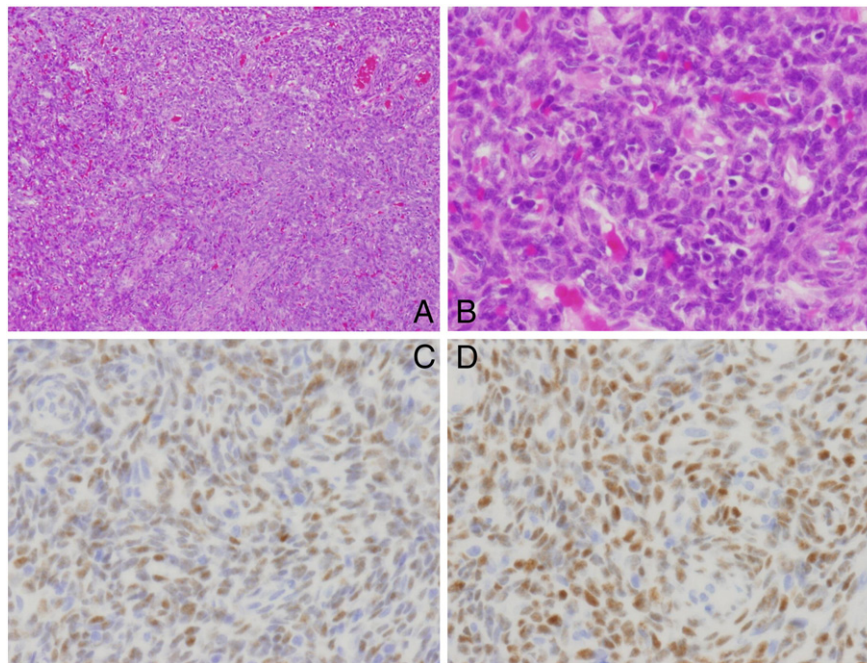


Fig. 1. Microscopic and immunohistochemical analysis of tissue obtained by partial pneumonectomy. A: hematoxylin and eosin stain (40 \times), B: hematoxylin and eosin stain (200 \times), C: estrogen receptor (200 \times), D: progesterone receptor (200 \times).

reported to occur with power morcellation (Park et al., 2011; Badia and Karini, 2010). However, we cannot prove that morcellation was the definite cause of dissemination of ESS in this case, for late recurrences are common in ESS and for the fact that lung metastases were present. Recurrences, which can occur in about one-third of patients with early-stage disease, present most commonly in the abdomen and lungs (Amant et al., 2014). However, considering the median time to recurrence of 65 months in stage I disease (Amant et al., 2014) and the relatively short time to recurrence in our case, morcellation may have resulted in subsequent pelvic dissemination. Park et al. recently reported that tumor morcellation in early-stage ESS was associated with a higher rate of recurrence (31.4%) and a lower rate of 5-year disease-free survival (58%) compared to the counterparts with no morcellation (7.4% and 89%, respectively) (Park et al., 2011). Besides some case reports (Badia and Karini, 2010), most studies do not clearly state whether morcellation was performed with or without an

electromechanical morcellator (Park et al., 2011; Yoon et al., 2014). Our case indicates that dissemination of ESS could result from a simple incision on the uterus without a power morcellator.

Furthermore, our case shows the efficacy of BSO for recurrent ESS. In the setting of initial surgery, hysterectomy is the cornerstone of treatment for ESS, but preservation of the ovaries remains controversial. Ovarian preservation can be considered for young women because it does not affect overall survival (Amant et al., 2014; Yoon et al., 2014; Bai et al., 2014; Amant et al., 2009; Amant et al., 2007), while BSO is associated with longer recurrence-free survival (Amant et al., 2014; Bai et al., 2014; Beck et al., 2012). On the other hand, BSO has gained consensus for recurrent ESS (Amant et al., 2014; Bai et al., 2014). However, there has been no report that actually proves the efficacy of BSO. Our case is the first report that confirmed, by laparoscopic second-look surgery, the complete remission after BSO for recurrent ESS.

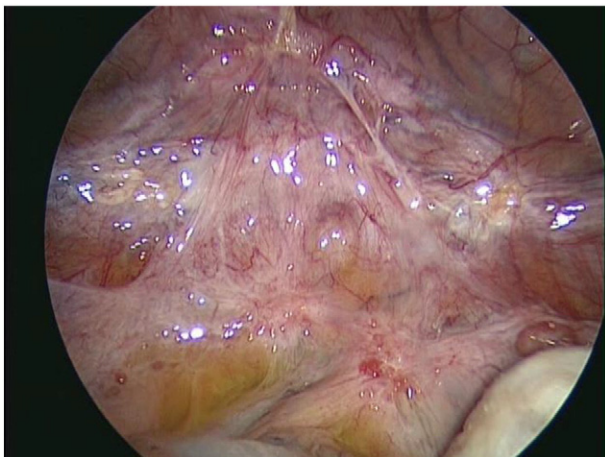


Fig. 2. Pelvic peritoneal disseminations. Occult recurrence was detected during laparoscopic exploration.



Fig. 3. Disappearance of pelvic peritoneal disseminations 6 months after bilateral salpingo-oophorectomy.

Morcellation of ESS should be avoided, but inadvertent morcellation of ESS can occur in the absence of reliable imaging modalities for accurate preoperative diagnosis. Informed consent to the possibility that uterine sarcomas including ESS could be diagnosed after morcellation of presumably benign uterine tumors is important. When ESS is diagnosed after laparoscopic surgery, consultation with gynecologic oncologists is necessary for further observation and treatment. To avoid iatrogenic malignant dissemination, vaginal morcellation in a pouch has been employed (Giovanni et al., 2012). Furthermore, BSO for recurrent disease might be effective in achieving complete remission.

In conclusion, an incision on the uterus using a harmonic scalpel could cause peritoneal dissemination of ESS, and remission might be achieved by BSO for recurrent ESS.

Conflict of interest

Written informed consent for this report was obtained from the patient. The authors have no conflicts of interest to declare.

Acknowledgment

Part of this case has been reported in *J Obstet Gynaecol Res* (Inoue et al., 2014).

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