

EXTRAUTERINE LOW-GRADE ENDOMETRIAL STROMAL SARCOMA

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SUMMARY

Objective: Endometrial stromal sarcoma is a rare cancer that accounts for 0.2% or less of all female genital tract malignancies. We present a case of extrauterine low-grade endometrial stromal sarcoma arising from endometriosis, which was managed by unilateral salpingo-oophorectomy with postoperative high-dose progesterone adjuvant therapy.

Case Report: A 28-year-old nulligravid woman had suffered from progressive abdominal distension accompanied by a palpable firm mass for about 3 months. An abdominal pelvic mass, measuring 13 × 12 × 8 cm, was seen on pelvic sonography and abdominal computed tomography. The CA125 titer was also elevated. Left salpingo-oophorectomy was performed when frozen section examination of the tumor indicated a benign tumor. However, the pathology of the tumor was extrauterine low-grade endometrial stromal sarcoma with extensive endometrioid glandular differentiation arising from endometriosis. The resection margin was also involved. The patient has been receiving high-dose progesterone therapy for 2 months without any adverse effects, except for an increase in body weight of 2 kg.

Conclusion: Low-grade endometrial stromal sarcoma typically has an indolent clinical course and favorable prognosis. Surgical resection is the primary therapeutic approach, and adjuvant therapy with radiotherapy, chemotherapy, or progesterone therapy should be considered for the management of residual or recurrent low-grade endometrial stromal sarcomas. [*Taiwanese J Obstet Gynecol* 2005;44(4):387-390]

Key Words: endometriosis, low-grade endometrial stromal sarcoma, progesterone

Introduction

Endometrial stromal sarcoma is a rare tumor of the uterus and is even rarer outside of the uterus. It accounts for 0.2% or less of all female genital tract malignancies [1]. The majority of extrauterine endometrial stromal sarcomas arise from endometriosis, which suggests a process of malignant transformation [2]. Endometrial stromal sarcomas with less than 10 mitotic figures per 10 high powered fields (HPF) are classified as low grade, and those with 10 or more/10 HPF are classified

as high grade [3]. Here, we present a case of low-grade endometrial stromal sarcoma arising from endometriosis, and managed by unilateral salpingo-oophorectomy with ongoing high-dose progesterone therapy.

Case Report

A 28-year-old nulligravid woman with an unremarkable medical history complained of progressive abdominal distension accompanied by a palpable firm mass of 3 months' duration. She first visited a local hospital where abdominal computed tomography revealed a large solid pelvic mass. She was transferred to our hospital under the impression of ovarian malignancy. On pelvic examination, the mass was found to be firm, non-mobile, non-tender, and engaged at the cul-de-sac. The complex septated mass measured 13 × 12 × 8 cm on pelvic sonography, and had a heterogeneous pattern

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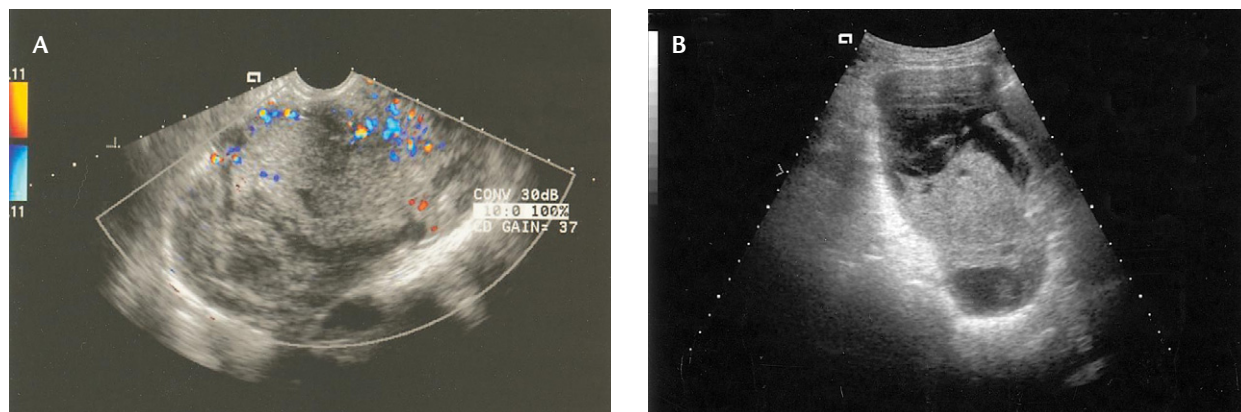


Figure 1. (A) Transvaginal sonography and (B) abdominal sonography of the left adnexa shows a heterogeneous and complex septated mass, measuring about 13 × 12 × 8 cm, with low impedance flow (resistance index, 0.33).

with high density, hypoechoic areas (Figure 1); there was no obvious ascites. Pulsed Doppler examination revealed low-resistance blood flow (resistance index, 0.33). CA125 level was elevated (348 U/mL). Chest X-ray was negative.

The patient underwent exploratory laparotomy via a vertical low abdominal incision. A left-side adnexal mass about 12 cm in diameter was noted. It demonstrated dense adhesion to surrounding organs, including the uterine wall, retroperitoneum, and sigmoid colon. The tumor consisted of flesh-like tissue mixed with tumor hemorrhage. The right ovary was slightly enlarged but grossly normal. The uterus was also slightly enlarged to about 2 months' gestation. A small amount of ascites (50 mL) was noted. The viscera including the liver, small and large intestines, peritoneum, mesocolon, mesentery, and omentum were all grossly free of tumor involvement. A frozen section examination of the tumor indicated that it was benign, so the patient received a left-sided

salpingo-oophorectomy. The pathology of the tumor, however, was extrauterine low-grade endometrial stromal sarcoma with extensive endometrioid glandular differentiation arising from endometriosis. Vascular tumor emboli were seen and the resection margin was involved (Figure 2). The mitotic activity in the spindle cells in the most cellular area was about 2/10 HPF. Both the spindle cell part and glandular part expressed estrogen and progesterone receptors. Ascites cytology tested negative for malignant cells. The patient has been receiving high-dose progesterone therapy for 2 months (medroxyprogesterone 500 mg/day) without any adverse effects, except for an increase in body weight of 2 kg.

Discussion

Endometriosis is one of the most common benign gynecologic conditions and has been estimated to affect

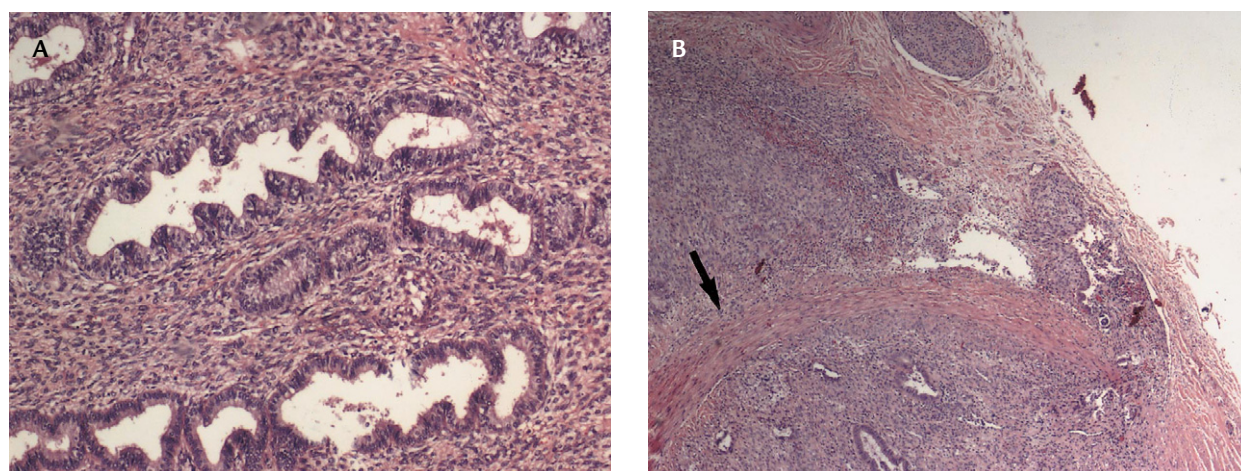


Figure 2. (A) Extensive endometrioid glandular differentiation arising from endometriosis (immunohistochemical [IHC] stain, original magnification × 200). (B) Vascular tumor emboli were seen in the vessels (arrow) (IHC stain, original magnification × 100).

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