



Fertility-sparing management combined with photodynamic therapy for endometrial stromal sarcoma: A case report



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KEYWORDS

Endometrial stromal sarcoma;
Fertility sparing;
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Summary

Background and objectives: Low-grade endometrial sarcoma (LGEES) has a favorable prognosis after standard surgical treatment. The conservative fertility-sparing treatments in young patients with LGEES have been reported; however, the role of conservative therapy is not well defined.

Study design/patient and methods: A 31-year-old nulliparous woman was diagnosed with LGEES after resection of a cervical polyp with resection margin positive for malignancy. She underwent fertility-sparing surgery including laparoscopic pelvic lymph node dissection, hysteroscopic endometrial polypectomy, endocervical curettage, and photodynamic therapy (PDT) on the endometrium and uterine cervix. And she had received adjuvant therapy with a non-steroidal aromatase inhibitor.

Results: She conceived by in vitro fertilization and delivered twins at 32 + 2 weeks gestation by Cesarean section 32 months after conservative treatment. She has no evidence of recurrence after 99 months of follow-up.

Conclusion: Conservative management of LGEES may be attempted in selected patients who want to preserve fertility. Conservative surgery combined with PDT has shown effective results in long-term follow-up.

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Introduction

Endometrial stromal sarcoma (ESS) is a rare entity of uterine malignancy, accounting for 0.2% of all uterine cancers [1]. The standard treatment of ESS consists of surgery with hysterectomy and bilateral salpingo-oophorectomy. Generally, ESS occurs in perimenopausal women [2], and it

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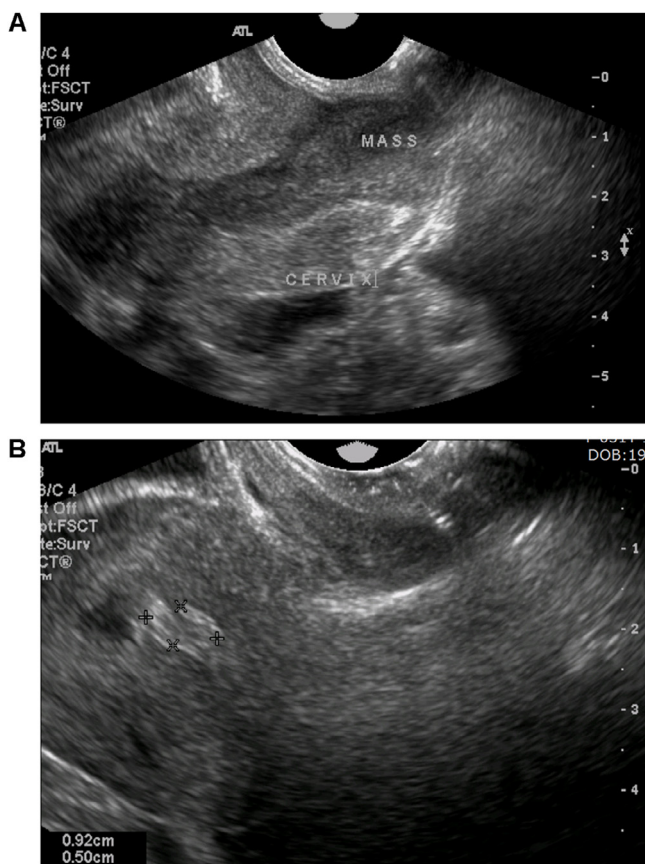


Fig. 1 Transvaginal sonography of the patient. (A) Protruding cervical mass to the upper vagina. (B) 0.9 cm \times 0.5 cm sized endometrial mass.

rarely occurs before the age of 40 years. Standard surgery causes irreversible sterility in patients who have not yet had children. Low-grade (LG) ESS has a favorable prognosis compared to that for other uterine sarcomas, with a 5-year survival rate of 92% [3]. Therefore, the use of conservative fertility-sparing treatments in young patients with LGESS has been reported [4–7]. Here, we present a case of LGESS treated by conservative surgery combined with photodynamic therapy (PDT).

Case

A 31-year-old woman (gravida 0) presented to our institution with 1 year history of menorrhagia. Medical and surgical histories were unremarkable. On pelvic examination, a 3 cm \times 3 cm polypoid mass was detected at the external cervical os that appeared to be either cervical myoma or polyp. No adnexal mass was palpated. Ultrasound examination revealed a 2.9 cm \times 3.1 cm pedunculated cervical polypoid mass and a 0.9 cm \times 0.5 cm endometrial polyp (Fig. 1A and B). The cervical polypoid mass was removed at first visit without any difficulty. The pathologic result of the cervical mass revealed LGESS (2–3 mitoses/10 HPF) with focal extension to the resection margin (Fig. 2). Immunohistochemical analysis showed that the tumor cells were strongly positive for estrogen/progesterone receptor, and CD 10.

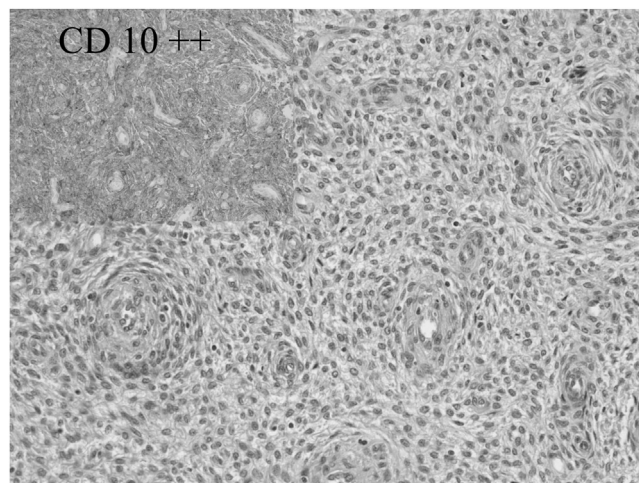


Fig. 2 The main microscopic image shows endometrial stromal sarcoma with hematoxylin–eosin stain (\times 200). Immunostaining for CD10 was strongly positive (\times 200).

Diagnostic imaging studies included abdominal computerized tomography (CT), pelvic magnetic resonance imaging (MRI), chest X-ray, chest CT, and positron emission tomography (PET)-CT were performed. Pelvic MRI confirmed a small endoluminal nodule at the endometrial cavity and a 1.6 cm left obturator pelvic lymph node enlargement. There was no evidence of intra-peritoneal disease or metastases. Other imaging studies were unremarkable. Laboratory tests showed normal levels of serum tumor markers (CA 125, CA 19-9 and CEA).

After discussions with the patient and her family, conservative management was planned because of the patient's strong desire to preserve fertility and nulligravidity. After confirming that the pelvic lymph nodes harvested by laparoscopic lymph node dissection were negative for malignancy including enlarged left obturator node on pelvic MRI, the patient underwent hysteroscopic endometrial polypectomy, endocervical curettage, and PDT on the endometrium and cervical canal. The PDT procedures for endometrial cancer have been described previously [8]. Histological examination of the endometrial polyp was also revealed LGESS. No remaining malignancy was detected in other specimens. Because of the risk of recurrence, conservative surgery was followed by adjuvant therapy with a non-steroidal aromatase inhibitor (letrozole 2.5 mg, by mouth every day) for 6 months. In consecutive clinical examinations and radiographic studies, the patient has no evidence of recurrence for 99 months.

After 32 months, a twin pregnancy was achieved by in vitro fertilization (IVF) because of male factor combined with female factor infertility. Preterm delivery occurred at 32+2 weeks of gestation by Cesarean section, and the patient gave birth to twin neonates (two boys weighing 2075 and 1755 g). The placental pathology during delivery showed no evidence of tumor.

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