Adenosarcoma of the uterine body initially presenting as an interstitial small tumor of the uterus: a case report

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Summary

Adenosarcoma of the uterine body is a rare mixed tumor in which a benign epithelial component is mixed with a malignant stromal element. It has been considered that this tumor originates from the endometrium and its most common finding of imaging is a polypoid tumor occupying the uterine cavity. The authors herein present a case of 37-year-old female with a complaint of abnormal vaginal bleeding. At the first visit, transvaginal ultrasound and magnetic resonance imaging (MRI) showed a round mass with a diameter of one cm in the uterine wall. No malignant pathological finding was detected. The patient visited the authors again one year later, because of continuous bleeding. At that time, they found a polypoid tumor in the uterine cavity, which turned out to be adenosarcoma with sarcomatous overgrowth. The round mass in the uterus detected at first time seems to have been incipience of adenosarcoma. Prodromal sign of adenosarcoma has not been reported previously.

Key words: Adenosarcoma; Diagnosis; Magnetic resonance imaging; Prodromal sign; Ultrasound.

Introduction

Adenosarcoma of the uterus is a rare mixed tumor in which a benign epithelial component is mixed with a malignant stromal element. This tumor has been considered to be present as a solid polypoid mass usually arising in the endometrium [1-4]. Therefore, it is sometimes misdiagnosed as a benign endometrial polyp, which can lead to a delay of the accurate diagnosis. The authors experienced a case presenting with an interstitial small tumor of the uterus which finally turned out to be adenosarcoma. This tumor seemed to be the prodromal stage of uterine adenosarcoma.

Case Report

A 37-year-old gravida 4 para 1 female visited a physician with a complaint of abnormal vaginal bleeding and hypermenorrhea for couple of years. She had had hormonal therapy of dysmenorrhea. She had not had any other past medical history, such as tamoxifen use, pelvic radiation, and so on. She had no family history of malignancy and gynecologic disease. Transvaginal ultrasound showed a hyperechoic round shadow with a diameter of one cm at uterine fundus muscle (Figure 1). To diagnose the mass and the cause of abnormal vaginal bleeding, magnetic resonance imaging (MRI) were performed. It revealed a mass with low intensity on T1-weighted images and high intensity on T2-weighted images compared with the myometrium (Figure 2) and diagnosed as adenomyosis. She visited this hospital for further examination.

Her cervical and endometrial biopsies were negative. The authors therefore diagnosed the tumor as benign, and proposed that the patient continue to submit to personal observation, and to take a low-dose oral contraceptive to control the irregular bleeding and hypermenorrhea. Although she continued taking the oral contraceptive for about one year, her symptoms did not improve. Then, she was examined again in detail.

Revised manuscript accepted for publication October 10, 2013

Eur. J. Gynaec. Oncol. - ISSN: 0392-2936
XXXV, n. 4, 2014
doi: 10.12892/ejgo24722014

At that time, transvaginal ultrasound showed an irregularly-shaped mass measuring three cm in diameter in the endometrial cavity (Figure 3). MRI demonstrated this mass to be a polypoid tumor in the uterine cavity. The mass contained solid components with low intensity on T1-weighted images and high intensity on T2-weighted images compared to the myometrium and areas of small cysts at the lower pole. The polypoid tumor seemed to be continuous with the previously detected fundus mass (Figure 4). It was not typical as adenomyosis or endometrial polyp. FDG-PET/CT was positive for the mass in the uterine cavity (SUV max = 6.4), suggesting that the mass could be malignant. The patient's tumor markers: LDH, CA125, CA19-9, CEA, SCC, and NSE were all within normal limits.

To diagnose the polypoid tumor, the authors performed hysteroscopy. Hysteroscopic examination showed that there was a dark red polypoid mass arising from the fundus of the endometrium. They resected this polypoid mass and a pathological examination was performed.

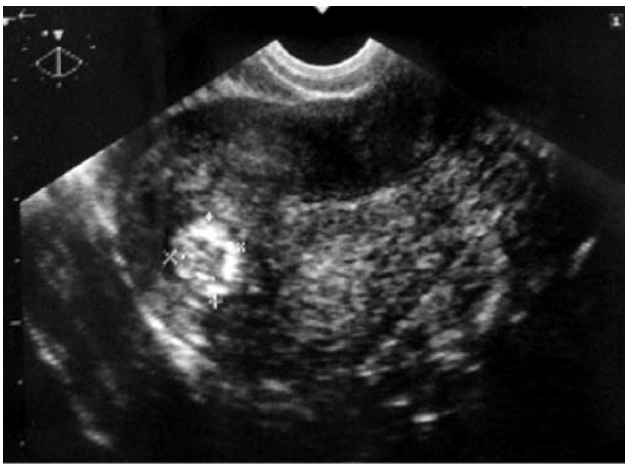

Figure 1. — Transvaginal ultrasound showing a hyperechoic round shadow measuring one cm in diameter in the uterine fundus wall.

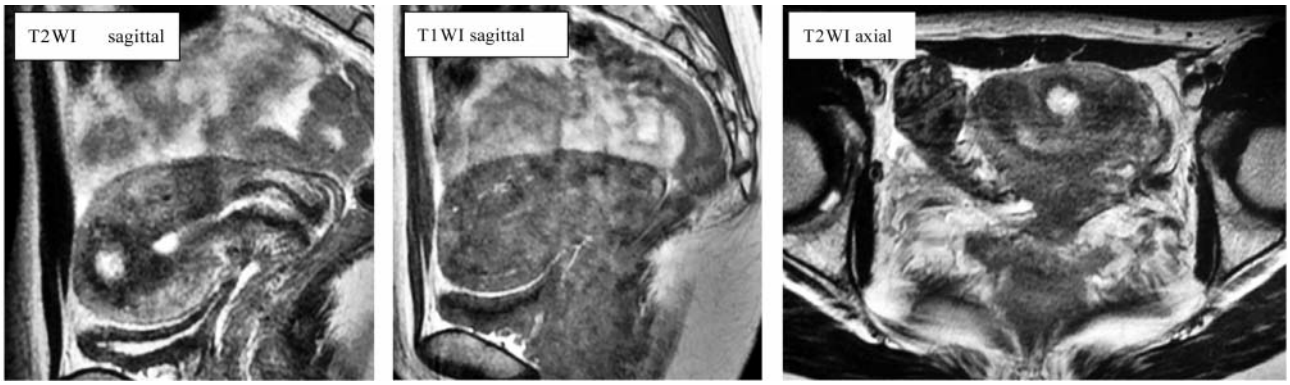

Figure 2. — Magnetic resonance imaging revealing a mass with low intensity on T1-weighted images and high intensity on T2-weighted images compared to the myometrium.

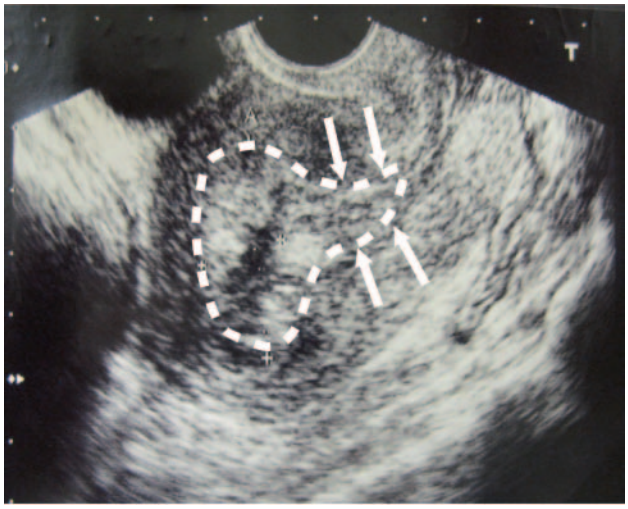

Figure 3. — Transvaginal ultrasound showing an irregularly-shaped mass with a diameter of three cm from the fundus (indicated by a white dotted line). The mass extending to the intrauterine cavities is indicated by white arrows.

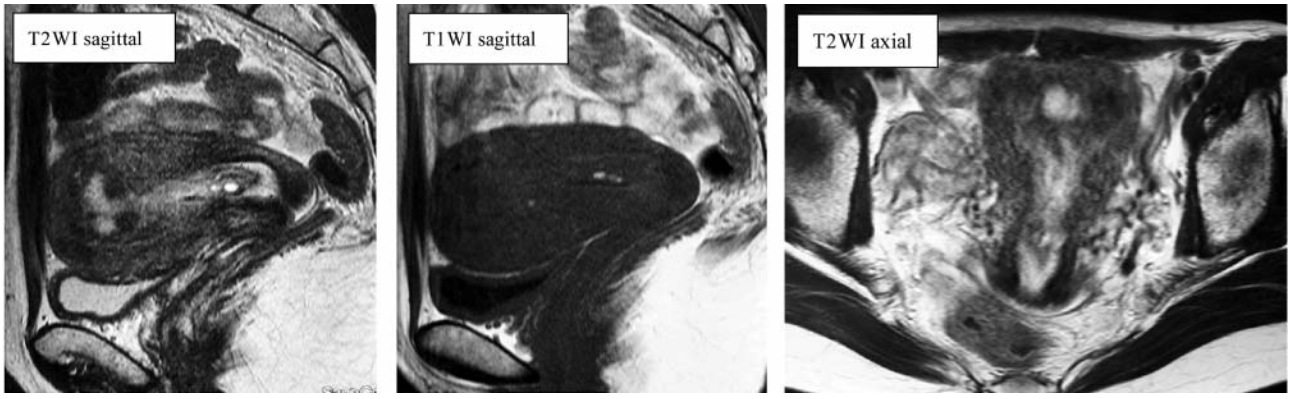

Figure 4. — Magnetic resonance imaging demonstrating a mass to be a polypoid tumor in the intrauterine cavity. The tumor has small cysts at the lower pole, and seems to be continuous with the previously detected fundus tumor.

Microscopically, this mass had irregularly enlarged glandular epithelial components, lined by epithelium of the endometrial type. These glands were surrounded by low-grade sarcomatous stroma. The tissue was positive for MIB-1 and CD10 staining (Figure 5). These findings suggested that this polypoid mass was an adenosarcoma or endometrial stromal sarcoma. A simple total abdominal hysterectomy and bilateral salpingo-oophorectomy were performed. A small remnant of the polypoid mass was seen in the left corner of the uterine cavity (Figure 6).

Microscopically, there was adenomyosis, benign glandular epithelial elements surrounded by low-grade sarcomatous stroma, and pure sarcomatous components. The pure sarcomatous compo-

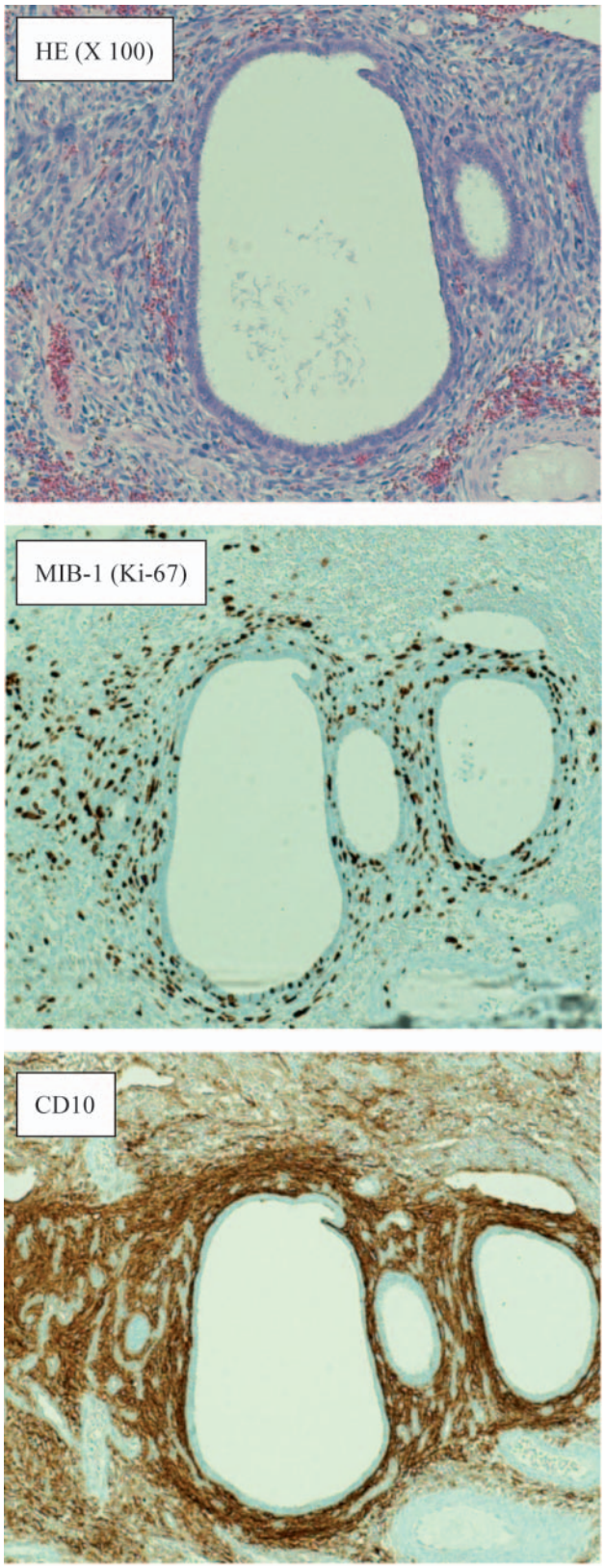

Figure 5. — Microscopically, this mass has irregularly enlarged glandular epithelial components, lined by epithelium of the endometrial type. These glands are surrounded by low-grade sarcomatous stroma. The tissue was positive for MIB-1 and CD10 staining.

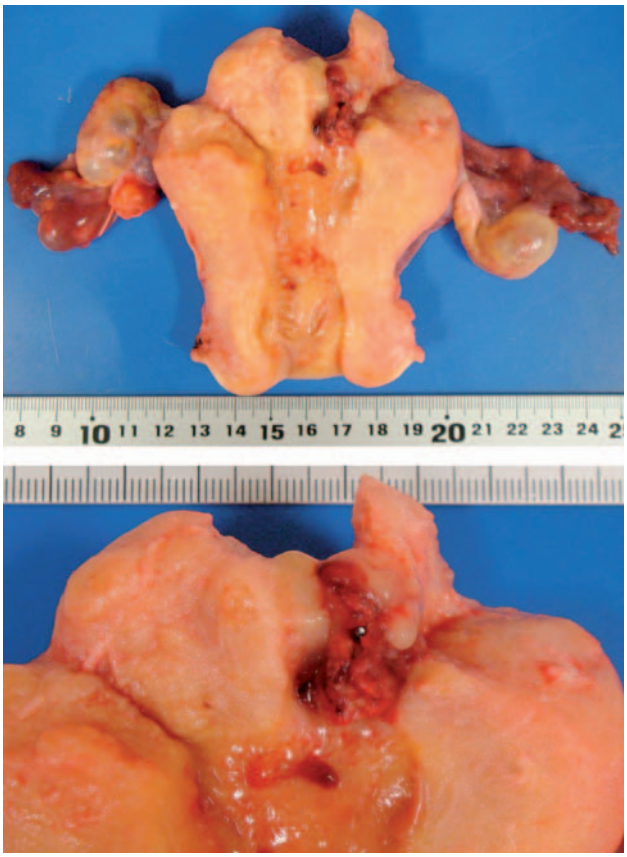

Figure 6. — A simple total abdominal hysterectomy and bilateral salpingo-oophorectomy were performed. A small remnant of the polypoid mass is seen in the left corner of the uterine cavity.

nent comprised more than 25% of the tumor. Therefore, the authors diagnosed this tumor to be adenosarcoma of the uterine body with sarcomatous overgrowth. Tumor invasion was revealed in about one-third depth of myometrium. After surgery, adjuvant therapy was not administered. The patient has been alive with no evidence of disease for about 48 months after the surgery.

Discussion

Adenosarcoma of the uterine body is a rare mixed tumor in which a benign epithelial component is mixed with a malignant stromal element. The tumor usually affects postmenopausal patients, and its most common symptom is genital bleeding. Several papers have reported that adenosarcoma usually originates in the endometrium and grows as a polypoid mass within the endometrial cavity [1-4].

In the present case, the small round tumor in the uterine fundus muscle seemed to turn into a polypoid tumor, which was found to be adenosarcoma. There have been no previous reports of prodromal signs of adenosarcoma. As this case shows, the incipience of adenosarcoma may possibly be detected as a hyperechoic mass by ultrasound and high intensity on T2-weighted images by MRI. Takeuchi M *et al.* [4]

reported that the presence of small hyperintense cysts scattered within the mass on T2-weighted imaging, reflecting glandular epithelial components, may be a characteristic finding of adeno- sarcoma. Therefore, when examining a patient who complains of genital bleeding and has a uterine intramuscular tumor as seen in this case, adenosarcoma should be considered when making at differential diagnosis.

Adenosarcomas of the uterus with sarcomatous overgrowth are aggressive tumors frequently associated with postoperative recurrence or metastases and a fatal outcome [1]. The authors will continue to follow this patient carefully.

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