Adenomyosis with extensive glandular proliferation simulating infiltrating malignancy on magnetic resonance imaging

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Abstract
We report a case of multicystic adenomyosis, which is an exceedingly rare benign tumor. The patient complained of an irregular menstrual cycle and abnormal genital bleeding that gradually increased in amount and frequency. The patient finally became severely anemic, and a hysterectomy was therefore performed. T2-weighted magnetic resonance imaging (MRI) indicated hyperplasia of the endometrium, with a myometrial lesion, where a high signal intensity multicystic mass was observed. The preoperative diagnosis was complicated by confusing MRI results. Postoperative macroscopic examination revealed a villous endometrium and a myometrium thickened with multiple small cysts containing serous transparent fluid. The final diagnosis, based on the hysterectomy specimen, was adenomyosis coexisting with simple endometrial hyperplasia. The MRI and positron emission tomography images are presented.

Key words
Adenomyosis · Multicystic adenomyosis · Endometrial hyperplasia · MRI · PET

Introduction
Adenomyosis is a common uterine disease characterized by intramyometrial invasion of basal endometrial glands associated with hyperplasia of smooth muscle. Magnetic resonance imaging (MRI), which provides high-resolution imaging in the pelvis, shows typical adenomyosis as thickening of the junctional zone forming an ill-defined area of low signal intensity, with occasionally embedded bright foci in T2-weighted images. However, the appearance of adenomyosis on MRI varies widely with the growth pattern, and the images may mimic those of uterine myomas, uterine malignancy, or ovarian cancer.

We report here a case of adenomyosis with a multicystic appearance, with simple endometrial hyperplasia. Unlike common cystic adenomyosis, the adenomyosis cysts contained serous, rather than bloody, fluid. Preoperative MRI examinations suggested an endometrial stromal sarcoma (ESS) based on the suggestion of myometrial invasion from the endometrial lesion. This report describes rarely observed MRI results from a patient with adenomyosis with a multicystic appearance.

Case report
The patient was a 40-year-old woman who first visited our hospital reporting an irregular menstrual cycle. Pelvic examination revealed a fist-sized, enlarged uterus and swelling of the left ovary. MRI of the pelvis confirmed a slightly enlarged uterus with a multicystic tumor in the posterior uterine wall and a serous cyst in the left ovary. We recommended further examinations, but the patient chose to visit another hospital for a second opinion.
One year after her first visit, the patient visited our hospital again complaining of abnormal genital bleeding. Cytological examination revealed no evidence of malignant tumors in the uterine cervix or uterine endometrium. T1-weighted MRI showed an enlarged uterine body and a multicystic mass isointense with the myometrium and endometrium (Fig. 1A). T2-weighted MRI revealed the mass to be hyperintense and extending to the myometrium (Fig. 1B). T1-weighted enhanced MRI showed the cyst wall to have similar enhancement to the surrounding myometrium (Fig. 1C). Diffusion-weighted imaging with $b = 1000 \text{ s/mm}^2$ showed a marked signal increase throughout the tumor, suggestive of a malignant uterine tumor (Fig. 1D). The serum tumor marker cancer antigen (CA) 125 level was high at 161.5 U/ml (normal <35 U/ml), and the carcinoembryonic antigen (CEA) level was 1.9 ng/ml (normal <6.0 ng/ml), CA19-9 was 25.6 U/ml (normal <37.0 U/ml), and lactate dehydrogenase was 145 U/ml (normal 124–226 U/ml; isozyme: within normal limits).

Malignant tumors could not be excluded on the basis of the MR images, and enucleation of the myometrial and endometrial lesion was likely to be difficult because of the lack of a clear margin separating it from the surrounding myometrium and endometrium. The abnormal uterine bleeding worsened. The bleeding might have been caused by the uterine tumors and would be difficult to cure by drug therapy. We therefore recommended hysterectomy. However, the patient did not return until 3 months later when she experienced substantial genital bleeding and had become severely anemic (hemoglobin 6.4 g/dl, hematocrit 21.9%).

A third MRI scan was performed, which showed the posterior uterine wall and endometrial thickness to be further increased. A multicystic lesion was observed with no clear distinction from an endometrial proliferative lesion. $^{18}$F-fluorodeoxyglucose-positron emission tomography and computed tomography (FDG-PET/CT) localized the abnormality to the endometrium and the vaginal cavity, and revealed moderately increased FDG uptake in the posterior wall of the uterus. The standardized uptake value (SUV) was 3.662, which gradually increased to 4.324 at the late phase (Fig. 2).

A hysterectomy with bilateral salpingo-oophorectomy and biopsy of the pelvic lymph nodes was performed to distinguish ESS from adenomyosis on the basis of the unclear borderlines of the high-intensity mass revealed by T2-weighted MRI and rapid histological examination. The results were unable to exclude malignancy.

In the hysterectomy specimen, the uterine volume was $125 \times 120 \times 70$ mm. The posterior uterine endometrium