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Case Report

Magnetic resonance imaging of adenoma malignum of the uterine cervix with pathologic correlation: a case report

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ABSTRACT

Adenoma malignum (AM) is considered a rare subtype of cervical adenocarcinoma. Although previous reports have described magnetic resonance findings, none of these reports evaluated the utility of diffusion-weighted imaging in the differential diagnosis of AM and other multicystic cervical lesions. We present a case report of an AM that did not show restriction on the apparent diffusion coefficient map, which can be explained by the low cellularity of the tumor. This is consistent with the proper correlation between the diffusion imaging and histopathology of the tumor. In this way, AM can present with high apparent diffusion coefficient values, as in benign cervical lesions. Therefore, the combination of a solid multicystic lesion that invades the cervical stroma on T2-weighted magnetic resonance images and the absence of restriction on the apparent diffusion coefficient map are very suggestive of AM.

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Introduction

Adenoma malignum (AM), also termed minimal-deviation adenocarcinoma, is considered a rare form of mucinous adenocarcinoma of the cervix. AM was first described and named by German gynecologist Gusserow in 1870 [1]. In 1975, Silverberg and Hurt et al. [2] considered its pathologic form to be a type of well-differentiated cancer, and thus suggested the term “minimal-deviation adenocarcinoma.” In 2003, the World Health Organization further described the neoplasm as a “very well-differentiated endocervical-type mucinous

adenocarcinoma composed mostly of lobular endocervical glandular hyperplasia-looking glands but with the characteristics of invasive adenocarcinoma” [3].

Very few studies have described the imaging characteristics of AM, and there is only one study on the utility of diffusion-weighted imaging (DWI) in the diagnosis of AM [4].

The purposes of presenting this case were to: (1) illustrate magnetic resonance imaging (MRI) findings of AM with a focus on DWI; (2) discuss the differential diagnosis; and (3) correlate radiological and pathologic findings.

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Case report

A 37-year-old woman presented with low abdominal pain and vaginal bleeding. The patient had sporadic Peutz–Jeghers syndrome with de novo mutation. Her medical history included intestinal hamartomatous polyposis, small bowel resection that was performed for intussusception, left breast lumpectomy for intraductal papilloma, and bilateral oophorectomy for ovarian sex cord tumor with secondary iatrogenic premature ovarian failure and subsequent treatment with hormone replacement therapy.

Two months before the initial diagnosis, the patient presented with metrorrhagia and profuse, watery vaginal discharge initially thought to be secondary to hormone therapy.

Gynecologic examination revealed that the cervix was enlarged, but the mucosa was normal in appearance.

Transvaginal ultrasound identified a poorly vascularized and heterogeneous submucosal lesion in the cervix and fluid accumulation in the endometrial cavity secondary to cervical stenosis produced by the lesion (Fig. 1).

Cytology from endocervical canal curettage was nondiagnostic.

Pelvic MRI revealed a distended endometrial cavity with fluid accumulation (Fig. 2A) secondary to diffuse thickening of the cervical stroma with disappearance of the normal low-signal intensity on T2-weighted sequence. These changes were secondary to a 65 × 32 × 30-mm cervical mass showing isointensity on T1-weighted sequence (Fig. 2B) and heterogeneous hyperintensity on T2-weighted images (Figs 2C–E). The lesion was composed of multiple cervical microcysts (>20) and a solid component. Cysts were small in size, measuring up to 5 mm in diameter and with smooth walls. The solid component infiltrated deeply into the cervical stroma on the right side, and infiltration was less evident on the left side suggesting right parametrial extension and probably left parametrial involvement (Figs 2C and E). The solid portion also invaded the lower third of uterine body, uterine isthmus, and superior and anterior third of the vagina (Fig. 2A). On contrast-enhanced T1-weighted images, the lesion was hyperintense (Fig. 2F).

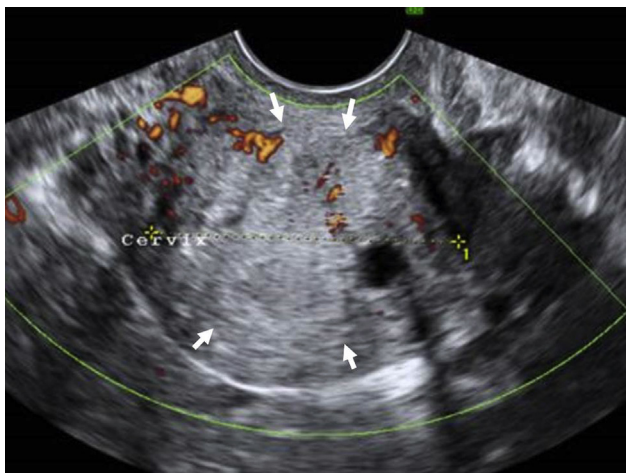


Fig. 1 – Transvaginal ultrasonography demonstrates a poorly vascularized and heterogeneous submucosal lesion in uterine cervix (arrows).

On DWI, the solid and cystic components of the tumor were hyperintense on the high-*b*-value images (*b*, 800; Figs 3A and B) but showed no restriction on the apparent diffusion coefficient (ADC) map (Fig. 3C).

Despite the absence of restriction on the ADC map, the presence of mixed cystic and solid lesion with stromal invasion in a patient with Peutz–Jeghers syndrome was highly suggestive of malignancy, particularly of AM.

Preoperative deep biopsy showed proliferation of well-differentiated endocervical glands with mucinous metaplastic changes, showing very little cellular atypia and low mitotic activity. Deep extension of these aberrant glands was associated with a desmoplastic stroma. Immunohistochemical study demonstrated focal but intense positivity for carcinoembryonic antigen (CEA). The overall appearance was considered highly suspicious of AM. The patient underwent a modified radical hysterectomy with pelvic lymphadenectomy and adjuvant chemotherapy and radiotherapy. Final surgical pathology confirmed the diagnosis of AM (Fig. 4). Histologically, the cervical mass showed tumor extension to the parametrium, vagina and uterine body, and vascular and perineural invasion. Microscopic metastases in the pelvic lymph nodes were found bilaterally.

Vaginal cuff recurrence and nodal and peritoneal dissemination were found seven months after the initial diagnosis.

Discussion

AM is a rare carcinoma of the cervix with an estimated prevalence of 3% of all cervical adenocarcinomas and of 0.3% of all cervical carcinomas of the uterus [5]. The prognosis of AM is poor with lower survival rates than conventional adenocarcinoma because dissemination into the peritoneal cavity and distant metastasis occurs during the early stage of the disease. AM is associated with Peutz–Jeghers syndrome, ovarian mucinous tumor, and ovarian sex cord tumor.

Clinically, the most characteristic symptom is a watery vaginal discharge, but this is nonspecific and may be the main complaint of patients with benign cystic uterine lesions. Furthermore, in early stages of the disease, the gynecological examination may be normal.

The detection rate of AM by cytologic evaluation is only around 32% [6]. Because the tumor is located deep in the endocervix and exhibits an endophytic growth pattern, it is difficult to make an accurate cytological diagnosis by a routine Papanicolaou test.

Therefore, imaging techniques and particularly MRI play an important role in the preoperative diagnosis and evaluation of AM. MRI shows the most detailed features and correlates them with histologic findings. At MRI, AM is seen as a multicystic lesion with solid component invading the cervical stroma and marked hyperintensity on T2-weighted images [7]. On contrast-enhanced images, there might be enhancement of the solid component, as in cervical carcinomas, including adenocarcinomas.

AM features on DWI have been previously described by only one paper [4]. In contrast to that paper [4], our case did not show diffusion restriction on the ADC map despite having a solid tumor component showing hyperintensity on DWI b

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