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Case Report

A case of uterine adenomyoma of endocervical type that was suspected as minimal deviation adenocarcinoma on biopsy, MRI findings, and literature review[☆]

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ABSTRACT

Uterine adenomyomas of endocervical type are rare benign tumors of the uterine cervix commonly presented as cyst-like, dilated glandular structures within polypoid masses. A premenopausal woman in her 50s was referred to our hospital because of an increasing watery vaginal discharge. A multifocal cyst measuring 5 × 4.5 cm in size projecting into the endocervical canal was revealed on a contrast-enhanced MRI. The fluid within the tumor showed a hypointense signal on T1-weighted imaging (T1WI) and a hyperintense signal on T2-weighted imaging (T2WI). On T2WI, most of the septa within the tumor showed a slightly hyperintense to hypointense signal, whereas some areas revealed a strong hypointense signal; the contrast effect on the septum was satisfactory. On the T2WI taken 2 years previously, the tumor was a 4.5 × 3.5 cm polypoid mass protruding from the posterior endocervical wall. Contrastingly, the current T2WI showed that the stem was no longer identifiable because of tumor growth. Because previous imaging showed that the tumor was a stalked tumor protruding from the posterior endocervical wall, the imaging diagnosis was uterine adenomyoma of the endocervical type. A biopsy suggested the possibility of a minimal deviation adenocarcinoma (MDA). Hence, a total hysterectomy was performed. The final diagnosis confirmed the uterine adenomyoma of endocervical type. Uterine adenomyoma of the endocervical type might be difficult to differentiate from MDA in small biopsy speci-

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mens; therefore, evaluation of morphology by MRI is considered important in preoperative diagnosis.

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Introduction

Uterine adenomyomas of endocervical type are rare benign tumors that commonly present as polypoid masses [1] and are often misdiagnosed as minimal deviation adenocarcinoma (MDA) in pathology [1,2]. We report a case of magnetic resonance imaging-diagnosed uterine adenomyoma of endocervical type that was suspected to be a minimal deviation adenocarcinoma (MDA) on biopsy.

Case report

A premenopausal woman in her 50s was referred to our hospital because of an increasing watery vaginal discharge. An MRI was performed 2 years earlier when the patient visited our hospital with the same chief complaint. In addition, she had a history of abnormal vaginal bleeding caused by a submucosal uterine myoma 11 years ago and had undergone uterine artery embolization (UAE) at our hospital. She has ulcerative colitis which is currently under control or treatment or she is receiving medication for. Blood and biochemical tests, including those for serum tumor markers, revealed no abnormalities.

A contrast-enhanced MRI was performed (Fig. 1). The tumor was a multifocal cyst sized 5 × 4.5 cm with a clear border projecting into the endocervical canal. Multiple cysts of up to 1 cm in diameter were observed within the tumor. The content fluid demonstrated hypointense and hyperintense signals on T1-weighted and T2-weighted imaging, respectively. On T2-weighted imaging (T2WI), most of the septa within the tumor showed a slightly hyperintense to hypointense signal, whereas some areas showed a strong hypointense signal. No hyperintense signal was observed in the septa on diffusion-weighted imaging (DWI); the contrast effect on the septum was satisfactory. T2WI images obtained immediately before UAE 11 years ago, 2 years after UAE (9 years ago), and at the last visit 2 years ago are shown (Fig. 2). Since the tumor was a polypoid mass of 3.5 × 1.5 cm, protruded from the posterior endocervical wall and exhibited a hypointense signal on T2WI 11 years ago, the patient was diagnosed with a submucosal uterine myoma (Fig. 2A). The patient had excessive menstrual cycles, and at the time of the visit, she had persistent abnormal vaginal bleeding and worsening anemia. Although submucosal uterine myoma was not a good indication for UAE, after consulting the obstetrician, UAE was performed to control the bleeding. After UAE, abnormal vaginal bleeding improved. The tumor shrunk after UAE but subsequently re-enlarged with multiple cysts inside (Fig. 2B). The T2WI 2 years ago showed that the tumor had a polypoid morphology with multiple cysts and a stem from the tumor to the posterior en-

docervical wall (Fig. 2C). Therefore, it was diagnosed as a uterine adenomyoma of endocervical type on MRI, and the patient was monitored. However, on the current T2WI, the stem was no longer identifiable because of tumor growth (Fig. 1B). Because previous imaging showed that the tumor was a stalked tumor protruding from the posterior endocervical wall, the imaging diagnosis was uterine adenomyoma of the endocervical type.

Curette biopsy suggested lobular endocervical hyperplasia (LEGH) or MDA. Therefore, a cervical conization and an additional biopsy were performed under direct vision. Given that the preoperative pathology indicated MDA, as evidenced by the image showing poorly atypical glands invading the stroma (Fig. 3), a total hysterectomy was performed.

Macroscopically, the tumor appeared as a polypoid mass protruding from the posterior wall of the uterine cervix (Fig. 4A), which was 4.5 cm in length and contained multiple cystic structures. Microscopic images showed numerous gland proliferations in the stroma, which were mainly smooth muscle (Fig. 4B). The glands varied in size and morphology, including round, oval, lobulated, and dendritic. The markedly dilated glands exhibited a cyst-like morphology (Fig. 4C). Some stromal areas, mostly smooth muscle or fibrous tissue, had sparse glands (Fig. 4D). The glands were lined with mucinous epithelium without nuclear atypia. There was no invasion into the surrounding stroma or desmoplastic stromal reaction. Thus, the final diagnosis was uterine adenomyoma of endocervical type.

Discussion

The classical histopathological features of uterine adenomyomas of endocervical type are well-circumscribed lesions composed of large glands lined with a single layer of benign endocervical mucinous epithelium [1,2]. Histologically, its differentiation from MDA is often challenging. Notably, the absence of nuclear atypia, glandular invasion, and desmoplastic response in the stroma, rich in smooth muscle, are key points for differentiation. The pathological diagnosis in this biopsy was suspected MDA, but it was deemed difficult to accurately diagnose this lesion with biopsy specimens that could only be viewed to a limited extent.

We performed a literature review using PubMed and searched papers using the English keywords “adenomyoma of endocervical type” and “polypoid endocervical adenomyoma.” A total of 26 cases were selected [1–8]. The ages of the patients ranged from 21 to 56 years (mean, 42 years). The most common symptoms are asymptomatic and abnormal vaginal bleeding; 4 patients presented with mucoid vaginal discharge [1,2,4], and one had a watery vaginal discharge [3]. The most

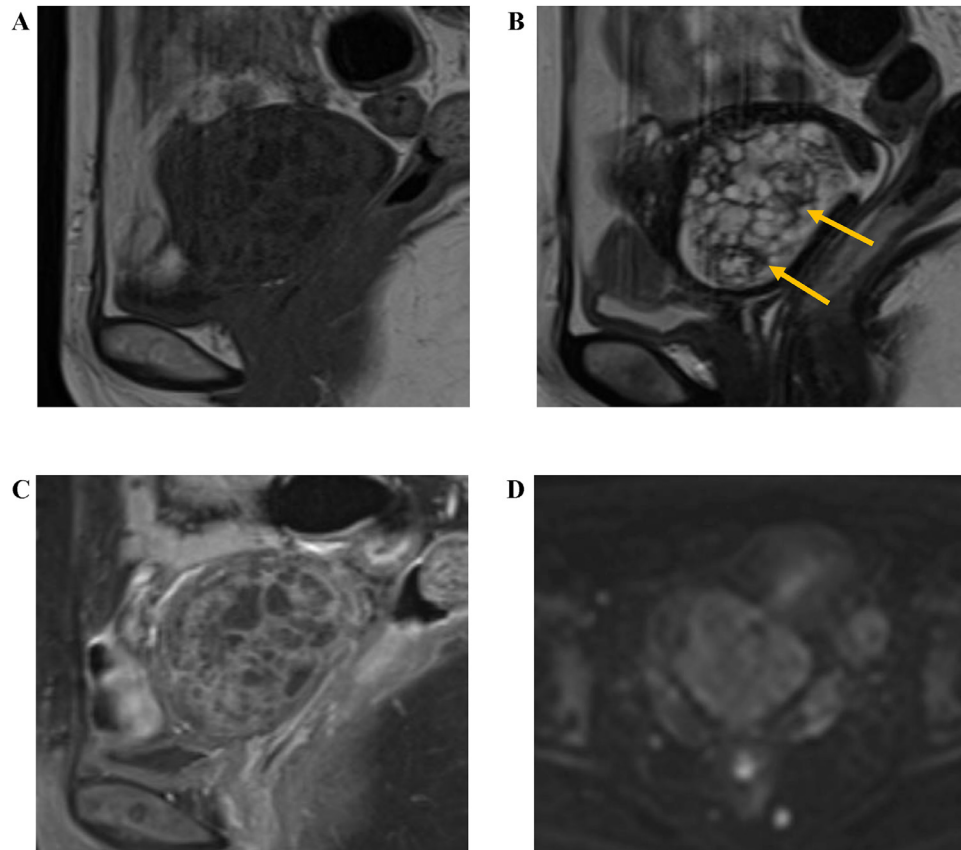


Fig. 1 – MRI of the uterine cervical tumor (A) sagittal T1WI, (B) sagittal T2WI, (C) sagittal contrast-enhanced image, (D) DWI. The tumor measures 5 x 4.5 cm in size and contains multiple cysts. The fluid content appears as a hypointense signal on T1WI and a hyperintense signal on T2WI. On T2WI, most of the septa within the tumor show a slightly hyperintense to slightly hypointense signal; however, some areas show a strong hypointense signal (arrow). The contrast effect on the septum is satisfactory. There is no hyperintense signal in the septum on DWI.

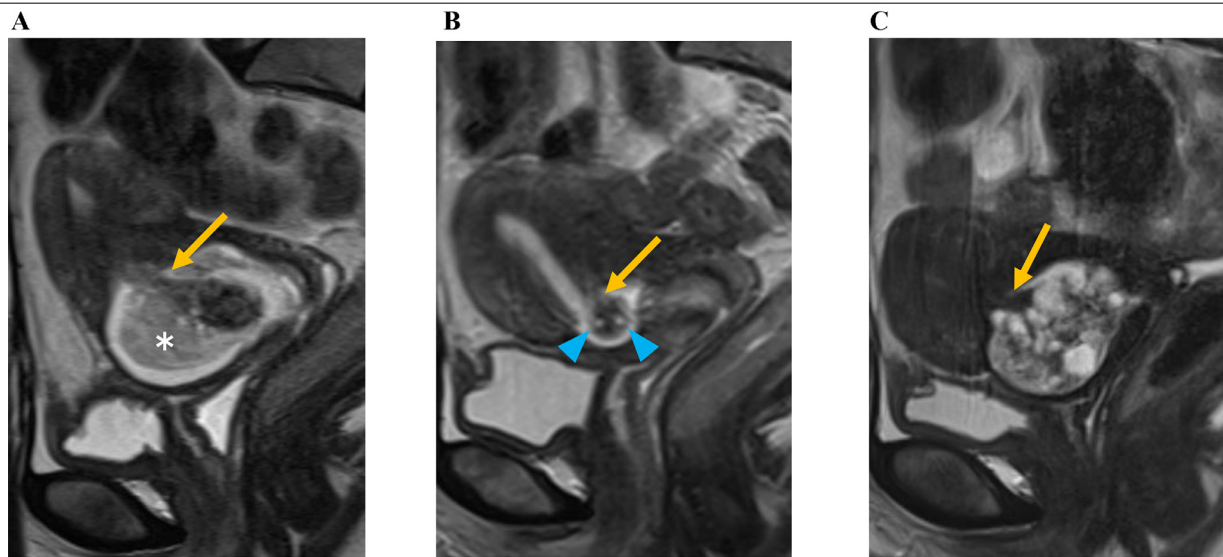


Fig. 2 – Previous sagittal T2WI of the tumor in the uterine cervix. (A) immediately before UAE (11 years ago), (B) 2 years after UAE (9 years ago), (C) 2 years ago. The tumor is a polypoid with a stem from the posterior endocervical wall (arrow). T2WI prior to UAE shows a slightly hyperintense signal area around the tumor (asterisk). Contrast-enhanced image shows no contrast effect in the same area (unshown). Therefore, the area is considered a hematoma. The tumor shrunk from 3.5 x 1.0 cm in size once after UAE but subsequently re-enlarged to 4.5 x 3.5 cm in size. The multiple cysts inside the tumor appear after UAE (arrowhead) and then increase with tumor growth.

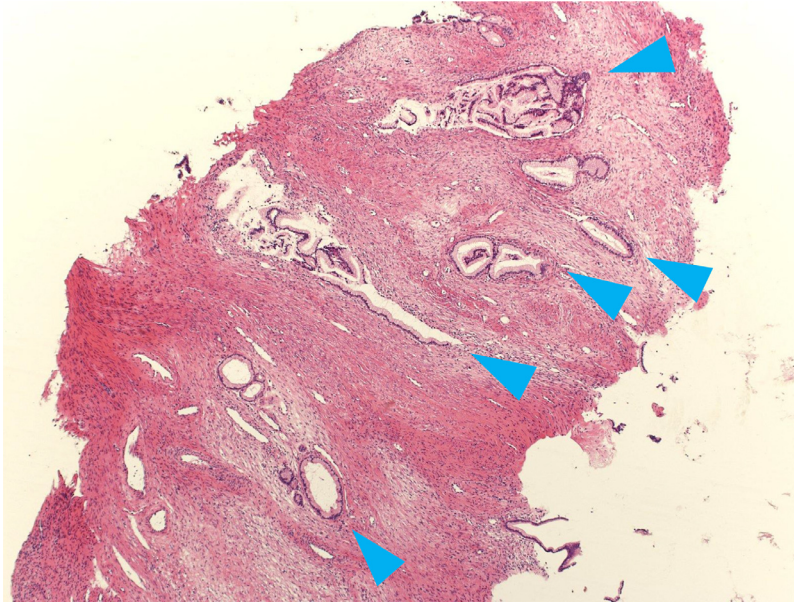


Fig. 3 – Direct visual biopsy specimen. Atypia of glandular structure is scant, but stromal infiltration is suspected (arrowhead).

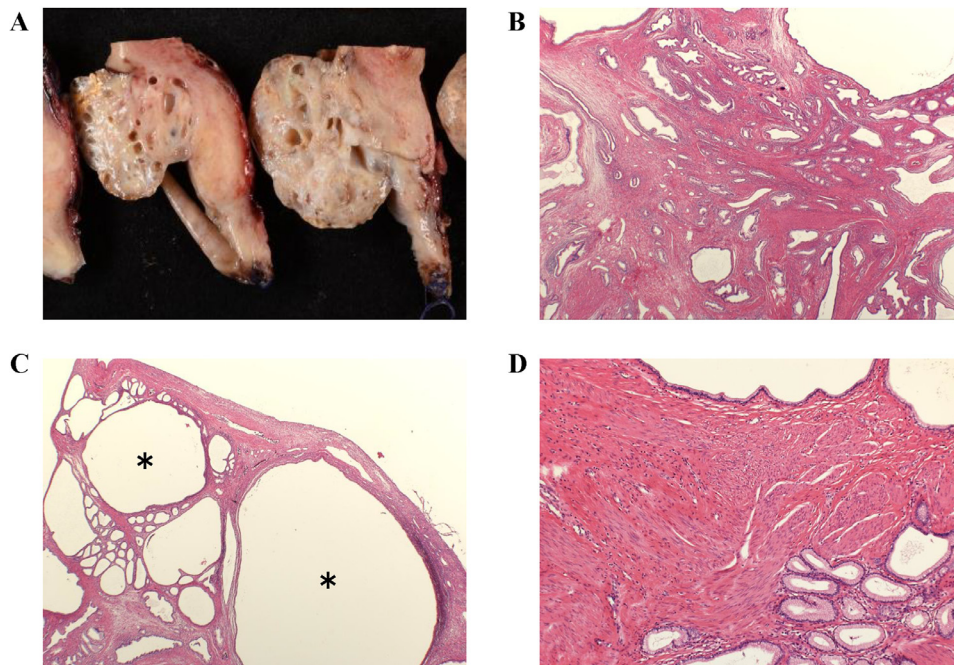


Fig. 4 – Resected specimen (A) and pathological specimens (B-D). (A) Macroscopically, the tumor is a polypoid mass protruding from the posterior wall of the uterine cervix. It is 4.5 cm in length with multiple cystic structures inside. (B) There are numerous glandular proliferations in the stroma, which is mainly smooth muscle. The glands are lined by mucinous epithelium without nuclear atypia (unshown). There is no invasion into the surrounding stroma. (C) The markedly dilated glands show a cyst-like morphology (asterisk). (D) Some areas of the stroma have very sparse glands, and the same areas are mostly smooth muscle or fibrous tissue.

common tumor morphology was a polypoid mass protruding into the endocervical canal. The tumor range in size from 1.3 to 23.0 cm. Four tumors were >10 cm in diameter [1,5,8]. Markedly dilated cyst-like glandular structures were observed in at least 9 pathology cases. MRI was reported in 3 cases: 2 on T2WI and 1 on T2WI and contrast-enhanced image [3–5]. All were polypoid masses protruding into the endocervical canal, with multiple hyperintense signal cysts in slightly hyperintense to strongly hypointense signal stroma on T2WI. This review revealed that tumors often contained cyst-like, markedly dilated glandular structures and have a polypoid morphology. These findings were evaluated using MRI. MDAs do not usually have a polypoidal shape. Therefore, MRI evaluation of tumor morphology is imperative for the preoperative diagnosis of uterine adenomyoma of endocervical type. However, it is difficult to differentiate uterine adenomyomas of the endocervical type from submucosal uterine myomas if cyst-like dilated glandular structures within the tumor cannot be identified on MRI. Our case was misdiagnosed as a hemorrhage from a submucosal uterine myoma 11 years ago because this cyst-like dilated glandular structure could not be identified within the tumor on MRI (Fig. 2A). Moreover, in our case, the signal on T2WI of the stroma was slightly hyperintense to strongly hypointense, similar to other reports. Pathological examination confirmed the presence of multiple small glands within the stroma, which was consistent with MRI findings. The distribution of these small glands in the stroma was not uniform, with some areas showing a high percentage of smooth muscle (Fig. 4D). Therefore, T2WI showed a slightly hyperintense signal in the stroma with many small glands and a strong hypointense signal in the area with few small glands and smooth muscles.

Uterine adenomyomas of the endocervical type often present as polypoid masses protruding into the endocervical canal with cyst-like dilated glandular structures, usually confirmed by MRI. Therefore, evaluation of its morphology on MRI is vital for preoperative diagnosis.

Patient consent

In preparing this case report, the author explained the case to the patient and obtained written consent from the patient.

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