MRI and Transvaginal Ultrasound Findings of Atypical Polypoid Adenomyoma: A Case Report

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ABSTRACT

Atypical polypoid adenomyoma (APA) is an uncommon type of polypoid characterized by fibroid stroma and endometrial glands. It occurs mostly in premenopausal women and rarely in postmenopausal women with irregular vaginal bleeding. In our current case, a 76-year-old woman presented with irregular vaginal bleeding. The final pathological diagnosis of the mass was APA. APA is not easy to diagnose before surgery. On the one hand, there was no obvious particularity in imaging features and clinical features, especially for uncomfortably identifying endometrial cancer. On the other hand, APA has a pedicle, attaching to any part of the uterine cavity, which can cause pseudocoel between the mass with the uterine cavity wall. So, when it comes to getting the pathological tissue in the absence of hysteroscopy, it is easy to access to the pseudocoel and obtain endometrial tissue rather than the pathological tissue of the mass. Therefore, preoperative imaging examination is of great significance diagnosis way of thinking to clinicians for APA. In the meantime, pathological tissue of APA can be obtained by hysteroscopy in visual conditions.

Key words: postmenopausal; atypical polypoid adenomyoma; irregular vaginal bleeding; MRI; transvaginal ultrasound

Endometrial polyps are the most common benign masses in the uterine cavity, whereas atypical polypoid adenomyoma (APA) is an uncommon condition. Only 14 of 1100 endometrial polyps were confirmed to be polypoid adenomyoma^[1], among which APA is even more rare. The interstitium of APA contains smooth muscle fibrous tissue, which is covered by a single layer of inner epithelium^[2]. APA may be associated with long-term estrogen effects^[3]. For postmenopausal women, approximately 1/10 of the patients with abnormal vaginal bleeding are finally diagnosed with endometrial cancer^[4]. For postmenopausal women, however, endometrial polyps are usually asymptomatic, and patients with large endometrial polyps may also show symptoms of vaginal bleeding. In our current case, a postmenopausal woman with irregular vaginal

bleeding was found to have a large and heterogeneous mass in the uterine cavity on imaging; however, no obvious abnormality was found by endometrial biopsy. Subsequently, the lesion was excised by hysteroscopy and pathologically confirmed to be an APA.

CASE DESCRIPTION

A 76-year-old woman presented with irregular vaginal bleeding for 5 days. The small amount of bleeding was bright red in color and persisted at admission. She had no abdominal pain, embolism, leucorrhea abnormality, vulva pruritus, vaginal swelling, frequent, urgent or painful urination, diarrhea, constipation, or bloody stool. The patient was in good mental status, with good appetite and sleep. She reported

that she had lost 1 kg in weight and slightly lost his physical strength in the past 3 months. She had been suffering from coronary heart disease, hypertension, and diabetes in the past two decades, with a history of major excision of thyroid for hyperthyroidism. Her thyroid function remained normal currently.

Gynecologic examination showed: vulva was normal; vagina was unobstructed, although a small amount of dark red blood stain was seen; cervical hypertrophy and cervical polypoid neoplasm were observed; the uterine body had smooth surface and good activity, without tenderness; and no obvious abnormality was found in the uterine adnexal area. Ultrasound findings included: the uterine body was sized about 78mm × 78mm × 59mm, the light spots in muscular wall were evenly distributed, the thickness of endometrium of the uterus was about 39mm (including anterior and posterior walls); multiple irregular cysts of different sizes were visible, among which the largest cyst was sized about 19mm × 14mm (**Fig. 1**). MRI revealed long strips of soft tissues mass in the uterine cavity, approximately 64mm × 50mm × 78mm in size, which has an uneven signal with a clear boundary, and slightly high intensity on T2 weighted image. Multiple small follicular cysts with high intensity were unevenly distributed within the mass on T2WI (**Fig. 2**). Endometrial curettage and biopsy were performed, and the pathology only revealed hematoma. Hysteroscopy was later arranged. Hysteroscopy revealed a heterogeneous and soft mass with uneven surface and hemorrhage. The mass was sized about 40mm × 50mm, located at the base of the uterus, and prolapsing into the middle of the cervical canal. It could not be peeled off, and then biopsy of the tumor by hysteroscopy was performed (**Fig. 3**). Tumor histology showed adenomyosis. Transabdominal hysterectomy and adnexal resection were performed (**Fig. 4A**). The patient was successfully discharged in 38 days.

Pathology showed that there were hyperplastic interstitial fibrous tissues in the endometrium, which had plenty of endometrial stroma and glands. Local glands were enlarged by different sizes and densely arranged, in which some of neutrophils and chronic inflammatory cells were diffusely distributed. Some parts of the mass showed polypoid growth and hyperplasia of glands; the ratio of glands to intercellular substance increased, the sizes were unequal, the arrangements were irregular, and cystic expansion was visible in some parts (**Fig. 4B**).

The pathological diagnoses were: a) APA; and b) endometrial polyps with simple endometrial hyperplasia.

Figure 1. Colour Doppler sonographic image of a 76-year-old woman diagnosed with APA.

The endometrium is markedly thickened, with multiple irregular cysts of different sizes and with a few spotty blood flow signals. APA, atypical polypoid adenomyoma.

Figure 2. A sagittal T2WI MRI image of a 76-year-old woman diagnosed with APA.

The mass fills the uterine cavity. The arrows indicate that multiple small follicular cysts with longer T2 signals are unevenly distributed within the mass.

Figure 3. Hysteroscopy image of a 76-year-old woman diagnosed with APA.

A large solid mass occupies the uterine cavity. The arrows indicate the joint where the APA lesion interconnects with the base of the uterine cavity is partially convex.

Figure 4. Postoperative specimen and pathology. (A) Gross pathology of hysterectomy and adnexal resection. A large solid mass occupying the entire uterine cavity. (B) Microscopy shows there are plenty of fibrous hyperplasia, with abundant endometrial stroma and glands. Local glandular hyperplasia varies in size and is densely arranged (HE $\times 20$).

DISCUSSION

Polypoid adenomyoma is rare uterine endometrial polyps featured by close mixing of endometrial glands with smooth muscle and thick-walled blood vessels. It accounts for about 1.3% of endometrial polyps reported in the literature^[1], among which APA is even more rare. Although endometrial polyps are usually small, APA can be as large as 17cm^[5]. Most of polypoid adenomyomas originate from the base of the uterus; when it is large enough, it can also fill the entire uterine cavity and protrude from the cervix, as reported in our current case. The most common clinical symptom of APA is abnormal vaginal bleeding, followed by increased menstruation, dysmenorrhea, and infertility. Protrusion of the cervical mass has been reported in a small number of cases^[5]. Our current patient presented with abnormal vaginal bleeding, which was consistent with literature^[5]. However, for postmenopausal women, when abnormal vaginal bleeding occurs, about 1/10-1/5 of the patients are finally diagnosed with endome-

trial cancer or cervical cancer^[4]. In addition, clinicians often lack experience in diagnosing APA. Therefore, clinical diagnosis of APA in women with postmenopausal bleeding can be challenging.

Only a few APA cases have been reported regarding its imaging manifestations^[6, 7]. On the one hand, APA has diverse radiographic findings, making it indistinguishable from endometrial polyps and endometrial carcinoma. Ultrasonographic findings of APA are mainly pedicled or un-pedicled neoplasms in the endometrial cavity, featured by heterogeneity of internal echoes, cystic areas in the neoplasm, and clear boundaries with the muscular layer^[8]. The boundary with the muscular layer is helpful in distinguishing APA from an intrauterine malignancy, as the latter has poorly-defined boundary between the lesion and muscular layer. About 20% of patients with polypoid adenomyomas also have myometrial adenomyomas^[8]. Although solid mass in the uterine cavity combined with multiple cystic lesions may be suggestive for the diagnosis of APA^[7], malignant mass in the uterine cavity can cause uneven endometrial thickening and small anechoicity in postmenopausal women, with a low speed and low resistance of the blood flow^[9]. On the other hand, as reported in this article, a large APA can fill the entire uterine cavity and protrude from the cervix; as a result, transvaginal ultrasound cannot distinguish between solid mass and diffuse endometrial dysplasia. On MRI, polypoid adenomyoma mainly shows single or multiple nodular masses in uterine cavity and may involve the uterine body and cervix^[10]. T1WI shows equal or slightly longer T1 signals, T2WI shows slightly higher or higher mixed signals, and T2WI also shows scattered small high-signal cysts in the mass, reflecting the epithelial components of the gland, which may be the characteristic MRI features of APA. The contrast-enhanced images in our current study showed uneven enhancement in the mass. Therefore, it is difficult to distinguish between APA and endometrial malignant hyperplasia by imaging alone in clinical practice, which is consistent with some experimental observations^[11, 12]. Therefore, when a large polypoid mass occupies the endometrial cavity and protrudes into the vaginal cavity, and imaging examinations detect intratumoral cysts in variable size, with clear boundary between the lesion and muscular layer, a differential diagnosis of polypoid adenomyoma should be considered.

To confirm the diagnosis in the current case, di-

lation & curettage of the cervix and uterine cavity was further performed. Diagnostic curettage is one of the most commonly used diagnostic tests for endometrial disease. Although curettage plays an important role in the preoperative diagnosis of endometrial samples, Visser *et al.*^[12] found 13% of patients who had a preoperative diagnosis of atypical hyperplasia by endometrial sampling were finally diagnosed as endometrial cancer after surgical resection. In our current case, the curettage missed the diagnosis, which may be due to the lack of knowledge about the imaging of APA, radiologist did not provide the corresponding information of the diagnosis way of thinking to the clinician of APA. As reported in the literature^[7], APA has a pedicle, attaching to any part of the uterine cavity, which can cause pseudocoel between the mass with the uterine cavity wall. So, when it comes to getting the pathological tissue in the absence of hysteroscopy, it is easy to access to the pseudocoel and obtain endometrial tissue rather than the pathological tissue of the mass. It is able to enter the uterine cavity under direct vision with hysteroscopy. So it were more precise to excise parts of the cervix and uterine lesions for histopathological examination, and the diagnosis of APA was established preoperatively.

As APA is mainly a submucosal polypoid mass that can be removed directly by hysteroscopy, gonadotropin-releasing hormone (GnRH) agonists may be used as a pretreatment to reduce the size of APA, especially for large tumors^[3]. However, although APA is a rare benign uterine tumor, it can coexist with endometrial atypical hyperplasia and/or endometrioid adenocarcinoma^[13]. For patients with large APA, hysterectomy or even radical hysterectomy is suggested^[14].

It remains difficult to distinguish APA from other malignant polypoid endometrial tumor, such as endometrial stromal sarcoma and adenocarcinoma of endometrium, especially in postmenopausal women with abnormal vaginal bleeding. Imaging examinations can help a clinician to characterize the entity and refine treatment strategies. While curettage specimens sometime fail to provide the essential information to establish a correct diagnosis, hysteroscopy plays an important role in the pathological diagnosis of APA.

Conflict of interests

The authors declare no conflict of interests.

Patient consent

Written informed consent was obtained from the reported patient.

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病例报道

非典型息肉样腺肌瘤的 MRI 和经阴道超声表现：1 例报道

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摘要

非典型息肉样腺肌瘤 (atypical polypoid adenomyoma, APA) 是一种罕见类型的息肉，其病理特征主要是肌瘤间质和子宫内膜腺体相间。APA 主要发生在绝经前女性，很少发生在伴有不规则阴道出血的绝经后女性。本例 76 岁绝经女性表现为不规则阴道出血，最终病理诊断为 APA。APA 在手术前不易诊断：一方面，APA 的影像学和临床特征无明显的特殊性，尤其是在与子宫内膜癌的鉴别方面；另一方面，APA 有一个蒂，可以附着在宫腔的任何部分，这可能导致肿块与宫腔壁之间形成假腔，因此，在没有宫腔镜检查的情况下获取病理组织时，很容易通过假腔获得子宫内膜组织，而不是肿块的病理组织。因此，术前影像学检查对临床医师诊断 APA 具有重要意义；同时，可在宫腔镜直视条件下获得 APA 的病理组织。

关键词：绝经后；非典型息肉样腺肌瘤；不规则阴道出血；MRI；阴道超声

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