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Case Report

A case of rapidly-growing atypical polypoid adenomyoma which was histologically diagnosed before operation and removed by a laparoscopic resection

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ABSTRACT

Objective: Atypical polypoid adenomyoma (APAM) is an epithelial-mesenchymal mixed tumor which often develops in the uterine cavity of reproductive age women, requiring preservation of the reproductive functions. Preoperative endometrial biopsy may not yield histological diagnosis as the tumor is a solid smooth muscle tumor. The standard treatment option is a hysteroscopic resection for the diagnosis and the treatment at the same time.
Case report: We report a case of rapidly-growing APAM successfully diagnosed preoperatively via transcervical punch biopsy followed by a laparoscopic resection. The mass was relatively large, had been located in the lower segment of the uterus, and the area of contact with the muscular layers was large. It was a complete removal and no recurrence had been observed 9 months after the operation.
Conclusion: This is the first report of APAM treated by laparoscopic resection. The method may be a useful alternative when hysteroscopic surgery is inappropriate.

Introduction

Atypical polypoid adenomyoma (APAM) is a benign disease of epithelial-mesenchymal mixed tumor which develops in the uterine cavity. The first report was made in 1981 by Mazur MT [1]. Hysteroscopic resection is the main stay for the diagnosis and treatment. However, the secondary resection may have to be performed in case if the primary resection had been incomplete [2,3]. This is a case report of a rapidly-growing APAM which was histological diagnosed preoperatively followed by a laparoscopic resection.

Case report

The patient was a 37-year old nulliparous woman with height of 158 cm and body weight of 63.8 kg. There was no remarkable past medical history. She had been under regular check-up for fibroid at a nearby clinic for the past 4 years. She visited our clinic for the first time with a chief complaint of irregular vaginal bleeding. Internal examination showed uterine body mass. Transvaginal ultrasonography revealed the mass size of 50 mm at low intensity of subserosal location and another mass of 20 mm with high intensity in the submucosal location. MRI confirmed a subserosal myoma of 52 × 38 mm and a sub-mucosal myoma of 20 × 20 mm. We recommended hysteroscopic resection but she declined. After 3 months of observation, the subserosal mass showed no significant growth but the diameter of the submucosal mass had been increased to 40 mm (Fig. 1). The patient therefore had another MRI. A mass of 41 × 34 × 31 mm was detected inside the uterine cavity at slightly high intensity in a T2-weighted image and at high intensity in a T1-weighted image with some blood components (Fig. 2 A, B). The result of endometrial cytology was negative. The level of CA125 was 13 U/ml and CA19-9 was 13 U/ml and was both within a normal range. In order to obtain adequate specimen to establish diagnosis, the cervix was dilated and the punch biopsy of the submucosal mass was performed. The material showed irregular growth of the glandular tissue in the interstitial tissue consisting of spindle cells.

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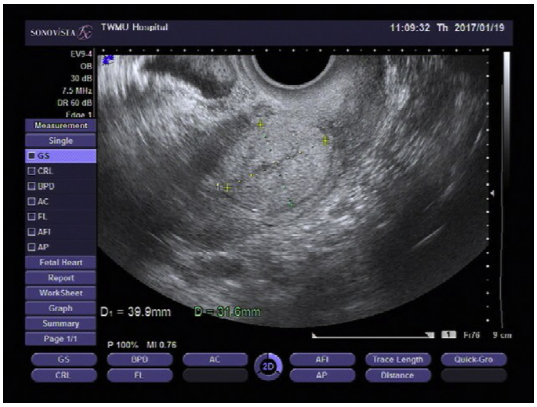

Fig. 1. The sagittal section of transvaginal ultrasonography. A mass of 40 × 32 mm was detected at low intensity in the lower segment of the uterus.

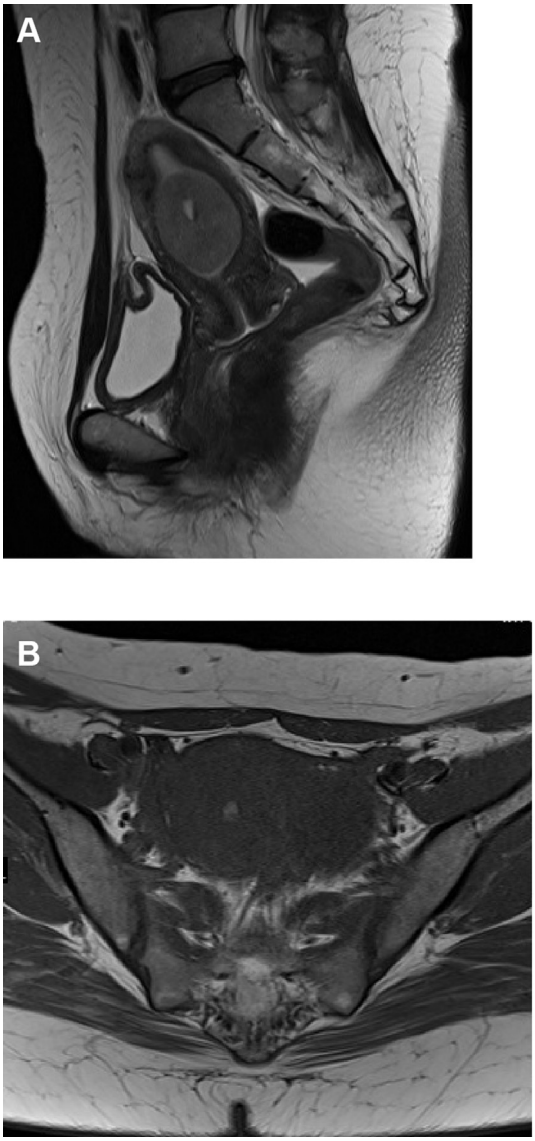

Fig. 2. Pelvic MRI images (A: T2 weighted image, sagittal section, B: T1 weighted image, horizontal section). The mass was located in the uterine cavity with slightly high intensity on T2 weighted image and low intensity on T1 weighted image. There was a high intensity area inside the mass on T1 weighted image.

Also squamous cell-like morula was observed in some glandular tissues (Fig. 3A). Those spindle cells were immunohistochemically stained with SMA. The endometrial stroma and morula were strongly positive for CD10 (Fig. 3 B, C). These pathological results suggest APAM. In hysteroscopy, a mass protruding in the lower segment of the uterus was observed. The base of the mass was widely in contact with the uterine wall, and no peduncle was found. As it appeared difficult to have a clear visual field and detect the border between the mass and the muscular layers in hysteroscopic approach, we decided the resection by a laparoscopic surgery.

After obtaining consent, laparoscopic surgery was performed with pneumoperitoneum using 4 ports (the parallel method) under general anesthesia. We first resected the subserosal myoma on the posterior wall of the uterus. Then the uterine manipulator

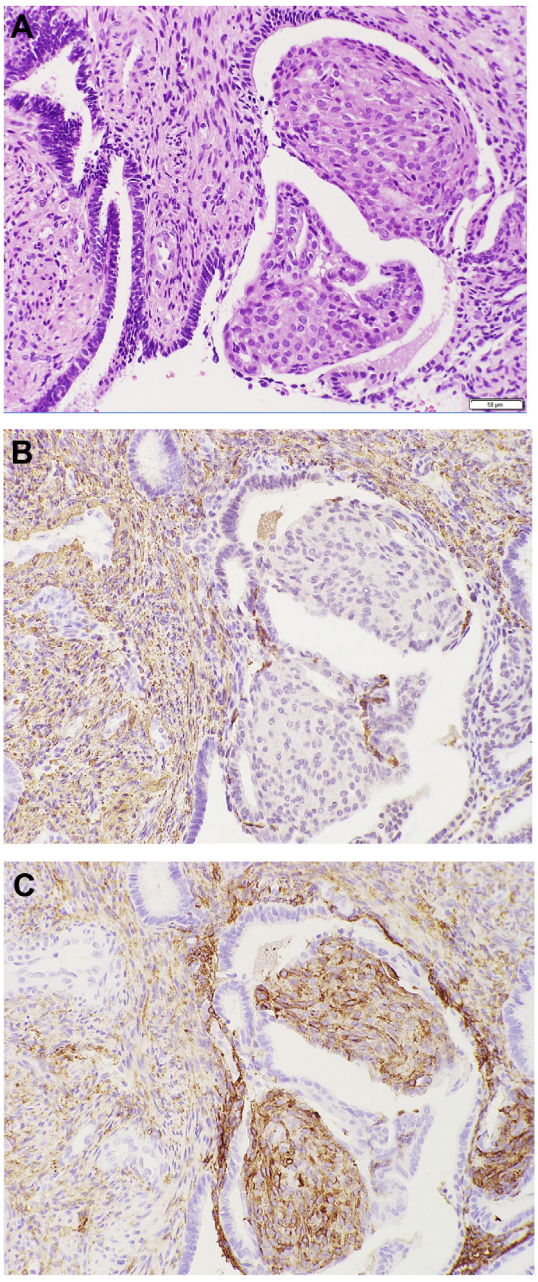

Fig. 3. Pathological findings. A: HE stain, ×200. Squamous cell-like morula was observed in the interstitial tissue consisting of spindle cells. B: SMA stain, ×200. SMA, which indicates smooth muscle cells, was positive in the interstitial tissue. C: CD10 stain, ×200. The endometrial stroma and morula were strongly positive for CD10.

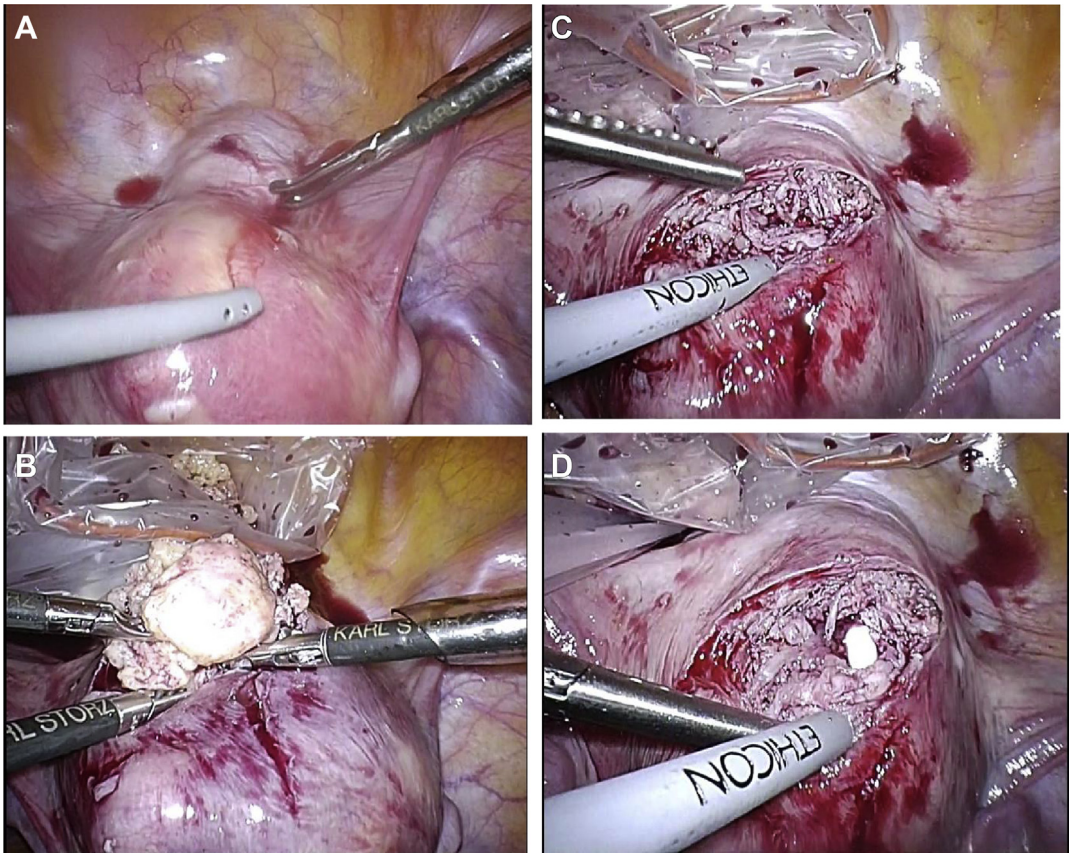

Fig. 4. Findings in the laparoscopic surgery. A: APAM was displaced by a uterine manipulator and a part of muscular layers was slightly elevated. B: The mass and muscle layers were separated by blunt operation. C: APAM was removed macroscopically without leaving any residues. D: A part of endometrium was damaged, and the manipulator was exposed.

was flexed posteriorly. This maneuver made a part of the front uterine wall slightly bulged facilitating the identification of the submucosal mass (Fig. 4A). A transverse incision of approximately 30 mm was made at this site. We reached the mass at a depth of about 5 mm, and separated it from the myometrium by blunt dissection (Fig. 4B). The mass was fragile, and multiple fractional removals were necessary to complete the resection (Fig. 4C). The damage of the endometrium was about 5 mm (Fig. 4D), and it was repaired by Z-suture, performed once, using an absorbable suture of 3–0. The weight of myoma resected was 81 g and APAM 8 g. The amount of bleeding during the operation was small. The histological study of the resected sample confirmed APAM. Hysteroscopic examination showed no APAM recurrence 9 months after the operation.

Discussion

Matsumoto et al. reported a study of 29 APAM patients [4]. The mean age of 38.0 (22–58) years old at the time of onset, and 93.1% was before menopause and 75.9% was nulliparous women [4]. The mean diameter of the tumor was 23 mm (5–65 mm), and 58.6% were pedunculated and 41.4% was sessile [4]. The sites of onset were in the fundus of the uterus for 58.6%, the lower segment of the uterine body 34.5%, and cervical canal 6.9% [4]. The tumor diameter was 2 cm at the first visit and it grew up to 4 cm in 3 months period. This suggests that it was an unusually rapidly growing APAM.

Pathological features of APAM include proliferation of atypical endometrium glands and smooth muscle cells, often with morula. Meanwhile, Longacre TA et al. reported that APAM with more than 30% of the structural atypia, undistinguishable from well-differentiated endometrial adenocarcinoma recurred by 60%. Therefore APAM with this feature is called APAM with low potential malignancy [5]. In addition, APAM has often co-exists with endometrial carcinoma. Matsumoto et al. [4] and Healey et al. [6] reported that endometrioid adenocarcinoma was observed in 17% and 8.8% of APAM patients respectively.

As for the diagnostic approach, size and location of APAM can be confirmed by imaging studies such as ultrasonography, CT and MRI. However, it is still difficult to differentiate APAM from degenerated fibroid even if it shows slightly high intensity on a T2 weighted image as in this case; it may be diagnosed as submucosal myoma on an imaging study alone. Moreover, although endometrial cytology and histology may reveal presence or absence of endometrial hyperplasia or endometrial cancer, it is difficult to obtain tissues necessary for diagnosis of APAM with curettage as the mass is hard. Endometrium cytology was negative in this case. However, as it had been rapidly growing, we obtained a biopsy from a large area so that it would contain not only endometrium but also subendometrium tissues by dilating the cervical canal and used a biopsy forceps. The biopsy was relatively easy as the mass was large and located near the internal ostium of uterus. We predicted a need of hemostatic procedure for vaginal bleeding, but the amount of post-biopsy bleeding was very small.

Hysterectomy may be considered if patient does not wish pregnancy because of risk of recurrence or coexisting endometrial cancer. If patient wishes to preserve fertility, hysteroscopic surgery and intrauterine curettage are performed. Matsumoto et al. reported that out of 21 patients who preserved fertility, recurrence was observed in one patient who received hysteroscopic surgery

(1/10) [4]. There are some reports that APAM was not completely resected by hysteroscopic surgery by one attempt due to ambiguity of the border between the lesion and the muscular layers [2], or technical difficulty due to the location of APAM near the internal ostium of uterus [3]. Therefore those cases required secondary surgery. In the current patient, we judged that complete resection of the mass would be difficult with a hysteroscopic approach as the mass was relatively large, had been located in the lower segment of the uterus, and the area of contact with the muscular layers was large. The thickness of the muscular layers had also reduced to about 5 mm, with a risk of perforation of the uterus. Although no report has been published on laparoscopic surgery for APAM, it was effective in our case as the mass was removed without leaving any residues and no recurrence occurred nine months after the operation.

Conflicts of interest

The authors have been no conflict of interest relevant of this article.

Acknowledgement

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