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CASE REPORT

Hironori Tashiro · Hidetaka Katabuchi
Yukitoshi Fukumatsu · Nobuyuki Tanaka
Yasuyuki Yamashita · Kohei Matsuura · Hitoshi Okamura

Endometrial adenocarcinoma following the conservative treatment of an atypical polypoid adenomyoma

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Abstract
We present a case of well differentiated endometrioid adenocarcinoma of the endometrium in a 29-year-old woman, developing 6 months after an atypical polypoid adenomyoma (APA) was diagnosed by curettage. Four cases of APA with coexistent endometrial adenocarcinoma were recently reported. To our knowledge, this is the first reported case of endometrioid adenocarcinoma following conservative treatment for an APA. Since APA may be associated with the development of adenocarcinoma, careful follow-up with hysteroscopy and endometrial sampling is recommended in a woman with APA who desires continued fertility.

Key words Uterus · Atypical polypoid adenomyoma · Endometrial adenocarcinoma · Hysteroscopy

Introduction

Atypical polypoid adenomyoma (APA) is a rare endometrial lesion which usually occurs during the reproductive and perimenopausal years. The term APA was first introduced by Mazur in 1981.[1] APA is histologically characterized by atypical endometrial glands encompassed by swirling and interlacing fascicles of smooth muscle cells. This lesion is generally not aggressive, and it is accepted that conservative management can be attempted.[1–3] However, we encountered a case of well differentiated adenocarcinoma of the anterior uterine wall in a 29-year-old woman 6 months after an APA was diagnosed by curettage.

H. Tashiro (✉) · H. Katabuchi · Y. Fukumatsu · N. Tanaka · K. Matsuura · H. Okamura
Department of Obstetrics and Gynecology, Kumamoto University School of Medicine, Honjo 1-1-1, Kumamoto 860-8556, Japan
Tel. +81-96-373-5269; Fax +81-96-363-5164
e-mail: obgyn@kaiju.medic.kumamoto-u.ac.jp

Y. Yamashita
Department of Radiology, Kumamoto University School of Medicine, Kumamoto, Japan

Case report

The patient was a 29-year-old Japanese female, gravida 0, para 0, with a history of infertility, irregular menstruation, and menorrhagia. Magnetic resonance images revealed a protruding hypointense mass[4] consistent with a filling defect noted on hysterosalpingography (Fig. 1). Rigid hysteroscopy disclosed an exophytic, polypoid mass with bosselated surface protruding from the anterior fundus. A dilatation and curettage was performed. The curettage specimen contained a round fragment of polypoid tissue measuring 1.5 × 1.0 × 1.0 cm (Fig. 2), as well as smaller pieces of tissue. The polypoid tissue had a rubbery consistency, but was less elastic than submucosal myoma. The cut surface was yellow-grayish homogeneous material. Microscopically, the specimen consisted of hyperplastic endometrial glands with nuclear atypia and squamous morules surrounded by smooth muscle cells (Fig. 3). The stromal cells had elongated nuclei with blunt ends and eosinophilic cytoplasm. Their mitotic count was only one per ten high-power fields. Immunohistochemical studies showed that the stromal cells were stained with desmin and alpha smooth muscle actin as well as with vimentin. These histological and immunohistochemical findings are characteristic of an APA.

Three months after the curettage, hysteroscopy disclosed two broad-based nodules extending from the fundus to the anterior wall of the cavity of the corpus. A directed biopsy and an endometrial smear test did not show any malignant findings. Six months after the initial curettage, hysteroscopy disclosed diffuse thickening with a knobby surface over the anterior uterine wall. A directed biopsy showed glandular crowding with nuclear atypia, and endometrial carcinoma could not be ruled out. Hysteroscopic resection was technically not possible. Considering the patient's youth, we attempted resection of the uterine masses with

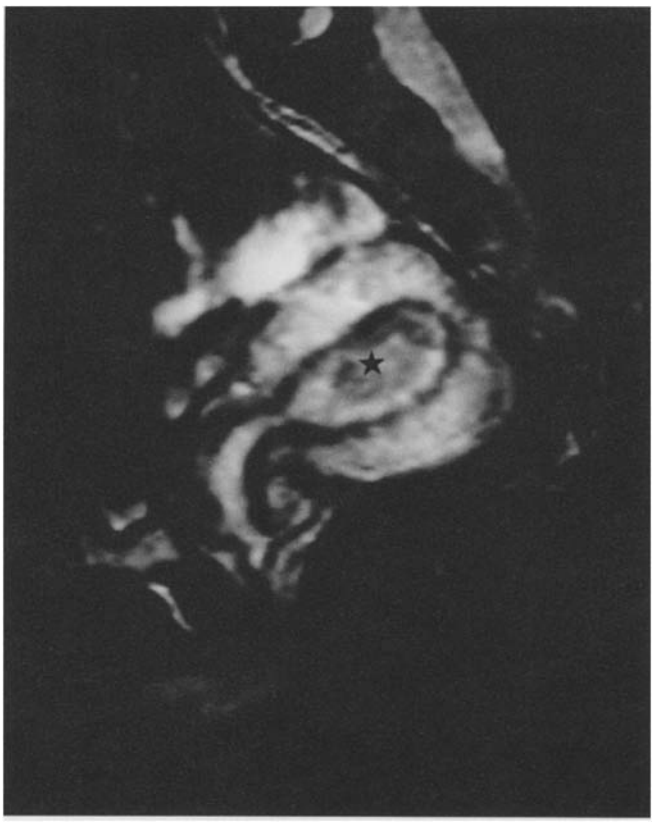

Fig. 1. Magnetic resonance sagittal image (TR = 2.0 s; TE = 70 ms) of the uterus, showing protruding hypointense lesion (★) within the uterine cavity

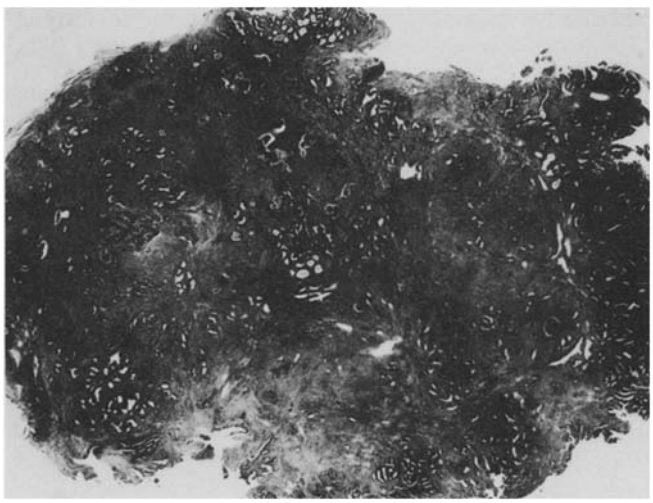

Fig. 2. Cross section of atypical polypoid adenomyoma in specimen from initial curettage, showing round shape. H&E, ×10

laparotomy. Multiple polypoid lesions, each measuring less than 1 cm, were noted over the anterior wall from the fundus to the internal os. The boundaries between the foci and the myometrium, unlike those of a myoma nodule, were indistinct.

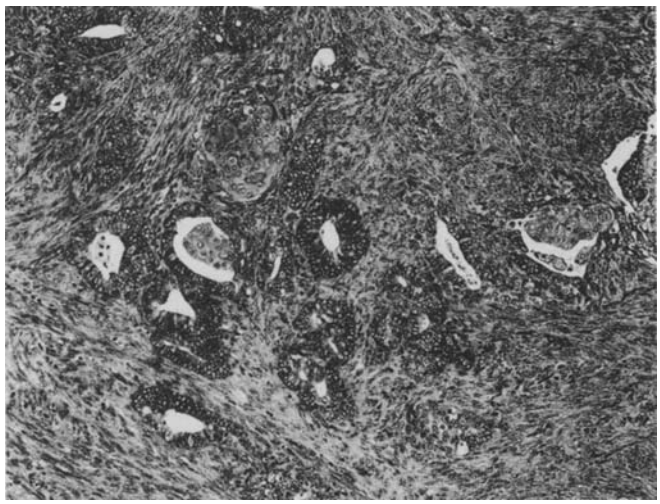

Fig. 3. Atypical polypoid adenomyoma in specimen from initial curettage. H&E, ×100

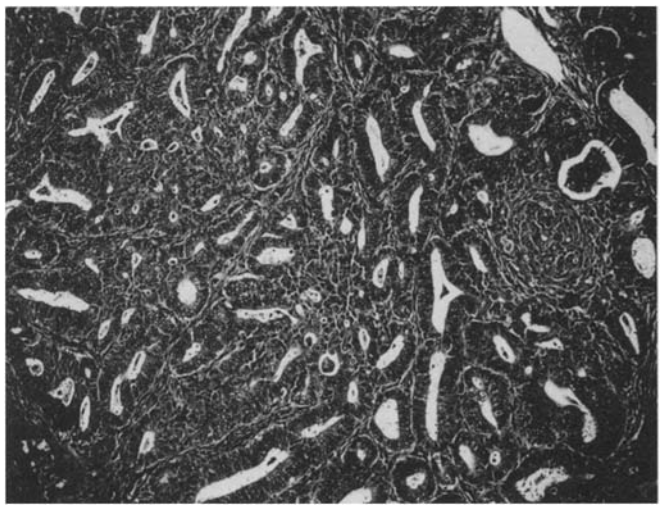

Fig. 4. Well differentiated adenocarcinoma in hysterectomy specimen. H&E, ×100

Intraoperative frozen section showed adenocarcinoma; total abdominal hysterectomy, bilateral salpingo-oophorectomy and pelvic lymphadenectomy were therefore performed. The uterus weighed 115 g. The bilateral ovaries were slightly enlarged and contained multiple follicles resembling polycystic ovary (PCO), although the characteristic endocrine disturbance of PCO had not been noted. The consistency of the polypoid masses was softer and less rubbery than that of the prior curettage specimen. Microscopic observation of the multiple polypoid tissues confirmed the glandular atypia accompanying squamous morules, similar to the epithelial component of the prior APA. In addition, there was ovoid or tongue-like architecture consisting of a cluster of confluent glands (Fig. 4). The neoplastic glands also elicited a desmoplastic stromal reaction, consistent with stromal invasion. The fascicles of smooth muscle cells in many of the polypoid lesions were less prominent than previously noted in the prior APA

specimen (Fig. 4). The patient is alive and without recurrence 4 years after the hysterectomy.

Discussion

APA is an unusual tumor which occurs as a polypoid mass in the uterine cavity. Since its first description by Mazur in 1981,[1] studies of the clinical features of APA have been reported.[2,3] Histologically, this neoplasm has an intimate admixture of hyperplastic endometrial glands and smooth muscle fascicles. The glands exhibit architectural and cytologic atypia, but this lesion has not been considered to display aggressive behavior. It is clinically accepted that APA can be managed by conservative treatment. However, hysterectomy has been recommended when APA cannot be distinguished from endometrial adenocarcinoma.[5]

APA usually occurs in pre- and perimenopausal women, and some cases of APA have been reported to be a complication of long-term estrogen exposure.[6,7] The histological features are basically similar to those seen in atypical endometrial hyperplasia. In addition, it has been reported that the endometrium adjacent to the APA is hyperplastic in some individuals.[1-3] These findings suggest biological correlates between APA and atypical endometrial hyperplasia. APA may be a precursor of or associated with endometrioid carcinoma. While nearly one-quarter of all cases of atypical endometrial hyperplasia progress to well differentiated endometrial carcinoma,[8] the current case report suggests that endometrial carcinoma may develop following an APA. Moreover, Longacre et al.[3] reported that 45% of APA specimens they examined contained markedly complex glands that were indistinguishable from well differentiated carcinoma, and in some instances the stromal component was less prominent in the recurrent or persistent lesions. Our findings of a decreased stromal component in many of the subsequent lesions in our patient supports these findings. In recent years, four cases of APA associated with adenocarcinoma have been reported.[6,9-11]

In our patient, the time elapsed from the first treatment to reappearance of the nodules in the uterine cavity was just 3 months. Such an interval suggests the co-existence of the adenocarcinoma and APA at the time of the curettage. Serial sections of the entire curettage specimen revealed no carcinomatous lesion. Further, the directed biopsy and the endometrial smear showed no malignant cells 3 months after the first treatment (the curettage). Judging from the patient's history, it is unlikely that the adenocarcinoma was not associated with APA at the time of the first treatment.

In conclusion, we believe this is the first report of APA that was observed to progress to adenocarcinoma after conservative treatment had already been undertaken.

As for the treatment of APA, hysterectomy is the most effective treatment, but may not always be desirable. If a patient with APA is conservatively treated because of her desire for continued fertility, careful follow-up, with hysteroscopy and possible endometrial sampling is recommended.

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