

# Adenomyosis completely encapsulated by muscle-like cavity in the mesorectum: a case report

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## Summary

A case of adenomyosis, completely encapsulated and located in the mesentery between the sigmoid colon and rectum, was admitted our hospital. We have reported the symptoms, diagnosis, and treatment of the present case.

**Key words:** Adenomyosis; Muscle-like cavity; Mesorectum.

## Introduction

Adenomyosis is a benign disease of the uterus caused by ectopic translocation of endometrium to the myometrium. A typical symptom of adenomyosis is aggravating dysmenorrhea, and is frequently accompanied by menstrual disorders, anemia, infertility, and sexual pain [1, 2]. Adenomyosis is not considered as a tumor per se because it does not have a capsule and cannot be completely removed by regular surgery [3].

## Case Report

The patient (female, 39 years old) was admitted to the hospital due to five years of dysmenorrhea and three months of aggravating symptoms. The patient had regular and average amount of emmenia without abdominal pain. Over the past five years, she also experienced dysmenorrhea and the symptoms had gradually worsened, but the patient could still work and live normally without treatment. During the recent three months, dysmenorrhea had particularly become aggravated and pain was not even relieved during the non-menstrual period. Analgesic drugs were needed and the patient had difficulty in working normally, which severely affected the quality of life. One month ago, she was found to have an encapsulated mass with a diameter of 5 cm in the left annexal area during the ultrasonic and gynecological examinations in another hospital. Surgical treatment was recommended. During the course of disease, there were no changes of menstrual amount, stool quality and quantity. The patient did not have other discomfort except for pain caused by dysmenorrhea. The patient had been pregnant four times and gave birth twice. Currently, she has two healthy daughters. The patient had had two abortions with the last abortion in 2006. Since 2006, a contraceptive intrauterine device was placed. Gynecological examination revealed that the uterus was slightly enlarged without tenderness. There was a mass with a diameter of 5 cm in the left back side of the uterus. The mass was solid and motile with obvious tenderness. No abnormalities were observed in the right accessories. Laboratory examination showed that both electrocardiogram and chest X-ray were normal. Blood CA125 concentration was 13.4 U/ml (normal value < 35 U/ml). B-ultrasonic examination indicated

that the right accessories had a mixed mass. The mass (size: 5 × 5 × 4 cm<sup>3</sup>) with a thick capsule had spotted strong echoes in the low intensity areas. There was no adhesion seen in abdominal laparotomy examinations. The uterus was slightly full and purple in color, which is similar to adenomyosis. Accessories on both sides were normal. A capsulated solid mass with a diameter of 5 cm could be detected in the mesentery of retro-peritoneum between the sigmoid colon and rectum. There was no anatomical adhesion between the mass and uterus. Mass cystectomy was performed and the uterus tissue on the bottom side was collected for biopsy examination. During mass cystectomy, the capsule of the mass was complete and the mass itself was completely removed. The surface of the mass was smooth. A 1 cm-thick muscle-like cavity structure of the mass was cut open and approximately 4 ml of chocolate-like viscous liquid came out from the cavity (Figure 1). Pathological examination on both cryo-sample intraoperative and postoperative paraffin-embedded sample confirmed that the patient had adenomyosis. A biopsy of the uterus also reconfirmed adenomyosis. Pathological examination of the mass and biopsy showed that endometrial glands and stroma had an island-like distribution in the uterine myometrium (Figure 2). Endometrial glandular epithelium showed proliferation including expansion of the glands and flat epithelium surrounded by endometrial stroma. Immunohistochemical analysis showed that estrogen and progesterone receptors were positive. It was recommended that the patient use ovarian function suppressive drugs after surgery. However, the patient refused to apply any medication. The patient was observed for two months. Menstruation normalized and dysmenorrhea disappeared.

## Discussion

Adenomyosis is a benign gynecological disease caused by invasion of endometrium to the myometrium, and is accompanied by diffuse hyperplasia of myometrium [4, 5]. The cause of the disease is the direct expansion of the basal layer of endometrium to the muscle layer, but without cultivation or metaplasia [6]. Ectopic endometrium is diffused in the mural muscle of the uterus (most commonly in the posterior wall). The muscle fiber has diffusive and reactive proliferation. The uterus is evenly enlarged with hard quality, and its size varies before and after men-

Fig. 1



Fig. 2

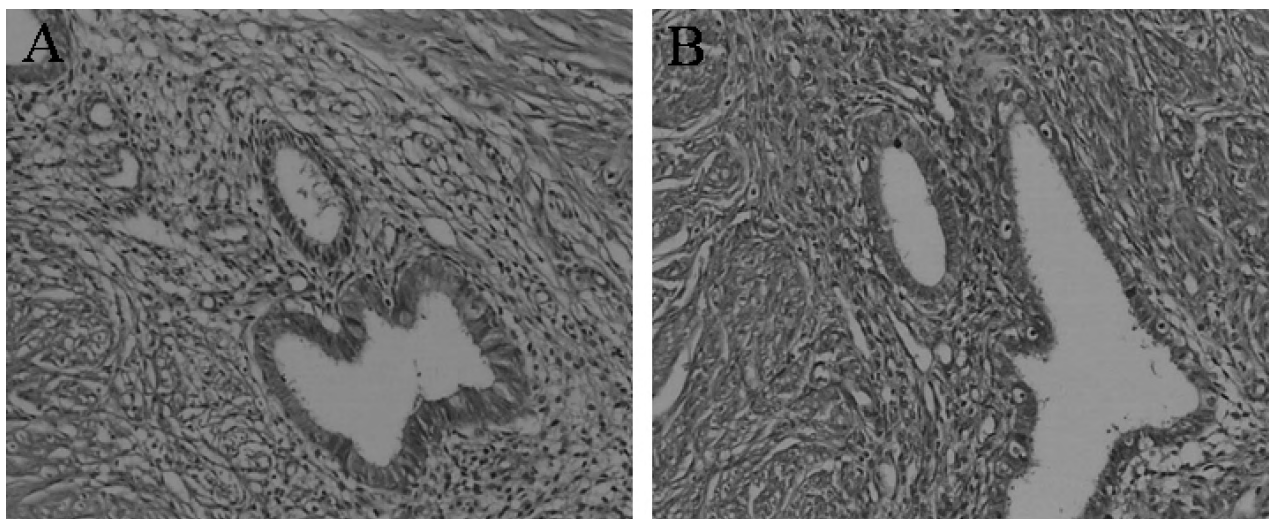


Figure 1. — The signs of mass displayed in each figure. A shows the smooth surface of the mass. B and C show the 1 cm-thick muscle-like cavity structure of the mass that was cut open and approximately 4 ml of chocolate-like viscous liquid that came out from the cavity.

Figure 2. — Histopathologic signs in mesentery adenomyosis in each figure. A and B show detected smooth muscle, endometrial gland, and stroma. Original magnification  $\times 200$ .

struation. Adenomyosis is often associated with endometriosis, fibroids, and pelvic adhesions [7]. One of the characteristics of adenomyosis is an undefined border from the surrounding tissues [8]. The main clinical manifestation of adenomyosis is secondary progressive aggravation of dysmenorrhea and extended time of menstrual period with increased amount. Biopsy during surgery is the gold standard for the diagnosis of adenomyosis [9]. Currently, extremely valuable biochemical markers for diagnosis and monitoring the progression of adenomyosis are unavailable. For example, CA125, anti-endometrial, and anticardiolipin antibodies may be increased, but not specifically in adenomyosis [10]. The clinical symptoms of the patient were secondary progressive aggravation of dysmenorrhea, which was consistent with the manifestation of adenomyosis. In addition, dysmenorrhea disappeared after surgery. We speculated that dysmenorrhea caused the encapsulated mass of adenomyosis. The mass with a smooth surface was located between the mesentery and had a clear border with the surrounding tissues. The features of the mass can hardly be explained by the presence of adenomyosis. Some reports showed that leiomyomatosis peritonealis disseminata (LPD) was a very rare disease and could be manifested by the symptoms of

endometriosis, e.g., progressive aggravation of dysmenorrhea, infertility, and sexual pain [11, 12]. White or gray red nodules can be diffusively distributed on the surface of the peritoneum in the abdominal and pelvic cavities. The color of the nodules is associated with the course of the disease. The surface of the nodules is smooth and the quality of the nodules is hard. The nodules have a clear border with a size of several micrometers to millimeters. There could be several hundred nodules commonly located in the omentum, mesentery, serosa surfaces of the small intestine and colon, and on the surface of the uterus. Histological examination manifests benign smooth muscle changes. Adenomyosis with similar characteristics to the patients in this study has not yet been reported. Currently, there are a few number of adenomyosis cases reported, and epidemiological studies for adenomyosis are unavailable. Therefore, factors that cause this disease are generally unknown. As histological analysis showed that estrogen and progesterone receptors were positive, we believed that the disease occurring in this patient was related to estrogen or progesterone. The occurrence of LPD is believed to be associated with the increased level of female hormones. We consider that adenomyosis can occur in both diffusive uterus fibroids and myometrium.

The patient reported in this study refused any drug medication and was only followed by outpatient service. We continue to follow-up the clinical symptoms and monitor mass recurrence.

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