

# Endometrioid adenocarcinoma arising from adenomyosis: a case report

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## Summary

**Background:** Endometriosis is most commonly found in the peritoneum of the lesser pelvis and in the genital tract (in the ovaries). Its malignant transformation is quite rare, which usually appears in patients who previously underwent surgical procedures aimed at treating endometriosis. Years of hormone substitution (unopposed estrogen therapy) is also considered to have a role. According to the present authors' current knowledge, these are mostly well-differentiated tumors with low malignancy, which are primarily treated surgically. **Case:** In the present case the authors present a 73-year-old female patient who underwent a laparotomy due to abdominal pain and a mass in the lesser pelvis. The authors performed hysterectomy along with bilateral adnexectomy and omental resection. The histological examination of the specimens verified an endometrial adenocarcinoma formed on the ground of adenomyosis and the endometrial adenocarcinoma of the left ovary. **Conclusion:** The malignant transformation of endometriosis is rare, and the mechanisms how it develops on the grounds of adenomyosis is currently unclear.

**Key words:** Endometriosis; Adenomyosis; Adenocarcinoma.

## Introduction

The prevalence of malignant transformation of endometriosis is unknown, but about 1% of those who are suffering from it develop a malignancy linked to endometriosis [1-3]. The majority of these malignant tumors originate from the ovaries (~78%), but extragonadal origins are also known [1, 3]. The most common localization within this is the pelvis, followed by the intestinal origin (endometriosis-associated intestinal tumor/EAIT), which usually appears to be in patients between their late 30's and early 50's with symptoms of abdominal pain, flatulence, change of bowel movements, haematochezia, dyschezia, and tenesmus [1, 3-5]. In the recognition of every lesion formed in the place of an endometriosis, the imaging examinations have an important role, such as ultrasound-diagnostics and MR scan, but the gold standard diagnostic procedure is laparoscopy (or in cases laparotomy) [4, 6]. Malignancies formed in the place of endometriosis have mostly low malignant potential and in cases of genital occurrence are usually contained within the anatomic borders of the given organ [1, 3]. The two most frequent histological forms are clear-cell and endometrioid carcinomas [2, 6]. Endometrial stromal sar-

coma formed in the place of endometriosis is a very rare entity [2, 7].

## Case Report

A 73-year-old female patient in menopause for 23 years presented with abdominal pain and frequent need to urinate, and these symptoms mandated a physical examinations. Endometriosis was not known in her medical history. Clinical examinations revealed a mass in the lesser pelvis reaching up to the umbilicus. Free abdominal fluid was not visible. An elevated CA-125 value (238 U/ml) was noted in the laboratory examinations.

The patient underwent laparotomy in the present department. A small uterus was visible with an intact right and a cystic left adnexum. During the removal of the left adnexal tumor, a significant amount of brownish fluid emptied from it. Total hysterectomy and right adnexectomy was carried out. Cytological sampling of the free abdominal fluid and resection of the greater omentum were also performed.

A right ovary macroscopically containing no abnormal parts, the greater omentum, the 6.5 cm long uterus, and a 9×4.5×7-cm left cystic adnexum were examined. While opening the uterus and next to the atrophic endometrium, an adenomyoma with a diameter of two cm was visible in the myometrium. During the microscopic examination of the area, invasive endometrioid adenocarcinoma foci were verified in place of adenomyosis. Immunohistochemical examination showed estrogen receptor (ER),

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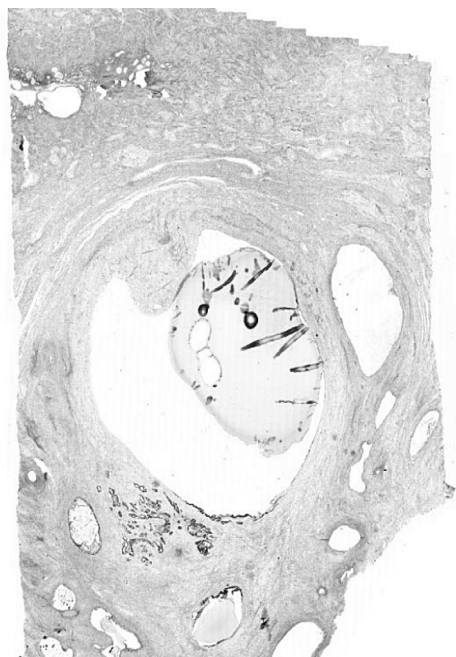


Figure 1. — Low power magnification of MIB-1 stained slice of the uterus showing atrophic endometrium, extended adenomyosis, and MIB-1 positive endometrial adenocarcinoma nests associated with adenomyosis at a distant location from the orthotopic endometrium.

progesterone receptor (PR), cyclin-dependent kinase inhibitor 2A (p16), tumor protein p53, and MIB1 positivity (Figures 1, 2). The right adnexum showed no pathology, however in the left ovary endometrioid ovarian adenocarcinoma was found along with a cystadenofibroma. The omentum and the free abdominal fluid sample showed no malignancy. Following surgical treatment, the tumor board recommended chemotherapy (taxol-carboplatin) [8].

### Discussion

The relationship between adenomyosis and endometrial cancer has been examined by several preceding publications [9, 10]. The cumulative occurrence of the two pathologies in patients who underwent hysterectomy is varies between 10% and 70% [10]. The diagnosis is often late due to the tumor free eutopic endometrium.

The exact mechanism of the formation of endometrial cancer is not known in the case of patients suffering from adenomyosis. Malignant transformation and other factors such as hormonal and growth factors, inflammation, genetic predisposition, the changing of the immune system, and oxidative stress play a role in carcinogenesis [1, 3, 10]. Age, infertility, and duration of the disease can also function as co-factors in malignancy [10]. Opposed to endometriotic cells, explicit p53 expression was described in atypical endometriotic cells, which can have a significance in the recognition of endometrial lesions with premalignant potential [11].

In the present case, a low-grade tumor, showing ER+,

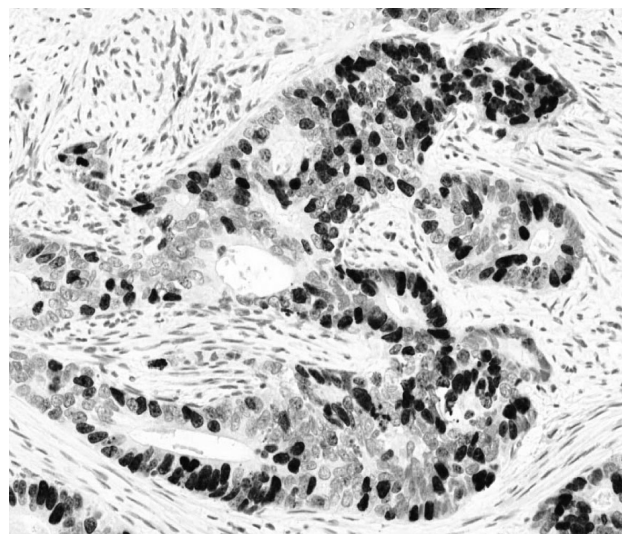


Figure 2. — High power magnification of MIB-1 positive adenocarcinoma cells.

PR+, p16, p53, and MIB1 positivity has been presented without any hormonal anamnesis.

Based on previously published cases, the endometrioid adenocarcinomas formed in the place of adenomyosis are low grade, mostly ER+, and hormone-sensitive tumors with good prognosis [10]. Although years of hormone therapy (mostly estrogen monotherapy) is listed in most malignancies formed in the place of endometriosis, an unequivocal cause and effect relationship between the hormone substitution and the carcinogenesis is currently unknown [1, 3]. Because of the rarity of such cases, it is highly recommended to gather and publish all available data of similar diseases to provide grounds for better understanding of its development, as well as to establish appropriate therapeutic modalities for these kinds of malignancies.

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