

## Case Report

# Malignant transformation of abdominal wall endometriosis with lymph node metastasis

## Case report and review of literature



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## Introduction

Endometriosis is defined as the emergence and growth of endometrial tissue (glands and stroma) outside of the uterus. It affects 8–15% of the population and is most frequently found in the ovary. Extrapelvic implantation may occur in any organ or tissue, but is rare in the abdominal wall. When it does occur there, a history of surgery, whether laparotomy or laparoscopy, is usually present.

Malignancy of this type of endometriosis in an abdominal wall scar may occur from just a few months until up to 18 years after surgery (Hensen et al., 2006). The case we discuss below deals with the malignant transformation of cesarean scar endometriosis with nodal metastasis at preperitoneal level. This is interesting because no other known cases have ever been reported.

## Case presentation

We herein present the case of a 49 year-old patient with a history of appendectomy, two births, a cesarean section, and removal of the endometriotic mass in the cesarean scar, 15 years before.

The patient was seen for her routine annual gynecologic examination, in which she reported the presence of a painful abdominal mass, which had increased in size during the previous few months. She described the pain as cyclical, corresponding to her menstrual cycles, and that it reminded her of the lesion for which she had previously been operated on. Her check-ups prior to this consultation had been normal and no abdominal nodule had been observed.

A physical examination revealed a mass located in the midline of the abdominal wall, 3 cm above the Pfannenstiel scar. The size of the lesion was roughly 6 cm × 6 cm, and it was painful upon movement and was Fothergill sign positive. The rest of the gynecological examination was normal.

Ultrasound scan and mammography were normal. Soft tissue ultrasound (abdominal wall) revealed a lesion measuring 70 × 60 mm located in the abdominal wall, confined within the mesogastrium, caudal to the umbilical region, compatible with fibrous soft tissue tumor (desmoid tumor). Due to the history of the patient we were not able to rule out endometriotic implants. Fine-needle aspiration was performed with pap smear revealing a benign process compatible with endometriosis (Fig. 1).

Tumor marker levels were: CA-125: 243 U/mL and CEA: <0.5 ng/mL. With a diagnosis of abdominal wall endometriosis, surgery was scheduled for removal of the wall tumor and exploration of the abdominal cavity.

## Surgical procedure

The intraoperative pathology of the abdominal wall nodule revealed a mass located at the preperitoneal level with histopathologic result of serous papillary adenocarcinoma (Fig. 2). During the exploratory laparotomy two preperitoneal nodules were observed in the right iliac fossa and another two in the left iliac fossa, which were resected and observed to be nodal metastases of serous papillary adenocarcinoma (two right and one left). The surgery proceeded with bilateral adnexectomy, omentectomy, a removal of the endometriotic mass located in the uterine fundus, endometrial biopsy, and a thorough assessment of the abdominal cavity with intraoperative examination of all the specimens, none revealing neoplastic infiltration.

The case was evaluated by the Gynecology and Oncology Committee, which decided that an extension study with a PET/CT was to be

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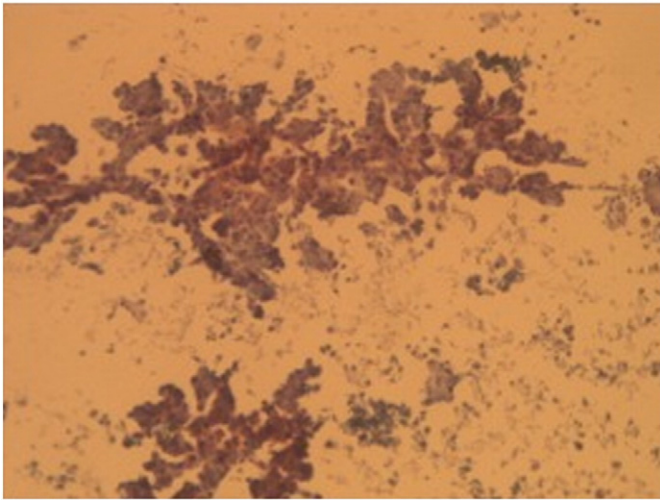


Fig. 1. Citological study.

performed. Two hypermetabolic images were observed, one at the level of the abdominal wall mass and another one in the splenic flexure. Surgical laparoscopy was performed with removal of the splenic flexure, which was reported as steatonecrosis with chronic inflammation. The fibrotic area in the abdominal wall mass was also resected, with the pathology providing a benign result as well.

Subsequently, adjuvant treatment was administered with 6 cycles of carboplatin and Taxol, plus quarterly follow-ups for clinical examination and laboratory tests (CA-125), and semi-annual follow-ups for radiological tests. After a 48-month follow-up the patient is disease-free.

## Discussion

The probability of developing endometriosis in a surgical scar is roughly 0.03–1% (Hensen et al., 2006; Blanco et al., 2003) and malignant transformation is likely to occur in 0.3 to 1% of the cases (Matter et al., 2003; Leng et al., 2006), 80% of which are located in the ovary. The principal risk factors of malignant transformation of endometriosis include: advanced age of the patient, if they are postmenopausal, and if the tumor diameter of an endometriotic lesion is >9 cm (Kobayashi et al., 2007). The probability that it exclusively affects the rectus abdominis muscles without peritoneal infiltration or aponeurosis is also rather

low, there have only been 18 well-described cases, and there haven't been any cases published at all documenting extraperitoneal nodal metastases (Gianella et al., 2010).

Abdominal scar endometriosis presents as a mass in the abdominal wall, usually adjacent to a scar from previous surgery, and is painful, with or without increased size of the scar. The abdominal pain described by the patient is usually cyclical, correlating with her menstrual cycles, if hormonal reserve is still present (Hensen et al., 2006).

Surgical history must be taken into account for the proper diagnosis. Imaging techniques can also be useful. As a first choice, soft tissue ultrasound is recommended, complementing it with ultrasound-guided fine-needle aspiration of the mass. When there is a suspected diagnosis, as in our case (due to the history of the patient), serum levels of CA-125 are requested. Non-invasive diagnostic tests have a very low sensitivity in such cases.

In spite of all these tests, the definitive histopathological diagnosis was serous papillary adenocarcinoma.

The differential diagnosis with palpable masses in the abdominal wall should include: hernia, hematoma, lymphadenopathy, lymphoma, lipoma, abscess, subcutaneous cyst, neuroma, soft tissue sarcoma, desmoid tumor or metastasis.

The most frequent extragonadal sites of malignant endometriosis are (>50% of the cases): the rectovaginal septum, the colon and the vaginal wall (Bats et al., 2008). In our case, the tumor was located in the preperitoneal space, affecting the rectus abdominis muscles. Therefore, the Fothergill sign was not useful in differentiating it from a rectus sheath hematoma or any other pathology exclusively affecting the abdominal wall muscles.

The most interesting aspect of the case is the nodal metastases, since it is the first case described in literature. Being a preperitoneal tumor, the nodal metastases follow the pattern of tumor spread that affects the abdominal wall muscles. When the primary lesion is located in the infraumbilical region, these metastases drain to the superficial inguinal chains. When the lesion is in the supraumbilical region, nodal metastatic spread may drain to the axillary chains.

CA-125 levels in the blood might be slightly elevated however it is a less specific biomarker. Many studies have evaluated different biomarkers for predicting or excluding endometriosis.

Recently, one interesting study showed that CRP serum levels were significantly higher in patients affected by endometriosis than in healthy patients. Similarly, other studies have shown that patients with mild endometriosis had lower anti-Müllerian hormone levels compared to patients without disease.

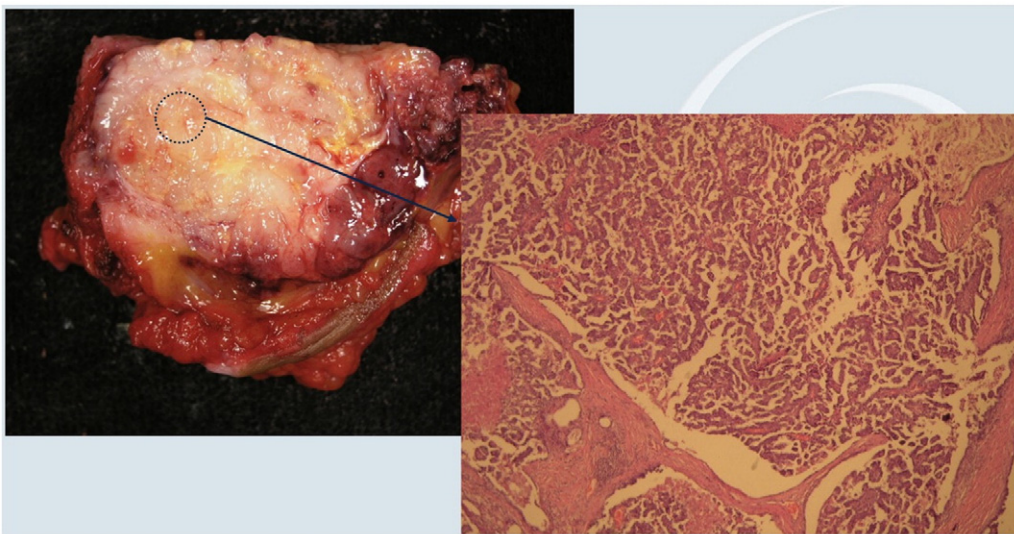


Fig. 2. Pathological study of the tumor (macroscopic and microscopica study).

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