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Case Report
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Endometrioid Adenocarcinoma after Atypical Rectosigmoid Endometriosis – A Case Report

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Abstract

Introduction: Malignant transformation of endometriosis is rare; however, studies have shown a correlation between endometriosis and some types of cancer.

Clinical Case: A case involving a 42-year-old primiparous patient with a metastatic intestinal endometrioid adenocarcinoma diagnosed due to a hemorrhagic acute abdomen surgery. This patient had a history of previous conservative surgery for leiomyomata, adenomyosis and endometriosis 2 years before, and 38 weeks delivery a year before this endometrioid adenocarcinoma diagnosis. The histological report of the first surgery revealed atypical endometriosis within the intestine.

Discussion: The most common extragonadal sites associated with endometriosis and cancer are the pelvic peritoneum, rectovaginal septum, vagina and colorectal serosa. Among the intestinal tumor sites associated with endometriosis, the rectosigmoid colon is the most common site.

Keywords: Carcinoma; Endometrioid; Endometriosis; Infertility

Introduction

Malignant transformation of endometriosis is rare, and the most common site is the ovary; however, studies have shown a correlation between endometriosis and breast, thyroid, and colorectal cancers, among other types [1].

Herein, we report a case involving a patient with endometriosis who presented hemorrhagic acute abdomen with a histopathological and immunohistochemical diagnosis of colorectal Endometrioid Adenocarcinoma (EA).

Case Presentation

A 42-year-old woman was admitted to the emergency department with intense and diffuse abdominal pain, more. Physical examination revealed diffuse abdominal decompression pain with greater intensity in the left iliac fossa. The patient underwent computed tomography of the total abdomen in the coronal view, revealing a moderate amount of peritoneal fluid with different densities and expansive formation indistinguishable from the sigmoid colon, which had a heterogeneous density and hematic areas.

Exploratory surgical laparotomy was promptly indicated and performed. During the procedure, a large hematic mass with clots was observed extensive and bloody infiltrative lesions were present in the mesosigmoid, with infiltration to the intestinal wall. The intestinal tract contained necrotic nodules, some of which were hardened while others were friable, with active bleeding. Radical rectosigmoidectomy was performed with a 15 cm extension, and the entire macroscopically compromised mesocolon was removed, with Hartmann colostomy on the left flank. The patient was progressed without complications and was discharged five days after the surgical procedure.

Histopathological analysis with Hematoxylin and Eosin (H&E) diagnosed a well-differentiated EA morphology and a papillary architecture, within the rectosigmoid and mesocolon, infiltrating the intestinal wall (Figure 1). Immunohistochemistry was compatible with EA with preserved immunoexpressing of DNA repair enzymes (MLH1, MSH2, MSH6 and PMS2), p53 (+) (wild-type, nonmutated), CK7 antibody (+) and estrogen and progesterone receptors (+) (Figure 1).

The results of germline genetic testing for BRCA 1 and BRCA 2 genes from peripheral blood samples in a panel for breast and ovarian cancer did not reveal pathogenic variants or likely

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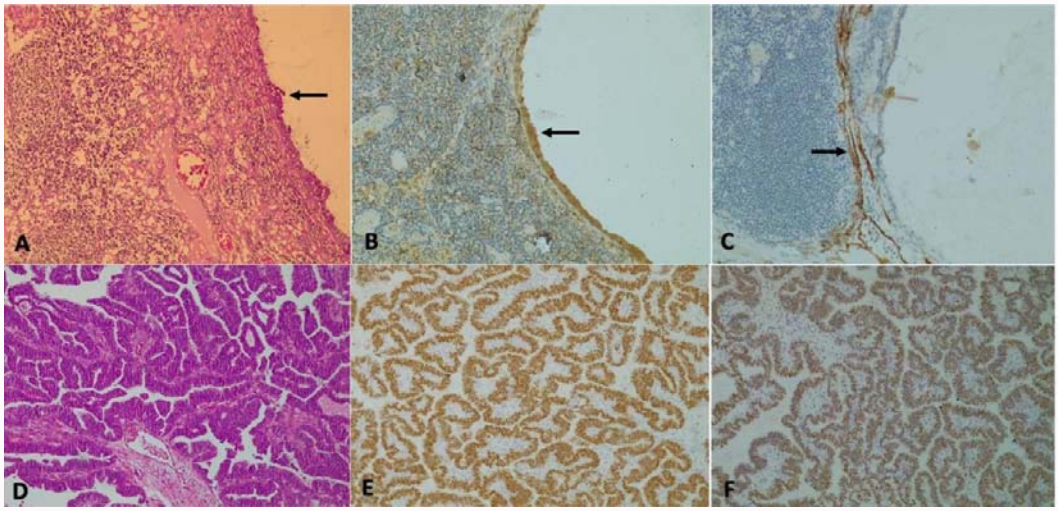

Figure 1: A) Photomicrograph of lymph node tissue affected by endometriosis reactivity for the CD10 antibody in lymph nodes (arrow indicates positive stromal component). CD10, 20x. D) Photomicrograph of papillary formations typical of endometrioid carcinoma detected in a rectosigmoid specimen, HE, 20x. E) Photomicrograph of positive immunohistochemical reactivity for the PAX8 detected in a rectosigmoid specimen, PAX8, 20x. F) Photomicrograph of positive foci with mild atypia (arrow indicating part of the cystic epithelial component). HE, 20x. B) Photomicrograph of positive immunohistochemical reactivity for the estrogen receptor antibody in lymph nodes (arrow indicates positive epithelial component) ER, 20x. C) Photomicrograph of positive immunohistochemical reactivity for the estrogen receptor antibody in endometrioid carcinoma detected in a rectosigmoid specimen, RE. 20X.

pathogenic variants of undetermined significance or copy number variations.

A PET-CT scan with FDG-18F showed hypermetabolism in hepatic lesions, the perirectal peritoneum and abdominal and pelvic lymph nodes, in addition to a slight increase in metabolism in a small pleural effusion on the right (Figure 2). Based on these data, the chemotherapy protocol for ovarian and endometrial cancer (paclitaxel 175 mg/m^2 and carboplatin AUC 5) was indicated and started. The oncology service recommended total hysterectomy, bilateral adnexectomy and lymphadenectomy, which were performed without complications and with pathological anatomy without atypia.

Regarding personal history, two years before the EA diagnosis, she had pelvic pain and infertility, this patient was then submitted to a laparoscopic procedure to remove fibroids, deep endometriosis and segmental rectosigmoidectomy. The histological report indicated endometriosis and leiomyomas without atypia. Also, bowel endometriosis was diagnosed with an area of atypia measuring 0.3 cm. The surgical margins were free, and 11 isolated lymph nodes were identified within the bowel segment, one of which was compromised with atypical endometriosis.

After seven months of the elective surgery for endometriosis, the patient became pregnant and developed oligoamines at 38th weeks, resulting in a cesarean delivery. Family history included paternal grandparents and maternal parents with lung cancer and cervical cancer.

This case was submitted to the ethics committee (CAAE: 50592921.7.0000.5085) and approved under opinion No. 4,909,625.

Discussion

Herein, we present a clinical case involving a patient in menacme with a history of myoma without atypia and deep endometriosis with intestinal and lymph node involvement, the latter two of which showed atypical endometriosis, who underwent surgical treatment and progressed with pregnancy and later sigmoid EA with hepatic, perirectal, abdominal and pelvic lymph node metastasis.

Similar cases in menopausal and postmenopausal patients have been reported, but in this report, the adult patient was 42 years old, was in menacme and had no family history of intestinal cancer [2,3]. Some cases in the literature show progression from endometriosis to EA associated with hereditary nonpolypoid colorectal cancer and Lynch syndrome, but the patient herein reported has no family history of these two conditions. At the age of 40, the patient presented with atypical endometriosis in the rectosigmoid and lymph node, after what radical surgery was not performed due to the patient's desire for pregnancy [3,4]. The results of germline genetic testing for *BRCA1* and *BRCA2* genes in a panel for breast and ovarian cancer did not identify pathogenic variants or likely pathogenic variants of undetermined significance or copy number variations, indicating that the patient does not carry a gene with a genetic mutation for an inherited predisposition to the development of breast or ovarian cancer.

During emergency surgery due to hemoperitoneum, freezing biopsy was not performed due to the urgency of the case; however, the Histopathological diagnosis (H&E) indicated an EA. Pathological staining *via* immunohistochemistry for CK7, CK20, estrogen receptors is useful to distinguish between endometriosis-related adenocarcinoma and primary intestinal adenocarcinoma [2,4]. Among primary colon adenocarcinomas, 75% to 95% have a CK7-negative and CK20-positive phenotype, while 80% to 100% of endometrioid adenocarcinomas have a CK7-positive and CK20-negative phenotype [1]. In the clinical case reported herein, immunohistochemistry revealed that the samples were CK7 (+), estrogen receptor (+) and CK20 (-), suggesting that this case was of gynecological origin, which was supported by the normal appearances of the uterus and its appendages during emergency surgery and exams. Suspected colonic endometriosis of gynecological origin led to initiation of a chemotherapy regimen for gynecological cancer.

Another finding favoring rectosigmoid endometriosis as the cancer origin in this clinical case is the fact that the biopsy from the

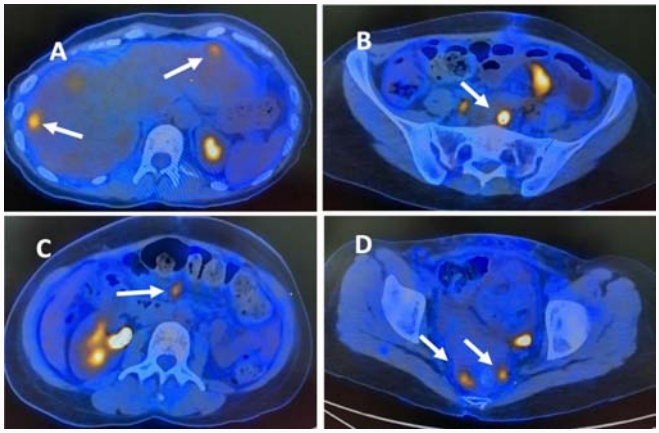

Figure 2: PET/CT with FDG-18F. A) Hepatic nodular lesions (arrows) with hypermetabolism. B) Presacral lymph node enlargement on the left with hypermetabolism. C) Lymph node in the mesogastrium with hypermetabolism. D) Perirectal fat densification and the presacral region associated with hypodense lobulated tissue with hypermetabolism.

previous surgical treatment of endometriosis showed endometriomas without atypia, and atypical endometriosis in the intestinal wall and in a mesocolon lymph node. Atypical endometriosis is associated with increased recurrence and malignant transformation of ovarian endometriosis [5]. The diagnosis of endometriosis with architectural atypia is important because it may be a precursor lesion of ovarian cancer; therefore, pathologists who identify endometriosis should carefully examine samples to further identify hyperplasia-type endometriosis, as patients with these conditions may have a higher risk of developing endometriosis associated with ovarian cancer [6,7]. However, this case study shows that the atypical pathology of malignant transformation occurred in the rectosigmoid colon and mesocolon.

In a systematic review and meta-analysis of cancer associated with endometriosis, regarding the association of endometriosis with colorectal cancer, five studies were evaluated. This finding indicates that although rare, an association may exist between endometriosis and colorectal cancer [8].

In a study following 37,434 participants from 1986-1989 in which 3.8% reported endometriosis at the beginning of the study, endometriosis was not associated with a high risk of colorectal carcinomas [9]. A similar finding was found in another long study [10], reinforcing the rarity of the clinical case presented in this report.

In the clinical oncological evaluation, the patient presented unresectable metastatic disease based on PET-CT showing mild hepatic hypodensity, with increased metabolism related to secondary neoplastic involvement, hypermetabolism in the lymph nodes and abdominal and pelvic lymph node enlargement, probable neoplastic involvement, hypermetabolism in peritoneal fat, which was most evident adjacent to the rectum, probable secondary neoplastic involvement, and a slight increase in metabolism in a small undetermined pleural effusion on the right. Immunohistochemistry confirmed a tumor of gynecological origin, which was possibly due to malignant transformation of endometriosis infiltrating the intestine from the outer layers to the muscle. Palliative treatment with a chemotherapy protocol to treat both ovarian and endometrial cancers was indicated: 175 mg/m^2 paclitaxel and carboplatin AUC 5 every 21 days for a total of six cycles, with a response evaluation after the 3rd cycle [2]. The patient in this clinical case showed excellent tolerance.

Conclusion

The disease evolution reported here is rare. The initial objective was to describe the relationship of atypical endometriosis and its evolution to cancer. However, the patient died three years after the diagnosis of EA.

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