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A case of clear cell adenocarcinoma arising from endometriosis of the rectum treated by laparoscopic surgery

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ABSTRACT

INTRODUCTION: Malignant transformation of intestinal endometriosis occurring in the extraovarian sites is extremely rare. We report a very rare case of clear cell adenocarcinoma arising from endometriosis of the rectum.

PRESENTATION OF CASE: An 83-year-old woman was admitted with the complaint of hematochezia. Colonoscopy revealed a tumor around about half of the rectal circumference. Biopsy of the tumor revealed a well-differentiated adenocarcinoma. Low anterior resection was undergone laparoscopically under the diagnosis of rectal carcinoma. Histopathological examination revealed clear cell adenocarcinoma, invading the sub-serosa of the rectum, but no metastasis of the lymph nodes. Immunohistochemical staining showed strong positivity for cytokeratin 7, but no staining for cytokeratin 20 and CDX2. The tumor existed adjacent to the endometrial glands, which were stained positive for Estrogen receptor. Ultimately, the patient was diagnosed with clear cell adenocarcinoma arising from endometriosis. Eighteen months after surgery, there are no signs of tumor recurrence.

DISCUSSION: Clear cell adenocarcinoma arising from intestinal endometriosis has been reported in 7 cases, including our case. Careful observation is required because the prognosis of endometriosis after malignant transformation remains poor.

CONCLUSION: We report a very rare case of clear cell adenocarcinoma arising from endometriosis of the rectum treated by laparoscopic surgery.

1. Introduction

Malignant transformation in endometriosis is rare. While the precise incidence remains unknown, a 0.3–1.0% incidence has been reported.[1] About 75% of the cases of malignant transformation have been reported to occur in the ovary, with the remainder occurring in extragonadal sites, most commonly the pelvic peritoneum, rectovaginal septum, colon/rectum, and vagina.[2] About 70% of the histological types of endometriosis are endometrioid adenocarcinoma. The incidence of clear cell adenocarcinoma in the extraovarian sites of endometriosis is 4.5%.[2–5] This paper reports a very rare case of clear cell adenocarcinoma arising from endometriosis in the rectum.

2. Presentation of case

An 83-year-old woman was admitted in June 2012, with the complaint of hematochezia. She has not suffered from endometriosis, and has not been treated with any hormonal agents.

Preoperative serum tumor-marker, carcinoembryonic antigen (CEA), carbohydrate antigen 19–9 (CA19-9), carbohydrate antigen125 (CA125) levels were not elevated. Barium enema showed a 45-mm-long polypoid lesion, accompanied with a trapezoidal change, located at the anterior wall of the rectum (Fig. 1). Abdominal computed tomography showed a heterogeneous enhanced wall thickness of the rectum (Fig. 2a). No metastasis was found in the liver, and lymph nodes in the mesorectum were not swollen. Pelvic magnetic resonance imaging showed a heterogeneous intensity mass in the rectum on T1 weighted imaging, with no suspicion of invasion to the uterus and sacrum (Fig. 2b). Bilateral ovaries were not swollen. Colonoscopy revealed tumor sizes around half of the rectal circumference, at the upper rectum, 13 cm from the anal verge. Biopsy of the tumor revealed a well-differentiated

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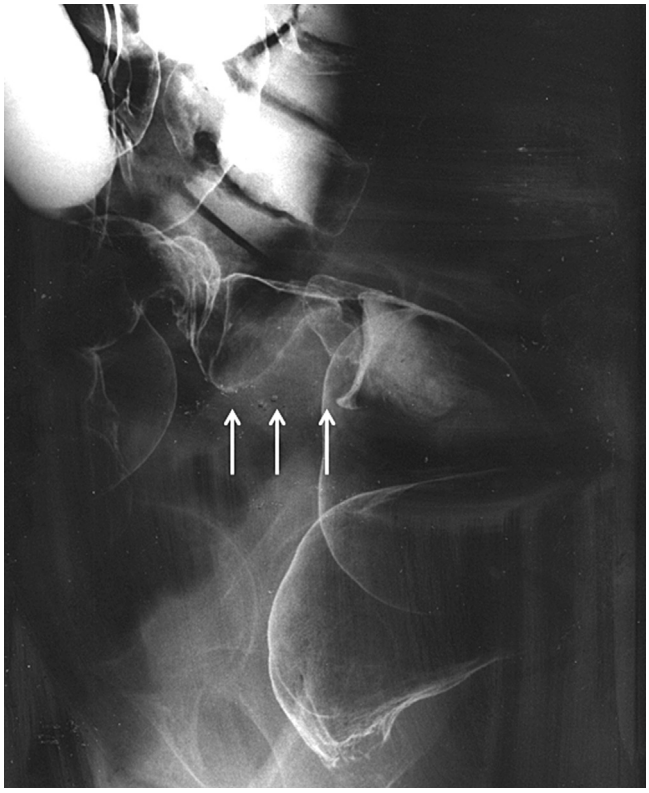

Fig. 1. Barium enema showed a 45-mm-long polypoid lesion (arrow), with a trapezoidal change at the anterior wall of the rectum.

adenocarcinoma. Although, it was histologically atypical of a carcinoma of the colon, we could not perform the further histological examination. Consequently, we diagnosed the patient as having rectal carcinoma, and planned to perform laparoscopic low anterior resection. Under general anesthesia, the first port for the camera was created at the umbilicus. Next, a 12 mm port was inserted through the right lower quadrant, three 5 mm ports were placed at the right upper, left upper and lower quadrants, allowing placement of a total of 5 ports. Upon observation of an intraperitoneal cavity during laparoscopy, metastatic liver tumor, peritoneal tumor, and swollen lymph nodes were not recognized. Both ovaries appeared normal. The rectal tumor was partially adherent to the visceral peritoneum of the left side of the uterus. It was suspicious that the tumor invaded to the peritoneum. Thus, the adhered part was resected. Frozen section was performed and demonstrated no invasion to the peritoneum. After that, we proceeded to dissect the mesorectum circumferentially, and resect of the distal rectum by the endoscopic liner stapler. Reconstruction was done with the circular stapler, using the method of the double stapling technique (DST). In the resected specimen, a polypoid ulcerated tumor, measuring 38 mm × 38 mm in diameter (Fig. 3). Histopathological examination showed that clear cell adenocarcinoma was contiguous with endometriosis, infiltrating through the sub-serosa (Fig. 4a and b). In immunohistological staining, the tumor was positive for CK7, and negative for CK20, CDX2, Estorogen receptor (ER), progesterone receptor (PgR). Endometrial glands and stroma adjacent to the tumor were positive for ER (Fig. 4c and d). Finally, it was diagnosed as clear cell adenocarcinoma arising in endometriosis of the rectum. No metastatic lymph nodes were recognized pathologically.

After surgery, she did not receive any chemotherapy of her own will. At 18-month follow-up, there was no evidence of recurrence.

3. Discussion

While adenocarcinoma arising in endometriosis has occasionally been reported in the ovaries, there has been a report that the rate of malignant transformation in endometriosis is 0.7–1.0%, with 80% occurring in the ovary.[1] However, malignant transformation of endometriosis at extragonadal sites remains rare; the rates of occurrence are 5.7%, rectovaginal septum are 4.3%, colorectum are 4.3%.[1] Moreover, the malignant transformation of clear cell adenocarcinoma occurring in the extraovarian sites continues to be extremely rare; with an incidence of 4.5%.[1–5,7–8] Malignant transformation in endometriosis was first described by Sampson in 1925,[9] who recommended that three

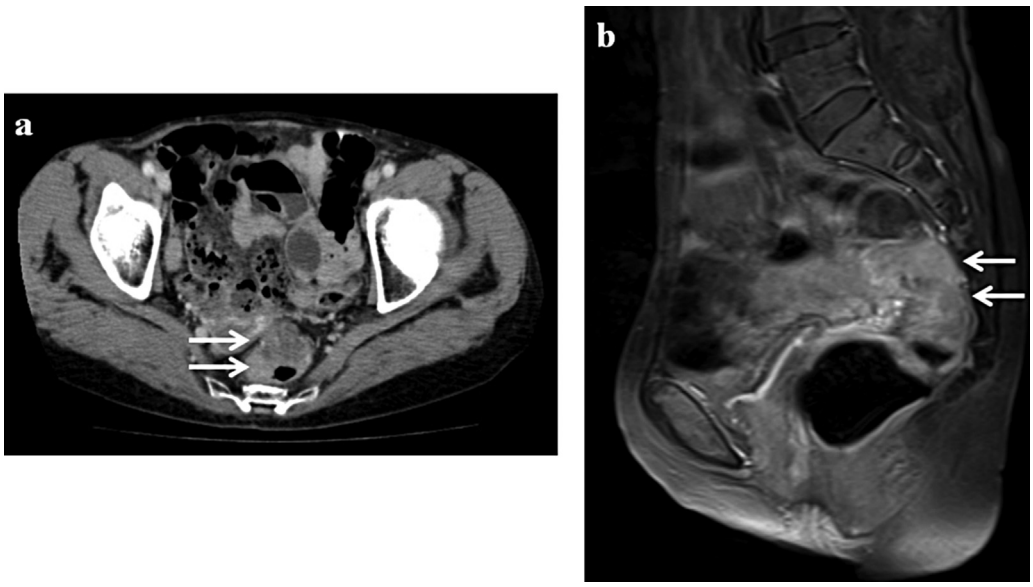

Fig. 2. Abdominal computed tomography examination showed enhanced wall thickness of the rectum with no suspicion of invasion of the surrounding organs (arrow). There was no sign of swelling of the lymph nodes in the mesorectum (a). Pelvic magnetic resonance imaging (T1 weighted image) showed a heterogeneous intensity mass of the rectum, with no suspicious uterus or sacrum (b).