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Letters to the Editor

Endometrioid adenocarcinoma in the lower rectum

Key words: Endometriosis. Endometrioid adenocarcinoma. Rectal cancer. Rectorrhagia.

Dear Editor,

Endometriosis is a common disease that usually involves the ovary (1) and generally has a benign course (2). Malignization is exceptional (fewer than 1 %), affecting the rectum in only 5 % of reported cases (2).

Case report

A 39-year-old woman with an uneventful history presents with abdominal pain in the left iliac fossa, weight loss, and rectorrhagia lasting 6 months. A mass effect is palpated 6 cm away from the anal margin during digital examination. CA125 is elevated (522.2 U/mL) whereas CEA and CA19-9 remains normal. Colonoscopy reveals friable mucosa at 7 cm from the anal margin, which suggests extrinsic compression and precludes echoendoscopy because of stenosis. Biopsies were negative for malignancy. An abdominal CT scan unveils a big, partially necrotized rectal growth, perirectal adenopathies, and uncertain uterus infiltration (Fig. 1). Finally, transvaginal ultrasound shows a vascularized solid mass, 65x56x68 mm in size, of likely digestive origin.

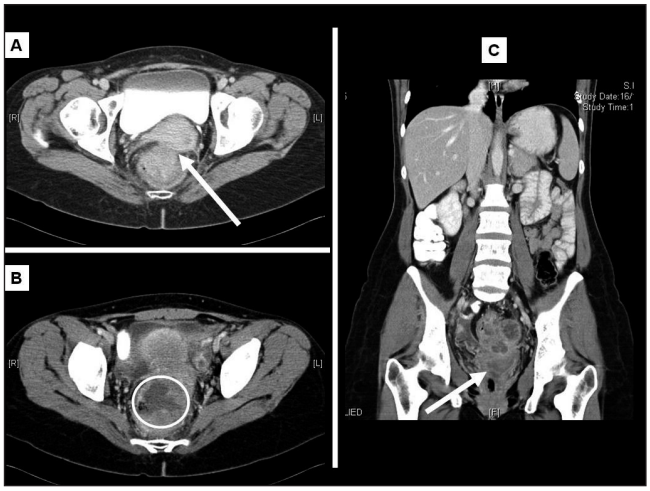

Fig. 1. Abdominopelvic CT scan showing a mass (arrow) at the mid-lower rectal level, seemingly infiltrating the uterine body (A). The lower picture (B) is an axial slice where extensive tumour necrosis areas (circle) may be seen. The right-side picture (C) shows a coronal section through the pelvis and the tumour in the mid-lower rectum.

In the absence of a histological diagnosis an exploratory surgery is decided upon. Following the opening of the peritoneal reflection a tumour is observed in the mid-lower rectum, and a rectal anterior resection is performed. Histopathological examination documents a poorly-differentiated, infiltrating, endometrioid adenocarcinoma over endometriosis sites within the rectal muscularis layer with intestinal wall infiltration (Fig. 2A), nodal metastases, and a cytokeratin 7-positive and cytokeratin 20-negative immunohistochemical pattern with estrogen receptors positivity. In view of these results surgery is completed with hysterectomy and double annexectomy, and the presence of an endometrioid adenocarcinoma infiltrating the uterine wall from the rectum by contiguity is confirmed, with no endometriosis sites in the uterus (Fig. 2B).

The postoperative course was uneventful; the patient received adjuvant radio-chemotherapy following discharge and remains disease-free after one year.

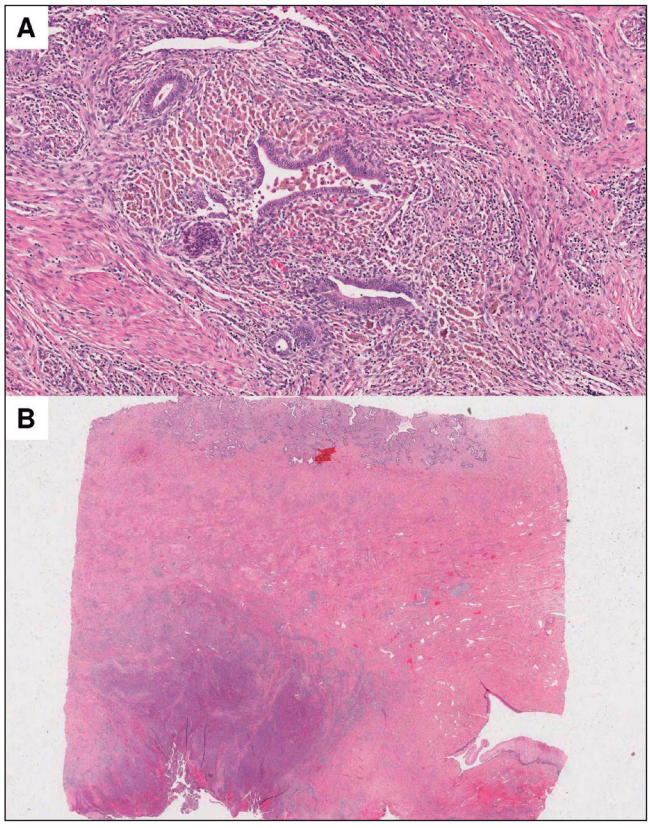

Fig. 2. Endometrioid adenocarcinoma histopathology. A. Endometriosis sites in the muscularis layer of the large bowel, where endometrial-looking glands with hemosiderin-laden macrophages and stroma may be seen. B. Uterus with endometrioid adenocarcinoma infiltrating from the rectum by contiguity. The uterine secretory endometrium with no pathological changes may be seen in the upper part of the image, whereas the poorly-differentiated carcinoma sharing rectal features appears on the lower part.

Discussion

Fewer than 50 cases of intestinal tumours associated with endometriosis have been reported, with endometrioid adenocarcinoma being the commonest variant. Most cases occur in postmenopausal women receiving estrogen therapy following hysterectomy and double annexectomy for endometriosis. The clinical picture usually presents with abdominal pain, metrorrhagia and/or rectorrhagia (3). Tumours are usually found in the sigmoid colon, upper rectum, and rectovaginal septum, being exceptional in the extraperitoneal mid-lower rectum.

The etiopathogenesis is unknown, and in most cases endometriotic implants are secondary to retrograde menstruation (4). Hyperestrogenism is a potential risk factor (5), and genetic predisposition is suggested for cases without it (5). On the other hand, the neurological hypothesis described by Possover and Anaf (6,7) correlates endometriotic implants with sympathetic nervous system distribution; this might account for our patient's case since the mid-lower rectum's location is extraperitoneal and in intimate contact with the sympathetic plexus.

This case meets all the histological criteria defined by Sampson (8) for endometrioid adenocarcinoma, with tumour cells growing from endometriotic foci within the rectal muscularis coat, hence not from invasion from the uterus. Furthermore, the histology of the hysterectomy specimen confirms uterine infiltration by contiguity from the rectum, with no endometriotic sites in the uterus or annexes, and an endometrium with normal characteristics, which rules out a uterine origin.

There is no consensus protocol regarding the treatment of choice. At any rate, the need for radical resection of the involved intestinal segment plus hysterectomy and double annexectomy is indisputable. Adjuvant therapy is warranted by the presence of nodal metastases and uterine infiltration.

To conclude, endometrioid adenocarcinoma of the rectum is a rare tumour that requires a high index of suspicion for diagnosis. Importantly, it should be differentiated from colorectal tumours in order to optimize management and for its prognostic value. Our case is atypical in terms of age at presentation, absence of relevant history, and tumour location.

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