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Case report

About a degeneration of a colon endometriosis cystic foci

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ABSTRACT

Introduction: Malignant transformation of endometriosis is a rare entity. Its occurrence in the digestive system is exceptional. In fact, due to the atypical symptoms, the diagnosis is often delayed at an advanced stage. So, the treatment strategy should be discussed in a multidisciplinary meeting.

Aim: Our objective was to describe a challenging diagnosis of an in situ carcinoma developed on a cystic foci of endometriosis of the colon in a woman without a particular past medical history.

Case study: A 32-year-old female patient presented with abdominal pain for about six months. The different explorations have concluded to an ovarian cystic tumour. Intraoperatively, the mass seemed to be developped in the right colon. This led to perform a right hemicolectomy. Gross findings consisted in a sub-mucosal 10-centimeters cyst and microscopic features were consistent with an in situ carcinoma developed on a cystic endometriosis foci.

Discussion: This case illustrates the malignant potential of endometriosis especially when it is misdiagnosed.

Conclusions: Besides the fact that this case was illustrated by radiological and microsocpic features, it puts emphasis on the non consensual management of a rare lesion of the colon.

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1. INTRODUCTION

Endometriosis is defined as a localization of hormonereresponse endometrium mucosa outside the endometrium. It can be gonadal or extragonadal.[1–3] The frequency of extragonadal endometriosis in the bowel is estimated to involve 3% to 37% of women with pelvic endometriosis and most lesions are found in the sigmoid colon and rectum.[4,5] The malignant transformation of these lesions accounts for 0.3%–1% of the cases and their diagnosis is based on microscopic features. In fact, no clinical or radiological features are specific.

2. AIM

Our objective was to describe a challenging diagnosis of an in situ carcinoma developed on a cystic foci of endometriosis of the colon in a woman without a particular past medical history.

3. CASE STUDY

A 32-year-old female was admitted to the department of surgery for a 6-month lasting abdominal pain. The patient was multiparous with two pregnancies and she had no history of gynecological surgery. The patient did not report a change in the intestinal transit or catamenial change in symptoms. Moreover, she wasn't using estrogen therapy. Gynecological and abdominal examination was normal. Abdominal ultrasound showed a 10-centemeter pelvic mass with dual solidocystic component (Figure 1). Abdominal and pelvic computed tomography (CT) scan showed a heterogeneous pelvic mass measuring 9 cm with a dual component and a significant contrast enhancement at the wall. According to the radiological features, this mass was supposed to be linked to the right ovary. No distant metastasis was detected (Figure 2). Ovarian tumor markers were negative. A subumbilical midline laparotomy was performed and showed a mass of the right colon

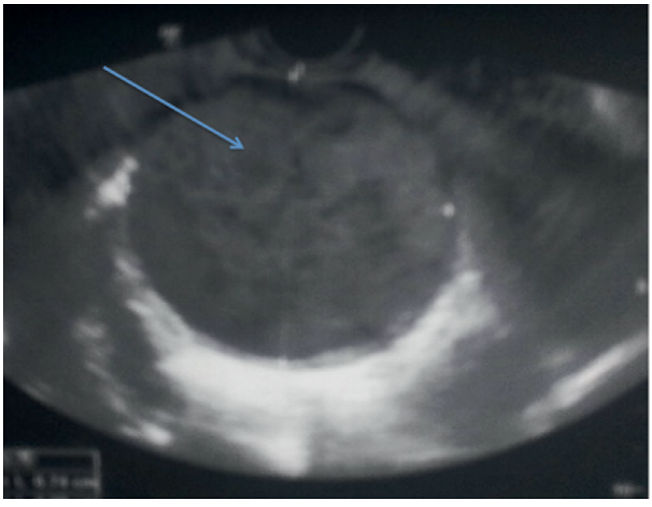

Figure 1. Abdominal and pelvic ultrasound examination showing a pelvic mass with solid and cystic component making 10 cm long axis.

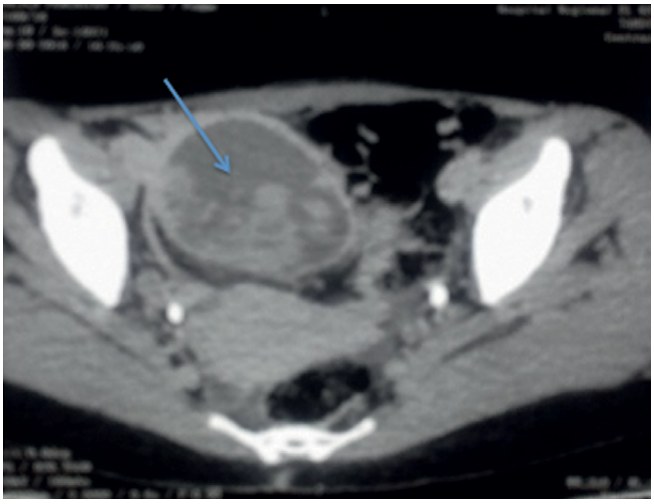

Figure 2. Abdominal and pelvic computed tomography scan showing a heterogeneous pelvic mass of 15 cm long axis, with dual component and significant contrast enhancement at the wall. Its origin was linked to the right ovary.

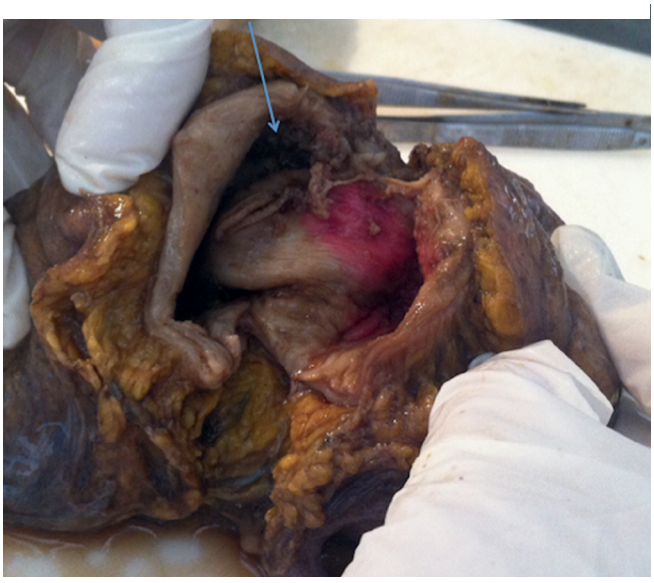

Figure 3. Gross findings showing a 10-centimeter cystic submucosal lesion (arrow).

with multiple lymph nodes of the right meso-colon. The exploration of the rest of the abdominal cavity did not show other abnormalities. The uterus and the adnexa were normal. A right hemicolectomy with a latero-lateral ileocolic anastomosis was performed. Extemporaneous exam was performed on an ileocolectomy specimen characterized by a sub-mucosal cystic lesion measuring 7 cm and filled with necrotic material and hemorrhage (Figure 3). It concluded to a cystic benign lesion. The definite exam showed a colonic mucosa with a submucosal cystic lesion dilacerating the muscularis mucosa, with a normal colonic layer (Figure 4a). The cystic lesion was covered by a unistratified epithelium mimicking endometrium mucosa (Figure 4b). The epithelium was characterized by focal papillary projections (Figure 4c). These formations were line by atypical epithelial cells characterized by hyperchromatic and atypical nuclei. These atypical cells were limited to

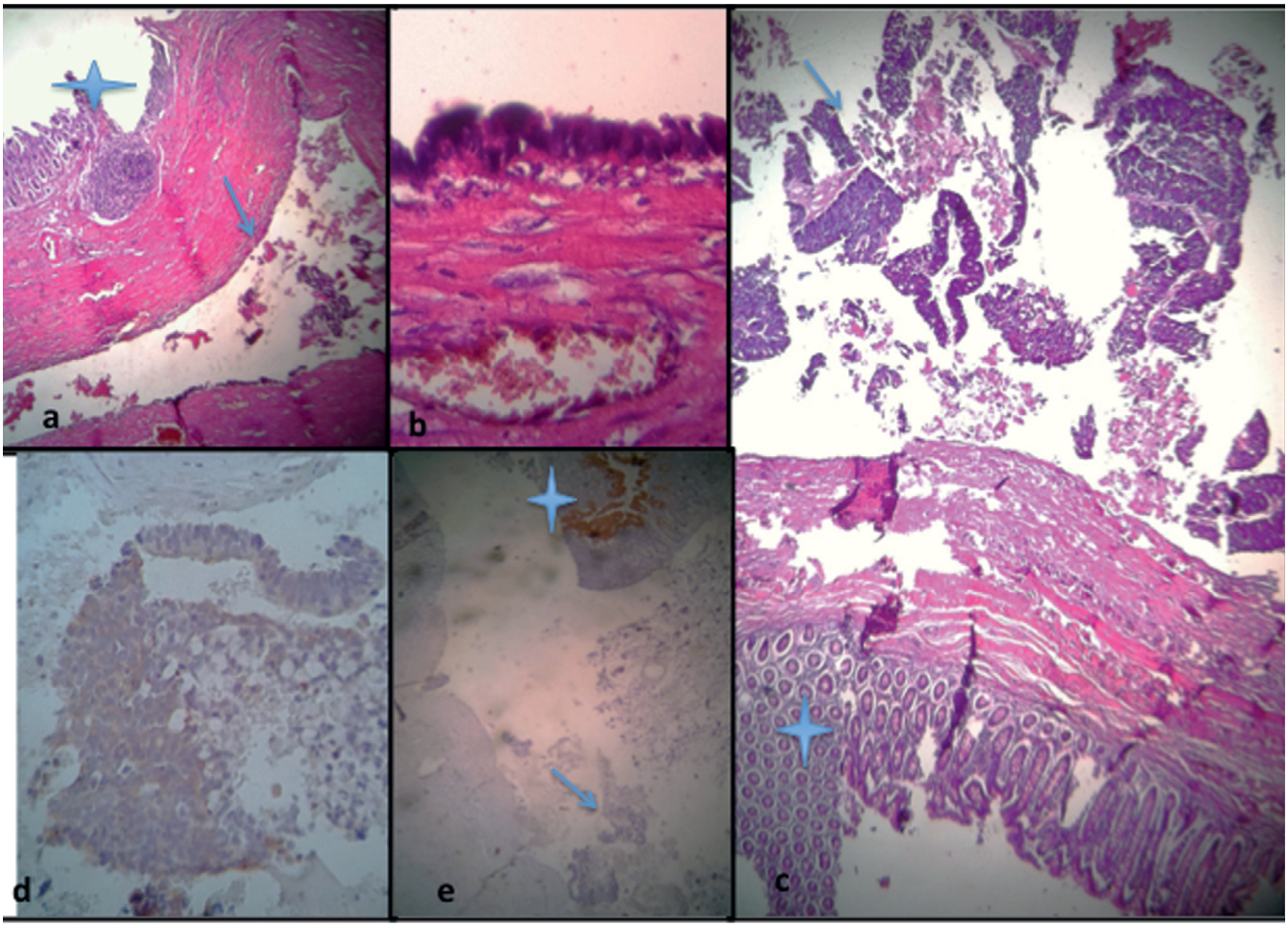

Figure 4. Microscopic findings showing: (a) the localization of the lesion (arrow) beyond the mucosa (star) (HE, ×250), (b) a fibrous cystic lesions lined by atypical cells with hyperchromatic nuclei (HE, ×400), (c) surface cells forming papillary projections (arrow). The upper colonic mucosa is showed using a star (HE, ×400). Immunohistochemical study showing: (d) the expression of CK7 antigen by the tumour cells (HE ×400), (e) the absence of expression of CK 20 antigen by the tumour cells (arrow). The intestinal mucosa expressed the CK20 antigen and highlighted the fiability of the technique (star) (HE, ×400).

the surface and didn't infiltrate the basal lamina. All the cystic lesion were included and studied in order to rule out a possible infiltration. Sixteen lymph nodes were sampled from the meso-colon and were benign. An immunohistochemical study was performed using antibodies againt cytokeratin (CK) 7, CK20, hormonal receptors and mib-1. Tumour cells expressed diffusely and intensely the CK7 (Figure 4d). On the other hand, the tumour cells didn't express the CK20 antigen (Figure 4e). The diagnosis of *in situ* carcinoma developed on a cystic endometrioid foci was retained and a biannual observation of the patient was established without a particular event.

4. DISCUSSION

This case illustrates a rare and challenging cystic lesion of the colon in a young woman without a particular past medical history and especially no endometriosis. The diagnosis of in situ carcinoma developed on a cystic endometriosis foci was established and was particularly challenging.

A few cases of degeneration of endometriosis foci of the colon have been reported in the English literature.[4,5] Almost all patients presenting such a lesion had a past medical history of endometriosis or have a history of cyclic intestinal symptoms during years. Radiologic features aren't specific of these lesions.[6] Transvaginal ultrasound after bowel preparation is reported to be the best initial imaging method for endometriosis, since it can detect foci of deep endometriosis as an irregular hypoechoic mass with or without hypoechoic or hyperechoic foci penetrating into the hypoechoic muscularis propria layer wall. It may also show as long, nodular hypoechoic lesions along the intestinal wall. A number of characteristic appearances of endometriosis of the rectosigmoid area have been reported, including the 'pyramid sign,' the 'comet sign,' and the 'Indian headdress sign.' Ultra-sound examination is also known to be operator-dependent, time-consuming. On contrastenhanced CT imaging, endometrioid foci typically appear as soft-tissue density masses, indistinguishable from other gastrointestinal pathologies either benign or malignant. Magnetic

resonance imaging shows eccentric mass/masses infiltrating into the intestinal wall causing luminal narrowing with associated fibrosis and smooth muscle hyperplasia appearing at times as irregular, speculated, hypo-intense lesions on T2-weighted images. Fat saturated T1-weighted images show a mass or thickening, which is isointense to muscle, possibly with interspersed hyper-intense foci that reflect hemorrhagic blood products.[7] Recently, some authors reported the utility of contrast enhanced MR-colonography in the diagnosis of lesions of endometriosis.[6] Because of the lack of inter-observer agreement and the necessity of very skilled radiologists, the gold standard for diagnosis of endometriosis remains the laparoscopic visualization of suspicious lesions. In our case, the diagnosis was misleading because the lesion was large, cystic and seemed to be linked to the ovary. The colonic localization of the lesion was a peri-operatively discovery. The extemporaneous exam concluded to a benign cystic lesion because of the absence of a stromal invasion and the surgical resection was performed because of the large size of the lesion and in order to avoid complications like intestinal occlusion. Microscopic examination was quite challenging. The diagnosis of endometriosis was easy because of the presence of typical cystic foci of endometrial mucosa. The difficulty in our case was represented by the presence of in situ lesions that were hard to classify. In fact, invasion is defined by an infiltration by the stroma and if present, the lesion is classified as an adenocarcinoma. When establishing the diagnosis of adenocarcinoma, the main challenge faced by pathologists consists in proving the development of such a tumour on lesions of endometriosis and the immunohistochemical study plays a key-role in that case when showing the negativity of CK20 antibody which is quite specific of a primary colonic adenocarcinoma. In our case, there was no infiltration proved despite the inclusion of the totality of the lesion. What was particular to our observation was the atypical cells forming papillary projections on the surface which were characterized by a high proliferative index. This kind of lesion was reported in a patient by Schutz R and colleagues.[8] In fact, they reported a similar non infiltrating lesion classified as an in situ carcinoma. The treatment of digestive endometriosis is mainly based on surgical resection according to many authors.[9] On the other hand, the mainstay treatment of malignant transformation of digestive endometriosis is based on multimodal therapy including surgery resection and chemotherapy. Indeed, only oncologic resection optimizes cure but even though passing on healthy margin, recurrence remains possible. In our case, there was no infiltration, the surgical margins were healthy and all the lymph nodes analysed were benign. These findings made our surgeons and oncologists advocate for a close observation without adjuvant chemotherapy.

Our patient hasn't presented complications since a 2-year follow-up period.

5. CONCLUSIONS

Besides the fact that this case was illustrated by radiological and microsocpic features, it puts emphasis on the non consensual management of a rare lesion of the colon.

Conflict of interest

The authors declare that they have no competing interests and disclose any personal or financial support.

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