CASE REPORT

Endometrioid adenocarcinoma arising from colonic endometriosis

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Introduction

Malignant transformation is a rare but well-documented complication of endometriosis, occurring in 0.7% to 1% of cases. The ovary is involved in 76% of such cases, and extragonadal sites constitute the remaining 24%; the colon and rectum are involved in only 5%. Several previous papers have discussed risk factors for and immunohistochemical studies of malignant transformation of intestinal endometriosis, but few include imaging features. In this report, we demonstrate and discuss the imaging features of a case of malignant transformation of intestinal endometriosis.

Case report

A 58-year-old, gravida 1, para 1 woman was admitted to our hospital complaining of lower abdominal pain. She had undergone a hysterectomy and bilateral salpingo-oophorectomy for severe endometriosis at 47 years of age. At operation, the right ovary was found to be densely adherent to the rectosigmoid colon. She had not received hormone replacement therapy after the operation. On admission, the serum tumour markers showed highly elevated CA 19-9 (4460 U/ml) and CA 125 (154 U/ml).

Barium enema demonstrated involvement of the sigmoid colon by an extracolonic mass. The mucosal surface was slightly irregular (Fig. 1), but colonoscopy revealed no abnormalities of the mucosal surface. MRI of the pelvis showed an intrapelvic pear-shaped mass, similar in signal intensity to sigmoid colon serosa, measuring 6×6×8 cm (Fig. 2). The anterior solid component of the mass was isointense on T1-weighted images and heterogeneously hyperintense on T2-weighted images; it was heterogeneously enhanced after administration of Gd-DTPA. The posterior cystic component was hypointense on T1-weighted images, very hyperintense on T2-weighted images, and was not enhanced by Gd-DTPA. The muscular layer of the sigmoid colon was hypointense on T2-weighted images, and partly interrupted by this mass; the mucosa, enhanced by Gd-DTPA, was continuous and uninterrupted. A malignant tumour such as malignant transformation of intestinal endometriosis, sarcoma, malignant GIST, or extraluminal mucinous cystadenocarcinoma growing outside the intestine was suspected.

An anterior resection of the rectosigmoid colon was carried out. The mass, 6 cm in diameter and densely adherent to the sigmoid colon serosa, was found to be a firm, solid tumour featuring both necrosis and haemorrhage (Fig. 3(a)). The colonic mucosa was intact. The final histological diagnosis was well-differentiated endometrioid adenocarcinoma adjacent to benign endometriosis of the sigmoid colon serosa and muscularis propria, with no invasion into the mucosa (Fig. 3(b)). The patient received five courses of chemotherapy comprising paclitaxel and cis-diaminedichloroplatinum after the operation, and has remained well for 17 months without recurrence. The tumour markers have remained within normal limits.

Discussion

Intestinal endometriosis has been found to comprise 12% of 7177 reported cases; the rectosigmoid
A colon has been found to be the most frequent site in 72.4% of 497 reported cases. The following hypotheses have been proposed concerning the causes of endometriosis occurring in the intestine.

1. According to the transtubal regurgitation and implantation theory, endometrial cells reflux through the Fallopian tubes, flowing out to the pelvic cavity and implanting on the peritoneum or serosal surface of the intestine.
2. The serosal or coelomic metaplasia theory is based on evidence that the serosa of the peritoneum in the embryo develops from the same coelomic epithelium as the endometrium.
3. In the metastatic theory, spread is by lymphatic or haematoogenous dissemination.
4. The direct implantation theory is self-explanatory.
5. The embryonic rests theory is similarly self-explanatory.

According to the most widely accepted transstubal implantation theory, intestinal endometriosis

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**Figure 1** Barium enema study shows that the sigmoid colon is compressed by an extracolonic mass. The surface of the mucosa is slightly irregular.

**Figure 2** (a) Axial T1-weighted image shows a mass similar to sigmoid colon serosa as two components: the anterior isointense and posterior hypointense areas. (b) Axial T2-weighted image shows an anterior heterogeneous hyperintense area and a posterior very hyperintense area. The hypointense muscularis propria of the sigmoid colon is partly interrupted by the mass. (c) Axial gadolinium-enhanced T1-weighted image shows an anterior heterogeneously enhancing area and a posterior non-enhancing area. The enhancing mucosa of the sigmoid colon is uninterrupted.
starts at the serosa and invades the muscularis propria, only occasionally extending into the mucosa.

Malignant transformation of intestinal endometriosis is very rare; fewer than 40 cases have been reported, as small series or single-case reports. Of these, 22 were adenocarcinomas and the remainder included endometrioid stromal sarcomas, mixed Mullerian tumours, clear-cell carcinoma and endometrioid adenofibroma. Our case fulfills all of Sampson's criteria for carcinomatous development in endometriosis, as follows.

1. There were clear examples of the endometriosis in close proximity to the tumour.
2. There were no other primary sites identifiable.
3. Histologically, the tumour and endometrial lesions resembled each other. In our case it is reasonable to assume that endometrial tissue was implanted from the original ovarian endometriosis into the serosa of the sigmoid colon.

Intestinal endometriosis usually mimics the features of a submucosal tumour as long as the lesion is localized in the serosa or muscularis propria. Some reports, describing the appearances of this lesion on barium enema, have shown more asymmetrical constrictions and long filling defects with sharp irregular margins and transverse ridging of mucosa produced by fibrosis of the submucosal layer, compared with primary colonic carcinoma. Because the mucosa is frequently intact or shows minimal changes, the chance of a biopsy positive for malignancy at endoscopy is low, at about 10% to 30%. In addition, a malignant tumour arising from intestinal endometriosis often shows as a submucosal tumour similar to intestinal endometriosis; it may therefore not be possible to distinguish these two lesions at barium enema or endoscopy. In this respect, CT or MRI are very useful in assessment of the whole wall of the intestine. MRI in particular is able to depict the mucosa, muscularis propria and serosa, and in the present case was able to demonstrate that the muscularis propria of the sigmoid colon was partly interrupted by the tumour although the mucosa remained intact. The heterogeneous enhancement of the anterior solid component of the tumour suggested a malignant tumour can be suspected.

When a submucosal or solid intestinal tumour is demonstrated in a middle-aged woman, particularly if there is a history of endometriosis or unopposed oestrogen therapy, malignant tumours arising from intestinal endometriosis should be considered in the differential diagnosis, together with GIST, sarcoma and malignant serosal implants.

References