Isolated sigmoid colon metastasis from a primary fallopian tube carcinoma: a case report

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Summary

Isolated metastasis of primary fallopian tube carcinoma (PFTC) is extremely rare. We describe a case of a 41-year-old asymptomatic woman who was referred three years after the initial treatment for PFTC due to elevated serum CA-125 levels. The abdominal and pelvic CT scans revealed a pelvic mass near the top of the vaginal vault. On surgery, a sigmoid colon tumour was found and a sigmoidectomy was performed. On histopathology the tumour involved the bowel wall from serosa to submucosa, without involvement of the underlying mucosa. Immunohistochemical staining was positive for cytokeratin 7 and negative for cytokeratin 20, and the tumour was determined to be a metastatic müllerian neoplasm, consistent with the initial PFTC. Although this is the first reported case of colon metastasis of PFTC, the possibility of such an unusual site of metastasis should be kept in mind, as PFTC may recur as isolated bowel lesions even in the absence of peritoneal disease.

Key words: Fallopian tube carcinoma; Metastasis; Sigmoid colon.

Introduction

Primary fallopian tube carcinoma (PFTC) is an uncommon tumour accounting for approximately 0.14%-1.8% of female genital malignancies [1]. Although knowledge about the natural history and recurrence of PFTC is limited, distant metastasis is considered to be exceptional. Metastatic colorectal cancer on the other hand, occurs in only 1% of colorectal cancers and an isolated colonic metastasis is very rare [2]. To our knowledge this is the first reported case in the literature of PFTC with an isolated metastasis to sigmoid colon.

The patterns of spread of PFTC have long been considered similar to those of epithelial ovarian cancer (EOC), principally by the transcelomic exfoliation of cells that implant throughout the peritoneal cavity. In approximately 80% of patients with advanced disease, metastases are confined to the peritoneal cavity [1]. Tumour spread can also occur by means of contiguous invasion, transluminal migration, and haematogenous dissemination.

We present a case of PFTC in which an isolated sigmoid lesion limited to the serosa and muscularis propria was found without any evidence of mucosal involvement, three years after surgical debulking and platinum and paclitaxel-based combination chemotherapy.

Case Report

A 38-year-old Caucasian woman with a two-year history of primary infertility and no family history of cancer, was initially referred to our Department of Obstetrics and Gynaecology in March 2006 for treatment of progressively enlarging bilateral ovarian cysts, found during infertility investigation. The patient had already had seven unsuccessful intrauterine insemination cycles and three in vitro fertilization attempts, according to the short protocol of ovarian stimulation with GnRH agonist and gonadotrophins, but the pregnancy outcome was negative. Transvaginal (TVS) ultrasound as well as abdominal computed tomography (CT) scans showed bilateral adnexal masses; a right cystic mass (4.2 × 2.6 cm) and a left side complex mass (13 × 10 cm). The serum CA-125 and CA 15-3 concentrations were 600 U/ml and 32.6 U/ml, respectively (normal ranges < 35 U/ml, < 30 U/ml, respectively). The serum CA 19-9 and CEA were normal. The Papanicolaou cervicovaginal smear was negative.

Exploratory laparotomy at that time revealed bilateral adnexal masses. Frozen section evaluation of the left adnexal mass showed carcinoma. Staging laparotomy with total abdominal hysterectomy, bilateral salpingo-oophorectomy, infracolic omentectomy and retroperitoneal lymph node sampling were performed. There was no obvious tumour spread elsewhere in the abdominal cavity. Peritoneal cytology was positive. Pathologic examination revealed a grade 2, moderately differentiated right endometrioid PFTC. In the left ovary, an endometriotic cyst and metastatic implants from the controlateral PFTC were found. All 24 resected bilateral lymph nodes were free of disease. No evidence of neoplastic involvement of the uterus, the ovaries or the omentum was found. Estrogen and progesterone receptors were identified by immunochemistry (monoclonal antibody 6F11 for estrogens receptors and 1A6 for progesterone receptors, Novocastra, Newcastle, UK) in 40% and 10% of the neoplastic cells, respectively. The tumour was a FIGO Stage IIC PFTC.

Surgery was followed by six cycles of combination chemotherapy using paclitaxel (175 mg/m^2) and carboplatin (AUC 6) without significant complications. The patient has been followed-up with clinical examination and measurement of serum CA-125 levels, which have remained less than 15 U/ml for three years. A CT scan of the upper abdomen and pelvis, which was conducted biannually, showed no evidence of disease.

In January 2009, the patient while asymptomatic was referred

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Fig. 1

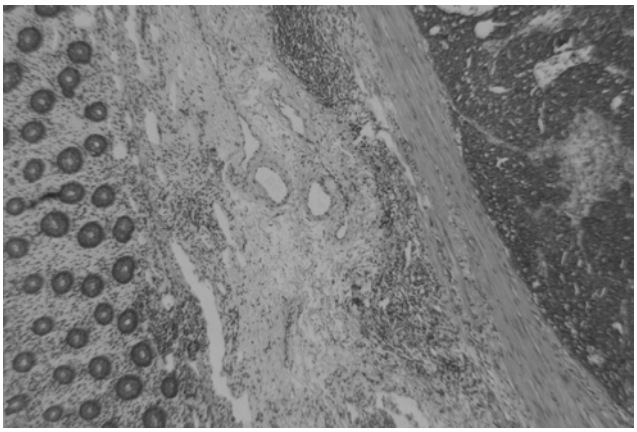

Fig. 2

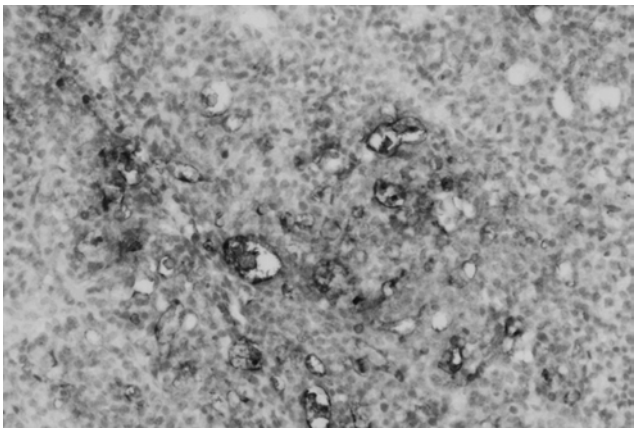

Fig. 3

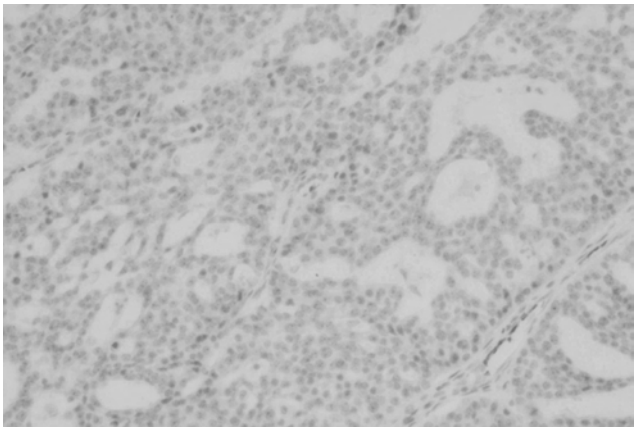

Figure 1. — Pathological examination showed a tumour in sigmoid colon wall, where the mucosa is unremarkable (H&E, original magnification x 25).

Figure 2. — Neoplastic cells are positive for cytokeratin 7 (immunochemistry, original magnification x 100).

Figure 3. — Neoplastic cells are negative for cytokeratin 20 (immunochemistry, original magnification x 100).

again to our department due to progressively elevated serum CA-125 concentrations (95 U/ml to 130 U/ml in a week interval) during a routine follow-up. Serum CA 15-3, CA 19-9 and CEA concentrations were normal. She reported no history of abdominal discomfort, constipation or diarrhoea. Abdominal CT scan revealed a complex pelvic mass (5 × 4 × 3 cm) near the top of the vaginal vault, and no other evidence of recurrent disease. On exploratory laparotomy all peritoneal organs and epithelial surfaces were free of gross disease. A 5-cm tumour growing in the sigmoid colon serosa was found and a sigmoidectomy with primary end-to-end anastomosis was performed. Peritoneal cytology was negative.

Pathologic assessment of the 14-cm-long bowel segment revealed a 5 × 3.5 cm yellow-tan solid tumour on the serosal surface. On opening the bowel, the mucosa was grossly normal, stretched over the tumour, but otherwise unremarkable. Microscopically the tumour involved the bowel wall from the serosa to the submucosa, without involvement of the underlying mucosa (Figure 1). The surgical resection margins and the donut specimen from the anastomosis were free of tumour. No endometriosis was present. The sigmoid colon lesion was determined to be a metastatic müllerian neoplasm consistent with the initial PFTC. Immunohistochemical analysis confirmed the H & E findings; the colon tumour was found to be positive for cytokeratin 7 (Figure 2) and CA-125 and negative for cytokeratin 20 (Figure 3), CEA and CA 19-9. These results confirmed that the colon tumour had an epithelial and müllerian origin.

The patient was treated for recurrent fallopian tube carcinoma with six cycles of carboplatin (AUC 5) and paclitaxel (175 mg/m2). The serum CA-125 level after she completed treatment was 8.5 IU/ml. A CT scan at that time showed no evidence of abdominal or pelvis masses. At the time of this report, one year after the patient completed her chemotherapy, she remains asymptomatic. She is followed-up every three months with clinical examination and measurement of serum CA-125 levels, which up to now have been within the normal range.

Discussion

PFTC is a rare tumour that resembles EOC histologically, clinically, as well as in surgical staging, management, indications for adjuvant chemotherapy and recurrence pattern. The most common encountered spread pattern is intraperitoneal dissemination through the transcoelomic route to neighbouring organs and peritoneal surfaces. Despite this fact, most metastases are considered to be extrapelvic, and half or more of them are extraperitoneal, usually in combination with intraperitoneal recurrence [1]. Lymphatic and haematogenous dissemination is exceptional, found late in the course of the disease and presents as an inguinal lymph node [3], bone [4], or brain metastases [5]. To the best of our extensive literature search this is the first reported case of isolated sigmoid colon metastasis from a PFTC.

Given that all knowledge concerning colon metastasis comes from reports regarding EOC, it has been documented that the serosa is initially affected and then invasion extends from serosal and subserosal tissues into the muscularis propria and mucosa of the bowel wall. Reed

et al. in their review of 77 autopsy records of patients with ovarian cancer, found metastasis to bowel serosa in only 86% and involvement to bowel mucosa in 36% of cases [6]. However, when mucosal involvement occurs, it may reflect either invasion from the serosal surfaces or haematogenous dissemination with infiltration of the submucosal capillary network. An atypical sigmoid metastasis from a high-grade mixed adenocarcinoma of the ovary has been reported, where the lesion was limited to the mucosa and muscularis propria without any evidence of serosal involvement [7]. More recently, Shibahara *et al*. reported a caecum metastasis in the muscularis propria and subserosa with only focal invasion of the colon mucosa and invasion of the retroperitoneum, which developed more than 20 years after the treatment of the primary bilateral ovarian cancers [8]. In the present case, the bulky tumour infiltrated the serosa extending to the colonic muscularis propria without involvement of the colonic mucosa. The pattern of metastasis was thought to have spread by the peritoneal route and one could expect the presence of advanced intraperitoneal tumour burden.

It is often difficult to distinguish between PFTC (especially the endometrioid and mucinous types) with sigmoid infiltration and primary gastrointestinal tract tumours. With regard to immunohistochemical staining which may be useful in the differential diagnosis, all the information comes from EOC. PFTC similar to ovarian carcinomas, other than those of mucinous type, are almost invariably positive for cytokeratin 7, while colonic carcinomas show noticeable positivity for cytokeratin 20 [9]. Loy *et al*. reported a cytokeratin 7 positive/cytokeratin 20 negative immunophenotype to be nearly 100% specific for an ovarian origin, and conversely a cytokeratin 7 negative/cytokeratin 20 positive immunophenotype was seen in 94% of the tumours of colonic origin [10]. Other tumour markers such as CEA and CA-125 can also be used; CA-125, estrogen receptors, and progesterone receptors are generally positive in ovarian and fallopian tube cancers, and CEA is generally positive in colorectal cancers [11]. In our case, the colonic tumour was positive for cytokeratin-7, CA-125, and estrogen receptors, and negative for cytokeratin-20, CEA, and progesterone receptors. This pattern was consistent with a müllerian rather than a colonic origin. In addition, a comparison of the patient's PFTC with the colonic tumour revealed that the two tumours had similar histologic features.

In conclusion, we present the first case of PFTC with isolated sigmoid colon recurrence, diagnosed on the basis of increased serum CA-125 levels. The possibility of such an unusual site of metastasis should be kept in mind, even if the patient is asymptomatic and in a disease-free state. A high index of suspicion is needed, so that a colonoscopic evaluation – which was not performed in our case – could be considered as part of workup studies. Given that lack of peritoneal surface involvement does not exclude recurrence of PFTC, the treatment of choice for the solitary colon metastasis may be surgical, followed by adjuvant chemotherapy, while intensive surveillance is essential for an early diagnosis and appropriate treatment with the perspective of the best prognosis.

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