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Case Report

Small bowel intussusception in metastatic endometrial carcinoma

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Endometrial carcinoma is the second commonest gynaecological malignancy. Survival is generally better than with other genitourinary malignancies on account of its overt presentation with post-menopausal bleeding and its tendency for local progression as opposed to early systemic dissemination. Consequently, surgery is the gold standard therapeutic strategy, and to date there is little evidence to support the routine use of adjuvant chemotherapy.[1,2]

Adult intussusception is very uncommon and unlike its paediatric counterpart is usually associated with a significant underlying organic focus.[3,4] When due to secondary malignant disease, the primary neoplasm is characteristically aggressive with a propensity for haematogenous spread. Similarly, splenic metastases are rare and characteristically occur in tumours which have a propensity for blood-borne dissemination.[5-7]

We report a unique case of recurrent endometrial carcinoma presenting with small intestinal intussusception and the incidental finding of a solitary splenic metastasis. This pattern of behaviour is very atypical in endometrial carcinoma. We discuss adult intussusception and present the argument for selective systemic chemotherapy in endometrial carcinoma.

CASE REPORT A 64-year-old multiparous woman presented with a three-month history of episodic post-menopausal bleeding. Cervical dilatation and endometrial curettage was performed and histology confirmed the presence of a poorly differentiated endometrial carcinoma. At operation the disease appeared to be confined to the uterus and cervix. Hysterectomy and bilateral salpingoophorectomy was performed with excision of the upper third of the vagina; lymph nodes were sampled from the internal iliac, external iliac and obturator group. Histology confirmed the presence of a poorly differentiated

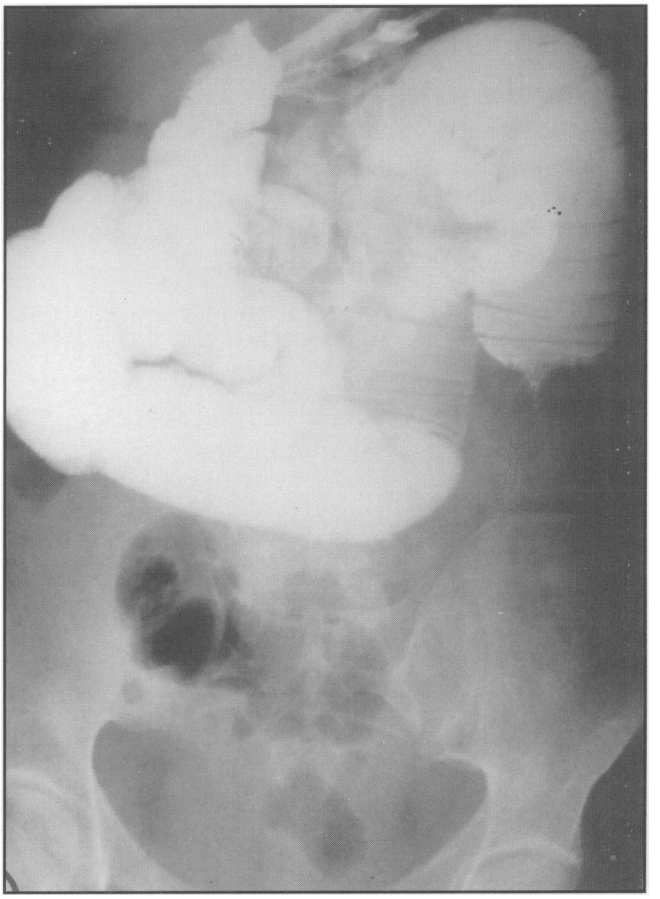

Fig 1. This radiograph shows an obstruction in the mid ileum and the blunted appearance at the site of the obstruction is typical of an intussusception.

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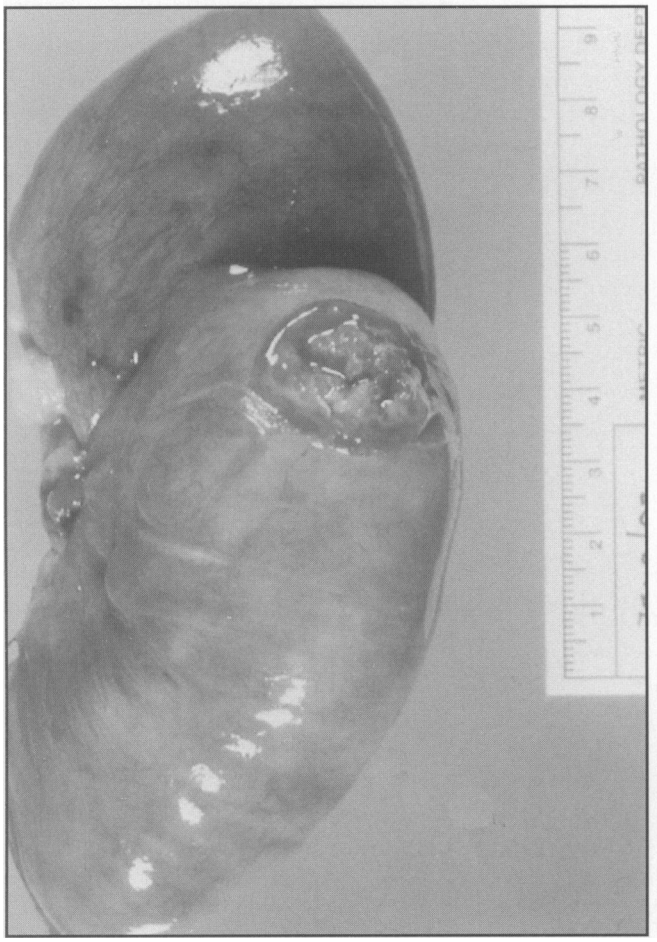
endometrial carcinoma. There were areas of microvascular invasion with lymph node metastases to the internal iliac lymph nodes.

Post-operatively the patient received 50 Gy of external beam radiotherapy and oral progestogen; the receptor status of the tumour was unknown. She remained well and a CT scan performed six months postoperatively showed no evidence of macroscopic disease recurrence.

Ten months later she presented with intermittent crampy abdominal pain and melaena. Clinically she was anaemic and displayed the physical and radiological features of small intestinal obstruction. A small bowel series confirmed the presence of a mid-ileal obstruction and the "elephant foot" appearance, suggestive of an intussusception [Fig 1]. Abdominal ultrasonography showed no evidence of ascites or hepatic infiltration; however a 3 cm solitary metastasis was noted in the upper pole of the spleen. At laparotomy the small intestine was found to be distended proximal to the mid-ileal region where there was an irreducible intussusception [Fig 2 a+b]. Consequently a limited small-bowel resection with primary anastomosis was performed, and histopathological assessment revealed a solitary focus of recurrent endometrial carcinoma acting as a " lead point".



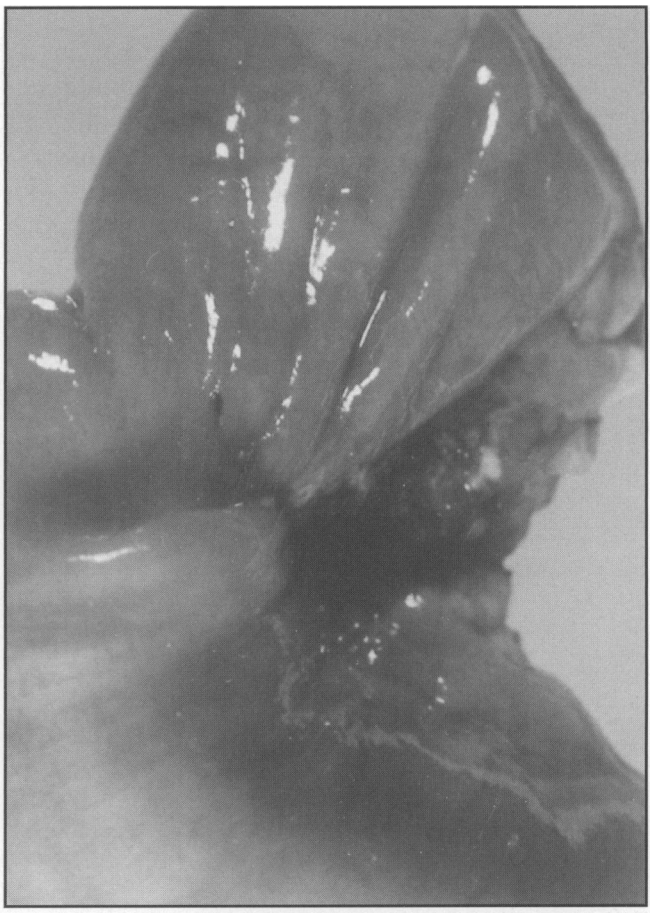


Fig 2a & b. These operative specimens show the ileal intussusception both before and after reduction. The ulcerated area in picture 2a clearly shows the endometrial metastases acting as a lead point for intussusception.

The patient made an uncomplicated recovery and was discharged five days later. She remained well for several months but died from disseminated disease within nine months.

DISCUSSION

Endometrial carcinoma occurs most commonly in postmenopausal women in the 6th decade of life, and the overall five year survival is approximately 65%.[1] The majority of tumours are confined to the uterus and present at an early stage with postmenopausal bleeding. Most of these patients have an excellent prognosis when treated by total abdominal hysterectomy and bilateral salpingoophorectomy alone, providing extraperitoneal disease is absent, the carcinoma is well differentiated and the depth of myometrial

invasion is less than one third.[2] Radiotherapy is used as adjuvant treatment for more advanced endometrial carcinoma, with a reduction in local recurrence rates. The long-term survival remains unchanged.[8] As in this case, hormonal manipulation with progestogens can be used as a first line adjuvant therapy. The basis for this is the presence of tissue-receptor sites in as high as 70% of tumours. Their routine use is of doubtful value. A large randomised placebo controlled trial of 1084 patients showed no survival advantage or progression-free period in patients receiving adjuvant progestogen therapy.[9] In carefully selected patients however, they may be of some value. Neijt [10] reviewed current literature on the systemic treatment of endometrial carcinoma and suggested that first-line treatment for those patients who are receptor positive should be hormonal therapy, providing their life expectancy was greater than four months. If the receptor status is unknown, but the tumour well differentiated, then again first-line treatment should be hormonal therapy. The response rate however is only about 10-15% and is usually short lived.[11] With gynaecological malignancy small bowel obstruction occurs commonly. This is due to direct spread of tumour to small bowel lying within the pelvis. Endometrial carcinoma has not previously been described in association with adult intussusception or with solitary splenic metastases.

Adult intussusception is difficult to diagnose preoperatively although the triad of melaena, intestinal obstruction and the presence of an abdominal mass has been described.[12] Barium studies, ultrasonography and computed tomography have all been employed with limited success.[1, 13, 14]

Adjuvant chemotherapy aims to ablate microscopic bloodborne tumour deposits and has been successfully employed in breast carcinoma, and more recently in colorectal carcinoma.[15, 16] The use of chemotherapeutic agents is associated with well-recognised morbidity and their global administration in the treatment of cancer may be inappropriate in many patients. It would be ideal if we could identify those patients who would benefit most from adjuvant chemotherapy. To this end some investigators have used specific clinical and histological criteria to select patients who may benefit from adjuvant chemotherapy. Lymph node metastases, initial tumour burden, and microscopic parameters of the primary specimen, such as the presence of lymphovascular invasion, a high mitotic index and a poor host immune response may all have positive predictive potential for those individuals who may benefit.[9, 10, 15, 19]

To date, cytotoxic therapy has not been of benefit in the management of endometrial carcinoma with extrauterine spread. Burke et al[18] treated 62 high-risk patients who had stage II and stage III disease . They were given a combination regime of cisplatin, doxorubicin and cyclophosphamide. As expected those with extrauterine disease had a poorer outcome; their results were found to be disappointing and at three years one third of patients had recurrent disease, with a high proportion of cases being extrapelvic metastases. Evidence however is now accruing that a role may exist for adjuvant cytotoxic therapy in the treatment of uterine carcinoma which is either confined to the uterus or resectable but has certain histological criteria.[19] Smith et al in their retrospective review, showed a survival advantage when they looked at 39 high risk patients given combination chemotherapy and radiotherapy. The two-year progression free interval was 72% in those patients with non papillary serous carcinoma, and 22.5% in those with papillary serous tumours. Their data however was limited by small numbers and short follow up, but it did further emphasize the need for prospective randomised trials to evaluate this form of treatment. Our case is of particular interest because of its unusual presentation. It emphasizes that local therapies for such disease are unlikely to be effective without adjuvant systemic therapy to treat and or prevent haematogenous dissemination. Unfortunately the correct timing of this therapy and the patient population likely to benefit are not yet known.

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