

Bowel metastases from primary leiomyosarcoma of the gluteal region

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ABSTRACT

Metastases to the stomach and small bowel from primary tumours in extraabdominal sites are uncommon. The usual primary tumours are breast, lung and malignant melanoma. Bowel metastasis from sarcoma is very rare. We present an unusual case of primary leiomyosarcoma of the gluteal region in a 62-year-old man, who developed stomach and small bowel metastases seven and half years after surgical excision of a primary tumour.

Keywords: bowel metastases, gluteal leiomyosarcoma, intussusception, leiomyosarcoma, small intestine metastases

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INTRODUCTION

Metastases to the stomach and small bowel from primary tumours in extraabdominal sites are uncommon. The common primary tumours that metastasise to the bowel are malignant melanoma, carcinoma of the breast and lung. We present an unusual case of leiomyosarcoma of the right gluteal region, presenting with melaena due to stomach and small bowel metastases, seven and half years after surgery and radiotherapy.

CASE REPORT

A 62-year-old man presented with swelling in the right gluteal region seven and a half years earlier. He was investigated and found to have leiomyosarcoma arising from the soft tissues of the right gluteal region. The tumour was confined to the gluteal region and he underwent surgical excision followed by radiotherapy. The patient was on regular follow-up since then, and remained asymptomatic for seven years. A routine chest radiograph revealed a solitary right lung mass which was confirmed on computed tomography (CT) of chest. A right upper lobectomy was performed and histopathology showed metastasis from leiomyosarcoma.

The patient presented four months later with melaena, followed by abdominal pain, nausea and occasional vomiting. Laboratory investigations showed a low haemoglobin level of 7 g/dL. Other biochemical and liver function tests were within normal limits. Upper gastrointestinal endoscopy revealed an ulcerative lesion in the fundus of stomach. Biopsy was taken from this lesion.

Ultrasonography (US) of abdomen revealed few

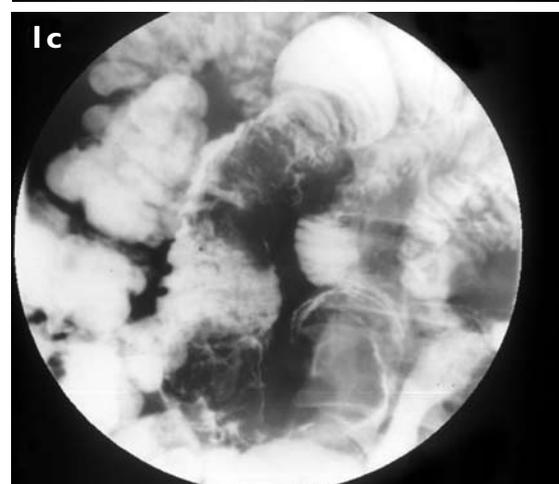
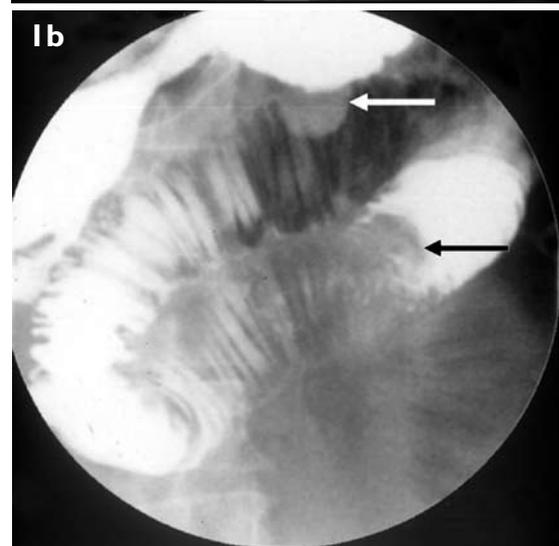
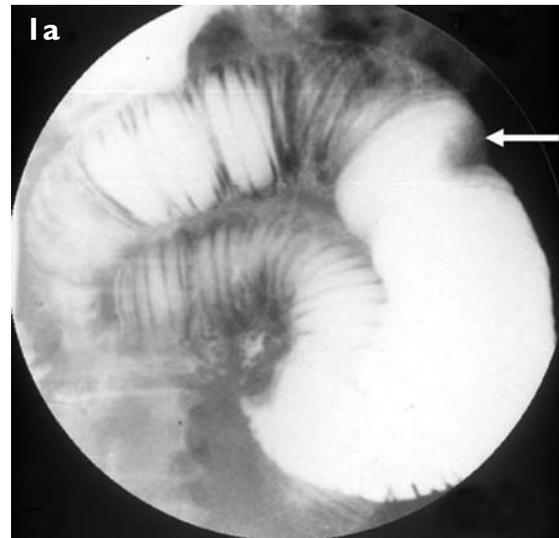


Fig.1 Barium meal and follow-through examination shows (a,b) multiple mural filling defects (arrows) in the jejunum suggestive of submucosal metastases, as well as (c) a jejunojejunal intussusception.

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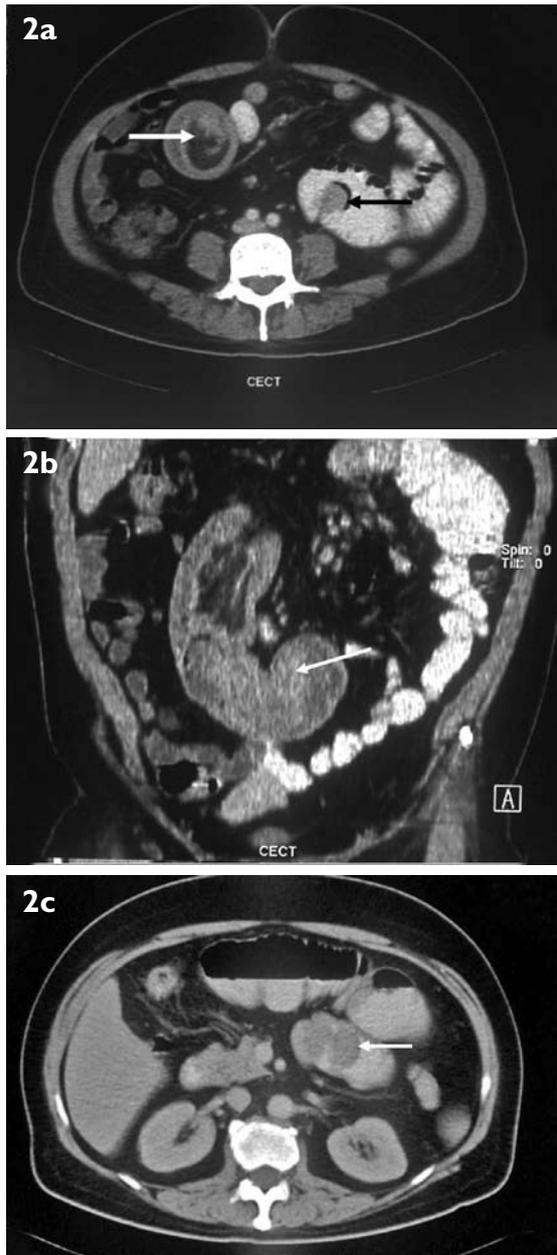


Fig. 2 (a) Axial and (b) reformatted coronal CT images show a jejunojejunal intussusception with a large polypoidal mass acting as the lead point (white arrows). Another polyp was seen in proximal jejunal loop (black arrow). (c) Axial CT image also shows other jejunal metastases (white arrow).

dilated jejunal loops. A target lesion was seen in the mid-abdomen. Barium meal and follow-through examination showed multiple filling defects in the proximal jejunum without mucosal destruction (Figs. 1a & b) and jejuno-jejunal intussusception with a coiled spring appearance (Fig. 1c). CT confirmed the presence of jejunojejunal intussusception and a large polypoidal mass was acting as the lead point, causing small bowel obstruction (Figs. 2a & b). Two separate polypoidal lesions were seen in other jejunal loops (Fig. 2c). In addition, both adrenal glands and mediastinal lymph node were enlarged, suggesting disseminated disease. No recurrent mass was seen at the right gluteal region (Fig. 3). Biopsy slides from the right

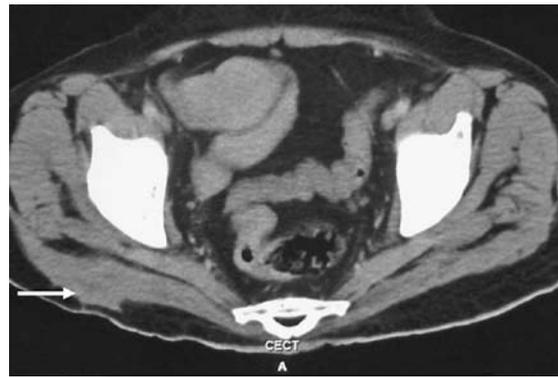


Fig. 3 Repeat axial CT image shows postoperative changes in the right gluteal region (arrow). No recurrent mass was seen.

gluteal mass, right lung and stomach lesions were reviewed. All three tumours were found to be leiomyosarcoma. The histopathological and imaging findings are suggestive of stomach and small bowel metastases.

Surgery for the stomach and small bowel lesions was not considered due to disseminated disease. The stomach lesion was treated with radiotherapy followed by chemotherapy. As the patient had severe anaemia, eight units of blood was transfused. He responded well to the treatment and was discharged after 20 days of hospital stay and he is, to date, followed-up regularly.

DISCUSSION

Leiomyosarcoma of somatic soft tissues is an uncommon neoplasm. It is more often found in the female genital and gastrointestinal tracts than in the somatic soft tissues.⁽¹⁾ Leiomyosarcoma may involve the bowel secondarily. Secondary involvement occurs by contiguous infiltration from the primary tumour in female genital and gastrointestinal tracts, or by distant metastases via lymphatic or haematogeneous route from the primary site in somatic soft tissues.⁽²⁾ The present patient had primary leiomyosarcoma arising from the soft tissues of the gluteal region with subsequent involvement of the lung and bowel by haematogeneous spread.

Bowel metastasis from soft tissue sarcoma is very rare, and there are only few reports in the literature.⁽²⁻⁴⁾ Although bowel involvement is a part of haematogeneous dissemination, it may give rise to serious complications, like gastrointestinal bleed or small bowel obstruction, which require active intervention. Identification of bowel involvement in these patients is therefore important. Behranwala et al reported four patients with small bowel metastases and one patient with stomach metastasis from soft tissue sarcomas.⁽³⁾ Myxoid liposarcoma was the most common soft tissue sarcoma to cause intra-abdominal metastases in their series, and subacute intestinal obstruction was the most common presenting feature. Sondenaa et al reported a case of small bowel metastasis from subcutaneous lower limb leiomyosarcoma, which presented with features of intestinal obstruction 22 months after the resection of primary tumour.⁽⁴⁾ At

surgery, multiple tumours along with intussusception due to a 3-cm mass was noted in the small bowel. Metastatic leiomyosarcoma was confirmed on histopathology.

Okada and Walts et al reported dual intussusception in a patient with leiomyosarcoma arising from the presternal location nine months after initial diagnosis. Their patient also presented with intestinal obstruction, and CT revealed ileal intussusception. At laparotomy, the excised ileum contained two intussusceptions and each had a 2.5-cm pedunculated, polypoid, intraluminal lesion at its leading edge. Between these lead points, there was a similar polypoid lesion and three submucosal nodules.⁽²⁾ In our patient, gastrointestinal bleeding was the presenting feature. Upper gastrointestinal endoscopy showed an ulcerative lesion in the fundus of stomach, which was the cause of the bleed. Biopsy from this lesion revealed leiomyosarcoma. The gastrointestinal bleed was severe enough to cause a fall in the haemoglobin level, requiring blood transfusion. The patient also developed features of subacute intestinal obstruction, due to intussusception caused by jejunal metastasis.

Intussusception is an infrequent cause of intestinal obstruction in adults. The majority of small bowel intussusception in adults are caused by benign tumours. Among malignant causes of small bowel intussusception, metastases are the most frequent cause.⁽²⁾ Surgery is the treatment of choice for these patients who present with features of acute intestinal obstruction.⁽³⁾ In the present patient, surgery was not considered, as he had features of subacute intestinal obstruction, and he also had disseminated disease.

The usual common primary tumours causing bowel metastasis are malignant melanoma,^(5,6) breast and

lung carcinomas.⁽⁶⁾ In malignant melanoma, multiple submucosal filling defects with central ulceration are seen in the barium examination.⁽⁶⁾ These lesions are located along the antimesenteric border of the small bowel,⁽⁶⁾ and they may be specific in arterial distribution. Metastasis from carcinoma of the breast usually involves the stomach and causes linitis plastica⁽⁶⁾ due to submucosal involvement. Metastasis from carcinoma of the lung causes large mesenteric masses with infiltration of the bowel wall, fixation and angulations of the bowel.⁽⁶⁾ The present patient had the characteristic imaging features of small bowel metastases. In summary, bowel metastasis from somatic soft tissue leiomyosarcoma is very rare. It should be considered in patients who present with intestinal obstruction or with gastrointestinal bleed. Barium meal with follow-through and CT are useful to detect multiple sites of small bowel involvement.

REFERENCES

1. Gerst PH, Levy J, Swaminathan K, Kshetry V, Albu E. Metastatic leiomyosarcoma of the uterus: unusual presentation of a case with late endobronchial and small bowel metastases. *Gynecol Oncol*.1993; 49:271-5.
2. Okada DH, Walts AE. Pathologic quiz case: dual intussusceptions in the small intestine. metastatic leiomyosarcoma as the cause of dual intussusceptions. *Arch Pathol Lab Med* 2000; 124:169-70.
3. Behranwala KA, Roy P, Giblin V, et al. Intra-abdominal metastasis from soft tissue sarcoma. *J Surg Oncol* 2004; 87:116-20.
4. Sondenaa K, Søreide JA, Erichsen C, et al. Metastases in the small intestine from a subcutaneous lower limb leiomyosarcoma. *Acta Oncol* 1992; 31: 865-6.
5. Richie RE, Reynolds VH, Sawyers JL. Tumor metastases to the small bowel from extra-abdominal sites. *South Med J* 1973; 66:1383-7.
6. Meyers MA, McSweeney J. Secondary neoplasms of the bowel. *Radiology* 1972; 105:1-11.