CASE REPORT

Osteosarcoma metastasising to the duodenum and pancreas

The incidence of metastatic osteosarcoma is increasing because of improved results following multi-agent chemotherapy and resection of the primary tumour. Metastases occur most commonly in the lungs, whereas bowel metastases are rare. We describe a 25-year-old female who presented with melaena six years after successful resection of an osteosarcoma of her right femur, and one year after resection of a solitary pulmonary metastasis. Imaging revealed a lesion arising within both the duodenum and the pancreas for which a Whipple’s pancreatoduodenectomy was carried out, achieving complete resection. Histological examination confirmed the diagnosis of metastatic osteosarcoma. We believe this is only the second such case reported. At 11 months post-operatively she had no detectable disease. Although rare, osteosarcoma can metastasise to the intestine. The surgeon must be aware of this complication, and that bowel metastases are potentially resectable.

Osteosarcoma is a rare neoplasm, accounting for less than 0.2% of all malignant tumours in the United Kingdom, but up to 4% of childhood malignancies. It has high metastatic potential, most commonly spreading to the lung, followed by the pleura, bone (other than the primary site) and heart. Treatment comprises a combination of chemotherapy and surgical resection. With developments in treatment survival is increasing, and a five-year survival of up to 92% has been reported. This has resulted in the delayed appearance of metastases, and metastases at unusual sites. The incidence of metastases to the liver, brain and regional lymph nodes is increasing, but it is rare to the bowel.

We describe the case of a 25-year-old woman who, following resection of an osteosarcoma of her right femur at the age of 19 years, and excision of a solitary metastasis in her right lung at the age of 23 years, subsequently developed a metastasis in her duodenum and pancreas, which was successfully resected.

We also present a review of the literature available on bowel metastases from osteosarcoma, identifying previously reported cases and commenting on the common features shown amongst these patients.

Case report

In February 1999 a 19-year-old female presented with a history of pain and swelling of the right knee. A diagnosis of osteosarcoma of the distal femur was made with further imaging showing this to be an isolated lesion. She was treated with six cycles of neo-adjuvant chemotherapy followed by resection of the tumour and distal femoral replacement.

In July 2005 she was admitted as an emergency to her local hospital with acute anaemia and melaena. Her abdomen was soft and non-tender, with no palpable masses, but routine blood tests revealed a haemoglobin of 3.8 g/dl. She responded well to resuscitation and blood transfusions and, once stabilised, further imaging revealed two masses arising from within the duodenum and pancreas.

We were treated with an emergency pancreatoduodenectomy, at which the abnormal tissue was completely excised, and no other abnormality was noted. Macroscopically, the two masses, which measured 10 cm × 4 cm and 5 cm × 3 cm × 3 cm, were in continuity and shaped like a dumb-bell. The tumour had infiltrated both the pancreas and the wall...
Osteosarcoma rarely metastasises to the bowel and this is only the second reported case involving the duodenum and pancreas.

The usual site to which osteosarcoma metastasises is the lung, for which pulmonary metastectomy has resulted in patients with long-term survival. Over a quarter of patients with pulmonary metastases now survive for more than five years, and 15% have a ten-year survival. Due to the rarity of bowel metastases there are no similar data for the jejunum, three in the ilium, two in the duodenum and one in the stomach. This case is the third report of an osteosarcoma metastasising to the duodenum, and the second in which the pancreas was also involved.

A review of the 12 case reports together with this case has shown common features in these patients. All but two developed pulmonary metastases prior to presentation with bowel metastases or had pulmonary metastases at initial presentation. Seven patients had their pulmonary lesions treated, six of whom, including our patient, were treated by metastectomy, and one by radiotherapy. Of all 13 cases of intestinal metastases from osteosarcoma, the disease-free interval prior to presentation with bowel metastases ranged from 0 to 6 years, with a median of 15 months.

Of the 13 cases, ten presented acutely with intussusception, obstruction or haemorrhage (as in this case) requiring emergency laparotomy. Seven of these patients had widespread disease at presentation with bowel metastases and in each case, survival was limited to several months.

Three of the 13 cases presented indolently with non-specific gastro-intestinal symptoms, and complete resection of the tumour at emergency laparotomy. Of these six with isolated bowel metastases, the post-operative survival of three patients has been reported. One patient died two days after emergency laparotomy from a further gastrointestinal haemorrhage, one patient is reported to be alive and disease-free five years later, and our patient is alive and well 11 months post-operatively.

This case is an important illustration that bowel metastases from osteosarcoma do occur, despite being rare. Widespread metastases at presentation herald a universally poor prognosis, but isolated lesions can be resected with good results. Aggressive surgical management of pulmonary metastases is advocated by many, but a surgical approach to bowel metastases (other than for the acute abdominal emergency) is less well established. Clinicians responsible for patients with osteosarcoma should be aware of the potential disease-free survival following resection of an isolated bowel metastasis.

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References


