CASE REPORT

Gastric liposarcoma resected by laparoscopic total gastrectomy to achieve a wide surgical margin

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SUMMARY

Gastric liposarcoma is an extremely rare tumour that usually affects the extremities and retroperitoneum. Preoperative diagnosis is difficult, and operative procedures are not well standardised. A 61-year-old woman presented with melaena, epigastric discomfort and palpitations. Upper endoscopy revealed a submucosal tumour at the posterior gastric fundus with an actively bleeding ulcer on the top. Our preoperative diagnosis was lipoma, and we performed laparoscopic intragastric surgery. However, the histopathological diagnosis was liposarcoma. Laparoscopic total gastrectomy was performed to achieve a wide surgical margin. Several recent series have shown that a positive microscopic margin is associated with a higher rate of local recurrence than a negative margin. We have added a staged operation to obtain a wide margin in cases involving a positive surgical margin. Preoperative diagnosis of liposarcoma is still challenging. Gastric lipoma-like tumours should be resected with a wide margin because of their possibility of malignancy.

BACKGROUND

Liposarcoma is the most common soft tissue sarcoma and accounts for 15%–20% of all mesenchymal malignancies.^{1–3} Liposarcomas usually affect the extremities and retroperitoneum, and gastric liposarcomas are extremely rare. The preoperative diagnosis remains challenging, and treatment, including the most appropriate surgical approach, is not well standardised. The mainstay curative treatment is surgical resection, and some patients may be candidates for multimodal treatment with chemotherapy and radiation.⁴ We report a case involving a patient who was diagnosed with a gastric submucosal tumour and underwent laparoscopic intragastric surgery with additional resection due to a

Figure 2 Urgent upper gastrointestinal endoscopy showed a submucosal tumour with an ulcer on top.

histological diagnosis of an atypical lipomatous tumour with a positive surgical margin. A carefully planned surgical strategy is required to obtain a wide margin for gastric lipomatous masses because an accurate preoperative diagnosis, including gastric liposarcoma, is difficult.

CASE PRESENTATION

A 61-year-old woman presented with melaena, epigastric discomfort and palpitations. She had a history of appendicitis and a duodenal ulcer. A gastric submucosal tumour with a smooth regular appearance in the gastric fundus was detected 18 months previously and followed up by a physician (figure 1). An endoscopic biopsy was performed 3 months previously because the tumour had grown during follow-up. No evidence of malignancy was detected. Endoscopic ultrasound with biopsy was not performed during follow-up. The patient's serum haemoglobin concentration was 5.0 g/dL. Urgent upper endoscopy revealed a submucosal tumour at the posterior gastric fundus with an actively bleeding ulcer on top (figure 2).

An enhanced abdominal CT scan revealed a 5 cm tumour arising from the gastric fundus and protruding beyond the gastric wall. The tumour appeared as a lipomatous mass with a hypoechoic area of fatty density, no intratumorous vascularity and no involvement of the major vessels (figure 3). These findings suggested the possibility of a lipoma, gastrointestinal stromal tumour or gastric schwannoma.

TREATMENT

Laparoscopic intragastric surgery was performed. The tumour was lifted and resected with a laparoscopic stapling device (figure 4A). The specimen was soft and yellow (figure 4B). Histopathological examination revealed that the tumour mainly

Figure 1 Upper gastrointestinal endoscopy showed a submucosal lesion with a smooth, regular appearance in the gastric fundus. The lesion was suspected to be a lipoma.

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Figure 3 (A) Abdominal CT showed a lipomatous mass with a signal intensity similar to that of fat. (B) Part of the tumour had a heterogeneous fat density.

consisted of adipose tissues of various sizes and pleomorphic cells containing atypical nuclei. Immunohistochemistry findings revealed that no MDM2 expression was observed in the patient, though amplification of MDM2 sometimes provide clues that help with the diagnosis of atypical lipomatous tumour. MIB-1 index was 10-20% indicating relatively high proliferative activity of the tumour cells (figure 5). The surgical margin was positive. Laparoscopic total gastrectomy was performed 3 months later as a second operation to achieve a negative surgical margin. Histopathological examination of the specimen obtained by total gastrectomy revealed no residual tumour because the staple cutline and an associated foreign body reaction obstructed the evaluation of the margin. The patient recovered uneventfully after the second surgery.

OUTCOME AND FOLLOW-UP

No tumour recurrence was detected 14 months after the second surgery.

DISCUSSION

This patient's course presents several clinical issues. The optimal treatment strategy for gastric liposarcoma has not been defined. The most appropriate procedure and whether it is necessary to achieve a negative surgical margin are unclear.

Surgical therapy is the first option for resectable liposarcomas. However, the local recurrence rate is as high as 60% at 5 years because the tumour often infiltrates into the surrounding organs.⁵ Liposarcoma is classified into four types: atypical lipomatous/

Figure 5 Histological findings. (A) The tumour mainly consisted of adipose tissue with some connective tissue. The nuclei were variously sized, and the pleomorphic cells had slightly atypical nuclei. The surgical margin was positive. (B) Representative S-100 immunohistochemistry findings are shown. (C) No MDM2 expression was observed. (D) The MIB-1 index was 10%–20%.

well differentiated, dedifferentiated, myxoid/round and pleomorphic.⁶ The diagnosis of an atypical lipomatous/well-differentiated tumour is difficult because the imaging findings reveal more lipoma-like tissues. Although atypical lipomatous/well-differentiated tumours are associated with a lower risk of local and distant recurrence than are the other types, previous reports have demonstrated that the 5-year local recurrence-free survival rates were significantly lower for negative than positive microscopic margins.^{7,8} In the present case, we performed laparoscopic total gastrectomy as the second operation to achieve a negative surgical margin because the margin was positive after the first operation. Additional wedge resection was difficult because the staple cutline of the first operation was too wide to identify the positive resection stump. The second operation was delayed for 3 months at the patient's request. We considered that a 3-month interval between the first and second operations was reasonable because atypical lipomatous tumours do not have an unfavourable tumour biology. Additionally, for soft tissue tumours, a diameter of >5 cm is a more unfavourable prognostic factor than a diameter of <5 cm because of the higher risk of positive margins and local recurrence.⁸ In the present case, the patient was considered to be at risk of a positive surgical margin. At the time of the first operation, we did not consider the possibility of malignancy because CT revealed a hypoechoic homogeneous lesion and the frequency of gastric liposarcoma is extremely low. An intraoperative rapid-frozen section biopsy might be necessary to obtain an accurate diagnosis and evaluate the margin.

The first operation in the present case was laparoscopic intragastric surgery. Laparoscopic intragastric surgery was first reported in 1994.⁹ This procedure is suitable for resecting submucosal tumours and early gastric cancers located near the oesophagogastric junction or gastric fundus that are difficult to remove by partial gastric resection. Laparoscopic intragastric surgery allows for full-thickness partial gastric resection, is minimally invasive and is cosmetically beneficial due to the use of only a single incision. For patients with submucosal tumours located in the upper gastric portion near the oesophagogastric junction with an endophytic growth pattern, major surgery would be excessive if the postoperative pathological

Figure 4 (A) The tumour was lifted with forceps and resected by a laparoscopic stapling device. (B) The specimen was soft and yellow.

examination shows that the tumour is benign. Another procedure for partial resection of tumours located in the upper portion of the stomach is laparoscopic and endoscopic cooperative surgery (LECS). Hiki *et al*¹⁰ reported that LECS involves seromuscular resection by laparoscopy with endoscopic dissection and offers the possibility of resection independent of the tumour location, such as in the vicinity of the oesophagogastric junction. LECS also allows the surgeon to achieve a wide margin because the surgical margin can be determined by intraoperative endoscopic marking. The positive margin after the intragastric surgery in the present case was a result of the characteristics of the tumour, which was not encapsulated. We often perform intragastric surgery for gastrointestinal stromal tumours, which are encapsulated. During intragastric surgery, the tumour is lifted with forceps and cut with a laparoscopic stapling device. Encapsulated tumours are easily lifted with a safe margin. Lipoma-like tumours, however, which are not encapsulated, can be sandwiched by the stapling device because the tumours are not lifted well; this is especially true for large tumours. The present case illustrates that non-encapsulated

large tumours should be treated by LECS to obtain a wide margin.

There are several approaches for resection of gastric submucosal tumours. Because the preoperative diagnosis of liposarcoma remains challenging, gastric submucosal tumours such as lipoma-like tumours should be resected with a wide margin.

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Learning points

- Gastric liposarcoma is extremely rare but should be considered as a differential diagnosis.
- Preoperative diagnosis of liposarcoma, especially atypical lipomatous tumours/well-differentiated liposarcomas, is still challenging.
- Non-encapsulated large submucosal tumours should be resected by an appropriate procedure such as LECS to obtain a certain margin.
- Surgical treatment of liposarcomas should involve an aggressive approach to achieve complete surgical resection with a wide margin.

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