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ABSTRACT

Herein, we describe a case of a 41-year-old woman with two giant leiomyomas located in the lower thoracic oesophagus and gastric cardia that were treated by minimally invasive thoracoscopic and laparoscopic surgery. We first resected the gastric cardia and laparoscopically prepared a gastric tube, and then we resected the lower thoracic oesophagus and intrathoracically anastomosed the oesophagus and gastric tube using thoracoscopic surgery with the patient in the prone position. Two concurrent giant leiomyomas of the oesophagus and stomach are rare, and the choice of surgical procedure to address the tumour from the mediastinum into the abdominal cavity was particularly challenging. We selected a minimally invasive thoracoscopic approach with the patient in the prone position. This strategy seems effective for resecting these giant tumours in the lower thoracic oesophagus and gastric cardia.

Key words: Oesophagus, Laparoscopy, Prone position, Thoracoscopic surgery

INTRODUCTION

Leiomyoma is the most frequently occurring nonepithelial tumour in the oesophagus⁴⁾. However, most are generally solitary and small^{1,4,8)}, and the incidence of more than one giant leiomyoma in the gastrointestinal tract is very low^{12,16)}. Herein, we describe a patient with multiple giant leiomyomas of the oesophagus and stomach that were treated using laparoscopic and thoracoscopic surgery.

CASE REPORT

A 41-year-old woman diagnosed with giant leiomyoma around the oesophagogastric junction was referred to our hospital for treatment. The patient and her family reported no relevant health history, though she did have dysphagia.

An upper gastrointestinal series showed a smooth crescent-shaped filling defect without mucosal abnormalities in the narrowed lumen from the lower thoracic oesophagus to the gastric cardia, suggesting a giant tumour with a smooth surface (Figure 1A and B). Esophagogastroduodenoscopy (EGD) revealed two giant tumours covered with normal mucosa in the lower thoracic oesophagus and gastric cardia (Figure 2A and B). Contrast-enhanced computed tomography revealed homogeneously enhanced, giant tumours lying across the oesophagogastric junction from the lower thoracic oesophagus and gastric cardia. The greatest diameter of the tumours was > 10 cm (Figure 3). According to biopsy specimens of tumour mucosa obtained after EGD, the pathological diagnosis was leiomyoma.

Surgical resection was planned because although the tumour was benign, it was causing dysphagia. Therefore, we resected the lower oesophagus and gastric cardia using minimally invasive laparoscopic and thoracoscopic surgery. The patient was placed in the supine position with a broad base. A 12-mm infra-umbilical port for laparoscopy was introduced using open insertion, and the pneumoperitoneum was established. Two 5-mm trocars were then placed immediately subcostal in the right and left mid-clavicular line, and two 12-mm trocars were positioned about 7 cm below the right and left costal margin. A liver retractor was introduced via a 5-mm stab wound positioned under the xiphoid (Figure 4).

We laparoscopically resected the greater omentum with preservation of the right gastroepiploic artery and vein, the left gastroepiploic artery and vein below the spleen, and the short gastric arteries and veins. We then resected the lesser omentum and dissected around the tumour in the gastric cardia. The stomach under the tumour was resected using a linear stapler except for part of the greater curvature of the stomach and almost all of the gastric tube. The abdominal wound was closed

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Figure 1 Upper gastrointestinal series. Smooth crescentshaped filling defect without mucosal abnormality is located in the narrowed lumen from the lower thoracic oesophagus (A) to the gastric cardia (B).

to complete the abdominal portion of the procedure.

The patient was then placed in the prone position with the right arm raised and the left arm lowered under bilateral ventilation. A 12-mm port was introduced in the posterior axillary line of the 7th right intercostal space using open insertion, and a pneumothorax of 8 mmHg was established. Two 12-mm trocars were then placed in the posterior axillary line of the 5th intercostal space and in line with the inferior tip of the right scapula of the 10th right intercostal space, for the operator and camera, respectively. Thereafter, 5-mm trocars for the assistant



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Figure 2 Oesophagogastroduodenoscopy findings. Two giant tumours are covered by normal mucosa in the lower thoracic oesophagus (A) and gastric cardia (B).

were positioned in middle axillary line of the 8th left intercostal space (Figure 5). The surgical field was difficult to visualize because the giant tumour in the lower thoracic oesophagus obscured the view. However, we resected around the tumour and transected the proximal side of the lower oesophagus using a linear stapler, and the tumours of the gastric cardia and gastric tube were drawn from the abdominal to the thoracic cavity. The residual greater curvature of stomach under the tumour was resected using the linear stapler, and an entire gastric tube was fashioned. The resected specimen was pulled outside the body via the wound at the 5th right intercostal space after expansion to 7 cm, as the tumour was relatively soft. We ultimately anastomosed the lower oesophagus and gastric tube using an overlap method for oesophagogastrostomy using the linear stapler.

The amount of blood loss was 112 mL, and the surgical duration was 645 min. The postoperative course was uneventful, oral feeding was started on postoperative day



Figure 3 Contrast-enhanced computed tomography findings. Relatively homogeneously enhanced, giant tumours lie across the oesophagogastric junction between the lower thoracic oesophagus and the gastric cardia. Greatest tumour diameter is > 10 cm.



Figure 4 Sites of the trocars for laparoscopic surgery. A 12mm infra-umbilical port for laparoscopy was inserted. Two 5mm trocars were placed at the bilateral subcostal sites, and two 12-mm trocars were positioned about 7 cm below the bilateral costal margin. A liver retractor was introduced via a 5mm stab wound positioned under the xiphoid.



Figure 5 Sites of four trocars for thoracic procedure. A 12mm port was introduced at the 7th right intercostal space (ICS) on the posterior axillary line (PA), and a pneumothorax of 8 mmHg was established. Two 12-mm trocars were then placed in the PA line of the 5th ICS and in line with the inferior tip of the right scapula of the 10th right ICS, for the operator and camera, respectively. Thereafter, 5-mm trocars for the assistant were positioned in the middle axillary (MA) line of the 8th ICS.



Figure 6 Histopathological findings. Haematoxylin and eosin staining shows proliferation of spindle-shaped tumour cell fascicles without mitosis or atypia that are consistent with leiomyoma.

10, and the patient was discharged on postoperative day 17. Haematoxylin and eosin staining revealed the proliferation of spindle-shaped tumour cell fascicles without mitosis or atypia (Figure 6). Immunohistochemically, the tumour stained positive for desmin and sm-actin and negative for CD34 and c-kit, and the pathological diagnosis of leiomyomas arising from the muscle layer of oesophagus and stomach was confirmed. Two years after the surgery, the patient continues to have adequate oral intake and is followed up as an outpatient. Follow-up endoscopy has shown no reflux esophagitis at the anastomotic site.

DISCUSSION

Leiomyomas are the most common benign type of oesophageal mesenchymal tumours. Most reported leiomyomas have been derived from the inner circular muscle layer of the middle and lower thirds of the oesophagus, particularly at the oesophagogastric junction. Most oesophageal leiomyomas are small, slow-growing, and asymptomatic, and approximately 50% of them are < 5 cm in diameter^{1,4,8)}. Oesophageal leiomyomas > 10 cm are generally designated as giant leiomyomas, and they occur at a reported rate of 17% of all leiomyomas⁸⁾. Leiomyomas usually present as solitary tumours, rarely as multiple tumors^{12,16)}, and they rarely undergo malignant transformation^{1,4,8)}. Dysphagia and epigastric discomfort are the most common symptoms of leiomyomas^{4,8)}.

A preoperative diagnosis of oesophageal leiomyoma is often unclear. Radiography of the upper gastrointestinal tract typically shows a smooth filling defect in the oesophageal lumen without mucosal abnormalities⁹). EGD reveals a protuberant mucosal lesion with normal and smooth oesophageal mucosa. Leiomyoma is detectable by endoscopic ultrasonography as a homogeneous, hypoechoic lesion with clear margins surrounded by a hyperechoic area¹⁸⁾. Computed tomography is useful for determining the relationship between tumours and other organs. While histological assessment can provide a definitive diagnosis, the obtaining of preoperative tissue biopsies remains controversial due to the possibility of mucosal damage and inflammation, which are potential risk factors for mucosal perforation during surgical enucleation^{8,11)}. Tissue biopsies can only partially rule out malignancy since concurrent leiomyoma and leiomyosarcoma have been recognized, and standard endoscopic biopsies cannot reach the muscularis propria. Therefore, only resection can rule out malignancy and provide the means for complete and accurate histological assessment.

Once a clinical diagnosis of leiomyoma is confirmed, many factors must be considered to determine the optimal treatment strategies. Not only the size and location of the tumours, but also the subjective symptoms, general status, and comorbidities of each patient should be considered. While a consensus has been reached as to the optimal surgical approach to treating symptomatic leiomyoma, the management of asymptomatic patients remains a matter of debate. Periodic follow-up with endoscopy and an upper gastrointestinal series have been recommended for asymptomatic patients with oesophageal leiomyomas < 5 cm because malignant transformation is extremely rare^{4,8)}. Although surgical trauma might be more harmful than no treatment, some have recommended that oesophageal tumours be resected since malignancy cannot be completely denied^{11,13)}.

On the other hand, oesophageal resection and reconstruction are preferred for giant oesophageal leiomyomas over conventional tumour enucleation. This is due to certain technical difficulties, the lack of ability to ensure wound healing of oesophageal muscle that has become thin and membranous due to pressure from the tumour, and the potential for giant leiomyomas to transform into leiomyosarcoma-like tumors¹⁴. Oesophageal resection may also be necessary owing to extensive, tight adhesions between a tumour and the oesophageal mucosa, a circular growth pattern, or a tumour lying across the oesophagogastric junction. Up to 10% of reported leiomyomas require oesophageal resection. Herein, we described a patient with very rare, giant, concurrent leiomyomas in the lower thoracic oesophagus and gastric cardia. We treated this patient using minimally invasive resection of the lower oesophagus and gastric cardia with thoracoscopic and laparoscopic reconstruction. Indications for the thoracoscopic approach remain controversial, as it depends on the size and location of the tumour as well as the experience of the surgeon^{5–7}). We introduced complete thoracoscopic surgery for oesophageal cancer at our institution in 2011 and the prone position in 2013. The major advantages of minimally invasive surgery included decreased postoperative atelectasis, pleural effusions, pain, and hospital stay^{10,17}).

With respect to the position of the patient during surgery, Cuschieri²⁾ first described thoracoscopic oesophagectomy in the prone position for oesophageal cancer in 1994. Palanivelu et al.¹⁰ considered selective intubation or single lung ventilation to maintain optimal exposure of the oesophagus to be unnecessary since gravity causes the lungs to fall anteriorly, and they reported a low incidence of postoperative pneumonia and decreased surgical duration¹⁵⁾. Although several reports describe thoracoscopic enucleation in the prone position for oesophageal leiomyoma^{3,10}, oesophageal resection for giant leiomyoma in this position has not previously been reported. One advantage of the prone position is good visualization of the lower mediastinum, and this position was suitable for our patient with a tumour in the lower oesophagus. Minimally invasive procedures and the prone position helped to reduce the postoperative hospital stay for this patient by avoiding respiratory complications.

CONCLUSION

The outcome of minimally invasive surgery for two concurrent giant leiomyomas was good. The minimally invasive thoracoscopic approach in the prone position appears to be practical and safe for giant tumours in the lower thoracic oesophagus and gastric cardia.

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