CASE REPORT

Laparoscopic pancreas-preserving subtotal duodenectomy for gastrointestinal stromal tumor

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Abstract
Gastrointestinal stromal tumors (GISTs) of the duodenum are rare neoplasms. The optimal surgical procedure is debated and several options ranging from limited resections to pancreaticoduodenectomy have been reported. The laparoscopic approach has been validated for gastric GISTs, but it does not yet represent a standard technique for tumors of the duodenum. We report the case of a localized duodenal GIST that was successfully treated by totally laparoscopic pancreas-preserving subtotal duodenectomy. This procedure may represent a feasible and effective treatment option for localized GISTs of the duodenum. Large series with long-term follow-up are needed.

Key words: Laparoscopic duodenectomy, GIST, pancreas-preserving

Introduction
Gastrointestinal stromal tumors (GISTs) are mesenchymal, usually spindle-shaped neoplasms of the digestive tract, showing immunopositivity for the KIT protein (CD117) (1). They are rare tumors with an estimated incidence of 1.5/100,000/year (2) and most commonly affect the stomach (40–60%) and small intestine (30–40%) (1), but only 3–5% are located in the duodenum (3). Surgical resection with clear margins is currently considered the standard treatment for localized tumors (1,2). Various surgical procedures, ranging from local resection to pancreaticoduodenectomy, have been described for duodenal GISTs, depending on the size and site of the lesion (4). The laparoscopic approach has been validated for gastric tumors (5), but it does not yet represent a standard technique for GISTs of the duodenum.

Case report
A 77-year-old woman with a body mass index of 35 Kg/m² was admitted with a history of abdominal pain for about four months, anemia and melena. Upper digestive endoscopy revealed a submucosal tumor in the proximal third duodenal portion, shortly distal from the papilla, suggestive of GIST; the overlying mucosa showed an ulceration that was the origin of the bleeding. Several endoscopic biopsies were obtained but were reported as nonspecific. Endoscopic ultrasound confirmed a submucosal tumor, 36 x 25 mm in diameter. CT scan showed no invasion of adjacent structures, lymph node involvement or distant metastases.

A pancreas-preserving subtotal duodenectomy was performed using a totally laparoscopic approach. The patient was placed supine in a reverse Trendelenburg position with legs abducted. The surgeon stood between the patient’s legs with the two assistants on each side. Pneumoperitoneum was created using the “Open Veress Assisted” technique (6). A 30” telescope was used and four trocars were placed as follows: a 10- to 12-mm telescope trocar (T1) in the midline, 1 cm above the umbilical scar; two 10- to 12-mm trocars along the left (T2) and right (T3) midclavicular lines, lateral to the rectus muscles, 3 cm
above T1; one 5-mm trocar (T4) in the midline below the xiphoid process (Figure 1). Intraoperative ultrasound was performed to localize the tumor from the papilla and to identify the proximal line of duodenal section, which was marked with a titanium clip. The duodenum was detached performing a Kocher maneuver. The duodeno-jejunal angle was dissected by division of the Treitz ligament and the proximal first jejunal limb was then divided with a 45-mm vascular cartridge linear stapler. Uncrossing of the third and fourth duodenal portions was carried out.

Then the detachment of the duodenum from the pancreatic head was carefully completed using bipolar forceps (T3) and harmonic scalpel (T2), up to the proximal line of the duodenal section. The second portion of the duodenum was divided with a 45-mm vascular cartridge linear stapler, about 1 cm below the papilla of Vater, by the titanium clip previously positioned (Figure 2). The sectioned proximal jejunum was passed beneath the mesenteric vessels and the intestinal tract was reconstructed in a side-to-side duodenojejunostomy using a 45-mm vascular cartridge linear stapler (Figure 3). The anastomosis was tested with methylene blue dye and fibrin glue was applied. The surgical specimen (Figure 4) was removed through a minimal enlargement of the T1 site using a retrieval bag.

Operative time was 150 minutes. The immediate postoperative course was uneventful. Oral intake was started on the 5th postoperative day after a gastrografin swallow control. The patient was successfully discharged on the 7th postoperative day. Histological
examination of the surgical specimen showed a low risk GIST, with clear surgical margins. As a consequence, our patient did not receive adjuvant therapy with imatinib mesylate. No late complications were observed and no recurrence was detected after a 38 month-follow-up.

Discussion

GISTs of the duodenum are extremely rare neoplasms, although they represent approximately 30% of primary duodenal tumors (7). They most frequently involve the second and third duodenal portions and only 30% show a malignant appearance (3,4).

Clinical presentation is variable according to the size of the tumor and the existence of mucosal ulceration (4). Upper intestinal bleeding is reported with increased incidence compared to other localizations; other symptoms include epigastric pain, palpable mass and intestinal obstruction (7). Small tumors, especially without mucosal ulceration, are usually diagnosed incidentally (8).

Endoscopy may detect most duodenal GISTs with the typical features of a partly intraluminal mass with the overlying mucosa ulcerated (7), as in this case. However, endoscopic biopsy may be not very useful and differential diagnosis from other gastrointestinal tumors may be difficult preoperatively (9,10). Endoscopic ultrasound can be used to visualize submucosal tumors and some findings, such as tumor size and irregular borders, suggest malignancy (3,10). CT scan is able to assess the extension of the primary lesion and to detect metastases; lymphadenopathy, if present, should raise an alternative diagnosis of lymphoma or adenocarcinoma (11).

Complete surgical resection with clear surgical margins and no tumor rupture is currently considered the ideal treatment for localized GISTs (1,2). Moreover, since GISTs are associated with negligible submucosal spread and lymph node invasion is extremely unusual even for high risk tumors, extensive margins are not required and systematic lymph node dissection is not recommended (1,9,12).

The optimal surgical procedure for duodenal GISTs is still debated due to the complex and peculiar anatomy of the pancreaticoduodenal region and several options, ranging from limited resections to major procedures, have been described (3,4,12). Biological features of GISTs make limited resections of the duodenum preferable to pancreaticoduodenectomy (PD) when technically feasible, in order to reduce operative morbidity and to allow more functional preservation and better quality of life, while providing comparable oncological results (3,12,13). Goh et al. (9) reported similar mean disease-specific survival and recurrence rates in a retrospective comparison of fourteen patients who underwent PD or limited resection for duodenal GIST; limited resection was associated with a significantly shorter operative time, but a similar morbidity rate. Another retrospective analysis of nine patients showed excellent disease-free survival following limited resection with clear margins (4). In a prospective study, Buchs et al. (12) compared five cases of segmental duodenectomy to two PDs, reporting long-term disease-free survival in the segmental duodenectomy group. They demonstrated that, after complete surgical resection, prognosis of duodenal GISTs is not influenced by the pancreatic margins, but depends on tumoral malignant potential. Also Tien et al. (13) reported that the type of operation was not correlated to operative risk and disease recurrence in their retrospective review of twenty-five patients. The only significant predictor for disease recurrence was high-risk GIST classification.

The type of surgical procedure is based on the size, site and proximity of the tumor to the duodenal papilla (12,13). Small tumors may be treated by wedge resection, if the resulting lumen is adequate and the ampulla of Vater can be preserved (4,9). Partial duodenectomy with Roux-en-Y duodenojejunostomy has been proposed for larger tumors involving the antimesenteric border of the second, third and fourth duodenal portions (14). Segmental duodenectomy with duodenojejunostomy can be performed for large inframucular tumors (3,4,15). Finally, tumors close to the papilla may require a PD (13).

In our case, since the tumor was relatively large (>3 cm) and located in the proximal third duodenal portion, shortly distal of the papilla, we performed a
pancreas-preserving segmental duodenectomy with side-to-side duodenojunostomy opposite to the papilla. We precisely planned the operative tactics by laparoscopic ultrasound, in order to estimate the exact distance of the tumor from the ampulla, to set the limits of surgical resection and to exclude any pancreatic involvement.

Similarly, Mennigen et al. (3) reported the case of a GIST located 3 cm distal of the papilla treated by segmental duodenectomy with latero-terminal duodenojunostomy, thus confirming the feasibility of limited resection even for periampullary tumors, if they can be resected with an anastomosis just below the ampulla. Also local excision with primary closure has been reported for periampullary GISTs (16).

The laparoscopic approach has been validated for gastric GISTs (3), but it does not yet represent a standard technique for tumors of the duodenum. In a series of nine patients, Chung et al. (4) reported two laparoscopic wedge resections with primary closure for low-risk and very low-risk GISTs of the first duodenal portion, respectively 2.5 cm and 1.9 cm in size, without evidence of recurrence after 17 and 22 months. Laparoscopic pancreas-sparing duodenectomy with duodenojunostal anastomosis has been previously described by Ammori (17) for a benign peptic stricture of the third and fourth duodenal portions. The author demonstrated the feasibility and the benefits of this approach in terms of quick recovery and short hospital stay. More recently, Poves et al. (18) for the first time described laparoscopic subtotal duodenectomy for primary adenocarcinoma over a villous adenoma of the third duodenal portion. They recommend this procedure for the treatment of benign and pre-malignant infra-ampullary duodenal pathologies and also as an alternative treatment option to PD in very selected cases of tumors confined to the duodenum.

Laparoscopic pancreas-preserving duodenectomy represents a feasible and effective treatment for localized duodenal GISTs, even for tumors close to the papilla. Minimally invasive resection performed with clear margins allows low morbidity and quick recovery and has comparable oncological results to PD. Certainly, expertise in both pancreatic surgery and laparoscopic technique is required. Large series with long-term follow-up are needed.

Declaration of interest: Prof. F. Corcione, Drs. F. Pirozzi, A. Sciuto, F. Galante, U. Bracale and F. Andreoli have no conflicts of interest or financial ties to disclose.

References