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ORIGINAL SCIENTIFIC REPORT WITH VIDEO

Laparoscopic Partial Sleeve Duodenectomy for the Infra-Ampullary Gastrointestinal Stromal Tumors of the Duodenum

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Abstract

Background Although organ-preserving operations are regarded as effective strategies for duodenal gastrointestinal stromal tumors (GISTs), laparoscopic partial sleeve duodenectomy (lap PSD) has not been fully evaluated. The aims of this study were to evaluate the effectiveness and technical feasibility of lap PSD.

Study design Between January 2011 and March 2016, we reviewed 13 patients who underwent laparoscopic approach among 22 patients who underwent PSD. PSD for the infra-ampullary lesions was defined as infra-ampullary duodenal resection including the first portion of the jejunum. After resection, all patients underwent reconstruction via side-to-side duodenojejunostomy.

Results The total mean operation time was 273 min (range 160–346 min), and estimated mean blood loss was 80 ml (range scanty-200 ml). One patient was converted to open laparotomy because of mesocolonic tumor involvement. The median postoperative hospital stay was 10.5 days (range 4–36 days). There were no postoperative mortalities. Postoperative complications included 2 instances of delayed gastric emptying (DGE), 1 duodenojejunostomy stricture, and 2 intestinal obstructions. No patient was treated with adjuvant therapy. One patient experienced hepatic metastasis 28 months after surgery during a mean follow-up period of 48.6 months.

Conclusion Lap PSD might be an oncologically effective strategy for duodenal GIST, and the laparoscopic approach is a technically feasible and appealing surgical modality in terms of safety and perioperative results. However, DGE and anastomosis strictures are concerns for postoperative complications, which need to be further investigated.

Introduction

Gastrointestinal stromal tumors (GISTs) are mesenchymal gastrointestinal tumors that can be found throughout the digestive tract. However, duodenal GISTs comprise only a small subset of all GISTs. The unpredictable prognosis of

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these tumors has been expressed as "uncertain malignant potential" [1]. Complete surgical resection with clear margins is the best known curative treatment for duodenal GISTs [2, 3]; however, surgical treatment for duodenal GISTs is problematic due to the ambiguity of their clinical behavior and their proximity to adjacent organs. In contrast to adenocarcinomas of the gastrointestinal system, previous studies have shown that the incidence of submucosal extension and lymph node metastasis is negligible in GISTs [4]. Furthermore, DeMatteo et al. [4] showed that microscopic surgical margin did not influence survival. These factors support a limited local resection approach rather than radical surgery for GISTs. Meanwhile, certain anatomical constraints in pancreaticoduodenal tumors may require a pancreaticoduodenectomy (PD). Despite improvements in the morbidity and mortality rates associated with PD in recent decades, the procedure may be an extreme measure for these relatively indolent tumors. A recent case series [5–7] demonstrated that limited resection of the duodenum without deterioration of prognosis was feasible. Therefore, partial sleeve duodenectomy (PSD) for duodenal GISTs offers a good surgical option when wedge resection is infeasible [8].

However, there have been few case reports published on laparoscopic partial sleeve duodenectomy (lap PSD) [9–13]. This study is the largest reported case series of lap PSD to date. Herein, we reviewed the surgical outcomes for patients undergoing lap PSD to evaluate whether lap PSD is an effective and technically feasible surgical option for some cases.

Materials and methods

Patients and methods

Medical records of patients in a prospectively collected database were retrospectively reviewed. A total of 13 patients underwent lap PSD among 22 patients who underwent PSD for a infra-ampullary duodenal GIST from January 2011 to March 2016 at Severance Hospital, Yonsei University College of Medicine, and Bundang CHA Medical Center, CHA University. The goal for all patients was complete tumor removal, and the extent of operation and type of procedure were determined based on tumor size, proximity to neighboring structures, and proximity to the ampulla of Vater. Deciding whether there was enough space for the introduction of a surgical stapler between the ampulla of Vater and the tumor was critical to preserving the pancreas in infra-ampullary duodenal GIST cases.

The use of a laparoscopic approach for PSD was determined based on the safety of the resection margin, risk of tumor rupture, each patient's performance status, and patient and surgeon preference.

PSD for the infra-ampullary lesions was defined as infra-ampullary duodenal resection including the first portion of the jejunum. Postoperative complications were graded using the Dindo–Clavien classification system [14]. Delayed gastric emptying (DGE) was diagnosed when no anatomical problems were observed with intestinal luminal passage on upper gastrointestinal series using a gastrografin; evidence of DGE was recorded according to severity and was graded using guidelines from the International Study Group of Pancreatic Surgery (ISGPS) [15]. GIST risk classification was stratified according to the Fletcher criteria [16].

This study was approved by the Institutional Review Board and Ethics Committees of Severance Hospital in the Yonsei University Health System, Seoul, Korea, and was conducted according to the principles of the Declaration of Helsinki.

Localization

If the tumor was located close to the ampulla of Vater (Fig. 1a), we localized the ampulla of Vater using various methods, including preoperative endoscopic tattooing (Fig. 1b), intraoperative endoscopic guidance, and preoperative endoscopic clipping with intraoperative ultrasonography.

Surgical techniques

The surgical procedures were performed as follows:

1. The patient was placed in the reverse Trendelenburg position with right-side elevation, similar to positioning for conventional laparoscopic cholecystectomy. The operator and assistant surgeon who handled the laparoscope stood on the left side of the patient.
2. A 12-mm trocar for the laparoscopic camera was inserted into the umbilicus, and two main working 5-mm trocars were placed in the subxyphoid area and the right abdomen around the anterior axillary line. Another 12-mm trocar was added at the left abdomen around the anterior axillary line for application of the laparoscopic surgical stapler (Fig. 2).
3. The procedure was initiated by dividing the gastrocolic ligament and kocherization, which exposed the pancreatic head and the duodenal loop. Next, the localized ampulla of Vater and the tumor could be identified (Fig. 3). At this point, we checked for the possibility of wedge resection and primary repair. If wedge resection was not feasible for reconstruction, we proceeded with the following steps:

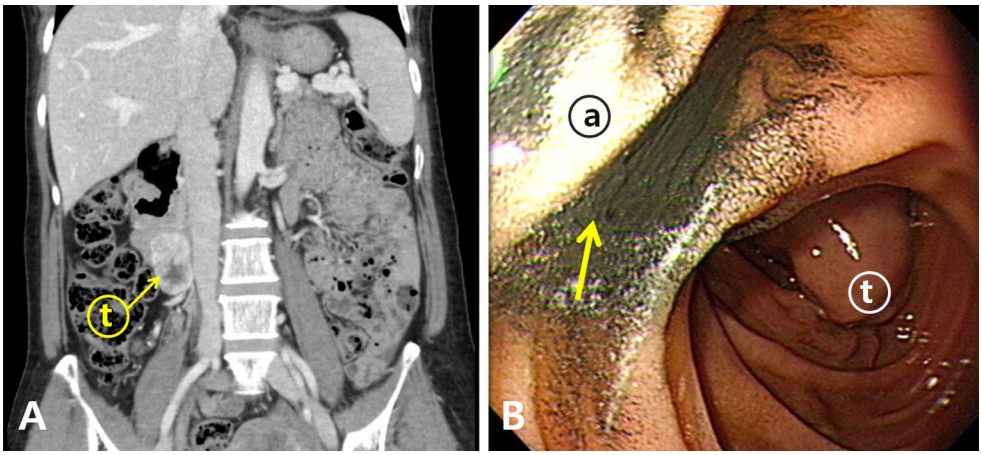

Fig. 1 Computed tomography (CT) and endoscopic findings show an oval mass in the duodenum. The tumor (ⓣ, thin yellow arrow) is located just distal to the ampulla (**a**). The ampulla of Vater (ⓐ, bold black) is localized with tattooing (thick yellow arrow), and the distance between the ampulla and the tumor (ⓣ, bold white) is confirmed on preoperative endoscopy (**b**)

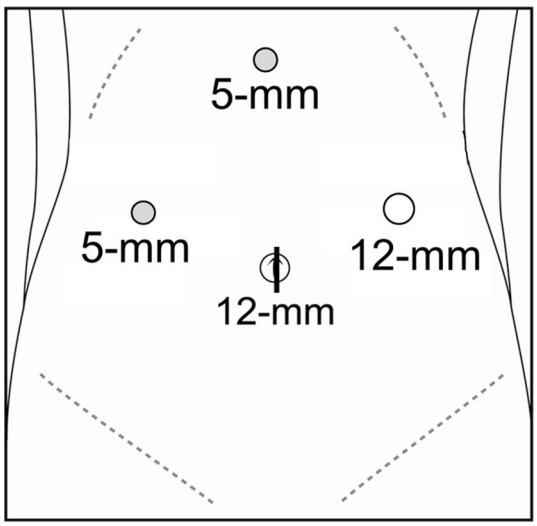

Fig. 2 Port placement: the laparoscopic camera is introduced through the umbilical 12-mm trocar, and the specimen is retrieved through the umbilicus. Two main working 5-mm trocars are placed in the subxyphoid area and the right abdomen around the anterior axillary line, respectively. Another 12-mm trocar is added at the left abdomen for application of laparoscopic surgical stapler

4. Distal duodenum from the ampulla was completely separated from the retroperitoneum and mesocolon. Then, small duodenal vessels at the inferior border of the pancreatic uncinate process were carefully resected (Fig. 4).
5. Next, the proximal jejunum was divided approximately 15 cm distal to the Treitz ligament using a laparoscopic 60-mm linear stapler (Endo GIA™; Covidien, Mansfield, MA, USA).
6. Then, the resected proximal jejunum was placed to the right side of the duodenum through the retromesenteric root.
7. Another laparoscopic 60-mm linear stapler was introduced between the localized ampulla and tumor to resect the distal duodenum (Fig. 5). The surgical specimen was completely removed.
8. Finally, the distal jejunal limb was brought alongside the remnant proximal duodenum in a retrocolic manner and a side-to-side duodenojejunostomy was made using a laparoscopic 60-mm linear stapler (Fig. 6).
9. After ascertaining the integrity of the ampulla of Vater, the anastomosis opening was closed with suture.
10. Usually, cholecystectomy is performed at the end. Intraoperative cholangiography was omitted because the location of the ampulla was checked by preoperative localization and directly visualized through the duodenal opening after anastomosis stapling. If the location of the ampulla is uncertain, an intraoperative cholangiography may be a good option.

Results

Patient demographics and operative data (Table 1)

The mean age of the 13 patients was 52.9 years (range 20–75 years), and there were 3 males and 10 females. The most common symptoms were melena in 3 patients and anemia in 3 patients. Incidental detection of the tumor occurred in 5 patients. One patient presented with

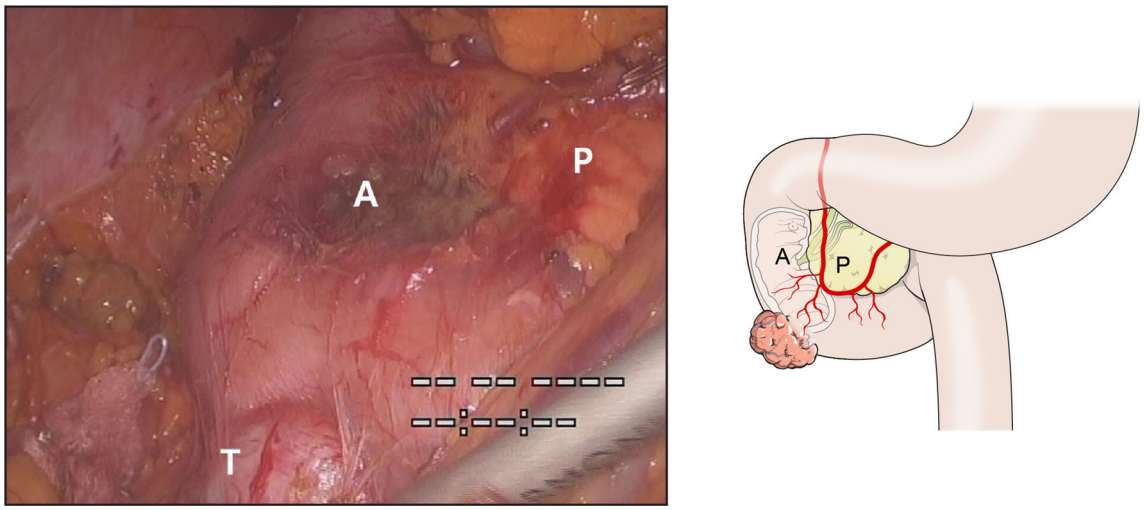

Fig. 3 The pancreatic head (P) and duodenal loop are fully exposed by the division of the gastrocolic ligament and kocherization. Next, the localized ampulla of Vater (A) and the tumor (T) are identifiable. At this point, we check for the possibility of wedge resection and primary repair. If wedge resection was not feasible for reconstruction, we proceeded to the next steps

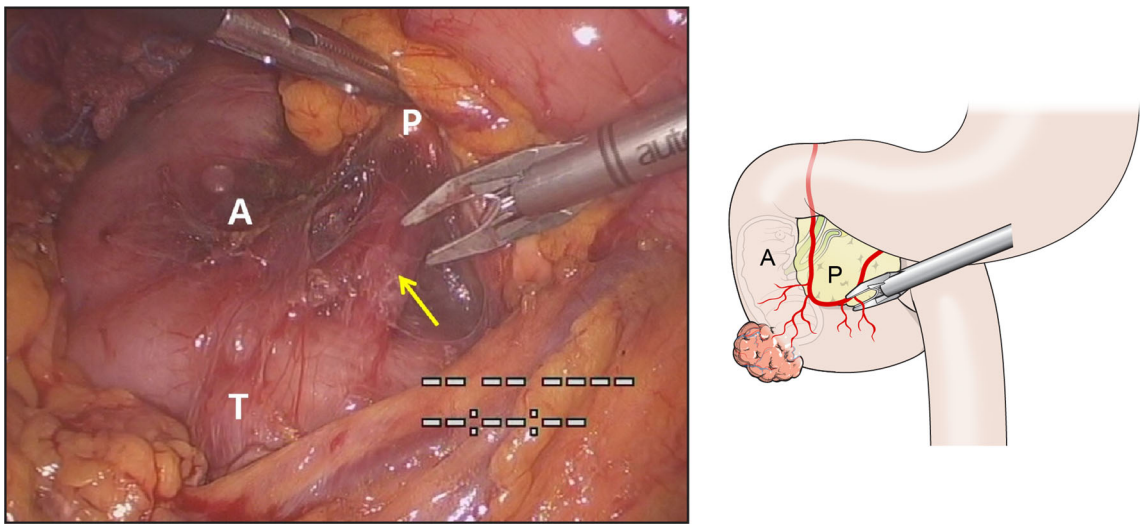

Fig. 4 The second and third portions of the duodenum is fully liberated from the retroperitoneum and mesocolon. Next, the small duodenal vessels (yellow arrow) at the inferior border of the pancreatic uncinate process (P) are carefully resected (A, localized ampulla of Vater)

abdominal discomfort and another with a palpable abdominal mass. The mean distance from the ampulla of Vater was 4.0 cm (range 1–15 cm). No patient was treated with neoadjuvant imatinib therapy. Tumors location in the second portion of the duodenum to the duodenojejunal junction was indications for this procedure. Eight tumors were located in the second portion of the duodenum, 2 in the third, and 3 in the fourth and duodenojejunal junction. Localization of the ampulla was performed in 4 patients using concomitant intraoperative endoscopy, preoperative endoscopic tattooing, and preoperative clipping with intraoperative ultrasonography because of their close proximity to the ampulla. The total mean operation time was 273 min (range 160–346 min), and the estimated mean blood loss was 80 ml (range scanty-200 ml). One patient was converted to open laparotomy because of mesocolonic tumor involvement.

Postoperative outcome and follow-up (Table 2)

The mean tumor size was 3.6 cm (range 2.2–9.0 cm), and the mitotic count was 2.8 per 50 high-power fields (HPF) (range 1–5/50 HPF). Most of the patients were classified as low risk, but three patients (nos. 8, 9, and 12) were

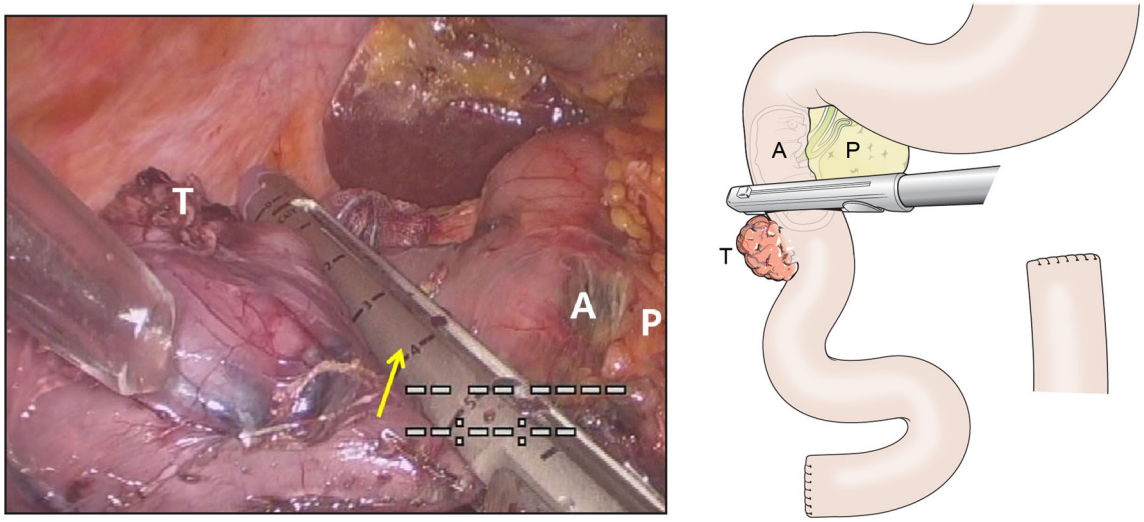

Fig. 5 Laparoscopic surgical stapler is introduced between the ampulla (A) and the tumor (T) to remove the distal duodenum (P, pancreatic head)

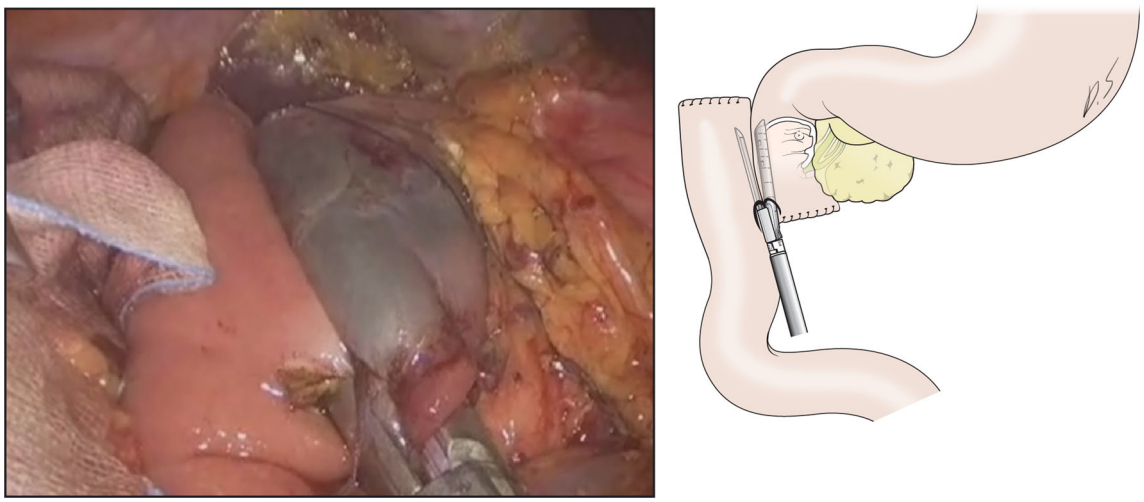

Fig. 6 Distal jejunal limb is brought alongside the remnant proximal duodenum in a retrocolic manner, and side-to-side duodenojejunostomy is performed using another laparoscopic surgical stapler

intermediate risk, according to the GIST risk stratification [17]. The mean surgical resection margin was 1.0 cm (range 0–3.5 cm), and the tumors were abutted at the resection margin in three patients. These three tumors were well-encapsulated tumors and were considered to be completely resected, although they were in contact with the resection margin. The mean postoperative hospital stay length was 10.5 days (range 4–36 days). Three patients (nos. 3, 4, and 7) were readmitted after discharge. The rate of postoperative complications greater than grade II was 38.5% (5 of 13 patients). Two patients had grade II delayed gastric emptying (DGE), which were primary DGE without other complications and which resolved after conservative management. Two patients had intestinal obstructions of the jejunal limb at the distal portion of duodenojejunostomy. Both were managed with repeat gastrojejunostomies: one laparoscopically and one with an open approach. There was one duodenojejunostomy site stricture, which we attempted to treat with endoscopic ballooning, but this approach failed. Finally, the patient underwent open gastrojejunostomy. There were no postoperative mortalities. No patients were treated with adjuvant therapy. In one patient (no. 9) with intermediate-risk GIST, the tumor invaded the mesocolon and open conversion was

Table 1 Demographic and operative data of patients

No.	Age (years)	Sex	Portion of duodenum	Distance from AOV (cm)	Localization of AOV	Operative time (min)	Blood loss (ml)	Open conversion
1	20	F	Second	2.0	0	280	200	No
2	21	F	Second	2.0	0	280	200	No
3	75	F	Second	2.0	Endoscopy	260	50	No
4	53	F	Second	2.0	Endoscopic tattooing	300	20	No
5	67	F	Second	1.0	0	260	150	No
6	75	M	Third	4.0	Clipping and US	290	50	No
7	52	F	Fourth	6.0	0	270	100	No
8	28	M	Second	1.5	0	280	50	No
9	54	F	DJ	12	0	320	50	Yes
10	58	F	Third	3.0	0	280	80	No
11	54	F	Second	1.0	0	160	0	No
12	70	F	DJ	15.0	0	226	0	No
13	61	M	Second	1.0	Clipping and US	346	100	No
Mean	52.9			4.0		273	80	

AOV ampulla of Vater, *DJ* duodenojejunal junction, *US* ultrasonography

Table 2 Postoperative outcome and follow-up

No.	Tumor size (cm)	Mitotic count (/50 HPF)	Risk	RM (cm)	Hospital stay (days)	Cx.	Re-OP	Recur	*F*/*U* (months)
1	2.3	3	Low	0	8	0		0	72
2	2.3	3	Low	0.3	8	0		0	73
3	3	4	Low	0.4	4	DGE (gr. II)		0	68
4	2.5	1	Low	0.5	7	Obst.	Lapa GJstomy	0	55
5	3.3	1	Low	0	8	0		0	53
6	3	4	Low	1.0	17	DGE (gr. II)		0	52
7	3.2	3	Low	2.5	8	Strict.	Open GJstomy	0	48
8	5.2	1	Intermediate	0.5	9	0		0	49
9	5.5	2	Intermediate	3.0	8	0		Liver (28)[a]	44
10	3.5	2	Low	1.5	8	0		0	47
11	2.2	5	Low	0	8	0		0	32
12	9	5	Intermediate	3.5	36	Obst.	Open GJstomy	0	24
13	2.3	2	Low	0.2	7	0		0	15
Mean	3.6	2.8		1.0	10.5	38.5%			48.6

HPF high-power field, *RM* resection margin, *Cx.* complication, *OP* operation, *Recur* recurrence, *F*/*U* follow-up, *DGE* delayed gastric emptying, *gr.* grade, *Obst.* obstruction, *Strict.* stricture, *Lapa GJstomy* laparoscopic gastrojejunostomy, *Open GJstomy* open gastrojejunostomy

[a]postoperative months

performed; hepatic metastasis occurred 28 months after surgery. This patient was treated with imatinib and showed partial response. Excluding this patient, all patients are alive without evidence of recurrence over a mean follow-up period of 48.6 months (range 15–73 months).

Discussion

Duodenal GISTs account for approximately 5% of all surgically resected GISTs. However, they represent approximately 30% of all primary duodenal neoplasms [18]. Because of the rarity of this disease, it is difficult to determine whether a duodenal GIST is malignant or benign. Additionally, the unique and complex nature of the anatomical location of duodenal GISTs makes determining the appropriate surgical strategy for them challenging. The most frequent onset sites are the second and third portions of the duodenum, which represents 56.2% of all duodenal GISTs [19, 20]. Therefore, surgical strategies for duodenal GISTs are largely dependent on tumor size and proximity to the ampulla of Vater. Common procedures for duodenal GISTs include wedge resection, segmental resection, and pancreaticoduodenectomy (PD). Currently, there are few data regarding the oncological safety of performing limited local resection compared to PD for duodenal GISTs. However, Connolly et al. [21] showed that encapsulated GISTs are rarely associated with wide infiltration and lymphatic metastases. Goh et al. reported that the oncological outcomes of limited resection were not inferior to PD [5], and a recent series concluded that local resection was not associated with an increased disease recurrence rate [7, 22, 23]. The short-term results of the current study indicate that limited resection may be an effective alternative that does not increase the risk of local recurrence or metastasis. However, despite their low malignant potential, Miettinem et al. observed late recurrence of duodenal GISTs—as late as 10 years after surgery [24]. Therefore, a large-scale study with long-term follow-up is still mandatory to demonstrate the oncological safety of local resection.

Short segmental resection of the duodenum with end-to-end duodenoduodenostomy is usually not eligible because the largest portion of the duodenum is attached to the pancreas and is, thus, immovable. Specifically, when removal of the segment of the duodenum that is distal to the ampulla of Vater is required, the whole distal duodenum and the first portion of the jejunum should be resected. This operation for infra-ampullary lesions was introduced in 1996 [25]. Indications for this procedure include the following: (1) an infra-ampullary lesion sparing the ampulla of Vater; (2) a duodenal defect that is too large for primary closure after wedge resection; (3) a tumor located on the mesenteric side that is not suitable for resection or repair; and (4) no tumor involvement in the pancreatic parenchyma. In addition to the above conditions, sufficient space (about 2 cm) for a surgical stapler between the ampulla and the tumor is a necessary consideration for a laparoscopic approach. If the tumor is located on the antimesenteric side, the space for a surgical stapler could be retained even when the vertical distance of the tumor from the ampulla is < 2 cm. When the lesion was too close to the ampulla, we used an open PSD to ensure protection of the ampulla or a pancreaticoduodenectomy.

For a successful and safe laparoscopic PSD, appropriate localization of the ampulla of Vater is essential. Common perioperative localization involved endoscopic clipping during gastrointestinal surgery [26, 27]. However, detectability by palpation was lost or limited during laparoscopy. Several localization techniques have been introduced to overcome the lack of tactile sensation during laparoscopic surgery, including preoperative endoscopic tattooing, intraoperative concomitant endoscopy, and preoperative endoscopic clipping combined with intraoperative radiography or laparoscopic ultrasonography to detect the clips [28–31]. We have tried various localization methods (Table 1). Although intraoperative duodenoscopy is usually a good option, it requires an experienced endoscopist and careful setting of the endoscopic instrument, which is costly in terms of both time and labor. In contrast, endoscopic tattooing immediately prior to the operation is simple and intuitive. The injected dye, however, spreads diffusely through the duodenal wall, which subsequently makes precise localization difficult and can cause a duodenal edema. We have recently used laparoscopic ultrasonography to localize the preoperatively placed clips on the antimesenteric side of the ampulla. Hyung et al. [31] described the efficiency and accuracy of laparoscopic ultrasonography to detect all localization clips at the gastric wall during gastric surgery. In our experience, the duodenal clips were easily detectable during the procedures. Despite the disadvantage that ultrasonography is dependent on the surgeon's familiarity, this technique is simple, accurate, and applicable in real time. Consequently, preoperative endoscopic clipping with intraoperative ultrasonography is currently our preferred localization technique.

The concept of this PSD might be attributed to the introduction of reconstruction technique. In fact, reconstruction techniques for laparoscopic side-to-side duodenojejunostomy have been introduced to treat superior mesenteric artery syndrome [32] and duodenojejunal junction GISTs [33]. Currently, however, minimally invasive PSD has only been described in small case series [9, 11, 12, 34, 35], specifically since Ammori et al. [10] reported the first laparoscopic surgery for duodenal stricture under the name of pancreas-preserving subtotal duodenectomy. To the best of our knowledge, this study is the largest series of laparoscopic PSDs for duodenal GISTs. Stauffer et al. [11] used the name of partial sleeve duodenectomy, but there is no commonly used terminology. This surgery is expected to become more common in the future, and consensus on the name of the operation will be needed.

PSD is an organ-sparing surgery to preserve the function of the ampulla and the pancreatic parenchyma. This procedure is supposed to prevent the severe morbidities associated with PD. Despite these advantages, we found that this technique was often associated with postoperative complications. Herein, we noted that the most common postoperative complications were with intestinal passage; two patients suffered from delayed gastric emptying (DGE) without evidence of any obstruction. The risk factors or causes could not be validated in this study because of the very small number of cases in our series. However, among the suggested causative factors for DGE, reduced motilin, which plays a key role in regulating gastric migrating motor complex (MMC), might be a possible cause [36, 37]. Motilin-immunoreactive cells are most abundant in the duodenal and jejunal mucosa, and duodenal resection decreases the plasma motilin level, resulting in disturbance of gastric contractions [36]. This theory is supported by evidence that DGE could similarly occur after PD [38] and segmental resection of the duodenum [19].

Another notable complication was stricture of the duodenojejunostomy site, which caused one of our patients to be readmitted, and was only resolved after operative gastrojejunostomy. We performed a duodenojejunostomy in the side-to-side manner using a surgical staple (Fig. 6). Factors affecting wound healing, including tissue tension and blood supply, should be carefully monitored, and accurate introduction of a stapler with the appropriate caliber for anastomosis is also crucial.

This is a retrospective study with selection biases for tumor size, tumor location, patient physical status, and surgeon preference. The number of subjects included in this study was too small to draw certain robust conclusions. DGE and anastomosis stricture are important postoperative complications to consider, but further investigations are needed to identify the causes and solutions for these complications. Nevertheless, PSD may be an oncologically effective strategy for managing duodenal and proximal jejunal GISTs based on the intermediate follow-up outcomes from this study. Furthermore, lap PSD is a technically feasible and appealing surgical modality in terms of safety and perioperative results.

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Compliance with ethical standards

Conflict of interest All authors declare that they have no conflict of interest or financial ties to disclose.

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