

Laparoscopic Pancreatectomy for Solid Pseudo-Papillary Tumors of the Pancreas is a Suitable Technique; Our Experience with Long-Term Follow-up and Review of the Literature

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ABSTRACT

Background. Solid pseudopapillary tumors (SPTs) are rare pancreatic neoplasms of low malignant potential that occur mainly in young women. Only 17 cases of SPT treated laparoscopically have been published in the literature and long-term follow-up data are still lacking.

Methods. Retrospective analysis of ten patients (8 women, 2 men; mean age, 25.4 years) (DS: 12.1; minimum 11, maximum 51) who underwent laparoscopic distal pancreatectomy with a definitive histological diagnosis of SPT. Long-term follow-up data were collected.

Results. The average tumor size was 43.8 mm (minimum 20, maximum 65 mm). The mean operative time was 177.5 minutes (DS: 53.7; minimum 120, maximum 255). In all, five patients underwent distal splenopancreatectomy; five patients underwent spleen-preserving distal pancreatectomy of whom three with splenic vessel preservation and two with the Warshaw technique. The conversion rate was nil and no case of perioperative mortality was recorded. The mean hospital stay was 7 days (DS: 2.7; minimum 4, maximum 12). Six patients had an uneventful postoperative course and four had postoperative complications. Two of them underwent reoperation, and the other two had non-surgical complications. After a median follow-up of 47 (range, 5–98) months, all patients were alive and disease-free.

Conclusions. Laparoscopic pancreatic resection is a safe and feasible procedure that could become the treatment of choice for patients affected by pancreatic SPT. Distal pancreatectomy should be performed, if possible, with spleen-preserving technique, especially in young patients. To avoid metastatic spread, laparoscopic or laparotomic biopsy should not be performed in patients affected by SPT.

The first laparoscopic pancreatoduodenectomy was performed in 1994 and distal pancreatectomy (DP) in 1996.^{1,2} The laparoscopic approach not only offers the operative advantage of better view in terms of magnification but also improves the patient's postoperative recovery, coupled with acceptable rates of perioperative morbidity.³ DP is well suited for the laparoscopic approach because of the absence of anastomosis. Moreover, the magnified view of the operative field facilitates separation of splenic vein and artery from the pancreatic parenchyma and adds a potential advantage in performing the spleen preserving technique.

A large, multicenter experience suggested that the laparoscopic approach should be reserved for benign tumors, even though recent studies have affirmed that laparoscopic distal pancreatectomy has similar outcomes as the open approach even for ductal adenocarcinomas.^{3,4}

Solid pseudopapillary tumors (SPTs) are one of the most uncommon histotypes (1–2%) of all exocrine neoplasms of the pancreas. They have a low malignant potential and occur mainly in young non-Caucasian women between the 2nd and 3rd decade, even though rare cases have been reported in children and men.^{5–10}

Described for the first time by Frantz in 1959, it was included in the World Health Organization's classification in 1996 with the name of SPT.^{11,12}

Its prevalence in young women indicates a connection between tumor development and hormones, probably more likely in its growth than in its pathogenesis.^{13–15}

Histopathogenesis of this tumor is still unclear; acinar, ductal, endocrine, and primordial cell origin has been proposed.¹⁶ Radiological features of these tumors are not specific and the immunophenotypic aspects do not correspond to those of normal pancreatic cells.¹⁷

Several studies have demonstrated the low malignant potential of these tumors with a specific mortality lower than 2%, and only 10–15% of patients with local recurrence or metastases after resection.^{9,18,19} Surgery represents the treatment of choice for these neoplasms. Moreover, a definitive diagnosis can only be done through pathological examination of the surgical specimen.^{7,18,19}

A uniform agreement on the use of laparoscopy as the surgical treatment of choice for patients with SPT still does not exist. In the literature, there have been published only 14 cases of SPT with good outcome after laparoscopic resection, however, data on long-term follow-up are still awaited. A recent paper, published by Matos et al., reported one case in which the patient was still alive and disease-free at 32 months after laparoscopic distal pancreatectomy.^{10,20–29} Considering the low malignancy of these tumors, the authors concluded that the laparoscopic approach might be a reasonable surgical option. Moreover, the laparoscopic resection offers the advantage of sparing pancreatic parenchyma, even if more studies and longer term follow-up are needed.²⁹

A recent study has questioned the appropriateness of the laparoscopic approach as a valid treatment for this kind of tumors.³⁰ In this study, three SPT patients underwent laparoscopic biopsy of the neoplasia and all of them developed recurrences.

The purpose of our study was to better define the long-term follow-up results after laparoscopic resection in patients affected of SPT by analyzing our experience between 2001 and 2009 and reviewing the literature.

PATIENTS AND METHODS

We retrospectively analyzed prospectively collected data regarding clinical presentation, laboratory findings, radiological imaging, intraoperative and postoperative parameters of ten patients (8 women, 2 men; mean age, 25.4 years) (DS 12.1, minimum 11, maximum 51) who underwent laparoscopic distal pancreatectomy with a definitive histological diagnosis of SPT, between August 2001 and May 2009 in two large centers (8 patients were operated on at General and Pancreatic Surgery Unit, Policlinico G.B. Rossi, Verona and 2 patients at General Surgery Unit Baggiovara Hospital, Modena, Italy). All patients

underwent laparoscopic pancreatic resection after informed consent. All diagnoses were histologically confirmed.

Long-term follow-up data were obtained during outpatient clinic or direct evaluation of CT or MRI scan (follow-up ended in October 2009).

The surgical technique used in all cases was the same. The patient was placed supine in the reverse 30° Trendelenburg position, with the surgeon placed in the middle between the patient's legs and the screen placed on the left side of the patient's head. A 10-mm camera port was placed at the umbilicus and two additional working 5 mm ports were placed in the xiphoid area and the right flank. A 12-mm port, for the linear stapler, was placed in the left flank. Dissection was made with electrocautery and Liga-Sure Atlas. The pancreas was exposed anteriorly by a window in the gastrocolic ligament, sparing the short gastric vessels. The inferior and superior border of the gland was mobilized, and the body/tail of the pancreas was detached from the retroperitoneum to visualize the posterior wall of the pancreas and the splenic vessels. The pancreas was transected with an endoscopic linear 45-mm stapler device. Five patients (50%) underwent spleen-preserving distal pancreatectomy; in three of them the splenic vessels were preserved, whereas in the remaining the procedure was performed with the Warshaw technique.

The specimen was placed in an endoscopic bag retrieval system and extracted through a Pfannenstiel incision. A soft nonsuction drain was placed on the pancreatic stump and drawn out through the 12-mm port site.³¹

A revision of the literature was done by a search in PubMed utilizing the terms laparoscopic pancreatectomy, solid pseudopapillary tumor.

RESULTS

In four cases the diagnosis was incidental (40%) and in six cases (60%) clinical symptoms were present. The prevailing symptom was continuous, vague abdominal pain. Presence of a palpable mass located in the upper abdominal quadrant occurred in two cases (20%).

The laboratory tests did not reveal any significant alterations. All patients underwent ultrasonography (US) and abdominal contrast-enhanced computer tomography (CE-CT) scan or magnetic resonance imaging (MRI) preoperatively.

The average tumor size was 43.8 mm (minimum 20, maximum 65 mm). The mass had a solid aspect in 40% of cases (4/10), whereas another 40% presented an internal necrotic-hemorrhagic area. Aspects of vascular infiltration and adhesion to mesenteric vessels were not observed in any of the cases. The median number of lymph nodes examined was 8 (range, 0–16), and the number of lymph

nodes involved was zero. Metastases were never detected, and all the lesions were potentially resectable at the end of the diagnostic workup. We never performed preoperative biopsy.

All surgical procedures were completed laparoscopically with no cases of conversion to open procedure. The mean operative time was 177.5 min (DS 53.7; minimum 120, maximum 255). Intraoperative blood transfusion was never needed.

No cases of perioperative mortality were recorded. The mean hospital stay was 7 days (DS 2.7; minimum 4, maximum 12). Six of 10 patients (60%) had an uneventful postoperative course with a mean hospital stay of 6.3 days (DS: 2.5 minimum 4, maximum 11). Four patients (40%) had postoperative complications. The mean hospital stay was in these cases 8 days (DS 3.1; minimum 5, maximum 12). Two of these patients had surgical complications that led to reoperation. In one case, reoperation was performed on the first postoperative day for bleeding after spleno-pancreatectomy. The procedure was done laparoscopically and consisted of hemostasis on the pancreatic stump by

Endoclip placement. This patient also had bronchopulmonary complications. The other patient was readmitted to the hospital after 15 days for fever and abdominal pain. She underwent reoperation for intra-abdominal hematoma after a spleen-preserving distal pancreatectomy. The procedure consisted of laparotomy, drainage of a retrogastric hematoma, and peritoneal toilette. Her subsequent postoperative course was characterized by a grade A pancreatic fistula. The other two patients had nonsurgical complications. One was affected by a chylous fistula, which was treated conservatively on an outpatient basis and resolved within a month after surgery. The last patient developed bronchopulmonary complications and wound infection and was treated by antibiotic therapy and wound cleansing. All patients were discharged as disease-free with a Karnofsky score of 100%. The lesion macroscopically appeared well circumscribed in all cases. After a median follow-up of 47 (range, 5–98) months, all patients were alive and free of disease. At time of writing, none of the patients is receiving pancreatic enzyme therapy and no case of diabetes has been observed. Follow-up ended in October 2009.

TABLE 1 Main characteristics, clinical features, localization, and size of SPT reported in literature and personal experience

Study	Age (year) and gender	Signs and symptoms	Tumor site	Tumor size (cm)
Kang ^{20,27,28}	25 F	No symptoms	Tail	4.5
	44 F	No symptoms	Body-tail	3.5
	32 F	No symptoms	Tail	4
	30 M	No symptoms	Tail	3
Han ²¹	—	—	—	—
Matos ²⁹	16	No symptoms	Body	2.5
	26	Back pain	Tail	5.9
Machado ²²	26 F	—	Uncinate process	4
Laxa ²³	—	—	—	—
Sokolov ²⁴	13 F	abd pain	Neck	—
	13 F	abd pain	Tail	—
Sasaki ²⁵	31 F	abd pain	Body-tail	5.5
Fernández-Cruz ²⁶	—	—	Body-tail	—
	—	—	Body-tail	—
Fais ³⁰	9 F	abd pain	Tail	—
	11 F	No symptoms	Body-tail	4.5
	12 F	hematemesis and melena	Head	—
Present series	35 M	No symptoms	Tail	5
	21 F	abd mass, abd pain	Body-tail	5
	11 F	abd pain	Body-tail	5
	35 F	abd mass, abd pain	Tail	5
	19 F	No symptoms	Body-tail	2
	51 F	No symptoms	Body-tail	4
	21 F	No symptoms	Body-tail	5
	17 M	No symptoms	Body-tail	3.5
	25 F	No symptoms	Body-tail	6.5
	13 F	abd pain	Body-tail	3

F female, M male, abd abdominal

A revision of the literature was done and a summary of the clinical characteristics and results of all patients are reported in Tables 1 and 2.

DISCUSSION

Only 17 cases of patients affected by SPT, who underwent laparoscopic distal pancreatectomy have been reported in literature (Tables 1 and 2).^{10,20–30} The authors

have reported good short-term results, with no postoperative complications. Unfortunately though, long-term follow-up is not available. Especially in case of pancreatic SPT, long-term survival is highly expected as long as surgery is safely performed. Therefore, surgeons dealing with this tumor need to take long-term outcomes into consideration.²⁷

In a recent study, Fais et al. reported three cases of recurrence after laparoscopic approach for SPT: two cases

TABLE 2 Therapy and outcome of SPT reported in Literature and personal experience

Study	Therapy	Postoperative complications	Outcome
Kang ^{20,27,28}	LPR (DP + splenectomy)	NO	–
	LPR (DP)	NO	–
	LPR (spleen-preserving DP with segmental resection of splenic artery)	NO	NED at 6 months
	LPR (spleen-preserving DP and cholecystectomy)	NO	NED at 6 months
Han ²¹	LPR (DP)	NO	NED at 19.8 months (mean of 5 resections)
Machado ²²	LPR (PD)	NO	NED at 2 months
Matos ²⁹	LPR (enucleation)	NO	NED at 13 months
	LPR (spleen-preserving DP)	YES (pancreatic-grade A-fistula)	NED at 32 months
Laxa ²³	LPR (hand assisted?)	–	–
Sokolov ²⁴	Central pancreatectomy (with pancreogastroanastomosis)	NO	NED after 6 months
	LPR (spleen-preserving DP)	NO	NED after 24 months
Sasaki ²⁵	LPR (spleen-preserving DP)	NO	–
Fernández-Cruz ²⁶	LPR (DP + splenectomy)	NO	–
	LPR (DP + splenectomy)	NO	–
Fais ³⁰	Laparoscopy biopsy, then PLR → peritoneal mets 43 months after → 5-FU + oxaliplatin + irinotecan → mets resection	NO	NED at 12 months after resection of recurrence
	Laparoscopy biopsy → open DP → peritoneal mets 40 months after → mets resection → 5-FU + oxaliplatin + irinotecan	–	NED at 43 months after resection of recurrence
	Laparoscopy biopsy → open PD (R2) → local recurrence 32 months after	–	Alive with local recurrence 2 years from the tumor resection
Present series	LPR (DP + splenectomy)	YES (reoperation for hemoperitoneum)	NED at 30 months
	LPR (spleen-preserving DP)	NO	NED at 62 months
	LPR (spleen-preserving DP)	NO	NED at 62 months
	LPR (DP + splenectomy)	YES (wound infection + bronchopulmonary complication)	NED at 45 months
	LPR (spleen-preserving DP)	YES (reoperation for retrogastric hematoma, blood transf., pancr. fistula)	NED at 66 months
	LPR (DP + splenectomy)	NO	NED at 98 months
	LPR (spleen-preserving DP)	NO	NED at 82 months
	LPR (DP + splenectomy)	NO	NED at 15 months
	LPR (DP + splenectomy)	NO	NED at 12 months
	LPR (spleen-preserving DP)	YES (chylous fistula)	NED at 5 months

LPR laparoscopic pancreatic resection, *DP* distal pancreatectomy, *PD* pancreaticoduodenectomy, *NED* no evidence of disease, *mets* metastases, *5-FU* 5-fluorouracil

after laparoscopic biopsy and the last after biopsy and laparoscopic resection.³⁰ These poor results are contradictory with the low risk of local or disseminated recurrence after open laparotomy without chemotherapy previously reported in literature.³⁰ The authors hypothesized that recurrences after laparoscopic management may have been due to diffusion of tumor cells caused by gas insufflation, especially during biopsy, and concluded that a larger series will be needed to evaluate the safety and efficiency of laparoscopy for the management of tumors such as SPT, without chemotherapy. On the basis of their experience, Fais et al. introduced doubts about the appropriateness of the laparoscopic approach for patients suffering from SPT. This report can increase the suspicion that the laparoscopic approach could be detrimental for these patients. In one of our previous studies, we reported good results after 1 year follow-up in an 11-year-old patient who underwent laparoscopic pancreatic resection for a SPT.¹⁰

In the present study of 10 consecutive resections for SPT, we reached a median follow-up of 47 months with no recurrence of disease. This data strongly support the hypothesis that laparoscopic approach in the surgical management of SPT is suitable and safe.

Data concerning preoperative cytological tests are conflicting.³² We do not submit our patients to fine-needle aspiration or preoperative biopsy; the presence of a "solid" resectable tumor in young patients has negatively influenced our strategy about the utility of fine-needle aspiration in our series. Moreover, some authors have reported that SPT of the pancreas may spread outside the pancreas, particularly in peritoneal cavity. Metastatic spread may be promoted by trauma, including tumor biopsies, which should never be performed.³³

According to the results from our cohort of patients and other previous studies, we consider the laparoscopic technique a safe and feasible procedure, which in expert hands can be completed in a reasonable time and yield good results.

The laparoscopic approach has always been curative for SPT whenever a radical resection, avoiding to break the mass, has been performed. In terms of complications, the literature data and this series show that the laparoscopic approach does not represent a risk factor for major complications.^{3,31}

Laparoscopic spleen-preserving distal pancreatectomy is a beneficial procedure for patients with benign and borderline malignancies of the pancreas. With such a procedure, it is best to try to preserve the splenic artery and vein, especially in young patients, such as those generally affected by a SPT.

Patients who undergo laparoscopic surgery have a faster recovery with a shorter hospital stay; even when complications occur the mean hospital stay is not significantly prolonged (6 vs. 8 days). This technique, however, should

be considered as an advanced laparoscopic procedure and its application in pediatric cases is still under development. Only surgical teams with advanced laparoscopic skills should to perform it.

In our opinion, laparoscopic biopsy should not be performed in patients affected by SPT.

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