

# Laparoscopic Surgery for Solid Pseudopapillary Tumor of the Pancreas

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## ABSTRACT

**Background and Objectives:** Solid pseudopapillary tumors of the pancreas are rare and occur most frequently in young women. They have an uncertain pathogenesis and unclear clinical behavior. Our aim was to evaluate the clinical presentation of solid pseudopapillary tumors and assess the efficacy of treatment with minimally invasive surgery.

**Methods:** From March 1997 to February 2011, 13 of 273 patients who underwent laparoscopic procedures on the pancreas were found to have solid pseudopapillary tumors. There were 12 female patients and 1 male patient. The median age was 21 years (range, 15–77 years). Abdominal pain was the most common presenting symptom (n = 9). Tumors were incidentally found in 3 patients on computed tomography scans obtained for other reasons.

**Results:** Enucleation of the tumor was performed in 4 patients, including 3 in whom the tumor was located in the head of the pancreas. Eight patients underwent distal pancreatectomy with splenectomy, whereas spleen-preserving distal pancreatectomy was performed in one case. The median tumor size was 6 cm (range, 1.5–11 cm), the median operative time was 197 minutes (range, 68–320 minutes), and the median blood loss was 50 mL (range, <50–750 mL). Distal resections were performed with a linear stapler. Four patients had postoperative complications. The median length of hospital stay was 5 days (range, 2–12 days). During a median follow-up period of 11 months (range, 3–121 months), no local recurrences or distant metastases were found.

**Conclusion:** Laparoscopic resections and enucleations of solid pseudopapillary tumors of the pancreas can be performed safely and with adequate resection margins even if the tumors are located in the head of the organ.

**Key Words:** Solid pseudopapillary tumor, Pancreas, Frantz-Gruber tumor, Laparoscopy, Enucleation.

## INTRODUCTION

Solid pseudopapillary tumor of the pancreas (SPT), also known as Frantz-Gruber tumor, was originally established as a separate entity in 1959<sup>1</sup> and represents 1% to 3% of all pancreatic tumors. It is most commonly seen in young female patients<sup>2–5</sup>, and only a few cases have been reported in male patients aged >60 years. The most frequent presenting symptom is abdominal pain, but patients may also remain asymptomatic.<sup>4,6</sup> Other symptoms include nausea, vomiting, weight loss, and abdominal fullness. On clinical examination, a palpable abdominal mass is the most common finding. The tumor size varies substantially (from 1.5 to 30 cm).<sup>3,7</sup> These tumors can occur in any part of the pancreas and are characteristically surrounded by a defined capsule. Rarely, the tumor cells may invade the capsule and metastasize, most commonly to the liver.<sup>4,8,9</sup>

Typically, findings of the laboratory tests including serum amylase and tumor markers (CA 19–9, CA 125, carcinoembryonic antigen [CEA],  $\alpha$ -fetoprotein) are normal. The treatment of choice is surgical resection. Because most of the patients affected by SPT are young female patients, the cosmetic concerns may be of significance. Therefore laparoscopic surgical removal of this tumor is appropriate. A low incidence of recurrence and good prognosis even after reoperation have also resulted in acceptance of laparoscopic surgery. Laparoscopic resection may therefore be appropriate for such pancreatic tumors if highly skilled surgeons are available.<sup>10</sup> Although recurrence can occur, the prognosis after surgical resection is excellent.<sup>7,11</sup>

This work describes 13 cases of SPT treated at Oslo University Hospital—Rikshospitalet, a reference hospital in Oslo, Norway.

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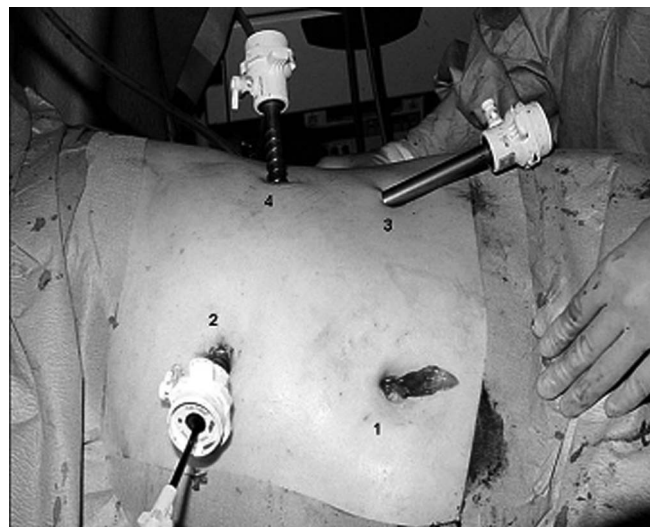
## MATERIALS AND METHODS

During the period from March 1997 to February 2011, we performed 273 laparoscopic procedures on the pancreas at Oslo University Hospital—Rikshospitalet. There were a wide range of laparoscopic operations, with 142 distal pancreas resections with splenectomy, 61 spleen-preserving distal resections, 32 enucleations, 2 Whipple operations, and 36 other procedures mainly presenting as diagnostic laparoscopies with curative intent. Among the 273 patients operated on, 13 were found to have SPT as the final histopathologic diagnosis. There were 12 female patients and 1 male patient. The median age was 21 years (range, 15–77 years). The most common complaint before surgery was abdominal pain and/or nausea and vomiting, which was present in 9 patients (69%). Six patients complained of weight loss (46%). Six patients presented with a mass in the abdomen (46%). Three patients were incidentally diagnosed on computed tomography (CT) scanning for other reasons (23%). In one patient, drainage for what was suspected to be a pancreas pseudocyst was initially attempted, which eventually was histologically confirmed to be an SPT.

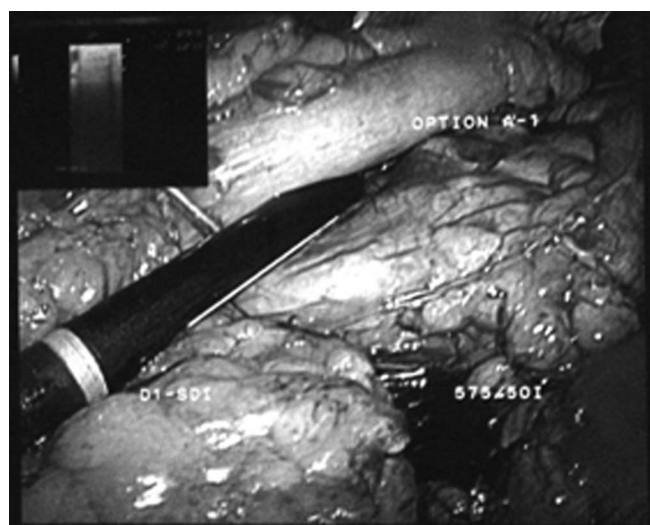
Preoperatively, the localization of the tumors was determined by abdominal CT scans. Occasionally, magnetic resonance imaging (MRI) and abdominal or endoscopic ultrasonography were used as supplements. Preoperative biopsy of the lesion in the pancreas is not routinely performed in our department, but 5 of the patients underwent cytology/biopsy at other hospitals before referral to our institution. The tumor was located in the head, body, and tail of the organ in 3, 5, and 5 patients, respectively.

Since the introduction of laparoscopic pancreatic surgery at our institution in 1997, open pancreatic surgery for lesions located in the body or tail of the pancreas and encapsulated tumors in the head of the pancreas has been used only in very few cases with suspected vascular involvement. The surgical technique for laparoscopic pancreas resection has previously been described in detail.<sup>12,13</sup> The typical placement of trocars is presented in **Figure 1**. Patients with lesions in the body or tail of the pancreas were placed in a 30° to 45° modified right lateral position. When tumors were localized to the head of the pancreas, the patient was placed in a supine position and trocar 2 was placed on the right side of the right rectus sheath, instead of the epigastrium, as shown in **Figure 1**.

The following preoperative and intraoperative criteria for safe enucleation were pursued: (1) clinical diagnoses, (2) CT scan confirming the tumor with a solid and cystic



**Figure 1.** Postoperative view showing the standard trocar placement for a distal (left-lateral) resection. Trocar 1 is introduced through the umbilicus by an open approach. This trocar and trocar 2 are the surgeon's working ports, whereas the camera is placed at trocar site 3. Trocar 4 is used for retraction for the assistant. The specimen is usually retrieved by an infraumbilical extension of trocar site 1.



**Figure 2.** Intraoperative laparoscopic ultrasonography.

pattern surrounded by a capsule, and (3) the relation between the tumor and the pancreatic duct. In case of a very close relation, the pancreatic duct may undergo stenting.

Intraoperative ultrasonography of the pancreas was performed to localize the lesion and ensure a complete resection (**Figure 2**).

When a left-lateral, distal resection was performed, a linear stapler (Endo GIA Green cartridge; Covidien, Mansfield, Massachusetts) was used to divide the pancreas, and the margin of the remnant pancreas was in most cases covered with a TachoSil patch (Nycomed, Pharmaceutical Co. Ltd, Roskilde, Denmark).

A silicone tube (Aquerius, Degania Silicone Ltd, Degania Bet, Israel) was routinely placed near the resection margins, and the concentration of amylase in the drainage fluid was checked daily. The decision to remove the drain was based on a combination of the amount of fluid drained and the amylase concentration of the fluid.

Results are presented as median (range). The severity of the postoperative pancreatic fistulas was evaluated according to the International Study Group for Pancreatic Fistula (ISGPF) definition: the amount of fluid drained on or after the third postoperative day with amylase content >3 times the serum amylase level. The fistulas were assigned 1 of 3 grades (grade A, B, or C).<sup>14</sup> Perioperative adverse events were assessed in accordance with standard grading systems—the Oslo classification of intraoperative incidents and the revised Accordion classification.<sup>15</sup> Statistical analysis was conducted with SPSS, version 18.0 (IBM, Armonk, New York).

The follow-up was performed by outpatient visits with abdominal CT scans as the preferred imaging modality. Ultrasonography and MRI were used when required.

## RESULTS

Of 13 patients diagnosed with SPT, 9 underwent laparoscopic distal pancreatectomy, 1 of whom had a spleen-preserving procedure, whereas 4 underwent enucleation of the tumor. In 3 of the patients who underwent enucleation, the tumor was located in the head of the pancreas, thus avoiding major surgery such as pancreaticoduodenectomy (Whipple resection). One of these 3 patients underwent reconstructive hepaticoduodenostomy after enucleation of the tumor with resection of the distal bile duct and cholecystectomy.

The median operative time was 197 minutes (range, 68–320 minutes) in total, including 143 minutes (range, 70–243 minutes) for distal resections and 205 minutes (range, 68–320 minutes) for enucleations. The median blood loss was 50 mL (range, <50–750 mL) in total, including <50 mL (range, <50–750 mL) for distal resections and 50 mL (range, <50–300 mL) for enucleations.

No metastases were verified intraoperatively. An abdominal suction drain was placed in 12 patients. There were no conversions to open surgery or other unfavorable intraoperative incidents.

All the specimens underwent histopathologic examination at our hospital. The median size of the tumor was 6 cm (range, 1.5–11 cm). All resections were reported as R0 resections. There were no cases in which infiltration of blood vessels, lymphatics, or other surrounding organs were described. Only one patient had a tumor with central calcification.

Postoperative complications were seen in 4 patients. Of these, one patient was reoperated on laparoscopically the same evening because of intra-abdominal bleeding (Accordion grade 4 complication). There was a small bleeding artery at the corner of the pancreatic resection area, which was treated with a clip. In one patient a left subphrenic abscess developed, which was managed conservatively with antibiotics (grade 2). One patient had minor hemorrhage from the umbilical incision, which was controlled by compression (grade 1). Furthermore, one patient was readmitted twice, on the 10th and 16th postoperative day, with abdominal pain. Abdominal ultrasonography showed a small fluid collection near the resection area, and the patient was managed conservatively without any intervention (grade 1). Pancreatic fistula did not develop in any of the patients.

Postoperatively, opioid medications were needed for a median of 1 day (range, 0–5 days). Eight patients were able to start oral intake of fluids the same day as surgery, whereas the remaining 2 and 3 patients started oral intake on the first and second postoperative days, respectively. The median number of days with the abdominal drain was 3 days (range, 0–6 days). The median duration of postoperative hospital stay was 5 days (range, 2–12 days), similar for distal resections and enucleations.

The median follow-up period was 11 months (range, 3–121 months). At the constellation of this article, all patients are alive without any recurrences or metastases.

Major perioperative outcomes are summarized in **Table 1**.

### Tumor in Head of Pancreas

Our series included 3 cases with tumors in the head of the pancreas. None of these patients presented with jaundice, in accordance with previous reports.<sup>6,8,11,16,17</sup> All 3 patients underwent laparoscopic enucleation of the tumors, thus avoiding the major surgical alternative procedure, which in most cases would be a pancreaticoduodenectomy. Of

**Table 1.**  
 Perioperative Data

Parameters	Distal resection (n = 9)	Enucleation (n = 4)	Total (N = 13)
Operative time (min)	143 (70–243)	205 (68–320)	197 (68–320)
Blood loss (mL)	<50 (<50–750)	50 (<50–300)	50 (<50–750)
Tumor size (cm)	6 (1.5–11)	5 (2.5–10)	6 (1.5–11)
Complications	2 (22.2%)	2 (50%)	4 (30.1%)
Postoperative stay (d)	5 (3–12)	4.5 (2–8)	5 (2–12)
Postoperative opioids (d)	1 (0–2)	1 (0–5)	1 (0–5)
Drain placement (d)	3 (1–5)	2 (0–6)	3 (0–6)

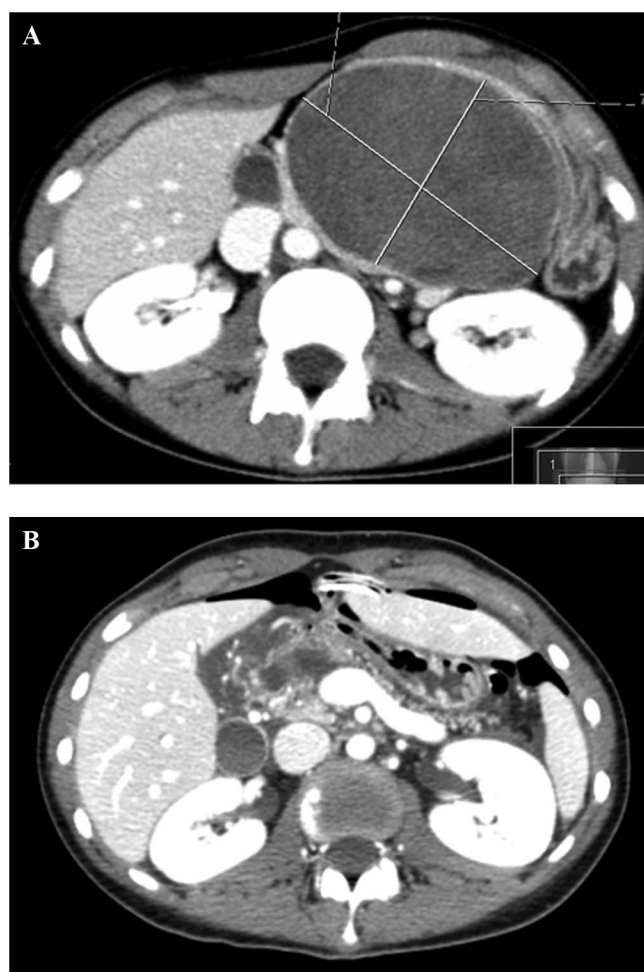
Data are presented as median (range) or number (percent).

particular interest is the case of a 15-year-old girl who presented with a 5-month history of abdominal pain and vomiting, as well as a gradually increasing abdominal mass. An abdominal CT scan showed a well-defined 7.8 × 10-cm mass in the head of the pancreas dorsal to the left lobe of the liver (**Figure 3**). It was difficult to localize the tumor because of peritumoral edema. The tumor was located to the right of the portal vein; it was carefully dissected free from the head of the pancreas and the surrounding vessels and enucleated in toto without rupture of the capsule. It was removed through a small infraumbilical incision. When sliced, it contained tumor tissue and signs of hemorrhage. The patient was discharged on the eighth postoperative day, and there were no signs of recurrence after 36 months' follow-up.

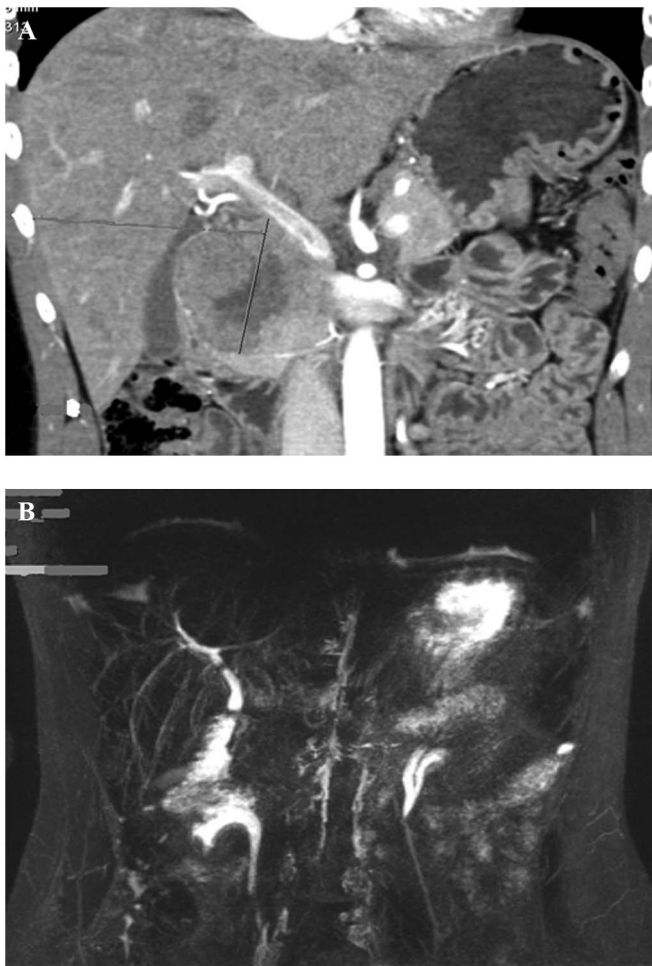
Another 15-year-old girl was admitted to the emergency department with a 1-year history of increasing abdominal pain. A CT scan showed a large tumor in the head of the pancreas (**Figure 4**). During the laparoscopic removal, the bile duct was shown to be adherent to the cranial portion of the tumor. Hence, separation without violation of the tumor integrity was impossible. Therefore, the choledochus was divided above the tumor, the gallbladder was removed, and a hepaticoduodenostomy with a 7-French stent was performed. A minor left subphrenic abscess developed, which was managed conservatively with antibiotics. The patient was discharged on the fourth postoperative day. Further CT and Magnetic resonance cholangiopancreatography controls showed no pathology and no signs of recurrence after 56 months' follow-up.

## DISCUSSION

Cystic pancreatic neoplasms are very uncommon entities. First described by Frantz<sup>1</sup> in 1959 as a distinctive tumor



**Figure 3.** Case X. A, Preoperative contrast CT scan showing 103.7 × 77.8-mm pancreatic mass under the left lobe of the liver pushing the stomach to the left. B, Postoperative CT scan.



**Figure 4.** Case Y. A, Preoperative CT scan showing an SPT in the head of the pancreas. B, Postoperative Magnetic resonance cholangiopancreatography showing hepaticoduodenostomy.

entity, this tumor was given the name solid pseudopapillary neoplasm by the World Health Organization in 1996.<sup>18</sup> Currently, nearly 1000 cases of established SPT have been documented in the English-language literature with a female-male ratio of 10:1. SPT is most frequently diagnosed in the third decade of life.<sup>3,4,8,19,20</sup> The most frequent symptom of these tumors is upper abdominal pain, seen in nearly half of the patients. Symptoms may be present for years before the correct diagnosis is made.

A CT scan was the main diagnostic imaging modality used in our study, although we supplemented this with MRI and ultrasonography in some patients. CT scan and MRI of abdomen show the characteristic features of this tumor, and the diagnosis can be made by combining these imaging modalities with the typical clinical presentation of SPT.<sup>21</sup> In addition, they can also show tumor calcifi-

cation. Tang et al<sup>11</sup> reported that 30% of cases showed tumor calcification in their series. In our study only one patient had tumor calcification. We also used intraoperative ultrasonography to verify the exact relations of the tumor to its surrounding structures.

In the literature, numerous SPTs have been reported to be found incidentally on CT scans performed for some other reason.<sup>22</sup> In our series we had 3 such cases, 1 of whom was a 77-year-old man who had an abdominal CT scan after removal of a superficial bladder tumor. On the CT scan, it was found coincidentally that the patient had a tumor in the pancreas. Successful laparoscopic resection of this tumor has been performed, and the patient is alive with a follow-up period of 12 months.

In clinical practice, SPT can be often mistaken for pancreatic pseudocyst. In our series there was one case in which the patient underwent endoscopic ultrasonography-guided drainage of suspected pancreatic pseudocyst at a local hospital. However, the stent could not drain the cyst, and biopsy findings showed SPT. The patient was referred to our clinic and underwent a distal pancreatectomy with splenectomy. The patient was discharged on the third postoperative day, and there were no signs of recurrence after 24 months of follow-up.

The diagnosis of SPT should be verified before planning and performing an enucleation. These are cystic pancreatic tumors, and it would be inappropriate if an enucleation was performed for certain tumors with a malignant potential, for example, intraductal papillary mucinous neoplasms. However, preoperative biopsy is not our hospital policy, and we avoid it because it has limited diagnostic accuracy and it may result in tumor dissemination in case of malignancy. We advocate a more accurate diagnostic approach based on clinical findings, CT scan, intraoperative ultrasonography, and intraoperative instrumental palpation of the tumor.

A recent report described 3 cases of SPT in which recurrences developed after laparoscopic biopsy and/or resection.<sup>23</sup> It has also been reported that preoperative biopsy has limited diagnostic accuracy.<sup>4,24–26</sup> Butte et al<sup>27</sup> reported on 45 patients, 18 of whom had preoperative biopsy, with a diagnostic accuracy of 56%. In a study by Reddy et al,<sup>6</sup> only 8 of 13 patients with SPT had their diagnosis confirmed by preoperative cytology. We did not investigate our patients with preoperative tumor markers such as CA 19–9, CA 125, CEA, and  $\alpha$ -fetoprotein because various previous studies have shown normal preoperative tumor marker levels.

Surgery, whether open or laparoscopic, is the treatment of choice for SPT.<sup>28,29</sup> Although the first laparoscopic pancreatic surgeries were performed >15 years ago, conventional open laparotomies are still used for most resections of pancreatic tumors. However, a trend toward laparoscopic pancreatic tumor resection is developing. Laparoscopic excision of the SPT is safe, feasible, more appropriate, and preferable to conventional open surgeries for these tumors in the pancreatic tail.<sup>5,30–32</sup> It has well-documented advantages as compared with open surgery. In this study all patients were treated with laparoscopic resections because we have not been performing open surgery for the isolated lesions in the corpus and the cauda of the pancreas since the introduction of laparoscopic pancreatic resections at our institution. For lesions located in the head of the pancreas, many factors play a role, but if a well-defined capsule is present and there is no ingrowth into the adjacent organs, laparoscopic enucleation should be seriously considered.

Li et al<sup>33</sup> reported on 14 patients out of 34 who had tumors located in the head and neck of the pancreas, 10 of whom underwent Whipple operations. With the availability of expertise, such extensive procedures can be avoided. Recently, Alvise et al<sup>24</sup> reviewed 10 cases of laparoscopic distal pancreatectomy with or without splenectomy with a median follow-up period of 47 months for SPT in the body and tail of the organ. At the same time, laparoscopic surgery of the pancreas requires highly equipped institutions and surgeons with knowledge in pancreatic surgery and skills in advanced laparoscopy.

The spleen is an important immunologic organ, but in this series the spleen-preserving rate is low. The reason is that SPT has an uncertain pathogenesis and it is classified as a malignant tumor with a potential to metastasize to the liver, lymph nodes, and spleen, as well as in the splenic hilum. Second, the inflammatory process around the tumor makes it difficult to perform a safe dissection of the splenic vessels.<sup>4,8</sup>

Pancreatic resections for any reason lead to a 2% to 4% mortality rate and 30% to 50% morbidity rate. In our study we had a few complications and no deaths. This is in accordance with our previous results in which the fistula incidence was 10% for 170 procedures, and to date, after 273 cases, it is still the same (10%). This could be because of operative technique and surgical expertise. We used a linear stapler (Autosuture, Norwalk, Connecticut, USA) to divide the pancreas in distal resections. Our transection method for the pancreas might be a reason for the low numbers of complications, by using a “green” cartridge (60 mm) instead

of blue and red cartridges and slowly closing the stapler during the transection, which is important to avoid bursting of the parenchyma proximal to the stapling line.

Unfortunately, long-term follow-up data are still lacking for laparoscopic resections in SPT patients. Matos et al<sup>17</sup> included two patients in their series who underwent laparoscopic procedures. One of these patients underwent laparoscopic distal pancreatectomy and remained alive and disease free for >32 months. Our study shows that all of our patients are alive and disease free with a median follow-up period of 11 months (range, 3–121 months). Although there are reports of using postoperative chemotherapy and radiotherapy, none of our patients received this treatment modality.<sup>7,29</sup>

## CONCLUSION

SPTs possess a malignant potential and must be treated aggressively. The known treatment for SPTs of the pancreas is surgical resection. Laparoscopic resection has the advantages of minimally invasive surgery along with cosmetic benefits, a quicker recovery, and a shorter hospital stay. With the availability of expertise, tumors in the head can be enucleated and extensive procedures such as pancreaticoduodenectomy can be avoided.

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