# Laparoscopic Spleen-Preserving Distal Pancreatectomy in a Solitary True Pancreatic Cyst

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### ABSTRACT

**Background:** Solitary true pancreatic cysts (STPCs), or epithelial cysts, are benign lesions that are extremely rare in adult patients. Advances in radiographic techniques have improved the ability to identify pancreatic cystic lesions. We report a case of a large and symptomatic STPC in a 47-year-old female patient who was treated successfully with spleen-preserving laparoscopic distal pancreatectomy. We also review the clinical and pathologic features of all reported STPCs within the past 25 years.

**Database**: To compose the review, we did a search of the international literature for STPCs that had occurred in adults. Fourteen related articles were found describing cases of STPCs. Clinical and pathologic information was collected for all of the reported pancreatic cysts, and a database was formed. STPCs are detected more frequently in women than in men. The mean age of occurrence is 43.2 years, and the mean cyst size is 5.6 cm. Fifty percent of true cysts are located in the head of the pancreas. Size and site are responsible for the symptoms caused, although 22.8% were asymptomatic. Diagnosis was made postoperatively in all cases by histopathologic studies. No case of malignancy was reported in any STPC.

**Conclusions:** STPCs are rare and benign lesions commonly discovered incidentally during abdominal imaging. Surgical treatment is considered the appropriate therapy for large and symptomatic STPCs. The definitive diagnosis is established by histopathologic and immunohistochemical studies.

**Key Words:** Pancreatic cyst, Laparoscopic pancreatectomy, Epithelial cyst, True cyst.

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## **INTRODUCTION**

Cystic lesions of the pancreas are relatively rare, although they are considered an increasingly important category with a challenging differential diagnosis.1 There is no formal classification of pancreatic cystic neoplasms.<sup>2</sup> Cystic lesions of the pancreas are commonly classified as pseudocysts, parasitic cysts, neoplastic cysts, and various types of congenital or acquired developmental benign cystic lesions such as retention cysts, duplication cysts, epithelial or true cysts, and lymphoepithelial cysts.1-3 Based on the estimated relative frequency of pancreatic cystic lesions, pseudocysts are the most common.<sup>1</sup> They are caused by alcoholic, biliary, or traumatic acute pancreatitis and are nonepithelialized and non-neoplastic cysts.3 Neoplastic cystic lesions are represented by cysts with true lining (mucinous and serous neoplasms) and cysts with degenerative/necrotic change in a neoplasm (solid pseudopapillary neoplasm, cystic ductal adenocarcinoma).<sup>2</sup> Solitary true pancreatic cyst (STPC) is described as a simple thin-walled cyst with cuboidal lining and clear fluid content,4 whereas a lympoepithelial cyst is a uni- or multilocular cyst with well-differentiated squamous lining surrounded by a band of dense lymphoid tissue composed of mature T-lymphocytes with intervening germinal centers formed by B cells.<sup>1</sup> Only a few cases of STPCs in adult patients are reported in the international literature.<sup>2,4–16</sup> The aim of this study is to report a new case of STPC in an adult patient. We also review the literature on STPCs to determine the clinical features, management, pathological features, and prognosis of these lesions.

### **CASE REPORT**

A 47-year-old woman was referred to our department with a history of discomfort and pain in her upper abdomen. Her physical examination was unremarkable. Hematological and biochemical test results were within normal parameters. Upper abdominal ultrasonography (US) revealed the presence of a 5.0-cm cystic lesion located in the tail of the pancreas. Blood results were negative for tumor markers (CEA, CA19–9). Abdominal computed tomography (CT) and magnetic resonance imaging (MRI) scans were subsequently performed and confirmed the pres-

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**Figure 1.** (a) CT image showing a cystic lesion in the tail of pancreas. (b-d) MRI images displaying a unilocular, homogeneous, low-attenuated and well-demarcated cystic mass.



Figure 2. Spleen-preserving laparoscopic distal pancreatectomy.

ence of a low-attenuation and well-demarcated cystic area,  $4.5 \times 3.5$  cm in diameter, located in the tail of the pancreas (**Figure 1**). The cystic mass was unilocular and its wall was thin. There was no communication with the pancreatic ducts.

Although the imaging features of the lesion appeared to be benign, surgical excision was done to rule out the probability of malignancy. Laparoscopic distal pancreatectomy was performed. The cystic mass was excised en bloc with the tail of the pancreas, and the spleen was preserved (**Figure 2**).

Macroscopically viewed, the lesion was 6.6 cm in major diameter, with a fibrotic wall and an inner nodular appearance (**Figure 3**). The cyst contained homogenous clear serous fluid. A sample was taken for cytological



**Figure 3.** Surgical specimen: tail of the pancreas with the cystic lesion.



**Figure 4.** (a) Inner surface foci of the cuboidal epithelium (hematoxylin and eosin stain, original magnification  $\times 100$ ). (b) Positive stain for CAM 5.2 (original magnification  $\times 400$ ).

and biochemical examinations and the results showed normal levels of amylase, CA 19–9, and CEA. Histopathologic studies showed that the cyst had a fibrotic wall and fibrosis in the inner surface foci of the cuboidal epithelium surface (**Figure 4a**). The surrounding pancreatic tissue was massively destructed and hemorrhagic. Immunochemical staining results showed the epithelial surface of the cyst was positive for CEA and CAM 5.2 (**Figure 4b**).

Based on histopathologic and immunohistochemical findings, the diagnosis of an STPC was made. At the postoperative course, a small peripancreatic collection was detected 4 days after surgery and was conservatively treated. Postoperative imaging (US and CT) showed normal blood flow in the splenic vessels. The patient was discharged on the ninth postoperative day. Two years after surgery, the patient remains free of symptoms.

#### DISCUSSION

STPCs are typically diagnosed in childhood, predominantly in infants, and can be found in association with von Hippel-Lindau syndrome or polycystic disease of the kidneys.13 The occurrence of STPCs in adults is extremely rare.13 To date, only a few cases of STPCs in adults have been described.<sup>2,4-16</sup> Table 1 shows in detail all of the reported STPCs within the past 25 years. Each article was carefully studied and a database was created that included the following STPC characteristics: gender, age, signs and symptoms, laboratory findings, diagnostic imaging modalities, localization, size, time of diagnosis, treatment, histopathology, fluid examination, and follow-up. Table 2 shows STPC features. Little is known about their etiology.10 Their predominant incidence in infants and children supports the theory that they are congenital.<sup>2</sup> It is believed that true cysts derive from abnormal segmentation of the primitive pancreatic ducts, with resultant seguestered endothelial cells.17

In the articles we studied, STPCs were found in 14 female and 4 male patients (F/M, 3.5:1). All patients were adults (>16 years) at the time of diagnosis. Most STPCs (50%) were located on the head of the pancreas. The mean diameter of the cysts was 5.6 cm, with range from 0.5 to 15.0 cm. STPCs cause symptoms related to their size and location that include epigastric pain, nausea, vomiting, and biliary obstruction. Pain is described within the epigastrium and the back. A significant proportion (22.8%) of the STPCs were detected incidentally and diagnosed during imaging for an unrelated medical problem.

The widespread use of abdominal imaging (US, CT, and MRI) and the advances in quality of these technologies have increased the identification of asymptomatic pancreatic cysts.<sup>18</sup> Typically, small (<2 cm), serous, cystic lesions of the pancreas are benign. Larger (>2 cm), mucinous, multilocular cysts, or cysts with a solid component, carry the risk of malignancy.<sup>1,19</sup> However, these diagnostic modalities usually fail to differentiate preoperatively among the histologic variants of pancreatic cystic lesions.<sup>20</sup> With increasing use, endoscopic US plus fine-needle aspiration (EUS-FNA) is currently becoming an indispensable tool in the diagnosis of cystic lesions of the pancreas.<sup>20</sup> This combination provides more detailed anatomic information about the cyst than conventional US and allows the sampling of both cystic fluid and any solid component in smaller lesions.<sup>2</sup> Nevertheless, in only one case of reported STPCs16 was an FNA preoperatively performed, with uncertain results.

The inability to accurately diagnose the pathologic lesions gives rise to a controversy in the management of these patients.<sup>20</sup> Management of asymptomatic incidental pancreatic cysts of  $\leq 2$  cm includes a follow-up initially at 6 months and then yearly by performing an imaging test at intervals, whereas fluid analysis is unnecessary.<sup>21</sup> Incidental cysts may enlarge over time, and an increase to 2.5 cm and/or a change in morphology is an indication for surgery.<sup>21</sup> The size criterion for resecting an enlarging incidental cyst without a change in morphology, however, is uncertain because cysts of 3 to 4 cm usually have a low potential for malignancy.<sup>18</sup>

Surgical intervention is considered the appropriate treatment for pancreatic cysts when symptoms are presented or if the cystic mass enlarges and compresses adjacent organs. In these cases, the risk of malignancy is high and surgery is considered mandatory.<sup>15</sup> Other factors that advocate for surgery are asymptomatic large lesions, a location in the tail of pancreas, and age (young patients 17–35 years).<sup>21</sup> Surgery includes cyst enucleation, cystoduodenostomy, cystogastrostomy, cystojejunostomy, and various types of pancreatectomies. Cyst enucleation is a safe and effective procedure for small lesions, especially if they are located in the pancreatic head.<sup>13</sup> Simple cyst excision is also described for STPCs (21.6%).

Laparoscopic surgery for pancreatic diseases has gained interest among surgeons in the past decade.22 Benign and premalignant pancreatic body and tail cystic lesions can be treated with laparoscopic distal pancreatectomy (LDP).<sup>23</sup> LDP can generally be performed with or without splenectomy.24 However, it is suggested the spleen be preserved if feasible, especially in young patients, because splenectomy can lead to life-threatening complications such as overwhelming postsplenectomy infection syndrome.24,25 There are two distinct approaches to perform a spleen-preserving laparoscopic distal pancreatectomy (SPLDP). The classic procedure is to identify, isolate, and preserve the splenic artery and vein.<sup>26</sup> Alternatively, the splenic artery and vein are ligated with the pancreas, and perfusion of the spleen is maintained by the short gastric vessels.<sup>27</sup> Vessel-preserving SPLDP was performed in our case. Cioffi et al.10 described a laparoscopic cyst enucleation, whereas Carboni et al.13 described an SPLDP to remove an STPC.

As with all pancreatic operations, wound infection, organ space infection, postpancreatectomy hemorrhage, and pancreatic fistula are all possible after LDP.<sup>28</sup> The preoperative patency of the splenic vessels should be evaluated carefully, especially when SPLDP is attempted.<sup>29</sup> Patients

							Tal	ble 1.					
						Well-Docume	ented Solit	tary True Pan	creatic Cysts				
Case	Author/Year	Gender/ Age	Location	Size (cm)	Symptoms and Signs	Laboratory	Imaging Modalities	Time of Diagnosis	Treatment	Histopathologic Findings	Fluid	Fluid Analysis	Follow-up (mo)
_	Mao et al. <sup>5</sup> / 1992	F/35	Neck	3.0	Nausea, weight loss	N/R	N/R	N/R	Excision	Cuboidal epithelium, STPC	Clear, serous	Normal	N/R
21	Mendez et al. <sup>6</sup> / 1995	NR	Head	NR	Epigastric pain, palpable mass, jaundice	N/R	CT	Histology	N/R	N/R	N/R	N/R	N/R
<i>6</i> 0	Mendez et al. <sup>6</sup> / 1995	NR	Head	NR	Epigastric pain, palpable mass, jaundice	N/R	CT	Histology	N/R	N/R	N/R	N/R	N/R
4	Sperti et al.7/ 1995	M/75	Body	2.5	Incidental finding	Normal	US, CT, MRI	Histology	Excision	Cuboidal epithelium, STPC	Clear, serous	N/R	N/R
10	Sperti et al.7/ 1995	F/66	Body	4.7	Pain, nausea	Normal	US, CT, MRI	Histology	DP	Cuboidal epithelium, STPC	Clear, serous	High CA 19–9	N/R
<u>\0</u>	Sperti et al.7/ 1995	F/40	Body	8.0	Pain, palpable mass	Normal	US, CT, MRI	Histology	Excision	Cuboidal epithelium, STPC	Hemorrhagic	High CA 19–9, CA 125	N/R
5	Tanno et al. <sup>8</sup> / 1998	F/53	Tail	7.0	Incidental finding	N/R	N/R	N/R	DP	Cuboidal epithelium, STPC	Clear, serous	High CA 19–9, SPAIN 1	N/R
~	Tanno et al. <sup>8/</sup> 1998	M/16	Tail	6.5	Pain	N/R	N/R	N/R	DP	Cuboidal epithelium, STPC	Clear, serous	High CA 19–9, SPAIN 1	N/R
6	Heindryckx et al. <sup>4</sup> /1998	F/46	Head	6.0	Nausea, weight loss, jaundice	High bilirubin, transaminase, ALP	US, CT, MRI	Histology	Excision	Cuboidal epithelium, STPC	Clear, serous	Normal	N/R
10	Kim et al. <sup>2/</sup> 2001	F/38	Neck	3.0	Incidental finding	Normal	CT, MRI	Histology	Central pancreatectomy	Cuboidal epithelium, STPC	Clear. serous	N/R	N/R
11	Takahashi et al.9/2001	F/50	Head	12.0	Incidental finding	Normal	CT, MRI, ERCP	Histology	PD	Cuboidal epithelium, STPC	Clear, serous	High CA 19–9, SPAIN 1	N/R
12	Cioffi et al. <sup>10/</sup> 2003	F/22	Head	4.0	Epigastric pain, dyspepsia	Normal	US, CT	Preoperatively	Laparoscopic enucleation	Cuboidal, cylindric epithelium, STPC	Clear, serous	Normal	Well 12 months
13	Fiamingo et al.'1/2005	F/26	Head	7.0	Postprandial dyspeptic disorder, palpable mesogastric mass	Normal	US, CT, MRI	Histology	D	Cuboidal epithelium, STPC	Clear, serous	High CA 19–9	Pancreatitis after 12 months
14	Sanada et al. <sup>12</sup> / 2007	M/81	Groove area	2.0	Epigastric and back pain, soft epigastric mass, gastric dilation	High amylase, lipase	CT, ERCP	Histology	PD	Cuboidal epithelium, necrotic change of epithelium, STPC	Floating debris	N/R	N/R
Table	e 1 continued	on next	page.										

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						Well-Docume	ented Soli	itary True Pa	J ncreatic Cysts				
Case	Author/Year	Gender/ Age	Location	Size (cm)	Symptoms and Signs	Laboratory	Imaging Modalitics	Time of Diagnosis	Treatment	Histopathologic Findings	Fluid	Fluid Analysis	Follow-up (mo)
15	Carbonei et al. <sup>13</sup> /2009	F/37	Head	3.0	Incidental finding	Normal	US, CT, MRI	Histology	Enucleation	Cuboidal epithelium, STPC	Clear, serous	Normal	Well 24 months
16	Carboni et al. <sup>13/</sup> 2009	F/21	Tail	8.0	Epigastric pain, nausea, dyspepsia, tenderness in upper left abdomen	Normal	CT	Histology	dCIJB	Cuboidal epithelium, STPC	Clear, serous	Normal	Pancreatic fistulae postoperatively Well 6 months
17	Khan et al. <sup>14/</sup> 2010	F/55	Head	12.0	Abdominal pain	High CA 19.9	US, CT	Histology	DD	Cuboidal epithelium, STPC	Clear, serous	Normal	N/R
18	Zentar et al. <sup>15</sup> / 2011	M/35	Head	6.0	Abdominal pain	N/R	US, CT	Histology	PD	Cuboidal epithelium, STPC	Clear, serous	Normal	Well 12 months
19	Dalal et al. <sup>16/</sup> 2012	F/35	Head	3.7	Abdominal pain	Normal	US, CT	Histology	Enucleation	Cuboidal epithelium, STPC	Floating debris	Normal	Well 24 months
20	Current study	F/47	Tail	5.0	Abdominal pain	Normal	US, CT, MRI	Histology	dCIdS	Cuboidal epithelium, STPC	Clear, serous	Normal	Well 36 months
F = ERCJ scop	female; $M = r$ P = endoscop: ic distal panc	nale; N/R ic retrogr	t = not r ade chol ay; STPC	$\frac{1}{1}$	led; ALP = alk opancreatogra olitary true pa	aline phosph phy; DP = d ncreatic cyst.	latase; US istal panci	= ultrasoun reatectomy;	d; CT = compu PD = pancreat	ited tomography toduodenectom	y; MRI = $m_2$ y; SPLDP =	Ignetic resona spleen-prese	nce imaging; rving laparo-

	Table 2.Solitary True Pancreatic Cysts' Features				
		Patients (n)	Percent	Mean	Range
Gender	Male	4	22.2		
	Female	14	77.8		
Age (y)				43.2	16-81
Symptoms and signs	Pain	11			
	Nausea, vomiting	2			
	Palpable mass	2			
	Biliary obstruction	1			
	Unrelated or no complaints	5	22.8		
Tumor size (diameter in cm)				5.6	0.5–15.0
Location	Head (+groove area)	9	50.0		
	Neck	2	11.1		
	Body	3	33.3		
	Tail	4	5.6		
Treatment	Open enucleation	2	11.2		
	Excision	4	21.6		
	Open pancreatectomy	5	28.0		
	Open pancreatoduodenectomy	4	22.4		
	Laparoscopic enucleation	1	5.6		
	Laparoscopic spleen- preserving distal pancreatectomy	2	11.2		

with poor vascular patency of the splenic vein had more postoperative pancreatic fistulas than did patients with normal vascular patency.<sup>29</sup> Even after SPLDP, not all spleens can be salvaged because of postoperative hypoperfusion of the spleen, which may result in infarction and infection.<sup>25,29</sup>

After excision, histopathologic studies establish the diagnosis. STPCs are typically unilocular, without communication with the ductal system or internal septa.13 The finding of a cuboidal epithelium layer is the key point in the diagnosis of an STPC. STPCs usually contain clear fluid with normal amylase and lipase concentrations.<sup>13</sup> In 83.3% of the reviewed cases, the fluid was clear and serous. When it was performed, fluid analysis of STPCs showed normal amylase and lipase levels. Cyst fluid analysis may be helpful, but it is not always a sensitive and specific enough test.<sup>13</sup> High levels of CA 19-9 were measured in 6 cases. Immunostains included positive results for CA 19-9, CEA, CA 125, and SPAIN 1. These are not specific, however, because they are also positive stain findings in neoplastic cysts, pseudocysts, and lymphoepithelial cysts.11 However, pseudocysts' lack of endothelial lining and lymphoepithelial cysts' lining is surrounded by a band of dense lymphoid tissue.<sup>1</sup> To date, no case of malignancy has been reported in STPCs.

### CONCLUSION

STPCs are extremely rare, benign pathologic entities, but the finding of a symptomatic or nonpancreatic cyst is still a diagnostic dilemma. Imaging, endoscopic techniques, and tumor marker assays may partially resolve the problem, but a final diagnosis is still made only by histopathologic examinations of the specimen after surgery. Minimally invasive operative techniques should be performed for cystic lesions of the pancreas. The spleen should be preserved, if possible, when distal pancreatectomy is performed.

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