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Pancreatic solid serous cystadenoma treated by laparoscopy: Presentation of a new case report and review of the literature

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ABSTRACT

Solid serous cystadenoma is an uncommon benign pancreatic tumor, with only, including this case, 21 cases published so far. It is often misdiagnosis with other malignant pancreatic tumors.

Below we report a new case of a solid serous cystadenoma of the pancreas treated by laparoscopic distal pancreatectomy in 53-year-old female who presented with epigastric pain. Histological and immunohistochemical examination revealed a solid serous cystadenoma of the pancreas. Preoperative diagnosis of this subtype of serous cystadenoma is difficult, and, due to its benign nature, conservative resection of the tumor is the recommended treatment.

After analyzing the literature, including this case from our department, we discuss clinical presentation, imaging characteristics and histopathological findings, considering in particular difficulties in preoperative diagnosis, feasibility of laparoscopic resection.

1. Introduction

The discovery of a solid pancreatic tumor can lead to the diagnosis of a very varied histological lesion and prognosis. Typically, solid tumors of the pancreas are associated with malignancy, whereas cystic tumors more often tend to be benign [1,2]. The challenge is to determine if there is a malignant tumor (adenocarcinoma, metastases. . .) or benign (solid pseudopapillary tumor (SPPT), neuroendocrine tumor (NET), autoimmune pancreatitis. . .) [2].

Perez-Ordonez and al, described in 1996, a particular form of pancreatic serous cystic tumor with solid appearance and called it solid serous cystadenoma (SSCA) [3], it is by far the rarest subtype of serous cystic neoplasm (SCN), with only, including this case, 21 cases published so far.

Apart from other subtypes of SCN with cyst morphology, SSCA is a solid pancreatic tumor, its architecture is different from that of a serous cystadenoma (SCA), but their cytological, immunological, and histopathological characteristics are identical [4], leading to difficulty in preoperative diagnostic by imaging studies [5]. SSCA is difficult to distinguish from other solid tumors[5–8]. The knowledge of this histological type is important because, in epidemiology and imaging, its characteristics resemble to those of other solid tumors such as pancreatic adenocarcinomas, NET, solid pseudopapillary tumor and renal cell carcinoma metastasis, but the management and prognosis of these diseases are totally different.

The preoperative diagnosis of SSCA is challenging because of its rarity. However, the absence of malignancy signs should consider a conservative management; as the lesion is benign, minimal invasive surgery should be [2,9].

After analyzing the literature, including this case from our surgery department, we discuss clinical presentation, imaging characteristics and histopathological findings, considering in particular difficulties in preoperative diagnosis, feasibility of laparoscopic resection.

The work in this case has been reported in line with the SCARE criteria [10].

2. Presentation of case

A 53-year-old woman presented to our institute with 6 mounts fixed epigastric pain, gradually increasing in intensity with episodes of nausea and vomiting. She had no other symptoms and was free from any underlying disease.

Her past medical history was unremarkable unless a caesarean section 16 years ago, and was not taking any medications. The patient had an unremarkable family history.

The patient was completely fit, BMI = 22.5 kg/m2, and her vital signs were normal. Physical examination revealed mild abdominal pain at the epigastrium.

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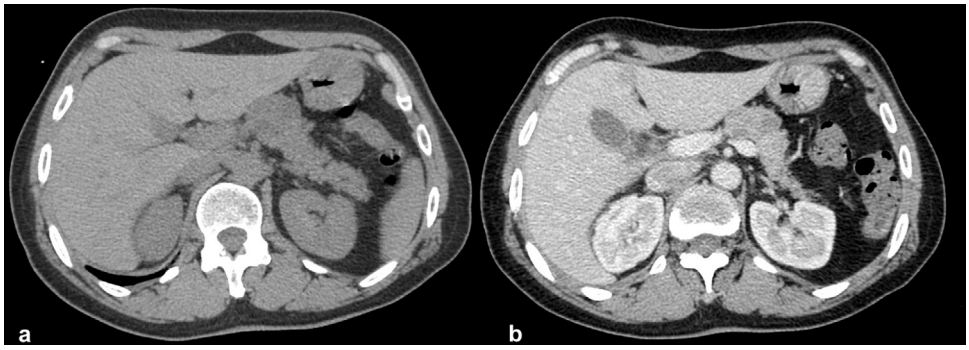

Fig. 1. Abdominal CT scan.
(a) Pre-contrast phase: A low density polycyclic mass is seen at the body of the pancreas.
(b) Portal phase: Weak tumor enhancement (80 HU) compared with the adjacent normal pancreatic tissues (100 HU).

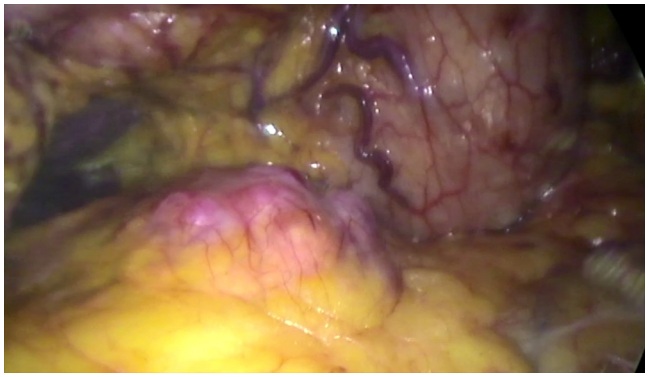

Fig. 2. Per-laparoscopic view. A well-encapsulated polycyclic mass was recognized at the pancreatic body after exposing the Lesser Sac by freeing the greater omentum off the transverse colon using Harmonic scalpel.

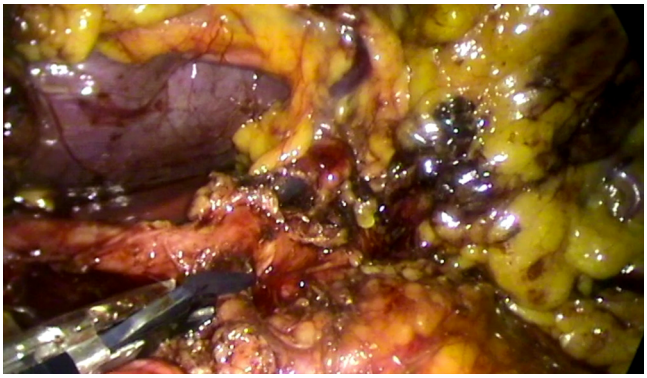

Fig. 3. Per-laparoscopic view. The splenic artery and vein were identified and clipped near to the celiac trunk.

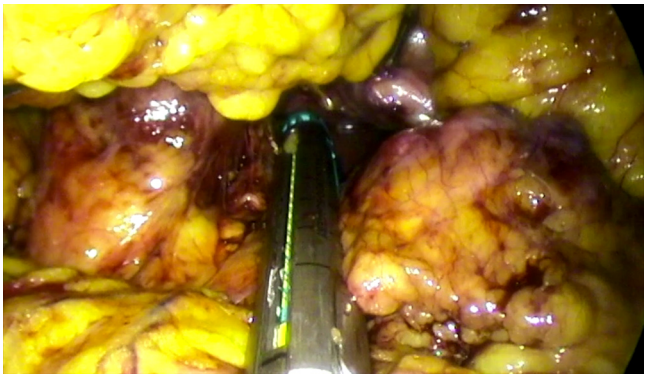

Fig. 4. Per-laparoscopic view. pancreatic body divided by a 60-mm green stapler.

Laboratory examination showed the following: WBC 4770/ml, Hb 12.3 g/dl, platelet count 263.000/ml, Albumin 42 g/l, IB 11 mg/l, DB 5 mg/l, Lipase 14 UI, Creatinine 7.2 mg/l, serum markers of exocrine and endocrine pancreatic tumors were normal (carcinoembryonic antigen (CEA): 3.9 ng/mL, reference range: 0–5 ng/mL; carbohydrate antigen 19-9 (CA19-9): 6.4 U/mL reference range: 0–27 U/mL).

Abdominal ultrasonography showed a tissular well circumscribed hypoechoic mass, 3 by 2 cm in diameter, at the pancreatic body.

An abdominal CT scan confirmed the presence of a 3.5 cm diameter well defined solid polycyclic mass at the pancreatic body, hypodense in the pre-contrast phase as compared to the surrounding pancreatic tissue (20–40 HU), without a dilatation of the distal pancreatic duct (Fig. 1a). In the portal phase, weak tumor enhancement (80–100 HU) compared with the adjacent normal pancreatic tissues (70 HU) (Fig. 1b). The central part of the tumor was consistently poorly enhanced throughout the scan. There was no honeycomb appearance, central scarring, or stellate calcification. There was no local invasion neither lymphadenopathies.

The diagnosis of solid mass of the pancreatic body was made. The differential diagnosis included pancreatic NET, solid pseudopapillary tumor and metastatic carcinoma. The diagnosis of ductal pancreatic adenocarcinoma was excluded in front of its morphological characteristics (well circumscribed, does not show contrast enhancement in the early phase, without vascular involvement neither coeliac lymphadenopathies). So, a laparoscopic approach is proposed.

The patient underwent a laparoscopic surgery. Intraoperatively, a well-encapsulated polycyclic mass was recognized at the pancreatic body (Fig. 2), the adjacent pancreatic tissue was completely normal, there was no local invasion neither macroscopic lymphadenopathies. A distal pancreatectomy with splenectomy via a medial-to-lateral approach was performed. The pancreas was approached through the gastrocolic ligament into the lesser sac. The retroperitoneum overlying the inferior border of the pancreas is dissected. The splenic artery and vein were identified and clipped near to the celiac trunk (Fig. 3). The short gastric vessels are taken down to fully retract the stomach. The pancreatic body was divided by a 60-mm green stapler (Fig. 4).

Macroscopically, the resection specimen showed a solid whitish, well circumscribed, encapsulated mass, measuring $3 \times 1 \times 1$ cm, located in the pancreatic body, 1.5 cm from the pancreatic surgical margin (Fig. 5a). The surrounding pancreatic parenchyma was normal and surgical margins were negative. The tumor contained a thick fibrous band without necrosis or hemorrhage. Histological

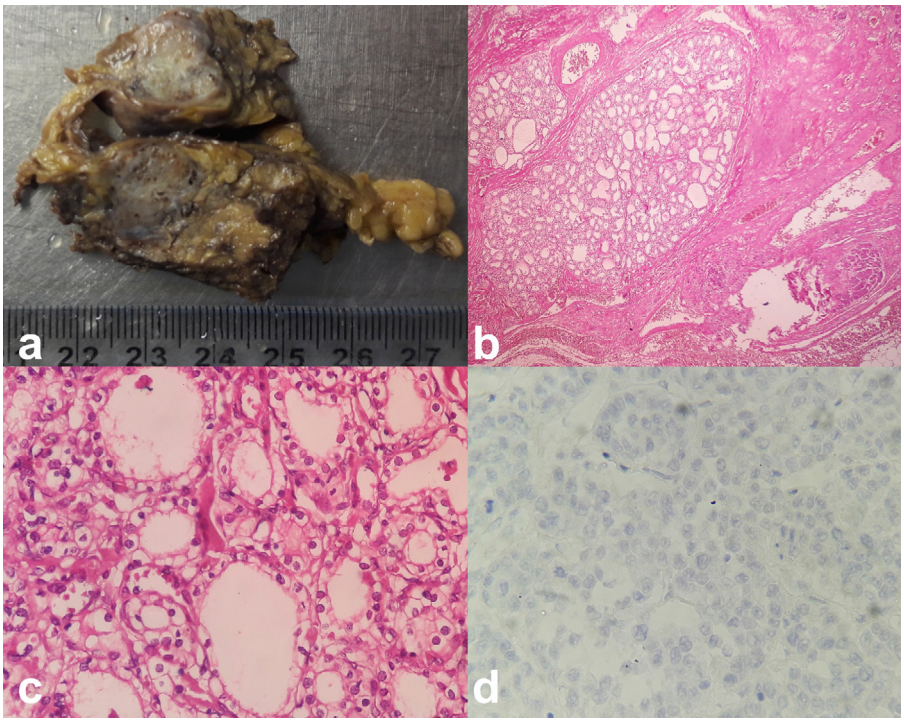

Fig. 5. Pathologic findings of solid serous adenoma of the pancreas.
(a) Macroscopic view shows a well-circumscribed whitish solid mass measuring 3 × 1 × 1 cm.
(b) Microscopy shows a tumor composed of small acini with glandular spaces with fine collagenous stroma (H&E, ×100).
(c) Tumor cells were polygonal with clear or pale eosinophilic cytoplasm with well-defined cell border (H&E, ×200).
(d) The tumor cells did not show staining to chromogranin.

examination showed a very limited and encapsulated tumor proliferation made of tubes of variable size within a hyaline and vascular stroma. The tubes are surrounded by medium sized cells with clear cytoplasm and regular nuclei, no pleomorphism or mitotic activity was identified (Fig. 5b and c). An immunohistochemical study was carried out to eliminate a well differentiated NET using chromogranin and synaptophysin. This study did not show tumor cell labeling to these two markers (Fig. 5d).

Postoperative course was uneventful and the patient was discharged from the hospital on the 12-postoperative day. The patient was followed up one month and eight months postoperatively, and we observed that she has recovered completely and remains disease-free.

3. Discussion

The recent improvements in abdominal imaging and invasive diagnostic techniques leads to detect a great proportion of SCN, it is a relatively rare disease, accounting for only 1% to 2% of all pancreatic tumors [5], and between 3–14% of all patients undergoing routine imaging [11].

SCN are actually subdivided histologically into five subtypes: serous microcystic adenomas, serous oligocystic ill-demarcated adenomas, solid-type serous cystadenomas or solid serous adenoma, von Hippel-Lindau disease-associated cystic neoplasms and serous cystadenocarcinomas [7,12,13]. SCA was also divided into four categories based on pathological findings by Kimura and al [5,14] (microcystic type (45–58%), macrocystic type (20–32%), mixed type (16–18%) and Solid type (3–5%)).

Table 1 shows the clinical characteristics of, in addition of this case, the 20 previously reported cases of SSCA based on a literature review. SSCA has the same demographic characteristics as the other subtype SCA, it occurs most frequently in elderly women 60 ± 9 years, it was reported in nine males and twelve females (sex ratio 0.75).

SSCA is usually discovered incidentally (52%) or during exams for nonspecific abdominal pain (31%), in the epigastrium (10%) left abdominal pain (5%). It can be located anywhere in the pancreas: head (40%), body (40%), or tail (20%) (Table 1). The median size is 2,8 cm [3–4 cm].

Clinical diagnosis of an SSCA is difficult because it cannot be distinguished from other solid tumors due to its radiologic characteristics. Among all of the previously reported cases including our case, only one case has had the preoperative diagnosis of solid serous adenoma [7,8,15]. Including this case, the most preoperative misdiagnosis is NET (76%), followed by other etiology of solid tumors (17%) SPPT, pancreatic ductal adenocarcinoma and metastasis.

On CT, SSCA has lower density on unenhanced phase as compared to the surrounding pancreatic tissue more frequently than NET, which was confirmed quantitatively as well by measuring their CT values [16]. Indeed, SSCA showed lower density relative to the surrounding pancreas more frequently than NET also on the delayed phase CT [16]. Characteristic SSCA image findings, such as honeycomb appearance, polycystic pattern, lobularity, central scar and hemorrhage is considered rather rare in SSCA [5,7,14]. On the contrary, Hayashi [16] reported the presence of fibrous capsule can be a sign to discriminate SSCA from NET.

Generally, the mostly accepted management options of pancreatic cystic tumors, is surgery, especially for patients with symptomatic, uncertain diagnosis, or have a high potential of malignant transformation [5,14,17]; however, pancreatic surgical resections are associated with high complication rate. In the other hand, asymptomatic SCA requires only regular observation if a sure preoperative diagnosis is made [7,8,16,18]; however, if there is high suspicion that a pancreatic tumor is an SSCA, based on radiological images, surgery can be minimized to more conservative

Table 1
Literature review of the characteristics findings of, in addition of this case, patients with solid-type serous cystadenomas.

Author	Year	Age	Sex	Location	Symptoms	Tumor size (cm)	Preoperative diagnosis	Operative procedure	Outcome/Follow
Perez-Ordonez [18,21]	1996	70	F	Tail	Abdominal pain	4	NET	DPS	5 years
Kosmahl [22]	2004	50	M	Head	Incidental	2	–	PPPD	–
Yamamoto [6]	2004	60	M	Uncus	Epigastric pain	2	NET	Enucleation	–
Gabata [15]	2005	59	F	Body	Abdominal pain	2	SSCA	DP	–
Matsumoto [23]	2006	39	F	Body	Incidental	4	NET	Enucleation	–
Yamaguchi [24]	2006	58	F	Body	Incidental	2	Malignant NET	DP	Uneventfully one year
Reese [1]	2006	66	M	Head/neck	Incidental	4	NET	PPPD	Diarrhea
Stern [25]	2007	62	M	Head and body	Abdominal pain	4.2	NET, PDA, SPPT, and metastasis	DPS	Uneventful
Sanaka [4]	2007	74	M	Body	Incidental	1.6	NET	Enucleation + YPJ	Uneventful 2 months
Casadei [21]	2008	59	F	Tail	Abdominal pain	4	NET, SPPT, and Metastasis	DPS	–
Yasuda [18]	2011	72	F	Head	Incidental	1.7	NET	PPPD	–
Hayashi [16]	2012 (2001 and 2009,)	74	F	Body	Incidental	4.2	–	–	–
		57	F	Head	Not mentioned	2.1	–	–	–
		58	F	Not mentioned	Not mentioned	3.2	–	–	–
Lee [9]	2013	56	M	Tail	Incidental	2.5	NET	Laparoscopic DP	Uneventful 12 months
Kishida [7]	2014	58	M	Body	Incidental	2.8	NET	DP	Uneventful 2 years
Wu [26]	2015	48	M	Head	Left abdominal pain	2.7	NET	PPPD	2 years
		65	M	Body	Incidental	2.3	NET	laparoscopic DP	–
Geramizadeh [8]	2015	68	F	Head	Abdominal pain	3	NET	PPPD	Uneventful 3 months
Katsourakis [27]	2016	72	F	Tail	Epigastric pain	3	NET	DP	PF six months
Current case	2017	53	F	Body	Abdominal pain	3.5	NET, SPPT	Laparoscopic DPS	Uneventful 9 months

NET: Neuroendocrine tumor; SPPT: solid pseudopapillary tumor; PDA: pancreatic ductal adenocarcinoma; DP: distal pancreatectomy without splenectomy, DPS: distal pancreatectomy with splenectomy, YPJ: Roux-en-Y pancreaticojejunostomy; PPPD: pylorus preserved pancreaticoduodenectomy; PF: pancreatic fistula.

procedure [12]. All the previously reported cases of SSCA have been treated by surgery. Various surgical procedures were performed according to the location of the tumor, they are mentioned in 18 patients are shown in Table 1; the procedures included a spleen-preserving distal pancreatectomy (33.3%), a pylorus-preserving pancreatoduodenectomy (27.7%), a distal pancreatectomy with splenectomy (22.2%) and an enucleation (16.6%). In three (16%) cases by laparoscopy and 15 (83%) by laparotomy. The laparoscopic pancreatectomy offers less morbidity, less intraoperative blood loss, and a shorter length of hospital stay [19–21]. The margin status, operative times, and the fistula rates are similar to open surgery [14,21]. Laparoscopic may be the surgery of choice for patients with benign neoplasms such SSCA [2,19].

4. Conclusion

Solid serous adenoma is an extremely rare subtype of serous pancreatic adenoma. Solid pancreatic tumors are typically associated with malignancy, whereas cystic tumors more often tend to be benign, therefore, it should be considered in the differential diagnosis of solid pancreatic tumors by both clinicians, radiologists, and pathologists in order to ovoid aggressive management. A conservative management should be proposed when malignancy signs could be eliminated. Laparoscopic resection offers both the anatomopathological diagnosis and minimally invasive resection for symptomatic tumor.

Competing interests

The authors declare that they have no competing interests.

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There is not any sources of funding.

Ethical approval

Not applicable. No research study involved.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and accompanying figures. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

The images are entirely unidentifiable and there are no personal details on the patient reported within the manuscript.

Author contribution

Mohamed Hamid: Data collection, drafting the paper.

Mohamed Tbouda:carried out the pathologic and immunohistochemical studies and drafted the manuscript.

Anas Mohamed Majbar: revising the manuscript.

Mohamed Raiss, Mohamed Ahallat: Surgeon performing the operation. Data collection. Coordination and helped to draft the manuscript.

Guarantor

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