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Successful treatment of a pancreatic schwannoma by spleen-preserving distal pancreatectomy

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Abstract

Schwannomas are neurogenic tumors that arise from the neural sheaths of peripheral nerves. These tumors can be located in any area of the human body; the most common locations are the head, neck, trunk and extremities. Pancreatic schwannomas are very rare. Over the past 40 years, only 67 cases of pancreatic schwannomas have been reported in the English literature. Here we present a case of pancreatic schwannoma in a 62-year-old male. The tumor was revealed by ultrasound and computed tomography in the neck and body of the pancreas. An accurate diagnosis was difficult to obtain preoperatively. The patient consented to the performance of a laparotomy, and the mass was found in the neck and body of the pancreas and successfully treated using a spleen-preserving distal pancreatectomy with splenic artery and vein preservation. The procedure has only been reported in one other case of pancreatic schwannoma; here we present the second reported case. Macroscopically, the tumor was well circumscribed, gray-white in color and 3.3 cm × 2.8 cm in size. Microscopically, the tumor cells were spindle-shaped and had a palisading arrangement with no atypia, which are results compatible with a benign tumor. Both hypercellular and hypocellular areas were visible. Immunohistochemically, the tumor cells were strongly positive for S-100 protein. The tumor was definitively diagnosed as a schwannoma of the pancreatic neck and body. The patient was followed for 72 mo and has been doing well without any complications.

Key words: Schwannoma; Pancreas; Spleen-preserving distal pancreatectomy; S-100; Mesenchymal tumor

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Core tip: Over the past 40 years, only 67 cases of pancreatic schwannoma have been reported in the English literature. An accurate preoperative diagnosis is difficult to obtain. Here, we present the case of a patient with a pancreatic schwannoma who underwent spleen-preserving distal pancreatectomy. This surgical intervention has only been previously reported in one case of pancreatic schwannoma. After surgery, the patient recovered quickly and had a good prognosis. In this case report, we share our experience in the diagnosis and treatment of a rare pancreatic schwannoma and performed a literature review to deepen the understanding of the subject.

Xu SY, Wu YS, Li JH, Sun K, Hu ZH, Zheng SS, Wang WL. Successful treatment of a pancreatic schwannoma by spleen-preserving distal pancreatectomy. *World J Gastroenterol* 2017; 23(20): 3744-3751 Available from: URL: <http://www.wjgnet.com/1007-9327/full/v23/i20/3744.htm> DOI: <http://dx.doi.org/10.3748/wjg.v23.i20.3744>

INTRODUCTION

Schwannomas are mesenchymal tumors that originate from the Schwann cells of peripheral nerves^[1]. Schwannomas are generally encapsulated, and over 90% are benign^[2]. These tumors can occur in patients of all ages, with an equal frequency in males and females, and are most often reported in patients between 20 and 50 years of age^[3]. A considerable number of these patients are asymptomatic, and the tumors are found incidentally^[4]. Schwannomas can show either monosomy 22 or loss of 22q material; the definitive pathogenesis of the tumor remains uncertain^[1]. Occasionally, the tumor can become cystic, hemorrhagic, calcified or even ossified^[5]. Surgery may be the optimal treatment for schwannomas, after which patients generally have a good prognosis^[6]. Although schwannomas can develop in any part of the body, the most common locations include the head, neck, trunk and extremities^[7]. Schwannomas in the pancreas are extremely rare. To our knowledge, over the past 40 years, only 67 cases have been reported in the English literature^[8-68]. In the present study, we present a case of a pancreatic schwannoma that was successfully treated using a spleen-preserving distal pancreatectomy technique and performed a review of the available literature.

CASE REPORT

On January 6, 2011, a 62-year-old male was referred to our hospital because of a pancreatic mass that was found on ultrasound during a routine health examination at the local hospital. His abdomen was soft, non-tender, and non-distended, with no evidence of a palpable or pulsatile mass. There was no history of weight loss or trauma and no family history of significant disease. Abnormal laboratory results included an international normalized ratio of 1.16 (normal range: 0.85-1.15) and a thrombin time of 21.7 s (normal range: 14.5-21.5). Other laboratory results, which included tumor markers, were normal. The ultrasound showed a well-defined, low-density lesion measuring 3.7 cm × 2.3 cm in the neck and body of the pancreas (Figure 1A). No blood flow signal was detected within the lesion on color Doppler ultrasound (Figure 1B). An unenhanced computed tomography (CT) scan revealed a well-marginated and hypodense mass measuring 2.8 cm × 1.9 cm in the pancreatic neck and body (Figure 2A). On contrast-enhanced CT, the mass was slightly and heterogeneously enhanced (Figure 2B). Endoscopic ultrasound-fine needle aspiration (EUS-FNA) was also performed. However, we failed to acquire a tumor sample. Although imaging results were obtained, the mass in the pancreatic neck and body was still unable

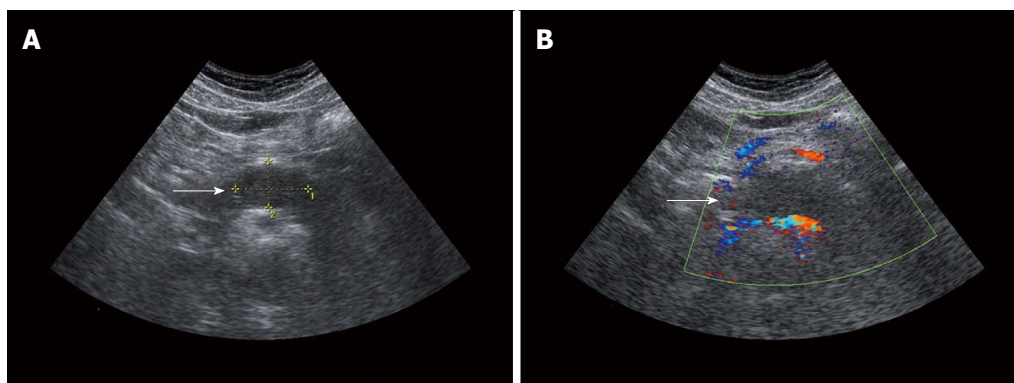


Figure 1 Ultrasound findings. A: Ultrasound showed a well-defined, low-density lesion (arrow) measuring 3.7 cm × 2.3 cm in the pancreatic neck and body; B: No blood flow signal within the lesion was detected by color Doppler ultrasound.

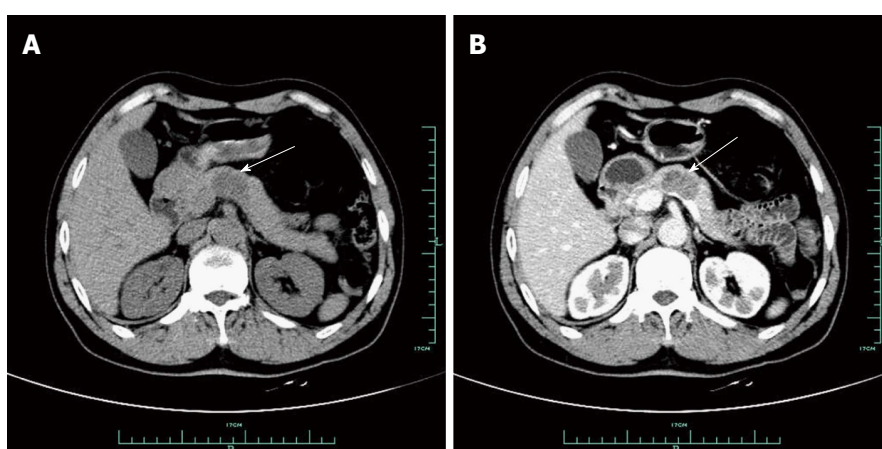


Figure 2 Computed tomography findings. A: An unenhanced CT scan revealed a well-margined and hypodense mass (arrow) measuring 2.8 cm × 1.9 cm in the pancreatic neck and body; B: On contrast-enhanced CT, the mass was slightly and heterogeneously enhanced.

to be accurately diagnosed.

The patient consented to the performance of a laparotomy, and the mass was observed to originate from the pancreatic neck and body. We performed a spleen-preserving distal pancreatectomy with preservation of the splenic artery and vein. Intraoperative frozen pathology revealed a pancreatic schwannoma. Macroscopically, the mass was observed in the pancreatic neck and body, gray-white in color and 3.3 cm × 2.8 cm in size. Microscopically, the tumor cells were spindle-shaped and had a palisading arrangement with no atypia, which are results that are compatible with a benign tumor. Both hypercellular and hypocellular areas were visible (Figure 3). Immunohistochemically, the tumor cells were strongly positive for S-100 protein (Figure 4), but negative for SMA, CD34 and CD117. The tumor was definitively diagnosed as a schwannoma of the pancreatic neck and body. After surgery, the patient recovered smoothly and was discharged from the hospital 10 d later. The patient was followed for 72 mo and has been doing well without any further complications.

DISCUSSION

Schwannomas are neoplasms that originate from Schwann cells in nerve sheaths^[69]. More than 90% of schwannomas are benign, and they account for approximately 5% of benign soft-tissue neoplasms^[3]. Malignant schwannomas are rare and are usually associated with von Recklinghausen's disease^[70]. Schwannomas can occur in patients of all ages, with equal frequencies in males and females, and cases are most often reported in patients between 20 and 50 years of age^[3]. Almost any site in the human body can be involved, although the head, neck, and extremities are the most reported areas of tumor development^[71]. Schwannomas that originate from the nerve sheaths of the pancreas are extremely rare. Intra-pancreatic innervation includes the perivascular plexus, periacinous plexus and peri-insular plexus. These three plexuses connect with each other to form a net-like structure. However, the type of nerve fibers that produce the origination of pancreatic schwannomas has not yet been described. To our knowledge,

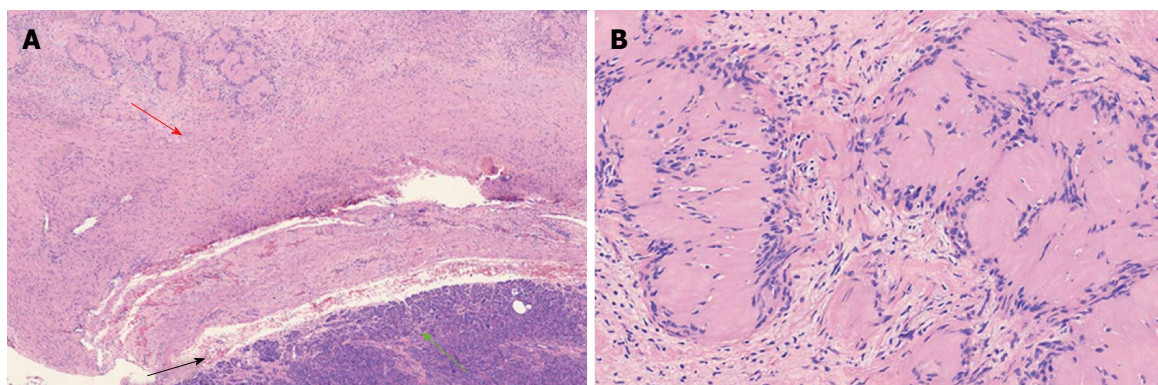


Figure 3 Microscopic examination. A: A thin capsule (black arrow) was found between the tumor (red arrow) and the normal pancreatic tissues (green arrow) (HE, $\times 40$); B: The tumor cells were spindle-shaped and had a palisading arrangement with no atypia, which is compatible with a benign tumor. Both hypercellular and hypocellular areas were visible (HE staining, $\times 200$). HE: Hematoxylin and eosin.

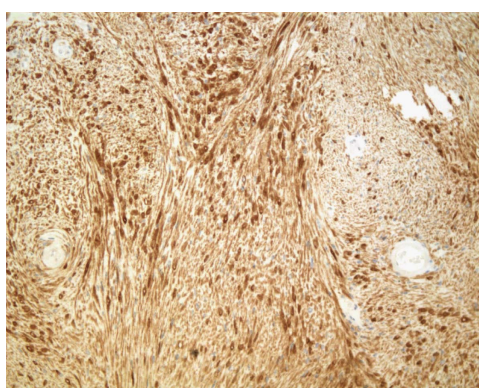


Figure 4 Immunohistochemical staining. The tumor cells were strongly positive for S-100 protein (HE staining, $\times 200$). HE: Hematoxylin and eosin.

over the past 40 years, only 67 cases of pancreatic schwannoma have been reported in the English literature^[8-68]. Table 1 summarizes the important available clinicopathological characteristics of these 68 cases, which include the present case. Continuous variables are summarized as the mean \pm SD and the range. Statistical analyses were conducted using SPSS version 20.0 for Windows (SPSS Inc., Chicago, IL).

Because the clinical symptoms and imaging characteristics of schwannomas are nonspecific, the accurate preoperative diagnosis of a pancreatic schwannoma is nearly impossible. A definitive diagnosis relies on the combined findings of the histopathological and immunohistochemical examination of surgical specimens. Microscopically, pancreatic schwannomas are usually encapsulated with varying relative amounts of the two histologic components of hypercellular Antoni A areas and hypocellular Antoni B areas^[22]. The former is composed of closely packed spindle cells with occasional nuclear palisading. The latter consists of loosely arranged tumor cells and abundant myxoid stroma^[13]. Over 90% of pancreatic schwannomas are benign. Thus far, only 5 (7.35%) malignant pancreatic schwannomas have been reported in the English

literature^[29,64,66-68]. Immunohistochemical staining showed that schwannomas are positive for S-100 protein, but negative for desmin, smooth muscle myosin, SMA, CD34 and CD117^[23,72].

Imaging modalities including ultrasound, CT and magnetic resonance imaging (MRI) have a certain diagnostic value but lack specificity. On ultrasound, a pancreatic schwannoma is usually shown as a well-margined hypodense lesion. On unenhanced CT scans, schwannomas are usually well-defined hypodense lesions with an associated capsule. Schwannomas with high Antoni A areas show a high density and appear inhomogeneous. Schwannomas with high Antoni B areas appear cystic and show a low density^[17]. On contrast-enhanced CT scans, Antoni A areas are enhanced, while Antoni B areas are unenhanced^[14]. On MRI, schwannomas usually appear hypointense in T1-weighted images and inhomogeneous and hyperintense in T2-weighted images^[23]. EUS-FNA may also be valuable for the preoperative diagnosis of schwannomas. Li *et al.*^[35] reported a pancreatic schwannoma that was definitively diagnosed by EUS-FNA. In the present study, EUS-FNA was also performed; however, we failed to acquire a sufficient sample of the tumor.

Surgery is the curative treatment for pancreatic schwannomas, and most cases are treated by laparotomy. Only one case of pancreatic schwannoma was treated using minimally invasive laparoscopic surgery (1.47%)^[21]. Enucleation has been reported in 10 (14.71%) cases. Patients treated by minimally invasive surgery might have less pain and faster recovery. Since the tumor can be located in different sections of the pancreas, surgical approaches may vary. In the present case, we performed a spleen-preserving distal pancreatectomy for the mass that was found in the neck and body of the pancreas. To date, spleen-preserving distal pancreatectomy has only been reported in one other case of pancreatic schwannoma^[38]; here we describe the second

Table 1 Summary of clinicopathological data from all 68 cases of pancreatic schwannoma

	<i>n</i> (%) or mean \pm SD (range)
Age (yr) (<i>n</i> = 67)	
Mean	55.67 \pm 15.13 (20-87)
Sex (male/female), (male %) (<i>n</i> = 67)	30/37 (44.78)
Symptoms ¹ (<i>n</i> = 67)	
Asymptomatic	25 (37.31)
Symptomatic	
Abdominal pain	29 (43.28)
Weight loss	9 (13.43)
Back pain	4 (5.97)
Nausea/vomiting	3 (4.48)
Anorexia	2 (2.99)
Anemia	2 (2.99)
Melena	2 (2.99)
Jaundice	2 (2.99)
Dyspepsia	1 (1.49)
Abdominal discomfort	1 (1.49)
Abdominal mass	1 (1.49)
Location (<i>n</i> = 68)	
Head	26 (38.24)
Head/body	3 (4.41)
Neck/body	1 (1.47)
Body	14 (20.59)
Body/tail	7 (10.29)
Tail	8 (11.76)
Uncinate process	8 (11.76)
Mean size (cm) (<i>n</i> = 63)	5.75 \pm 4.52 (1-20)
Benign	59 (5.21 \pm 3.89) (1-20)
Malignant	4 (13.75 \pm 6.24) (7-20)
Operation (<i>n</i> = 68)	
PD ²	20 (29.41)
PPPD	2 (2.94)
DP + splenectomy ³	16 (23.53)
DP + splenic preservation	2 (2.94)
Enucleation	10 (14.71)
Central pancreatectomy	1 (1.47)
Unresectable	2 (2.94)
Refused	1 (1.47)
Not specified	13 (19.12)
Histology (<i>n</i> = 68)	
Malignant	5 (7.35)
Benign	62 (91.18)
Not specified	1 (1.47)
Nature of tumor (<i>n</i> = 68)	
Solid	21 (30.88)
Cystic	27 (39.71)
Solid and cystic	14 (20.59)
Not specified	6 (8.82)
Mean follow-up months (<i>n</i> = 30)	22.23 \pm 19.56 (3-67)
No. of deaths	0

¹Some patients had two or several symptoms; ²One patient underwent resection of the portal vein; ³One patient underwent resection of the transverse colon. PD: Pancreaticoduodenectomy; PPPD: Pylorus preserving pancreaticoduodenectomy; DP: Distal pancreatectomy.

reported case. Compared with the traditional distal pancreatectomy and splenectomy for tumors in the body or tail of the pancreas, spleen-preserving distal pancreatectomy can not only offer complete resection of the tumor but also preserve the spleen. After complete removal of the tumor, patients usually have a good prognosis.

In conclusion, the finding of a schwannoma in the pancreas is extremely rare. Over the past 40 years, only 67 cases of pancreatic schwannoma have been reported in the English literature. Although multiple imaging modalities are currently available, it is challenging to obtain an accurate diagnosis prior to the performance of surgery, which is the optimal treatment for pancreatic schwannomas. Spleen-preserving distal pancreatectomy has only been reported in one previous case. After complete resection of the tumor, patients with pancreatic schwannoma usually have a good prognosis.

COMMENTS

Case characteristics

On January 6, 2011, a 62-year-old male was referred to the authors' hospital because of a pancreatic mass found during a routine health examination at the local hospital.

Clinical diagnosis

The patient's abdomen was soft, non-tender, and non-distended, with no evidence of a palpable or a pulsatile mass.

Differential diagnosis

Intraductal papillary mucinous neoplasm, mucinous cystic neoplasm, solid pseudopapillary tumor, pancreatic endocrine tumor or pancreatic ductal adenocarcinoma.

Laboratory diagnosis

Abnormal laboratory results included an international normalized ratio of 1.16 (normal range: 0.85-1.15) and a thrombin time of 21.7 s (normal range: 14.5-21.5). Other laboratory results, including tumor markers, were normal.

Imaging diagnosis

Ultrasound showed a well-defined, low-density lesion measuring 3.7 cm \times 2.3 cm in the pancreatic neck and body. No blood flow signal was detected by color Doppler ultrasound within the lesion. An unenhanced CT scan revealed a well-marginated and hypodense mass measuring 2.8 cm \times 1.9 cm in the pancreatic neck and body. On contrast-enhanced CT, the mass was slightly and heterogeneously enhanced. Although they obtained these imaging results, the mass in the pancreatic neck and body was still unable to be definitively diagnosed.

Pathological diagnosis

Macroscopically, the mass was shown in the pancreatic neck and body, gray-white in color and 3.3 cm \times 2.8 cm in size. Microscopically, the tumor cells were spindle-shaped and had a palisading arrangement with no atypia, which are results that are compatible with a benign tumor. Both hypercellular and hypocellular areas were visible. Immunohistochemically, the tumor cells were strongly positive for S-100 protein, but negative for SMA, CD34 and CD117. The tumor was definitively diagnosed as a schwannoma of the pancreatic neck and body.

Treatment

The patient underwent a spleen-preserving distal pancreatectomy.

Related reports

Schwannoma in the pancreas is extremely rare. Over the past 40 years, only 67 cases of pancreatic schwannomas have been reported in the English literature. To date, the use of a spleen-preserving distal pancreatectomy has only been

reported in one other case.

Experiences and lessons

Imaging modalities, including ultrasound, CT and magnetic resonance imaging, have a certain diagnostic value but lack specificity in the diagnosis of pancreatic schwannoma. Surgery is the curative treatment for the tumor. Since the tumor can be located in different sections of the pancreas, surgical approaches may vary. After complete resection of the tumor, patients with pancreatic schwannomas usually have a good prognosis.

Peer-review

This study shares the experience in the diagnosis and the treatment of a rare pancreatic schwannoma with an accompanying literature review to deepen the understanding of the subject. The information in this paper is useful for readers.

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