CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 36 (2017) 161–166

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

A case of pancreatic schwannoma showing increased FDG uptake on PET/CT

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ARTICLE INFO

Article history:
Received 3 May 2017
Accepted 27 May 2017
Available online 1 June 2017

Keywords:
Pancreatic schwannoma
Pancreatic tumor
FDG-PET

ABSTRACT

INTRODUCTION: Schwannomas are neoplasms originating from Schwann cells in nerve sheath, but pancreatic schwannomas are rare.
PRESENTATION OF CASE: A 59-year-old female incidentally pointed out a 2-cm-sized solid mass in the pancreas head by computed tomography (CT) which was performed for other reasons. Magnetic resonance imaging (MRI) showed a mass with hypointensity on T1-weighted images and a heterogenous hyperintensity on T2-weighted images. Magnetic resonance cholangiopancreatography (MRCP) showed no abnormal findings in the main pancreatic duct. The mass of the pancreas head showed a significantly increased uptake on fluorin-18 fluorodeoxyglucose positron emission tomography CT (FDG-PET/CT). Based on the radiological findings, a malignant potential tumor was preliminarily considered. Subtotal stomach-preserving pancreatoduodenectomy (SSSPD) was performed to make a definite diagnosis. The resected specimen showed a well-encapsulated circumscribed mass, measuring 2.0 cm, in the pancreas head. Histopathological examination revealed proliferation of spindle cells showing interlacing and palisading patterns in the tumor. The proliferating cells showed no atypia and few mitoses. Immunohistochemically, the spindle cells were positive for S-100 protein. The Ki67 labeling index was approximately 2%. Based on these findings, a diagnosis of benign schwannoma of the pancreas was made.
CONCLUSION: Although pancreatic schwannoma is a rare neoplasm, it is important to take into account this tumor in the differential diagnosis of pancreatic tumors. In addition, we should be aware that FDG-PET/CT shows abnormal accumulation in a benign pancreatic schwannoma.

1. Introduction

Schwannoma is a benign nerve sheath tumor originating from Schwann cells and they can arise in any part of the peripheral nerves. Pancreatic schwannomas are rare. Pancreatic schwannomas have a wide variety of imaging findings and tend to be confused with other pancreatic tumors [1]. Herein we describe a case of asymptomatic pancreatic schwannoma that was incidentally detected by computed tomography (CT) and showed increased fluorin-18 fluorodeoxyglucose (FDG) uptake. This case report has been prepared in line with the SCARE criteria [2].

2. Presentation of case

A 59-year-old female complained of lower abdominal fullness sensation and was referred to our hospital. Abdominal computed tomography (CT) revealed a large cystic ovarian tumor that was 18 cm in diameter. Simultaneously, a 2-cm-sized solid mass in the pancreas head was detected. Fluorin-18 fluorodeoxyglucose positron emission tomography CT (FDG-PET/CT) did not show a significantly increased uptake in the cystic ovarian tumor, while increased accumulation with a maximum standardized uptake value (SUV max) of 5.6 was seen in the mass of the pancreas head (Fig. 1a, b). After resection of the cystic ovarian tumor, further examination for the pancreatic mass was done. Laboratory data

Abbreviations: CT, computed tomography; FDG-PET, fluorin-18 fluorodeoxyglucose positron emission tomography; MRI, magnetic resonance imaging; MRCP, magnetic resonance cholangiopancreatography; SSSPD, subtotal stomach-preserving pancreatoduodenectomy; SUV max, maximum standardized uptake value; EUS, endoscopic ultrasonography; CEA, carcinoembryonic antigen; CA19-9, carbohydrate antigen 19-9.

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http://dx.doi.org/10.1016/j.ijscr.2017.05.031

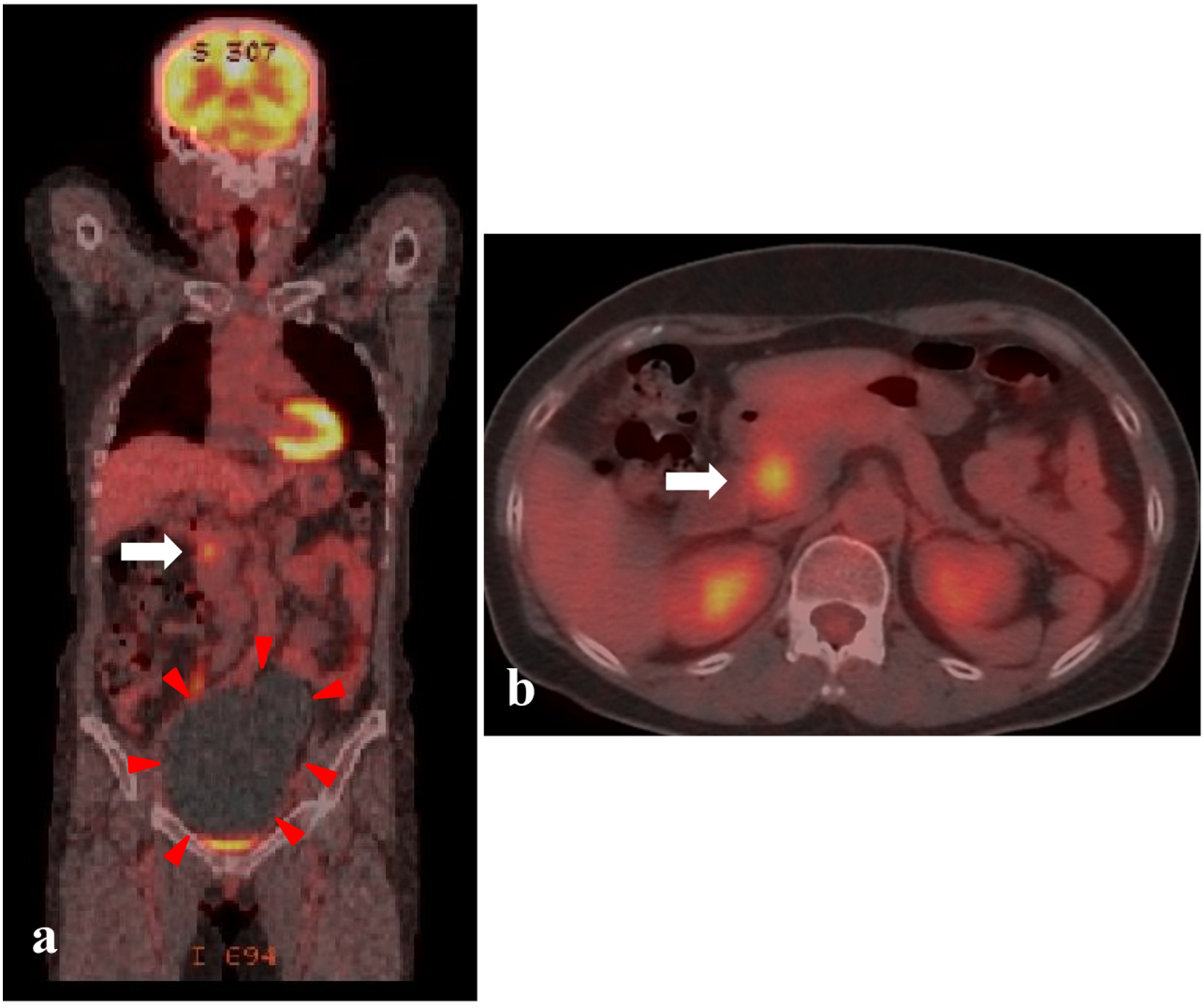

Fig. 1. Fluorin-18 fluorodeoxyglucose positron emission tomography CT (FDG-PET/CT) did not show a significantly increased uptake in the cystic ovarian tumor (red arrowhead) (a), while increased accumulation with a maximum standardized uptake value (SUV max) of 5.6 was seen in the mass of the pancreas head (white arrow) (a, b).

including carcinoembryonic antigen (CEA) and carbohydrate antigen 19-9 (CA19-9) were within normal ranges. A CT scan showed a low density mass, 20 × 20 mm in size, in the head of the pancreas (Fig. 2a). Dynamic CT showed a well-circumscribed solid mass presenting slight, gradual, heterogeneous enhancement (Fig. 2b–d). Magnetic resonance imaging(MRI)showed the pancreatic mass with hypointensity on T1-weighted images and heterogenous hyperintensity on T2-weighted images (Fig. 3a, b). Magnetic resonance cholangiopancreatography (MRCP) showed no abnormal findings in the main pancreatic duct (Fig. 3c). Endoscopic ultrasonography (EUS) showed a hypo-echoic mass with no blood flow. Based on the radiological findings, solid pseudopapillary neoplasm, neuroendocrine tumor or solid type of serous cystadenocarcinoma was preliminarily considered. Subtotal stomach-preserving pancreatoduodenectomy (SSPPD) was performed to make a definite diagnosis, because the possibility of malignant pancreatic tumor could not be excluded due to the increased FDP uptake on FDG-PET/CT. Macroscopically, a well-encapsulated circumscribed mass, measuring 2.0 cm, in the pancreas head was observed (Fig. 4a). Histopathological examination revealed proliferation of spindle cells showing interlacing and palisading patterns in the tumor. The proliferating cells showed no atypia and few mitoses (Fig. 4b, c). Immunohistochemically, the spindle cells were positive for S-100 protein (Fig. 4d) and negative for smooth muscle actin, c-kit, CD34 and desmin. The Ki67 labeling index was approximately 2%. Based on these findings, a diagnosis of benign schwannoma of the pancreas was made. The patient's postoperative course was uneventful and she was discharged on the 38th day after the operation. At a 10-month follow-up after resection, the patient did not have any recurrence.

3. Discussion

Schwannomas are mesenchymal tumors that originate from Schwann cells in peripheral nerve sheaths, and they commonly occur in the head, neck, trunk and extremities. Pancreatic schwannomas are rare. Recently, Xu et al. reviewed 65 cases of pancreatic schwannoma reported in the English literature over the past 40 years [3]. The male-to-female ratio was 1:1.2 with a mean age of 55.2 years, and the mean diameter of the tumors was 5.8 cm. The clinical symptoms are nonspecific. The symptoms include abdominal pain (43.8%), weight loss (12.5%), back pain (6.3%), and nausea/vomiting (4.7%). Thirty-seven percent of the patients were asymptomatic, and the lesions were incidentally detected by imaging modalities performed for other reasons. The most common location site was the head (40%) followed by the body (23.1%), the

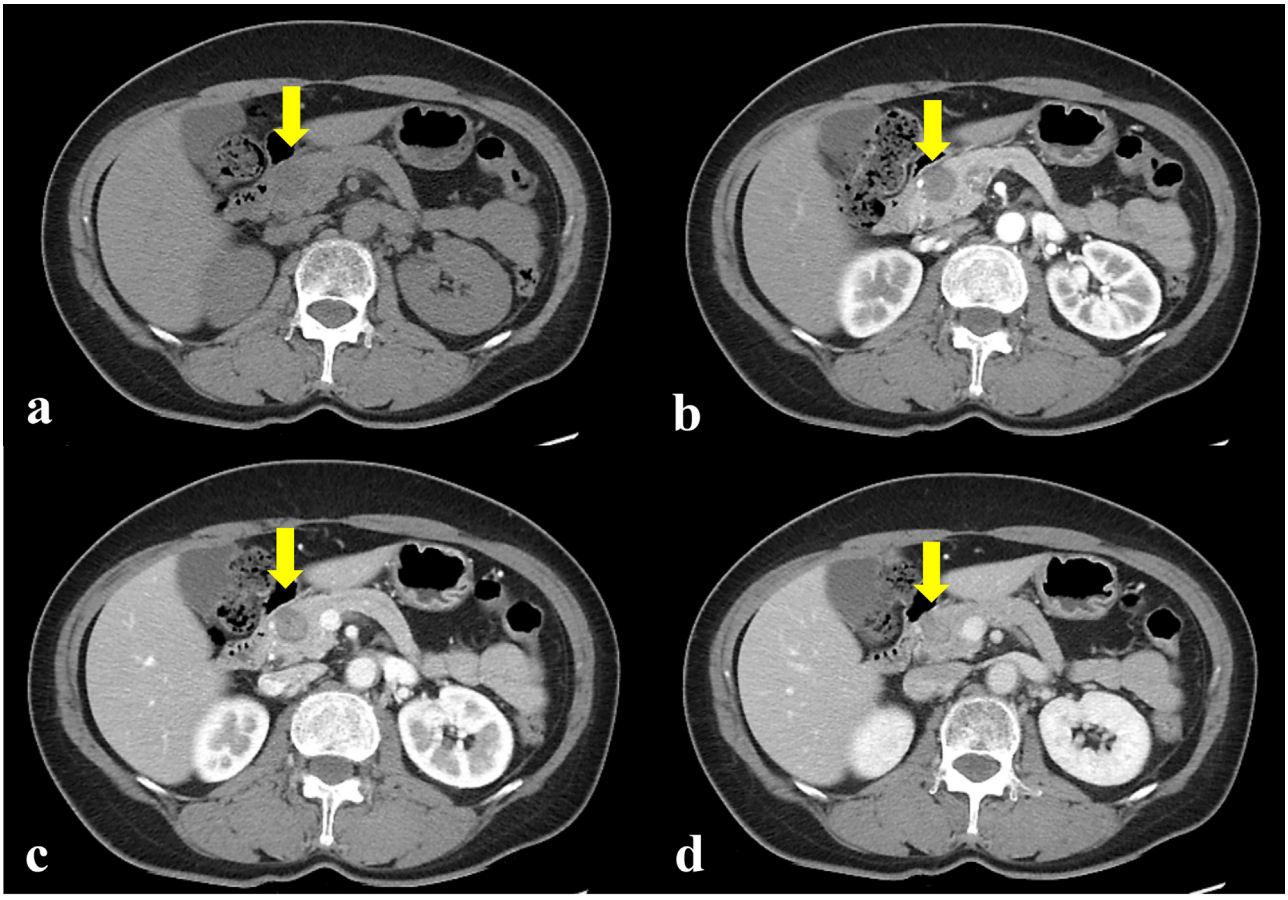

Fig. 2. Computed tomography (CT) scan showed a low density mass, 20 × 20 mm in size, in the head of the pancreas (a). Dynamic CT during the arterial phase (b), portal phase (c), and venous phase (d), showed a well-circumscribed solid mass presenting slight, gradual, heterogeneous enhancement (yellow arrow).

tail (10.8%), uncinate process (10.8%). The most common treatment was pancreaticoduodenectomy (34%) followed by distal pancreatectomy (25%) and enucleation (14%).

Generally, schwannomas are solitary, well-circumscribed and encapsulated [4]. Small schwannomas appear as a homogenous, encapsulated mass, whereas large schwannomas show cystic degeneration, hemorrhage or central necrosis [5]. Schwannomas are characterized by two histologic components: Antoni A and Antoni B areas. Antoni A area is hypercellular and characterized by closely packed spindle cells with nuclear palisading and Verocay bodies, whereas Antoni B area is hypocellular with degenerative changes and has loosely arranged tumor cells [1]. Antoni A area shows a high density and heterogenous appearance due to high cellularity and increased lipid content. Antoni B area appears cystic and shows a low density due to loose stroma and low cellularity. On contrast-enhanced CT, the Antoni A areas are usually enhanced, whereas the Antoni B areas are unenhanced [6]. Since most of the pancreatic schwannomas reported had both Antoni A and Antoni B areas in various proportions, pancreatic schwannoma shows a variety of image findings. Accordingly, preoperative diagnosis of pancreatic schwannoma, especially cystic pancreatic schwannoma, is difficult. Radiologically, schwannomas sometimes mimic cystic pancreatic tumors such as mucinous cystic neoplasms, solid pseudopapillary neoplasms, serous cystic neoplasms and pseudocysts.

FDG-PET/CT is a widely used imaging tool in oncology. Generally, malignant tumors tend to accumulate FDG because of the hypermetabolic nature of malignant cells. Actually, FDG-PET/CT is useful for assessing the malignant potential and bioactivity of gastrointestinal stromal tumors (GISTs) [7]. It has been reported that schwannomas in peripheral nerve sheaths and in the gastrointestinal tract occasionally show increased FDP uptake even though they are benign [8,9]. However, little is known about the results of FDG-PET/CT for pancreatic schwannomas. To the best of our knowledge, there have been only 3 reports including this case report in which the results of FDG-PET/CT are described [6,10] (Table 1). The mean diameter of the tumors was 4.0 cm and mean SUVmax was 4.4. Although the number of cases was small, there seems to be no correlation between tumor size and the value of SUVmax. Despite showing increased FDP uptake, all of the cases were histologically diagnosed with benign schwannoma. According to a previous study, factors having a significant association with FDG uptake include tumor size, cellularity defined on the basis of the ratio of Antoni A area to the whole tumor area, and microvascular density [11]. Another study showed that there was no correlation between tumor size or tumor proliferation rate (Ki67 labeling index) [12]. Miyake et al. investigated the clinical, morphologic and pathologic features associated with increased FDG uptake in benign schwannoma, and they demonstrated that schwannomas of gastrointestinal origin and schwannomas with peritumoral lymphoid cuffs were significantly associated with higher FDG uptake [13]. These results indicate that we should know the features of schwannomas associated with increased FDG uptake and avoid misinterpretation of hypermetabolic schwannomas as malignant tumors.

A definitive diagnosis of schwannoma can be achieved only by histopathological and immunohistochemical examinations of the resected specimen. Microscopically, the hallmark of schwannoma is the pattern of alternating Antoni A and B areas. Immunohistochemically, schwannomas stain strongly positive for S100 protein, vimentin and CD56, and negative for cytokeratin AE1/3, CD34, c-

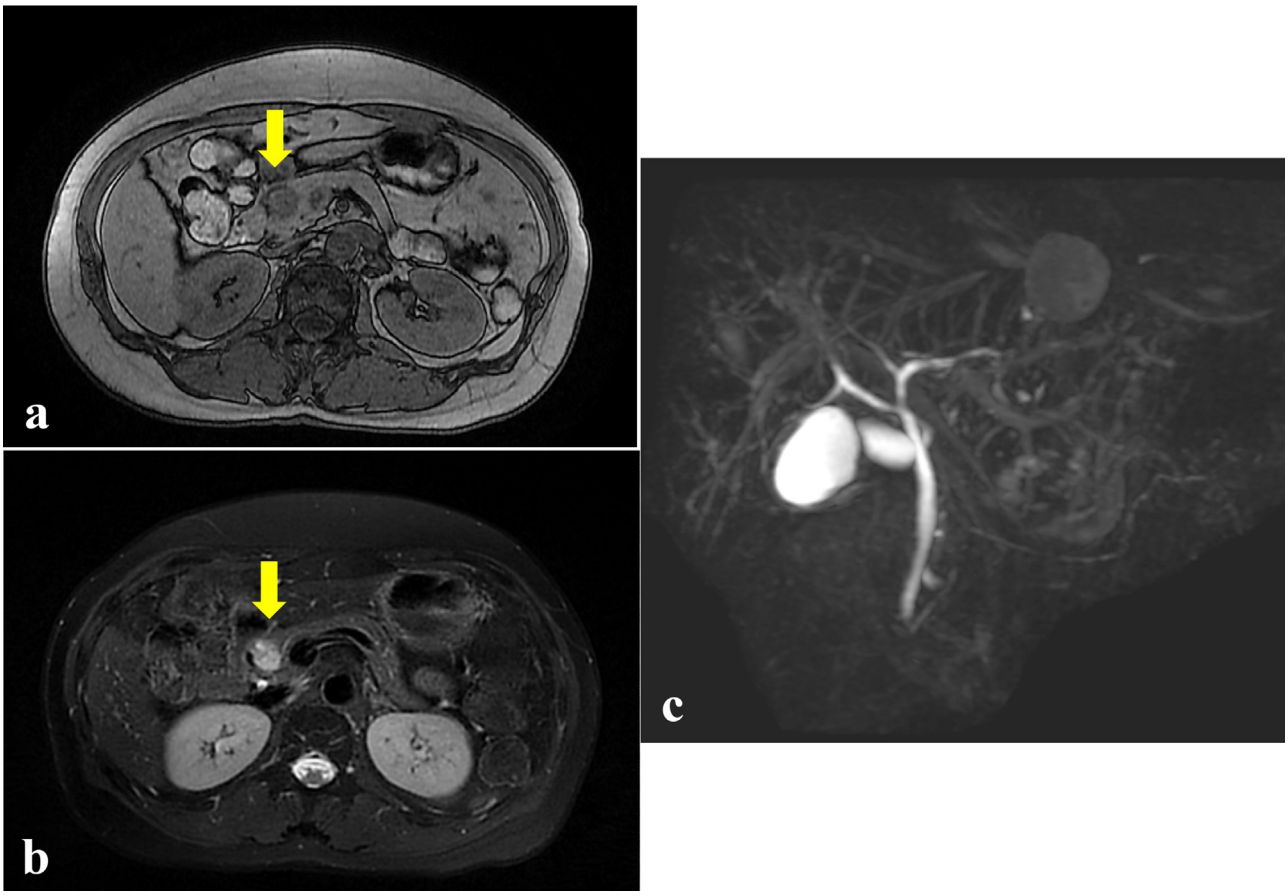

Fig. 3. Magnetic resonance imaging(MRI)showed a pancreatic mass with hypointensity on T1-weighted images (a) and heterogenous hyperintensity on T2-weighted images (b) (yellow arrow). Magnetic resonance cholangiopancreatography (MRCP) showed no abnormal findings in the main pancreatic duct (c).

Table 1
Reported cases of pancreatic schwannoma in which the results of FDG-PET/CT are described.

Case	Age Gender	Opportunity of findings	Location	Size (cm)	FDG-PET SUVmax	Treatment	Malignancy	Prognosis
1 [6]	79 male	incidental	head	5.0	3.9	SSPPD	benign	alive for 4.5 years no recurrence
2 [10]	40 female	incidental	head	5.0	3.6	PPPD	benign	ND
our case	59 female	incidental	head	2.0	5.8	SSPPD	benign	alive for 10 months no recurrence

FDG-PET fluorin-18 fluorodeoxyglucose positron emission tomography, *SUVmax* maximum standardized uptake value, *SSPPD* subtotal stomach-preserving pancreatoduodenectomy, *PPPD* pylorus-preserving pancreatoduodenectomy, *ND* not described.

kit, desmin, and smooth muscle myosin [14]. Recently, there have been several reports of pancreatic schwannoma that was accurately diagnosed preoperatively by endoscopic ultrasound-guided fine needle aspiration (EUS-FNA) [15,16]. If the diagnosis of pancreatic schwannoma is confirmed preoperatively, simple enucleation is sufficient since the vast majority of pancreatic schwannomas are encapsulated, slowly growing benign tumors. However, when the tumor is located in a periampullary site or close to the main pancreatic duct, simple enucleation is sometimes difficult. More invasive resection such a pancreatoduodenectomy or distal pancreatectomy might be necessary for an anatomical reason. Following complete tumor resection, patients with pancreatic schwannoma are reported to have a good prognosis. On the other hand, there have been several cases of malignant pancreatic schwannoma described in the English literature [17–19]. Diagnostic criteria for a malignant pancreatic schwannomas have not been established. Moriya et al. demonstrated a correlation between tumor size and malignant formation [14]. The authors emphasized the importance of early resection and accurate diagnosis. In cases in which the tumor shows malignant behavior (infiltration of tissue) or in close proximity to important vessels and in cases in which results of examination of a frozen section are inconclusive, an oncologic margin-negative resection has been recommended [17]. In our case, we could not make a definitive preoperative diagnosis of the tumor as pancreatic schwannoma and we could not exclude the possibility of a malignancy because of increased uptake FDP on PET/CT. Furthermore, the tumor was located close to the main pancreatic duct in the pancreas head, and we therefore performed SSPPD. Macroscopically, the tumor was a well-encapsulated circumscribed mass with no infiltration of surrounding tissue. Histopathologically, the proliferating cells showed no atypia and few mitoses with a Ki67

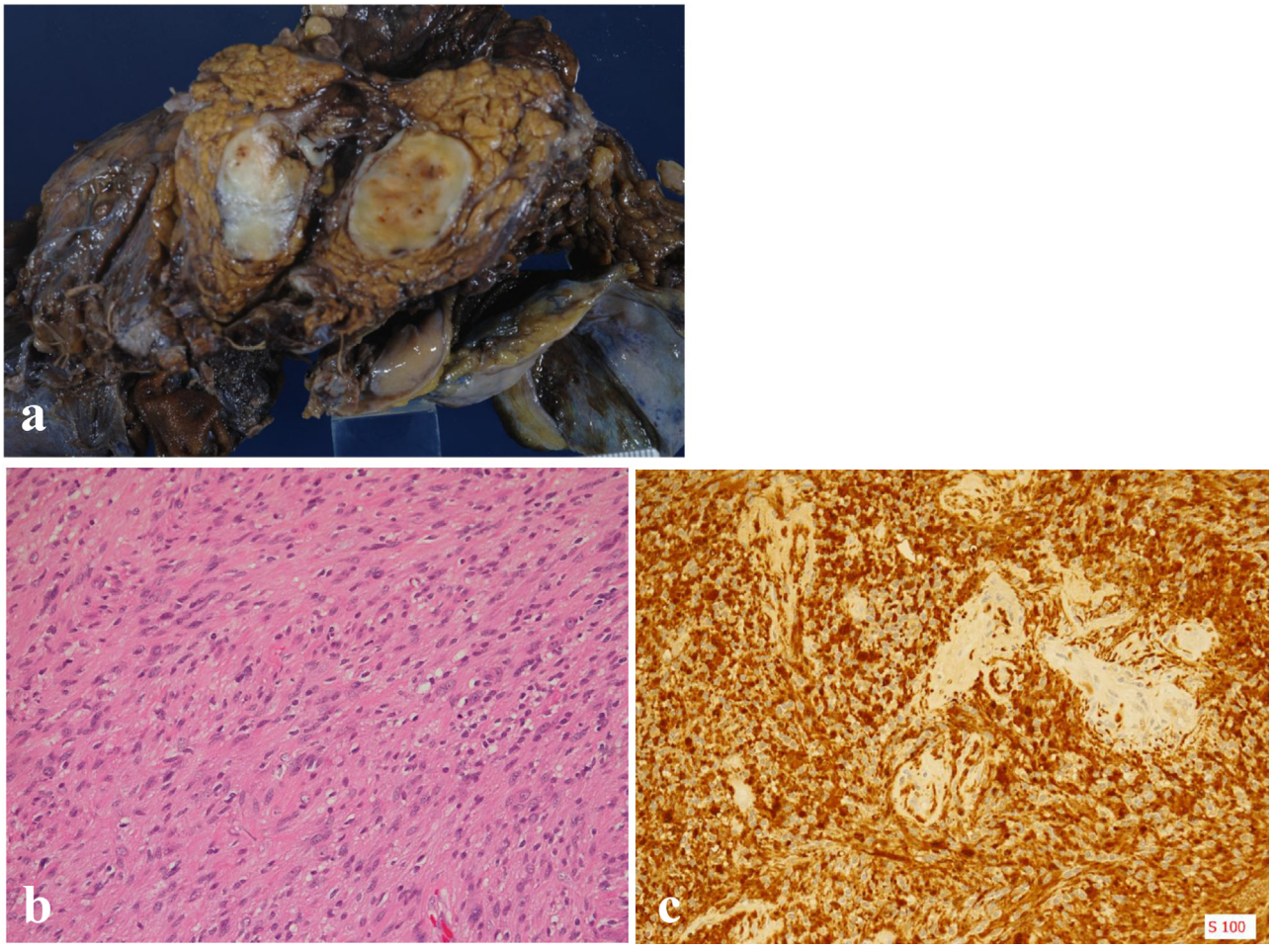

Fig. 4. Macroscopically, the tumor was a well-encapsulated circumscribed mass (a). Histopathological examination revealed proliferation of spindle cells showing interlacing and palisading patterns in the tumor. The proliferating cells showed no atypia and few mitoses (hematoxylin-eosin staining ×200) (b). Immunohistochemically, the spindle cells were positive for S-100 protein (c).

labeling index of 2%. These findings were consistent with a benign schwannoma.

4. Conclusions

Incidental detection of pancreatic schwannoma is predicted to increase due to the widespread use of CT and MRI. It is important to take into account this tumor in the differential diagnosis of pancreatic tumors. In addition, we should be aware that FDG-PET/CT shows abnormal accumulation in a benign pancreatic schwannoma.

Conflicts of interest

The authors declare that they have no competing interests.

Funding

None.

Ethical approval

This study does not include a research study.

Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Author contribution

S Fukuhara and S Fukuda made substantial contributions to conception and design and acquisition and interpretation of data. S Fukuhara and S Fukuda wrote the draft manuscript. HT, KI, TM, YH, S Fujisaki, MT, TN and HS performed the critical revision of the manuscript. S Fukuda gave final approval of version to be published. All authors read and approved the final manuscript.

Registration of research studies

This is not research study.

Guarantor

Saburo Fukuda.

Acknowledgement

None.

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